

**Mothers' Experiences of Caring for Children Receiving  
Growth Hormone Treatment**

**A Thesis Submitted in Fulfilment for the Degree of  
Doctor of Philosophy in Nursing**

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## **Declaration**

This thesis is submitted in total fulfilment of the requirements of the degree of Doctor in Philosophy. I declare that this thesis is entirely my own work unless otherwise acknowledged and has not been previously submitted as an exercise for a degree at Trinity College Dublin or any other university.

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## Abstract

The last five decades has seen a rapid expansion in the number of children requiring growth hormone treatment (GHT). Children generally begin GHT at a very early age which means parents often endure a complex and difficult pathway with regards to GHT decision making, administration, storage, medication adherence and costs. The literature revealed parents lack of choice in GHT devices, lack of support, lack of adherence, lack of reassurance about side effects, and organisational difficulties including; preparation, administration, storage and travel. Parents have significant responsibilities, yet there are no studies which reflect an empirical understanding of the essence of parents' experiences of caring for their child requiring GHT. The life-world of the parent that consists of social, practical, and taken-for-granted dimensions are largely unknown and therefore there was scope for this study. The aim was to explore parents' day to day experiences, the 'taken for granted' aspect of caring for children receiving GHT.

This study used the philosophy of hermeneutic phenomenology based on the approach of Gadamer. Sixteen mothers from the Republic of Ireland participated and the data were collected through in depth interviews and diary recordings. Sixteen in depth interviews and eight diaries were completed. The mothers' experiences of caring for their child receiving GHT were framed by three concepts that were: uncertainty, normalisation and stigma. These concepts were used to elaborate on the four major meanings encapsulating their experience of caring for their children receiving GHT. The four major meanings of their lived experiences which are explained in the form of themes are: (1) "It's the right thing to do" Striving for the security and the wellbeing of the child, (2) "Doubting yourself constantly" Constant uncertainty, (3) "But then you just get used to it I suppose" Adhering to GHT and lifestyle changes - the new normal, (4) "I hadn't been told anything about it" Information behaviour; looking for normality and certainty. Mothers experienced significant challenges coping with the uncertainties associated with GHT. The felt stigma of restricted growth, rare medical condition and GHT appeared to hinder some mothers from seeking support from their families, other parents and communities. In addition, the substantial need for information and support was noted. These findings indicate that healthcare

professionals and policy makers need to ensure that services for these children include the necessary supports for parents.

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## Structure of Thesis

The following summary outlines the structure of the thesis and the content of each chapter.

Chapter 1: Introduces the study background and the overall aim of the research that explores the experiences of parents caring for their children receiving growth hormone treatment (GHT).

Chapter 2: Provides a literature review and considers the existing published work relating to the overall context of children receiving GHT with a focus on studies describing parents' experience of caring for their children receiving GHT.

Chapter 3: Represents the chosen research methodology based on Gadamer's hermeneutic phenomenology. The chapter presents an overview of descriptive and interpretive phenomenology and provides an account of the research methods and the data analysis approach used for this study.

Chapter 4: Reports the findings and is separated into four sections, one for each of the four main themes emerging from the data.

Chapter 5: Offers a discussion of the findings and considers the overarching concepts of 'uncertainty', 'stigma' and 'normalisation'. This chapter reviews the main theoretical framework supporting the concept of 'uncertainty' and identifies areas of congruence with Mishel's seminal work (1988, 1990). Findings relating to the concept of 'normalisation' were also considered using Deatrick *et al.*'s (1999) five attributes of normalisation as a way of coping and adaptation. The chapter concludes with recommendations and the implications for clinical practice, nursing education and research.



# Table of Contents

Declaration.....	i
Abstract.....	ii
Acknowledgments .....	iv
Structure of Thesis.....	vi
Table of Contents.....	vii
List of Figures.....	xii
List of Tables.....	xiii
Abbreviations.....	xiv
<b>Chapter 1: Introduction.....</b>	<b>1</b>
1.1 Introduction.....	1
1.2 Background.....	1
1.3 Evolution of Research Interest .....	2
1.4 Research Purpose.....	3
1.5 Rationale for the Study .....	4
1.6 The Research Design.....	4
1.7 Conclusion .....	4
<b>Chapter 2: The Literature Review.....</b>	<b>5</b>
2.1 Introduction.....	5
2.2 The Place of the Literature Review in this Study .....	5
2.3 Source of Information.....	6
2.4 Importance of Child Growth .....	8
2.5 Growth Monitoring in Ireland.....	8
2.5.1 Measuring Technique and Equipment .....	9
2.5.2 Screening and Referral .....	9
2.6 Clinical Assessment and Investigations Aimed at Identification of a Primary Diagnosis.....	11
2.6.1 History and Physical Examination .....	11
2.6.2 Hormonal Status .....	11
2.6.3 Radiological Assessment.....	11
2.6.4 Genetic Testing.....	12
2.7 Growth Hormone Treatment (GHT).....	12
2.7.1 History and Devices .....	12

2.7.2	Indications for GHT.....	13
2.7.2.1	Growth Hormone Deficiency (GHD).....	14
2.7.2.2	Turner Syndrome (TS) .....	14
2.7.2.3	Prader-Willi Syndrome (PWS).....	15
2.7.2.4	Small for Gestational Age (SGA)/Russell Silver Syndrome (RSS) .....	15
2.7.2.5	Chronic Renal Insufficiency (CRI).....	16
2.7.2.6	Short Stature Homeobox-Containing Gene (SHOX).....	16
2.7.3	Prescribing of GHT and National Guidelines .....	17
2.7.4	Growth Hormone Treatment Side Effects.....	18
2.7.5	Discontinuation of GHT .....	19
2.7.6	Statistics .....	19
2.8	Parental Reaction to Child Growth.....	19
2.9	Parents and Administration of Injections .....	21
2.10	Parents and GHT Devices .....	23
2.11	Treatment Burden and Adherence.....	24
2.12	Parental Concerns About Growth Hormone Treatment - Allocating the Gap in the Literature .....	30
2.13	Growth Hormone Treatment Research Involving Ireland .....	32
2.14	Conclusion .....	34
	<b>Chapter 3: Methodology and Methods.....</b>	<b>36</b>
3.1	Introduction.....	36
3.2	Worldview .....	36
3.3	Choosing a Research Approach .....	37
3.4	Influences on the Choice of Research Approach .....	38
3.5	Philosophical Foundation of Phenomenology.....	38
3.6	Theoretical Lens .....	40
3.6.1	Choosing Phenomenology.....	40
3.6.2	Choosing Hermeneutics.....	41
3.6.3	Choosing Gadamer’s Hermeneutics.....	42
3.7	Research Method .....	44
3.8	Ethical Considerations .....	44
3.9	Sample and Recruitment .....	45
3.9.1	Targeted Sample Group.....	45

3.9.2	Recruitment Process .....	46
3.9.2.1	Phase One (May 2015-April 2016).....	47
3.9.2.2	Phase Two (April 2016 on June 2017) Successful Attempt .....	49
3.9.3	The Study Sample.....	51
3.10	Methods of Data Collection or Gaining Understanding .....	52
3.10.1	Indepth Interviews .....	52
3.10.2	Conducting Interviews.....	54
3.10.3	Diaries.....	56
3.11	Chosen Analytical Framework.....	58
3.11.1	Deciding Upon a Research Question .....	62
3.11.2	Identification of Pre-understandings (Before Data Collection) .....	63
3.11.3	Gaining Understanding Through Dialogue with Participants (Interview and Diary) .....	64
3.11.4	Gaining Understanding through Dialogue with Text (Transcribing and Analysing).....	65
3.11.5	Establishing Trustworthiness.....	67
3.11.5.1	Member Checking .....	70
3.11.5.2	Using NVivo.....	71
3.12	Conclusion .....	72
<b>Chapter 4: The Findings</b> .....		73
4.1	Introduction.....	73
4.2	Alteration to Transcripts .....	73
4.3	Sample .....	74
4.4	The Children’s Needs and Diagnosis .....	75
4.5	Theme 1: “It’s the right thing to do” Striving for the Security and the Wellbeing of the Child .....	78
4.6	It is a Mothers’ Duty to Protect.....	79
4.6.1	Advocating and Seeking Partnership.....	84
4.6.2	Decision to Start GHT /Normalising GHT.....	87
4.6.3	Adapting and Being Sensitive to the Child’s Needs .....	97
4.6.4	The Continuity of GHT as a Source of Wellbeing.....	102
4.6.5	The Future Well-being of the Child .....	105

4.6	Theme 2: “Doubting yourself constantly” Constant Uncertainty .....	107
4.6.1	Diagnostic Uncertainty .....	107
4.6.2	Treatment Uncertainty.....	117
4.6.3	Future Uncertainty .....	124
4.7	Theme 3: “But then you just get used to it I suppose” Adhering to GHT and Lifestyle Changes - The New Normal .....	129
4.7.1	Managing GHT Injections.....	129
4.7.2	Lifestyle Adjustments.....	142
4.7.3	Attending Endocrine Follow Up Appointments .....	153
4.7.4	Vigilance to Treatment Side Effects and Positive Outcomes .....	160
4.8	Theme 4: “I hadn’t been told anything about it” Information behaviour; Looking for Normality and Certainty .....	164
4.8.1	Information Needs .....	164
4.8.2	Seeking and Sharing Information .....	169
4.8.3	Need for Support .....	176
4.9	Conclusion .....	181
	<b>Chapter 5: Discussion</b> .....	182
5.1	Introduction .....	182
5.2	Uncertainty .....	183
5.2.1	Antecedents of Uncertainty .....	185
5.2.1.1	Stimuli Frame .....	186
5.2.1.2	Cognitive Capacity .....	187
5.2.1.3	Structure Providers .....	187
5.2.2	Appraisal of Uncertainty and Coping Strategie .....	188
5.3	Mothers’ Experiences of Uncertainty .....	189
5.3.1	Antecedents (Triggers) of Mothers’ Uncertainty .....	191
5.3.1.1	Diagnostic Uncertainty.....	192
5.3.1.2	Treatment Uncertainty.....	196
5.3.1.3	Future Uncertainty.....	202
5.3.1.4	Social Stigma Uncertainty.....	204
5.3.2	Mothers’ Appraisal of Uncertainty.....	206
5.3.3	Mothers’ Coping and Adaptation.....	207
5.4	Normalisation as a Significant Adaptation Technique .....	213
5.5	The Temporary Interpretation is Achieved “Fusion of Horizons”.....	217

5.6 Recommendations .....	223
5.6.1 Clinical Practice.....	223
5.6.2 Nursing Education.....	227
5.6.3 Further Research.....	228
5.7 Limitations.....	228
5.7.1 The Methodological Approach.....	228
5.7.2 The Sample .....	229
5.7.3 Data Collection Methods .....	230
5.7.4 Being a Parent and a Single Researcher .....	231
5.8 Conclusion .....	224
<b>References</b> .....	234
<b>Appendices</b> .....	263
Appendix 1- Participant Information Leaflet.....	263
Appendix 2- Informed Consent Form.....	266
Appendix 3- Introductory Letter to the Study .....	268
Appendix 4- Diary .....	269
Appendix 5- Pre-understandings Prior to Starting Data Collection .....	271
Appendix 6- Example of a Reflective Diary Log for One of the Interviews .....	273
Appendix 7- Example of the Initial Preliminary Interpretation of Texts.....	274
Appendix 8- Example of Open Codes in a Sample Interview .....	275
Appendix 9- Codes to Categories Done Manually .....	276
Appendix 10- Codes to Categories Using NVivo 11 .....	276
Appendix 11- Example of Finding Subcategories .....	277
Appendix 12- Core Categories and Subcategories.....	279
Appendix 13- Synthesis and Initial Theme Development .....	283
Appendix 14- Example of a Reflexive Log .....	288
Appendix 15- Examples of Annotations in NVivo.....	290

## List of Figures

Figure	Title	Page
1	The Process Leading to the Final Themes	77
2	Mishel's Uncertainty in Illness Model (1988)	185
3	Mishel's Reconceptualised Model of Uncertainty in Chronic Illness (1990)	185
4	Part One of Mishel's Uncertainty in Illness Model (1988) (Antecedents)	180
5	Part Two of Mishel's Uncertainty in Illness Model (1988) (Appraisal and Coping)	188
6	Mothers' Experiences of Uncertainty Adapted from Mishel's Uncertainty in Illness Model	190
7	Completing the Hermeneutic Circle (The Final Interpretation)	218
8	Initial Themes Evolving to Final Themes as They were Challenged by the Literature	219
9	Uncertainty May be Ongoing or Evolving Overtime Adapted from Mishel (1990)	220
10	The Relationship Between Uncertainty and Reaching Normality Adapted from Deatrck <i>et al.</i> (1999)	220
11	Final Framework: Fusion of Horizons	222

## List of Tables

<b>Table</b>	<b>Title</b>	<b>Page</b>
1	Inclusion and Excision Criteria	6
2	Description of the Sample	51
3	Medical Conditions Involved in the Study	52
4	Analytical Framework Used for Data Analysis; Modified from Fleming <i>et al.</i> (2003) and Ajjawi & Higgs (2007)	60
5	Pseudonyms	74
6	Geographical Location of Sample	75
7	Table Representation of the Study's findings Detailing the Main Concepts, Themes and Subthemes	77

## Abbreviations

BiPAP	Bilevel Positives Airway Pressure
BSPED	British Society for Paediatric Endocrinology and Diabetes
CGF	Child Growth Foundation
CPAP	Continuous Positive Airway Pressure
CRI	Chronic Renal Insufficiency
ESPE	European Society for Paediatric Endocrinology
GHD	Growth Hormone Deficiency
GHT	Growth Hormone treatment
HSE	Health Service Executive
IGHD	Idiopathic Growth Hormone Deficiency
IGF-1	Insulin-Like Growth Factor-I
IM	Intramuscular
IUGR	Intrauterine Growth Restriction
MAGIC	Major Aspects of Growth in Children
NHS	National Health Services
NICE	National Institute for Clinical Excellence
NICU	Neonatal Intensive Care Unit
PWSAI	Prader-Willi Syndrome Association Ireland
PWS	Prader-Willi Syndrome
rhGH	Recombinant Human Growth Hormone
RSS	Russel Silver Syndrome
SCBU	Special Care Baby Unit
SDS	Standard Deviation Score
SGA	Small for Gestational Age
SHOX	Short Stature Homeobox-Containing Gene
SQ	Subcutaneous
TCGI	Turner Contact Group Ireland
TS	Turner Syndrome



# **Chapter 1: Introduction**

## **1.1 Introduction**

In this chapter the background, motives for the study, purpose, rationale and research design will be provided.

## **1.2 Background**

The last five decades have seen a rapid expansion in the number of children requiring growth hormone treatment (GHT), a 'biosynthetic' growth hormone which aims to increase height to achieve normal height in early childhood (Kirk 2012). In the past, children who needed GHT had to receive it in the form of intramuscular injections two to three times a week at clinics (Moore *et al.* 1987). Then in the 1980s, due to technological advances, GHT became available in the form of self-administered subcutaneous injections that could be delivered at home (Kastrup *et al.* 1983). In addition, children could commence GHT at a very early age which meant that parents had to undertake responsibility for GHT administration (Van Dongen & Kaptein 2012, Kaptein 2013). Consequently, parents often have to undertake complex care with regards to GHT administration, storage, medication adherence, and costs.

The involvement of the parents may be a contributory factor in adherence to GHT administration (Laing 2014). Unfortunately lack of adherence to GHT is still a reported problem in the literature (Van Dongen & Kaptein 2012). It is known that lack of adherence can lead to more repeat visits to the hospital for more diagnostic tests and unnecessary changes to the treatment course that may lead to worse overall outcomes and increased health care costs (Fisher & Acerini 2013). Moreover, parents' perceptions of their child's illness and GHT benefits can play a role in their acceptance and adherence to GHT (Van Dongen & Kaptein 2012).

The literature review revealed that parents lack choice in type of GHT device, support, adherence and reassurance about side effects. The literature also

showed many organisational difficulties associated with GHT including; medication preparation, administration and storage (Van Dongen & Kaptein 2012, Kremidas *et al.* 2013, Marini *et al.* 2016, Brod *et al.* 2017). These existing challenges highlight the significance of the needs of parents and thereby the extent of responsibility of the healthcare providers to ensure they support the individual needs of children on GHT and their parents.

Therefore, is a need to understand and reveal the life-world of parents who care for their children needing GHT. By offering further understanding of the experiences encountered by parents, it is hoped that professionals will have greater understandings of the issues faced by these families and thereby improve current practice which will help to improve the commencement and maintenance of GHT regimen.

### **1.3 Evolution of Research Interest**

In 2005, I graduated with a bachelor's degree in Nursing in the Kingdom of Saudi Arabia (KSA). In 2007, I was employed as a clinical nurse educator in the Department of Child Health Nursing at the School of Nursing at King Abdul-Aziz University. I have always enjoyed teaching and working in children's nursing. In 2009, I was blessed with my second son who was diagnosed with Russell Silver syndrome (RSS) at the age of 6 months. Between having a background in child health nursing and mothering a child with a growth disorder, I was inspired to research this area. As my son's condition causes him to be very short and petite, growth hormone treatment (GHT) was suggested as a potential treatment since he was diagnosed in 2010. A year later he commenced GHT to help him reach his potential adult height. In 2012, for my MSc in Child Health at the School of Nursing and Midwifery in Trinity College Dublin, I systematically reviewed the effect of GHT on the height of children with RSS for my thesis. After this project, I became aware of how GHT can help many children grow to an optimal height and the potential side effects, adherence issues and technical difficulties.

My husband and I had to make the decision to start our son on GHT when he was one and a half years old after it was prescribed by his endocrinologist to enhance his potential adult height. When we made the decision to commence him on GHT, we

had mixed feelings; feeling hopeful and happy to start our son on a treatment that may help him reach his potential adult height, but also feeling overwhelmed with making this big decision to commit to this invasive treatment. Being both a mother and a nurse, I was anxious about giving my son daily injections. Even with seeing good outcomes from the treatment, I still faced emotional challenges. I always wondered how other parents with no medical or nursing background managed GHT administration.

In addition, through social media and support pages, I had read about the experiences of other parents caring for children receiving GHT. It seemed to me that parents have many questions and concerns related to their child's GHT that are unanswered. I also recognised a dearth in literature available around parents' experiences of giving GHT or, caring for a child receiving GHT which does not capture the intensity or the totality of the experiences that I know of. I also encountered professionals who work in this area and they seem to underestimate the phenomena. This confirmed that some professionals may not understand the reality as lived by individuals experiencing these phenomena. I soon realised that parents' experiences may never be visible and may always be taken for granted. Thus, with my experience of having a child that needs GHT and my nursing background, I sought to investigate parents' experiences.

I cannot help but hold prejudices, assumptions and have the mindset of an experienced parent caring for a child on GHT for the last seven years and a previous researcher in the field of GHT. Therefore, I expected to have some issues in common with the participants but endeavoured to be critically aware of my positioning in the research process.

#### **1.4 Research Purpose**

The purpose was to explore parents' experiences of caring for a child receiving GHT in Ireland. The aim was to explore the parents' lifeworld and from this, develop a greater understanding of the nature of their experiences and identify potential interventions that may support parents. The intention was to deliver rich, indepth descriptive and interpretive evidence by uncovering these experiences, and consequently, to offer direction to professional children's nursing practice and to inform and add to the evidence base for healthcare services.

## **1.5 Rationale for the Study**

There is little evidence from an international and an Irish perspective of the experiences of families of children receiving GHT. Although the child's perspective is equally as important as the parents, little is known about parents' caregiving experiences of providing GHT for their child. The care given by parents to a child needing GHT requires skilled practice that can make physical and psychological demands on them. Yet these demands and consequences from the parent's perspective are limitedly identified. The lifeworld of the parent that consists of social, practical, and taken-for-granted dimensions are largely unknown and therefore there was scope for a new study. It is timely that appropriate services are developed from a user perspective. By offering further understanding of the experiences encountered by parents it is hoped that professionals will have greater understanding of the issues faced by these families and thereby improve current practice which will help with the commencement and maintenance of GHT regimen.

## **1.6 The Research Design**

This study was guided by the philosophy of hermeneutic phenomenology, using the approach developed by Gadamer (1975). This offers a way to investigate subjective phenomena and is based on the belief that truths about reality are grounded in everyday experiences (Van Manen 1977). Phenomenology inspects the meanings that lived experience has on people's lives. The fundamental purpose is the study of the individual's experiences with a foremost concern directed at the person and his/her views. The emphasis throughout was on the subjective experience of the world as it was experienced by each parent and attention was given to a full picture of their experiences. Phenomenology is discussed further in the third chapter.

## **1.7 Conclusion**

This first chapter has provided the background, my personal situation, rationale and approach for this study. The next chapter provides a review of the literature and appraises empirical studies which explored the experiences of parents caring for their child commenced on GHT.

## **Chapter 2: The Literature Review**

### **2.1 Introduction**

In this chapter, the literature related to the topic will be critically discussed. This review begins with the importance of child growth, monitoring, detecting, and referral for growth failure in children in Ireland. It then provides an overview of growth hormone treatment (GHT), the indications, safety and prevalence. Following this, studies reporting parents' experiences in relation to child growth, use of injection and GHT devices, treatment burden, and adherence. The studies conducted in Ireland in relation to GHT are discussed and the existing gap in the literature is highlighted.

### **2.2 The Place of the Literature Review in this Study**

The timing and completion of a literature review that forms the basis for ideas before or during the study to identify the aim and the research question, vary in accordance with the research method used (Grbich 1998). Although an indepth literature review is suggested prior to conducting a quantitative research, there is a debate regarding its use in qualitative research (Parahoo 2014). It is suggested that researchers should not generally begin with an extensive review in qualitative research because this will limit the possibility of the researchers' presumptions or biases about the topic emerging (Speziale & Carpenter 2007, Wojnar & Swanson 2007). Furthermore, Koch (1995) recommends that researchers should thoroughly consider the philosophical underpinnings of their chosen methodology prior to commencing the review as this will impact the approach taken. Based on the chosen research philosophy of interpretive phenomenology, a literature review should not be conducted. However, others suggest that knowledge of existing work is beneficial to guide the direction of the study (Silverman 2010).

Therefore, it was necessary to identify literature known about the topic and confirm an absence of similar studies to provide justification for this study. A review of studies concerning children receiving GHT and those exploring parents' experiences of caring for a child on GHT was conducted. Later, another review was carried out near the end

of the research to contextualise the findings and to reveal how the findings compared to existing knowledge about the phenomenon. Using this flexible approach, it was feasible to let findings emerge with limited potential contamination with other studies. When the data were analysed, the literature was used to confirm and contextualise the findings. This helped understand the parents' experiences by applying a holistic approach without letting external influences or pre-existing structures from other work influence the findings of the study.

### 2.3 Source of Information

A literature search was conducted using electronic databases including British Nursing Index (ProQuest), Cumulative Index to Nursing and Allied Health Literature (CINAHL), Cochrane Database of Systematic Reviews, Lenus, PsycINFO, and PubMed. Key search terms included: attitudes, caring, children, experiences, fathers, growth hormone treatment, growth hormone therapy, human growth hormone, mothers, parents, perceptions, recombinant human growth hormone. Generally, literature from 1985-2018 was sourced though earlier seminal literature was included if appropriate. This was based on the recognition that GHT in the form of subcutaneous synthetic GH to treat children was introduced in 1985. Only studies published in English were included. Searches were undertaken of major texts and publications from the UK and Irish Government (British Society for Paediatric Endocrinology and Diabetes (BSPED), European Society for Paediatric Endocrinology (ESPE), Health Service Executive (HSE), National Health Service (NHS), National Institute for Health and Care Excellence (NICE)). Literature from voluntary organisations, including the Major Aspects of Growth in Children (MAGIC) Foundation, Prader-Willi Syndrome Association Ireland (PWSAI), Turner Contact Group Ireland (TCGI) and Child Growth Foundation (CGF) were sourced. The advice of the university specialist librarian was obtained. Information was also sought from main growth hormone treatment manufacturing companies in Ireland. See Table 1 for the inclusion and exclusion criteria of the literature review.

Table 1: Inclusion and Exclusion Criteria

Inclusion Criteria	Exclusion Criteria
Document Type:	

<p>Research articles from electronic databases</p> <p>Major texts and publications from the UK and Irish Government.</p> <p>Literature from voluntary organisations.</p> <p>Information from main Growth hormone treatment manufacturing companies in Ireland.</p> <p>Dates: literature from 1985-2018</p> <p>Language: Studies published in English</p> <p>Population: Mothers and fathers (parents) and Caregivers</p> <p>Content: Studies that refer to the parents' or caregivers' management, views, experiences, attitude and perceptions on/with GHT.</p>	<p>Dates: Literature before 1985</p> <p>Language: Studies published in non-English</p> <p>Population: Children, siblings and healthcare providers</p> <p>Content: Studies on GHT that do not include parents or caregivers. Studies on GHT that include parents or caregivers as proxies for the child's experience with GHT and not their own.</p>
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After extensively searching the literature, only four relevant studies were found. These studies were conducted in the following countries: Germany, Italy, the Netherlands, United Kingdom and the United States of America. One of the four studies was conducted in three of these countries. This review begins with the importance of child growth and the monitoring, detecting, and referral of growth failure in children in Ireland. It is then followed by a description of GHT, reasons for initiating it including the common diagnoses, the safety of the treatment and its prevalence. Finally, literature specifically exploring the parents' experiences with GHT administration, treatment burden, and adherence are discussed.

## **2.4 Importance of Child Growth**

Growth is one of the most fundamental tasks of childhood development. Height and weight are primary indicators of child's health and physical development (Growing Up in Ireland 2013). Normal growth only happens when a child is healthy, sufficiently nourished and emotionally secure (Hoey 2014). Adequate lengthening of the skeleton relies on the intricate collaboration between genetic, hormonal, nutritional and psychosocial variables especially during the first two decades of a human's life (Smith & Werther 2005). Monitoring growth carefully is essential for the early recognition of disease, particularly a broad range of endocrine abnormalities in which restricted growth would be the initial or the only sign of a medical problem (Drake & Kelnar 2006, Laing 2014). Therefore, growth assessment and monitoring are an essential part of the health examination or medical investigation of any child and are usually done as part of good routine clinical care and child health visits like immunisation visits and developmental assessments (HSE 2012, Hoey 2014). Through early detection of underlying growth failure, the diagnosis and management of treatable growth disorders, the reduction of complications and/or associated comorbidity and the improvement of health, height prognosis and quality of life can be attained (Léger 2017).

The European Society of Paediatric Endocrinology (ESPE) categorises factors affecting growth in its ordering of paediatric endocrine diagnosis and groups them into three broad categories which are: primary growth failure, secondary growth failure and idiopathic short stature (Laing 2014). It has been noted that short stature in childhood is one of the most common growth problems after childhood obesity (Hoey 2014). Reaching the maximum height potential for each child is essential (Smith & Werther 2005). One of the ways doctors can improve impaired growth in children is by prescribing growth hormone treatment (GHT).

## **2.5 Growth Monitoring in Ireland**

This section provides an overview of the current recommendations in Ireland drawn from the Health Service Executive Training Program for Public Health Nurses and Doctors in Child Health Screening, Surveillance and Health Promotion report (HSE



2012). Height and weight should be measured, and charted on a growth chart, whenever parents or professionals hold concerns about a child's growth. Plotting on a growth chart should be routine practice in the follow-up of children with chronic disorders or disabilities. A single measurement will identify children who are particularly short or tall outside the limits of the 0.4 and 99.6 centiles. In addition, children should have their height measured and charted at school entry (HSE 2012).

### **2.5.1 Measuring Technique and Equipment**

- Using proper equipment that is frequently calibrated and maintained reduces instrument variation. The recommended equipment by the HSE (2012) are:
  - Electronic self-zeroing scales, Grade 3 clinical electronic scales in metric setting.
  - Supine length measure (infantometer or baby mat).
  - Leicester height measure (self-calibrating)/stadiometer.
  - Non-stretchable/lassoo 0.5 cm tape for head circumference.
- Taking measurements at approximately the same time of day under the same conditions lessens subject variation. These errors can be reduced by standardisation of methods and training (HSE 2012).

### **2.5.2 Screening and Referral**

Including 'cut-off points' in the growth-monitoring programme determines the sensitivity and specificity of screening. Setting the 0.4th centile as the cut-off-point for short stature will increase the chance of identifying children who are below the 0.4th centile having a growth problem but will increase the risk of overlooking a child with disordered growth (number of missed cases or false negatives) (HSE 2012). While setting the 3rd centile as the cut-off-point reduces the chance of missing or delaying the detection of those with short stature, but will increase the number of false negatives, that is, children who are being referred for a possible growth disorder but whose growth is normal (Hindmarsh 1996).

The choice of referral criteria is debatable in all screening programmes (HSE 2012).

The endocrinologists that took part in the Growth symposium in 2005 to develop the referral criteria had strong views, proposing the 3rd percentile be selected as the cut-off point for better early recognition and referral for those with growth disorders (HSE 2012). There is an apprehension that setting the 0.4th centile will result in missing some children with pathological causes of short stature including Turner syndrome, celiac or thyroid disease etc (HSE 2012). Concern has also been raised in the UK that this cut-off level is too strict (Agwu *et al.* 2004). However, the general view was that the 0.4th percentile would be the standard cut-off point (HSE 2012).

As a result, it has been stipulated that children must be referred at any time if there is a clinical concern or parental concern about their growth regardless of their centile line (HSE 2012). Communication of growth concerns to both family and general practitioner (GP) are a vital constituent and any child whose measurements are outside the estimated cut-off centiles should be considered for further evaluation and/or specialist referral (HSE 2012). The HSE (2012) has indicated that with the lack of nationally agreed referral guidelines, it is recommended that referrals are made in the first instance to the GP, then to the local paediatric service if no clear reason for the problem is recognised.

However, even though these guidelines and criteria are drawn from the growth-monitoring 'Training program for public health nurses and doctors in child health screening, surveillance and health promotion' manual for healthcare providers in Ireland to follow, it should be noted that Ireland continues to lack standardised growth monitoring and referral guidelines for growth hormone deficiency (GHD) in children (Hawkes & O'Connell 2016). Hawkes & O'Connell (2016) reviewed the current practice in Ireland surrounding child measurement and diagnosis of GHD and found a significant variation in practice related to childhood measurement and GH prescribing in Ireland. The authors concluded the need for national guidelines in the evaluation and treatment of children with possible GHD and accurate measurement of children in clinics to guarantee that axiological techniques and equipment are in agreement with national guidelines.

## **2.6 Clinical Assessment and Investigations Aimed at Identification of a Primary Diagnosis**

### **2.6.1 History and Physical Examination**

In relation to what has been mentioned above, the history and physical examinations are fundamental and great attention should be paid to premature and/or small for gestational age (SGA) (low birth weight or birth length) birth (Léger 2017). The occurrence of a chronic condition should be considered, and dysmorphic features should be recognised (Barstow & Rerucha 2015). Parental growth and adult heights are pertinent and known to be connected to the response to GH (Savage & Bang 2012).

### **2.6.2 Hormonal Status**

The noting of genetic defects in the GH–insulin-like growth factor (IGF)-axis has emphasised the significance of endocrine assessment, comprising an identification of serum insulin-like growth factor-I (IGF-I) and GH secretion (Léger 2017). Growth hormone deficiency (GHD) should be diagnosed early in patients with neonatal symptoms of hypoglycaemia and extended jaundice, a characteristic growth pattern and likely of more pituitary hormone deficiencies (Savage & Bang 2012). Growth hormone deficiency in these children and in those with less severe idiopathic growth hormone deficiency (IGHD) can be confirmed by a low IGF-I concentration and GH stimulation testing with a GH cut-off set at 7 or 10 µg/l (Savage & Bang 2012, Léger 2017). Serum IGF-I levels are largely GH-dependent but also influenced by age, pubertal development, malnutrition, chronic inflammation or hepatic diseases (Savage & Bang 2012).

### **2.6.3 Radiological Assessment**

Magnetic resonance imaging (MRI) of the hypothalamic-pituitary region must be done when GHD is diagnosed, to eliminate an organic cause such as a tumour (Barstow & Rerucha 2015, Léger 2018). A skeletal survey is designated for body

disproportion and an X-ray of the left hand and wrist for bone age, though not diagnostic, may be significant for management (Savage & Bang 2012).

#### **2.6.4 Genetic Testing**

Genetic testing is significant to identify conditions that are associated with a short stature such as Prader-Willi syndrome, Turner syndrome, Russell Silver syndrome and short stature homeobox-containing gene (SHOX) (Savage & Bang 2012).

### **2.7 Growth Hormone Treatment (GHT)**

The terms recombinant human growth hormone (rhGH), human growth hormone (hGH) or somatotropin are all terms used to refer to growth hormone treatment (GHT). Recombinant human growth hormone (rhGH) is produced by the pituitary gland, which stimulates growth, cell reproduction, and cell regeneration (Salem 2009). Human growth hormone is vital for normal growth in children. It influences by direct action on the growth plates and by the production of insulin-like growth factors (IGF-1), mostly in the liver, which in turn increases growth (NICE 2010).

#### **2.7.1 History and Devices**

Growth hormone treatment was first used for growth hormone deficiency (GHD) in 1958; cadaveric GH was replaced with a more human-like growth hormone – recombinant human GH (rhGH) in 1985 (Kemp & Frindik 2011). Cadaver growth hormone was used to treat children with GHD (it was extracted from the human pituitary gland of cadavers) until it became an officially recognised cause of Creutzfeldt-Jacob disease in some children in 1985 (Taback & Van Vliet 1999). Soon after, it was produced synthetically in commercial labs and called ‘biosynthetic’ growth hormone which had the identical amino acid sequence as that found in human growth hormone ‘somatotropin’ (Salem 2009). GHT has now been accessible for over five decades, with all GHT now biosynthetically manufactured, and administered by daily subcutaneous injection (Kirk 2012).

Growth hormone delivery devices have greatly evolved since the 1960s when patients used to receive GH intramuscular injections two to three times a week in clinics (Moore

*et al.* 1987). Then in the 1980s, GHT became available in daily subcutaneous injections that patients could self-administer at home (Kastrup *et al.* 1983). Growth hormone treatment delivery devices progressed from using syringes and needles to injection pens, electronic injections, and needle-free injections. This evolving process was underlined by the goals of simplifying the delivery of the drug and reducing needle phobia and anticipated injection pain (Rohrer *et al.* 2017).

### **2.7.2 Indications for GHT**

Impaired growth velocity caused by either insufficient GH production such as in growth hormone deficiency (GHD) or by an impaired response to physiological GH levels such as Turner's syndrome are the most important reasons why children are prescribed GHT (Kappelgaard & Laursen 2011, Kirk 2012). The aim of GHT is to increase height to achieve normal height in early childhood and then preserve a growth pattern to normalise adult height that is suitable for age, sex and within the genetic target range (Kappelgaard & Laursen 2011). Not only is it used in conditions to improve height but also for other reasons such as in Prader-Willi syndrome (PWS) for improvement in body composition, bone density, cardiovascular risk, respiratory function, behaviour, socialisation, and self-esteem (Kirk 2012).

Growth hormone deficiency (GHD) remains the main indication for GHT in children and can be due to various aetiology, with both congenital and acquired causes (Audi *et al.* 2013). GHT has also been approved in the USA and Europe for other paediatric conditions with non-GH deficient states characterised with short stature, such as Turner syndrome (TS), small for gestational age (SGA) with failure to catch-up, Russell Silver syndrome (RSS), Prader-Willi syndrome (PWS), short stature homeobox-containing gene (SHOX) deficiency, chronic renal insufficiency (CRI) and, in the USA but not Europe, Noonan syndrome (NS) and idiopathic short stature (ISS) (Kirk 2012, Van Dongen & Kaptein 2012, Cappa *et al.* 2016). Once the condition is diagnosed or height velocity falls below the normal level, GHT is usually prescribed (Kirk 2012).

### **2.7.2.1 Growth Hormone Deficiency (GHD)**

Growth hormone deficiency is the most common indication for GHT in childhood and it is said that a quarter of all children with a height below  $-3$  standard deviation score (SDS) have GHD. If untreated, final height is estimated to be 134–146 cm in males, and 128–134 cm in females (Kirk 2012). The incidence of GHD is estimated to be between 1 in 3500 and 1 in 4000 children and for about half of the children with GHD, the cause is unknown (idiopathic) (NICE 2010). About 70% of patients with GHD suffer from an isolated deficiency of GH, but it can also manifest as part of multiple pituitary hormone deficiencies. It is most commonly caused by congenital (due to midline embryonic defects and transcription factor mutations), acquired (due to a pituitary/hypothalamic tumour, trauma, irradiation, infiltration, infection) or idiopathic (Kirk 2012). Short-term and long-term studies have revealed that starting GHT early (before the age of 3) in children who are diagnosed with GHD, had a noticeable effect in early catch-up growth and in a major improvement in height (Antoniazzi *et al.* 2015).

### **2.7.2.2 Turner Syndrome (TS)**

Turner syndrome is a chromosomal disorder which complete or partial lack of one X chromosome in girls occurs (Nadeem & Roche 2014). Girls with TS manifest with the two most common features that are, short stature and ovarian failure (NICE 2010). They do not have a deficiency in GH but may have a relative lack of sensitivity to GH because of haploinsufficiency of the short stature homeobox-containing gene (Nadeem & Roche 2014). Not all girls with TS need GHT (NICE 2010). Turner syndrome happens in between 1 in 1500 and 1 in 2500 live female births. If untreated, these girls usually have a final adult height of 136–147 cm. Adult women with TS are typically 20 cm shorter than other adult women (NICE 2010). GHT has proven to have a significant effect on bettering the height outcomes for girls diagnosed with TS regardless of starting age or duration of GHT (Nadeem & Roche 2014) while, other studies have indicated that the sooner GH is started the better the height outcome (Baxter *et al.* 2007, Kirk 2012).

### **2.7.2.3 Prader-Willi Syndrome (PWS)**

This is a dysmorphic syndrome with clinical features comprising short stature, obesity, obesity-related diseases, abnormal body composition, hypogonadism, hypotonia, hyperphagia, hypoventilation, and behavioural and learning difficulties (Kirk 2012). This genetic disorder is caused by the loss of paternally derived genes on 15q, due to deletion in 70%, but also maternal uniparental disomy, translocations and imprinting defects (Chen *et al.* 2007, Kirk 2012). Prader-Willi syndrome occurs in between 1 in 15,000 and 1 in 25,000 live births with men having a final adult height of about 154 cm and women having a final adult height of 145–159 cm (NICE 2010). It is uncertain whether children with PWS are truly GH deficient or not, although PWS does share some phenotypic features: short stature with poor growth, abnormal body composition (decreased muscle mass, increased fat mass) and hypothalamic dysfunction (hyperphagia, central hypogonadism) (Kirk 2012). In PWS the aim of GHT is to improve both body composition as well as enhancing growth, and GHT results in correcting both height (approximately 1 SD in the first year), body composition (less fat and more lean mass) and muscle strength/tone (Kirk 2012). The benefits GHT has on the cognitive and adaptive abilities in children with PWS has also been reported recently (Dykens *et al.* 2017).

### **2.7.2.4 Small for Gestational Age (SGA)/Russell Silver Syndrome (RSS)**

Growth hormone treatment is proven to optimise growth and improve short stature in children who are small for their gestational age SGA (NICE 2010). These children are born with weight and/or length at least 2 SDS below the mean of their gestational age and have not shown any evidence of growth catch-up two to four years after they are born (Houk & Lee 2012). Licensed indication by the NICE (2010) is for growth disturbance (current height [SDS]  $-2.5$  and parental adjusted height SDS  $-1$ ) in short children born SGA, with a birth weight and/or length below  $-2$  SD, who unsuccessfully catch-up in growth (height velocity SDS less than 0 during the past year) by the age of 4 or later. Nearly 10% of children born SGA do not get to the normal height range (Houk & Lee 2012). Those whose growth has not caught up by 4 years of age will need GHT (NICE 2010). Children born SGA are a heterogeneous group, including normal children, and children with growth delay due to maternal,

placental and foetal factors. Approximately 80% of children born SGA show catch-up growth in the first 6 months of life, which ends by 2 years of age in most, though this may be postponed until up to 4 years of age in some (Kirk 2012). 10 - 15% of SGA children who fail to catch-up spontaneously are linked to genetic conditions such as Russell Silver syndrome and chromosomal disorders (Ong *et al.* 2005). In addition, the Food and Drug Administration in 2001, as well as the European Agency for the Evaluation of Medicinal Products in 2003, decided to approve GHT for short SGA children, including children with RSS who show no catch-up growth by 2-4 years of age, regardless of the GH secretor (Johnston 2008).

#### **2.7.2.5 Chronic Renal Insufficiency (CRI)**

Growth failure associated with CRI typically manifests when the glomerular filtration rate falls to 50% of the normal rate (NICE 2010). In the UK, renal registry specifies that 29% of children who endure renal transplantation and 41% of children on dialysis are under the 2nd percentile for height within their first year and continue so throughout childhood due to a more marked slowing in height velocity (NICE 2010). It is also suggested by NICE (2010) that GHT is suggested for prepubertal children with CRI, if nutritional status and metabolic abnormalities have been improved, and steroid therapy has been reduced to a minimum. Growth hormone treatment should be stopped after a renal transplantation, and only restored after 1 year if it has been determined that catch-up growth has not happened (Kirk 2012).

#### **2.7.2.6 Short Stature Homeobox-Containing Gene (SHOX)**

The short stature homeobox-containing gene (SHOX) is situated on the distal ends of X and Y-chromosomes and works on long bone growth (NICE 2010). Normal growth needs two functional copies of the gene; therefore, growth impairment can arise if one copy of the short stature homeobox-containing gene has been incapacitated by mutation or deleted (haploinsufficiency) (NICE 2010). SHOX deficiency results in short stature in people with conditions such as Turner syndrome, Leri-Weil syndrome and dyschondrosteosis (Kirk 2012). Grounded on a small study (26 people with SHOX haploinsufficiency compared with 45 of their unaffected relations), children with SHOX haploinsufficiency were 3.8 cm shorter (-2.1 SD) than



their unaffected relatives and this variance stayed throughout their childhood (NICE 2010). It has been proven that GHT positively effects these children's growth velocity nearly in the same way as shown to be with TS (Kirk 2012).

### **2.7.3 Prescribing of GHT and National Guidelines**

It is recommended that treatment should be introduced and monitored by a paediatrician with special expertise in the management of children with growth disorders (NICE 2010). Growth hormone is mainly prescribed in the UK on a shared care basis between primary and secondary/tertiary care with the general practitioner taking responsibility for prescribing, and the lead centre taking responsibility for initial diagnosis, monitoring and adjustment therapy (Kirk 2012). They usually follow the national shared care guidelines for the use of paediatric GH, prepared by the British Society for Paediatric Endocrinology and Diabetes (BSPED 2015). In Ireland, complex and rare conditions are commonly managed in specialised paediatric endocrinology services at tertiary centres or on a shared care basis with a local GP who has specialised training in paediatric endocrinology (HSE 2018). No national guidelines have been put in place for prescribing GHT in Ireland. Irish healthcare providers prescribing GHT follow the same guidelines used in the UK, which is endorsed by the National Institute of Health and Clinical Excellence (NICE) who have reviewed the use of GHT in childhood twice, first being in 2002 and most recent one in 2010. The latter is due for a review in 2020 (NICE 2010).

The license indications in the NICE (2010) GH in childhood guidelines are as follows:

- Growth disturbance in children due to insufficient secretion of growth hormone (Growth hormone deficiency).
- Growth failure in girls linked to gonadal dysgenesis (Turner syndrome).
- Growth retardation in prepubertal children associated with chronic renal insufficiency (CRI).
- Enhancement of growth and body composition in children with Prader–Willi syndrome (PWS). The diagnosis of PWS should be established by suitable genetic testing.
- Growth disturbance (current height standard deviation score [SDS]  $-2.5$  and

parental Adjusted height SDS  $-1$ ) in short children born small for gestational age, with a birth weight and/or length below  $-2$  SD, who failed to show catch-up growth (height velocity SDS less than 0 during the past year) by 4 years of age or later.

- Growth retardation associated with SHOX deficiency, as set by DNA analysis.

#### **2.7.4 Growth Hormone Treatment Side Effects**

The British Society for Paediatric Endocrinology and Diabetes (BSPED) states that treatment should only be started in specialist centres that habitually take part in a national audit of their clinical activities, any possible side effects, and benefits of the therapy should be discussed comprehensively with the parents and child before commencing treatment (BSPED 2015). Growth hormone treatment in childhood is mostly considered safe, with serious adverse events rarely reported especially with currently approved doses (Sävendahl *et al.* 2012). However, evidence of specific conditions such as benign intracranial hypertension, scoliosis and slipped capital femoral epiphysis have been noted shortly after commencing GHT in a small sample of treated patients (Darendeliler *et al.* 2007). Apprehensions have also been raised about the likelihood of long-term alterations in glucose metabolism and connection with the incidence of neoplasms, but there is little support for excluding patients with pre-existing risk factors (Ong *et al.* 2005, Cappa *et al.* 2016). NICE (2010) also advise paediatricians to pay particular attention when prescribing GHT to children with diabetes mellitus or its risk factors, slipped capital epiphyses, idiopathic intracranial hypertension or malignancies.

Sustained surveillance studies on the efficacy and safety of GHT in children remain important, and long-term outcomes and safety have been reported in large international databases of GH-treated patients. These include the Genetics and Neuroendocrinology of Short Stature International Study (GeNeSIS) sponsored by Eli Lilly and Company, the Kabi International Growth Study (KIGS) sponsored by Pfizer, the National Cooperative Growth Study (NCGS) sponsored by Genentech and the NordiNet®International Outcome Study (IOS) sponsored by Novo Nordisk. These multinational studies deliver global information, giving a summary of the impact of GHT in children (Cappa *et al.* 2016).

### **2.7.5 Discontinuation of GHT**

The recommendations to stop are: growth velocity increases less than 50% from baseline in the first year of treatment; final height is approached and growth velocity is less than 2 cm total growth in one year; there are insurmountable problems with adherence or final height is attained (NICE 2010, Kirk 2012).

### **2.7.6 Statistics**

In the Netherlands, around 2,000 children receive daily GHT (Van Dongen & Kaptein 2012), while in the UK in the year 2006 there were 4,712 children on GHT and in the year 2009, 760 new patients commenced on GHT (Langham & Kirk 2011). However, in Ireland there is no registry of the number of children on GHT (Hawkes & O'Connell 2016). So, after extensively searching and asking relevant professionals in the country for a statistical count, I was able to get an estimate of 1,000 children in Ireland on GHT from one of the five GH manufacturers in the Republic of Ireland (ROI) back in 2015.

## **2.8 Parental Reaction to Child Growth**

When a child does not grow and fulfil the expectations of their families, then there are consequences for the child and family (Thomlinson 2002). Parental anticipation and anxiety will often occur with growth issues and needs to be addressed while dealing with worries about height potential (Woolley 2011). Parents are also more likely to worry about short stature than tall stature when it comes to their child's growth (HSE 2012). In most cases of parental concerns, it is the mothers' concern about the child's height that is usually the main reason why children are brought to clinics for investigation (Naiki *et al.* 2013). In Canada, Thomlinson (2002) examined the lived experience of families (n=12) of children who were failing to thrive. Twelve families agreed to participate, consisting of 11 mothers, one stepmother, six fathers, and three grandmothers. Participants' stories were collected through in-depth audio-recorded interviews. They were asked to think and talk about their everyday experience as a

family as broadly as they wished. The findings showed that parents (both mothers and fathers) saw their capabilities as parents being questioned. The physical growth restriction that manifested in their children, regardless of the cause, severely affected family members. They were distressed by the comparisons of the children made by others and themselves. They also faced difficulties in the process of seeking care for their children, as the families expressed how their concerns often were not heard by the professionals. They also felt responsible for their children's growth failure and this added to a feeling of isolation and helplessness. They appreciated nurses and doctors who listened and acknowledged them as valuable members of the care team. Parents invested considerable effort, energy, time and financial resources into finding a diagnosis and treatment for the underlying condition that was causing the lack of growth. These parents efforts were similar to the efforts employed by families of children with cancer or other chronic life-threatening illnesses which were discovered in Cohen's (1995) study which explored the triggers for heightened parental uncertainty in chronic, life-threatening childhood illness.

Daniel *et al.* (2005) in the UK investigated families (n=9 mothers) decision-making for their child to undergo an elective surgery as a treatment for short stature due to Achondroplasia. It was noted in this study that the decision process was based on the perception that the child would be functionally, socially and potentially psychologically disadvantaged in the future because of their short stature. Mothers, their children and other important family members involved in the decision process were confronted with a dilemma because the treatment proposed long-term benefits but short-term hardships and suffering. It appeared that mothers were trying to do what would be best for their children to improve their final height. In the USA, Grimberg *et al.* (2015) used surveys to examine parental concerns about child growth and factors that drove their decisions to medically intervene or not with their child's height. By collecting surveys from parents (n=1820) of children aged (9-14), they identified 22 distinct concerns expressed by parents that were grouped into seven categories: treatment characteristics, health, psychosocial functioning, physical appearance, adult success, comparison and cultural/demographic features. The categories rated as having the greatest influence on parents' decision to seek GHT were: GHT efficacy and side effects; child health; and psychosocial function as a child and as an adult.

## 2.9 Parents and Administration of Injections

Some degree of needle anxiety and phobia affects about 22% of the general population, nevertheless it has been noted as a neglected diagnosis (Wright *et al.* 2009). Multiple members of the same family may share needle phobia. When they experience needle phobia, they may have a vasovagal response manifested by hypotension that causes sweating, nausea, dizziness, and fainting (Howe *et al.* 2011). Children and parents with needle anxiety can find regimens involving daily injections intimidating, severely impacting their ability to self-manage the condition and attain glycaemic control in the case of diabetes; they may be unwilling to test blood glucose, to increase the number of insulin injections, and to attend diabetes visits often (Deacon & Abramowitz 2006). It could be concluded that similarly with GHT regimen, parents and children with needle phobia may be reluctant to administer a daily injection, comply with the regimen and attend endocrine follow-ups regularly which may involve blood testing.

In addition, when parents perform complex clinical procedures on their children at home such as administering injections, they may feel a sense of dislike towards this 'nursing role' that may overshadow their parental role. Parents like to define themselves as parents and not nurses and have a normal parent-child relationship (Kirk *et al.* 2005). Acting as the child protector is one of many roles parents hold (Bowlby 2005). That sense of protection that the parent holds may be lost when their child has to undergo invasive medical procedures (Bernard & Cohen 2006, Karlsson *et al.* 2014). However, parents' attendance is essential during a Needle Related Medical Procedure (NRMP), according to children and by using supportive measures during NRMP, parents can help reduce pain and distress (Salmela *et al.* 2011).

Notwithstanding the parents' own feelings, they have a key role in supporting their child through these invasive procedures. They may experience different emotions that could affect their capacity to safeguard their child (Karlsson *et al.* 2014). They may mask their own feelings of anxiety and worry and put them aside so the child's needs can be put first during invasive medical procedures, like injections, to maintain control over the situation. It should be noted that the negative feelings that parents feel during these procedures, if left unsupported, might affect their supportive capacity, which will

directly affect the treated child (Karlsson *et al.* 2014).

Since there are limited studies that explore the perspectives of parents around managing GHT injection administration, other studies that investigate parents' use of injections for other medical conditions that require children to receive regular injections were used in this review. Many studies have examined the perspectives of parents around the management of their children with Type 1 diabetes and switching from insulin injection to insulin pump. Howe *et al.* (2011) looked at the prevalence of fear, distress, pain, and level of cooperation with insulin injections and blood glucose finger sticks in children with newly diagnosed diabetes and their mothers. In this study, they found that a high percentage of mothers reported high fear and distress with needles at the time of diagnosis. Although most improve in time, 13.6% of the 23 mothers in the study continued to report high fear and distress six to nine months after diagnosis and their continued report of high distress correlated with the poor cooperation of children, which correlated with poorer diabetes control. A study by Sullivan-Bolyai *et al.* (2004) examined parents' experiences of caring for a child aged <12 years using insulin pump therapy (for 3–36 months) by indepth interviews with 21 mothers and fathers of 16 children. They found that parents who willingly accepted the pump therapy were motivated by the avoidance of the painful injections (insulin injections). Another qualitative study explored parents' experiences of managing their child's diabetes using an insulin pump (Rankin *et al.* 2015). They conducted indepth interviews with 19 parents of children (aged  $\leq 12$  years) with Type 1 diabetes who used an insulin pump and found that the most significant benefit for parents from switching to a pump was the avoidance of the painful injections (Rankin *et al.* 2015). These studies illustrate the preference of parents to avoid directly injecting their children, which indicates the emotional struggle associated with the administration of injections for parents. This phenomenon is also seen in other medical conditions that require children to receive regular injections or as needed such as in the case of using EpiPen during an allergic reaction. Kim *et al.* (2005) explored whether underuse of EpiPen might be attributed to parental discomfort with injection administration using surveys. It was reported that parents (n=75) who used EpiPen for their children with food allergies claimed that they would be uncomfortable administering the EpiPen to their child and 40% of parents noted that the reason for

the uneasiness was because they feared hurting their child (Kim *et al.* 2005).

A study carried out by Rosenfeld and Bakker (2008) tried to identify key factors that influence adherence and persistence in patients receiving GHT and to promote the development of interventions to support continuous GHT use. They found after surveying patients on GHT along with 398 parents, that agreement with the statement, 'Injection is painful' was predictive of non-adherence for parents of children on GHT, but not for paediatric or adult patients on GHT (Rosenfeld & Bakker 2008). Likewise, in Kremidas *et al.*'s (2013) study, 47.7% of caregivers (n=235) were more likely to agree that injections are painful. These findings also support the emotional turmoil that parents may encounter when they have to administer daily injections for GHT.

## **2.10 Parents and GHT Devices**

Growth hormone treatment is administered by daily subcutaneous injections and may be given via administration devices that are needle or non-needle. The literature search revealed some studies that examined patient and caregivers' preference with regards to GHT devices. A study by Dumas *et al.* (2006) aimed to identify which attributes of GHT administration devices were most significant to doctors, nurses, teen patients, and parents of young children receiving GHT. Using a survey that was distributed to parents (n=18) from France, Germany, Italy, the UK and the USA. They were asked to rate 19 device attributes, and rank four GH administration devices in order of preference. Reliability, ease of use, lack of pain, safety, storage and number of steps before, during and after administering the GH were reported to be the most favourable five attributes of a GH administration device. Parents in the UK and Italy ranked the lack of pain as the most central attribute. In addition, when they were asked about device preference, an electronic device was favoured by 71% of the parents over automatic injection devices or prefilled syringes (Dumas *et al.* 2006). It was suggested that the ideal device design has features that can lower both the real pain (e.g. finer needles and adjustable injection speeds) and the perceived pain (e.g. needle-free or hidden-needle options and noiseless operation) felt by the patient. Minimal pain was also alongside reliability and ease of use (Dumas *et al.* 2006).

The parents need for GHT devices that are easy to use also resonated in a Swiss study that examined parents' preference for easy to use growth hormone injection devices quantified by the willingness to pay and found that they were willing to pay for device features facilitating ease of use (Meinhardt *et al.* 2014).

However, with the number of the devices available that have different features to choose from, parents may not be able to choose or select their preferred device that features their favoured attributes which could lead to lack of adherence (Kapoor *et al.* 2008). In the Netherlands, (Van Dongen & Kaptein 2012) study on parents' views on GHT for their children identified a need for support and participation of parents in the practice of selecting a GH device. 48% of the 69 parents involved in the study reported a lack of freedom to choose the type of GH device that best suited their needs.

## **2.11 Treatment Burden and Adherence**

Daily subcutaneous injections can be an overwhelming experience for many parents. Andrews (2000) who is a mother of a child prescribed GHT stated that:

*'The Growth Hormone replacement was another story. Injections of Growth Hormone would need to be given several times a week. All I could think of were the trips we would need to make for these injections. Then I was told that I would be trained to administer them. There was no way I was going to stick my child with a needle! I was a mother, not a doctor or a nurse. I would not and could not do it; at least that was what I thought. Of course, I had to do it. My husband, who is a great father and supporter of his children, would not even consider learning to give injections. It would be my complete responsibility. I truly understood his unwillingness to stick his child. Why should both of us suffer?'* (Andrews 2000, p. 1)

Daily subcutaneous injections may seem like a burden because they may cause pain and bruise the child. Health care professionals carefully monitor the effect of therapy, and the dosage may be regulated every 3–6 months. However, these hospital visits can be stressful for both children and parents because X-rays and blood tests may be necessary (Van Dongen & Kaptein 2012). In addition, GHT is associated with poor adherence as frequently noted in the literature, as skipping of injections is not



followed by immediate negative effects (Van Dongen & Kaptein 2012). A study undertaken in the UK assessed 75 children's concordance to GHT and reported that 23% of the children in the study missed more than two doses a week (Kapoor *et al.* 2008). It was also noted that failure to comply with the financially and physically costly GHT in GHD may be as high as 50% of all cases (Ahmed *et al.* 2007). Non-adherence can lead to more repeat visits to the hospital for more diagnostic tests and unnecessary changes to the treatment course that may lead to worse overall outcomes and increased health care costs (Fisher & Acerini 2013).

Numerous studies have evaluated factors that influence GHT adherence for both adult and child patients. Some studies have investigated the impact of the device used on adherence and attributes of comfort, ease of use and choice (Desrosiers *et al.* 2005, Wickramasuriya *et al.* 2006, Kapoor *et al.* 2008, Rosenfeld & Bakker 2008, Fuchs *et al.* 2009, Hey-Hadavi *et al.* 2010, Bozzola *et al.* 2011, Rubin *et al.* 2011, Kaptein 2013, Tauber *et al.* 2013, Meinhardt *et al.* 2014, Spoudeas *et al.* 2014). Other studies looked at motivation, acceptance and satisfaction with therapy, family surrounding, educational level, training and administration technique for GHT, patient-doctor relationship and level of understanding about the disorder and therapy (Smith *et al.* 1993, Sigüero *et al.* 1995, Haverkamp *et al.* 2008, Kapoor *et al.* 2008, Rosenfeld & Bakker 2008, Norgren 2009, Pfützner *et al.* 2010, Cutfield *et al.* 2011, Kappelgaard & Laursen 2011, Van Dongen & Kaptein 2012).

Despite the availability and safety of GHT to treat children, and the current developments in product features, such as pen delivery, hidden needles, needle-free devices, auto-injectors and electronic injectors; clinical outcomes remain suboptimal (Rosenfeld & Bakker 2008, Kirk 2009) and patients and caregivers may experience a significant burden related to injection practices (Kremidas *et al.* 2013). The reported adherence to GHT is variable since as many as 36%–49% don't take it as prescribed, and this is majorly because of the considerable efforts needed in treatment management (Haverkamp *et al.* 2008). Younger children are more adherent to GHT than adolescents and adults (Rosenfeld & Bakker 2008). Younger children may need more help from their parents or carers to inject (Kaptein 2013) and poor adherence in children may reflect a lack of understanding of their treatment by their parents (Rohrer *et al.* 2017).

It was noted by Van Dongen & Kaptein (2012) that the patients' perceptions of GHT benefit play a role in the patient acceptance of GHT. However, the key to better understanding of patient acceptance of GHT is to understand parental perceptions and beliefs regarding illness and treatment, because parents are in control of the first years of treatment and beyond. Few studies have explicitly examined the opinions of parents and parents' views regarding GHT for their child (Van Dongen & Kaptein 2012). The involvement of the parents may be a leading factor in adherence as younger children or children with learning difficulties may be unaware of the need for treatment and can be unwilling to receive the injection (Laing 2014). However, not many studies have looked at treatment burden from the perspectives of parents when it comes to GHT. Only four studies assessed GHT burden from the perspective of the parent (Van Dongen & Kaptein 2012, Kremidas *et al.* 2013, Marini *et al.* 2016, Brod *et al.* 2017).

In the Netherlands, Van Dongen & Kaptein (2012) evaluated parents' (n= 69) opinions, beliefs and perceptions about GHT and level of communication and support received from healthcare professionals via an online survey. They aimed to gain a better understanding of parents' perceptions and views on self-management of GHT. The survey addressed acceptance of the diagnosis and treatment and included questions on the quality of information, involvement in the decision-making process, operational aspects, emotional problems and support. It was found that of the parents surveyed, 48% reported a lack of choice regarding the GHT device that best suited their needs, 92% believed that they and their children would benefit if the children self-administered injections, and 65% noted that training to support self-administration would be useful. According to 79%, the accessibility and availability of support from another parent with experience of caring for their own child with GHT, together with their doctor, would be valuable. 37% of the parents specified that their children felt anxious about injections, and 83% of parents would welcome psychological support to overcome their anxiety. An increase in unwillingness to receive GHT by the adolescents were witnessed by 40% of the parents and 57% of these parents highly appreciated psychological support to overcome their adolescents' reluctance to receive GHT (Van Dongen & Kaptein 2012). The study participants were recruited from the

Dutch Society of Growth Hormone Treatment (NVGG) and were promised a €10 charitable donation to the NVGG society as an incentive. Therefore, the participants might have been more motivated than others to participate which suggests that the sample may not be representative of the global patient population.

In the USA, Kremidas *et al.* (2013) assessed administration burden associated with GHT from the perspectives of patients (n=61) (13 or older with GH disorder) and caregivers (n=239) (almost all parents and female who responded were caring for a child of an average age of 9.5 years) using a webpage survey. This study aimed to develop an understanding of the GHD treated patient and caregiver experiences and attitudes towards GHT administration practices including storage requirements, administration challenges, and reasons for non-adherence. After descriptively analysing the variables and using statistical measures, the authors reported that more than a third of caregivers experienced the burden of storage at home or while not traveling (36.0%) while the burden of storage worsened when traveling (51.0%). Caregivers wanted or preferred more flexibility with storage requirements and wished to be able to store GHT outside the fridge for longer periods of time (69.9%). In addition to storage requirements being a burden, 44.2% of the quarter of the caregivers who had to reconstitute GHT before use, found this inconvenient and burdensome. Also, more than half of caregivers agreed that injections caused bruising and stinging and 47.7% acknowledged that GHT injections are painful. It was also confirmed that caregivers know the significance of adherence, as nearly two-thirds of caregivers opposed the statement, 'Missing a couple of GH injections per month is not a big deal'. However, most of the same participants stated that normally they would miss at least one, and sometimes up to three, doses per month. The results imply that awareness of the significance of adherence may only be one of the numerous factors associated with adherence. The involved relationship between the parent and child receiving GHT may also add to adherence, both including children who are self-injecting and those who are not. Though assessment of adherence was not the focus of the study, the discordance shown in reactions concerning the importance of not missing doses and the account of missed doses necessitates further research. Nonetheless, it is important to note that the caregivers in the Kremidas *et al.* (2013) study were affiliated with the Major Aspects of Growth in Children (MAGIC)

Foundation which are a global patient advocacy organisation in the area of human growth disorders (USA) and may have been more motivated to participate in the study. In addition, the participants had to be U.S. residents and therefore, this sample may not reflect the global patient population. Furthermore, the study covered aspects of care associated with administration practices but did not investigate the emotional, physical and psychosocial burden that may affect parents. Finally, the data were collected and analysed using quantitative methods which limits a holistic understanding of the phenomena.

An Italian study by Marini *et al.* (2016) used a narrative-based approach to collect testimonies from patients with GHD (n=67) (8-17 years old), their parents (n=72), siblings (n=7), and providers of care (n=19) collected from eleven Italian centres. They aimed to explore their experience with GHD by understanding their perceptions regarding the management of GHT and identifying the impact the condition had on their family, social and school life. After the narratives were analysed using a qualitative analysis, three clusters were identified: 'Disease-centred' narratives: focused on the clinical point of view; 'Illness-centred' narratives: focused on living with a particular condition; and 'Between disease and illness-centred' narratives: containing both technical and emotional elements. Results from each target groups (patient, parents, siblings and health provider) were compared and interpreted together. They found that although care management and outcomes were deemed satisfying in the 72 collected narratives from the parents, their narratives covered topics such as: the first signals of GHD (n=65; 'I remarked the slowing down by myself and looked for a centre of care'), the waiting for the diagnosis (n=48; 'Lived with anxiety and concern'), the communications of the diagnosis (n=48; 'I was already prepared'), the communication of the therapy (n=51; 'I was worried and not convinced'), the difficulties of the therapy today (n=89; 'It is an organizational issue'), the outcomes of the therapy (n=75; 'My son/daughter grows up very well'), the centres and care teams (n=56; 'I totally trust the centre and the team of care'), expressed fear and worries (n=42; 'I am worried about the side effects of the therapy') and style of narration (n=69; Disease-centred: 23 (25%)). From the parents' narratives, 67% of them revealed persistent worry about potential side effects and not being well informed about side effects. The study concluded that the lack of reassurance could signal an alarm bell for decrease adherence. Although Marini *et al.*'s (2016) study aimed to

examine the real world and needs of patients and their parents, a deeper possible understanding of the parents' needs was not fully captured. This is due to the use of semi structured plots to collect narratives as a data collection tool in addition to the study not solely being focused on the parent's lived experiences. Also, the study captured only the experiences of parents of children (aged 9-17) and diagnosed only with GHD which leaves the experiences of parents who have much younger children and/or have other medical indications needing GHT unexplored (Marini, *et al.* 2016).

A more recent study by Brod *et al.* (2017) aimed to explore the GHT burden for children and their parents. The authors interviewed children/adolescents (n=39) aged (8 to <13) years, with GHD and parents of children aged (4 to <13) year old (n=31) from Germany (5 focus groups), United Kingdom (UK) and United States of America (USA) (individual phone interviews). Of the 31 parents who participated, 19 were parents of child respondents who participated in the study. Data were collected using semi-structured interviews that were designed based on the literature and clinical expertise, to elicit the perceived symptoms, burden, and impacts of the GHD as well as the GHD treatment on social, physical and emotional aspects of daily living. Parents were asked to report only actual and not perceived or presumed impact on children. The interview transcripts were analysed thematically and underpinned by modified grounded theory principles. Based on the findings of the analysis, a conceptual model of major and minor treatment burden impacts was developed. The two treatment burden domains identified for parents were emotional well-being and interference. However, the findings of the study were a combination of descriptions presented by both children and parents. Parents self-reported being emotionally affected by GHT for their children. They reported their own worry (n=21, 62%), comprising worry about GHT administration (n=20, 59%), inflicting pain to their child (n=13, 38%), and GHT costs (n=5, 15%). They also noted feeling sadness about their child's need for GHT (n=7, 21%), guilt (n=6, 18%), and frustration with injection administration (n=5, 15%). Parents also self-reported being impacted by interference resulting from GHT. Half of the parents (n=17, 50%) noted that GHT interferes with family travel and travel planning. Around one-third of parents (n=11, 32%) reported the length of time for injection preparation being interfering, and that

GHT affected their daily and social life (n=4, 12%). It was concluded that GHT had significant impacts on both children and their parents and the self-reports revealed the significant impact GHT had on their daily lives. Brod *et al.*'s (2017) study focused on exploring the experience of children who are GHD and their parents and therefore excludes the experiences of parents who have children receiving GHT for other reasons. Also, the study did not focus primarily on the impact of GHT on parents. In addition, the results only reflect experiences of parents who have children aged 4 to 13 years and excluded those who have children either younger or older than the age range in this study. Moreover, participants had to have not more than a year's experience of being on GHT which may not be sufficient time to develop a rich experience of the phenomenon. In addition, the data collection tool used (semi-structured interviews by phone) could have restricted the depth of the experiences conveyed and therefore the extent of understanding of the phenomenon. Nonetheless, a broad description of the extent of burdensome on parents was provided while lacking a detailed description of how GHT was interfering with their travel planes, medication preparation, daily activities and social lives.

## **2.12 Parental Concerns About Growth Hormone Treatment - Allocating the Gap in the Literature**

Other studies have investigated health-related quality of life (HRQL) in relation to children who are short prior to and while on GHT where parents were asked to fill in questionnaires about their insights and opinions (Eiser *et al.* 2005, Sheppard *et al.* 2006, Stheneur *et al.* 2011, Otero *et al.* 2013). The understanding of the child's HRQL was largely based on parents' views on the shortcomings of being short and small and perhaps having an unrealistic expectation about GHT (Otero *et al.* 2013). When the child is short, parents seem to rate their child's HRQL much lower than what the child would rate him/her self (Erling *et al.* 1994). In fact, in the UK, Otero *et al.* (2013) wanted to explore the relationship between the perspectives of the child and parent in regards to rating HRQL before initiating GHT to better understand how parents and children view the consequences of short stature and the potential benefits of GHT. Results indicated that parents rated lower on the physical and psychosocial HRQL than children rated themselves.

Parental concerns and psychosocial functioning of young children born (SGA) treated with GHT was explored (Lagrou *et al.* 2008). Forty parents of children were interviewed and given a questionnaire at the start of GHT and after 2 years of follow up. They found that before starting, 85% of the parents were concerned about short stature, 76% anticipated an increase in adult height of 610 cm and 81% expected a positive impact on well-being. Half of the parents articulated fears regarding GHT. After 2 years, more parents of treated children reported obvious growth and physical changes, and fewer parents noted teasing due to short stature. An improvement of well-being was noted by half of the parents of treated and untreated children and worries about GHT ended almost completely. It was concluded that before the start of GHT, fears about side effects and painfulness of injections were present in half of the parents. Once GHT was commenced, concerns about short stature and worries about GHT diminished.

Parental concern can influence the decision-making process and increase both referrals to specialists and the prescribing of GHT by endocrinologists and affect their acceptance of physician treatment recommendations (Cousounis *et al.* 2013). Grimberg *et al.* (2015) looked at parental concerns about child growth and factors that drive their decisions to medically intervene or not with their child's height. They concluded that GHT efficacy, side effects, child health and psychosocial function as a child and as an adult had the greatest influence on their decision to start GHT. Because parents have such great influence on the decision-making with regard to commencing GHT, Cousounis *et al.* (2013) in the USA sought to investigate how parents learn about GHT and the information that is readily available. In 13 open focus groups with parents (n=71) of children ages 9-14 randomly selected, the majority indicated the internet as the leading source for information. It was concluded that clinicians should provide parents with tools to critically evaluate the content on the internet which can often be incomplete and/or biased and recommended providing a list of pre-reviewed websites or their own material, which could greatly help them make an informed decision.

Most previous research related to the views and perspectives of parents around GHT took a positivistic perspective where scales were used to measure the reactions and perceptions of the parents. For example, studies that measured child health-related quality of life (HRQL) prior to and while on GHT asked parents to complete questionnaires (Eiser *et al.* 2005, Sheppard *et al.* 2006, Stheneur *et al.* 2011, Otero *et al.*

2013). However, other studies looked at the effect of GHT on the behavioural, psychosocial and cognitive characteristics of children on GHT where parents were asked again to answer listed questions (Stabler *et al.* 1998, Ross *et al.* 2004, Van Pareren *et al.* 2005, Lagrou *et al.* 2007, Chaplin *et al.* 2011, Naiki *et al.* 2013). Also, studies that specifically looked at the parents' perspectives around GHT management (Van Dongen & Kaptein 2012, Kremidas *et al.* 2013) also used questionnaires to investigate. Two studies did use a qualitative based approach to collect and analyse data from parents, however, the studies did not primarily focus on parents and results were usually combined and interpreted collectively with the other targeted populations in the study (the patient child/adolescent, siblings and health provider). In addition, they focused purely on the experience from the context of GHD where other GHT indications and conditions were ignored (Marini *et al.* 2016, Brod *et al.* 2017).

Kremidas *et al.* (2013) suggested that a better appreciation of parent/caregiver administration practices of GH and inconvenience accompanying the use of GHT products will help educate health care professionals and potentially improve patient education and training. A better understanding of real-world product administration practices would help nurses recognise areas of need in relation to patient education and training (Bhosle *et al.* 2011). A fundamental part of the role of paediatric endocrine nurse specialist (PENS) is to maintain long-term relationships with families and this relationship is strengthened by recognising and respecting the developing expertise of families as they gain confidence and competence to manage GHT (Collin *et al.* 2016). While results from previous studies (Van Dongen & Kaptein 2012, Kremidas *et al.* 2013, Marini *et al.* 2016, Brod *et al.* 2017) provide valuable information for healthcare providers, they leave the lifeworld of the parent that consists of emotional, psychological, psychosocial, practical and taken-for-granted dimensions largely unexplored. The literature review search revealed a limited number of relevant published studies, therefore, there was scope for this study that explored parents' experiences when delivering care and treatment for their children receiving GHT.

### **2.13 Growth Hormone Treatment Research Involving Ireland**

There was a paucity of research studies into GHT in general in Ireland (n=9 found). The physiological effect of the treatment has been the main focus of GHT research in



Ireland. Two studies, Parvin *et al.* (2004) and Nadeem & Roche (2014) examined the effect of GHT on the height of girls with Turner syndrome (TS) and found that GHT has a positive impact on height in girls with TS. While, two studies by Jensen *et al.* (2013, 2015) were parts of the '(NESGAS) North European Small-for-Gestational-Age Study' which Ireland was part of. They evaluated insulin-like growth factor -1(IGF1) titration of GH dose in contrast to current dosing strategies and explored insulin and glucose metabolism in small for gestational age (SGA) children treated with a fixed GH dose over a period of 1 year. They also examined genetic markers of insulin sensitivity and insulin secretion.

In a very dated study, Wild (1988) conducted a Somatonorm (GHT) post-marketing surveillance study in Ireland 1986-1987 and concluded that GHT was safe. Whitehead *et al.* (1989) explored the Northern Ireland experience of GHT for short stature in adults and found that GHT is safe and efficient but continues to be commenced late in terms both of age and height standard deviation score. Because the study focused on the experiences of adults receiving GHT and not children and more importantly their parents, the study is not relevant to the researched problem.

Kirk & Clayton (2006) who assessed the provision of specialist and transitional paediatric endocrine services in Ireland found that although transition services are established in many larger units, current guidelines are not always adhered to. Furthermore, a national audit by Langham & Kirk (2011) assessed free patient choice for paediatric patients commencing GHT within Ireland. This survey indicated that only one of three units involved agreed with the principle of free patient choice for GHT devices and offered it. Results are worrying, as lack of patient choice of device used is directly related to lack of adherence to GHT (Fisher & Acerini 2013). The most recent national survey on the diagnosis and treatment of paediatric GHD described the current practice in Ireland with regards to children measurement and approach to the diagnosis of GHD (Hawkes & O'Connell 2016). In this study, a questionnaire was sent to 139 paediatricians in Ireland and 35 responded (9 being paediatric endocrinologist). When deciding the GHT preparation to be used, the most vital factor was a fair rotation between all brands (n=5, 50%), and the least important factor specified was the patient choice (n=4, 40%). This resonates with the results of Langham and Kirk's (2011) study conducted

five years later. All respondents signified the need for guidelines for GHT prescribing in Ireland (Hawkes & O'Connell 2016). Criteria noted as essential in the guidelines include patient selection criteria (n=7, 70%), dose adjustment and discontinuation of GHT in failed response (n=8, 80%), restrictions on who should prescribe GHT (n=8, 80%) and limitation to least costly products (n=5, 50%). Preserving the ability of trained endocrinologists to use clinical judgment in grey areas, or to have a committee for such cases was also suggested (Hawkes & O'Connell 2016).

## **2.14 Conclusion**

The literature search concerning children commenced on GHT provided a number of studies which were considered to be of importance. The literature presented a review of the importance of child growth, child growth monitoring in Ireland, GHT history and devices, GHT indications and outcomes, GHT side effects, GHT discontinuation, GHT statistics in Ireland, parent's reaction to child's growth, parents handling injections, parents and the use of GHT devices, parents and GHT burden and adherence, parent's concerns and finally GHT research in Ireland. There were many studies that examined the pharmaceutical aspects of GHT and its physical effects. Studies have also looked at psychosocial effects of the treatment. However, in relation to parents and GHT, studies have generally focused on issues of adherence to GHT and technicality and usability of GHT devices. Only four studies examined parents' perspectives and experiences when delivering GHT care for their child on the GHT regimen. The literature revealed that parents lack; GHT device choice, support, adherence to GHT (regardless of their awareness of the importance of adhering to the treatment) and reassurance about GHT side effects. The literature also highlighted some organisational difficulties associated with GHT including; medication preparation, administration and storage. With the existence of these challenges, it highlights the significance of parents' needs.

There are limited studies that focus specifically on parental experiences of caring for children commenced on GHT. Therefore, there was a need to undertake a study of the lived experiences of parents of children receiving GHT. Moreover, no study was found that explored the lived experience of parents caring for their children receiving GHT regardless of the indication for GHT and from an Irish perspective. The next

chapter will provide an overview of the research methodology chosen for the study and the philosophical approach of phenomenology. The chapter also presents the methods used for data collection and concludes with a review of the study's trustworthiness and rigour.

## **Chapter 3: Methodology and Methods**

### **3.1 Introduction**

This chapter provides a rationale for the choice of methodology for the study. It also provides an overview of the research methodology chosen based on the philosophical underpinnings of phenomenology and shows how this philosophy influenced the methods and approach used for data collection and analysis. The chapter begins by clarifying my worldview based on beliefs, values and attitudes around the nature of reality and truth and how this influenced the decision to choose phenomenology. It then summarises the choice of the appropriate methodology followed by an overview of the philosophical foundation of phenomenology. Further consideration is then given to the theoretical lens used and outlines how they were considered to finally reach Gadamer's philosophical concepts (Gadamer 1975) that underpin the philosophical underpinning of this study. The chapter then describes the research methods used in completing the research study and finally concludes with a review of the study's rigour and trustworthiness.

### **3.2 Worldview**

Before starting this research, the philosophical and methodological underpinnings of both descriptive and interpretive hermeneutic approaches were carefully examined. I needed to self-reflect on my own ontological and epistemological beliefs before deciding which approach to follow in this study journey. I was leaning towards appreciating differences and embracing uniqueness and ambiguity instead of seeking similarities, patterns, and universality in human experiences.

My ontological stance accords what I think reality is. I believe that no single truth exists, as one person's reality may not be the reality of another's. As an interpretive researcher, assuming that reality is, as we can know it, is construed intra subjectively and inter subjectively within the meanings and understandings garnered socially and experientially (Angen 2000, Van manen 2016). Epistemology is the study of knowledge

and how we know what we know (Wellington *et al.* 2005). I strongly believe that knowledge is interpreted and can be generated through experience, co-constructed by common language and dialogue and is inclined by its context and position in time and place. The next section will elaborate on my personal motives behind this study as it is important to maintain authenticity and transparency in line with the philosophical underpinnings of the chosen theoretical lens (Gadamer 1975, Gadamer *et al.* 2004).

### **3.3 Choosing a Research Approach**

As stated before, this research study aimed to gain a deeper understanding of how parents experience caring for their children receiving GHT. Research methodology generally consists of either qualitative or quantitative, or sometimes a combination of both, but this study was proposed to create data about experience, and so, was more matched to qualitative research methods 'the perceived paradigm', rather than quantitative methods 'the received paradigm'. In addition, a qualitative research method was selected because earlier research in this area was limited, and qualitative research is ideal for constructing deep understanding of human experience.

It may be argued that experience and understanding are not measurable objects that can lend themselves easily to measurements and thus methods that emphasis making rich data, are more fitting to the focus of this research. So, a qualitative research methodology was chosen for this study. Qualitative research can be in the form of grounded theory, ethnography, case study or phenomenology, but the choice relies on the researcher's claims on knowledge (Cresswell 2003). In order to focus on the individuals' own meaning of their experiences with the belief that individuals make sense of their world based on their own perspectives and that this is influenced by time and context, a methodology based on realist ontology would have not suited this study. The aim was to understand what it means to be a parent of a child receiving GHT, and how parents experience caring for a child receiving GHT. Because seeking to generate theory was not intended, grounded theory was not suitable. Ethnography was also eliminated, as resources to observe and/or interact with study participants in their real-life environment and to be fully immersed in their day-to-day lives - in their homes where usually GHT injection is administered at bed time - were not feasible

(Sangasubana 2011). This study sought deep information and insights from the parents' perspectives and a full understanding of their experiences, therefore, a phenomenological approach (interpretive) was the most suitable approach to follow.

### **3.4 Influences on the Choice of Research Approach**

The purpose of the phenomenological approach is to clarify the details, to recognise phenomena through how they are alleged by those who are living in a situation. The choice to study a topic in a specific way involves a philosophical choice (Shih 1998). When a researcher wants to use phenomenology, he/she needs to check their personal beliefs, attitudes and values about the nature of reality and truth. The notion that knowledge is gained through understanding and cannot be captured through empirical analytical sciences but reached by sharing a common meaning, was adopted (Van Manen 1977). Through phenomenology, parents' experiences could be illuminated to understand the context of their experiences and how they are described. Through parents' used language and collected text, the power of disclosing the world as they live it may be achieved (Van der Zalm & Bergum 2000).

### **3.5 Philosophical Foundation of Phenomenology**

Phenomenology is an umbrella term that covers both a philosophical movement and a variety of research approaches (Kafle 2013). The philosophical beginnings of phenomenology can be found in the ancient Plato' allegory of the cave (Smith 2013). This allegory offered the notion that what people comprehend to be reality is only a shadow of the phenomena of the true reality and that these two things are distinctly individual (Converse 2012). The philosophy behind phenomenology is to return and explore the reality of life and living referred as the study of the 'life world' or 'lived experience' and explores the experience 'pre reflectively' (Dowling 2007). When applied to research, phenomenology studies the nature and the meanings within that experience. It focuses on the way things seem to us through experience, or in our consciousness and the phenomenological researcher tries to provide rich textual description of the lived experience (Finlay 2009).

The word phenomenon originates from the Greek word 'phaenesthai' which means to flare up, to show itself, or to appear (Moustakas 1994). Philosophers used the phrase phenomenology in the 18th century; such as Kant and later by Hegel, who made noticeable use of the term when it appeared in the title of his 1807 work 'Phenomenology of Spirit' (Dowling 2007). The concept of understanding human experiences, which is the basis of a phenomenological approach to research, was first introduced by the German philosopher Husserl in the early 1900s (Pringle *et al.* 2011). Phenomenology is said to have started at the start of the twentieth century with the work of Husserl & Lauer (1965) although Moran (2000) determined that Husserl's work was inspired by and grounded on the earlier descriptive psychology of Franz Brentano (Moran 2000). Franz Brentano extended the concept of phenomena to incorporate thought (Moran 2000). His 'principle of intentionality' indicated that 'every mental act is related to some object and implies that all perceptions have meaning' which was the central concept in the phenomenology of his student, Husserl, who is referred to as the 'father' of phenomenology (Dowling 2007).

Husserl's phenomenological enquiry aimed to understand the human thought and experience by following an unbiased approach to studying things as they appear (Dowling 2007). By bracketing the researchers' perceptions and presumptions of a particular phenomenon in order to experience the true essence of the phenomena which lies in the object, made his approach unique from the naturalistic science (Converse 2012). Husserl's methodology is often called 'description phenomenology' because it focuses on coming to know through experience with the goal of describing that experience (Dowling 2007).

Husserl's student, Heidegger, rejected this subject-object duality and developed this view into a more interpretive form along with Gadamer (Hekman 1983). Heideggerian phenomenology is hermeneutic and focuses on how to use this preconception to interpret meaning of a phenomenon (Mak & Elwyn 2003). Heidegger and Husserl influenced Hans-Georg Gadamer's work. He advanced Heidegger's earlier work, having been a student of his at Marburg and Freiburg in the 1920s, where he studied practical application. Gadamer saw hermeneutics not as developing a way of understanding, but a means to illuminate the conditions in which understanding, perception, experience and knowing itself takes place (Gadamer 1975). In alignment

with Heidegger's view that language and understanding are indivisible structural aspects of human 'being-in-the world,' Gadamer indicated that language is the common medium in which understanding occurs and that understanding follows interpreting (Lavery 2008). Gadamer considered interpretation as a fusion of horizons, a dialectical interaction between the beliefs of the interpreter and the meaning of the text (Lavery 2008). A 'horizon' is a series of vision that incorporates everything seen from a specific vantage point. A person with no horizon, in Gadamer's opinion, does not see far enough and overestimates what is nearest, while to have a horizon means being able to see past what is familiar. This is where questioning is essential, as it helps the person create new horizons and understandings which is a critical aspect of the interpretive process (Lavery 2008). Gadamer emphasised that we are all part of history and consciousness and he recognised the merging of individual's horizon within the prejudices of history, including those offered by people and text in the development of knowledge and understating.

Rodgers (2005) remarked that Gadamer foresaw the hermeneutic circle as a process of movement between the aspects of the text and the interpreter of the text. In this movement; the preconceptions of the interpreter are fused into the process and transferred from preconceptions to new understandings as the interpretive process progresses onwards. Annells (1996) adds that Gadamer believed that understanding and interpretation are compelled together, and interpretation is always a growing process, thus a conclusive interpretation is likely never possible. Finally, following in Heidegger's tracks, Gadamer's ontological view was that researchers are deeply inclined by the traditions of their culture and therefore act on that way of being in the world, thus understanding is a way of being in the world rather than as a way of knowing about the world (Converse 2012). For his epistemological view, knowledge originates through interpretation, which is a way of being, rather than a way of knowing (Converse 2012).

### **3.6 Theoretical Lens**

#### **3.6.1 Choosing Phenomenology**

The concept of understanding human experiences, which is the basis of a phenomenological approach to research, was first introduced by Husserl in the early



1900s (Pringle *et al.* 2011). Husserl's phenomenological enquiry aims to understand the human thought and experience by following an unbiased approach (bracketing) to studying things as they appear (Dowling 2007). He claimed that only through eliminating one's own bias could one possibly look at consciousness in an objective way so he advocated bracketing as a central step whilst exploring phenomenon. Thus, bracketing the researchers' perceptions and presumptions of a particular phenomenon is essential to experience the true essence of the phenomena which lies in the object (Converse 2012). Husserl employed the reduction process and consequently this led me to explore the works of Heidegger as I found bracketing unrealistic because of my personal situation.

Heidegger rejected this subject-object duality and developed a more interpretive form (Hekman 1983). He deems the person exists as 'being-in-the-world' and that the persons' historical and traditional backgrounds are already incorporated into their experience to become part of their reality, without parting of subject-object (Hekman 1983). Heidegger's strong connections to Gadamer as his teacher then led me to study Gadamer's works. Gadamer sees understanding as a state of being human and that understanding is interpretation (Schwandt 2000). Schwandt (2000) indicates that socio-historically bias or prejudice should not be contemplated as a feature that an interpreter must control to get a clear understanding.

### **3.6.2 Choosing Hermeneutics**

Hermeneutics is the art of understanding and the theory of interpretation. It focuses on the human experience as it is lived. It reveals details and apparently marginal aspects within an experience that may be taken for granted, with a purpose of generating meaning and attaining a sense of understanding (Kafle 2013). It is a research methodology designed to produce rich textual descriptions of the experiencing of a phenomena where a deeper understanding of the meaning are sought, through progressively layered reflection while using rich descriptive language (Kafle 2013). The interpretivist approach seeks culturally derived and historically driven interpretations of the social life world, therefore, an interpretivist can be seen as someone who tries to clarify the meaning of a phenomenon (Crotty 1996). If it were intended to take this

approach, it is deemed helpful in explaining the experience and understanding of being a parent of a child receiving GHT. It does not consider understanding as a way of knowing but as a form of being and as a result of this ontological importance, the epistemological stance is not the main feature of this philosophy (Converse 2012). This felt more ontologically comfortable and led to my decision to use hermeneutics for this study.

### **3.6.3 Choosing Gadamer's Hermeneutics**

Gadamer's version of hermeneutics is that, firstly, our own preconceptions are fragments of our individual linguistic experience which makes understanding likely, and secondly, people who express themselves, and those that understand them are connected by human consciousness - 'universality' a 'fusion of horizons' - which makes understanding conceivable (Morse 1994). He viewed hermeneutics not as developing a way of understanding, but a means to illuminate the conditions in which understanding, perception, experience and knowing itself takes place (Gadamer 1975). Gadamer's ontological view was that researchers are deeply inclined by the traditions of their culture and therefore act on that way of being in the world (Converse 2012). He emphasises that understanding is a state of 'being-in-the-world' and that tradition plays a significant role in interpretation. Therefore, as a nurse and a mother of a child on GHT, being more empathetic with participants and having a clearer understanding of their experience due to the commonalities that may arise while looking for contradictions, may be reachable.

Gadamer believed that there is always more than one worldview for us to respect and in the same time we should be true to our own perspectives and experiences (Gadamer 1975). With his major concept of pre-understanding, one may have the advantage of acknowledging and using their pre-understandings to understand and interpret more deeply. I could empathise more with mothers, having a clearer understanding of their experience because of the commonalities that may emerge. I also share the historicity and language of mothers with children on GHT that would facilitate the interpretation, analysis and co-construction of our findings.

The fusion of horizons and the hermeneutic circle consequently play a vital role in hermeneutic philosophy, and Gadamer considers interpretation as a fusion of horizons, a dialectical interaction between the pre-understandings of the interpreter and the meaning of the text (Polkinghorne 1983). This concept will not only assist in acquiring the commonalities between each story told, but also the contradictions that may arise as they also need to be highlighted. This would broaden my horizon, as I would enter this study with my mind not set on only what I have experienced with having a child on GHT but be open to other experiences and even be surprised by the outcomes. Gadamer foresaw the hermeneutic circle as a process of movement between the aspects of the text and the interpreter of the text, in this movement; the preconceptions of the interpreter are fused into the process and transferred from preconceptions to new understandings as the interpretive process progresses onwards (Rodgers 2005). This is done by understanding parts of the text and relating them to the whole (context) and vice versa initiating a dialectical analysis of parts of the data to understand better the whole (Cohen *et al.* 2000). Gadamer believed that this process of coming to an understanding is a vital component in Gadamer's hermeneutics, and this plays a vital role in data analysis.

Gadamer implied that to understand is to understand differently and that a fusion of horizons does not mean reaching consensus (Gadamer 1975). He also claims that it is essential to be surprised by your research. Besides, like Gadamer, it is thought that perspectives are dependent on historicity and context. In addition, understanding and interpretation are compelled together, and interpretation is always a growing process; thus, a conclusive interpretation is likely never possible (Annells 1996). Finally, Gadamerian hermeneutics has proven to be well established and utilised in numerous nursing research studies (Walters 1995, Ford & Turner 2001, Fleming *et al.* 2003, Geanellos 2005, Ajjawi & Higgs 2007, Grassley & Nelms 2008, Fex *et al.* 2011, McCloud *et al.* 2012). In summary, hermeneutic phenomenology underpinned and informed by Gadamer's hermeneutics fits the study's aim. It provides a framework for exploring the experiences of parents' and getting a deeper understanding of the phenomena, which is currently unknown. The next section of this chapter describes the research method used to conduct the study with reference to Gadamer's philosophical hermeneutic principles (1975).

### **3.7 Research Method**

As discussed earlier phenomenology is the study of the 'lived experience' and aims at gaining a deeper understanding of the meaning of our everyday life world (Van Manen 1984, Van Manen 2016). To reach this goal the research methods selected for data collection and analysis were built on the methodology of interpretive phenomenology as this approach seeks to gain understanding and involved clarifying data from the 'lived world' of the participants. A description of the process follows and begins with an explanation of ethical considerations, participant sample and recruitment. This section will also offer a description of the data collection tools and analysis technique used and will conclude with a review of the study's rigor and trustworthiness.

### **3.8 Ethical Considerations**

Ethical approval was obtained from the School of Nursing and Midwifery Ethical Research Committee and ethical approval to recruit parents from three main children hospital in the Republic of Ireland was granted on the 27th of Feb 2015. This was followed by ethical approval to recruit from three hospitals in the ROI and four child and parent support organisations that deal with many childhood growth disorders and syndromes affected with growth restriction worldwide. Although ethics committees are part of the ethics process in moving the study forward, being conscious of one's own ethical orientation to the study was also considered. This orientation was built on respect for the participants at all times. I had thought about the impact and the potential harm this study might have on participants. I also valued their trust in sharing their stories and experiences with me. The promise of anonymity and the promise of confidentiality were always maintained. Participants' information was not passed to another person or agency or used by others and was kept between the researcher and the participant at all times. However, participants were informed that only when it was legally impossible to maintain confidentiality such as in the case of breaching the child right act - as clarified in the information pack - that I had a professional obligation in this matter and therefore, the matter would be reported. Fortunately, I did not have any cases that I needed to report.

Students using research interviewing are obligated to maintain ethical standards (Keats

1999). I always presented myself as a researcher in face-to-face interviews with participants. I provided a clear, verifiable statement in the information pack indicating my identity and role. All participants were provided with a Participant Information Leaflet (See Appendix 1) that fully explained the study and further explanation was provided in the case of questions being put forward by the participants before the start of the interview. They also had time to read the consent form prior to signing it upon commencement of the interview (See Appendix 2). They were advised of the risks and the support put in place should they need it. They were informed of their right to withdraw from the study at any point with no repercussion.

Participants were assured that pseudonyms would be used instead of real names throughout the study in order to maintain confidentiality. This involved field notes and transcripts. Any documents or tape recordings that had real names mentioned in them were kept in secure locked storage cabinets which only I would have access to. Participants were given the choice of having their interview transcript sent to them as an opportunity for them to change anything or make comments and clarifications. However, only one of the 16 mothers interviewed requested this to be done. Due to the sensitivity of the topic and emotional issues that could arise from the research question, a list of potential resources and support organisations that parents could contact for psychological and psychosocial support was prepared.

### **3.9 Sample and Recruitment**

#### **3.9.1 Targeted Sample Group**

In qualitative studies, the aim in participant selection in hermeneutic phenomenological research is to select participants who have lived experience who are eager to talk about their experience and who are adequately diverse to provide rich stories of the specific experience (Laverty 2008). Kleiman (2004) adds that the typical sampling strategy in phenomenology is a snowball and purposeful method. The goal of phenomenological research is not to generate results that can be generalised, but to appreciate the meaning of an experience of a phenomenon. Therefore, the number of participants can be fairly small (Kleiman 2004). So, it was decided to focus on a small sample of mothers'

experiences who fitted the criteria of being a mother older than 18 years of a child receiving daily GH injection for at least 6 month who was between the ages of 2-18 years. Mothers were initially chosen as the targeted sample group as it was believed that mothers are the primary care givers for children with chronic conditions that need complex illness related treatment regimen (Starke & Möller 2002, Sullivan-Bolyai *et al.* 2004, Daniel *et al.* 2005, Nicholl & Begley 2012, Van Oers *et al.* 2014). However, fathers of children with special health care needs have been found to be highly involved in the care of their children, often advocating for their children's medical needs even if it means putting themselves in the health care system as “unpopular” family members (Chesler & Parry 2001, McNeill 2004, Yogman & Garfield 2016). Although in contemporary society significant progress has been made toward gender equality, more subtle inequalities remain. While both parents work, mothers are still more likely to make child care arrangements, schedule doctors' appointments and sign the permission slips (Schoppe-Sullivan 2017). However, father's involvement in care has been connected to higher adherence to treatment, better child psychological adjustment, and enhanced health status compared with families with nonparticipating fathers (Wysocki & Gavin 2004, Yogman & Garfield 2016). So, the targeted sample group was revised to include fathers as I believed that their voice is equally as important and to expand the inclusion criteria and increase recruitment rate which will be explained in more detail in the following section.

### **3.9.2 Recruitment Process**

A recruitment strategy was initially put in place to recruit mothers. In order to select a sample from across the Dublin area, the three main children hospitals in Dublin city were selected as potential recruitment sites as most children who are on GHT from the Republic of Ireland (ROI) are followed up at the endocrine clinics at these hospitals. Negotiating access to mothers through these three hospitals was implemented initially through emails to potential key personnel in each of the hospitals with a brief outline of the study (See Appendix 3). After receiving replies indicating interest to the study from the lead key personnel in each site, an introductory letter outlining the study and recruitment criteria was sent. Follow up meetings were arranged and agreement to support the recruitment process was sought from staff in two of the facilities in the

Dublin area. One of the key personnel in the third site expressed some concerns and was reluctant to support the study as I was not a member of the healthcare staff working at that site. After two attempts to negotiate access were not successful, I had to exclude that site. However, ethical approval and internal collaboration was achieved from the two other sites and recruitment was initiated very soon after that in May 2015. I was informed by one of the sites that agreed to allow recruitment of mothers, that I could not recruit children who were Small for Gestational Age (SGA) from that site as they were currently involved in a national research study.

### **3.9.2.1 Phase One (May 2015-April 2016)**

Recruitment began in May 2015 at the first site. They agreed to distribute 25 information packs I would provide them containing both a Participant Information Leaflet and an Informed Consent Form to be distributed to mothers at the endocrinology clinic. However, as packs were being tracked, it was found that they were not being distributed, because gatekeepers were very busy and faced difficulty doing so. Despite the effort of hanging up posters and flyers in both the waiting room and treatment room, and repeatedly following up on packs at the first recruitment site, no one came forward showing interest to participate in the study.

Meanwhile, recruitment started at the second site in October 2015 where gatekeepers did not agree to distribute the provided packs due to also being busy. Therefore, I was given permission to distribute the packs. I informed them that by doing so, patient confidentiality and autonomy may be breached but they did not find this to be an issue and were willing to fully support me doing so. A total of 14 packs were initially given to the gatekeeper at the second site after some persuasion to distribute them on my behalf. Posters and flyers were also displayed on the waiting room walls to enhance the recruitment strategy. However, after much waiting and not obtaining any responses from the mothers who received the first 14 packs distributed by the gatekeeper, it was finally decided to attend clinics and hand the packs in person to potential participants because the gatekeeper was not willing to do this anymore.

Having received permission to distribute the packs in person, I was mindful of how my

presence could affect recruitment. I considered that approaching the mothers in person could hinder potential participation because of how I looked being a Muslim woman wearing a head cover (Hijab). I thought parents could be hesitant and was aware that the trust factor could hinder recruitment. This was potentially due to the latest worldwide terrorist attacks in the name of Islam which I and the majority of people embracing Islam, absolutely condemn and consider these acts insulting to our faith. I was worried that I would have difficulty gaining the participants' trust and didn't want my external appearance to affect my study. So, I needed to work extra hard to try gain their trust. I was faced with this dilemma and needed to make a quick decision as I needed to start recruiting as soon as possible and relying on gatekeepers to distribute packs was not efficient. Consequently, I decide to go ahead with attending clinics at the two sites.

As recruitment was initiated in two of the sites mentioned above and with zero participants, I initiated an ethical application to a third site in a regional city after receiving support from the key personnel at that site. This was initiated to broaden my recruitment area and eventually in January 2016 ethical approval was achieved and a gatekeeper in that facility agreed to distribute the information packs to mothers who visit the endocrine clinic with their children. I also provided the gatekeeper with many posters and flyers to display in the waiting area as extra means of recruitment. As this site was situated in another city far from my residency, constant contact with the gatekeeper was maintained via email, so as to keep track of the number of packs that had been distributed from the 10 packs initially provided.

By February 2016, I reviewed the low participation rate from the three sites mentioned previously and hence reapplied for ethical amendment to my recruitment strategy to add parent support organisations and support groups. After gaining ethical approval to recruit from support organisations and support groups, recruitment was initiated there. Also, in order to widen the sample inclusion criteria, I sought both parents instead of only mothers. I also felt the need to mention to potential participants I met at the clinics that not only was I a children's nurse but also a mother of a child on GHT to try gain their trust and enhance participation rates. This was based on the notion of taking an insider role in qualitative research which is said to assist in building the researcher's legitimacy and often provides easier access to participants (Adler & Adler, 1987). Moreover, participants are more likely to be open with who they consider has shared



certain aspects of an experience or phenomenon, which may result in more richer data (Corbin Dwyer & Buckle, 2009). This caused a lot of back and forth thinking and much confusion ethically, but after discussing it with my supervisor, I was convinced that this was the only way to remove the authority of being a researcher, which put me at the same level as parents and more approachable because of my lived experience.

I am glad I made that decision along with reducing the interviews from three interviews for each parent to one and reducing the diaries from two diaries for each parent to one. These steps were taken after consulting my supervisor to find ways in which to improve participation rate and to help increase my sample size. I was advised to do so as encouraging parents to attend three face-to-face interviews and writing up two diaries each, may have served as obstacles for parents who live busy lives. So, with the new strategy put in place (adding parent support organisations to the list of recruitment sites for this study, updating all research materials such as posters, flyers, information leaflets and consent forms to include parents, reducing interviews to one interview each and reducing the diaries to one diary each) which was applied in April 2016, led to significant improvement in the recruitment rate.

### **3.9.2.2 Phase Two (April 2016 on June 2017) Successful Attempt**

From April 2015, participants were primarily recruited from three hospitals in the ROI, via support organisations, and snowballing. The sample was successfully reached after I had attended a couple of endocrine clinics at the first two recruitment sites and personally introduced myself and the study to potential participants at the clinics. I introduced myself as a research nurse and a parent with the same lived experience to gain additional trust. After verbally discussing the study with them, I gave out information packs reinforcing the verbal information given. My contact details (professional email and research phone number) were all included in the pack for participants to get back to me. I chose to ask the potential participants I met at the clinic, if they would like me to call them a week later to see if they still wanted to participate. This only happened when they didn't mind me doing so as I was conscious of not coercing potential participants. I only did so because I knew that some of them might have needed a single reminder, to remind them of the study, as they were busy

parents. After implementing this strategy, a single telephone call back a week later helped increase the study sample significantly.

Study posters and flyers were kept in reach of potential participants view for the whole period of recruitment and many personal follow-ups were made to make sure they were still hung and visible. The sample was also successfully recruited through study posters or flyers hung at recruitment sites. After the participants had seen the posters hung at the waiting rooms, they contacted me by email or text to show interest in participating in the study. I then organised to send them the information pack through post or email so they would be fully informed before following up with them in seven days' time to get their final informed decision to participate.

Later, participants were successfully recruited through four support organisations that help parents caring for children receiving GHT. After receiving their approval, gatekeepers from each organisation agreed to distribute study information through emails containing information leaflet and consent form, advertisement through their Facebook page, or allowing me to attend their annual group conference to present a brief introduction to the study to invite attending parents who were members of the organisation. By these means, mothers then contacted me through contact details which I had provided. Further along the study, social network was also used via snowballing technique as a participating parent shared the study information among her support group Facebook page. In addition, some participants approached me through email, text and/or call via snowballing through already participating mothers.

With all efforts to recruit a decent sample size of parents and changing the sample group from only mothers to both parents, the final reached sample included 16 mothers. This result strengthens the position I held at the beginning of the study and supports why I had initially chosen mothers as the preferred sample group for the study, as I believed from the very beginning that mothers are most often the primary care givers for sick children (Sullivan-Bolyai 2004, Daniel 2005, Starke 2002, Van Oers 2014, Nicholl 2012). Therefore, changing the sample group from only mothers to both parents did not make a difference with the study sample size.

### 3.9.3 The Study Sample

A total of 16 mothers comprised the final study sample. These mothers were caring for children receiving GHT for different reasons. The children in the study had a variety of conditions including growth hormone deficiency (GHD), Prader Willi syndrome (PWS), Russell Silver syndrome (RSS), small for gestational age (SGA) and Turners syndrome (TS). Most would have received GHT for growth failure and to improve short stature however, in the cases of PWS, the treatment was to improve muscle strength, maintain normal body weight, prevent obesity and improve short stature.

The mothers of these children were mostly stay at home parents and were caring for more than one child. Some of them were working mothers and with different educational backgrounds. The length of the experience of caring for a child with GHT ranged from six months to eight years. So, the sample provided a wide range of experiences (See Table 2 and 3 overleaf for further clarification-pseudonyms are used).

Table 2. Description of the Sample

Mothers	Age	Occupation	Number of Children	Duration of GHT	Indication
Francis	51	Child minder	3	2 years and 9 months	GHD
Sandra	44	Nurse	5	1 year	GHD
Joan	40	Home parent-PhD	2	8 years and 4 months	PWS
Rose	39	Home parent (Cardiac physiologist by training)	3	2 years and 3 months	GHD
Emma		Nurse	3	6 months	RSS
Michelle		Photographer	1	7 years	PWS
Berny	43	Special Needs Instructor	1	8 months	TS
Steph	51	Home parent (non-practicing Engineer)	3	2 years and 7 months	RSS
Liz		Home parent	1	4 years	GHD
Louise	46	Home parent (Non-Practicing Solicitor)	4	4 years	PWS

Mary	51	Marketing Consultant	3	6 years	TS
Nora		Home parent	2	8 months	SGA + failure to catch up in growth
Bridget	41	Home parent	3	1 year and a half	GHD
Loran	38	Home parent	5	1 year	GHD
Kelly	55	Home parent	3	1 year	PWS
Loretta	42	Midwife	4	2 years and 2 months	SGA+ failure to catch up in growth

Table 3. Medical Conditions Involved in the Study

Number of Participant	GHT Indication
6	GHD
4	PWS
2	RSS
2	TS
2	SGA and failure to catch up in growth

### 3.10 Methods of Data Collection or Gaining Understanding

Indepth interviews and diaries were used to gain an understanding of each participant's experience of caring for their child receiving GHT.

#### 3.10.1 Indepth Interviews

The hermeneutic phenomenological methodology was underpinned and informed by Gadamer's hermeneutics. A qualitative research method was selected because earlier research into parents' experience caring for children receiving GHT was limited. Qualitative research is ideal for exploring the lived experience and for constructing a deep understanding of human experience. Therefore, a phenomenological approach would best suit a study that seeks deep description, interpretation and insights from the parent's' perspectives. In the human domain this typically represents collecting 'deep' information and perceptions by inductive, qualitative methods such as interviews,

participant observation, and expressing it from the perception of the research participants (Lester 1999). Qualitative interviews are one of the most common forms of data collection tools in qualitative research and are usually considered the 'golden standard' tool in qualitative research (Mason 2002). In addition, interviews are considered the primary method of data collection in phenomenology research because it explores, illuminates and gently examines the participants' description (Wimpenny & Gass 2000).

As this study's aim was to explore parents' experiences, interviews fitted with the chosen ontological position as participants' views, knowledge and interpretations of their experience are meaningful. Interviewing was interpretive and understanding was sought through interpretation where meanings were co-created between the interviewer and interviewee. This was achieved by presenting an interpretation of the interviewee's data, looking for more clarification by asking questions and exploring an aspect of the conversation instead of another (Geanellos 1999). I finally chose to interview parents only once after initially planning to conduct three consecutive interviews that would be no longer than an hour each. This change of plan was influenced by the failure to get parents to participate in the first full year of starting recruitment, as aforementioned in recruitment (phase one). Many parents during that period contacted me using the contact details provided on the posters that were displayed in waiting and treatment rooms at recruitment sites. However, after sending them the information leaflet and consent form, they declined participation by either expressing how busy they were or simply never replying. It was immediately noted that there was a pattern of rejection to participate in the study especially after providing them with the study information pack. It was then concluded that those parents must have found it too difficult to participate in the study. I kept in mind that I am inviting busy parents who may already have enough weight to carry on their shoulders and may find committing to this study very demanding. So, my next plan was to facilitate the participation process as much as possible and decided to reduce the number of interviews to one interview each that lasted up to an hour after gaining approval from my supervisor.

### 3.10.2 Conducting Interviews

Since many parents lived outside the city and some in remote areas of Ireland, all efforts were made to meet them at their home or a local café of close proximity to them. However, most interviews were conducted at the participant's home. A small number were conducted at a location chosen by the participant, such as a local café. Making long distance journeys to reach some of the participants who lived across the country proved to be of great challenge. Commuting using a personal car usually lasted between 1-3:30 hours to reach the participant's location and vice versa back home. Usually, to avoid delays I would leave very early in the morning at around 5-6am before morning rush hour and heavy traffic. As for the interviews that were in the city, I met mothers at their homes or local café according to their preferences.

In order to break the ice and make meeting the mothers at their homes easier, I brought along a box of chocolate as a gesture of appreciation for accepting to be interviewed at their home. I also prepared an information pack consisting of an information leaflet, consent form and a diary. Prior to each interview and after meeting the participant, I went through the information leaflet and reminded her that she had the right to withdraw at any time. I also asked the participant if she had further unanswered questions that need to be addressed and/or inquiries about my professional background. The participants also had the chance to read through and prepare questions prior to the interview as the information packs were sent to them by post or email, at least seven days in advance to the interview date. When the participant did not have any questions to ask and was happy to go ahead with the interview, the recorder was powered on.

Only a very few times did a participant whom I had recruited through meeting at the clinic asked me how my son was doing on GHT, or how long he had been on it before starting the interview. In this situation I gently redirected her to the interview and gave very vague answers such as 'He is fine' or 'he is on it for quite some time now' and reminded her that I am very interested to hear her story and will get back to mine after the interview had ended. This did make me feel at times pressured and uncomfortable as to how my identity and role as a researcher would be maintained, or how my experience giving GHT to my son would overshadow theirs if mothers wanted to know my experience with GHT. I was quite aware and conscious of this happening so I felt the

best way to stop my own experience influencing their own stories prior to their interviews, was to make sure I mentioned only the shared experiences I had with them after the interview concluded and only if they asked me. This was to maintain the trust given to me in the first place by the mothers and to enhance the relationship between us. I felt ethically obligated to share the commonalities with some mothers more than others, if they asked me about my situation and I would have been the first parent who has a child on GHT or/ and with the same very rare condition for them to meet.

When commencing the interview, I explained to each mother that I did not have a set of questions and that I would only ask one general question ‘Tell me about your experience caring for your child on GHT?’ and that I might ask questions later on to clarify things that they had mentioned. The recorder was turned on only when they were ready to start. Interviews lasted between 15-77 minutes and only ended when the participant did not have anything more to share. Throughout each interview, I kept a warm, non-judgemental attitude towards the mother, mindful to be responsive, and contemplate on what was said. I embraced an active listening attitude and encouraged the parent to be reflective. These actions demonstrate an attitude of being open, which invigorated a pathway toward understanding and interpretation. The manner in which the participants responded to questions was noted such as laughs, pauses, sighs, sarcasm, and confidence with which they recounted their experiences. Body cues were also noted for signs of uneasiness to a topic or question, or even hesitation to give more information. I listened carefully and with much empathy, and I avoided saying ‘I know’ to the mothers, although I could very much relate to their experiences. During the interviews, I wrote key words of issues to investigate further while the interview proceeded. Since meanings are co-created, I knew I needed to be fully aware of my pre-understandings and conscious of the influences throughout the interview. Having two cultural backgrounds (Saudi Arabian and Irish), I am fluent in both Arabic and English. I was mindful of sometimes facing difficulty trying hard to word some of the ‘previously unprepared’ prompt questions correctly during interviews, in an effort to convey clear and simple language questions. For example, as I listened back to some interviews, some questions were very long-winded and could have been asked more concisely. On the other hand, I found myself relating to these mothers easily and effortlessly, understanding what they were telling me as being Irish myself helped me understand the humour, and slang that I believe I would have struggled to understand if

I was not.

At the end of each interview, the mothers were thanked for their time and participation in the study. I then provided them with a diary each and asked them to fill it in within two months and instructed them how to do so. They were asked to send the diary when they were finished to my postal address at the School of Nursing and Midwifery at TCD. I also asked for their permission to contact them again in case I needed to remind them of diary completion or if I needed any clarifications when analysing their data. I also ensured them that they may contact me at any time if they had any questions or felt the need to change or remove any shared information. After leaving the participant, important points like time, setting, issues brought up during the interview, interruptions, thoughts and feelings about the interview, issues raised and decisions made were written down as field notes. This helped me understand and address what was needed to co-create the text. In total 16 interviews were completed at the end of the data collections stage from May 2016 - June 2017.

### **3.10.3 Diaries**

In addition to the 16 interviews, eight diaries were collected from the participating mothers to gain insight into the social process and the rationality that underpins actions and events (Alaszewski 2006). By using unstructured diaries, these diaries could be treated as documents that represent the naturalistic life stories of the parents' experiences (Alaszewski 2006). Diaries offered a valuable adjunct to the interviews in this qualitative research study, particularly when it is desirable to collect data unaffected by the researcher's presence, and gather stories that are part of the participant's everyday life that are not controlled or influenced or provoked by (I) the researcher (Nicholl 2010). In addition, diaries provided a natural way of using language because they create texts that make use of social conventions to understand what is happening to an individual (Alaszewski 2006). The data generated from diaries can form an important part of the overall body of knowledge. However, the use of diaries requires careful considerations as success depends on many factors such as diary structure, guidelines, participants' attentiveness in the research topic and their ability to meet the demands of recording (Nicholl 2010).



In this study, diaries were used with the interviews to better understand the phenomena, as they can facilitate access to hard to reach or hard to observe phenomena. They also provided confirmatory evidence of the information obtained from interviews and also helped overcome memory obstacles so that information that the mother forgot to mention in the interviews wrote down in a diary at her convenience. However, using diaries in research could have limitations. I had to rely on the participants to complete the diary. The initial plan of the study was to invite parents to complete two diaries each (see recruitment phase one). However, as mentioned previously, I felt that asking parents to fill in two diaries would be demanding and could reduce their desire to participate in the study. Therefore, mothers were only asked to complete one diary each as this was found to be more manageable and workable (see recruitment phase two).

A diary template was used that had been modified from the diary template used by Nicholl (2008) and it has been peer reviewed for clarity and readability (see Appendix 4). Even though the diary was easy to follow, I instructed each mother after the interview that she could use the diary to share her experiences in her own time and space. I also gave them the freedom to fill in as much or as little they wanted. I asked if two months to fill it in was enough and if they needed more time to do so, they could. I was not very strict on following up too soon because I felt that by restricting time and amount of information needed to complete the diary, would put unnecessary pressure on the mother. Despite giving them a minimum of two months to complete it, many did not send them back in that time frame and were then reminded to complete the diary many months after their interview. Some mothers after the interviews told me that they would not have anything more to add but will still take the diary to have a think and try adding something. Several expressed that they hoped they would get the time to fill it in, and some expressed difficulty in writing things down on paper. After the two months follow up with some mothers, a few asked me to clarify again what I needed them to do with regards to the diary and expressed how nothing new has come up or nothing extra was to be added. Others expressed how every night had been the same since the interview and stated that they definitely didn't have more to add and did not wish to complete the diary. Thomson & Holland (2005) also explained how a huge variation in the depth and quantity that study participants want to write in diaries exists. Mothers in this study wrote as little as half a page to as much as 4-5 pages. The content of the diaries also

varied. One mother wrote down the points discussed in the interview, which emphasised the importance of those discussed points to that particular mother. Others wrote detailed diary entries of many past challenging encounters.

It was noted by Corti (1993) that it cannot be expected that participants will keep their diaries in close reach to them at all times to write down the moment despite all the efforts of the researcher. This was soon noted when I followed up with mothers to remind them of the diary and ask if they had faced any problems completing them. Some asked for a new copy of the diary to be resent to them - with comments like “I misplaced the diary’ or ‘I can’t find the copy you gave me’. Every effort was made to resend them a new one to be completed. Having said that, eight mothers did complete their diaries and had them sent to my college postal address or college email address. I received them mostly in the form of hand-written documents. One was given in the form of a typed word document. By October 2017, I had received the last diary.

### **3.11 Chosen Analytical Framework**

Interpretive work has been known for its intriguing feature of lacking a prescribed method for analysing data (Fleming *et al.* 2003). Instead of strictly adhering to a specific method, hermeneutic-phenomenological approaches are based on a body of knowledge and understandings, a history of lives of philosophers and authors, which, taken as an example, symbolises both a basis and a methodological ground for current human science research practice (Van Manen 2016).

Although Gadamer (1975) proposed valued insights into how one may acquire a deep understanding of texts, he did not suggest either a methodology or a method for doing so. Nevertheless, it is within Gadamer’s view that in order to understand, a systematic approach is needed. Gadamer (1975) clarified that hermeneutics is not a simple matter of endorsing a method, however this positions a struggle for the hermeneutic researcher, who wants structure to begin the research process. A number of authors, notably (Colaizzi 1978, Van Manen 1984, Giorgi 1989, Diekelmann 1992, Koch 1993) have proposed methods for initiating phenomenological or hermeneutic studies. I found Colaizzi’s (1978) method not quite suitable as he encourages researchers to identify their

preconceptions only to generate a research question and not as a prerequisite for understanding, which opposes Gadamer's work.

While Van Manen (1984) suggested that added knowledge from using one's pre-understandings would be turned against itself, therefore, inconsistent with the role of pre-understandings which did not suit my views. This is also seen in Giorgi's (1989) approach, which is firmly based on the phenomenology of Husserl that requires reduction or bracketing of pre-understandings, which in my opinion would be difficult if not, impossible. Diekelmann's (1992) method requires a team approach in data analysis in order to reveal contradictions and inconsistency in the analysis between each team members to control bias rather than to identify them, which I think is necessary for full understanding. Although Koch's (1993) six steps of analysis seemed very appealing to me, she failed to express a clear relationship between her applications of the work of Gadamer (Koch 1993). So the most recent analytical framework by Fleming *et al.* (2003) who offered a template for analysing Gadamerian-underpinned research was sought, as it helped guide many nursing researchers through the application of Gadamer's philosophical traditions in five clear stages.

This analytical framework was chosen because it provided a clearer guide to how to use the major concepts of Gadamer's work and how to integrate them into the research study. The process starts from as early as choosing a research question right down to maintaining the trustworthiness of the findings, which seemed structured in a sense. So for this reason, Fleming *et al.*'s (2003) analytical framework was chosen over other frameworks that were found in the literature used by Gadamer's underpinned research studies such as (Koch 1993, De Sales 2003, Austgard 2012). All frameworks covering the main concepts of Gadamer's philosophy (pre-understandings, fusion of horizons and the hermeneutic circle) previously mentioned were found to be very similar and too vague to follow. I also chose to integrate Ajjawi & Higgs's (2007) stages of data analysis developed for their research study on using hermeneutic phenomenology to investigate how experienced practitioners learn to communicate clinical reasoning. It was found that the steps were clear especially on how to apply the fourth step of the chosen framework of Fleming *et al.* (2003), which is 'Gaining understanding through dialogue with text'. This will be emphasised more in the fourth step of the framework as illustrated in Table 4. Fleming *et al.*'s (2003) analytical framework's main five stages

includes; deciding upon a research question, identification of pre-understandings, gaining understanding through dialogue with participants, gaining understanding through dialogue with text and finally establishing trustworthiness which will be explained in more detail.

Table 4. Analytical Framework Used for Data Analysis; Modified from Fleming *et al.* (2003) and Ajjawi & Higgs (2007)

<b>Fleming <i>et al.</i> (2003)</b>	<b>Ajjawi &amp; Higgs (2007)</b>	<b>Fleming <i>et al.</i> (2003) &amp; Ajjawi and Higgs (2007) “steps followed”</b>
<b>Step one:</b> Appropriate open research question.		Appropriate open research question.
<b>Step two:</b> Identification of Pre-understandings.		Identification of Pre-understandings.
<b>Step three:</b> Gaining understanding through dialogue with participants (interviews and diaries).		Gaining understanding through dialogue with participants (interviews and diaries).
<b>Step four:</b> Gaining understanding through dialogue with text (hermeneutic circle and fusion of horizons).	<b>Stage one:</b> (Immersion)	Gaining understanding through dialogue with text:  Organising the data-set into texts (transcribing).  Identifying the participant’s horizon:  Iterative reading of texts.  Preliminary interpretation of texts to facilitate coding.



	<p><b>Stage five:</b> (Illumination and illustration of phenomena)</p> <p><b>Stage six:</b> (Integration and critique)</p>	<p>circle. (Aggregation)</p> <p>Themes are challenged:</p> <p>Themes to be challenged by the researcher and in turn challenged the researcher's pre-understandings.</p> <p>Linking the literature to the themes identified above.</p> <p>Reconstructing interpretations into stories.</p> <p>"Rich and more detailed understanding is achieved"</p> <p>Interpretation (fusion of horizons):</p> <p>Critique of the themes by the researcher and externally.</p> <p>Reporting final interpretation of the research findings at this point in time.</p>
<p><b>Step five:</b> Establishing trustworthiness.</p>		<p>Establishing trustworthiness.</p>

### 3.11.1 Deciding Upon a Research Question

In this stage it is essential to examine the appropriateness of the research question to the methodology assumptions. Research conducted through a Gadamerian tradition is established from an aim to attain deep understanding of a phenomenon. So according to (Gadamer 1990) the essence of the question contributes to the opening up of

possibilities for deep understanding. He highlighted the significance of choosing the correct questions for elaboration of the hermeneutic situation. Thus, the original or opening research question impacts the whole research process. He additionally mentioned that there is no understanding without the activity of questioning and that there was a very narrow relationship between the two that gives sense to hermeneutic research (Gadamer 1990).

When I met with my participants for the interview, I explained that I did not have a set question to ask and they were invited to reflect openly and deeply on their experiences through asking them an open-ended question (Kvale 1983). The interviews were in the form of conversations and to deepen the understanding, the opening question was followed with clarifying questions (Fleming *et al.* 2003). I generally started the interview with this opening question ‘Tell me about your experience of caring for your child receiving GHT?’ and then might have used probing questions like ‘Tell me more about this?’ or ‘What is this like for you?’ Consistency on using this main question throughout the interviews was generally maintained to keep myself and the participants oriented to the subject being researched and to continue asking relevant questions throughout the study to try reach a deep understanding (Geanellos 1999).

### **3.11.2 Identification of Pre-understandings (Before Data Collection)**

Researchers using the philosophy of Gadamer need to identify their pre-understandings or prejudices on the topic of interest (Fleming *et al.* 2003). It is recommended remaining open to hidden prejudices and that unexamined prejudices may impact or limit the horizon of understanding so one’s prejudices need to be provoked (Fleming *et al.* 2003). Prejudice can limit the individual’s horizon which consequences in not seeing far enough, underestimating the familiar, which results in not seeing or closing off further understandings by assuming it is already known (Geanellos 1999). With my personal and professional experience in the field of GHT and underpinning my study with the philosophy of Gadamer, I knew I had to bring my pre-understandings and prejudices of the topic into consciousness. Reflecting upon my prejudices will enable me to move past my pre-understandings and transcend my horizons to better understand the phenomena, which will influence the research findings (Fleming *et al.* 2003). I

found this process not as easy as it might seem. In order to provoke my pre-understandings, I needed to bring out hidden prejudices and ones that I might underestimate myself. An appropriate approach I found to help me bring my pre-understandings to the surface was having conversations with my supervisor, colleagues and family members. Then the pre-understandings that became visible within the discussion would be described and analysed in the research report (Fleming *et al.* 2003). During the research process my pre-understandings changed from the point of data collection until the write up stage and in order to actively focus on this transformation, I periodically reviewed them through holding a reflective diary to help me remain oriented to the phenomena and enable me to enter the hermeneutic circle (Geanellos 2000). To further clarify this step, a written note of my pre-understandings prior to starting data is shown in Appendix 5.

### **3.11.3 Gaining Understanding Through Dialogue with Participants (Interview and Diary)**

Gadamer did not like the expression ‘collecting data’ or ‘gaining information’ and preferred to use the term ‘gaining understanding’ (Fleming *et al.* 2003). He remarked that understanding may only be potential through dialogue, one being open to the opinion of the other (Gadamer *et al.* 2004). Dialogue means not only a conversation between two people, but also a dialogue between reader and text. In both cases language is considered the mean and it is through language that understanding becomes potential (Gadamer *et al.* 2004). In order for the text to be co-created by the researcher and participants, I sought to capture the temporality of every remark within a conversation and so I played an active research instrument. Understanding would come into view when my horizon infuses with the horizon of the participant (Polkinghorne 1983, Morse 1994).

A ‘horizon’ is a series of vision that incorporates everything seen from a specific vantage point (Gadamer *et al.* 2004). A person with no horizon, in Gadamer’s opinion, does not see far enough and overestimates what is nearest at hand, while to have a horizon means being able to see past what is familiar at hand. This is where questioning becomes practical, as it helps the person create new horizons and understandings



possible which is a critical aspect of the interpretive process (Lavery 2008). He emphasised that we are all part of history and consciousness and he recognised that the merging of individual's horizon within the prejudices of history, including those offered by people and text in the development of knowledge and understating (Gadamer *et al.* 2004). Therefore, it is important for the researcher to try to understand how personal feelings and experiences influence the research, and then incorporate this understanding into the study and this was encouraged by maintaining a reflective diary throughout the phase of data collecting. An example of keeping a reflective diary of each interview is illustrated in Appendix 6.

Hermeneutic differences is an essential part of hermeneutic understanding which was noted by Gadamer and that a researcher should not attempt to see through the eyes of the participants to understand the phenomena (Lamnek 1995). Gadamer argues that by putting oneself into someone else's shoes, he/she is ignoring one's own pre-understandings (Gadamer *et al.* 2004). It is not possible for two people to have the same pre-understandings, and reach the same understanding at that point in time (Gadamer *et al.* 2004). Understanding is always considered a historical, dialectic and linguistic occurrence and is achieved through the 'fusion of horizons' (Pascoe 1996). Our horizons are constantly being shaped and changed by our past and awareness of the present (De Sales 2003). Geanellos (1998) claims that the only way to reach interpretive understanding is by engaging with text inside the hermeneutic circle (Geanellos 1998).

#### **3.11.4 Gaining Understanding through Dialogue with Text (Transcribing and Analysing)**

Gadamer emphasised the necessity of writing down the interviews (transcribing) even though he echoes the power of spoken words over the written. However the researcher should not totally rely on the written words (transcripts and diaries) but should also listen to the spoken words (tapes) (Gadamer *et al.* 2004). 'Text' not only signifies to the written transcript, but also to recordings, comments made by the researcher about the interview situation and observations. Field notes describing the context and emotions not noted on the recordings were conveyed to the interview text.

### *Interpretation is in its infancy*

Interviews were transcribed verbatim and repeatedly read to get a good grasp of the whole text. Gaining understanding of the whole text was the starting point of the analysis because the meaning of the whole text will impact the understanding of every other part of the text. Already my pre-understandings could have caused the build-up of a sense of anticipation that influenced the first encounter with the text. However, I was fully aware of trying to keep an open mind and reflecting on my influences. This step (immersion) was implemented by reading through the transcripts while hearing the recordings several times before exploring the parts. This step is correlated with the 'Preliminary interpretation of texts to facilitate coding'.

Transcripts were considerably annotated with identification of features of significance, as well as narratives of my feedback to what they said and what I said. I carefully examined each transcript for ideas, which seemed to add voices to the meaning of the phenomena. Also, I attentively reviewed the transcripts for clues of my own transformation. When articulating the meaning units, the question put to the text concerned meanings of caring for a child receiving GHT. Across this dialogue with the text, main meanings were sought. Both my horizon and my participants' horizons were fused into a tentative interpretation. An example of the initial preliminary interpretation of texts to facilitate coding is illustrated in Appendix 7.

### *Rich and more detailed understanding is achieved*

Next, every single section or sentence (the part) is investigated to expose its meanings to understand the subject matter. This was initially done by identifying first order 'participant constructs' (open codes) as shown in Appendix 8. These constructs represent participant's horizon referring to participants' ideas expressed in their own words or phrases, which capture the precise detail of what the person is saying. This step is followed by abstraction phase, where second order 'researcher constructs' (categories) are identified as displayed in Appendix 10. Due to the massive number of open codes, it was decided to use NVivo 11 to assist in the organisation of open codes and forming the categories as shown in Appendix 10. Then, subcategories under these categories are generated manually as displayed in Appendix 11. Subsequently, core categories and subcategories are grouped manually into sub-themes as shown in Appendix 12. These sub-themes are representing researcher's horizon, which are

generated using the researchers' theoretical and personal knowledge; these were abstractions of the first order constructs (Integration).

Then the synthesis and theme development phase was initiated (meshing the horizons) where grouping of sub-themes into themes are made. Afterward, further elaboration of themes and relating them to the whole meaning of the whole text to try in turn expand the meaning of the whole. This movement from the parts back to the whole text is the core of the hermeneutic circle (Aggregation). It is expected then with the deepened understanding of the whole text, meaning of the parts can expand. When articulating the themes, the question put to the text concerned meanings of caring for a child receiving GHT. Themes are constantly being challenged by myself and in turn challenged by my pre-understandings. This step is illustrated in Appendix 13 and Figure 1. After that, the illumination and illustration of phenomena phase trails. This is where I started linking the literature to the themes identified above and finally reconstructing interpretations into stories which will be illustrated in Chapter 4.

*Final interpretation is achieved "fusion of horizons"*

The final phase of this step is integration and critique. This is where the critique of the themes by the researcher and reporting final interpretation of the research findings are completed which will be discussed in the final discussion chapter (see Chapter 5).

### **3.11.5 Establishing Trustworthiness**

Hermeneutic phenomenological research aims to highlight the experience under exploration as close as possible to how it was lived by the participant, while noting the interactions and overlapping horizons between researcher and participant (Cohen *et al.* 2000). As a researcher underpinning my study on the philosophy of Gadamer, I was responsible for establishing the trustworthiness (rigour) of the research process and the truthfulness of my analysis (Fleming *et al.* 2003), otherwise, the findings of the study would be meaningless. Criteria laid out by Lincoln & Guba (1985) applicable to a Gadamerian research process were used to establish trustworthiness. These criteria are credibility, dependability, conformability and transferability.

Credibility indicates the confidence to the truth of the data and its interpretation (Lincoln & Guba 1985). In this study, methodological coherence, and researcher's responsiveness were strategies for achieving this. Methodological coherence is reflected throughout the process of the study in a non-linear way as the researcher moves back and forth between framework and application to guarantee congruence amongst deciding on a research question, literature, recruitment, data collection and analysis (Kvale 1997). My responsiveness was achieved by sustaining truthfulness, staying open, and being sensitive while listening to each participant's story to ensure her perspective was clearly noted and represented as clearly as possible. And then the use of direct quotes upheld from the used text allows the reader to partake in the validation of the data. Dependability is the stability of data over time and conditions (Lincoln & Guba 1985). It is however uncertain whether dependability can be indorsed to a study that is underpinned by Gadamer's philosophy as my horizons and those of the participants will change, therefore, a final interpretation will not be achievable, and interpretation of data will change over time.

Conformability is the objectivity of the data (Lincoln & Guba 1985). Coherent with hermeneutic phenomenology, I didn't hold an objective position, but included my pre-understandings as part of the data. Objectivity in hermeneutic research can be comprehended as being open to the study text. Transferability according to Lincoln & Guba (1985) is generalizability of the data. To what extent can findings be transferred to other settings or a group? As this study is a hermeneutic one, the aim of this study is not generalized by the findings; but to provide deep description of the phenomenon of caring for a child receiving GHT from a parents' perspective.

Understanding according to (Gadamer 1990) can only be achieved by creating harmony between the whole and the parts of the text. This proposes a standard for trustworthiness in relation to the development of understanding rather than simply to the end result of the study. My responsibility as a Gadamerian researcher was thus to deliver adequate detail of the processes, and the findings in the research report. It is suggested by Denzin & Lincoln (1998) that reflexivity plays an essential part in amplifying the integrity, credibility and trustworthiness of a qualitative research study. This involves ways of questioning own thoughts, attitudes, reactions, and habitual actions in an attempt to understand our role in relation to others (Bolton 2018). Being self-aware involves

thinking about positionality such as ethnicity, age and previous life experiences, social identity, role and personality (Dowling & Cooney 2012). This did help with looking more critically at situations and relationships and finding ways to revise and enhance being and relating (Cunliffe 2009). As a novice doctoral qualitative researcher using reflexivity, I found this practice rather challenging and chaotic. It was a learning curve and a tool I learned quite gradually along the way. I came across the article 'Reflexivity and Professional Use of Self in Research: A Doctoral Student's Journal' (Valandra 2012) and she offered tips and guidelines that I found useful to spur self-affords with reflexivity. Valandra (2012) offers suggestive reflexive questions at every phase of a research study.

The pre-research phase involves being honest with myself about my biases and limitations, my knowledge or lack of it, around the topic and population of inquiry, and the possibility of my perspectives and experiences to affect the research process. Some pre-research phase questions considered in alerting myself to the topic, idea, or population include:

1. What do I already know about this topic?
2. How do I know what I know?
3. How have my personal and professional experiences shaped what I know?
4. What assumptions, biases, attitudes, and beliefs shape my construction of this idea?
5. What am I passionate about regarding this topic/idea?
6. How are my life experiences shaping the design of this study?

I tried engaging in dialogue with self to experience answering these questions to highlight my awareness of my experience in relation to the object, topic, and intended audience. I wrote down my pre-understandings and knowledge around the topic of GHT, which stemmed from my personal and professional experiences (as previously noted under evolution of research interest in Chapter 1 and in Appendix 6. During the implementing phase, I kept a diary to capture and document my thoughts, feeling, insights and field notes and such questions were considered along the way:

1. How do my life experiences shape the implementation of this study?
2. What motivates the participant to talk to me?
3. In what ways can what I disclose about myself potentially influence what study participants share or not share about themselves?
4. What am I noticing about study participants' communication patterns?
5. What kind of information do study participants share about themselves without solicitation from me?
6. What do study participants share before and after the formal interview/study begins and ends?

An example of a reflexive log that was noted on my research diary while recruiting mothers at one of the recruitment sites is shown in Appendix 14. In the analysing and writing up phases of the study, reflexive logs were mainly written as side notes on the interview transcript documents as well as annotations (see Appendix 15) along the way that were entered into NVivo 11. Such questions were considered:

1. Whose stories are represented?
2. Whose voices are missing?
3. What are the similarities?
4. What are the differences?
5. In what ways did my presence influence the participants' responses?
6. In what ways am I invested in the study's findings?
7. How did participants' responses after the formal interview influence my interpretations of their stories?

### **3.11.5.1 Member Checking**

Much time was spent reflecting on whether the transcripts should be returned to the mothers and after lots of reading, there appeared to be opposing views on this as 'member-checking' has been one of the techniques to improve rigour in qualitative studies (Grbich 1998). However, Caelli (2001) proposes returning transcripts to participants to review, clarify, or validate findings depending on one's theoretical stance. McConnell-Henry *et al.* (2011) and Webb (2003) suggest that member-checking

is unsuited for phenomenology as there is no edict in interpretive research to verify or generalise findings. This is grounded on the belief that the participants' story being true at that specific time and on reflection can change their recollections and change their initial beliefs and perceptions. Thus, in agreement with this principle, transcripts of the interviews were not returned to the parents for checking. However, participants were asked if they wanted to remove anything disclosed in the interview after the interview was over and only one mother asked to remove one piece of information from the transcripts. The findings were discussed on many occasions with my supervisor as it is recognised that 'experts' can help to corroborate the findings (Whitehead 2004), enabling further insight and depth through challenge and discussion (McConnell-Henry *et al.* 2011). Nonetheless this belief was used knowing that multiple truths exist and interpretation in hermeneutic phenomenology is subject to change depending on the context and experience of those intricate in interpretation.

#### **3.11.5.2 Using NVivo**

Using (QSR NVivo 11) the qualitative data analysis programme facilitated the ease of returning to the transcripts to see the code or key phrase in the context of the interview and equivalent text. It also assisted in the management of the data and allowed easy retrieval of quotes from the interview transcripts (Denscombe 2014). Building connections between one piece of datum with another was simpler. The computer software package was also used to identify key words but the need to visually and manually work with the data meant moving beyond the computer software (QSR NVivo 11). Although using models and diagrams was feasible using the program as themes and links developed, writing and collating ideas on paper (word documents) in schematic ways was much easier. The software was used initially to open code the data collected. Then after that open codes were gathered into categories (see Appendix 10) and at this point the software was mainly used to manage the data, followed by visually and manually working with the data using word document to complete the final analysis. As much as the software tool was helpful in terms of managing the data, it couldn't do the analysis, nor could it perform the reading and thinking for me (Gibbs 2002). So, generating subcategories and themes was mainly done manually. Emerging ideas and results of my thinking were recorded using the software (see Appendix 15) however

trivial they seemed. This was useful but given the changing and evolving nature of thinking in this study, it was hard to record all insights, so a research diary was also used. Feeling distant from the data was also noted as a disadvantage in using the software. However, by returning to the transcripts and listening back to the recordings from time to time, I was able to keep close to the participants' worlds.

### **3.12 Conclusion**

This chapter has considered the philosophical underpinnings of this research study based on Gadamer's interpretive phenomenology and has provided detail on the research process undertaken. It provided a justification for the choice of methodology and an overview of the research methodology chosen based on the philosophical underpinnings of phenomenology. It explained my worldview based on beliefs, values and attitudes around the nature of reality and truth and how this swayed the decision to choose phenomenology. It summarised the choice of methodology and the philosophical foundation of phenomenology. It also considered Gadamer's philosophical concepts that supported the philosophical underpinning of this study. The chapter described the research methods used to complete the research study and finally concluded with a review of the study's rigour and trustworthiness.

The following chapter presents the research findings based on the four main themes identified from the mother's interviews which were: (1) "It's the right thing to do" Striving for the security and the wellbeing of the child, (2) "Doubting yourself constantly" Constant uncertainty, (3) "But then you just get used to it I suppose" Adhering to GHT and lifestyle changes - the new normal, (4) "I hadn't been told anything about it" Information behaviour; looking for normality and certainty.



## **Chapter 4: The Findings**

### **4.1 Introduction**

The aim of the study was to use a hermeneutic phenomenological approach to achieve a greater understanding of the nature and meanings of the experiences of parents caring for children receiving growth hormone treatment (GHT). The experiences unfolded from the participating mothers comprise three main concepts that were central to their lived experience which were: uncertainty, normalisation and stigma. These concepts were used to elaborate on the meanings of their experience of caring for their children receiving GHT. This chapter includes a description of the mothers and children and outlines the four major meanings of their lived experiences which are explained in the form of themes. These themes were: (1) striving for the security and the wellbeing of the child, (2) constant uncertainty, (3) adhering to GHT and lifestyle changes - the new normal and (4) information behaviour; looking for normality and certainty. The details of the parents inside world are illuminated and quotes from participants are used throughout to support the findings.

### **4.2 Alteration to Transcripts**

Within this chapter, identifying markers (parents, child, hospital, city, support organisation, health personnel and growth hormone manufacturing company) have been removed. Pseudonyms have been used for the mothers and their children for example [Sam]. Personnel and sites have also been anonymised. This is indicated by the sites being changed to X or Y and put between brackets “[ ]” for example [X hospital]. Impersonal pronouns have replaced other names throughout to help with clarity in reading; for example, father’s name is replaced with [the father], drug company name or nurse are replaced with [drug company or drug company nurse], health personnel’s name replaced with [endocrinologist, nurse and GP]. I have removed some text and this is indicated by... (3 dots). Brackets “( )” that don’t replace names, signal the addition of my text to provide context and the removal of other possible identifiers. Quotations are indented without parentheses.

### 4.3 Sample

This study was intended for both parents as the chosen sample. Both mothers and fathers were invited to the study however; the sixteen participants who provided data were only mothers from the ROI. Three mothers had more than one child in the family that were diagnosed with a medical condition non GHT related such as (Autism and Langerhans Cell Histiocytosis). One mother had a brother who was diagnosed with growth hormone deficiency (GHD) who received GHT as a child. Seven mothers were working outside the home in healthcare or own businesses during the period of data collection. The remaining nine were home carers and three were non-practicing professionals. Background information and geographical location is provided in Table 5 and 6.

Table 5. Pseudonyms

Mothers	Child's Gender	Child's Age	Duration on GHT	Condition
Francis	M	11 years old	2 years and 9 months	GHD
Sandra	M	12 years old	1 year	GHD
Joan	M	12 years old	8 years and 4 months	PWS
Rose	M	8 years old	2 years and 3 months	GHD
Emma	M	9 years old	6 months	RSS
Michelle	M	8 years and 3 months old	7 years	PWS
Berny	F	5 years old	8 months	TS
Steph	M	6 years old and 9 months old	2 years and 7 months	RSS
Liz	M	8 years old and 11 months	4 years	GHD
Louise	M	5 years old	4 years	PWS
Mary	F	11 years old	6 years	TS
Nora	F	6 years old	8 months	SGA
Bridget	M	10 years old	1 year and a half	GHD
Loran	M	9 years old	1 year	GHD
Kelly	F	≥ 13 years old	1 year	PWS
Loretta	M	8 years old	2 years and 2 months	GHD

Table 6. Geographical Location of Sample

Geographic Location (ROI)	Number
Co Cork	4
Co Dublin	7
Co Galway	1
Co Mayo	1
Co Meath	1
Co Sligo	1
Co Wicklow	1

#### 4.4 The Children's Needs and Diagnosis

The children had a range of medical conditions which required growth hormone treatment (GHT). These medical conditions included growth hormone deficiency (GHD), small for gestational age (SGA), Turner syndrome (TS), Russell-Silver syndrome (RSS) and Prader-Willi syndrome (PWS). One child was also diagnosed with autism.

The children had a range of medical needs and requirements of care. These varied from needing only GHT to needing other medical treatment grouped with GHT such as, additional medication and medical devices or machines (back brace, BIPAP and CPAP). One child needed a back brace to correct scoliosis, three children needed other medication besides GHT, and one child needed to use a CPAP at the time of data collection while another two had used it previously. One child at an earlier stage required assistance with feeding using percutaneous gastrostomy (PEG) and nasogastric (NG). A few children after their birth needed assistance with feeding via a nasogastric tube in the NICU.

These children required ongoing hospital visits and admissions to obtain a diagnosis. Some who were given another diagnosis prior reaching the diagnosis they held at the time of interview. Many of the children had undergone a range of surgical interventions before the period of data collection such as (tonsillectomy, adenoidectomy, circumcision, PEG placement, and grommets insertion). In addition, all children had experienced at least one of the GHT related medical testing such as growth hormone

stimulation test, magnetic resonance imaging (MRI), sleep test for Prader Willi syndrome (PWS) and blood tests. Some children had undergone other GHT non-related medical tests such as endoscopy, lumbar puncture, genetic testing.

The mothers gave very detailed descriptions of their children and their needs, which gave an initial reference point from which to explore their experiences of caring. Mothers often chose to talk from birth, the point of diagnosis, or from the first suspicion that something was wrong, to the present and beyond. They wanted to provide a biographical account to tell 'their story'. For mothers, noticing that something was different about their child was the beginning of their experiences of caring.

The mothers' experiences are bounded by time and their experiences can be noted on a trajectory starting from their lives before GHT to their future with GHT. These experiences are contextualised firstly within a period of their lives before GHT. This period involves the mothers noting a problem from as early as the prenatal stage and/or birth of the child to the mothers noting a problem some years after. It also includes mothers seeking medical attention and reaching a medical diagnosis which lead them to GHT. Secondly, there is a period of their lives where they explained the journey of starting GHT, where the mothers make the decision to start the treatment regimen supplemented with challenges of crossing this stage. Thirdly, adjusting to their lives with a child with GHT; this involves managing GHT within the home environment and the outside environment. The home environment includes the process of starting injections and the technicalities of administering the drug every night and being adherent with the regimen. The outside environment includes the child receiving GHT outside the home environment when normal routine was altered. It also involves travelling outside the home to any venue outside the physical walls of the home. This period also includes attending endocrine follow up appointments to monitor GHT. Fourthly, the future of the child in relation to his/her condition and GHT with much uncertainty attached.

The treatment pathway explained above was influenced by three major concepts that were 'normalisation', 'stigma' and 'uncertainty' and are central to the mothers' lived experiences caring for children receiving GHT. Their experiences were finally encapsulated into four key themes that elaborated the essences and meanings attached

to their experiences caring for their children receiving GHT. The four major themes emerging from the mothers' experiences are titled starting with a direct quote taken from the mothers' descriptions followed by a theme name - given by the researcher. The final themes were: (1) "It's the right thing to do" Striving for the security and the wellbeing of the child, (2) "Doubting yourself constantly" Constant uncertainty, (3) "But then you just get used to it I suppose" Adhering to GHT and lifestyle changes - the new normal, (4) "I hadn't been told anything about it" Information behaviour; looking for normality and certainty. The process of reaching the final four themes is visually demonstrated in Figure 1 and in Table 7 overleaf for further elaboration. Each theme will be discussed and explained with mothers' accounts in the following paragraphs.

Figure 1. The Process Leading to the Final Themes

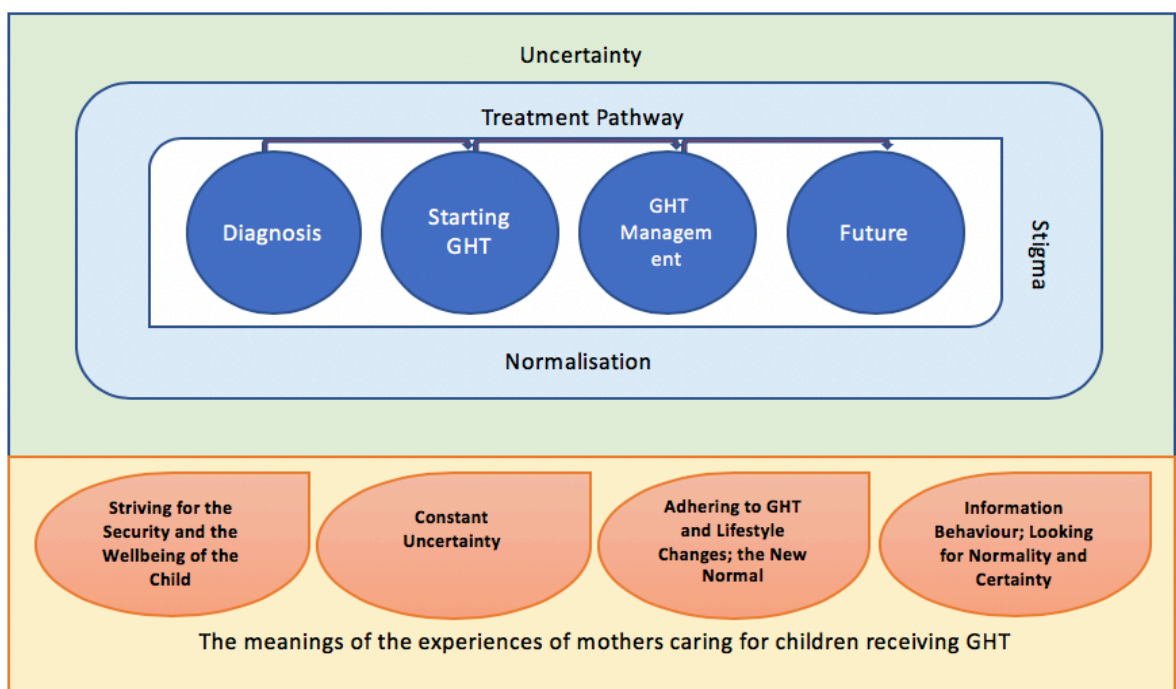


Table 7. Table Representation of the Study's Findings Detailing the Main Concepts, Themes and Subthemes

Concept	Themes (meaning)	Subthemes
Normalisation, stigma and uncertainty.	4.5 "It's the right thing to do" Striving for the Security and the Wellbeing of the Child	It is a mothers' duty to protect Advocating and seeking

		partnership Decision to start GHT /Normalising GHT Adapting and being sensitive to the child's needs The continuity of GHT as a source of wellbeing The future well-being of the child
Uncertainty, normalisation and stigma	4.6 "Doubting yourself constantly" Constant Uncertainty	4.6.1 Diagnostic uncertainty 4.6.2 Treatment uncertainty 4..6.3 Future uncertainty
Uncertainty, normalisation and stigma	4.7 "But then you just get used to it I suppose" Adhering to GHT and Lifestyle Changes - the new normal	Managing GHT injections 4.7.2 Lifestyle adjustments Attending endocrine follow up appointments Vigilance to treatment side effects and positive outcomes
Uncertainty, normalisation and stigma	4.8 "I hadn't been told anything about it" Information Behaviour; looking for normality and certainty	4.8.1 Information needs Seeking and sharing information 4.8.3 Need for support

#### **4.5 Theme 1: "It's the right thing to do" Striving for the Security and the Wellbeing of the Child**

Mothers expressed a great sense of parental duty toward protecting their children which emerged as one of their main concerns. This concept of being a 'good' parent could be detected in accounts spanning prenatal, infancy and childhood through to the potential future of their children as they approach adulthood. Prior to starting GHT, mothers were constantly trying to help their children maintain a normal life and were challenged to find ways to meet the challenges that were as a result of their children being different.

#### **4.5.1 It is a Mothers' Duty to Protect**

The notion of constantly trying to protect the child from harm was clear throughout their experiences. The strong sense of protection towards the children was noted as early as the prenatal stage. One mother alerted the midwife that she wasn't growing as big as her previous pregnancies. She knew that there was something not quite right with the size of her foetus after comparing with her previous pregnancies and to her, that was a deviation from what is normal and therefore sought medical attention.

As [Sam] was my number three, I felt very early on that he was going to be a small baby. I alerted the midwives probably about 22 weeks. I was putting clothes on that I was wearing with the other two and I wasn't filling them. I just felt small (Steph).

As children grew, many of the mothers began to observe how small or thin their children were, and they constantly compared their children with other children their age in the society, in an attempt to determine what was normal. Many mothers did so by comparing their affected child with his/her siblings. Others constantly compared with children in the extended family, friends and class mates within the same age group.

The first few years of [Frank's] life, his height never worried me. He was always small it only became apparent when he started school, how small he was. That is when I began to worry about him (Francis's diary).

For some, they constantly compared after passing a stage of thinking that their child was small due to genetic factors. Some parents reassured themselves as they were reminded of how small they were when they were that young, however, when the size issue was no longer ignorable, parents looked for answers to why their children were not growing normally. Mothers noted the differences in their children being smaller than others and this was also obvious to others in the community.

There was a history in our family of very petite. I have loads of petite cousins. I've about 40 cousins and about 25, 26 of them are female and they're very petite (Mary).

Some mothers expressed great concern about their child's underweight/thinness more than their short stature. Being underweight may have caused the mother great stress as it is a mother's instinct to nurture her children. For these mothers, seeing their children too thin was hard and may have questioned their nurturing duties 'being a good mother' towards their children, especially when it was apparent to others how thin these children were.

I think actually, all along it was his weight bothered me more. He was so thin. There was nothing of him there really, you could put your hand around his legs, your fingers nearly and I suppose that's the part that always bothered me. Not so much the height, but it was his whole physique (Sandra).

Now if you saw him in his swimming togs you'd say, "There's something wrong with that child," but you wouldn't when he's clothed. He looks the same as everyone else, he's just slight, but you wouldn't see how slight he is until he's in his swimming togs and then you'd think that you don't feed him. He looks like one of those kids that's neglected (Emma).

A strong desire to protect their child from social exclusion and promote acceptance in order to protect the child's self-esteem was obvious. Mothers expressed a strong desire to protect their children from hurtful comments. Many mothers expressed how comments from others (for example; teachers, lollypop lady, and random people) in their community regarding their child's size caused frustration at times. They expressed feeling annoyed, vigilant and over protective to not let their child hear such comments as this could have made their child more aware of the difference 'being small' which they may have already noted themselves at that stage.

Shortly before [Ryan] started school a teacher in school (he would have been 4 when this comment was made) "Ah look at him he looks like he should be in a buggy with a dody in his mouth"! Then the shock when I told her his age and that I hoped he hadn't heard her say that. She didn't mean it in a mean way but it was still hard to hear (Rose's diary).

As he got older other boys and girls commented about his height. This always annoyed me especially older people saying things. In my mind they should have



more sense. I remember once I told the person that he wasn't deaf. I knew once they went by he would ask why did they say that he was very small. I would always say to him that they said he was getting tall. I felt very protective (Francis).

Prior to starting GHT many children were already aware of being the smallest in class or even in the whole school and this appeared to have had a great psychological toll on them. The child's height and size made it more difficult for the child to pursue normal activities such as swimming and being assumed not competent enough. Mothers expressed distress about the lack of understanding and how judgemental people were because of their child's size. The psychological aspect of trying to maintain the child's self-esteem and confidence was a huge worry for most of the parents and trying to protect the child was very noticeable.

He totally understands why and he understands that he's smaller. He was more aware of it than I am because he's in a class and he's the smallest in the class and other kids are, he was always aware that his other friends are much bigger than him... you're aware that you're different and you're aware that you're smaller than your other friends, he didn't have much energy, he was tired (Liz).

[Ryan's] swimming lessons: He was constantly moved down to the younger group of children who couldn't swim even though Ryan was able to swim by then it was just presumed that he couldn't because of his size and it made me realise how much people judge someone's ability based on their height/size. It really knocked his confidence and I felt so angry and frustrated about it (Rose's diary).

Some mothers held their children back a year in school so that they could blend in more easily with their peers in class and avoid standing out as much due to their small size. Mothers felt that at least they would be the oldest in the class and this could help them make up for being the smallest. It was noted especially for the boys in the study that the mothers put the child in a mixed class (boys and girls) which made it easier for the child (boy) to blend in. Another mother expressed that when her son started a higher level and moved to an all-boys school, it was then very obvious that he was very small which highlighted the difference to the previous mixed classes, where he would have blended

in fine. It was apparent that these mothers were trying to protect their children from feeling different and tried to minimise the social exclusion as much as possible with the goal of protecting their child's self-esteem.

But the kids in his class used to tease him about being small, definitely. Definitely... so in my head I thought, if I hold him back then he'll be the oldest, he may be the smallest, but at least he'll have then that he's the oldest (Loretta).

Surprisingly, I think he just came out of the womb with confidence. So he thinks he's the best in the class at everything. I don't think he notices and I think that's a good thing about having a mixed class. I think he's the eldest in the class. So some of the boys are taller than him, but he doesn't cop that they're a year younger (Emma).

Some children, however, had started to isolate themselves from doing activities that normally children would do because they were aware of the difference and as a way of avoiding hurtful comments and rejection from other children. Mothers noted when their child avoided playing football in a football team, attended birthday parties, joining a choir and other activities because of their small size and being assumed to be younger (juvenile). Some mothers tried to help them to overcome their fears and to fit in as much as possible by signing their child to a football club or attending a birthday party with the child.

He won't play football. He won't actually go and play football with the others. There's a local club and they do training every week and I've signed him up for the last three years. He's a great little footballer, he plays it out the front with his buddies, but he won't go to the club to play it and I think it's because, I don't know whether they're bigger than him or what, but he just won't do that at all. A little bit of low self-esteem I would say (Loretta).

Trying to reassure the child when they questioned and sought answers from their mothers about why they were different was challenging. Mothers tried to answer these sorts of questions while being mindful of protecting their child's self-esteem. Trying to normalise the size issue or downplay the concern to reassure the child were some of the methods used. Many mothers used genetic factors to explain to their children and to

rationalise the size difference. The sense of acceptance and normalising the issue was apparent in these experiences which also reflected the parental duty of constantly trying to protect their children from harm.

Then he started to ask why he was much smaller than the other boys his age. My answer was always “Did you see the height of their fathers”. You are like us so don’t worry... I used to say to [Frank] but their daddies are all very tall look at their daddy’s and look at your daddy you know you would be trying to explain to him that’s just the way it is but not knowing then that it was something else you know what I mean (Francis).

Due to the fragile physique (short and thin) of the children being a main concern, some mothers expressed great apprehension about their children’s physical safety. The children were at greater risk of being physically injured by other children around them. Some injuries occurred while the children were playing contact sports (for example football) such as having a chipped ankle, head bang and a pulled groin. Another concern from a mother was her child’s risk of falling down the stairs in school due to the heaviness of his school bag and that the child was not physically able to carry it due to his size. This led to the mother accompanying the child to school and assisting him.

The school had two small classes and there was another girl in [Megan’s] class that had extreme special needs. She was about twice, three times, no, four times the size of Megan, both in height and in weight and even with her being in the same class as Megan was a safety hazard (Mary).

His school bag was too big for him and heavy so I would be trying to go in to the school with him and get him up the stairs so his bag would be kind of falling back down the stairs you know what I mean (Francis).

Under this category, the mothers’ sense of protection was clearly detected through their experiences. As early as the prenatal stage, mothers noted differences that could potentially affect their child’s health. Then as their children developed, mothers tried to maintain and protect their child’s physical and emotional wellbeing. This leads to the next category discussing mothers advocating and seeking partnership in order to meet their children’s medical needs.

#### 4.5.2 Advocating and Seeking Partnership

All mothers were frequent users of the health care services many years before starting GHT and continued to use them. They had visited the hospitals quite frequently looking for answers to why their child was not growing well or causes of undefined manifestations. Many were left with unanswered questions which caused considerable frustrations along the journey of trying to find answers to concerns. Some mothers reached a diagnosis not long after their children were born as manifestations of their condition 'syndrome' were well-defined and enough for health care providers to investigate further.

So from there he went straight to the neonatal care in ICU and then they did a whole load of tests and then on day five we were moved, well, he was moved to [X hospital] actually, straightaway and then I went of course, with him and stayed there. So we were there for the next ten days approximately, eight to ten day. Then on day 15 he was diagnosed. So they did a whole load of tests, lumbar puncture the whole shebang. So he was diagnosed, they did the genetic testing and all the rest and he was diagnosed with Prader-Willi syndrome (Michelle).

However, most mothers had a not so straight-forward experience reaching their child's diagnosis as manifestations were not as well-defined resulting in years of follow-ups and monitoring with no definite answers. Children had presented with many indications for mothers to seek medical attention, including failing to grow (height and weight) to other health concerns such as feeding issues, blackouts, sleep apnoea, gastrointestinal problems, vision problems, feeding problems, milestone delays and autism.

The process of finding a diagnosis for the children who had not been diagnosed shortly after birth, influenced mothers' experiences as they persisted in seeking the cause for the lack of growth and other health manifestations noted in their children. They felt responsible for making health care decisions with the best interest of their child in mind. They tried either actively or passively to advocate to find answers and reach a final diagnosis for their children. Some mothers were passive about trying to find answers as to why their child did not grow well or were delayed catching up developmental

milestones. They felt comfortable and confident to leave it in the hands of the health care providers to solve the issue and trusted their decisions regarding their child's health. However, most mothers appeared to play a more active role in seeking answers and finding solutions. This will be discussed in more detail in Theme 4 section 4.8.2.

Noting the lack of growth was apparent in mothers' experiences that involved being delayed diagnosed well after the birth of the child - in the case of GHD. Most mothers brought their concerns to the professionals' attention. Because they seemed worried about the lack of growth manifested at the time, they sought medical attention to tackle the problem and find answers to why their children were not growing normally. When they did draw the attention of health care providers to the issue, some mothers were given false reassurance that their children were small but would soon catch up.

I kept pointing it out to her, but because he was low on the growth scale, it was he's a little bit on the small side and it wasn't until he was four... and I said, "He's small, he isn't growing. His feet don't grow, his nails don't grow. Nothing grows. He never has a haircut." and she (paediatrician) said, "No that's perfectly normal." (Loran).

Mothers were advised to adopt a 'wait and see' position and to be held up in a 'wait and see' approach proved to be very frustrating for mothers. Helplessly waiting for long periods of time to attend follow-up appointments and witnessing their child failing to grow or deteriorating in health in between appointments was extremely challenging and stressful. Some mothers were given a wrong diagnosis e.g. abdominal migraine, constitutional short stature, autism, and cerebral palsy. Others questioned if their children were correctly measured at the time. Some felt completely dismissed by the healthcare provider when they brought up their concerns, which at times made them feel 'stupid' and 'over the top'. This led them to find other ways to get their questions answered such as changing their healthcare provider.

Now I don't know if that was relevant later, but for me as a mother it is relevant. Even though I have told that to the medical profession and none of them thought it was relevant. So that was fine, we got over that... He had speech delays when he was two, three, four, five and he had to get a lot of speech therapy which to me is connected but none of the professionals agreed that they were

connected... We just began noticing that he was very small... I asked the doctor (GP) and he would say yes okay he is small but he is healthy, come back in six months and see how we go. Every time I come back he says it is kind of borderline. He was five per centile, four per centile and the doctor said that three and half per cent was his definition of what was a problem and what wasn't a problem. So, at one visit he was finally down to three and a half per centile and then he referred us to a private paediatrician (Bridget).

Many mothers had to fight to obtain their child's diagnosis and were forced to take an active rather than passive role in their child's medical care. They felt obligated to advocate for their child and therefore challenged their child's health care providers' decisions. They felt that if they had not done so, that their children would have not been diagnosed or commenced on GHT. Mothers tried to influence the child's care by arguing for what they thought to be in the child's best interest and in accordance with the child's needs.

When he was a baby there was months, until he was about five or six months, I felt I was being fobbed off,... sending him home and then he was really sick again and we were back in hospital... at the time I felt I hadn't done enough, I didn't speak up enough... I was a little bit meek and I don't know, did we do the best thing for him... we were months here in [X hospital] asking for a gastroscopy and they wouldn't do it and I suppose I had that fight in me because I had regrets back then when he was a baby... that probably fuelled it a little bit because I was disappointed with how I managed him when he was a baby... I just let myself be pushed from pillar to post without anyone looking for proper answers (Sandra).

The first encounter with an endocrinologist... was a very negative experience. I was told that it's likely just constitutional growth delay... but also that there were children a lot smaller than him attending at the clinic made me feel so stupid for even getting a referral. It was as much as to say what have I got to complain about when he is still on the growth chart. It was a very stressful experience... At his next follow up 6 month later to check his growth again, I met with the same consultant just by chance and at this appointment he was moving towards a decision to discharge [Ryan] from their care. This was so

difficult as I knew in my heart there was a problem and to know that he wouldn't be helped was so difficult. I had to fight for another appointment and was made to feel so over the top for asking for this... Thank goodness I did push for another appointment as it was at his next appointment a year later that I met with a different consultant and had a completely different experience. He decided at that appointment that we need to do stimulation test for GHD (as Ryan's growth rate had dropped significantly as he had fallen very low on growth chart). He was so kind and dealt with all in such a different manner. To feel like I was actually being listen to for the first time was such a relief. Had I just accepted what the first consultant was edging towards, Ryan would no longer be in the system and would not have been diagnosed (Rose's diary).

It was apparent that mothers felt obligated to advocate for their children's medical needs and tried to play a significant role in the attempt to find a diagnosis and seek answers to the lack of growth or other health manifestations witnessed on their children. After reaching a final diagnosis, mothers were faced with the decision to start GHT which leads to the following category.

#### **4.5.3 Decision to Start GHT /Normalising GHT**

For most mothers, it was a 'no choice situation' to start GHT. Test results from the stimulation test and MRI confirmed for many the need for GH replacement, while, others just felt it was the right thing to do to give their children the best outlook to their future by boosting their height and wellbeing. Motives for starting the treatment varied for each mother depending on the child's condition, mother's personality traits and background knowledge. For the children who were diagnosed with Prader-Willi syndrome, their mothers were definitive about starting GHT as soon as possible because they were very aware and very confident of the benefits it held for the condition. So, they were eager to start it as early as possible.

So we contacted some consultants in England and the United States and they did agree that it would be beneficial to get him on it as soon as possible... My own reassurance I suppose is having gone to all of the various meetings of the association where there are a lot of older people with Prader-Willi syndrome

present, that they seem to be either, just developmentally they don't seem to be as strong or there's a notable difference physically between the older people with Prader-Willi syndrome who wouldn't have had the benefit of growth hormone and the younger generation, which as far as I can see, predominantly have been on it and to be honest, you wouldn't notice the difference between them and typical children (Louise).

So I suppose we were introduced to the idea of growth hormone and the advantages and how it would help. I guess it just seemed to be the right thing to do. I don't know if we ever seriously thought about not doing it, but a really good friend of ours is the consultant and we spoke to him about it and he was saying. "Look, it's definitely the way to go." So I guess we never thought that we wouldn't do it really. I suppose we didn't do a huge amount of investigation into it I suppose, but we felt for [Mark's] sake that we had to do this to help him have any kind of proper life (Michelle).

Some mothers felt they had no choice as their children were GHD and needed the GHT replacement. Other parents of children who were not deficient in GH but were generally small, felt that GHT would give their children a chance to better their expected adult height and needed to take advantage of the narrow window of opportunity to do so. They all eventually followed their healthcare providers' advice (endocrinologist) to start GHT and felt it was the best thing to do at that time to help their children.

I felt that for [Leo's] sake the decision was taken. I made the decision, but it wasn't really a decision that I had many options on because when they showed me the results of the stim test (stimulation test), he was never going to grow and he was going to be forever the size of a two-year-old (Loran).

I felt it was the right thing to do for him... it was only a narrow window in which to start and I said I think he'll appreciate it when he's older (Emma).

Starting GHT was sometimes worrying and confusing for the mothers' especially around the effectiveness of the treatment in helping their child improve and consequently boost their confidence and self-esteem which will be discussed in more detail in Theme 2 section 4.6.2. However, the treatment was going to be a daily reminder of the child's difference i.e. 'small size' which needed fixing. Therefore, the



treatment at times was like a double-edged sword and potentially raised the child's self-consciousness of their difference.

I suppose I felt because [Nancy] was small I didn't want to have to say to her it's something that needs to be fixed, but I didn't want her to think it's not okay to be small. So I struggled with how to explain... I said to the doctor-, she said, "What's your biggest concern?" and I said, "My biggest concern is how to tell my daughter there's something wrong with her that we need to fix and it might not work and how do I explain to her then that we tried to fix it-, but it's something that we tried to fix because it's not okay and we can't fix it and it's actually okay after all (Nora).

I just hated that this was kind of, not that it (GHT) was going to define him but was going to be such a daily reminder of being small (Sandra).

Mothers were trying to help 'fix' what they thought was not normal due to societal expectations and were looking for ways to normalise the difference in size. Starting GHT was a means to help their children fit in more into society and to reduce the potentiality of social exclusion. But at the same time, being aware that daily GHT injections could encourage the stigma and reduce the child's acceptance of him/her being small and in turn psychologically harm them. Consequently, they struggled with an internal ethical dilemma when making the decision to start GHT. They questioned if accepting the treatment meant they were not accepting of their child's appearance and in turn, inflaming the stigma attached. However, this might have not been the case for other mothers who had a different opinion on starting GHT. Some mothers expressed how fathers sometimes had an opposing view which made the decision-making process even more difficult. But eventually, all children started the treatment after both parents agreed together what would be best for their child.

The most traumatic thing of the whole experience was-, I don't know how to articulate it. Just that if he'd a broken leg, of course you'd go to the doctor and get a cast for it, but if somebody's eyes were blue and you'd rather they'd be brown, you wouldn't go to the doctor about that and in as much as that's a continuum where his level of severity was and whether it was appropriate to

follow up on it and whether it wasn't. That was the hardest thing. Especially because I and my husband felt differently about it. That was the hardest thing I would say (Bridget).

Mothers appeared to fear social exclusion because of the stigma attached to being different and worried that their children would not be able to fit in or be accepted (normalised) by the society which may have influenced their decision to start their child on GHT.

I could be out in the public and people move seats in a coffee shop. They don't want to sit beside a couple of people with Down's syndrome or whatever. So I know people can be cruel. It's not very nice (Berny).

The notion of stigma being attached to the child was noted clearly when it came to the mother and father deciding to start the treatment for their child. Mothers expressed the idea of a boy being small was hard to accept because society expects boys/men to be big and tall. They expressed that this reality was sad but true and worried about the future of their sons in that sense. They also expressed that they might not have chosen to start the child on GHT if the child was of the opposite gender (girl). One mother expressed that a girl could wear high heels and the problem would simply be solved. Another mother conveyed that the notion or stigma of males needing to be big and strong was not only witnessed in society but supported by some healthcare providers. She noted that it influences the healthcare provider's decision-making and called it being 'sexist'. Another mother who herself was very small as a child and has a brother who was treated with GHT as GHD runs in her family, expressed the strain of getting her brother started on the treatment as she was ignored because she was a 'girl'.

I brought his twin up with me... I brought her to [endocrinologist] and I would say she's small, but she was on the chart. She was on the second. So they said, "Well she's on the chart and girls are okay." Nobody sees her anymore. Whereas, he's going all the time. Yes, it is a bit sexist definitely and we've laughed at that all the time saying girls can be smaller (Loretta).

He's my older brother (who needed GHT for GHD) and it was like, "Oh she's a girl it's okay she's petite. It's fine." It seems to be a bigger issue if it's a boy. I don't understand that (Liz).

He predicts if we keep going on growth hormone, he could reach five ten, when he had said five four originally. If it was a girl I wouldn't have started therapy I don't think (Emma).

He [geneticist] brought the news to me (son diagnosed with Russel Silver syndrome) and my immediate reaction, obviously I was processing all this information. He mentioned dwarfism and all this sort of thing and he was saying-, my immediate reaction was, "How short is he going to be?" That was the first thing I said and he said, "Well he might be about five foot four." I went, "God!" like this and he said, "What's wrong with a man being five foot four? Napoleon was five foot four." and I looked at this guy and he was stood there talking to me and he was down here on me. He was about five foot four. He's obviously done very well, but that was my immediate reaction and I would say that's most people's reaction and he was very defensive. He had the Napoleon counter play, but I'm sure if you asked him in the pub one time if he preferred to be taller, I'm sure he would prefer to be taller. So that would be my only thing, I just want to try and maximise this opportunity (Steph).

It was noted that mothers tried to put themselves in their child's shoes when making the decision to start GHT and many times they felt accountable if they declined the treatment. They worried that refusing GHT would be questioned at a later stage by the child as an adult. They also thought by accepting GHT, they had opened a door of opportunity for their children. They didn't want any future regrets for not starting GHT. To them GHT was a chance and an opportunity to help their children grow and as a parent they had the duty to do what is best for their children.

Probably, for his sake yes. I thought would I want to be five foot one if I had a chance of being five foot five? I'd prefer to be five foot five in today's society. Irish are much taller now (Emma).

You're taking a chance. Yes, but at least when she's 16 and she's grown up she can't come back to us and go, "Why am I four foot eight or whatever and you could have helped me." At least we can say we tried. We tried to help her because it's hard I am sure being very short in society today. People are so judgemental and they're horrible. So it's just to give her that. At least we did

something... I just put myself in her position really and I thought, if I was a 16-year-old girl and I knew my parents didn't give me this treatment and I could have been taller, I would have been quite angry with them I suppose. So I just put myself in her shoes and thought how I'd feel. I think [child's father] did the same as well (Berny).

It was clear that mothers wanted to try to normalise the child's condition and find ways to help the child fit in and boost their confidence and self-esteem, by making them not too different from the norm in the society or even in the family itself.

He (her child on GHT) has two other brothers so I would always hate for him to be way behind them I mean he will always be shorter than them there is no doubt about that but just for his own confidence his own self esteem... He's starting secondary school in September so, I'm really glad that it happened before he started in secondary school because at least he's got the security of-, okay, he is going to be the smallest boy when he starts there, but at least he's the security of knowing that maybe he won't be the smallest boy when he's leaving there (Sandra).

While making the decision to start GHT, parents had sought others (family, friends and professionals) opinions and thoughts around GHT to help guide them in the decision-making process; and this will also be discussed in Theme 4. However, sometimes parents who sought advice made the process more confusing and difficult for them. The decision to start GHT was to benefit their child despite others not approving or witnessing the need for this treatment.

So we had discussed it quite a lot and discussed it with our own families and there was a lot of mixed feelings about it, but we both felt that it would benefit her in the long run and we'd give it a go and if it didn't work out, then maybe after six months or a year that we'd stop doing them, but there was a lot of people suggesting that she doesn't need them. There's nothing wrong with being small and all that. It was hard, but we just said we're the parents and we're going to decide... There was family, sister, father, your mum. There was people, but I think everyone was, "Ah, she doesn't really need." and people say, "If that was my daughter I wouldn't give it to her." but it's very hard when it's you have

to make that decision and you have to think the benefits (Berny).

So when I went back in, we still hadn't fully decided if we were going to do it or not and we didn't fall out about it, but my husband basically thought we shouldn't do it and I thought we should do it... He thought [Bill] is fine, he's in good form and running around and he's got friends and he's a normal, fine little boy. Why do such a big medical intervention to make him into the six footer, does it matter? (Bridget).

The degree of wanting to start GHT varied amongst mothers. Some were happy with starting when it was permitted by the healthcare provider to start according to the UK national guidelines (NICE 2012). Others were frustrated for not starting sooner; this will also be mentioned under Theme 2 section 4.6.2. Some also expressed their willingness to seek the GHT elsewhere if they had been denied the treatment in the Republic of Ireland (ROI). They would overcome any obstacles to achieve that goal.

If we didn't get approval for growth hormone in this country, we would have gone elsewhere. I just felt that it was the right thing for him. Everything I'd read, everything about the [support organisation], but I was also mindful that it was very expensive and we'd have to put money aside because we felt that if he didn't get it on the health service here that we would have to go and try and fund it outside of Ireland (Steph).

It was noted that many of the mothers felt particularly frustrated with the timing of their child starting GHT. Some felt that it was unreasonably delayed and had wished to have started much earlier. Many were very aware of the national guidelines for starting GHT in Ireland and elsewhere in the world (USA and UK) and they constantly compared guidelines. They generally felt let down by the Irish system starting much later than the USA. This delay made them feel frustrated and angry that they had to wait for no reason because they didn't understand why it was licensed in other countries to start much earlier than in Ireland. They felt that their children may have missed out on the opportunity to start sooner and gain the benefits that comes with that, which will also be discussed in Theme 2 section 4.5.2.

We were told anecdotally from other parents that the Irish health-, the HSE or the Irish system wouldn't approve growth hormone for children under the age of

two and this seemed to be completely contrary to what international literature was suggesting, which said that the benefits-, the sooner that the child went on growth hormone, the greater the benefits... The only thing that annoys me is that the Irish medical system won't even consider putting a child on because my impression from other parents who have gone through the conventional system is that they won't even entertain it until two years of age and I don't know why... I just think that's criminal... I think it makes such a difference from the parent's point of view, peace of mind. I know parents who have gone to the States. I know parents who have gone to England and I don't see that that should be necessary because if it's accepted everywhere else in Europe and the States, why should Ireland be different? I think that's really unfair and I think the demographics here and the licensing of growth hormone should be completely revisited and I get really, really annoyed at the delay in administering growth hormone (Louise).

Louise also emphasised in her diary:

The initial anxiety at the possibility delay that there might be getting him on it and the long-term impact that this may have had on his development (Louise's diary).

Not being happy with the timing of starting GHT was due to not only the national guidelines for starting GHT but it was also due to other frustrating reasons mothers faced in this journey. Some delays were due to health care providers delay in performing necessary tests and surgical procedures prior to starting GHT (i.e. sleep apnoea test, tonsillectomy, MRI, GHT stimulation test and bone age x-ray). However, very long waiting periods to obtain tests, receive results or attend appointments was very frustrating to mothers. This was especially noted when they were eager to start GHT as early as possible (in cases of Prader-Willi syndrome) so that their child could gain the benefits of the treatment as soon as possible.

The only thing I can think of that wasn't straight forward in relation to growth hormone was actually getting him started on it... We knew from when he was probably six months old that we wanted him to start... there were numerous delays due to waiting lists for sleep apnoea sleep studies and then he was

waiting for ages to get his tonsils out and that was really frustrating waiting to get him started... So that was frustrating (Joan).

With all that said, many mothers were comfortable with the timing of their children starting GHT.

I'm just glad that something was done that we didn't leave it until he was about 16 and then go, "Jesus." we noticed how small he is... So it was addressed when it needed to be addressed... he was put on it at the right age (Sandra).

As the benefits of GHT were normally considered when making the decision to start GHT, the potential harmful side effects of the drug were also considered by mothers at that stage. They expressed feeling very concerned about the safety of the drug and potentially harming their child; this will be discussed in more detail in Theme 2 section. 4.6.2.

If it was something like if you get an injection every year, but because it was every day and I was very unsure about the side effects and as I said, I'd looked it up on the internet and it was really bad side effects that you can get and I was frightened that something would happen to her I suppose (Berny).

Not only the potential side effects of the treatment bothered most mothers, some felt it was a gamble to start the treatment as potential benefits were not completely guaranteed; this issue is emphasised more in Theme 2 section 4.6.2. Starting their children on a quite invasive treatment such as GHT could psychologically harm their children with no significant benefits gained in the long term. On this note, many mothers felt obligated to risk it, take the chance and try the treatment for a period of time despite of their uncertainties. They were determined that if the treatment proved beneficial to the child, then they would be happy to continue the treatment. If proved otherwise, then they would be very quick to stop.

How do I explain to her then that we tried to fix it-, but it's something that we tried to fix because it's not okay and we can't fix it and it's actually okay after all (Nora).

Involving the child in the decision process was common. Mothers described involving

the child when it came to decide on starting the treatment, especially when the child was old enough to understand, and when they didn't, they spoke about taking their child's best interest always into consideration. Parents made sure to explain to their children what GHT was for and why they needed to start GHT as parents were their children's main source of information. They tried to reassure their children that GHT was like every other treatment and taking it was not going to make them any more different (normalising). They always conveyed to the child the need for the treatment as beneficial and never as harmful so that the child would start and continue to accept the GHT.

He's old enough to be involved. So we explained to him why... He totally understands why and he understands that he's smaller... We don't give kids enough credit, I think they can accept and understand a lot more than we give them credit for. I've even tried to explain to him what the pituitary gland was, shown him pictures and he kind of gets it. I think you just have to give them the information and I think they can understand and accept a lot more, we don't need to shelter them as much (Liz).

I said look you have two choices you can be small and do nothing about it or you can be small and do something about it so... I had to explain to him then, I was saying, "It's not so much your size, but it's puberty and you're not going to be getting the beard, you're not going to be getting the hairy legs way later than other kids." He still will be way later, "But if you don't go on the treatment, it may not happen at all." So he knew all that and he never said he didn't want to go on it. He was always game on for it. I think he just saw it as being, if something can be done let's just do it (Sandra).

Mothers strongly invested hope in the power of GHT to enable their child to fit in by 'normalising' their function and appearance. It was hoped that GHT would aid growth, normal development as well as enhance their child's emotional well-being in the longer term. Mothers often claimed to understand their child's inner emotions, making direct connections between a child's negative self-image and the power of GHT to enhance their confidence and sense of well-being. There was an assumption that GHT which aimed to 'normalise' a child's function/appearance and enhance their self-esteem would enable them to more easily fit in with society. It was comforting for mothers that such



'normalising' could provide their children with a degree of protection from anticipated social stigma and this belief sustained them during the treatment course. Mothers stressed their belief that the end results justified the costs involved and that they had done the 'right' thing in sanctioning GHT for their child. The sense of protection and striving for the wellbeing of the child continues into the following category discussing mothers adapting and being sensitive to their children's needs.

#### **4.5.4 Adapting and Being Sensitive to the Child's Needs**

The mothers expressed how they tried to adapt themselves to reduce fear or concerns for the child which in turn gave the child a sense of control. All the mothers described how well their children adapted to the GHT once they started. Although, the first couple of weeks of commencing proved to be difficult for most, they soon adapted and adjusted to the treatment regimen. However, occasionally children questioned the necessity of the treatment which was hard for mothers to hear as this only proved the extent of burden the treatment had on their children. The children occasionally refused to receive the injection especially when they were tired as this was commonly reported by the mothers. When this happened, instead of threatening the child, the mothers tried to motivate him or her to take GHT. Mothers adapted to the child's wishes and mostly handed full responsibility to the child to make these decisions, however, to counteract their behaviour, mothers reminded the children of the need for the GHT, why they started in the first place and the consequences of not receiving it. Some mothers used downwards comparison to help their child cope with the treatment burden as a way of normalising the treatment; which will be noted later in Theme 3 section 4.7.1.

I suppose, put things into perspective, she may give out at times, "Why do I have to do this?" and I would put it into perspective by saying, "[Megan], everyone has something going on. Every family has something going on. This is small in the overall scheme of things." and as she's got a bit older she can see that (Mary).

I suppose the advice we had been given, behaviourally, for handling Prader-Willi syndrome is that attack isn't always the best way forward. That it's much better to keep an even keel and let the child come around to it himself.

Otherwise, knowing that he'd have a lifelong or certainly a long time on growth hormone, we didn't want to start setting an unpleasant precedent for him. We wanted it to be part of his daily routine. So he is fine with it now and it really was just, not forcing it on him, talking to him. Distraction, a lot of distraction and it was quite gentle and letting him come to us. That was the main thing, rather than chasing him around with it (Louise).

You do have the very odd time and it would only be every couple of months, he would go, "I wish I didn't have any growth problems." and you just feel awful, you feel so bad for him. I just wish he didn't have it. You explain to him, "This is to help you and it's to help you to be able to grow." and all that kind of thing and he gets it, but you still feel so bad for him. I just wish he didn't have to have it (Rose).

Mothers tried to adapt to how and when their children wanted to receive the GHT. When they wanted the injection in a certain body area, their wishes were respected despite the healthcare provider's recommending varying the injectable areas. The mothers' knowledge of their own child, helped them to adapt to minimise the occurrence of fear by choosing the right time for GHT injection when the child was not too tired and upset.

The endocrinology nurse really wants him to start using it independently and has been saying that now for a few follow ups in a row and even, she wants him either to screw on the top or put his hand here or just something and he just won't. He hates it and he just won't and he only wants it in the bottom, even though she wants us to vary between the legs, the tummy and the bottom, but he prefers the bottom I suppose because it's fatter and so, we just try and vary it around the bottom. We always do it so, we're not fully following her instructions from that point of view, but the hard thing is I suppose because it's in the evening. So you're at your most tired, he's at his most tired. You just want to get it done and not upset him (Bridget).

The mothers also gave descriptions of using distraction techniques, hiding the needle, administering a topical anaesthetic on the skin (Emla cream) beforehand, using buzzy bee - a vibrating bee with icepack wings that decreases sharp pain when placed

upstream while injecting, warming up the medication to room temperature, administering while child was asleep and adjusting the size of the needle to the smallest gauge they could find in order to alleviate discomfort and fear related to injection pain. Mothers self-discovered ways to help facilitate the injecting experience for their children. Some also used professional recommendations from psychologists and nurses on methods to reduce injection phobia; distraction and reword charts. They also described trying to offer some positive experiences in relation to the child accepting to receive the injection or visiting the hospital for follow ups. This was done as rewards, offered after the treatment or as incentives mentioned in advance.

So I've actually changed my procedure now and I wait until he's asleep and I go up to bed and I'll do the jab then because it's just easier for all of us (Michelle).

They recommended blowing bubbles, giving her treats like chocolate. That kind of thing or doing breathing exercises. She was doing all of it and blowing her bubbles and doing all that and then she'd still be screaming when the injection came. So it didn't seem to calm her really. It was like a game and then when she saw it coming she'd still be the same. So I just tried my own thing with her, singing silly songs. I'd just hold her and sing a silly song. She still doesn't like getting it done, but she's not screaming she's not crying (Berny).

Some mothers broke the normal routine of administering GHT to adjust to normal life activities such as the child going on sleep-overs. Especially if the child was not self-administering the injection, they would skip a dose, give it earlier than usual before they leave home or even accompany the child for GHT administration purposes; which will be discussed in Theme 3 section 4.7.1. In making these arrangements, the mothers considered the risk of distress, pain, and social isolation and balanced this against the benefits to the child's emotional well-being.

Now the fact that we've been told by the nurse now that she can skip a day, you can skip a night. We skipped our first night only about a month ago when she had a sleepover because she was going to her friend's house at 3 o'clock in the day and it would have been awkward for me to come in and she didn't want it in the fridge, she doesn't want her friends knowing about it (Mary).

Mothers also expressed that to not frighten the child while administering GHT

injections or during medical tests, the mothers tried to keep their feelings hidden without showing their fear or sadness to their children. This would have been more apparent in the first few weeks or months of commencing GHT. Despite the mother's fear of needles, worry about hurting her child, or disappointment about the experience, they hid their emotions and got on with it for the sake of the child; this will also be noted in Theme 3 section 4.7.1.

I didn't want to be thinking every evening and I could feel myself getting tense beforehand. Your children can pick up on those things. So he could probably sense that I was being a bit nervous about it and then maybe then I was. Maybe I was jabbing it in a bit too hard because I was nervous or something and that was only for about a week and then I thought this isn't a long-term solution, I'm not going through this argument every night. In general he's fantastic, he's a great little boy and we get on really, really well, he's brilliant and he's a very happy little fellow. In general, I think his pain threshold is quite high actually. So I was like, "No, I need to find a solution (inject while the child is sleeping)" (Michelle).

Whereas, it's me that's putting on the brave face all the time and even she comes to me and she says-, she senses that I don't like giving blood, but I've got to be strong and show her because she's giving so regularly (Mary).

Although mothers usually explained to their children why GHT injections had to be performed and what could result if the treatment regimen was not followed, they were sometimes challenged with their children refusing injections especially in the early days of the treatment. Sometimes mothers and fathers were emotionally challenged as they had to forcibly inject GHT by physically restraining the child. In addition, guiltning them into taking their injections also occurred. These means were used when mothers felt that there were no alternatives than to be assertive and strong. They described themselves as doing everything they could to make it easier for the child but also realising that the child had to receive the GHT for their own good.

Well, one or two people did ask me, "Jesus." or they'd say when they heard when he had to be on it until he was 17, "Holy god, every day." and I'd say, "Yes." but to be honest, the reaction I got was, "Jesus, fair play to you, I don't

know if I'd do that." and my answer would be, "Well, it's easier when you're a nurse." because that stuff wouldn't phase me at all, having to hold him down to do injections. I know they'll get over it and it's the right thing for them in the long-run, but I imagine if I wasn't a nurse, I think it could be different. I probably would have caved in those first three weeks. I probably would have caved and said, "No, I'm not doing this." because there was a couple of friends that came to help me were traumatised. They were like, "Jesus, how do you do that?" I was upset myself doing it, but I didn't want [Even] to see me upset. I hated holding him down and doing it (Emma).

We had a late night on Saturday night. [Leon] was exhausted on Sunday when it came to injection time, he was just so tired he cried and cried, he just didn't want the injection. I felt extremely guilty trying to force him to take it. I tried to bribe him, cajole him but nothing worked. In the end, I said he didn't have to love it. After about 5 minutes of me ignoring and he did but I felt awful that I made him feel that he was some kind of failure because he wouldn't take the injection. I am not sure what this will do to his self-esteem in the future. I did praise him endlessly after but still, it's not fair on both of us! (Loretta's diary).

Mothers explained that many of the children preferred to keep their GHT a secret and conceal it from others especially at the very beginning. Most appeared to respect their children's wishes and supported them in doing so.

The odd time when somebody might say, "Have you done your injection tonight [Bill]?" so with all the comments they (family members) know not to mention it now and he hides under a tea towel if we're at granny's house and we have to get to the loo and do it really quietly with the door locked and then put it back (Bridget).

I always said to him, "If you want people to know we'll tell them. If you don't, you don't have to."... he doesn't mind me talking to you about it, but if I was to meet a friend for coffee and we started talking about it, he'd be just no... he's happy about it and he tells some people, but he doesn't feel the need to tell everybody and that's fine too (Sandra).

Mothers constantly tried to adapt as a result of being sensitive to their child's needs.

When children refused to take the injection, they turned to mostly self-discovered ways that proved to help in this situation. They constantly reminded them of the benefits and the consequences of not adhering to the regimen. They tried to adapt to how and when to inject their children and respected their wishes. They knew their children well enough to know how to minimise or avoid distressing them. Sometimes they considered the risk of distress, pain, and social isolation and balanced this against the benefits to the child's emotional wellbeing. The mothers' motivation for maintaining their children's wellbeing was also felt as they continued on the journey of GHT which leads to the next category about continuity of GHT as a source of wellbeing.

#### **4.5.5 The Continuity of GHT as a Source of Wellbeing**

Almost all mothers articulated how they saw much improvement in their children's health and wellbeing after commencing GHT. Change for the better was noted in the children's growth (weight and height), appetite, energy levels, physical appearance and development by their parents, the children themselves and others in the society. GHT served to help mothers and children to have a more positive outlook to the future.

Most positive experience I had was the September after [Ryan] first started GHT. When he went back to school after the summer holidays a number of other moms from the school commented on how much he has shot up over the summer and how tall he had got. These were people who knew nothing about his diagnosis and his treatment and so I knew they weren't saying it to make me feel better. It was the first time ever since Ryan had been born that anyone had ever said how much he had grown. It was really important to me to let Ryan know how well he was doing on GHT and how much it was helping him and he was so thrilled too. I know it is about so much more than his height and his outward appearance, but his growth is a reflection of how his health is improving overall on GHT (Rose's diary).

Experiences were mostly positive after starting GHT in regard to witnessing the positive effects of the treatment. Mothers were delighted to see their children thrive on the treatment.

So since starting growth hormone, we had our first six-month revisit and she'd grown seven centimetres and put on two kilos. So her growth velocity had shot up. So it was brilliant and we were delighted. She just looks great. It's helped her muscle tone. She looks stronger and her appetite has increased. She definitely seems to have more stamina. She doesn't seem as tired in the evenings and she hasn't missed a day of school. It just seems to be all positive really since we started (Nora).

[Leon] is doing really well over the summer, he has well passed out his twin sister. Everyone is commenting on how big he is getting. He is really nearly at the same height as his friends which makes it all worthwhile. He is thrilled with himself (Loretta's diary).

Mothers corroborated how their children started to blend in more in school and with other children their age after months of commencing GHT. They were happy to see that their children were not so different anymore and were not grabbing unwanted attention from others. They also witnessed how other people around them reacted differently which only served to confirm the social stigma that once was witnessed before starting GHT.

He's catching up on the boys even though he's a year ahead of them. When he went in, all the way up-, he's a year older than them, but he was always smaller than them, but now you wouldn't pick him out of a crowd that he's smaller. So he's had that much of a catch up that he blends now. Whereas before, it was obvious that he was way smaller than everybody else before he was on the growth hormone (Loretta).

When people stopped speaking to him like a baby that was huge. His image changed. Everything about himself changed and now after spending three or four years on growth hormones he is typical of average size... For a long time he was called stumpy. These are all the little labels that were attached to him because of his height and now he has grown out of all these labels and he's delighted and he's quite proud that he gets injections. He's quite confident about the whole experience (Loran).

Some mothers felt at ease when they saw the positive effects after months of starting

GHT. For them, these positive results confirmed the necessity of the treatment especially after doubting making the right choice for their children, this will be discussed in more detail under Theme 2 section 4.6.2.

We went back about a year later and [endocrinologist] said-, and she doesn't spend ages with us, but she said something along the lines of, "From where he is on the percentile now and where he started on the percentile, he never would have made that leap without the hormone. So yes he absolutely needed it." and there was such a relief because a part of me still felt-, even a month into it, I still felt like I was giving him something artificial to make him perfect. I still had that fear and I more or less, don't anymore (Bridget).

However, only one mother still had reservations around starting GHT. She had not witnessed any significant changes to better her daughter's health. She expressed still waiting to see improvement in her daughter's weight as this was her major concern when starting GHT. The treatment was her last hope to help control her daughter's weight, but with no impact. Therefore, she questioned the continuation of the treatment:

I have looked up how long does it take growth hormone to work? In every case it's going to be different as well. She's coming on to it very overweight. So I can only imagine I still have to keep the strict diet and exercise and hopefully, the three will marry up and I'm looking for the slightest change. I'm not looking to find that she's like Twiggy overnight, I'm looking for something... but I am now looking at it-, having done my best with diet and exercise, I'm now leaning very heavily on what growth hormone is going to do on top of that because the two of them on their own weren't working (Kelly).

Kelly also mentioned in her diary:

A curve has been detected on [Kim's] spine giving her the appearance of a hunchback. I am concerned the GH has made her grow taller but in her case the growth has not been upright but more into a curve on her spine. Awaiting an appointment with orthopaedic surgeon- I will tell him I think GH was causing curve. Also I am going to ask Kim's consultant if continuing GH is of any good as in my opinion Kim's condition in general (physically) has deteriorated since commencing GH (Kelly's diary).



Mothers and children noting the benefits of GHT served to help them have a more positive outlook to the future. After much doubt, physical and psychosocial improvements reassured many mothers that they made the right decision to start the treatment regimen. Not only were the mothers concerned about the wellbeing of their children at the present, but the future wellbeing of the child was also considered which leads us on to the next category.

#### **4.5.6 The Future Well-being of the Child**

Mothers expressed great concerns about the future wellbeing of their children. They were worried about potential side effects that may only appear at a later stage. They also were worried about their children being capable to adapt to the treatment regimen when they are old enough to take full responsibility of the treatment. They often questioned their children's quality of life as young adults receiving GHT.

I was afraid of premature puberty, I know and then I was going, "Oh god will there be an impotence or a fertility issue in years to come?"... Me as a mother, I suppose my big thing really is that I hope to god that in years to come there isn't a side effect (Loretta).

Mothers were apprehensive about reaching the adolescent stage where their children may not cooperate with them and the healthcare providers. They may also refuse to continue receiving treatment or challenge taking full responsibility of the treatment regimen. Mothers feared voluntary discontinuation of the treatment by their children.

I think once he gets to a certain level and maybe his growth might plateau, I'm not sure if he will be as happy about it. I think the teenage years might be hard because I think they get to a certain level and it's like a maintenance dose then. When the plates fuse, I think that might be the, "I don't need this anymore." I will just have to deal with that when I get there (Liz).

Some mothers also expressed feeling uneasy about the uncertainty around the continuity of their child's healthcare services as they enter adulthood. Also, transitioning from the child services to the adult services was unclear. Whether they wished for the

continuation or discontinuation of GHT, they were always concerned about their child's future wellbeing which will also be discussed later in Theme 2 section 4.6.3.

The only worry is when he reaches a certain age and he is a way of [current children's hospital] then that is my concern is what happens next you know so what happens for the future you know... Will they continue his treatment or will they just make him stop and that's the worry you have of what his future is like you know... because people kids have written on those sites (support groups) "oh yeah I felt very tired (after discontinuing GHT) I really wished I could have had it." (Francis).

The other thing is, we were writing Wills last week. My father died and I said, we better start doing our own Will and who's going to be his guardian and husband's sister lives in America, all our family are living abroad so we're going who will be guardian? If he goes to America can she manage the health? It's just stuff like that you have to think about, god forbid. Who's going to be guardian, who will be able to cope with this (cost and continuity of GHT)? (Liz).

Some mothers articulated their apprehension about the potentiality of their children missing more hormones in the future and/or the worsening of their children's medical conditions.

I know she'll be starting oestrogen soon because she needs oestrogen in her body for her bones and we have an appointment actually at the beginning of December to determine-, I'd say they'll start her with the oestrogen because her ovarian function has failed (Mary).

I worry a bit about puberty in regards whether he'll need testosterone, but I think that's just part of the hypopituitary. I'm not sure if that's specific to the growth hormones. It's kind of-, you can't look at it in a vacuum, you're going to have to see the whole thing (Liz).

The theme 'striving for the security and the wellbeing of the child' held many issues that revolved around the notions of balancing, fitting in, normalisation, protection, society expectations and stigma. This section illustrates how mothers are constantly

trying to protect their children and fulfilling their children's physical and emotional needs. Their stories revealed how they detected problems with their children's health and development, nurturing their children, maintaining their children's self-esteem and/or guarding them from physical harm. It also shows the mothers' will to protect their children was continuous as they advocated for their children to obtain medical help and fight to reach a diagnosis. The mothers making the decision to start their children on GHT in the hopes of helping their children overcome being different and its consequences was also highlighted. Moreover, mothers' inclination to protect their children while adjusting to GHT was also noted as they were always conscious of their children's emotional and physical needs regarding the GHT regimen burden. Also, the sense of protection that appeared as they talked about their worries around uncertainties regarding the future wellbeing of their children with or without GHT is also covered. Stories describing distress and the sacrifices, but then victory, emanates as mothers witness acceptable growth and development in their children that leads to better wellbeing and social inclusion. This also indicates their strong motive to protect and seek the wellbeing of their children. This leads to the second theme which discusses the uncertainties faced by the mothers which could directly affect their ability to optimally protect their children and fulfil their parental duties and be in control.

#### **4.6 Theme 2: "Doubting yourself constantly" Constant Uncertainty**

In this theme, areas of uncertainties that appeared highly significant to mothers are going to be illustrated. Mothers expressed many doubts around their journey of reaching a definite diagnosis. Then after reaching the diagnosis, their uncertainties continued as they decide to start their children on GHT and thereafter. They also expressed unknowingness around future matters in relation to their children's wellbeing with or without GHT.

##### **4.6.1 Diagnostic Uncertainty**

Many mothers reported difficulty obtaining a diagnosis, such as delays, misdiagnosis and lack of consensus among medical professionals in diagnosing the condition treated

with GHT. This uncertainty in reaching a diagnosis stemmed from the prenatal stage to the point of diagnosis. During the interviews, the health and well-being of the children was notably highlighted by the mothers. They witnessed their children's health, development and/or growth deteriorate, and as a result they were either admitted to hospital or had various medical testing performed. They were also referred to many different specialities but eventually were seen by an endocrinologist. Many mothers experienced a rocky start to their journey as their children were either born premature, small, or had a medical reason to be admitted in the hospital for further investigations which at times was a scary experience for the new mother.

When I was 20 weeks pregnant, I had my scan and that stage she was only measuring 18 weeks. So we were diagnosed with severe IUGR, but otherwise she seemed perfect in the womb. She was just small for her dates. So the pregnancy progressed okay and at 34 weeks, the placenta wasn't performing as well as they would have liked so they did a C-section and she was taken out and she weighed 1.25 kilos. So she was very SGA, but otherwise she was very healthy, she was breathing on her own. She did great, but when she was 10 days old she developed NEC and sepsis and we had a tough few weeks in the neo (Nora).

Michelle was faced with being told that a C-section was needed to deliver her son because he remained in a breech position until near the end of the pregnancy. Then immediately after he was born she expressed how her son was 'not lively' as other babies would be, but she didn't think the healthcare provider had concerns at that stage. However, on the fifth day post-partum, her son was 'taken away' for further investigations.

He wasn't as lively as other babies might be, but I don't think even at that stage I don't think they had major concerns... I was in for five days... and on day three I remember they took him away. I didn't realise it at the time, but under the guise of having jaundice, even though he didn't look particularly jaundiced or anything, but I guess that was just an excuse (Michelle).

Some of the children were admitted to special care units (NICU and SCBU) just after birth or even transferred to another hospital in the country. Joan experienced

transferring her new born baby to another hospital located in another city for further investigation to reach answers.

When my baby was born he was in intensive care in [X hospital] for about two or three weeks and then he was transferred to [Y hospital] and he was diagnosed there (Joan).

In addition to having experienced a difficult pregnancy until the birth of their babies, many mothers also faced difficulties with providing the normal mothering care giving activity of feeding their babies and were constantly worried about their baby's weight gain as this was a clear marker of growth, development and normality.

He was a prem baby, but he was failure to thrive he didn't put any weight on and he was bad at feeding and stuff like that (Liz).

Some mothers faced difficulties feeding their babies either by breast or bottle. They have previously tried to manage these difficulties, which at times proved challenging, especially when uncertain about the cause of the failure to thrive that was evident in their children.

I'd have to wake him to feed him, yes and you just felt that if you hadn't have woken him he'd have been fine. So it was a tiring time and after that we struggled on for probably about four months and he would feed and then he would be sick and that's where the expressed milk came in. You'd have this ritual to go through. You'd feed him at first and he would throw the first lot up. So then I had milk expressed to give him the second time. The second lot would usually stay in. It's only when I'm talking about it, it's coming back. It was a fulltime job. I was either feeding or expressing (Steph).

Feeding difficulties such as holding down food in some cases would have persisted for many years. Some mothers illustrated how they still had to deal with difficulty feeding as their child grew older with the help of professionals, which also exhibited some challenges. Supplements and enteral nutrition were used to treat failure to thrive in many of the cases.

From the age of about one my child had major problems with holding down

food. She was constantly vomiting... she was getting sick about three times a day... this went on for a couple of years quite honestly until she was about four and a half and she'd been on various infant formula to try and fatten her up and nothing was happening. Even, they (GP) got me to attend a dietician. I would have a science background so I was aware of the foods that would put on weight, but that wasn't the problem. It was the fact that she just wasn't holding down food (Mary).

He was becoming very weak and they didn't really know what was going on and they were saying, "Look, I think you should stop breastfeeding or trying to breastfeed." They tried to put in different supplements, but that really wasn't working either and the paediatrician said, "I don't think you should continue. I think we need to try other formulas and just see if we can go that route." So we looked at it, we went that route and it didn't make a blind bit of difference really... He was still vomiting all the time and probably, about eight months old there was a decision made that he should have an NG tube. So we had the NG tube and the NG tube was only going to be for a couple of months just to get him over-, he'd had a bug and it had just laid him really low and he'd been admitted to hospital and we ended up having the NG tube for about six or seven months, constant... we just basically said we want to have a PEG fitted as soon as (Steph).

In many of the stories illustrated above, one can see how evident it would have been for mothers' not to note something was wrong with their child. Not only for the mothers who had a difficult start which gave flag signs to something worrying about their child's health, but the knowingness that their children were different through constant comparing with other children in the family or community. For many, this was the impetus to seek medical attention and seek answers to why their children were different. Others did not react as quick and would have found ways to reassure their uncertainties among themselves. They noted their children being sick for many years and could also witness a delay in reaching their milestones such as crawling, rolling, walking, talking or even teething late compared to other siblings in the family. Many children were described being 'very sick' and 'miserable' for many years but with no definite answers as to why they were sick.

He'd been really sick and we'd been looking for years to see what was wrong (Emma).

He ended up being very miserable and sick for a long time then (Sandra).

Although many mothers had noted their children's lack of growth at an early stage, some tried to reassure their uncertainties by putting the lack of growth down to genetics. Some did not worry so much because their children were generally healthy.

I just thought I was the same I was a big baby and didn't really grow I remember my mother saying you were always small so (Francis).

There were some delays reaching an infinite diagnosis that lasted for some a couple of years and this really had an emotional impact on many mothers. Some mothers were given a different diagnosis to the one they had at the time of interview (e.g. abdominal migraine, constitutional short stature, autism and cerebral palsy). Some were falsely reassured that their children would catch up in growth. In addition, a couple of mothers questioned if their children were even correctly measured. Many felt dismissed by the healthcare providers when they noted their concerns, which led them to find other ways to get answers such as changing the healthcare provider, as mentioned earlier under Theme 1 section 4.5.2.

They thought autism, they thought loads of different things and he was originally given cerebral palsy as a diagnosis from a young age (Emma).

He said we will weigh him but my doctor is laid back, had a measuring tape laid out like this. I said you don't want to be very accurate you know it wasn't very accurate he laughed I said when you are going through this every mm counts so at least the hospital one was perfect a said would you not try and get and he said I have one but just never put on the wall surgery probably too busy and then he has a family of his own so he is probably up you know between work and kids so yeah he just measured him and he had an old scales like my own bathroom one would probably be better you know and I was saying same thing every ounce counts (Francis)

Noting or recognising the manifestations of the diagnosed condition proved challenging

for some mothers. They questioned if their children's symptoms were not typical to the asserted condition. The symptom pattern was not consistent for them and when the final diagnosis was reached, it raised a sense of shock and disbelief because it was unexpected.

I Googled it when I went home and I thought no, it couldn't possibly be this because she didn't exhibit any physical signs, other than she wasn't growing much... So eventually I rang up the paediatrician in [X city] and I said that I had heard about Turner syndrome and could [Megan] possibly have that?... I had an emergency appointment with him and he said that 99% sure it's not Turner syndrome, but just to keep you happy and to rule it out we'll run a test for Turner syndrome and a sample was sent to London for analysis... in the meantime, I had gone to a gastrointestinal consultant in [X hospital], who was excellent and throughout the course of the conversation- ' he'd referred her for an acid reflux test in [Y hospital] and he said had we done a test for Turner syndrome and my husband and I looked at each other and we just said, "Yes we're waiting on a result." and he said it would be good to have that. So we thought oh no, it couldn't be... So then after that... we obviously got the result and couldn't believe it (Mary).

To reach a final diagnosis, these children underwent a lot of investigations and tests. These tests did not prove to be easy on both mother and child. Children had blood drawn while experiencing needle phobia alongside entering an MRI machine and feeling very ill during stimulation tests. Mothers also faced difficulties as they mainly accompanied their children throughout these tests. Not only were tests hard on mothers but the waiting to have medical test done upheld many worries and concerns. After the tests were completed, mothers were faced with the anticipation and worries as to what the results hold. Waiting for the test results caused a resurgence of fear that mothers tried to ignore.

I suppose the only negative after that then was waiting for the results. We waited a long time for the results and that was very hard. He had the stim test early April and oh God, it was after the May weekend before we got the results. There was a problem with the lab... Anyway, it meant all the bloods if they weren't urgent bloods weren't being done for weeks on end and his wasn't in an urgent



category. So that was the hard time waiting and he'd be asking, "Did we get my results yet?" "Did we get them yet?" (Sandra).

Sometimes multiple healthcare professionals (GP, paediatricians, geneticist and endocrinologist) were involved in diagnosing the condition, increasing the potentiality for inconsistencies amongst medical opinions, labelling of the condition and delays. Suboptimal referral to the endocrinologist was also expressed by some of the mothers especially who had children who were diagnosed with GHD. Some mothers also questioned proper measuring techniques used by the general practitioner who would have been the first healthcare provider to be noted of the growth restriction. In addition to the inconsistency in growth monitoring of the children which was also noted in some cases, it was also presumed by some mothers that the rarity of the conditions may have contributed to delay in diagnosis of growth disorders due to poor recognition and understanding of the medical conditions by general practitioners and paediatricians. But there was also a proportion of mothers who reported a delay of the medical diagnosis because of the district healthcare professionals' underestimation of the problem.

1st time I approached a GP about my concerns he was completely dismissive. I felt stupid for thinking there was a problem and then relieved at the time that he must be ok as GP had said he was fine (in hindsight I know he never measured his height correctly). Down the line I felt really annoyed at this experience as all it served to do was to delay getting a referral to an endocrinologist when I again knew there was definitely something wrong (Rose's diary).

Mothers reported that the diagnostic process was lengthy and frightening. The lengthy 'wait and see' approach in order for some children to finally be diagnosed was noted. This 'wait and see' approach was very much hard on mothers when they acknowledged that their children were failing to grow. The feeling of not having control and waiting for the approval from the GP or paediatrician to get a referral to an endocrinologist was challenging, as discussed previously in Theme 1 section 4.5.2.

He saw him for about 18 month and again we would go every 6 month he was very well all this time and he was fine and healthy and he is a great eater and but he just very thin as well it wasn't just his stature he is very thin and so when he was 9 then no he was 10 he was the paediatrician in [X hospital] he had come as

far as he could with him and he referred him on to see (endocrinologist)... so we saw her and we saw her for about 16 months and again she just in kind of 6 monthly checks on him and then January of 2015 she said look I think there is probably ah something going on there that it's you know not just she was talking in terms in first about oh I'm just thinking of the word...constitutional growth delay (Sandra).

Some mothers recalled the events surrounding their children's diagnosis with all of them experiencing significant emotional turmoil. Test results were either reassuring for most mothers and gave them ground for their children's diagnosis, or in some cases not reassuring as they were inconclusive to the suspected condition and did not answer the doubts and uncertainties around their children's diagnosis. With the presence of uncertainties regarding the underlying cause of the illness or diagnosis, added stress and anxiety was faced by many mothers.

When he had his MRI done as well, the awful thing was, his came back normal and part of me was going-, which is a terrible thing to say because it's great because it means the likelihood of him having other hormone deficiencies is very low with him, but I would love to have a thing to go, "Do you know what, this is what's wrong. He's an ectopic pituitary." or there's something wrong that goes, that's why he has it and then you know that there's a physical result that you can say because you doubt yourself again then and you're going, "Gosh, if his pituitary is very normal, then why does he have it?" (Rose).

The awful thing for me was, I looked at the chart and I saw, "Query Prader-Willi." Went home, I did leave her in hospital. I went home, Googled it and one of my friends was the anaesthetist in a hospital, I knew him for years and he kept saying to me, "She's probably just premature." because they brought her on a little bit early, but the test came back as a false negative. So when I asked did she have it, they said, "No it's come back from [X hospital] genetic testing, no she doesn't have it, but we're concerned." So when I went for the six-week check-up I was brought in last. There were a good few mothers in front of me and I thought, "This is strange I'm on time." and he just looked at me and said, "She has a syndrome," and I said, "What?" "She's Prader-Willi." (Kelly).

Some children even needed more than one 'stimulation test' in order to finally reach the concurrent diagnosis of GHD as part of the diagnostic process.

So they did a stimulation test and they tried him with a few different agents and it was non-conclusive. He didn't react at all, he didn't go up or down as regards the test. So they did another one and he totally reacted (Liz).

As hard as reaching a definite diagnosis was for many of the mothers, receiving it was also hard for many. Whether this was straight forward soon after birth or after a long fight many years later, anticipated or not, it came as a shock for some mothers and as expected to others.

So then after that what we did was, we obviously got the result and couldn't believe it... You're in a state of shock (Mary).

It wasn't the best experience to be honest because the doctor called me in and I was on my own, husband wasn't there... So it was very upsetting... I just remember the room spinning and he was saying, "The circumference of his head and this, that and the other." My whole world just fell apart because you're sitting there hearing all these things and having had no idea that there was anything. So I still remember that day like it was yesterday, it was pretty horrible... it's such a big deal to find out there's something wrong with your child (Michelle).

We were very upset when it came... it didn't come as a complete surprise... but I just find it extraordinary that it's all one child in the family (Sandra).

For most mothers, however, the diagnosis was met with relief, marking the turning point from a state of uncertainty. However, for one mother receiving the diagnosis only perpetuated uncertainty. She doubted the validity of the final diagnosis because she continued to see her son's health deteriorate after the commencement of GHT and witnessed other manifestations that are unfamiliar to the final diagnosis.

Now, they didn't check anything else as far as I am concerned. It was like, he doesn't produce growth hormone that's what we're replacing and none of the other symptoms, none of the other signs were followed. So when he started

experiencing difficulties in the last year and they were coming up with new things and when I researched it, I was like that points-, that would cause lack of growth hormone. He was born with Hypospadias so he had to have circumcision. Loads of things that fit... When he had the stim test he was on the ward and everybody on the ward was having stim tests and the endocrinologist said to me, "Look around the ward, all these children are typical of children with a growth hormone deficiency and [Leo] doesn't fit the physical characteristics." which were a wide forehead, wide nose, low ears. Elfin-like and I was like no, he actually doesn't look-, the other kids all had a familiar look about them. Whereas, he was different and at that point I thought maybe they're wrong, but at the same time we did know, concrete evidence, he's not producing growth hormones and we replaced it, but I didn't feel that it was researched enough. I think that the medical profession tends to find a problem and deal with the problem and the extra problems-, that couldn't possibly be that. We have a solution and they don't look further and now, four years down the road we have other difficulties that are presenting themselves. His eye sight is failing, his absent seizures are increasing. His mood is low and that all has to connect to something. It's not normal for a nine-year-old to be losing their sight at a rate of knots. So that's where we're at now (Loran).

Mothers encountered many uncertainties in their journey to obtain a final diagnosis for their children. Uncertainties in noting a problem was apparent in regard to symptom patterns and manifestations. Uncertainties were also present while they sought medical attention, especially when their concerns were dealt with by the health providers at a lower standard than preferred. Their concerns and uncertainties were mainly undermined or ignored, and they felt dismissed by several healthcare professionals. Discrepancies in healthcare provider's means and standards of finding a diagnosis were also a major reason in aggravating their uncertainties. Finally, reaching a final diagnosis helped settle uncertainties attached at this stage for most, but not all. As they approached commencing GHT for their children, mothers faced many uncertainties regarding the treatment which is discussed in the next category in more detail.

#### 4.6.2 Treatment Uncertainty

After reaching a final diagnosis, mothers became aware of the treatment options available for their children according to their condition. Some underwent many tests and treatments to resolve health issues and symptoms non-related to GHT. They had other medical interventions grouped with GHT such as, additional medications, back brace, non-invasive ventilation such as; BiPAP (Bilevel Positive Airway Pressure) and CPAP (Continuous Positive Airway Pressure) and dietary restrictions. However, many children only needed GHT. This depended on the condition and the severity of the condition.

My son, [Liam] takes growth hormone treatment and he also takes thyroid treatment as well. He's got hypopituitarism (Liz).

He wears a back brace (for scoliosis)... he uses a BiPAP machine (for sleep apnoea) (Joan).

As mothers went through the decision-making process for starting GHT - as mentioned previously in Theme 1 section 4.5.3 - they considered making the best decision for their children at that time. Mothers expressed doing the right thing for their children and genuinely holding the best interest of the child in mind. The main goal for most mothers when it came to make the decision, was to improve the child's growth which was not improving. There were other reasons for commencing GHT other than improving growth or replacing the hormone, such as improving muscular hypotonia, earlier attainment of developmental milestones, reducing sleep apnoea and enhancing body fat composition in PWS. However, making the decision proved difficult for most mothers, as they expressed feeling hesitant, unsure and worried about starting their children on the treatment. This was due to the unknowing side effects of GHT. Although they all acknowledged the many benefits of the treatment when making the decision, at the same time they spoke about potential harmful side effects which they dreaded happening to their children.

With reference to the communication of GHT, some mothers were worried and not completely convinced about the treatment. In time, since they believed this option was the only solution, they chose to follow the therapeutic plan, but in their deepest thoughts they disclosed feeling scared about the possible consequences of GHT administration.

So I think it was last November they'd (endocrinologist) mentioned it (starting GHT) and we were very hesitant. When we heard it we were like, "Oh I don't know." So they said they would come back to us again in February and then we'll have a look again and I think they did a bone scan and all that kind of stuff and said come back again. So when she came back again in the February, he said, "Yes I'd like to start"... I don't know, I just wanted advice I suppose. I didn't really know what to do. I was very unsure (Berny).

Almost all mothers trusted their children's endocrinologist, but despite this general positive attitude toward the treatment and its potential positive effects, they expressed worries around possible consequences of the therapy.

Well, I was afraid I'd given him something that was going to cause problems, but I took that chance. I did some research at the time into the side effects, but I just trusted the consultant in the end. I went with what he told me. He said it was okay so, I said alright... She wanted him to go on it. I know her (paediatrician) and she knew [endocrinologist] and she said, "Come on, do it." She wanted him to go on it. So by the time [endocrinologist] made his decision, I said I would because he was so slow at making it. I thought he's not making the decision lightly. So I was quite happy to go with him when he decided, okay it's time to give it. I was happy for him to get it... I couldn't find anything that frightened me enough not to use it. I was taken the chance one way or another, but you don't know. I mean, I don't know. I looked up at the time and anything I asked [endocrinologist], he didn't know that much about it either (Loretta).

We obviously got the result and couldn't believe it and the result went to the GP and the paediatrician and obviously, the gastrointestinal consultant in [X hospital]. The [Y hospital] paediatrician got on to us and he said get her in immediately and we'll put her on growth hormone immediately. I didn't want to go anywhere near him because we'd been with him for two years and he should have been floating that up two years' ago, being honest with you (Mary).

The articulated doubts revealed an overall confusion and lack of information about the consequences of GHT. This implies that while mothers were supported and empowered through the diagnosis and the starting of GHT, regardless of their satisfaction with the

results and living with the daily injections without dangerous impacts, in their deepest thoughts they persistently worried about side effects especially ‘cancer’. Terms such as ‘leukaemia’, ‘negative growth’ were also used to describe their fear of cancer as a potential side effect - sometimes wondering and questioning their decision. Mothers were not reassured enough about the secondary features of GHT that might surface in the future. Almost all mothers, except for one, were reassured with witnessed positive results, but were doubting whether there was a price to pay in the future. As well as thinking of the negative effects of GHT when deciding to start the treatment, many mothers were uncertain about seeing definite positive results by commencing the treatment and questioned its worth. However, in the end it was a chance they felt that they needed to take.

I’m injecting my son with injections that are exacerbating growth and I hope whatever that tumour is, is not growing, if there’s a tumour. Is it growing? I don’t know, I really don’t know (Loran).

It should work, but it may not. Some people it doesn’t work on. So it’s kind of knowing, is this nightly injection actually going to benefit her at all or is just going to-, we won’t know. We’ll never know really (Berny).

Some mothers questioned their decision to start GHT and doubted its need and necessity. This was especially noted when children were put on GHT after a prolonged and delayed period of fighting, pushing and advocating by many mothers. It was also noticed when mothers didn’t have a conclusive test result to rationalise the need for GHT. In a couple of cases mothers doubted this, especially when their children were prescribed and commenced on GHT before receiving a conclusive MRI result. The delay in receiving test results was mainly due to long waiting periods for the test to be performed or for results to come in.

All the negative experiences made the guilt worse when I 1st started giving [Ryan’s] injections as I felt if a consultant (and others) can be so dismissive, am I really doing the right thing... especially, with something like this when you’re doubting yourself constantly because you’re thinking, “Am I just being over the top? I don’t want to go and get my kid treatment for something that he doesn’t need.” because it’s not obvious, it’s not just a blood test and you can go, “Oh

yes grand, that's what's wrong." or you have an x-ray (Rose diary).

I know of people who have got kids with much bigger problems where the parents weren't researching it and so it did feel like-, I felt like I was the one who was niggling and niggling and niggling until I got the answer and other parents weren't doing that and then, was I wrong to do that or were they wrong to not do that? I don't know. So we couldn't decide and the consultant was very nice about it and she basically said, "It's your decision, but yes you should do it (Bridget).

I was like, "Are we doing the right thing? Does he really need this?" It was only when we got the MRI I went okay, I know he needs it now. I knew by the fact he wasn't growing and he reminded me a lot about my brother when he was his age and I knew myself that he needed it, but it would have been nice to have-, it's quite unusual for the doctors to have to go and treat you without having the full background. It's not their fault, he would have preferred to have the MRI as well, but then he said, "Look the MRI is only going to confirm what I know already." but it would have been nice to have it, from a parent point of view, to know that this is exactly it's all been done and checked and we definitely need this. Whereas, for a while it was like, "Do I have to do it, does he need it? Am I injecting him with something he doesn't really need?" but then when I saw the results and he started growing (Liz).

All mothers eventually overcame their doubts and fears when making the decision to start GHT apart from one who refused to start her daughter on GHT when it was first communicated to her by the endocrinologist. She was overwhelmed by her uncertainties around potential side effects which interfered with her decision to start the treatment at an earlier stage.

It was just a diagnosis that you went home and looked up yourself and then I did a little bit of research through the Prader-Willi [support organisation] in America and all the stories with growth hormone were good, but one of the things that plays on my mind is that it shouldn't really be given to a child who is obese and [Kim] is... but the reason I was so reluctant was I felt it was probably just another guinea pig treatment that hadn't been fully researched and yet,



there's loads of children taking it... Anybody said anything negative I jumped on that because nobody knew anything about it and my parents are elderly and they were of the view, the poor child this is just another guinea pig treatment, I wouldn't do that to her (Kelly).

However, Kelly did eventually start her daughter on GHT as a final resort. She felt helpless and the only last hope to get her daughter's weight controlled was to commence her on GHT.

So I was stuck between a rock and a hard place and growth hormone was the only option I had because despite maintaining a pretty strict diet and putting in a certain amount of exercise, she just continued to gain weight. So it was really, really stressful and I was praying and I was doing everything and finally, I decided to give in and say okay (starting GHT) (Kelly).

But a year into the treatment, Kelly is still bombarded with uncertainty about the benefits of GHT and questions her timed decision to start the treatment with great guilt as she cannot witness any of the positive effects she hoped for yet.

It's just now a year later looking at her, I was even awake last night thinking, "When is it ever going to kick in?" I know he said it's not a miracle, it's not a magic wand, but I am sorry I didn't put her on it earlier. I think it would have been a big help to her, but we'll have to wait and see (Kelly).

For many of the mothers, seeing positive results helped lift the doubts and uncertainties that they had about their decision to start their child on GHT. Growth, physical and developmental improvements were their main goals to attain and they were the observers to the first successful outcomes, and the value of GHT was underlined in many testimonies as mentioned previously in Theme 1 section 4.5.5. However, when a child manifested a headache, hip pain, excessive pubic hair, moodiness, night terrors, clumsiness, changes in facial features 'angry looking' or scoliosis, mothers were reminded of the potential side effects of GHT and associated these symptoms to GHT treatment. The lack of reassurance was also witnessed when mothers turned to healthcare providers for answers. Yet, in many cases these questions raised by mothers around possible side effects were given non-reassuring answers. Therefore, being uncertain about side effects and feeling threatened could directly affect the continuity of

the treatment regimen. Also, after dose adjustment, some mothers noted unfamiliar symptoms and questioned their relevance to the new GHT dose. In many cases mothers reached their own conclusion to these symptoms being directly related to GHT without the reassurance of a health provider.

He has a pain in his groin they are saying the men on the pitch he is too young to have groin strain and then you are conscious oh is it his hip because he went up a dose... last September he went up a dose so you were conscious then did he you know is he getting you know the way if they increase the dose too much his hips could displace (Francis).

So then I thought there's obviously some sort of correlation then because it's happening when they up his growth hormone levels. So I don't know if it's related or not. I don't know if it's any affect. I guess with [Ryan], it was particularly obvious because he'd never had nightmares (Rose).

Questioning and doubting the timing of commencing the treatment surfaced for many of the mothers. They expressed never knowing the validity of the timing of their children starting GHT and if the decision made by the endocrinologist to start GHT was ever delayed. This also led to the confusion that many mothers held around the inconsistencies and discrepancies in starting GHT from one case to another which was also previously mentioned in Theme 1 section 4.5.3. They observed other children in the community or in support groups who were put on GHT at an earlier or later age compared to their own and this only aggravated their uncertainties around the treatment, healthcare providers' credibility and the validity of the guidelines for commencing GHT in the ROI.

I was a little surprised that lots of kids are started on growth hormone very early on and I wonder would we be in a better situation if [Megan] had started the growth hormone early on, but I know that research is constantly changing... If there was one central area that you could go to. That there's a summary of what is the optimum age for starting growth hormone would be really wonderful for other parents who are about to start it because clearly, when we started it, depending on which endocrinologist you're dealing with, there's a different opinion on it and still you're meeting with other parents for instance, I met a

parent and they had kids starting on growth hormone at three and four, your own situation and it's like, who's right and who's wrong? (Mary).

I guess looking back on it, I wish he'd started earlier, but I suppose one, we'll never know if it would have made any difference... but he was behind in a lot of things in playschool. Just taking height out of it completely, he was immature for his age, not very coordinated in terms of holding scissors and that kind of thing and definitely, immature for his age in terms of being bold and getting into trouble. That kind of thing and then, age four to six was all his speech therapy and sometimes, I think maybe he would have never needed all that if he had had growth hormone, but that's only my own theory. Maybe all of his muscles and everything weren't getting what they should be getting so, everything sounded a bit sluggish and actually, it would have been okay if we'd started growth hormone earlier and instead of starting in regular junior infants, he started in a specialised speech unit just for that one year and then went with his friends in normal senior infants and it could have saved all that if he'd started earlier or maybe it wouldn't have. It's impossible to know (Bridget).

The inconsistencies in GHT guidelines across many countries was noted by the mothers as described previously in Theme 1 section 4.5.3. Many were aware and equipped with information regarding international GHT guidelines such as the one used in the US and the UK. They questioned the validity of the GHT guidelines followed by the endocrinologists treating their children. They also challenged the Irish system and found the delay unjustifiable. They could not understand why it was permitted to start GHT earlier in the US while in the UK and Ireland it is delayed.

The growth hormone started pretty much after his fourth birthday. We were hoping to try and get growth hormone earlier because in the States you can have growth hormone earlier, but we were told under no circumstances in the UK and Ireland could you have growth hormone until four years old... Well I was frustrated because the majority of information that I was seeing was from America and very positive stories about kids. Either failure to thrive or Russell-Silver or SGA or whatever, they were all doing very well on growth hormone and I could see this child, who even at that stage was falling behind his peers. So it was frustrating, but at the end of the day we had what we had (Steph).

They waited until he was about three or four I think, four maybe. They just monitored him. They said in Ireland they don't give growth hormone very early unless it's extremely necessary. They prefer to monitor them... Whereas, I know from talking to people on [support organisation] and other websites, in the US they give it much earlier than they seem to do it in the UK and Ireland. So I was kind of querying that (Liz).

Mothers were overwhelmed with many uncertainties related to GHT. They expressed feeling hesitant making the decision to start GHT due to their uncertainties around the probability of unwanted side effects of GHT. They also felt unsure about the potential side effects of the treatment. They had many doubts around the necessity of starting the treatment especially when not having a solid test result indicating the cause of the GHD. Many relied on trusting the healthcare provider to cope with their uncertainties. Most felt reassured seeing positive results of the treatment on their children, however, uncertainties attached to potential side effects were always lingering at the back of their minds. They also highlighted feeling uncertain about the timing of starting GHT due to the inconsistencies across different cases and countries. The many uncertainties associated with GHT could have minimised their sense of control which led to constant feelings of guilt and worry. The great sense of uncertainty noted at the diagnostic and treatment stage also runs into future matters which are discussed in the following category.

#### **4.6.3 Future Uncertainty**

For many of the mothers, the sense of uncertainty followed on to touch future matters related to their children. These matters included the length of treatment, discontinuation of treatment due to potential harmful side effects, transition of children smoothly into healthy adulthood and genetic predisposition to future offspring. The length of treatment and the potentiality of stopping GHT was also noted as areas of uncertainty for mothers. All mothers were unsure when GHT would be discontinued. Some did not know if the treatment was going to be a life time treatment or temporary. Some would have had an estimation, but it was not definite.

She (the child) asks how long more do I have to be on it? I suppose I don't

really know. I know that she'll be on it for a couple more years, but as to what that timeframe is, I don't really know (Mary).

She keeps asking me, "How long am I going to be doing this? Am I going to be doing this until I die." and I say, "Don't speak about dying." "You know what I mean, when I'm an old woman will I still be taking injections." and I don't even know about that and I don't even think because I think we're fairly new on it if we're only a year, but for her it's a lot of injections (Kelly).

For many, the possibility of unwanted side effects showing in the near or far future also meant that the length and continuity of treatment was unpredictable. Mothers expressed 'trying GHT out' when making the decision to start as mentioned before in Theme 1 section 4.5.3. Mothers were also definite in taking action to stop the treatment if any serious side effects were to occur at any time.

I know, there were the side effects, but I think generally they happen in the beginning, but I'm not totally sure, could they crop up? That's the only thing, but if there was any side effects that affected her greatly. No, I'd stop (Berny).

Some mothers were not sure of the definite prognosis of their child's diagnosis such as GHD and the likeliness of missing more hormones in the future as covered previously in Theme 1 section 4.5.6. Moreover, their children's continuity on GHT as adults was also unknowing for many which was also noted in Theme 1 section 4.5.6. Furthermore, mothers of children diagnosed with PWS were not aware of the length of treatment and the possibility of the continuity of treatment into adulthood. The only mothers who seemed to have some kind of estimation of GHT length were the mothers of children diagnosed with Russell silver syndrome (RSS), Small for gestational age (SGA) and Turner syndrome (TS) but again, without a definite length to follow. However, in Turner syndrome cases, the possibility of discontinuing GHT due to starting other hormonal treatment (oestrogen replacement) in the future was also unknown.

There would be a chance he could miss loss more (hormones) ... they (endocrinologists) will have to wait till I suppose he reaches puberty and testosterone and that end of it the cortisone and all of that (Francis).

I do remember the [endocrinologist A] which I forgot to mention to you

actually, telling us that when [Megan] starts her oestrogen after a certain time she needs to stop the growth hormone. So I remembered that and my husband also remembered that and I would have said it to [endocrinologist B] in [X hospital] a couple of months' ago and she said no that she wasn't aware of that and so I thought, is it better delaying putting her on the oestrogen if you have to stop the growth hormone injections shortly after and she said no that's not the case. So being honest with you, I meant to do some further research on that to see when girls start oestrogen, what is the latest scientific research to suggest that you should stop the growth hormone (Mary).

Many mothers reported being unsure regarding their children's GHT when they transition to adult care in the future. Their children, leaving the care of their paediatric endocrinologists and starting elsewhere with adult endocrinologists at adult hospitals, was an area that proved worrying because of the 'unknown' for many mothers. The uncertainty regarding transitioning of care to adult services also directly affected their uncertainties about the length and continuation of their children's GHT treatment.

I just hope that he gets the treatment he needs as he goes into adulthood. It's a big worry for me that he won't get it when he does become an adult. Doctors can't give a proper answer so we will have to wait and see what happens (Francis).

I asked [endocrinologists] and they said, "We're here until he's 19." and noncommittal after that really (Bridget).

Mothers also claimed to be worried about the unknown regarding their children transitioning smoothly into adulthood and taking responsibly for their own GHT treatment. The adolescent stage was no different for some as they worried how their children would continue the treatment when entering this deemed difficult stage. As previously mentioned in Theme 1 section 4.5.6, mothers want to see their children transition smoothly to healthy adults in the future. When GHT is believed to be the reason their children were healthier, then being uncertain about its continuity in the future would only cause stress to these mothers. Therefore, for some mothers, the future health and wellbeing of their children as adults not receiving GHT was also unknown.

Now, when he goes to Cubs and when he goes on hikes with the Cubs, we have

to follow to give the injection because they wouldn't give an injection and he's not able to self-inject because he doesn't have the strength in his hands to press the plunger. So that will be the next-, is when he's doing it himself. Although then they'll be doing certain deals to-, is he actually doing them or are they getting flushed down the toilet or into his sister or into a teddy? It's too important for us to leave him responsible for it (Loran).

I don't know how long she's going to be on the growth hormone, but I would certainly like to be asking the endocrinologist, not when [Megan] is there, but for us to get some insight because in secondary school they can have trips away. So that's going to be a new chapter if she's going to be on the growth hormone she'll need to be taking it if she's away three or four days or whatever (Mary).

Some mothers' descriptions suggested that they were concerned about their children's ability to manage GHT independently as adolescents and young adults. This created ongoing uncertainty about mothers' future role and involvement in their child's GHT treatment. They also expressed being apprehensive about the effect managing GHT would have on their ability to live normally. So, mothers face many areas of uncertainties when it comes to their children transitioning into adulthood with or without GHT.

I wonder when he gets older himself how will he manage as regards if he wants to go travelling. We'll just deal with that when we have to (Liz).

What I think is going to be hard is when they're a young adult. I think when they're 17 to 23, just thinking about if there's a Scout camp or you're in college and you don't plan to come home and they decide, "Actually, let's go out straightaway from college." Those kind of flexible things, I don't know how all those things will be. I think that will be really hard. So I am hoping it won't be daily by then. That's wishful thinking (Bridget).

The extent of future uncertainties did not just revolve around the future of the children and GHT. One mother did report questioning and being uncertain about the genetic tendency of her son's medical condition to be passed on to future offspring. Therefore, it could be understood that having a child with an illness and requiring GHT and not having enough reassurance could also trigger worry about the possibility of it

happening to other siblings or future offspring which might affect their family planning.

I did mention to her (endocrinologist) the last day we were in, I said, “Is this a congenital thing? Is it a genetic thing?” because they’re three other kids younger and she said, “Probably not, it’s probably idiopathic.” Where she wasn’t saying all this one off, standalone thing, but she said, “No, there’s probably no reason to believe any of the other-.” Well, the other two are fine. He’s still young (referring to her youngest son in the room at the time of interview) (Sandra).

When it comes to future uncertainties, the length of GHT, its continuation or discontinuation due to harmful side effects, the transition into healthy adulthood and the genetic predisposition were the major areas of concerns.

Under this theme, it was evident that mothers held a variety of uncertainties that were prominent in their lived experiences. They had uncertainties throughout their journey to reach a final diagnosis for their children. Noticing the symptoms and manifestations of their children’s conditions raised many uncertainties about the presence of a problem. They then sought medical attention to reach answers, but their concerns were ignored at times. The variations noted in healthcare providers’ means of investigating and reaching a diagnosis also enhanced their sense of uncertainties. Reaching the final diagnosis helped most mothers feel a sense of relief and certainty. After the diagnosis, mothers were faced with the decision to start their children on GHT which proved to be difficult as many felt hesitant. They hesitated because of the uncertainties attached due to the lack of reassurance around potential treatment, positive and negative effects. The uncertainties continued after commencing GHT due to having doubts around its necessity. This was most evident when mothers did not have a solid test result to base their decision on such as an MRI result confirming an ectopic pituitary. Most mothers started their children on the treatment despite their uncertainties by placing their trust in the endocrinologist. As time passed and mothers and children witnessed positive results, their uncertainties were disregarded but were soon to be evoked when their children manifested unusual symptoms with or without GH dose adjustment. They usually associated the unwanted symptoms to GHT and were weary of the side effects. Also, being aware of different timing indications for starting GHT among different cases and countries, created more uncertainties regarding the treatment. Mothers also had unanswered questions associated with future matters related to their children’s



health and wellbeing with or without GHT. The duration of treatment trajectory for GHT was also unknown for many mothers. Moreover, the continuation and discontinuation of the treatment was questioned along with their children transitioning into adult healthcare in the future. Finally, the genetic disposition of the children's conditions to pass on to future offspring was also questioned. Despite their uncertainties, mothers tried to optimally manage their children's GHT regimen which leads us to the next category illustrating their adherence to GHT and making lifestyle changes accordingly to settle into the new normal.

#### **4.7 Theme 3: “But then you just get used to it I suppose” Adhering to GHT and Lifestyle Changes - The New Normal**

This theme addresses the mothers' adjustment to their lives with a child on GHT. This involved the management of GHT within the home environment and external environment, attending endocrine follow up appointments and continuously monitoring the effects of the treatment.

##### **4.7.1 Managing GHT Injections**

Mothers knew that commencing GHT entailed daily injections which would be either administered by the parent or the child. However, they described being hit by reality on the first night of administering a GHT injection at home. The process of starting the injection was always explained in terms of the drug company nurse paying a home visit and teaching both parent and child how to administer and manage the device chosen by either the parents or the endocrinologist. Participating in the choosing the device of choice varied across cases. It appeared that most mothers were given a choice regarding GHT injection devices before commencing GHT. However, some were not offered any choices.

I don't know that there's an alternative. An alternative has never been offered or discussed or optioned. I know other people are taking Nortropin and only because of America. I don't know (Loren).

They decided (endocrinologist) to go with Genotropin as our growth hormone. I'm not sure why they selected one over another. I wasn't really involved, it was just what they thought was best for him. I don't know why they decided one drug against another drug, but we got on okay with it (Liz).

Some mothers expressed being given a choice but then chose the recommended option.

Well, we were given the choice of a non-needle one, which we were very tempted to go for, but she (Endocrinology nurse) said really, it hurts as much or hurts slightly more than the needle one. That you're forcing the liquid into the skin and for a while we had a plastic thing to go over the needle (the needle device chosen) so he wouldn't see, but we found you couldn't fully know what you were doing, were we pressing it too far into the skin? We said tough, a needle is a needle, get on with it (Bridget).

One mother noted that she was offered many choices and chose the preferred device.

When we had various choices to make about the type of growth hormone and decided to go with Saizen because I hate needles myself (Mary).

After the device was chosen, the nurse from that drug company paid a visit to the family home where she provided them with an introductory session and a teaching session about the device and the practicality of using it. The teaching session proved to be very helpful and informative for children and parents including fathers as they were also involved in this process. The child is also involved and made aware of how to inject when found appropriate for his/her age in preparations to self-administer injections.

After getting this beneficial support at the beginning of starting GHT, many parents were left for the first couple of nights emotionally struggling to adapt to the new treatment regimen. The reality of their child starting treatment became real and having the daily injections became a daily reminder of the condition and the need for GHT. Many mothers had to adjust to a new role of injecting and it was a shift from the normal parental role. Adjusting to the new role was particularly difficult at the beginning of treatment.

So being back home (after birth and being diagnosed) I suppose you forget all

those things (the child's condition). You forget that there's this thing that's going to be with him for life. So then I suppose doing the job then every day brings that home, but then you just get used to it I suppose (Michelle).

A friend of mine is a nurse and initially she had said, "I will come and I'll do the injections. You need to be his mummy, not his nurse." and that was tough, but at the time I said no. If he's going to have these until he's 17, which is long-term, I can't have someone calling in every day to give him an injection. I need to soak this up. So it was tough (Loran).

Some mothers expressed being less capable and handing over the role of administering the injections to the father, but that was not always the case. Some couples shared the role of administering GHT between them. However, most mothers did take full responsibility of administering GHT from the very beginning or at a later stage as they adapted to their new role.

The first time doing it, I would have been very nervous. We definitely would have done it together. For the first probably month or so I'd say we did it together every night just to make sure and we would have taken turns to do it. We would have been very, which cheek? We do it into his bum and it was like, "Which cheek did we do last night?" to make sure it was alternate. So you were very hyper aware of everything. Yes, it would have been fairly nerve-racking to be honest (Michelle).

I hate needles myself. My husband would be much better at dealing with that. I'm completely a coward when it comes to even giving bloods (Mary).

Particularly, when he (Father) saw how he (the child) reacted. He thought, "Ah Jesus no, we shouldn't be doing this. This isn't right." (Emma).

I was asking him (Father) last night, I said, "Do you have anything?" He said, "Well, giving the injections freaks me out." It freaks him out, the thought of doing it, "Am I doing it right and am I hurting him?" For him it's like, am I hurting him? Does it hurt him and am I causing him pain? Whereas, to me it's like I'm more hardnosed, "Sorry we have to do this [Liam] whether you like it or not." Whereas husband is like poor thing. We'll buy him a present he's been

through a lot. Maybe mothers are more hands on, I don't know (Liz).

Many mothers disliked giving the injection to their children in general. Many felt bad for their children especially when they resisted or were ill. Expressions such as 'jabbing' and 'stabbing' were used by these mothers to describe the activity of the parent administering the GHT. Some mothers hated the thought of piercing their child's skin and inflicting pain to their loved one. This challenge involved not only the administration of the nightly GHT injections at home, but also getting regular blood samples from their child at follow-up visits which proved to be hard for many mothers to witness.

Initially I was anxious. Nobody wants to inject their baby with needles, but I'm quite clinical and when I had explained it to him. Initially, I had videos of him getting injections and it was like torture. It was horrible and very distressing and again, no one for me to talk to about, this is so stressful (Loran).

If they were to have a blood test or a biopsy of any description. It's not nice to see the child miserable and upset particularly, when you know it's justified. It's not that they're just being difficult, but it is genuinely unpleasant and not regularly, but often there would be occasions where he cries with the needle and he says it's sore, it's painful... I always hesitate sticking the needle in longer than I should because it's not nice... but I actually find it difficult to just find the courage to just poke it in to him. That gets easier. In the beginning that's probably the most difficult especially, when they're very young. It's a big ordeal (Louise).

It was terrible jabbing him with a needle (Francis).

[Becky] ran away and started saying "NO" when she seen her Easy pod coming out of the fridge for her nightly injection. I felt so bad for giving out to her. I forget sometimes how stressful this nightly injection can be for all of us... I really don't like giving Becky her injection when she is unwell. She has a cold and I feel bad for her (Berny's diary).

Although injecting was hard for some mothers to perform due to emotional and physical challenges, they felt that they had to conceal their fears to protect their children, which

was previously discussed in Theme 1 section 4.5.4.

The first night I would have cried after he'd gone to bed, a lot. I felt terrible. I felt so guilty... never in front of him, but when he would have gone to bed (Rose).

Coming to terms with the treatment requiring daily injections for a questionable period of time was difficult for many parents. However, with the unawareness of how long the treatment period would be, it could only be imagined that parents found making sense and adapting to the first period after starting the treatment (injection) even more difficult.

I guess my main concern was that I was going to hurt [Mark]. I suppose she'd given us the impression that maybe Mark could be on this for life. That's still the impression we're given. So I suppose just in your head psychologically, you're thinking, "Wow this is something we have to do forever." It's a big deal and it just seemed like a big deal that you were giving an injection every day to this tiny little person. So they were the main thoughts that were going through my head at the time. I suppose it seemed like a big deal because it's something you're doing and you could be doing this forever (Michelle).

Some mothers, after facing challenges, questioned the decision made to start GHT. They felt guilty and bad for the child especially when they faced many uncertainties at the diagnostic and the decision-making stage of the treatment. However, many mothers coped and overcame the treatment burden while still feeling confident of the decision made.

The reality of knowing I had to give him an injection every night, I felt awful... "Why did I do this? Why did I pursue it because now, I'm the one who has to give him an injection?"... it's still your child you're injecting every night so it's still very difficult... You're hurting your kid and you have to explain that it's to make him better (Rose).

I found the first couple of weeks traumatic myself doing it to him and he was crying and, "Why are you doing this to me?" that was horrendous, but I still think it's the right thing for him... but yes, very hard. When they're screaming

the place down it's dreadful... I think it was a good thing to do for him. I'd do it again. Even when it was tough for those first three weeks, I still kept telling myself it's the right thing to do. (Emma).

He's only six, I wouldn't expect him to want to do it. If I was six I wouldn't want someone poking me every night either. Sometimes I feel sorry for him, god love him. It's awful, but then I understand he needs it. Sometimes, when he gets very upset I say to my husband, "Is this worth it just for an extra few inches?" but then I know it's not just the height it's the heart, it's for other things as well, it's not just for the height of him. So I have to remind myself of that and sometimes I feel like I'm torturing him, but I know it's not just for the height, it's for other reasons as well. I know it's working for the other things needed as well. So I justify it, but it causes a lot of trouble at night time going to bed and I'd love to have a more relaxing bed routine (Liz).

Not all parents were fully responsible for administering GHT. Some children were self-administering at the time of interview and for their mothers to watch them self-administer the injection proved emotionally difficult.

I used to get a bit teared up when he used to be doing it (Sandra).

I have to watch him you know it's hard watching him (Francis).

Many of the mothers took full responsibility of the administration compared to the fathers. Rose expressed selectively choosing to administer GHT. It gave her a sense of accomplishment that she played a part in assisting her child to become healthier and took pride in her ability to assume full responsibility for the care of her son's condition.

My husband was a bit more, "Oh do we have to give injections every night?" and stuff like that and I'd be the main caregiver. I don't know why. We both tried, but he just couldn't-, he wasn't as-, well, wasn't as confident or I was there all the time because he was travelling with work and stuff like that. I kept trying to get him to be more involved. I don't know why he just left it up-, we weren't fighting or anything, it was just I ended up doing it most of the time (Liz).

Not necessarily fine giving it to him, but it doesn't make me squeamish or anything and I'm okay with that and maybe because [Father] might be more nervous doing it. He'd be fine with it now, but they both prefer that I do it. So I've taken in that role, but in a way I prefer that because I feel at least then when you go to his appointments and he's grown, you go, "Okay, I helped him a bit to do that. I had some role in it." I feel I need to be part of doing that (Rose).

Not only did most mothers express facing emotional turmoil when managing GHT injections, a lot of mothers also reported having technical and physical challenges. Mothers faced many difficulties at the beginning of the treatment regarding the technicality of preparing the injecting device used. Some lacked confidence doing so and chose to hand over the responsibility of preparing the injections to the fathers. Even when they had been prepared and well supported with the help of the drug company nurse earlier on, they still needed time to adjust to the technicality of preparing the device and using it (inserting the cartilage, priming, calibrating, inserting the needle, and storing). No mixing of medication was involved in any of the cases. Many mothers were responsible for preparing the injections or double checking when the child was self-administering. Some mothers didn't find any difficulty in prepping the injections and injecting. Moreover, device technicality errors (i.e. the needle getting stuck in the device or ceasing in the administering site 'leg') were also faced by many mothers.

The setting up of the pen, I wasn't very confident. We read the instructions every time in the beginning, but now it's fluent (Loran).

My husband normally prepares the needles, simply because I'm not technically minded and I have to look up the instructions every time, but it's perfectly manageable. He will normally prepare the needle and we leave it in the fridge (Louise).

So she started on that then about the age of five and a half I think and it was a rough time at the start and it took a while to get the needle right, the various parameters. I forget now, but the depth of the needle, the speed of the needle and so forth (Mary).

He (the child) has a habit of forgetting to tell you the needle didn't come out and putting it away and then when you go to put another needle in but I'm the one

that sorts that out I'm the one that ends up doing bits with it (Francis).

It can play up a little bit and they can get stuck if you don't handle it right. I know how to do it now. Often, I have to get tweezers to pull the needle out because the machine jams. So obviously, if that happens with somebody else, they haven't a clue how to fix it (Loretta).

No. My husband, whatever he does putting the cap back on, the needle gets jammed. So every time he has to use tweezers to pull the needle out...It happens with him all the time (Emma).

When administering the GH injection, some mothers expressed needing to restrain and hold the child down as they were resisting. This proved to be physically hard to do for many and would have required others' assistance to do so. This especially was noted at the beginning of the treatment when the child and mother were still settling into the new treatment regimen. Some children challenged the administration of the injection at a much later stage. Others had periodical phases of not wanting the injection.

It took three of us to hold him down to do it initially because he found it really sore... So the physical part of doing it was difficult... We've loads of friends-, unfortunately, it was bad timing when I started. My husband was going away the next day for a week and I knew it was going to take three adults. So neighbours and friends and my brother, who lives around the corner, he'd come one night and he'd hold him down and I'd do it (Emma).

Mothers found ways to overcome these administration challenges which was mentioned before in Theme 1 section 4.5.4. They tried ways to reduce the injection pain and would have used methods like applying anaesthetic cream (Emla) prior to injecting, using buzzy bee, choosing the smallest gauge needle, adding a needle shield to the injecting device, taking the medication out of the fridge in advance to its administration so it could adapt to the room temperature and finding the less painful site to inject. Others would have chosen an appropriate timing for administration while sticking to the GHT regimen for example, administering when the child is not tired or when the child is sleeping. They found ways to adjust to the regimen 'the new normal' and looked for ways to make the experience easier for both the child and parent.



The needle tip on them in longer so, he asks can we cut it and we tried that, but then it's blunt so, you can't. You have to just take it and because he's very thin, I don't know if that's quite common, but we were concerned about medical anorexia, it was suggested at one point, but he doesn't have enough, he can only take them in his legs and his bum because he doesn't have body fat anywhere else. So the MiniQuick (Genotropin injection), when you put it into his leg it hurts (Loran).

He lies on his back (while sleeping) quite a bit so, I have to turn him and sometimes that wakes him up and in the process he'll go, "Oh no, I don't want to do the jab." but in general, he's okay... The night time thing has been probably I would say the last three months, maybe not even. Between two and three months I've been doing it that way (injecting son while asleep). Up to that point my husband used to do it a lot. My husband has now started working in London Monday to Friday, he's only home at the weekend. So he used to do it a lot... my son does say, "It's better when daddy does it." but anyway, he seems to be getting over that I hope. So yes, about the last two months I think I've been doing it at night time yes (Michelle).

There was an ad for a smaller size and I was like, "Why is he not getting those smaller ones?" So I just rang up the-, I think I rang up the [drug company] nurse and said, "Can we not get a smaller needle?" and she said, "Oh yes, we'll order them for you?" and there was no pain then... Actually, I have to admit I tried poking myself with the needle as well just to see what it was like because I know if I do it a certain way it doesn't hurt. So it's just the technique, it's up to the technique whether it hurts or not. (Liz).

Many believed that the child would soon adjust to the 'new normal' despite the challenges. They decided not to use alternative ways to reduce injection pain and phobia and believed that sooner or later they would have to eventually adjust to the situation.

Only that it's good that he can't see the needle (a feature of the Easypod device) because for a long time he didn't-, it's not that he didn't know, he knew he was getting a needle, but he wasn't as afraid of it. What I used to do was take it out

afterwards and say, “Now that’s what you had put in.” Just so that he’d see the needle and register it because he has to have bloods done every few months. (Loretta).

For a while we tried-, there’s a cream you can get that numbs that little patch of skin, but that had to be on 30 minutes beforehand. So by the time that had been on 30 minutes and you had to watch your time and then get the thing out the fridge. It was more hassle, I just thought feck it he’s got to live with it. Do you know what I mean? I know somebody else I’ve seen online uses a Tens machine, little buzzy thing (buzzy bee) he’s tough and I thought he’s got to get on with it (Bridget).

The site of administration was a key concern for many mothers as discussed previously in Theme 1 section. 4.5.4. Many children were very thin, and mothers expressed injecting in some sites more concerning than others, such as the arms being very thin. Methods such as pinching around looking for the fattiest part they could find to inject were used. They tried following instructions to alternate the injection site every night. However, some children preferred one site over another due to comfort or discretion. This was demonstrated when some children didn’t want to be injected in the buttocks and preferred other parts such as the thighs. Mothers reported a few problems associated with the administration of GHT injections such as discomfort, medication leak, slight bleeding and bruising.

His arm he was too skinny he had no meat on him and he was very skinny so we kind of tried to pinch around (Francis).

[Endocrinologist] was trying to get me to do them on the cheeks of his bum because he’s pure muscle here (pointing at thigh). He (endocrinologist) was going, “There isn’t enough fat there to give it.” He just felt that it would be sorer on the thigh, but he’s always had it in the thigh and [Leon] will-, I said, “Why don’t we just try it?” He (Leon) will not allow me. He says, “No, no.” “He (endocrinologist) wants you to try it, he said it will be less sore.” He (Leon) won’t entertain it at all (Loretta).

Adjusting to the administration process was hard at the beginning for many, but not all. Some of the mothers were nurses or self-injecting as patients themselves and would

have had previous experience of injecting. This really helped with adapting to the situation much quicker. Others who did not have previous experience took much longer to adjust and to get used to injecting their children with GHT and overcome needle phobia. By the time they were interviewed, most mothers who were responsible for administering GHT were fluent and confident in the injecting procedure.

I suppose the fact that it seems to be benefitting her so much really helps as well and I don't mind doing it. I had to inject myself through my entire pregnancy as well, which was way worse and big needles. So I don't mind needles and all that kind of thing. It's fine. So yes, my husband is similar. We're quite happy with it all (Nora).

I work as a nurse so, I'm used to needles. So I wasn't afraid. So I think that made a big difference. He wasn't smelling fear off me. I was like, "Sit down I do this every day." So therefore, he just had to get on with that then. I said, "We have to do this." and so I did (Loretta).

Mothers reported different management techniques and coping strategies that helped them and their children adjust in the settling period. They often expressed believing that their affected child was very resilient and strong and well suited for this difficult situation. Others downplayed the treatment regimen and compared it to other more invasive and harsh medical interventions. Some found ways to avoid challenging situations as mentioned previously in Theme 1 section 4.5.4. Finding support from their partner/husband was also acknowledged as helpful. Having familiarity with injections and technicality was also noted as a facilitator. The support from the drug company nurse was also highlighted as helpful by many. Being open and involving the child helped with adjusting. However, parents and children witnessing the positive results of the treatment was noted as the major contributor to settling in fast.

My husband would have been better with all of that initially. I'm fine now obviously, but initially he was way more practical about it because it wouldn't bother him with the sight of blood or whatever. So you needed to have someone that was very level-headed and practical to implement this initially because if the two of us were like mush the child is going to pick up on that undoubtedly and she wouldn't have a very good approach. She's not bad at all about giving

blood (Mary).

I've never given it to her. That's one of the beauties probably, of her starting it older and she's also completely fascinated by 'One Born Every Minute', '24 hours in A&E', they're all her favourite programmes. So a needle or if her brothers get hurt in rugby or something she's on the scene straightaway with bandages. So to her all the preparing and the giving the needle and everything, all of that. Now, I dial it up and I do all that every month, but from time-to-time it's sore, but I've never had to do it. She's really great (Kelly).

Well, it's been pretty good so far, in the sense that I'm very lucky with [Ryan], he's my son who has it. He's very open to getting the injections and all that and he's very good with needles and with all that kind of thing. So from that point of view it's been quite good... out of my three kids it was lucky that it was him and not lucky for him, but it made it easier for us because of all of them, he takes it in his stride and I've never heard of a child like him, but he looks forward to blood tests (Rose).

[Leon] is taking his injections really well every night with buzzy bee which really helps. He and we are quite happy to continue using the GH as we can see the improvements in height coming along nicely (Loretta's diary).

Despite the challenges faced by the mothers, they appeared to adjust through these difficult and rebelling phases. They managed to commit and adhere as much as possible to the GHT regimen. Eventually they became more adapted to the treatment requirements and learned to normalise the treatment by finding helpful ways which were either self-discovered or suggested by a healthcare provider. The settling in period at the beginning of the regimen was difficult for many but eventually both mother and child adjusted well and GHT became a habit.

So yes, there's a little bit of a struggle at night, I would say, "Come on." and she would say, "I'm not ready, I'm not ready yet." So it's a bit of a-, but I don't delay we just go for it and then she has it and that's final. It's not pleasant, but it's way better than I ever could have hoped for. It's not something we think about much. Just brush your teeth give the injection (Nora).

Sometimes you could see him just a little wince and sometimes you'd get a little blood if you hit a blood vessel, but since he's older now he's fine with it. He actually quite enjoys the ritual I think, of having it done. We tend to still do it when he's asleep, but normal bedtime is half eight and then number two who shares a room with him is half nine. So normally, when number two is going to bed dad goes up and does the injection and puts number two to bed as well... I suppose they always said to give it at night time and I suppose in ourselves we just felt that it might be sore and it might be easier to do it when he was asleep. So it's just a habit that we've got into (Louise).

Mothers may have perceived the efforts used to adjust to the treatment regimen as part of fulfilling their normal parental role and felt responsible to minimise the disruptive aspects of condition management. Occasional misgivings and feelings of guilt were things that mothers experienced as a normal part of being a parent but were not a threat to their sense of competence.

When he was in [X hospital] actually for-, he had his tonsils, adenoids and other stuff done, I brought the medication with me and put his name on it and give it to the nurse to put in the fridge and then she gave it back to me ... Yes and I had to ask the people (nurses) that they had it in the fridge and they had to unlock the fridge and I had to put his (son's) name on it with stickers. I had stickers for school so I put them on the box instead. It wasn't up to them (nurses) to remind me, it was up to me to go and remember every night. No one else will remember (Liz).

Because I feel it's my responsibility... Maybe it's for myself because I ease the guilt then of knowing that I'm doing the best I can for him and I'm keeping up with it... if it was insulin or something like that, you wouldn't have a choice, you would have to do it. So it shouldn't be that different in his case because he's not producing what he needs to produce.... So it's my job to make sure that he gets it every night... So I would be very regimental in that (Rose).

As mothers tried to adjust to the requirements of administering GHT to their children, they also adjusted to GHT management and made lifestyle changes accordingly which are described in the next section 'Lifestyle adjustments'.

#### 4.7.2 Lifestyle Adjustments

Balancing treatment management with other aspects of family life was also noted. Mothers expressed how GHT became integrated into the normal bedtime routine and was parallel to normal activities such as brushing teeth and reading a bedtime story. They reported GHT being administered any time after 'tea time', before bed or while the child is sleeping. There was no set time for these families to administer GHT and usually was fitted into their individual lifestyle.

He hasn't missed one dose now in the year, ever. No matter where we are, what we're doing, he'll always-, but you know, it's just become a routine (Sandra).

After EastEnders it's time for the needle. So she's like, "The needle." If I'm on the phone or just reading or whatever, "The needle." (Kelly).

Mothers expressed altering the timing of injection according to the child's mood, location and their availability. Trying to adhere to the GHT was illustrated especially when the normal routine was altered. Mothers reported not skipping a dose in the most challenging situations such as when the child was tired, ill, having surgery or being admitted to hospital. The continuity of the treatment was significant to them.

No. Even when he's actually got a stomach bug, just to give it to him, he's never really skipped a dose. Even if he's fasting for an operation or such. When he was in [X hospital] actually for-, he had his tonsils, adenoids and other stuff done, I brought the medication with me (Liz).

Sometimes then if you're out, if you have a late night, he will not take it if he's tired. We were at communions over the weekend now and if he's coming home at 12 o'clock he will not take it. So what I do sometimes, depending on the night, I'll wait until he's asleep and I'll give it to him then (Loretta).

Some mothers reported situations where they might need to miss doses. In the case where the child is not self-injecting, a couple of mothers mentioned being on nightshifts and not being home to inject - one of the reasons for missing a dose. Also traveling or the child having a sleepover at a friend's house could affect the sustainment of adherence. Running out of medication and supplies was another reason for missing

doses. In addition, the child concealing the treatment from others would have also affected adherence especially for the children who were self-injecting. Having permission from the healthcare providers to miss a dose was also used in necessary situations to miss a dose.

We've only missed three doses in the 16 months and they were just totally forgotten. Not by me I might say. Three errors on my husband's part. Only three so, that's not bad and if I'm at work and my kids have to do it and he's having a wobbler or a hissy-fit, I would just say don't do it, just forget it. It doesn't matter for one day (Emma).

On one or two occasions we've gone over a bank holiday weekend or if we go away we may have forgotten the needle or we may run out of it. So the maximum in the last four years he may have missed maybe four injections as a result of circumstances, but we've just gone back to giving it to him as normal the following day and there haven't been any side effects (Louise).

Now the fact that we've been told by the nurse now that she can skip a day, you can skip a night. We skipped our first night only about a month ago when she had a sleepover because she was going to her friend's house at 3 o'clock in the day and it would have been awkward for me to come in and she didn't want it in the fridge, she doesn't want her friends knowing about it (Mary).

Mothers also reported missing doses due to forgetfulness on the parent or the child's part. They also reported trying to remind the child or vice versa in order to minimise the number of missed doses. Being immersed in life and what it brings may alter the parent and child's normal routine which occasionally results in forgetting the GHT injection. However, mainly there was a sense of being regimental and keeping compliant to the treatment emanating from the mothers.

She went to stay with her dad and they forgot to take it one night. I've never forgotten to give her it, ever and in fact, she'll say to me, "The needle." (Kelly).

I says am how you getting on (holidays) he (father) said grand he said nearly forgot the injection. I had to run up and get it out of the fridge they (father and son) were trying to remember. [Frank] (son) might have even forgotten himself

because he was enjoying himself so (Francis).

[Liam] was quite good, some nights if I'm very tired there's a lot of stuff going on I forgot and Liam went, "You forgot my injection." So he tells me to go and get it. So he does know it's part of his-, it's just something he knows. So he won't avoid it, he knows if I forget it he'll remind me to go down and get the injection, "Sorry Liam, I'll go and get it." and there's been one or two nights he's gone to sleep if there's a lot of stuff going on, we've been travelling and we go like, "Oh we forgot to give him the injection. Okay, I'll go and do it in his sleep." So that's fine. Some nights I have totally forgotten it and his doctors say he can skip one or two days a month if he has to, but other than that and I know I could change the pen and give him extra the next night as long as he gets it over the week. We've been lucky so far, we've remembered it (Liz).

As illustrated above, when doses were missed, mothers expressed handling this in a variety of ways. Some mothers didn't make a big deal out of it as they perceived missing a minimum of doses not to be significant or harmful to the child. Likewise, being reassured by the endocrinologists or nurses and having permission, helped downplay the importance of following or being strictly compliant. Some mothers counteracted the missed dose by spreading it out through the next few injections or giving it the next morning (if the device permitted them to do so, however in this case the easy pod doesn't allow).

Going on holiday... I know I've gone, but not too far, where I've forgotten to bring it with me. It's not like insulin where you're going to die. You just miss a few (Loretta).

But no if it's just a night we just do it in the morning or something or just miss one or something It's not the end of the world (Steph).

Francis expressed having an opposite reaction to missing a dose.

He was shocked there was a red line on the (easy pod indicating a missed dose) and he was devastated and I said [Frank] I nearly died as well because I could have sworn he got his dose but I don't know what happened (Francis).



Often mothers expressed finding it hard to find others to inject GHT when the child was not self-injecting. This was noted when parents wanted to travel or stay out late. In many of these situations, fathers, siblings, or family members would take over the responsibility of injecting GHT. Some mothers trained siblings to inject for situations like these. It was reportedly hard to find someone willing to perform an invasive procedure which may have restricted parents from pursuing normal activities. Many times, the mothers did not ask for support assuming that this was the case and in many incidences that reported it was tangible. One mother reported her mother (the child's grandmother) refusing to inject her granddaughter. Some mothers found it hard to hand over the responsibility of administering the injection to others. The issue of hesitating to trust others to do so was noticeable.

We have trained-, because we didn't always have weekends away, but we do now because the kids are older, but I can do them and my husband can do them and then the eldest are twenty, he can set up a needle and a new pen. We don't often leave it to him, but he can do it and brother, who is 14, we have trained him on how to set up the MiniQuicks, which is the daily disposable injections. We've taught him how to do that so that in the event that we're not here he still gets his injection and he has never had a break from injections (Loran).

Well, I suppose if I was to ask somebody who hasn't done an injection before. You know, most people haven't they get a bit scared. So if I'm out-, I've been out at weddings and I'm getting calls with, "The needle is stuck." because there's a few mechanical issues with it... So then I get the call when I'm out and sometimes he'll refuse it from whoever if I've a younger babysitter in, say. Sometimes what I do if I'm going out at 8 o'clock, I'll give it to him before (Loretta).

That's the only thing, nobody else will do the injections. We haven't had a night away from [Nancy] really because nobody wants to help us out. They're all a bit icky when it comes to it, but I'd say I could train either my sister or my brother if I needed to. My mum would be the person Nancy would stay with every so often, but she doesn't want anything to do with it. She's happy with it all she thinks it's great, but she doesn't want to be the mean granny giving her grandchild an injection. So that's how she feels about it (Nora).

Leaving the home environment and traveling in a car, boat or plane, caused mothers many practical difficulties. They had to try adapting to the change in the normal routine followed and manage GHT and adapt to unfamiliar environments. Preparation like thinking ahead and requesting enough equipment and supplies before perusing the planned journey was essential. Mothers planned and prepared for traveling with GHT in the form of; requesting a traveling letter from the GP, remembering all the needed supplies (device, needles and medication cartilages), ordering and having enough supplies to last the trip, maintaining GHT drug storage instructions while traveling and finally choosing suitable holiday accommodations to suit GHT storage requirements.

Once or twice we've gone away for the weekend and brought the medicine, but forgotten the needles and we might phone the [drug company] representative. The nurse who is new ... she's great as well and she might fax a request through to whatever local chemist we're going to in the locality we're going to with a request for the needles. So she's been very obliging as well (Louise).

I find that part of it a pain in the neck because say as an example, Monday is the day I work. My husband has to go to his mother's in the evening because she collects them from school and all this crap. Not today, but he has to remember to take it out of the fridge ten minutes before he leaves the house. You can't send it in the schoolbag because it can't be out of the fridge that long or if you forget it and leave it in the mother's house (Emma).

Storing GHT was an issue of great concern that caused utmost difficulties for mothers. In order to maintain the medication in exacting storage requirements, mothers had to anticipate how long the journey would take and planned accordingly. They often had to prepare a cooler that would keep the GHT stored under 8° Celsius to keep it safe and viable for the duration of travel. This may have caused reluctance to travel to far destinations due to possible failure to sustain the efficacy of the medication. Even after they arrived at a new destination, they had to ensure that they provided a fridge to store the GHT. The availability of proper storage facilities proved to be tricky as private fridges were not always available at holiday accommodations. Some mothers felt uncomfortable having the GHT stored outside of their view by the hotel staff in a main hotel fridge when there were no other options.

With the growth hormone itself as I say, the major thing was storage and travelling. Holidays. Travelling. We got the freezer bag and put it in, but I found going on holiday, if I booked a hotel they don't have a fridge. I was looking for portable fridges on the web and they don't seem to be available. They're a few hundred quid's worth and they're not really reliable. So it was a hassle of if we go away, we always decided we'd go self-catering so we'd have a kitchen and we have a fridge and that's how we've managed it, but it can be a bit awkward if you just want to go to a hotel and you ring them up because a lot of hotels used to have mini bars and now they don't. So it's like, "Do you have a fridge, don't you have a fridge?" and then they go, "We'll keep it in the staff fridge for you, you could ask for it." Other people I know are fine with doing that, but I wasn't very comfortable considering you can't shake it. When it's mixed up you're not supposed to drop it or shake it. I didn't know how it would be treated when it was out of my control. So I wasn't-, so on holiday we've always kept to self-catering and I've managed travelling and that (Liz).

Liz also expressed trying to manage unexpected situations while traveling which also extended the challenges of traveling with GHT.

There was one incident this year where we actually hired a boat. We said okay, let's try and do a boat trip for a week and I knew there was a fridge on the boat. I thought great and they said, "Oh yes the fridge is charged it's been going all night." but it turned out the boat didn't have any power sockets. So you had to run the boat-, it was running off the battery. I was in a panic ringing up people and asking-, ringing up the emergency hotline and saying the batteries are gone, I can't turn the boat on. That's my worst experience is keeping the stuff cold and travelling (Liz).

Some experienced delayed checkpoints at some airports and being questioned for handling medication and needles. In the case of Loran, carrying GHT made her traveling experience more challenging than it already was as she was traveling with more than one child with special needs.

Travelling with the medication is a nightmare as well. If the letter from the doctor isn't specific enough-, now, luckily for us I was stopped once and when I

explained what it was for and showed her my letters, they checked into it and they agreed, but you're not allowed to bring sharps on the plane, but I don't have an alternative because they can't travel in the language because if I haven't got it I can't get it. So that's a logistical nightmare as well. We travel every year, but my thoughts on it is there should be a specific-, for everybody, not just for growth hormones, but for anyone who uses sharps, there should be a specific travel case that a carrier would have that's deemed safe and secure by airports throughout the world and then we'd be allowed free without-, now, they don't charge, free travel through so we don't have the hassle of explanations because all my children have autism and if we're stopped to discuss these little injections, these little syringes that I'm carrying. I have to, I don't have a choice. It's just another logistical disaster, not really thought-out. Then it's probably because there's a very small percentage of the population that have to do this so, we're not priority (Loran).

However, it was interpreted that despite all of the challenges faced while traveling with GHT, mothers managed to adapt to GHT travel requirements.

Extra precautions have to be taken when traveling but to date this has never posed a problem (Louise).

The inconvenience of storing GHT was perceived, not only when parents left the home environment but also the storing requirements of GHT at home, was challenging. Issues like forgetting to put back the injection into the fridge after use was expressed by some mothers especially at the settling stage of the treatment. Also, the inconvenience of the injection needing to be stored in the kitchen fridge and not in the bedroom near the child was noted. One mother expressed GH injections taking up too much space in the fridge. In addition, maintaining GHT storage in case of electricity cut off was also mentioned. However, despite complaining about these difficulties, they usually found ways to adapt and adjust to the storage requirements of GHT at home.

The fridge thing definitely makes it a bit more of a pain, but obviously they can't help that, but that definitely-, about half the inconvenience I think is the fridge. Up and down to the fridge. Yes so, I pray that there will be a patch some year (Bridget).

In the early days is remembering to put it back in the fridge and then you feel terrible because it is just with four children running around at bed time and homework and brushing teeth, it can just get overlooked and then suddenly you realise, “Oh my god this is like gold dust and we forgot to put it back.” but I think it’s happened maybe twice and we phoned the drug company nurse and she’s been okay with it, but it does make you feel very guilty. That’s probably the worst experiences we’ve had with it. Feeling the guilt and forgetting to put it back. It hasn’t happened in years now, but in the early days you do have to remember to put it back and what we used to do then, we got an alarm and as soon as we take it out of the fridge we put an alarm on it. In fact, I think my husband pre-sets the alarm for 8 o’clock every night because we take it out at twenty to nine (sic) and then at 8 o’clock the alarm would go off to remind us to put it back in the fridge. So that was how we overcame that difficulty (Louise).

I find my fridge is full of pens. He’s on six pens a month now and it’s just the size of them, they take up the fridge (Liz).

There was also a sense of uncertainty regarding changes in GHT storage guidelines according to the device used. Some mothers expressed being informed of changes by the drug company nurse. The allowance of storing a used GHT cartilage outside of the fridge at room temperature below 25° Celsius facilitated traveling. However, Mary expressed feeling unsure about this and preferred following the old guidelines.

We went to Cork in the summer. Now, we only went for four days, but we were a bit-, because initially, you were supposed to keep it in the fridge every day, but the lady from the drug company said you can leave it out of the fridge for up to seven days. So that was fine because initially we thought, how are we ever going to go anywhere? We’re going to need to go someplace with a fridge, but no we were fine. We just administered it as normal (Berny).

I believe you can leave it out of the fridge now. The rules have changed with respect to storage of it. So you can leave it out of the fridge now for 24 hours. I feel uncomfortable doing it because you’re so used to having it refrigerated... That came about a year and a half ago, two years’ ago. She told us that. I’m a bit sketchy on what you can do because we still traditionally do the same thing all

the time (Mary).

Mothers' described expending time and effort on the organisational aspects of GHT management other than administration and storage, for example, organising prescriptions, sorting out the disposing of used needles, ordering supplies, requesting medical financial support and organising trips to follow up appointments that for many meant traveling long distances. Mothers mostly had to ask for prescriptions from their children's endocrinologists. They also had to order device supplies such as lithium batteries and disposable needles when needed from the drug manufacturing companies. However, some mothers expressed that the means of ordering needles and batteries for the devices through the drug companies had recently changed. Now they can order them from the pharmacy once they are included in the written prescription. Also, the disposing of used needles was done in a variety of ways across cases. Some expressed an uncertainty of the process of disposing them correctly. Much effort was thus spent mobilising multiple resources to help with the many practical, often administrative aspects of care.

I find the prescription is an absolute nightmare because I'm always without it and I'm always chasing my tail trying to make sure because they're a special license it's a bit of a disaster... the writing on the form has to be exact. If they do a spelling error it's turned down and I'm like, "My son needs his medication. We don't have time for spelling mistakes,"... Then getting rid of the needles is a nightmare. How that works I don't even know. We have a sharps container and if I want to get rid of it I have to give two weeks' notice. Then, when you bring it down if it's not full enough I'm in trouble and I'm like, "I don't know how I know when it's two weeks away." I have to go down to the clinic and the chemist can exchange it for the needle exchange if [Leo] was on heroine, if he was a drug addict, but he's not a drug addict he's a growth hormone user and for that reason he's not allowed to take them. So you have to go to the clinic and sometimes when you go they don't have a replacement container, which means you're holding the needles separate to your own waste. It's ridiculous. It doesn't make sense... So that's a nightmare and then the labelling has to be exact. So therefore, my thought is the containers should come pre-labelled and I can just fill in the details. She won't take hand written labels, they have to be printed. I

don't just have printed labels. Maybe it's just our person is awkward, but why does no one think of these things (Loran).

I suppose the hardest things like the blood tests that he's got to get in advance of the 12 month appointments, which have to be first thing in the morning. So it's not like we're just nearby and we can go in and take an hour. For us it will be four hours. Going down, getting two girls minded, getting the bloods taken. It's a bigger thing I suppose, but not so bad and certainly not as bad as other people would have it, journey wise and I'm at home with the kids all the time so that makes it easier. Gosh, even that kind of thing. If I was able to get those bloods taken locally it would be so handy, but I can't because the hormone needs to be checked immediately in the lab (Bridget).

So they basically order it in every month (GHT) and I have to get the prescription. It's a specialised script so, it has to come from the consultant, but they give it to me. I just have to remember when I'm in [X hospital], to get a prescription and they order it in and there's no problem. They've changed it now that you get the needles-, if you run out of needles they're hard to get them. So you have to be organised enough to know and the batteries are lithium batteries and you get those and the sharps and the sharps boxes off the company, but you can also order them now through the chemist. That's changed and you have a code. I haven't done that yet. I have just rung the company just because it's a bit easier for me. I just pick up the phone. Otherwise I have to try and route out to see what codes and the chemist can order them for me then. (Loretta).

The financial aspect of GHT was also highlighted as an organisational aspect of GHT management. Many mothers discussed how they paid for or were financially supported by the government to cover the cost of GHT each month. Most of the mothers paid the maximum fee of €144 (at the time of interviews) for each family under the drug payment scheme. Some mothers expressed having a medical card or a long-term illness card and as a result the complete costs were covered. All mothers noted the high cost of GHT and generally felt appreciative of the fraction of the cost they had to pay or being fully covered due to owning a medical card. This was especially noticeable when they mentioned other counties where GHT is not covered and would require families to either personally finance it or have medical insurance to cover the cost of GHT.

Another big thing for us as well is the cost of it. I know it's a high-tech drug and it's quite expensive and we're quite lucky in Ireland because of the 144 you pay the family whatever the-, drug scheme, yes we're lucky that's there, but my husband has been offered a transfer to a few different countries over the years and it's like, "We can't go to America, will we get health insurance?" That's kind of a worry for us because we may not end up living in Ireland forever. Wherever we go will they have the care, the cost of it because for four months you're fine, but when you go with a pre-existing condition to somewhere like America, will you get health cover, will you get health insurance? Will you get a consultant? That's a worry. We were chatting to his consultant and said, "What do you think?" He's worked in Canada and he's worked in the UK and he said, "Okay, I'll give you a list of countries, Canada, England, UK, France, Holland, Ireland have very good health systems that are part of it and be covered. After that it's going to be very complicated. If you go to America-" looking at maybe transferring to Asia, Singapore, I don't know. It's another thing you have to think about. So it's whether we'll be covered and the cost (Liz).

But the other thing is the expense is obviously, all covered on his long-term illness card, as well as the CPAP and the sleep apnoea equipment. Otherwise, my husband often says, "Could you imagine what it would be like if we were living in the States or somewhere where you had to pay for that?" I don't exactly know much, I think it's something like 22,000 a year, I don't know the ins and outs, but it's very expensive. So we're very privileged to have that (Louise).

However, one mother reported struggling to pay for the GHT but even that would have not affected maintaining her child's GHT regimen. Some reported their keenness to get a medical card or a long-term illness treatment card for their children so that they would be financially covered. However, most mothers noted that they would continue to cover the cost of GHT with or without external support especially after witnessing the positive effects of the treatment apart from one mother who was uncertain what she would do without a medical card.

I would get four of the injections. Sometimes he (pharmacist) gives me a fifth because he feels sorry for me and sometimes, I don't have enough money to pay him and he's great. I say I don't have enough, it would coincide when I'd have a



payment and he'd give me a couple of days to pay, but I think there's a medical card coming out for everybody on long-term illness treatment in March. So that will be great, but again the cost, I'd do anything (Kelly).

I've long had a squabble with the long-term illness scheme people because I think he should be entitled to one, so actually, there's a case with the Ombudsman and has been for the last two and a half years. So hopefully that will be resolved (Joan).

He has a medical card, but it will be interesting to see if it wasn't funded through the medical card we'd still go ahead I think. We would because we've seen the effects of it (Steph).

No, we have medical cards. Now, I don't know if we ever lost the medical cards what would happen then, but for the moment we all have medical cards (Berny).

With all the challenges faced and adjustments required to fit GHT to their new normal, they also made sure to attend endocrine follow up appointments. Experiences in relation to attending frequent endocrine follow up appointments are highlighted in the next section.

### **4.7.3 Attending Endocrine Follow Up Appointments**

With children being on GHT, all mothers mentioned attending endocrine follow up clinics at least twice a year. The appointments mainly involved weighing and measuring height before the consultant addressed concerns and issues related to the followed treatment. Blood tests and bone x-rays were also part of the monitoring process and this involved arranging the tests to be carried out in advance of the upcoming appointment. A sense of follow-ups gradually becoming a routine for mothers and children as time passed was felt.

I guess she'll (endocrinologist) check for-, I suppose the main thing they're looking at for is scoliosis, that's one of the things and they do his bloods every time to check for any abnormalities and he had to have his testicles lowered as well. So they check how that's all going and I guess just if there's any early

onset puberty or anything because of the growth hormone. They would be looking out for things like that and I guess we'd have a general chat about things relating to it. So that's about every six months (Michelle).

We're called back every six months and on the sixth month it's just a health check. Height and weight and how are we getting on? Then on the twelfth month he must get bloods taken first thing in the morning three weeks in advance of that (Bridget).

It was also noted that many of the unemployed mothers accompanied their children to most of the endocrine follow up appointments. These mothers felt that follow up appointments were very routine and familiar and therefore did not need the children's fathers to accompany them for support. When planning to attend an appointment (usually an early one), required taking time off work, taking the child out of school for the day and arranging to travel far distances which usually took a full day for many mothers. Sometimes for mother and child who live far from the hospital to be able to attend an early clinic, they would have to travel the night before and arrange accommodation in close proximity to the hospital. Therefore, mothers didn't see the need for both parents to attend the appointments. Altering the fathers work schedule to attend frequent appointments may only disrupt the normality and stability within the family routine and finances.

All the other visits we bring him (mother and father) so the last two I have gone I said to him (husband) there is no point in taking time off work because it's the same old you know you just wait and go in get weighed get measured (Francis).

He (husband) doesn't really go to the appointment with the consultants anymore. He used to come to the initial ones, but I found every time I went it's always just the mother. Very unusually you'd see a father in the waiting room. Maybe because it depends if you're off on maternity leave or you're off with young kids. If you're working, can't take the time off. If he can get the time off he will, but now it's such a routine appointment it's like, "I don't really need to go." It's the same, we weigh and we measure and we have a chat. I'm not working at the moment so it's easier for me to do it. I do notice the majority is always the mothers in the waiting room (Liz).

Well because of the logistics, we-, [Sam] quite enjoys that because we go up, usually the day before and we stay in the hotel and then always we get the earliest appointment possible the next morning so that we're in and we can find somewhere to park and then we have usually a morning there and we see who we need to see and then hopefully we're back on the road back (home) by lunchtime and we see either [endocrinologist A] or [endocrinologist B] when we're there and any bloods that we need to give or x-rays or things (Steph).

Despite being familiar with the hospital settings and clinics, attending an upcoming appointment created emotional turmoil for many mothers. They reported being anxious and worried when an endocrine appointment was due. In addition, attending the hospital frequently involved the children having a blood test which proved to be the most challenging part of visiting the hospital for many of the mothers due to their child's needle phobia.

I am getting a bit tense thinking about her endocrinology appointment next week. Just hope everything goes well and hope she doesn't have to get a blood test as she hates them! (Berny's diary).

I think what it was is he's quite slim. So sometimes it's hard to find a vein and when he was younger I think the blood test, they had to squeeze his hand a lot... so I think he thinks they're going to hurt him again and he's aware. We have different things, we keep him warm going in. I put extra clothes on him to keep the skin warm so the veins stick out and stuff like that, but every time we go to a consultant now it's like, "Do I have to have a blood test?" The first thing he says is, "Hello, do I have to have one?" and his consultant now is only doing blood tests every six months rather than every three months because he said he's stable now, he's growing, it's working he doesn't need it as regularly, but it's like, "Okay you don't have to have one now, but you have to have one-." but the whole way into the hospital in the car it's like, "Will I have to have a blood test?" It's constant anxiety about blood tests rather than anything (Liz).

In addition to the reported difficulties and challenges with attending the clinics, some mothers worried about which consultant they would be meeting on the day. This was mainly noticed with mothers who were going through the public sector which meant

they didn't know which consultant was going to be present at the appointment on the day. Favouring a consultant over another was also clear in many of the mothers' descriptions. It was expressed that the appointment experience would majorly depend on the consultant and that who they saw made a difference. Preference was mainly due to trust, patient and physician relationship and communication skills. Most mothers appreciated a consultant who was confident, willing to listen to their concerns and be sensitive to their needs. They also appreciated being involved. They valued consultants who were not paternalistic in their approach.

From talking to other parents they're like, "Oh you've got [endocrinologist A] you're very lucky." I don't know how you find dealing with hospitals, but it's kind of like it depends on which consultant you get. He's quite happy, "You know what you're doing. You're in charge, you're the main caregiver mum. Whatever you want." Whereas, I know from dealing with the consultants myself for different things I have, some of them are very preachy and I know better than you (Liz).

The first doctor I met, his comment was that, "We have children in here a lot smaller than him." So again, I felt really stupid and I'm bringing my child just because he's a little bit small or whatever... the other doctor I got was very good and he was really-, a completely different experience with him It just makes such a difference who you get to deal with. It really makes such a difference because-, especially, with something like this when you're doubting yourself constantly because you're thinking, "Am I just being over the top? I don't want to go and get my kid treatment for something that he doesn't need." (Rose).

Rose's trust issues were due to prior experiences.

At another of his follow ups appointments I unfortunately met with the 1st consultant again and his 1st question was why was [Ryan] put on GHT?! I couldn't believe he said this again I had to go through everything and felt like I had to justify Ryan being on treatment when if he had just read his chart he would have seen why he was put on GHT. When he looked through the chart he said "oh yeah it does show from his response to GH that he is definitely GHD"! As if it was all in my head up to that point. Also was again made to feel silly

over any concerns or questions I had in relation to feeling he might need a dose increase again having to fight for the best for my son. Very tiring. It made me realise how lucky I had been to have met with the other consultant when he was diagnosed. It's a shame though that this is the case that it is down to luck (Rose's diary).

Our public consultant is not particularly warm but I just trust her. Whereas, the first consultant who was private, I just did not trust her (Bridget).

Patient and physician relationship issues were mainly expressed in relation to how sensitive and considerate consultants are to the mothers' and children's needs. Not being sensitive enough was perceived by many. Also feeling rushed and not getting enough time at appointments to cover questions and concerns was also noted which will also be discussed in Theme 4 section 4.8.1. Another issue was feeling unworthy of the consultant's time. A sense of being less important and as minor cases for the consultant to deal with.

Yes and I think unfortunately, a lot of doctors lack that. Touch... sensitivity, empathy and understanding how difficult it must be for somebody. Particularly, when you see everybody else around you. I have three other boys so she wasn't my only child. I can imagine if a person has one child and they have problems, they can be a bit more sensitive because you tend to hang on to every word a doctor tells you. I stopped doing that after I had [Kim]. When I realised about the false negative (at diagnosis) (Kelly).

I suppose the other big problem I have with it is how doctors discuss everything right in front of the child. They don't seem to have much regard for little ears listening and what they're saying about the child... They would, except that I always bring my mother so that my mum can take her out of the room so that I don't let that happen... I suppose just talking about things that the child hasn't heard of before or doesn't understand or know how to process (Nora).

Small things like the three of us were sitting in her office and she said to us, "So what's the problem with [Bill]?" Now, he was six let's say. So in my opinion, definitely old enough that that kind of thing isn't acceptable to say that. I was the one to say, "Maybe husband and Bill could go outside the door for this part

or something.” So I didn’t think she was very sensitive to him and when she sent us the referral letter afterwards, her report letter, she got his name wrong and called him Ben. His name is Bill. Again, I know it’s a very small thing, but it’s like she didn’t bother (Bridget).

One mother felt she still had to prepare for appointments as she anticipated needing to fight for and push for action to advocate for her child’s needs. So, for her, advocating for her child’s healthcare needs didn’t end after reaching the diagnosis but continued with regards to managing GHT. This was only experienced when the mother was dealing with one particular consultant, which lead her to request for the favoured consultant at later appointments to avoid this.

I’ve actually started recently saying to the nurses-, asking for one particular doctor. I say, “If it’s possible-,” trying to be nice about it and I know they’re busy, “If it was at all possible to get this other doctor because I find him easier to talk to.” and they’ve been very good about it the last two appointments because I used to go in dreading [Ryan’s] appointments. I used to go in dreading them because you felt in some ways you were just fighting it a little bit (Rose).

Ineffective communication was another major aspect of the patient and physician relationship. The ways in which consultants approached and communicated with mothers and their children at the clinics were reported as a highlighting point of the experience. Some were described for their thoughtless manner when it came to address the mother or child’s concerns around the child’s lack of growth while others were more sensitive, thoughtful and actually listened. Positive descriptions mothers gave in their encounters describing their children’s endocrinologists were ‘warm’, ‘sensitive’, ‘friendly’, ‘lovely’, ‘have more time’, ‘very reassuring and helpful’, ‘very good’, ‘approachable’, ‘easy to talk to’, and ‘being open’. While negative descriptions used were ‘lack of sensitivity’, ‘lack of empathy and understanding’, ‘too busy’, ‘says very little’, and ‘not analysing how you feel’.

Now you are worrying as I said what happens to him (the transition to adult care) I did ask [endocrinologist A] (mother laughing) “oh don’t worry about that we will cross that bridge when we come to it” I could ask [endocrinologist B] and he would say “oh I know what you mean”. I did say to him will he loss any

of his other hormones “hum good question yes there could be a chance” you know he would answer you nicely the other fella [endocrinologist A] I only met twice and I was glad I never you know even though you are kind of under [endocrinologist A] but I see [endocrinologist B] or [Frank] sees but he is easier (Francis).

She suggested coming in for another appointment with the consultant who as I say, is very busy and I always get the impression that she deals with much sicker people than our son and I always get the feeling that she’s saying, “You don’t know what real sickness is, don’t waste my time.” She’s nice, but I think we’re a small problem compared to other things (Bridget).

Lack of effective communication was noted when results of blood tests were conveyed to mothers. They mentioned needing to remember to call the clinic to receive the results and add to the organisational aspects of care which was sometimes hard to remember as mentioned previously in Theme 3 section 4.7.2. This was significant to mothers as the dose depended on the results of the blood test. The change in the dose, if required, was reported two weeks post-test and mothers expressed needing to initiate finding this out. One mother complained of a delay in receiving a letter from the endocrinologist regarding adjusting the GHT dose. The delay resulted in her child receiving a higher dose than needed for a significant amount of time which would have been preventable with better communication put in place. Taking a higher dose than needed only aggravated the mother’s uncertainties around potential GHT side effects and only served to increase stress and worry.

[Leon] had IGF-1 bloods done in Feb 17 it took until yesterday to get the result from [X hospital]. Levels are on the higher side of normal. [Endocrinologist] wants to leave it until next March 18 to be revised at the clinic. I am concerned the spacing is a little far away to make the correct clinical judgment to me but this is my frustration and I don’t live in [X city] so it’s not so easy to attend review appointments. I had asked for a yearly appointment in [X hospital] and 6 monthly in [Y city] with [paediatrician] unfortunately she can’t make a decision so we have to wait! (Loretta’s diary).

Oh yes, there was a hiccup with the growth hormone, which is important to note

as well. She was taking it since January and we had a follow-up appointment. So six months into it, in the June and [endocrinologist A] said, "I'd like her to have a blood test." So she had a blood test and this was what really annoyed me, six weeks later-, she was on a dose of 1.65 that was what he started her on. So six weeks after the blood test we were due to go and see him. It was 10th November was the day we were due back. So she had the blood test six weeks prior to that... So anyway, on the day I was going to see him, a letter came in from the hospital telling me, immediately stop the dosage at this level as she's showing a high level of IGF-1 and commence her on a dose of 1.4. So I said, "Oh my god, six weeks since she had the test. She's had six weeks more of it after that being wrong." So I went into see him and I was absolutely furious because he would have had the results of the blood test at the very latest, two weeks and he met me in the corridor and he said, "You got my letter." I said, "I got your letter this morning." and he said to me, "You are joking." I said, "No, I wish I was." To make matters worse-, he said, "It's nothing to worry about, but you know we have to keep an eye on it." (Kelly).

The descriptions of ineffective communication between healthcare professionals and mothers were major sources of parental stress and contributed to the uncertainty experienced particularly when related to the treatment and care of their child. Despite communication being less than ideal, it was also important to note there were also recollections of healthcare professionals exceeding the parents' expectations and demonstrating excellent communication skills, providing valuable and much needed information (This will also be addressed later in Theme 4). The uncertainties often led mothers to be constantly vigilant to the outcomes of GHT which will be covered in the next section 'Vigilance to treatment side effects and positive outcomes.'

#### **4.7.4 Vigilance to Treatment Side Effects and Positive Outcomes**

In addition to attending endocrinology clinics as a feature of constant monitoring of GHT outcomes, mothers were constantly vigilant about the effects of GHT and continuously alert to not only the positive effects but the negative ones too. As mothers expressed their experiences around GHT side effects, it was noted that mothers did not



think of GHT side effects when their children were responding well and were generally healthy. However, as mentioned before both in Theme 1 section 4.5.5 and Theme 2 section 4.6.2, they were very quickly reminded of the potential side effects as soon as the child fell sick. They were hypervigilant when their children complained of unusual pain or were not their normal selves.

I also find that when he feels unwell, I worry is it something to do with his treatment. It is something we will always do as mothers of children with unusual medical condition (Francis's diary).

In addition to the reassurance given from the endocrinologist that the GHT was working, mothers generally looked for results and always measured their children's progress as signs of reassurance that GHT was working and worthwhile. They continually compared their children against other children their age or within their group (condition). They frequently measured their children's height at home while waiting in-between appointments. They also measured progress when their children were growing out of clothes and were able to engage in normal life activities. Other people in the community noticing the growth progress was also reassuring.

He is growing very well, he's on the 50th centile. His muscle tone is still poor, but I think it's much better than it would have been. His energy levels are very good. He still sleeps every afternoon for an hour and a half, but otherwise he's able to engage in pretty much any activity that we would do as a family or swimming, any sporting activity (Louise).

The consultant did tell me it's not a magic wand and I wasn't expecting her to turn into-, you know, to lose a huge amount of weight and immediately change, but I find it really frustrating because I see her every night taking an injection, being really good and it is sore. I know it's sore for her. I'm not even going to mention the cost of it. I don't have a medical card so, it's €144 a month. I don't mind that, I'd give anything, anything to see her even improve slightly. So I just feel-, I don't know. People are saying to me, "Oh she looks different, she looks better." but I feel they're humouring me. So for growth hormone, a year into it I don't know whether that's too early to see even a slight change, but at the moment I am really stressed and frustrated over it (Kelly).

So when you can think back to those images and see the way your child is and if you do as I do, attribute it predominantly to the growth hormone that is a major incentive to deal with whatever minor hassles come along with it. So I think it's good (Joan).

Mothers were very aware of their children's needs and especially when it related to GHT and dose adjustments. One mother reported she knew that her son needed his dose increased as she felt he was lacking energy. Another mother had an opposing opinion and demanded a decrease because she noted an unusual symptom on her son. This suggests that mothers are constantly hypervigilant to their children's responses to the GHT whether the responses are what they wish for or not. Nonetheless, this constant monitoring appeared to exacerbate mothers' uncertainties as discussed previously in Theme 2 section 4.6.2 and which will also be covered next in Theme 4 section 4.8.1. Loran sheds light at her concern that she was misinterpreting the perceived signs of responding appropriately to GHT and may have not reacted accurately to her child's needs.

I don't see any (side effects)... So when he was on the dose and his IGF reading was high and I said I think that he's getting pains, he was getting pains in his shoulders and complaining of headaches and I said I think it's because his growth is too fast and they said they can reduce his dose. Based on the blood tests he should have been within range, but I said I thought it was too high and they were quite happy to bring it down. Now since they've brought it down, that was in October and since they've brought it down his growth, development has stopped (Loran).

I find when we increase the dose-, every few months when we go see-, and weigh and measure him and checks the blood levels he says there's room for improvement so we'll increase it, but I know within a week of increasing it he's going to have awful pains in his legs and his hands. My son also has hypermobility syndrome. So he's what we used to call double-jointed. He's flexible everywhere. So I find when he does get growth spurts he gets lots of pains especially, in his wrists, his elbows and his knees (Liz).

He just didn't seem well in himself. I know his growth rate had slowed a little

bit and I had looked into the dose he should be on based on his weight and I knew he was in the next weight range up when I'd looked into it and he [endocrinologist A] wasn't upping his dose at all and he's doing fine and blah, blah, blah, very dismissive. I felt again I had to push things. I said, "Look, he's in the next weight range. I find he's just not doing great in himself, I find [Ryan] can get quite tired and different things as well." Very dismissively he said, "It just affects growth so, he shouldn't have any other symptoms." which is ridiculous really. If it just affects growth, my logic is then why do we have it as adults? I know it's minimal, but we still have it in adults because it fulfils other functions right (Rose).

Under this theme, several issues have been discussed in relation to the mothers' management of GHT. They compromised treatment regimens and tested limits to heighten normality for themselves and mainly for their children. They illustrated how GHT injections were managed and how they became easier and less time consuming over time. They described progress over the years in being able to do a good job of minimizing the disruptive aspects of GHT management. Mothers also described independently reviewing, altering the normal routines and adjusting GHT regimen to fit in with their planned schedule of activities on a day-to-day basis, particularly altering the timing of GHT injection. The management of GHT regimen also included organisational aspects which involved obtaining prescriptions and supplies which took lots of planning. Mothers also must deal with arranging to attend endocrinology follow up appointments and follow up on test results as part of managing GHT. In addition, continuous monitoring of GHT outcomes describes the work mothers undertook to continuously track their children's progress and wellbeing. They also sought medical advice and requested dose adjustment if necessary as a means of having control. However, having control to manage and adjust to their children's GHT proved difficult for many mothers, especially with much uncertainties lingering around as mentioned previously. These uncertainties were mainly a result of the dearth of information and support which will be elaborated in more depth in the following theme.

#### **4.8 Theme 4: “I hadn’t been told anything about it” Information behaviour; Looking for Normality and Certainty**

Mothers faced health challenges associated with their children’s condition and GHT management. This resulted in a breakdown in their notions of normality, thus creating a gap in knowledge which exacerbated uncertainty. Consequently, this stimulated information behaviours—including voicing information needs, and seeking and using information as mothers grappled to understand their lived experiences caring for their children on GHT and re-establish a sense of being normal. Mothers drew upon information behaviours from their reported stories regarding their experiences from the very beginning and which are still ongoing for many.

##### **4.8.1 Information Needs**

The mothers’ stories emphasised their uncertainties regarding many aspects of their children’s medical conditions, GHT and the management aspects of it. The lack of information available to these mothers was stressed at various stages of their experiences alongside their children’s treatment pathway, starting from the diagnosis of the condition followed by the decision-making process and starting the GHT regimen and subsequently managing all aspects of care related to the treatment regimen. It was apparent that most mothers had lots of unanswered questions that were never addressed.

I felt I had no support and I’m confident that he’s getting injections and he’s growing, but I am really unaware of potential future affects and that worries me. I’m not going to think on it because if I stop the injections he’ll stop growing. Do they stop growing? I don’t know. If he doesn’t get them, does his body now produce growth hormones? I don’t know. Do they ever review the stim test? I don’t know. When I ask they say, “He’ll be taking these until he’s 17 or 18.” If his body used to produce growth hormones and stopped producing them, why did it stop producing them and is it impossible for it to start producing them again? I don’t know (Loran).

Mothers described information-searching in the immediate post-diagnosis period. Specialists, general practitioners, nurses, pharmacists, charities support organisations,

friends, family and the internet were all key sources of knowledge and advice. However, lack of information from the healthcare providers was noted occasionally. As early as receiving the diagnosis of their children's medical condition, mothers expressed not receiving enough information about the condition and complained about the means of receiving the information. Many were given written documents in the form of leaflets and booklets and they found that these were not an effective means of conveying useful information. They preferred to have had more face to face conversations as a style of receiving the information. The lack of time spent between the mothers and their child's healthcare provider was noted and this was an issue when it came to get enough information and addressing concerns. Not only was the lack of information a major concern but also the credibility of the information provided by the professionals was also questioned.

The hospitals were appalling in dealing with it (communicating the diagnosis). There was no pastoral care, there was no advice. We were handed a printout from the internet by our consultant at 10 o'clock one night. He wasn't even our usual consultant. They had very limited information. We were misled into believing that it was a paternal genetic defect. Whereas, in fact [Lea] has uniparental maternal dicer homology. So for a long time my husband was feeling very guilty about the whole situation, until we met with the geneticist in [X hospital] who was excellent. So the hospitals were very poor on it. There was no real network that I could see at the time, in Ireland, other than the association and very limited information on growth hormone, other than what we managed to acquire from overseas consultants. They were very reluctant to help us or to advance us (Louise).

The lack of information was also visible as mothers expressed lack of preparation regarding diagnostic tests (i.e., stimulation test, MRI and blood tests). Many mothers lacked appropriate preparation for these tests before and after starting GHT.

It was horrible to sit there and to have to sit through that and we found there was no conversation, no preparation, it was just this is what we have to do and again, I took it on the chin because I thought if this is what it is then we have to do it... he did the first one (MRI) in [X hospital] under anaesthetic so that's easy, but the second one they decided they were going to do it up in the [Y hospital] in [X

city]... it was quite clear to me that they're not familiar with doing these tests on young children in that hospital. So when they brought him up they told him there was no injections, I had told him, be prepared for all of these things because these might happen and when they brought him in they said, "No not at all, there's no injections." and they needed to give him an injection, they needed to inject dye and he said, "No not today, I'm not having it today." and I said today we are going to do it because otherwise we have to come a different day and you'll have to take time off school and we'll have to pay for parking again and all these ridiculous things that are meaningless, but that made him think, "I need to do this." So they said, "He's not staying still enough." and I was like, "He's eight years old, you're putting him into a tube that scares the life out of adults." I don't know that it was the best idea, but it was the only plan we had and so they did it, but he did have to get injections and he did have to get two MRIs (Loran).

like a lot of my concerns that I had about the growth hormone prior to starting turned out not to be realities at all. So maybe if I had had information on the whole thing before we had to make a decision, it would have been really useful because I thought in my head we'd have to have blood tests every three months. I suppose I didn't realise that-, I couldn't believe when we had our blood test they were like, "You don't need these done for another year." (Nora).

As Nora mentioned in her story above, many mothers were not adequately informed to make an informed decision to start GHT. For most mothers, the lack of uncertainty due to lack of information regarding the treatment as previously mentioned in Theme 2 section 4.6.2, did not hinder them starting their children on GHT at the time it was considered, which was covered previously in Theme 1 section 4.5.3. However, it did hinder starting GHT sooner for Kelly who delayed starting her daughter on GHT because of the lack of support and information provided.

I hadn't been told anything about it, it was just being thrown at me, "Consider growth hormone." So I was looking it up on Google, which was a disaster and seeing how movie stars were taking it, like Sylvester Stallone and [Kim's] weight continued to escalate... [endocrinologist] was encouraging me to go for it, but I felt because I didn't know anything about it... [endocrinologist] did ask

me afterwards, what was my biggest fear. It was that she'd die if she took it... I was ignorant about it. That was the real reason. I don't think it's explained enough (Kelly).

Kelly expressed that she needed more information to make an informed decision and to overcome her concerns and worries. She expressed how she would have benefitted greatly from being redirected to support organisations and meeting other parents who had children receiving GHT. This need of shared experience was noted in all the stories as a very useful source of information.

The whole business about it would be, it's not explained in enough detail. In my case it should have been explained to me earlier when I was a bit nervous about it. I felt there should have been somebody taking me aside and saying, "Look I'll talk to you about it and you can meet other parents who are on it." It might be no harm to have a group of parents like this and for them to say, "My child is on it." they're not telling you to do it because obviously they can come back and say, "My child didn't get on okay on it." but I just feel for parents it might be no harm to have some kind of meeting for them or a way to explain it to them because I know doctors are busy and that's their manner and their way, but for parents to talk to each other (Kelly).

Lack of support and information was also noted after starting GHT and uncertainty was also noted when trying to manage GHT and adjust to lifestyle changes. Lack of information was observed in respect to healthcare providers addressing unusual symptoms that mimic GHT side effects and providing ways to manage GHT burden, which lead to self-searching for answers and solutions.

I did find from research these Go Quick pens that you can get that you don't have to keep in the fridge. I find that out myself from the web, no one told me that. No one at [drug company] told me that. No one at the hospital told me that and then when I asked the chemist, "Oh yes you can get them and they're single use only and you don't have to keep them in the fridge, but you'd have to get a prescription from the hospital for it." So I have to go back to the consultant and get-, so next time I'm in I might just ask him for a running prescription whether I use it or not, just to have the Go Quick pens... I wish there was a bit more-, if

I'd known about the Go Quick pens and I'd known about the travelling, if I'd known about the tips about warming it up before I give it to him. I have to hold it in my hand and heat it. If it's cold it hurts more. Stuff like that you find out if you do research (Liz).

I know he had one side -, after a while when he was on it, I know they can get that where the epiphysis in the hip can shatter because the bones grow too quickly and one day he couldn't get out of the bed. He just couldn't put his foot under him and I was going, "Oh!" Now, he wasn't that long on it, but in the end it must have been that he strained himself the day before because nothing turned out of it. I rang the girls in [X hospital] and nobody knew what to do with him because they hadn't had this before, but it disappeared over about two or three days and he hasn't had it since (Loretta).

The side effects of growth hormones as well is when he started to grow his body wasn't ready to grow and so he fell a lot. I don't know if other children fall a lot, but it was like his whole spatial awareness was skewed because now this body that he's in has grown so rapidly that he wasn't used to it. Things like that that they didn't prepare me for that I used to find myself scratching my head a lot (Loran).

Then just to explain about the upper respiratory thing (instructed to stop GHT when child is suffering from an upper respiratory infection). I think again, when I got the letter about the upper respiratory, that was something that hadn't been explained. I don't think the whole thing is explained properly (Kelly).

Future uncertainties conveyed a lack of reassurance provided to these mothers by the healthcare providers. Lack of information to reduce future worries regarding potential side effects and continuity of care was also noted in many interviews.

My understanding is lifetime (length of GHT), but I don't completely know. They said that he'll be in the paediatric system until he's 18, but they sometimes leave that go until 19, depending on exams and living at home and all that kind of thing and they were noncommittal about anything beyond that when I asked in the early days. From my reading of it, my understanding is that we all need growth hormone to be produced all the time for our wellbeing and my



understanding is it will be for life, but I don't know and again (Bridget).

This section 'Information needs' has considered the mothers' experiences of lack of information regarding many aspects related to their children's condition and GHT journey. It highlighted examples of inadequate and inconsistent information that contributed to the uncertainty and lack of reassurance felt by the mothers. It also demonstrates how lack of information affected the ability to manage the children's condition and GHT effectively which contributed to the stress and anxiety felt by mothers. The lack of information lead mothers to seek and share information which will be highlighted in the next category.

#### **4.8.2 Seeking and Sharing Information**

Mothers tried to bridge gaps in their knowledge and learned about their children's conditions, treatment management (GHT) skills and future matters expectations as a way of making sense of their experience. When they received limited information from the healthcare providers, they turned to other ways to source information. Many mothers self-searched for information and gained understanding from the internet and in many times found undesirable information which replaced their curiosity and sense of uncertainty with disappointment and frustration leading to more stress and worry. Rather than minimising mothers' uncertainties about their child's illness and treatment, information searching revealed negative stories, that in turn, created further uncertainty about the child's future. This led some mothers to abandon their search altogether.

That first day we got the diagnosis, I'd never heard of the term Prader-Willi syndrome, it meant absolutely nothing to me. So stupidly I Googled it and it really upset me. So I vowed after that that I wasn't going to Google it again and to be honest, I've done very little research about Prader-Willi syndrome because the little bit I have done I just find it upsets me and I worry too much about the future (Michelle).

I have Googled. I've read up on Russell Silver and stuff like that, but I didn't look too much into the actual growth hormone itself. I just wanted to know what the side effects are and is it natural or is it synthetic and what is it now? They

were the only questions I wanted to know (Emma).

However, many mothers explained how they are always wanting to be up-to-date and know more about GHT and always self-searching to find unanswered questions.

So we continue to research and keep reading about it and see if there's any new developments and when I saw that you were doing research into it I immediately thought, great. Get into anything that's going and understand and see if there's anything more that we could be doing, any more things that we can be pushing. That's where we're at (Steph).

I'm delighted that people are doing research on it. I actually do read some research papers myself and stuff and it is an area-, do you know what I've noticed actually now that you've mentioned it, I found that diabetics get a lot more help with pens and stuff. Any of the information I've found about the pen was I've actually looked up on diabetes sites and diabetes centres and stuff like that as regards how to give the injection because it's the same mechanism, but I find there's not as much information. Even though it's a different drug it's the same mechanism. So I feel lucky that there's diabetics there and they're using the same pens and stuff and maybe from a growth hormone point of view we're very lucky that we've got those pens because I feel like if diabetics didn't have those pens we'd still be using big needles. It's just that we've acquired the technology from some of those. I think a lot of the game and giving the injections is for growth hormones has come from the diabetic endocrine centres rather than the other way around. We're just piggybacking on their research as regards to the pens and the method of delivery (Liz).

The self-searching for information that many mothers pursued, helped them in receiving better and faster responses from the health services. Being equipped with information helped them advocate for their children's health needs.

It was only when we went armed with the information, to our own consultant (endocrinologist) that he agreed to go along with it (start GHT earlier), but it was an uphill road. It was an uphill road and we were lucky (Louise).

Receiving medical information from the healthcare providers (endocrinologist,

endocrinology nurse, drug company nurse and pharmacist) was considered the most reliable source of information when it came to GHT.

Definitely just seek professional advice and don't be Googling, I think is the best thing because you really don't get correct answers. Go to your GP or go to your local clinic or ring [X hospital] even and talk to somebody there. I think it's best to talk to a professional definitely (Berny).

However, most mothers expressed their need for experiential knowledge. This highlighted their need to share information with other parents experiencing similar situations. Mothers usually found other parents through support organisations (e.g. MAGIC foundation, Child Growth Foundation UK, Prader Willi Syndrome Association Ireland (PWSAI) and Turner Syndrome Contact Group Ireland (TCGI)), social media (Facebook pages) or at follow up clinics (speciality clinics).

She (GP) told us about [support organisation] and then I went home and I googled it and I got in contact with the Chair, who's wonderful... she explained about her daughter and she told us that there was a meeting coming up that both of us should go to. So we went to it... and it was just great to speak to other parents. Very sad, but good to network and being honest with you, talking to the parents is even more beneficial than listening to the formal talks, even though that's certainly good as well, but just meeting with other parents and just sharing what's going on with your child and learning from each other. Absolutely superb (Mary).

It doesn't make sense and again because there's no parent support or peer support, we don't have anyone we can speak to, I can speak to my sisters and my mother about it, but they don't really understand the reality of how extensively exhausting it is looking after medical needs constantly. Don't get me wrong, there's people worse off than us, his medical need is an injection every day and a follow, but it's like Jesus Christ give us support at some level, but we don't (Loran).

Yet, one mother declared how she found it hard to find experiential knowledge from parents who were living abroad as their experiences differed and, in many ways, didn't fit the Irish context regarding issues in healthcare, type of drugs, devices and financial

issues. So as a way of seeking and sharing information, Bridget had established a Facebook page for parents in Ireland.

I tried googling it which, I guess everybody does, but a lot of that stuff takes you to America and so, a lot of the information there is about insurer... so basically, it wasn't very relevant. So the UK and the Irish stuff is more relevant actually... So that's part of why I set up the group then because there's a UK Facebook group and so now there's a small Irish one. It's not very active, but still it's nice to have. We've asked each other the odd thing and it's helpful for the odd thing. It is nice to have it, but I guess your own individual case is so specific that there's not much information to be had really (Bridget).

The sharing of personal experiences with GHT facilitated emotional and cognitive engagement and this in turn assisted many mothers. However, not all mothers were fond of experiential knowledge and preferred avoiding support groups and organisations. A sense of being judged was noted as a reason for this shunned reaction, as well as, believing that experiences are individualised and differ from one case to another.

I'm not on Facebook so I've no clue. I've decided I'm just going to leave well enough alone. I don't know if I really want to know from anybody else. I'm not one of those-, even with my other child with autism, I never did anything with support group stuff. Just use my own friends and family as support. I probably work differently to other people... because everyone has a different way of doing it and I don't know if there's a right or a wrong and I did go initially to a few support groups for autism and because we chose to go down the diet road and natural therapies, people were looking at us like we had four heads. So I stopped going because I just thought-, you feel like you have to justify yourself. So I didn't want to start that with this either (Emma).

Fear of being judged and needing to justify to others was prominent in many of the mothers' stories. This draws on the preference of many mothers to restrict sharing information and disclosing about their children's condition and GHT treatment. The notion of others not understanding was prominent in many stories with some mothers remarking an inclination to conceal their experiences from others.

I wouldn't because people wouldn't get it so much. It's not nice, you'd like if

you were able to discuss it. If a child has a disease then you feel you can talk about it more, if it's something that people are aware of (Rose)

Mothers expressed that GHT could be a taboo topic. This might be due to the stigma attached to the treatment being used abusively for nontherapeutic reasons in order to reverse the effects of ageing and promote athletic powers. It could be elicited from this that the stigma attached made it more difficult for mothers to 'open up' about their needs and children's needs regarding GHT due to the fear of being judged. Also needing to justify to others who would not understand, was another reason. This lack of understanding was also expected due to the rarity of their children's conditions and seemingly looking healthy and well.

Why I haven't spoken to other-, like I said to you, I haven't met another parent. I suppose there's still probably a bit of a taboo about it (GHT) maybe. I don't know, maybe it's just very uncommon, I have never heard of any other kid that's on it, but then I suppose maybe they are and you just don't hear about it. I think maybe there is a bit of a-, it's not a spoken about thing. It's funny, my mother, when he was diagnosed she was saying, "Am I to tell people or is a secret?" and I was thinking, "Why would it be a secret?" If he was diabetic you'd say, "Oh god, my grandson has just been diagnosed with diabetes." If he was asthmatic. So that was her generation and she was saying, "I hope you don't mind now, but I did say to-." I said, "Tell anybody, I have no issue." (Sandra).

I also play down what his problem is because he is not sick looking it is hard to explain to people what is actually wrong with him. People only see height, they don't know all the other things that GHD causes (Francis's diary).

I found maybe because its growth hormone and not another hormone that [Ryan] is lacking that it is not viewed as seriously by some doctors and also by others outside of the medical profession not familiar with GHD as the effects of not treating are not as imminently serious as another hormone like say cortisol or insulin. Because of that I often feel I have to justify [Ryan's] treatment more than someone lacking a different hormone. People see that he's just small, what's the big deal but don't understand the real impact of not treating, as a result I don't discuss it much with other people (Rose's diary).

It was apparent that with the lack of certainty attached to the diagnosis to justify the need for GHT that some mothers originally felt (as discussed previously in Theme 1 section 4.5.4 and in Theme 2 section 4.6.2), could have made it more difficult for them to share their experiences with others, in addition to not feeling confident enough to share with others due to the nature of the diagnosis and treatment. This perception might be due to the belief that others may consider GHT to be a secondary treatment rather than a necessary one.

I was having a talk to my friend there a couple of weeks ago and she'd be a very understanding person, she wouldn't be the type of person who would question anything or make you feel bad, but she had a few questions for me because she was concerned about her little girl because she wasn't growing well and I was explaining a few things to her, but she was, "Is [Ryan] still on them (GHT)?" even though I'd said to people, "This is until he's fully grown and then possibly as an adult, we just don't know." I think there's still that lack of understanding, but he's growing and he's fine now, he looks okay so why you still giving it to him? I just think people don't fully understand it and you don't want to feel like you have to keep justifying everything all the time. So you're better off just-. Like I said, if his MRI had showed something, I think it would be easier. Not that I want that, but at the same time you feel like you can explain it then to other people as well as yourself (Rose).

Occasionally in order to maintain normality and protect the child as discussed in Theme 1, some mothers were selective in how much or how often they would disclose to others.

No. Well, I haven't told anyone. Don't want her judged. It was a recommendation from the genetic counsellor in [X hospital] who told us. Yes. Don't tell people about it. Don't tell anyone that she has Turner syndrome especially, when she goes to school. Don't let them know, she could be treated differently. Just the fact she's different to their kids. They will judge her. Why is she doing this? What else is wrong with her? I think it's just not as common and with diabetes or something, I think people are a bit more-, it's still scary, but it's a bit more widely known about. Whereas, growth hormone treatment isn't (Berny).

I'm not sure how confident I am to just openly discuss it with everyone. So I haven't. I wouldn't class it as a secret, if I came up. I've told a couple of friends, but I wouldn't be just bringing it up for no reason conversationally. I suppose if I ended up in a conversation with someone, I wouldn't mind sharing it. I don't think it's that much of a secret, but I suppose I feel protective of [Nancy] that it's her story. So I am not sure (Nora).

Having said that, some mothers felt the need to disclose to others. For example, some mothers believed being open about it and not keeping it a secret was a way of trying to normalise the treatment and help their children live with the condition and GHT and even get better support.

Because he does swimming and he might have little pin bruises and people say, "What happened to you, poor child?" and he says, "That's my injections, some days I bleed." People feel sorry for him because he has to get injections (Loran).

We had got very bad advice from the endocrinologist that we were dealing with in [X hospital] about not telling the school about Megan's health condition ...I had said it to some of the ladies in the Turners group about the advice from the paediatric endocrinologist in [X hospital] and they said that's dreadful and it wasn't good because it meant that we had to work really fast before Megan hit five and a half to get Megan the resources she needed. So it was the worst advice we got and his argument was that if the school know about the condition they'll treat her differently, but she needed to be treated differently (Mary).

Others felt that sharing information about their children's condition and GHT was curtailing to increase awareness and better the understanding of others. Especially when the wellbeing of their children and their parental duties were threatened or questioned by others.

The principle of [Kim's] school tells me she would like to have a chat. She tells me the staff of the school are very concerned about Kim's weight. They think she has ballooned and at her largest- I feel they are accusing me of making her fatter- very upsetting for me. I point out she is on GH and I am doing all I can (Kelly's diary).

We had got very bad advice from the endocrinologist that we were dealing with in [X hospital] about not telling the school about [Megan's] health condition and he had said that his daughter's friend had Turners and that she kept it a secret from the school and he said that was private to her. So we didn't say it to the primary school when Megan started and Megan had her diagnosis during the summer and we should have said it to the school... The school had two small classes and there was another girl in Megan's class that had extreme special needs. She was about twice, three times-, no, four times the size of Megan, both in height and in weight and even with her being in the same class as Megan was a safety hazard, but with the school not knowing about Megan's diagnosis, the two of them ended up in the same class, which they probably wouldn't have made that decision!... So it was the worst advice we got and his argument was that if the school know about the condition they'll treat her differently, but she needed to be treated differently (Mary).

This section concluded the means of seeking information and bridging the gaps in knowledge used by mothers. Encounters of them trying to cope with their uncertainties were demonstrated. Also, the sharing of information as a way of finding answers and seeking support were highlighted. In addition, the level of discreetness of information sharing used by mothers were touched. This leads us to the next theme which will cover their support needs.

#### **4.8.3 Need for Support**

The level of support received from healthcare providers (endocrinologist, endocrinology nurse, drug company nurse and pharmacist) varied across experiences. Both cognitive and emotional support appeared to be attached to their stories. Cognitive support was in the form of being provided with enough information from the endocrinology team when commencing their children on GHT and thereafter. It was also apparent in the form of being prepared by the GH drug company with a home visit and a teaching session given by the drug company nurse. In addition, continued support regarding device use and needed supplies that were delivered to the family by the drug company nurse was also noted. Moreover, some mothers received great support from



their local pharmacist in relation to information and practical provision.

Lack of information was greatly linked with a perceived lack of support. The lack of cognitive support might have led to the lack of emotional support for these mothers as dealing with gaps in knowledge only exacerbated the notion of uncertainty and increased their anxieties. Lack of support was closely linked to them feeling alone in the treatment journey.

I feel like the doctors are good at analysing what's wrong with the child and prescribing the medication, but after that it's up to you as a parent, as a caregiver to do the research and know what you're doing and how you're doing it and you have to advocate for your child a lot (Liz).

My husband works so he isn't here to discuss these things and nobody in my family has ever dealt with this before. The cousin that had the growth hormone deficiency was in America for treatment, but that was 40 years ago. It's quite different and I don't know support groups. The hospital doesn't put you in touch with other parents who are going through the same thing and that would be a huge benefit being able to compare, is my child now responding well to the medication? Has anyone else had these side effects? Is it a medication problem? The majority of the parent support is from America where there's far more treatments offered as opposed to here. So that's a bit of a struggle. It's not something that I'm fond of, the isolation that it brings and for [Leon] because he doesn't know anybody else that gets injections every day and he could do with peer support, but there isn't. It just doesn't add up (Loran).

Not only was insufficient information a sign of absence of support, the lack of constant support was also noted. Many mothers praised the support they got at the beginning from the drug company regarding the drug company nurse teaching them the technicality of the GHT injection, covering device issues and following up on their adjustments. However, the support was reported to be very strong at the very beginning for most and tapered off as time progressed. The lack of consistent level of support bothered some mothers more than others.

I got a call to say that the [drug company nurse] would be out to discuss the medication and to show me how to do the injection, set up the injection... she

came in and she gave me a school bag and there was a teddy bear and a story book and all these great things, but then that was it. That's the end of that support (Loran).

We had great service from the nurse at that time, but certainly as time has gone on that level of support is not there to the same extent as it was at the beginning, definitely not. They've had-, I forget the company that deals with-, is contracted to-, the aftercare service, I forget the name of them. [drug company name] is the company, but they use a company and since they contracted that out, certainly the level of support is not good at all. They've had major turnover with the nurses who come out and visit. In fact, I can't remember the last time-, maybe it's about a year, a year and a half ago somebody visited and the only reason that was the case was the fact that our device wasn't working anymore because it had slowed down and it's because the machine was two and a half years and you're supposed to replace it every two years (Mary).

Nonetheless it is also important to note that some mothers felt well supported and appreciated the great service provided by the drug companies.

The girl [drug company nurse], she was fabulous because I could ring her about anything and then she would get on to [X hospital] if I-, you know what I mean? It was going through her. So she was really the support, definitely the support. Now, [drug company] have rejigged in the last while and they've only one nurse now covering the country. So the girl [drug company nurse], is gone and there's another girl who is probably not as nice... Now, she's a lovely girl, don't get me wrong, it's just that she's now covering the whole country. Whereas, I had a very personal relationship with [the first drug company nurse]. She used to come to the house and she would have come any time I asked and anytime I rang she'd say, "I'm going to Donegal, I'll drop into you on my way." I never had to say, she was in the house maybe three times, four times maybe. That's it (Loretta).

The support that was received from the endocrinology team seemed to vary as well. Some mothers expressed not getting adequate levels of support from them. Mothers reported many encounters where the consultant or the nurse were too busy, which

resulted in rushed appointments as previously discussed in Theme 3 section 4.7.3. It appeared that some mothers even avoided seeking support from the endocrinology team because of this. Vice versa, many mothers praised and were happy with the support given by the endocrinology team.

I knew I could go to the consultant if I have to, but they're so under pressure, they don't need a mother ringing them worrying about an injection (Emma).

[X hospital] are fantastic. There's an excellent nurse... The level of care that she provides and she checks the sites and gives advice to [Nancy] about where to inject and for instance, we'd have been doing her backside and she was saying that maybe moving forward she was showing Nancy other areas that she could inject so that mum and dad are less involved in the injection and Nancy can give the injections herself (Nora).

One mother assumed that the type of service (public or private) could have a direct link to the level of support received.

The endocrine units in the hospitals are very good, the endocrine nurses are very good, but I don't know. We've always gone privately, the private clinic in [X hospital] and we just got referred there when he was young and I just thank god we've got very good health insurance so I'm quite happy to do that, but I find that because I'm going privately I don't have the-, other people say, "Ring your endocrine unit." I don't really see them, I go and see him in his clinic. I don't really go to the endocrine hospital. So I'm wondering if I'm missing out on a bit of support there. I've only ever been to the endocrine unit when he's had that test (GH stimulation test). So I've been wondering if we're missing out on the support because I'm private. I'm very lucky I don't have to wait (Liz).

There was a generalised sense of mothers needing more support in regard to GHT. Instead of being given the support they had wished for, they were left to support themselves and feeling alone and this is conveyed in statements such as:

The physical side of support with using the device, choosing the device, I thought the physical side of it was fantastic and the support regarding the medical side of things, but I guess the emotional aspect of it, I felt I was on my

own completely (Nora).

Yes, I'm quite happy with it because I'm quite comfortable with it, but it shouldn't be up to-, if there's anything new, I feel like someone should be contacting me and saying there's new pens out or there's new needles out or this is the latest, maybe we should try this drug. I feel like a lot of it is-, okay, the consultant might suggest a different drug or something if they whatever, but the day-to-day stuff you're on your own (Liz).

Most mothers sought support from other parents living the same shared experience. This was apparent as they searched and found ways to obtain experiential knowledge to fill in gaps in their knowledge. It was noted that this would have eased their experience in terms of making the decision to start and then beginning GHT. It was also said to be important in terms of having other similar situations to compare or bounce off and assess GHT's effectiveness. Similarly, it was a source for a sense of community which are willing to be helped and assist each other at times of need regarding GHT. For example, providing advice and recommendations based on experience. Parents also were a source of valuable information as they directed mothers to appropriate support organisations and resources. They were a source of reassurance when doubt and uncertainty were overwhelming. Finding the support from others who were described as 'being in the same boat', was very helpful.

On a practical note, I think that I would have benefited gratefully from having contact with another mother in the same situation as I, in those early weeks between receiving results and commencing treatment. It was a stressful time trying to come to terms with it and trying to be positive and upbeat about it for son's sake (Sandra's diary).

Other parents have sometimes helped us out. We've got the name from [support organisation] of other parents who are on growth hormone as well. So that's very useful as well to have a community of growth hormone parents that can help each other out (Louise).

Maybe have a meeting for the parents with other parents, which I think a lot of the syndrome support groups do because there's nothing like talking to somebody else who's in the same boat as yourself (Kelly).

This section on “Support need” has reflected mothers’ experiences of receiving support from healthcare providers and highlights provided descriptions of unsupportive encounters which resulted in conflict often arising due to sub-optimal communication. Also, mothers’ recommendations and support needs were considered. The findings under Theme 4; “I hadn’t been told anything about it” Information behaviour; looking for normality and certainty encapsulates the journey of lacking and needing information and support by these mothers. The findings highlighted challenges that came with suboptimal communication and support and while there were illustrations of good communication and support these did not feature as prominently in the descriptions of experiences encountered along their children’s treatment pathway. The lack of emotional and cognitive support were obstacles to making sense of their experience and, in turn, anticipate greater control of their children’s illness and GHT.

#### **4.9 Conclusion**

This chapter has presented the findings of the study, organised according to the themes (meanings) that have emerged. The chapter provides an insight into the experiences of mothers caring for children receiving GHT. The exploration of mothers’ experiences of caring for children receiving GHT has identified that this revolves around three concepts which are normalisation, stigma and uncertainty, and are central to the four major meanings of their lived experiences. These meanings were discussed under each theme titled: (1) “It’s the right thing to do” Striving for the security and the wellbeing of the child, (2) “Doubting yourself constantly” Constant uncertainty, (3) “But then you just get used to it I suppose” Adhering to GHT and lifestyle changes - the new normal, (4) “I hadn’t been told anything about it” Information behaviour; looking for normality and certainty. Each theme indicated the physical, social and emotional impact it had on mothers and their children receiving GHT. The following chapter will discuss the findings in relation to the related theoretical concepts and literature examined after the themes were identified.

## Chapter 5: Discussion

### 5.1 Introduction

The previous chapter presented the findings of mothers' lived experiences caring for their child receiving growth hormone treatment (GHT). Using hermeneutic phenomenology based on the principles of Gadamer's (1975) philosophy, the research concentrated on gaining a deeper understanding of mothers' first-hand experiences. The aim was to enter the 'lifeworld' of the parents and understand the meaning or 'essence' attached to their experiences. The mothers' experiences provided a wealth of data that represented three overarching concepts of 'uncertainty', 'stigma' and 'normalisation' that are central to the four major meanings of their lived experiences; (1) "It's the right thing to do" Striving for the security and the wellbeing of the child, (2) "Doubting yourself constantly" Constant uncertainty, (3) "But then you just get used to it I suppose" Adhering to GHT and lifestyle changes - the new normal, (4) "I hadn't been told anything about it" Information behaviour; looking for normality and certainty, as discussed in the previous chapter.

This final chapter critically discusses the findings and focuses on these three overarching concepts as they appeared as the main characteristics of the mothers' lived experiences. Due to their importance and in keeping with Gadamer's (1975) principles of hermeneutic phenomenology, additional consideration of the literature related to the concepts of 'uncertainty', 'stigma' and 'normalisation' was undertaken. A theoretical framework and a concept analysis were identified from the literature, these being Mishel's uncertainty in illness theory (Mishel 1988), reconceptualised theory of uncertainty in chronic illness (Mishel 1990) and Deatrick *et al.*'s (1999) five attributes of normalisation. Both were found to address key theoretical perspectives and shared some meaning with this study's findings. However, grounded on Gadamerian principles of exercising new and pre-existing knowledge to examine and interpret the data, new important findings relating to the 'social stigma uncertainty' was identified. As a result, a new model based on this study's findings and integrating aspects of Mishel's (1988, 1990) and Deatrick *et al.*'s (1999) work was created. This new model, seen in Figure 11

(in section 5.5) symbolises a fusion of the researched and researcher's thoughts and interpretations in what Gadamer (1975) terms as a 'Fusion of Horizons'. It delivers an original and alternative framework for healthcare professionals to understand parents and family's needs when caring for children commenced on GHT.

This chapter begins with an overview of the concept of uncertainty in both Mishel's (1988) original uncertainty in illness theory and Mishel's (1990) reconceptualised theory of uncertainty in chronic illness, followed by a critical discussion of the study findings under each of the original model's main concepts which are antecedents, appraisal and coping with uncertainty. Subsequently an outline of the concept of normalisation using Deatrick *et al.*'s (1999) five attributes of normalisation is outlined, and then followed by a critical discussion of the study findings under each attribute. Finally, the chapter concludes by considering the clinical implications and recommendations for practice and research and outlines the study's limitations.

## **5.2 Uncertainty**

The discussion will initially contemplate the findings of this study based on the concept of uncertainty and will critically discuss the reactions and coping strategies used by the mothers as they adjusted to caring for their children receiving GHT. It is significant to note, where this study's findings concur with existing studies the emphasis will be on the new inputs to knowledge concerning the mothers' experiences of caring for their children receiving GHT.

Mishel's original theory of uncertainty in illness (1988) is grounded on a cognitive appraisal theory and includes the work of several theorists. Factors both within the person and characteristics of the event initiating stress (stimuli) influence the perception of illness-related events. Initial appraisal happens when a person allocates meaning to the stimuli. Uncertainty occurs when the individual is incapable of assigning meaning and value to events and is unable to foresee outcomes (Mishel 1984). Mishel (1988) later developed her conceptualisation work on uncertainty and subsequently published her uncertainty in illness theory (Mishel 1988), which addressed the uncertainty occurring when individuals and caregivers confront doubt during the treatment and

diagnostic of an illness. Mishel's (1988) original uncertainty in illness theory was later reconceptualised to incorporate the constant uncertainty experienced by those with a chronic illness which not only implicates the person affected, but those caring for the individual as well (Mishel 1990). Mishel's (1990) reconceptualised theory addresses the incorporation of uncertainty in chronic illness into an individual's life. The reconceptualisation effort was primarily fuelled by questions about the outcome portion of the original uncertainty theory (1988) suggesting that, living with chronic uncertainty can be adjusted to and can be the favoured state when other options are reflected as having negative outcomes (Mishel 1990). These inferences backup the findings described in section 4.7 as the mother described learning to live with GHT despite their uncertainties. Therefore, in light of the findings of this study described in Chapter 4, much of the theoretical work of Mishel (1988, 1990) echoed with the mothers' descriptions of uncertainty relating to their child's illness and GHT.

Additionally, although Mishel (1988) acknowledged that uncertainty can be a positive as well as a negative concept, her original framework concentrated mainly on the negative aspects of uncertainty, overlooking the fact that sometimes individuals embrace strategies that focus on preserving uncertainty to foster hope. While her reconceptualised theory (Mishel 1990) addresses some of the uncertainties mothers face when dealing with chronic illness, it was recognised that the mothers in this study experienced other dimensions of uncertainty relating to their child's unique need to have GHT. Furthermore, although the mothers' uncertainty was induced by illness of their child and their need for GHT, the uncertainties experienced were from a much wider perspective and needed broader consideration.

The model used in the current study is the model description found in uncertainty in illness (Mishel 1988) (see Figure 2 overleaf) and Mishel's reconceptualised theory of uncertainty in chronic illness (1990) which reconceptualised the outcome portion of the original uncertainty in illness theory (see Figure 3 overleaf).



Figure 2. Mishel's Uncertainty in Illness Model (1988)

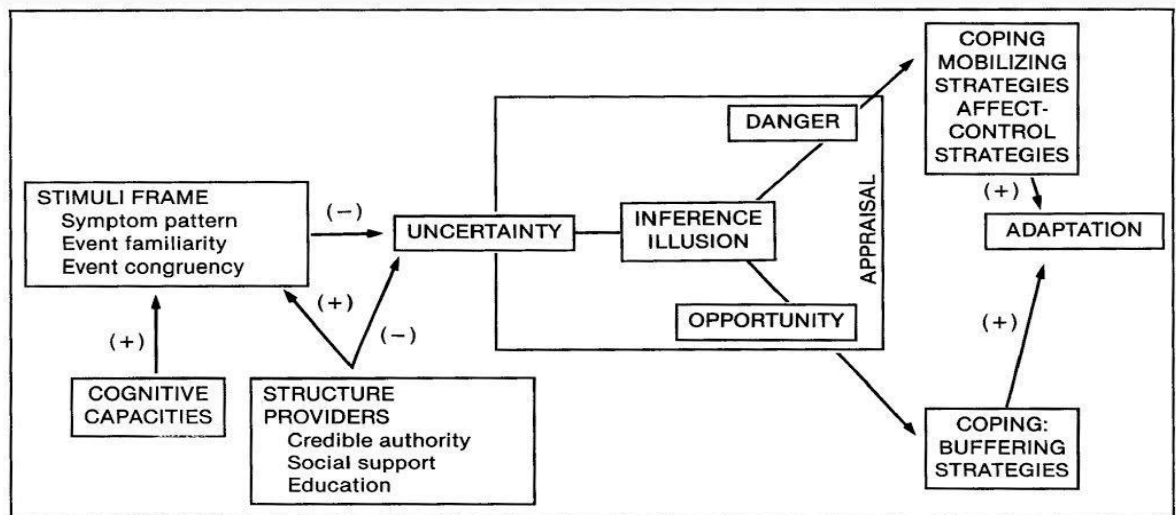
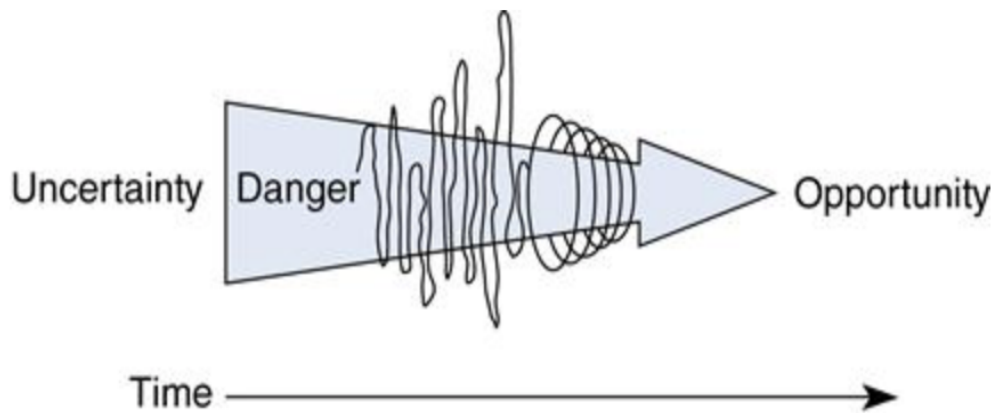


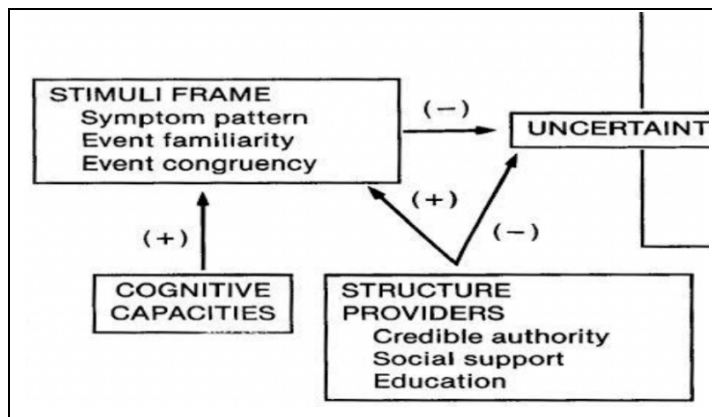
Figure 3. Mishel's Reconceptualised Model of Uncertainty in Chronic Illness (1990)



### 5.2.1 Antecedents of Uncertainty

The first part of Mishel's uncertainty in illness model (1988) (see Figure 4 overleaf) is the antecedents of uncertainty which consists of three components: stimuli frame, cognitive capacity and structure provider and these will each be explained in turn.

Figure 4. Part One of Mishel's Uncertainty in Illness Model (1988) (Antecedents)



### 5.2.1.1 Stimuli Frame

The stimuli frame is the level to which the illness is patterned, familiar, and congruent with expectations (Stewart *et al.* 2010). Symptom pattern indicates symptoms that occur in a pattern, which cause less uncertainty. Individuals process symptoms in the context of their own experiences, cultural, and social cues along with information from a healthcare provider. Symptom appraisal can be delayed when symptoms lack prominence (Mishel 1988, Mishel & Braden 1988). Event familiarity refers to the repetitive nature of the structured environment. Familiarity results from a cognitive process based on experience with the environment. New and novel symptoms or treatments threaten familiarity (Mishel 1988, Mishel & Braden 1988). Event congruence is when there is consistency between what is expected and what occurs. Lack of congruence from expectations result in questions, and challenges the predictability established (Mishel 1988, Mishel & Braden 1988).

The stimuli frame is influenced by the other two components of the antecedents of uncertainty which are cognitive capacity - the ability to process information and structure providers - the resources available to help interpret the meaning of the stimuli (Mishel 1988, Mishel & Braden 1988).

### 5.2.1.2 Cognitive Capacity

Cognitive capacity is the capability of a person to process information. When the

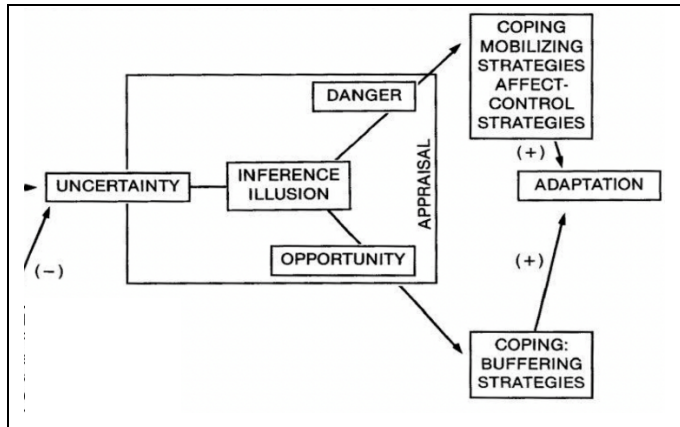
environment is perceived as a threat, cognitive efficiency is reduced, and the capacity to further process cues is lessened (Mishel 1988, Mishel & Braden 1988).

### **5.2.1.3 Structure Providers**

Structure providers in this model are credible authority of healthcare personnel, social support and educational level (Stewart *et al.* 2010). Credible authority is the extent of trust and confidence patients or caregivers have in the capability of healthcare providers to provide care. Trusted, reliable information provided to families and caregivers improves event familiarity and fosters event congruence. Trusting relationships with healthcare providers has been found to decrease uncertainty (Mishel 1988, Mishel & Braden 1988, Mishel 1990). Social support decreases uncertainty by acting as a feedback system to help understand the meaning of illness-related events. Social support reduces uncertainty by adjusting the ambiguity of the illness, the complexity of the treatment and the unpredictability of the future. Having someone to share information with aids in the appraisal process (Mishel 1988, Mishel & Braden 1988, Mishel 1990). Education has both a direct and indirect consequence on perceived uncertainty. Education offered can develop the parent/patient's knowledge about the stimuli frame which helps offer meaning and understanding. The educational level also plays a role in the capacity to modify uncertainty cognitively. Individuals with less education exhibit higher levels and more extended phases of uncertainty due to a lack of ability to comprehend complex treatments and rationale for care (Mishel 1988, Mishel & Braden 1988, Mishel 1990).

## 5.2.2 Appraisal of Uncertainty and Coping Strategies

Figure 5. Part Two of Mishel's Uncertainty in Illness Model (1988) (Appraisal and Coping)



Appraisal of uncertainty is defined as placing a value in the uncertain stimuli (Mishel 1988, Mishel & Braden 1988, Smith & Liehr 2014). There are two components of appraisal which are inference and illusion. Inference signifies the appraisal of uncertainty using related examples and is constructed on personality characteristics, general experiences, knowledge and contextual cues (Mishel 1988, Mishel & Braden 1988, Smith & Liehr 2014). Illusion indicates the construction of beliefs shaped from uncertainty that have an optimistic outlook (Mishel 1988, Mishel & Braden 1988, Smith & Liehr 2014). An appraisal will end in uncertainty being considered as either a danger or an opportunity. Loss or absence of a reliable authority can lead to a danger appraisal in which uncertainty encourages a fight or flight response. When danger is considered, a coping response is required so that it can be reframed into a positive illusion. Because uncertainty renders a situation which is vague, positive oriented illusions can be made from uncertainty, leading to an appraisal of uncertainty as an opportunity. It is considered that uncertainty can be seen as an opportunity when the alternative is negative certainty (Mishel 1988, Mishel & Braden 1988, Smith & Liehr 2014).

With danger appraisal, coping methods are focused on lessening the uncertainty if possible, and handling the emotion caused by the danger appraisal. There are two coping strategies associated with danger appraisal which are mobilising strategies and

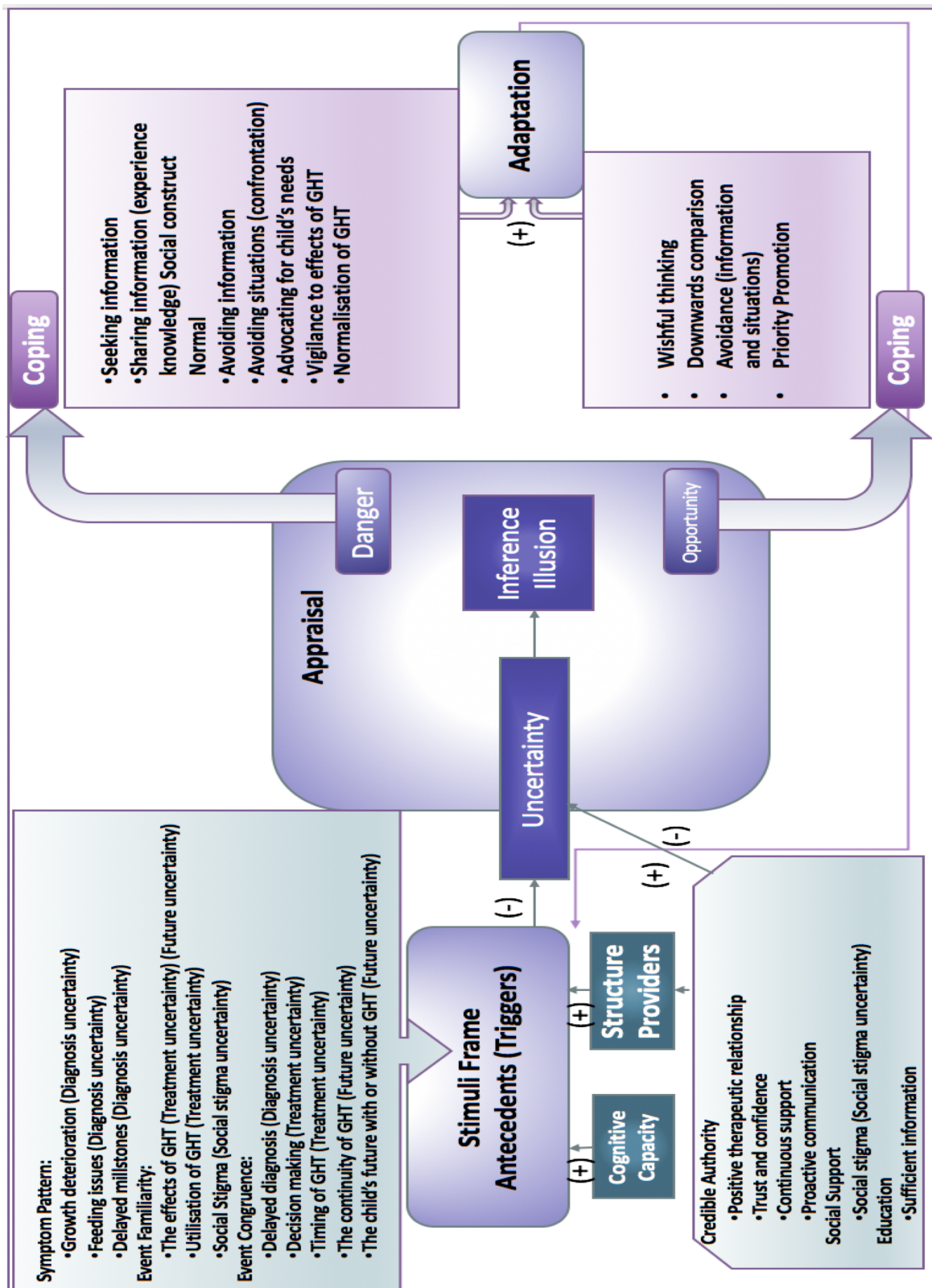
affect-control strategies (see Figure 5). Mobilising strategies include the use of direct actions, vigilance and information seeking, while affect-control strategies include faith, disengagement and cognitive support. Moreover, with opportunity appraisal, buffering methods are used to support the uncertainty (Mishel 1988, Mishel & Braden 1988, Smith & Liehr 2014). Buffering methods are utilised to block the input of new stimuli that could modify the individual's appraisal of uncertainty as an opportunity (Rice 2012). Buffering strategies incorporate avoidance, selective abstraction of information, minimisation of intimidating information and the rearranging of priorities (Rice 2012).

In the model (see Figure 4), opportunity and danger are parallel to each other, signifying the patient selects one and only one route (Mishel 1990). Although this may be suitable for certain clinical situations, it may not correctly reflect the changeability that occurs over the illness pathway and may not reflect the long-term illness situation (Mishel 1990). Over time, an appraisal of uncertainty as a danger, may progress into appraisal of uncertainty as a positive event (Mishel 1990) (see Figure 2). Choice of only one type of appraisal contradicts the idea of appraisal being a process that fluctuates over time. Instead, the theory mirrors a mechanistic orientation to a precise state and not a process (Mishel 1990). In the next section, mothers' experiences and the concept 'uncertainty' will be discussed.

### **5.3 Mothers' Experiences of Uncertainty**

Mishel's uncertainty in illness model (1988) helped to bring an understanding of the response and adjustment seen in the findings. Based on Mishel's categories of antecedents, appraisal and coping and adaptation to uncertainty; these classifications offered a helpful structure to critically discuss the mothers' experiences of uncertainty which is summarised in Figure 6 overleaf.

Figure 6. Mothers' Experiences of Uncertainty Adapted from Mishel's Uncertainty in Illness Model



### **5.3.1 Antecedents (Triggers) of Mothers' Uncertainty**

While theoretical work was undertaken by Mishel (1988, 1990), additional conceptual work on the uncertainty experienced by parents confronting their child's illness was being established by Cohen and colleagues (Cohen & Martinson 1988, Cohen 1993, 1995){Cohen, 1995, The triggers of heightened parental uncertainty in chronic, life-threatening childhood illness;Cohen, 1993, Diagnostic closure and the spread of uncertainty;Cohen, 1995, The triggers of heightened parental uncertainty in chronic, life-threatening childhood illness}. Cohen and her colleague's work were stemmed from a longitudinal study of the impact of childhood cancer on families and concentrated mainly on the nature of parental uncertainty (Cohen & Martinson 1988). Subsequent to the child's diagnosis, Cohen (1993) suggested that uncertainty was no longer limited to the single feature of not knowing what was wrong with their child and extended to other various areas of the parent's life. These dimensions of uncertainty were experienced as existential, etiological, biographical, situational, social and treatment uncertainties (Cohen 1993). Existential uncertainty correlated to the threat to the child's existence or survival and the parents' mindfulness of their child's future being open and undetermined. Etiological uncertainty was outlined as not knowing the cause or origin of the disease. Biographical uncertainty signified the unknown impact the child's illness and diagnosis would have on the personal lives and world of the child, parents and siblings. Social uncertainty related to the changes in the once taken-for-granted relationships between the parents and hospitalised child, the impact on the parents' spousal relationships and effect on the siblings. Situational (or environmental) uncertainty referred to the parents being confronted by an unfamiliar medical environment in which they were confronted by new rules and regulations that determined what they could or could not do. Treatment uncertainty related to the difficult decisions the parents had to make concerning the treatment for their child, which often had to be made under conditions of urgency (Cohen 1993).

In this study, multiple antecedents added to the mothers' uncertainty and following analysis and interpretation of the findings, the mothers uncertainties were noted as arising from the aspects of etiological (diagnostic), biographical (future), treatment and social stigma uncertainty, some of which supported Cohen's (1993) dimensions in her study of parental experiences of uncertainty with chronic childhood illness (life

threatening illness). After re-assessing the literature on the concept of ‘uncertainty’ and ‘stigma’ upon reaching the findings of the study, a relevant connection between the two mentioned concepts was found and a decision to add ‘social stigma uncertainty’ to the mothers’ dimensions of uncertainties was made. This additional dimension adds to the previous dimensions covered under (section 4.6) which were diagnostic, treatment and future uncertainty. Social stigma uncertainty was also one of the themes reported in a study by Kerr & Haas (2014) which explored uncertainty experienced by parents of children with ‘orphan’ illnesses requiring multi- disciplinary care. By adding social stigma uncertainty, a deeper elaboration of the findings was achieved and therefore, a richer understanding of the findings was attained. In addition, it helped highlight one of the three major concepts emphasised in the study which is ‘stigma’. The next sections will critically discuss the study findings under the four concluded dimensions of uncertainty experienced by mothers in the current study which were: diagnostic, treatment, future and social stigma uncertainties.

### **5.3.1.1 Diagnostic Uncertainty**

Symptom appraisal was usually hindered for the mothers, due to the unfamiliarity of the symptoms and the patterns of symptoms associated with the majority of the children’s conditions (section 4.5.1 and 4.6.1). With many of the children not being diagnosed soon after birth, many mothers were trying for many years to find answers to why their children were ‘different’ and not growing normally despite their efforts (section 4.5.2 and 4.6.1). They sought medical advice, and many were still left with heightened diagnostic uncertainty several years before the child was finally diagnosed (section 4.5.1, 4.5.2 and 4.6.1). The delay in diagnosis was mainly due to some of the manifestations of the growth disorders not being very well defined soon after birth, such as growth hormone deficiency (GHD) and Turner syndrome (TS). This delay in diagnosis has been noted in previous studies as one of the leading problems with managing these conditions (Stochholm *et al.* 2006, Saari *et al.* 2012, Murray *et al.* 2016).

Symptoms such as feeding issues and growth restriction with unknowing aetiology for a considerable amount of time and developmental delays in reaching normal milestones



were reported (section 4.5.1 and 4.6.1). While mothers compared their children's growth and development with what their perceived familiar 'normal' was and to societal expectation and by noting the discrepancies, the lack of congruence heightened mother's uncertainty which led them to seeking healthcare providers' help (section 4.5.1, 4.5.2 and 4.6.1). Many mothers were left to care for their children and implement unfamiliar treatments such as initiating feedings through a nasogastric tube (NG) or a percutaneous endoscopic gastrostomy (PEG) (section 4.6.1).

Diagnostic uncertainty was an essential element for the mothers and many of their stories and descriptions mention the uncertainty associated with the diagnostic stage. Many waited for an extended period with no confirmed diagnosis as to why their children were different (section 4.5.2 and 4.6.1). Delayed diagnosis in GHD is often due to the diagnostic process being multifactorial (Murray *et al.* 2016). The lack of any 'gold standard' test for GHD and/or the fact that other disorders may share similar auxology - is the science of human growth and development (Hermanussen & Boging 2014) - to GHD, could be the reason (Murray *et al.* 2016). Some mothers were given reassurance that the children would catch up in growth sooner or later (section 4.6.1). The false reassurance may have been due to the guidelines that provide leeway for the child to catch up in growth up to four years, especially for children born small for gestational age (NICE 2010). It also may possibly be that the healthcare providers were using the observation and reassurance technique, which is one of the strategies used to treat short stature with no prominent cause (Deal *et al.* 2013).

Another cause of diagnostic delay perceived by mothers was being given a different diagnosis altogether (section 4.6.1). Some mothers questioned the legitimacy of the measuring techniques used by the general practitioner (GP), which in their opinion could have caused unnecessary delays. This observation by mothers could be explained by the fact that Ireland continues to lack standardised growth monitoring and referral guidelines for GHD in children (Hawkes & O'Connell 2016). Perceived unnecessary delays were noted although it is recommended that referrals should be made in the first instance to the GP, then to the local paediatric service if no apparent reason for the problem is recognised (HSE 2012). However, mothers questioning the growth measuring techniques used by the GP, signposts a preventable delay in referral to specialised services and receiving the appropriate medical treatment.

The findings helped to elaborate on the burden faced by mothers of children with different growth disorders as they journeyed the road of finding answers to their uncertainties. Some mothers were dismissed entirely, and their concerns regarding their child's growth were ignored by the healthcare providers (section 4.5.2 and 4.6.2). This made them feel 'stupid' and 'over the top' and increased their uncertainties. They sometimes challenged the healthcare providers' decisions and, in many cases, if they had not done so, they firmly believed that their children would still be undiagnosed or diagnosed at a much later stage than when they were, which resonates with the findings in another study by Thomlinson (2002). Thomlinson (2002) examined the lived experience of twelve families of children who were failing to thrive and found that parents devoted great effort, energy, time and financial resources into seeking a diagnosis and treatment for the underlying condition that was affecting their child's growth. The efforts are similar to the efforts demonstrated by families of children with cancer or other chronic, life-threatening illnesses (Cohen 1995). The findings revealed that mothers sought to be heard and wanted their concerns to be acknowledged by healthcare providers but felt let down. This also adds to the extent of burden mothers had faced long before receiving a diagnosis for their child and commencing GHT.

This study suggests that there remains a critical need to minimise diagnostic delay to assist families that are at a risk of delay-associated distress which was also noted by Marini *et al.* (2016), who found that parents caring for children with GHD experienced emotional distress waiting for the diagnosis of their child's condition. This also resonates with Evans *et al.*'s (2015) findings that families experiencing delays expressed guilt and anger and adverse impacts on the family-clinician relationship and believed delays impacted on treatment and prognosis. The effects of the diagnostic experience in children with growth failure can be considerable on mothers as the period before receiving a diagnosis has been known to provoke significant uncertainty for parents (Lenhard *et al.* 2005). It was also noted by Brod *et al.* (2017) who examined the burden of GHD on children and adolescents and concluded that the early identification and commencement of GHT might lead to fewer impact in terms of their psychological and psychosocial wellbeing.

Misdiagnosis, underestimating the issue, lack of consensus among medical professionals and/or dismissing mothers' concerns (section 4.5.2 and 4.6.1) were

challenges experienced by mothers seeking a diagnosis for their children with a hidden or unknown chronic illness (Kerr & Haas 2014, Mihelicova *et al.* 2016, Benson *et al.* 2017) . There is evidence that supports the effects of a prolonged diagnostic stage and that parental uncertainty can cause anxiety, depression, cognitive disturbances, helplessness and fear (Jessop & Stein 1985, Grootenhuis & Last 1997, Hinton & Kirk 2017) which has substantial health costs (Mitchell & Hauser-Cram 2008, Raphael *et al.* 2010). It appears that when parents perceive greater uncertainty, they sense less control over their child's condition (Lipinski *et al.* 2006, Madeo *et al.* 2012). This could justify the felt frustration, anger and anxiety by many mothers in the current study due to the prolonged diagnostic stage in reaching a final diagnosis (section 4.6.1).

The credible authority of healthcare providers was questioned, and many times their decisions were challenged by the mothers (section 4.5.2 and 4.6.1). The lack of understanding alleged by some mothers regarding professionals (GP) not having enough knowledge about their children's condition influenced the delay in getting their children correctly diagnosed (section 4.6.1). The defect in the structure provider (credible authority) along with the lack of support and information provided (education) all heightened their uncertainty (section 4.8). These findings also resonate with the professional uncertainty noted in lived experiences of parents living with childhood multiple sclerosis (Hinton & Kirk 2017).

All mothers received their child's diagnosis with desolation, however it did not come as a shock for many as they watched and lived through the experience of their child failing to grow, which was also noted by Marini *et al.* (2016). While mothers who have children with a rare genetic disorder that contributed to the lack of growth, the diagnosis came as a shock and this, in turn, may have affected their cognitive capacity to process information. This may have resulted in raised uncertainty at the time of diagnosis which was again noted by Evans *et al.* (2015). Regardless of the cause or attribute of the uncertainty during the diagnostic stage, failing to address the psychological impact of delays may hinder families' capacity to adjust to the diagnosis and may have lasting implications for treatment and/or screening adherence (Oeffinger *et al.* 2006).

### 5.3.1.2 Treatment Uncertainty

The uncertainty faced during the diagnostic phase resulted in doubt about GHT for some mothers (section 4.7.2) which was also reported in a study with adult survivors of childhood cancer (Oeffinger *et al.* 2006). Growth hormone treatment (GHT) is a controversial treatment especially with the notion of it possibly causing cancer (Conrad & Potter 2004, Hintz 2004, DiVall & Radovick 2013, Nicholls *et al.* 2017). It has been used for other biomedical enhancement or non-medically unlicensed reasons such as to enhance sports performances which are called GH doping (Foddy & Savulescu 2017). With many controversial issues surrounding the treatment, and the novelty of the treatment, the unfamiliarity and taboo attached to it, could have influenced the mothers' sense of uncertainty to the treatment (section 4.6.2 and 4.8.2). This finding has not been reported before which makes it a novel contribution to the current knowledge in this area. Also, time was an essential element in regard to the starting of treatment. Many mothers felt uncertain about the timing of commencing GHT. The lack of consistencies about when to start in the GHT guidelines noted between countries and between cases made them question the accuracy of their child's GHT commencement timing. Also, the delay in diagnosing augmented their uncertainty about the accuracy of GHT commencement timing.

The lack of congruency of the treatment's positive outcomes when deciding to start GHT initiated uncertainty in mothers (section 4.5.3 and 4.6.2). Also, concerns about potential harmful effects from the treatment, occurring in the near or far future was another issue raised that heightened their uncertainty (section 4.5.3, 4.6.2, 4.6.3, 4.7.4 and 4.8.1) which echoes with Marini *et al.*'s (2016) findings that reported parents worry about GHT side effects and are not well informed about it. Marini *et al.* (2016) suggested that a lack of reassurance could negatively affect adherence to GHT which was also noted by Laing (2014). Since the commencement of GHT the mothers have lived with the struggle between the desire to offer to their children the solution of the growth problem, and their uncertainties regarding the hormonal administration (section 4.5.3 and 4.6.2). They spoke about their need for more reassurances about the outcomes of therapy and their children's future (section 4.8). In a long process, such as the treatment for GHD, which entails the need for patients to be patient to see results and persistent when faced with discouragement at times of little growth, the felt uncertainty

can threaten adherence to the therapy (Marini *et al.* 2016). If families are not confident and reassured about the treatment, they can choose to abandon the treatment, since results do not always show from the very beginning (Marini *et al.* 2016). This was evident in the findings of this study as one particular mother did not witness any positive results since her daughter commenced GHT and felt her child's health (diagnosed with PWS) had deteriorated and wished to discontinue GHT (section 4.5.5).

Lagrou *et al.* (2008) also concluded that parents feared GHT side effects before commencing their children who were born small for gestational age (SGA). Grimberg *et al.* (2015) also looked at parental concerns regarding child growth and factors that drive their decisions to intervene or not with their child's height medically. Parents remarked that GHT efficacy and side effects and child health and psychosocial function as a child and as an adult, had the most considerable influence on their decision to start GHT. These previous findings also resonate with the concerns of the mothers regarding the risks of GHT which illustrated the complex dilemma resulting from inadequate and inconsistent information about GHT and the future impact. The lack of information and support as they were deciding to commence GHT was reported many times. Having a strong alliance with the healthcare provider has been associated with higher levels of wellbeing, and greater perceived healthcare provider support (Trevino *et al.* 2013). Information from healthcare providers assists in reducing ambiguity, complexity, and unpredictability of illness and treatment, resulting in increased understanding and lowered uncertainty (Clayton *et al.* 2018). The lack of support and information from healthcare providers reported by the mothers (section 4.8) is consistent with the result of previous studies that information support from healthcare providers can influence uncertainty (Marini *et al.* 2016, Tiwaree *et al.* 2016).

The worry that GHT may heighten the pre-existing concern of 'being different', as one mother described GHT as being a daily reminder of her son being short - illustrates the uncertainty attached to the possibility of adverse psychological effects resulting from the child being treated with GHT (section 4.5.3). This notion of medicalisation of short stature contributing to psychosocial problems has been reported elsewhere (Conrad & Potter 2004, Naiki *et al.* 2013). The length of treatment was another aspect considered by mothers when commencing their children on GHT. However, the duration of treatment was mostly unknown for most of the mothers and an additional reason to feel

uncertain about the novel treatment. The lack of information in this regard was noted, and the answer to this question was left unanswered for many (section 4.5.6, 4.6.3 and 4.8.1). This again could hinder their adaptation to the treatment and reduce their adherence which is an area under researched.

Furthermore, the lack of support and understanding from others was seen by mothers as hugely associated with the stigma attached to GHT (section 4.5.2 and 4.8.2). The need to justify to others the need for the treatment when the child was healthy but small made it hard for mothers to disclose to others. The disclosure of the treatment proved to be even harder as time passed and the positive effects of GHT had been witnessed by others, but the child still required GHT. They claimed that since the visible stigma 'short stature' was not there anymore, people did not understand, and mothers felt the need to justify having their child on GHT. This also echoes with parents' experiences caring for children with chronic conditions such as multiple sclerosis (MS) (Hinton & Kirk 2017). They also faced interaction uncertainty as social interaction exacerbated their sense of uncertainty about their child's illness, because children and young people with MS frequently looked visibly well, so parents found it difficult to convince others that their children were ill and had specific health needs (Hinton & Kirk 2017). However, this finding in the current study contributes new knowledge and understanding with regards to mothers caring for children receiving GHT. This result is significant as it hindered some mothers from seeking support from family and friends which resulted in isolation, also noted in studies about other conditions (Benson *et al.* 2017). Lack of public understanding and knowledge around growth disorders and GHT was noteworthy in the current study. The negative public perceptions of GHT challenged coming to terms with the treatment for some mothers. Consequently, this is an area worthy of further research to explore the public understanding about GHT and its consequences on adaptation for patients and their families.

Grimberg *et al.* (2015) looked at parental apprehensions about child growth and factors that drive their decisions to intervene or not with their child's height medically. Parents reported that GHT efficacy and side effects and child health and psychosocial function as a child and as an adult had the most significant influence on their decision to start GHT which were also confirmed in the current study. Similarly, Guerriere (2003) explored these four factors contribution to mothers' uncertainty concerning gastrostomy

tube (G-tube) insertion for their children. The four factors were lack of information; unclear value trade-offs; lack of support; and social pressure in a temporary decision-making context. Mothers confirmed the existence and importance of these four factors in their experiences. Lack of information, need for experiential knowledge, taking a chance, balancing pros and cons, lack of support from healthcare providers, and social pressure were all findings that are congruent with this study. However, the social pressure felt by mothers in the current study was not perceived from healthcare providers as Guerriere (2003) suggested, but was from the social stigma attached and the need to try to normalise the child as previously mentioned (Grimberg *et al.* 2015). Nevertheless, mothers also reported agreement and disagreement from family members and friends similarly reported by Guerriere (2003) which sometimes challenged their decision-making process. However, despite these uncertainties, challenges, and feeling abandoned, they commenced their children on GHT. The process of decision-making appeared to place pressure on parents as they made a joint decision, therefore, this is another area that can benefit from further research.

Some mothers (section 4.7.1) testified a lack of choice in picking the GHT device that best suited their needs which was previously noted in Ireland, UK and the Netherlands (Langham & Kirk 2011, Van Dongen & Kaptein 2012, Laing 2014, Hawkes & O'Connell 2016). Van Dongen & Kaptein (2012) and Laing (2014) recognised the need for support and participation of parents in the practice of selecting a GHT device. Lack of choice in selecting an appropriate device has a significant implication for healthcare professionals. The role of the paediatric endocrine nurse specialist can be essential in assisting patients and parents in choosing the most suitable growth hormone device to meet their needs and to offer continuing support during the GHT pathway, and in turn enhance adherence (Laing 2014). Issues that are believed to be modifiable such as lack of information, lack of support and lack of choice, means healthcare professionals have the opportunity to possibly minimise the extent to which these factors contribute to uncertainty. Acerini *et al.* (2012) recommended a three-step approach for a more successful decision-making process for patients commencing GHT, which involves GHT education, device selection and follow-up after making a choice. These steps can be harmonised within the scope of the endocrine nurse specialist role. Further investigation into the decision-making process in the context of GHT is suggested.

Growth hormone treatment (GHT) implementation uncertainty (section 4.7.1) is a noteworthy finding that emerged from this study. Soon after starting GHT and as mothers and children were adapting to the new treatment, mothers encountered many technical and organisational issues to the treatment. Reducing injection pain, disposing of needles, and storing and traveling with GHT were all issues where mothers needed extra support and information. Brod *et al.* (2017) noted GHT burden for children diagnosed with GHD and their parents and found that parents worried about administration, inflicting pain on their child, and GHT costs. They felt frustration with injection administration and interference with family travel and travel planning. Marini *et al.* (2016) also concluded that parents of children with GHD had difficulties with GHT and its organisational issues. They reported GHT being painful and causing bruising and stinging in addition to storage inconvenience both at home and while traveling (Kremidas *et al.* 2013).

The lack of instructions or the inconsistency in the instructions on how to dispose used needles (section 4.7.2 and 4.8.1) caused additional frustration for many mothers in this study. The need to organise and plan to order needed supplies for GHT from multiple sources added to the burden of treatment. For example: asking for and receiving a prescription for GHT from an endocrinologist at follow up appointments (every six months); ordering medication cartilages and needle tips from a local pharmacy; ordering needle tips (if not requested from the pharmacy), lithium batteries, needle shield or a replacement of the injection device from the GHT drug manufacturing company. Also, the process of disposing of the used needles was also vague for some. Many mentioned the GHT drug manufacturing company connecting them with a company that provided a service to collect the filled needle disposing container. Others suggested bringing the filled container to a hospital to empty it there because they did not know any other way. Marini *et al.* (2016) also mentioned organisational issues, but the current study gives detail to the organisational burden associated with GHT to help target these problematic issues especially in the Irish context where this is occurring. It has been suggested by Laing (2014) that longer durations of GHT prescriptions, prescription collection and home delivery, reminders and calendars, in addition to immediate and extended family and social support, can help children and their carers to continue with GHT. Further research around methods to address these organisational issues is suggested to minimise associated GHT burden.



The change in the storage guidelines concerning a specific GHT drug caused some doubt for a few mothers (section 4.7.2). Not being allowed to store the medication outside of the fridge after the first use and then later being able to do so, aggravated their sense of uncertainty because they did not understand why it was now allowed, so the gap in knowledge again triggered uncertainty in mothers in the current study. Although mothers had sufficient teaching, support and follow up after the initiation of GHT, many described the support being stopped at some point (section 4.8.3). This implies the need for ongoing support from the drug company nurse or specialist paediatric endocrine nurse whose goal is to provide support using a variety of interventions and evidence-based practice (Laing 2014).

Many of the findings of this study are similar to other studies of parenting a child with complex medical needs. In Nygard & Clancy's (2018) metasynthesis of parents' experiences of caring for a child with special health-care needs at home, one of the eight categories that emerged from the synthesis titled "Managing the front line-a constant battle" confirmed that parents carry a huge responsibility for giving children with special health-care needs the best possible care. This responsibility involves understanding the child's health condition, organising and transporting to and from health appointments, providing information to health specialists, school care and administering medications and medical procedures. The parents defined their duty as overwhelming and time-consuming, and felt unprepared for the extent of obligation they had to take on and to meet the child's needs, parents had to navigate the health-care system, support organisations and the internet for information. "Someone to share my responsibilities with" was another key category in Nygard & Clancy's (2018) metasynthesis who concluded that support from family, friends, people in parallel situations and district nurses was expressed as necessary for motivation, relaxation and relief in parents' everyday lives. Communication and support from health-care professionals were also noted. Many parents had negative experiences of meetings with healthcare professionals. They described being ignored and mistrusted, which caused stress and a sense of despair. The quality of the relationship with health-care professionals was reflected as very essential.

In another review of parents' experiences of living with a child with a long term

condition (Smith 2015), it was noted that in order to take control of their child's condition, parents wanted knowledge of the condition and treatments; to learn from illness episodes and to use these understandings to recognise and respond to following illness symptoms in their child; and to improve effective relationships with healthcare professionals. Parents sought information about the disease and treatments; accessing services and support networks; and approaches that would help them cope. Parents explained difficulties in gaining information and many were disappointed with the information provided by health professionals. Parents developed significant expertise in handling their child's condition. They anticipated to be involved in care decisions but did not necessarily want exclusive responsibility for such decisions. Parents' also acknowledged communicating with professionals as stressful. These findings from both Smith (2015) and Nygard & Clancy (2018) were also evident in the current study.

### **5.3.1.3 Future Uncertainty**

Mothers struggled with the uncertainty to forecast a 'normal' future for their child. Future matters that were related to the child's health, functioning and wellbeing were also disrupted by the sense of insecurity. The future of the child, the length of GHT and the continuity of healthcare services as they transition to adulthood was questioned by some mothers (section 4.5.6 and 4.6.3). Mothers had experiences of future uncertainty regarding the impact of the condition and GHT may have on the child's ability to be 'normal' in the future. They thought of potentially harmful circumstances that are unknowing about the future (section 4.6.3) which was similarly found by Kerr *et al.* (2014) who explored uncertainty experienced by parents of children with 'orphan' illnesses requiring multidisciplinary care. Some mothers were concerned about how GHT could affect the child's functionality as adults and which would hinder their ability to pursue a healthy lifestyle regarding traveling, going to college, socialising and disclosing to others (section 4.6.3). Similarly, in Hinton and Kirk's (2017) study that explored the lived experience of parents caring for children with multiple sclerosis, parents' accounts proposed that they were worried about their child's ability to attain culturally normative milestones, such as moving out of the family home. This created continuing uncertainty about parents' future role and involvement in their child's illness. These findings also correlate with the wider literature of parenting a child with

complex medical needs. In Nygard & Clancy (2018) study the category “Always at the back of my mind” explained parents constant worry about the future. Parents worried about the progression of the disease and their inability to act. Parents also had difficulty letting go of the sick child, due to their reluctance to hand over the responsibility for medical care to others. A further concern was the child’s future ability to cope with studies, work and moving away from home.

The length of treatment was also an unclear topic, as most mothers were uncertain how long their children would remain on GHT (section 4.5.6 and 4.6.3). This could be caused by the nature of GHT monitoring and the possibility of its discontinuation at any time depending on the response to treatment and adherence (NICE 2010, Kirk 2012). In addition, mothers questioned the ability of their children to continue GHT if they reach the awkward stage of adolescence which could threaten a successful continuation of GHT regimen and therefore treatment positive outcomes. Their children’s acceptance or refusal of GHT when they reached adolescence was unpredictable. This echoes with the literature that adherence patterns change during the GHT pathway, with teenagers having the lowest rates (Rosenfeld & Bakker 2008). Laing (2014) mentions that barriers to adherence could be due to the psychosocial and emotional demands of adolescence, shared with boredom and lack of understanding and awareness of the long term effects and outcomes of the treatment together with its long term nature. Van Dogen & Kaptein (2012) explored the opinions of parents (n=69) in the Netherlands regarding GHT and examined their beliefs and perceptions about GHT and level of communication and support received from healthcare professionals. They also reported that parents noted an increase in the unwillingness of their adolescent children to accept GHT. The parents also suggested and welcomed psychological support to overcome their adolescents’ reluctance (Van Dongen & Kaptein 2012). The felt uncertainty about the successful continuity of GHT when children reached adolescence coming from the mothers indicates the need for healthcare providers to equip mothers with relevant information as to what to expect at this stage of development before it is reached (section 4.5.6 and 4.6.3).

The long-term consequences of GHT as one mother stated ‘is there a price to pay?’, illustrated the constant worry about the potential side effects appearing later on, was apparent (4.5.3, 4.5.5, 4.5.6, 4.6.2, 4.6.3 and 4.7.4). This was also previously reported

by parents in other studies (Lagrou *et al.* 2008, Grimberg *et al.* 2015, Marini *et al.* 2016). However, of note in the present study was that mothers felt reassured when they witnessed the positive effects of the GHT such as an increase in height and muscle strength. When the child manifested an unusual or unfamiliar symptom such as headache or hip pain, their worry about GHT side effects and their sense of uncertainty about the treatment were heightened again. So, it appears that uncertainty felt by mothers could fluctuate depending on the outcomes of the GHT, and healthcare providers should note these times of uncertainties and try support and reassurance.

Transition is the phase when paediatric endocrinologists should support and facilitate their patients' transition into the adult world and when adult endocrinologists should gain the confidence of these new patients and their paediatric endocrinologist (Cook *et al.* 2009). Lack of communication between the paediatric and adult endocrine services and lack of organised care for these patients is common (Cook *et al.* 2009). A high number of patients with growth hormone deficiency (GHD) like others with chronic conditions, drop out of therapy during the transition period. This is due to many factors such as lack of understanding of the disease process, inadequate knowledge of treatment options, and the patient becoming more independent and requiring interaction with a new set of health-care providers (Hauffa *et al.* 2017). It was suggested that education regarding disease management and treatment options should be available from an early age and right through the transition period (Hauffa *et al.* 2017). Children in the current study were prepubertal and their mothers were not prepared in advance for the transition period despite their curiosity, which caused ambiguity and uncertainty (section 4.5.5, 4.5.6, 4.6.3 and 4.9.1). This indicates the importance of discussing the possibility of the continuation of GHT and transitioning into adult services with parents of children with GHD and Prader Willi syndrome (PWS) from an early stage.

#### **5.3.1.4 Social Stigma Uncertainty**

Goffman (1963) conceptualised stigma as an indication that someone lacks social acceptance. When children do not grow and meet the expectation that their family and society hold, there are consequences for the child and family. The sense of protection in the findings of this study was prominent as mothers drew their attention to delays in

their children's development and growth. The children being underweight and skinny proved to be stressful for mothers. This may be due to their roles being questioned as being nurturing mothers and a sign of neglect on their behalf (Wright 2000, Surkan *et al.* 2014) as mentioned previously in (section 4.5.1).

Constant comparison was usually made by family members, friends, strangers and healthcare professionals regarding the child's size and what was expected of a child that age (section 4.5.1). The attitude and comments of others in the community often created more distress on mothers due to their lack of understanding (section 4.5.1). The impact of a chronic condition such as short stature on the wellbeing of children has been studied, with most of the studies looking at the psychological effect of short stature in children using parental reports of stress and adjustment problems in the child (Garganta & Bremer 2014). In a study by Sommer *et al.* (2017) the health-related quality of life for children with short stature was explored and found that the highest number of statements made by the children and parents were linked to social and emotional needs and concerns. Children emphasised about social exclusion and being teased and bullied by peers, similarly reported by mothers in this study (section 4.5.1). Some mothers tried to help their children blend into the society as much as possible to minimise isolation by encouraging them to attend social events and activities. However, this was difficult for some children, and instead, they avoided these situations (section 4.5.1). This is consistent with the findings of previous studies that found short stature to be associated with stigmatisation and a high degree of social isolation in comparison to taller peers (Bullinger *et al.* 2013, Naiki *et al.* 2013, Sommer *et al.* 2017). This was not applied to all children as many were praised for their confidence by their mothers (section 4.5.1) which was also reported by Naiki *et al.* (2013) who concluded that although the children with short stature were not satisfied with their height, this did not hinder their everyday life activities.

Most mothers faced not only social stigma about the child's condition but also with their child's GHT. Worrying about how people would react to their child's condition and GHT caused avoidance and isolation for some mothers (section 4.8.2). As discussed previously with GHT being a controversial treatment (5.3.1.2), other unauthorised and abusive uses of GHT such as sports doping and rejuvenation treatment could overshadow the significance of the treatment for medical indications as for the ones

mentioned in the present study (Conrad & Potter 2004, Hintz 2004, DiVall & Radovick 2013, Nicholls *et al.* 2017). Similarly “suffering in silence” was a category that resonates with the current study which noted that parents were scared that others had negative preconceptions (Nygard & Clancy 2018). The child’s special needs were subsequently a taboo that parents avoided speaking about. Many parents selected not to tell others, not even their close family or the child’s teacher about the disease, as they dreaded that the stereotypical attitudes would harm the child. In the current study, it was felt that some mothers were uncertain about how people would react to the disclosure of GHT because of the notion of the treatment being unfamiliar to people and/or being undervalued due to the scandal of doping. This social stigma uncertainty could have heightened their own uncertainty about the treatment and made them question the need for treatment (section 4.5.3, 4.6.2 and 4.8.2). Thus, further research into the idea of growth hormone abuse and its psychosocial effects on children receiving GHT for medical reasons and their caregivers is recommended.

This section has discussed the findings of this study in relation to the antecedents of uncertainty from Mishel’s (1988) model. The next section will discuss the findings in relation to part two of Mishel’s (1988, 1990) models (see Figure 5) which is appraisal of uncertainty and coping strategies.

### **5.3.2 Mothers’ Appraisal of Uncertainty**

Adaptation to uncertainty is directly influenced by how individuals appraise the uncertainty and how they implement ways to cope (Mishel 1990, Stewart 2000). The appraisal will end in uncertainty being considered as either a danger or an opportunity (Mishel 1988). As formerly indicated it is known that the costs of uncertainty are reliant on the appraisal of uncertainty (Stewart 2000) and the approaches used for coping are supported by the assessment of whether it is seen as positive or negative. Illustrative quotations show how mothers appraised situations which had an impact on coping strategies, evident throughout Chapter 4 in the two categories of: danger and opportunities. A wide range of coping strategies used by mothers are now considered in the next section under the coping section of both Mishel’s uncertainty in illness model (1988) and reconceptualised model of uncertainty in chronic illness (1990).

### 5.3.3 Mothers' Coping and Adaptation

The literature on uncertainty in illness focuses very much on how individuals engage with coping strategies to either reduce uncertainty such as evidence seeking behaviours or foster uncertainty by avoiding activities that could lower their hope (Stewart 2000). In the original - Mishel's model of uncertainty in illness (1988) - coping strategies used with danger appraisal are mobilising strategies that include the use of direct actions, vigilance and information seeking, while affect-control strategies include faith, disengagement and cognitive support. Moreover, with opportunity appraisal, buffering methods are used to support the uncertainty (Mishel 1988, Mishel & Braden 1988, Smith & Liehr 2014). Individuals use problem-focused strategies when the uncertainty is assessed as opportunity and emotion-focused approaches when there is a seeming danger (Mishel 1997). However, Penrod (2001) challenged this, stating that it is not always the case and proposed that behavioural approaches are also used to manage uncertainty along with cognitive and emotive strategies. Recognised emotive strategies for coping, comprised the mothers seeking information and support from healthcare providers similar to other parents who have been through similar experiences (Green *et al.* 2015).

In Smith's (2015) structured review of parents' experiences of living with a child with a long-term condition, adaptation and coping was recognised as a common feature of caring for a child with a long-term condition. Parent's adjustment emerged to be an ongoing process because of ongoing changes in their child's condition and stage of development, together with changeable family needs. The internet has been reported as the leading source of information for parents seeking information about GHT (Cousounis *et al.* 2013). Evidently, mothers used both problem-focused and emotion-focused approaches as they requested information from the internet and through social media to seek information on future expectations with regard to the condition and GHT (section 4.8). Yet some mentioned being provided with booklets and pamphlets from the hospital on the medical condition and how to care for a child on GHT. However, some mothers found them of minimal value and proposed alternatives such as DVD's and/or talking to other parents who have children receiving GHT as being more helpful. The need for experiential knowledge from other parents caring for a child receiving GHT was a notable finding. Most mothers wished they could have had this support

especially at the decision-making stage. These recommendations together with Cousounis *et al.* (2013) recommendations that clinicians provide parents with tools to critically evaluate the content on the internet and a list of pre-reviewed websites or their material could considerably help.

The behavioural strategy adopted by the mothers was constant vigilance (section 4.7.4). Vigilance as a concept that has been formally recognised as one of the coping strategies often embraced by parents when facing uncertainty in illness (Lee & Lau 2013, Meakins *et al.* 2015). Mothers have also been found to implement vigilant behaviours mainly when children are at risk of unpredictable symptoms, and their survival is considered precarious (Dodgson *et al.* 2000, Sallfors & Hallberg 2003, Sullivan-Bolyai *et al.* 2003). The mothers reported similar behaviours such as worrying about the appearance of negative and unwanted side effects of GHT especially ‘tumours’ and ‘cancer’. However, mothers were always vigilant not only about the adverse effects but also about the positive effects of GHT. Some mothers continually measured the child’s growth at home, compared their height with others and noted their child growing out of clothes faster than the norm. Vigilance involved monitoring the maintenance of the positive effect of GHT, for example, when the child stopped growing as much or had less energy, mothers asked for an increase in GHT dosage. With this invasive treatment, all mothers expected and hoped to see positive results (section 4.5.5 and 4.7.4). However, as mentioned previously, only one mother was not confident and reassured enough a year after her daughter’s GHT commenced. She did not see a notable change in her daughter’s weight which was the main concern. Instead, she reported GHT worsened the curvature in her daughter’s back. She also conveyed the possibility of abandoning the treatment as a way of coping with the uncertainty. GHT in this case was appraised as a danger to her daughter’s health and to cope, and so she plans to forsake GHT as part of her daughter’s treatment regimen for PWS.

It is established that gaps in understanding hinders the individual ability to cognitively and emotionally process personal circumstances (Dervin & Frenette 2003). They also signify obstacles to health management, for example, informed decision-making (Andsager & Powers 2001) and communication between patients and health practitioners (Faisal *et al.* 2013). As mothers faced health challenges associated with their children’s condition and GHT management, this resulted in a breakdown in their



notions of normality, thus creating a gap in knowledge which exacerbated uncertainty. Thus, this stimulated information behaviours including voicing information needs and seeking and using information as mothers grappled to understand their lived experience caring for their child on GHT and re-establish a sense of being normal. Information behaviour, which is noted in the findings (section 4.8), covered emotion-focused and problem-focused strategies comprising of seeking support from other parents who had previous experience of caring for a child receiving GHT and sharing information with other parents in similar situations.

Woodgate & Degner (2002) determined uncertainty as an element in supporting hope. In this study, despite the uncertainties held by mothers about GHT, they all decided to commence their children on GHT with the hope that this treatment would help them reach their optimal wellbeing. One mother avoided seeking information and avoided support from support groups as a way of coping with uncertainty. Consequently, these findings encourage the need for professionals to help mothers to deal with the uncertainty rather than trying to eliminate it (Mishel 1990).

Social stigma uncertainty not only adds to the magnitude of uncertainties faced by mothers, but it could also have impeded successful coping of mothers with uncertainty. This uncertainty hindered some mothers from seeking support from family and friends. Keeping the condition or GHT a secret and avoiding social situations that enhance the stigma were strategies used by some mothers (section 4.8.2). Many attempts to protect their children's psychosocial wellbeing were reported (section 4.5.1 and 4.5.4). This is in line with Naiki *et al.* (2013) who had shown that short stature often attributed to overprotection by families and aversive social experience related to the child's short stature. In the current study, the mothers tried to protect their children from hearing the perceived negative comments about the child's size. These negative comments were not only from people in public but also from healthcare providers as they communicated at follow up appointments. It is, therefore, crucial for healthcare providers to be aware and exercise more sensitive approaches while interacting with these children and their mothers.

With the felt responsibility for sheltering their children from harm and preserving their children's sense of normality (section 4.5.1), some mothers avoided situations that

would risk their children being exposed to an adverse reaction from people, and being stigmatised further (Haug Fjone *et al.* 2009) such as holding the child back in school at a lower level so that they do not stand out as much between younger peers. Alternatively, avoiding all-boy schools (in the case of the child being a boy) as boys are often taller than girls. They also avoided their children being exposed to hearing comments that could aggravate their sense of being different at clinical follow-ups. This was done by taking along another family member to the appointment and placing the child in their care outside of the room as healthcare provider communicated to the mother. These findings resonate with the conclusions of Benson *et al.* (2017) who looked at the challenges with parents disclosing their child's diagnosis of epilepsy with others.

Before receiving a final diagnosis, mothers expressed facing challenges trying to communicate with their children concerning their medical condition and size. Mothers regularly tried to reassure their children by normalising their size and downplaying the concern in order to cope (section 4.5.1, 4.5.4 and 4.7.1) which are noted strategies of coping (Mishel 1988). Silva *et al.* (2018) noted that height-related quality of life, weaknesses and more affecting complications in children and adolescents with short stature adds to caregiving stress, which in turn has an adverse effect on parents' quality of life, independently of the diagnosis, treatment status, and current height deviation. In the current study, mothers were not only concerned about their children's psychological wellbeing but also their physical wellbeing (section 4.5.1). They expressed great apprehension about their children's physical safety which could lead mothers to act over protectively in line with Naiki *et al.* (2013). Due to their size, these children may face more environmental barriers and therefore rely more on their parents (Bullinger *et al.* 2013). This overprotection from parents and over-reliance of the children on them could lead to exacerbation of the stigma attached to the child's height and in turn, infantilise the child which could negatively affect their psychosocial wellbeing (Lambert & Keogh 2015).

Mothers felt threatened by others reaction to their children's difference 'being small', 'having a rare condition' and 'receiving GHT' (section 4.5.1 and 4.8.2). It was noted by Kerr *et al.* (2014) that parents' perception of stigma might generate and heighten their sense of uncertainty leading them to seek as much information about treatment, or

impulsively decide on treatment that may challenge medical advice which suggests waiting. Most mothers illustrated trying to minimise the difference and maintain the child's confidence which explains their perception of 'social expectation' when it comes to being small. It is already known that the child's health and psychological wellbeing, and as an adult, had considerable influence on parents' decision to start GHT (Grimberg *et al.* 2015).

Nelson *et al.*'s (2012) study examined how parents' experience and manage decision-making for children's 'normalising' surgeries in the context of elective treatments which aim to 'normalise' a child's function, appearance, communication or identity. Findings disclosed a core category, 'doing the right thing,' that captured parents' key worries and concerns about their children's treatment and emphasised several emotional, social and cultural factors reinforcing their decision-making viewpoints. Parents justified a felt 'moral' obligation to be 'good' parents by pursuing the 'normalising' treatments, mainly surgeries, made accessible to their children. Such treatments were considered as a way of assisting their child's social inclusion and helping them attain their full potential. The findings of the current study concur with this as mothers put themselves in the shoes of their children and made a choice with their best interest in mind (section 4.5.3). This study adds to the extent of the dilemma faced by parents when deciding to start GHT. They feared resentment from their children in the future and always empathised with their children. However, it was also noted in the findings of this study that parents were also challenged with deterrents to commencing GHT which stretched the burden of making the decision. They feared GHT would backlash and negatively affect their children's psychological wellbeing as it was a daily reminder of the 'difference' they had which could accentuate the stigma attached. They questioned if by accepting GHT, they would have failed to accept their child's difference which also exacerbates the attached stigma (section 4.5.2).

Sex bias in referrals for poor growth evaluation was also reported in the current study (section 4.5.3). Based on the literature, social pressures emphasise more on growth in boys rather than girls (Grimberg *et al.* 2004). Studies have associated height to male student competence according to the perception of elementary school teachers' (Villimez *et al.* 1986) male success in initiating intimate relationships (Hensley 1994), work success (Hensley 1987, Hensley & Cooper 1987) and academic success (Hensley

1993). Growth hormone registries signify favouring treatment of boys with poor growth; boys receive GHT by ratios of about 2:1, subject to the diagnosis, with the highest for idiopathic short stature (August *et al.* 1990, Chatelain 1999). Anecdotally it has been stated that the more likely explanation is that boys are more commonly referred to growth clinics because it is socially more acceptable for girls to be short than boys (Chatelain 1999, Grimberg *et al.* 2015).

This is similar with the results of the current study as many mothers declared gender bias as being a contributor to the decision to start GHT. The notion of social pressure and expectation for males to be taller was stated. One mother who had a set of twins (a boy and a girl who were both small) felt that the healthcare provider gave more attention to her son who was followed up and treated with GHT, while, no equal attention was given to her daughter, though she was slightly bigger than her brother. This illustration could explain the increased number of boys being put on GHT because they contract more medical attention about their growth. For instance, young girls with Turner syndrome (TS) are often neglected and brought to the attention of medical help only for evaluation of delayed puberty rather than for short stature (Danda *et al.* 2017). Gender bias, long waiting periods for testing, the absence of standardised guidelines on starting GHT in Ireland, and delays in detecting and referring growth failure in children to specialist services were reasons for the delay in commencing GHT, as reported in the current study. Although this study managed to find reasons for the delay of the commencement of GHT from the mothers' perspective, it would be beneficial to explore patient and healthcare professionals' perspectives on factors contributing to the delay of the commencement of GHT in Ireland. This would be essential in resolving the delays that have been lived by the mothers.

Finally, uncertainty experienced by patients has been recognised as unavoidable in nursing practice (Vaismoradi *et al.* 2011). Therefore, nurses require skills and knowledge to manage uncertainty effectively. As mitigation of uncertainty is not always possible additional responses to the management are required (Macnamara 2014). By exploring the main sources of uncertainty healthcare professionals can try to resolve or reduce some of the uncertainties experienced. Nurses should also be aware of the individual's evaluation of uncertainty as resilience and tolerance of uncertainty have been found to be predictors of emotion-focused coping (Macnamara 2014). Therefore,

when mothers are struggling to cope, nurses can help to increase coping skills by recognising ways mothers have formerly coped effectively, for example, utilising approaches such as taking one day at a time and building small time-based milestones to aim for. The next section will discuss 'normalisation' which was one of the three overarching concepts, beginning with an overview and ending with a critical discussion of the findings according to Deatrick et al.'s (1999) five attributes of normalisation.

#### **5.4 Normalisation as a Significant Adaptation Technique**

The literature signifies that families must actively rebuild a healthy family life after the uncertainty of the pre-diagnostic phase has been resolved and the shock of the diagnosis has lessened (Deatrick *et al.* 1999). The means that mothers cope and adjust to their child's diagnosis is fundamental for the family's physical and emotional wellbeing and to the child's individual coping and adjustment to the condition (Hentinen & Kyngäs 1998). One of the main clinical standards used to judge the success of family management with chronic conditions is normalisation (Deatrick *et al.* 1999). Families who have significant members with chronic conditions are advised by healthcare professionals to establish their lives and interactions as close to 'normal' as possible (Deatrick *et al.* 1999).

In the healthcare literature, normalisation has considered how individuals and families cope with a wide-range of chronic conditions. Families conceptualise through a cognitive process which they define their lives as normal despite the demands of their child's long-term condition and through strategies to manage their child's care needs (Deatrick *et al.* 1999). Knafl & Deatrick (1986) laid the foundation for many of the work on normalisation in nursing with a classic concept analysis. Their conceptual analysis of normalisation recognised four key elements revealed by individuals using this process of normalisation. The elements recognised by Knafl & Deatrick (1986) involved: (1) acknowledging that the impairment is present; (2) defining life as basically normal; (3) minimizing the social consequences of the illness; and (4) engaging in behaviour that demonstrates normalcy to others. In other words, an individual or family who normalises acknowledges that there is a deviation from health but attempts to live as normally as possible in spite of these restrictions. To the extent

that parents want to express the normalcy of their child, they may seek out experiences and situations to highlight that normalcy. Parents may manipulate and control the environment and put their children in normal situations. Later, Deatrick *et al.* (1999) reviewed the four key elements of Knafl & Deatrick's (1986) conceptual analysis of normalisation to incorporate a fifth element that was the incorporation of treatment regimens consistent with normalcy, which may or may not agree with the suggested treatment regimen. For the sake of elaborating the findings of this study involving a medical treatment regimen, Deatrick *et al.*'s (1999) five updated attributes of normalisation will be used which are: (1) acknowledging the condition and its potential threat to lifestyle; (2) adapting normalcy lens for defining the child and family; engaging in parenting behaviours and family routine that are consistent with a normalcy lens; (3) engaging in parenting behaviours and family routines that are consistent with normalisation; (4) developing a treatment regimen that is consistent with normalcy; (5) interacting with others based on the view of the child and family as normal.

The first attribute reflects the disruption of normality and the need to know 'making sense'. It also echoes the significance of the diagnostic period, drawing attention to the family's transitions to accepting the chronic condition and its consequences for family life (Deatrick *et al.* 1999). The literature has suggested that diagnostic uncertainty triggers a situation of constant uncertainty that spreads across multiple aspects of family life (Cohen 1993). This in turn effects how families actively reconstruct a normal family life after the uncertainty of the pre-diagnostic phase has subsided. It has been revealed how mothers endured the turmoil and confusion of prolonged periods of reaching a definitive diagnosis with the uncertainties attached to it (section 4.5.1 and 4.6.2). Mothers have described how the presence of first symptoms and the subsequent journey for seeking a diagnosis which disturbed the constructions of everyday life marking the end of their earlier known normality (section 4.5.1 and 4.6.1). The reaching of a diagnosis was seen as a turning point that allowed mothers to start adjusting to their new normality. With prolonged delays in the diagnostic process, this could interfere with mothers' adjustment. The sooner the mothers know the cause of the deviation from normal, the sooner they can start adjusting to the new normal. Social stigma uncertainty also may have played a great role in how mothers acknowledged the condition and defined normal, and how they made sense of the first symptoms, subsequent journey of seeking a diagnosis and the process of initiating GHT as mentioned previously. In this

regard, starting GHT is seen as a normalisation treatment to bring their children to their best potential and closest to what is socially accepted (section 4.5.3).

The second attribute explains how mothers consciously looked at the treatment as being part of their lives which influenced how they adjusted to the GHT regimen gradually until it became a bedtime routine that was compared to ‘brushing their teeth’ (section 4.7.1). It was seen as a treatment that had to be incorporated into their lives with no option other than adjusting to it (section 4.7.2). Many of the mothers also acknowledged their children as being normal but ‘small’. Many used downward comparisons to look at their child’s condition as being ‘more normal’ than others with more profound health issues (section 4.5.4 and 4.5.5).

The third attribute elucidates the strategies used by mothers to create or sustain normal family life. Many mothers tried to influence a normal life for the child by protecting the child from social exclusion. Others managed to manipulate the GHT to not affect the child’s social life. Many handled the disclosure of the condition and treatment either by facing or avoiding social stigma in order to maintain normality for the child (section 4.5.1, 4.5.4 and 4.8.2).

The fourth attribute discusses developing a treatment regimen that is consistent with normalcy and highlights how the treatment regimen is modified to minimise disturbance to the child’s and family’s life (Deatrick *et al.* 1999). This attribute was identified in Chapter 4 (section 4.7). The notion of trial and error to reach normality in treatment (Deatrick *et al.* 1999) is also detected in the current study as mothers tried to find ways to minimise the pain and discomfort of daily injections on the children. They also tried adjusting time of injection with the child’s emotional and physical needs accordingly, to minimise disruption. Missing doses when the child had sleepovers were seen as a way to maintain a sense of normality which might help explain why parents may have missed injections despite knowing the importance of adhering to GHT (Kremidas *et al.* 2013). It has been reported that GHT had some impact and interfered with family travel and social lives for children with GHD and their parent (Kremidas *et al.* 2013, Brod *et al.* 2017) which is in harmony with the findings of this study. However, this study shows how mothers adjusted to these GHT management burdens. It is noteworthy that many mothers independently sought ways to fit comfortably to the new normal for both

child and mother without professional support.

The fifth attribute involves interacting with others in order to decrease the social consequences of the child's condition. Mothers were aware that their child's differences were readily apparent to others and often felt conspicuous in public because of their child's size. They felt sensitive to the reactions of others, and vigilant. They were most apprehensive about their child being treated as normal as possible and feared discrimination or inequality of treatment by others. This sometimes led mothers to restrict sharing information about their child's condition and treatment as people would not understand, as discussed previously in social stigma uncertainty (section 5.3.1.4). This avoidance to social stigma could result in isolation and impact mothers' coping and adaptation which in turn hinders them reaching normalisation.

In Germeni *et al.*'s (2018) study, they sought to clarify how the diagnosis of a rare disease, as compared to a common, chronic condition, may impact maternal experiences of childhood illness. They found in their qualitative study with 26 mothers in Italy that the diagnosis was perceived as being the first step towards treatment, a way of improving the child's wellbeing, and a turning point that could enable mothers to start adjusting to their new normal, a similar finding in the current study. In addition, the process of normality reconstruction includes a redefinition of normality as integrating the condition and treatment, also reordering family life, based on the needs of the sick child and attaining a sense of control over the disease (Gower *et al.* 2017, Germeni *et al.* 2018).

In Germeni *et al.* (2018) study, the mothers of children who had a rare condition were found to have more difficulty coping and reconstructing a new normality after receiving the diagnosis due to the rarity of the condition. Markedly the access to sufficient information, social support, and contact with other parents who have children with the same rare condition, were found to be significant stressors. The literature recognises information-seeking and sharing as central means of making sense of illness and re-establishing perceived normality (Genuis & Bronstein 2017, Germeni *et al.* 2018). With lack of sufficient information, social support, and contact with other parents were notable barriers expressed by mothers, they voiced their need for more relevant information and support from other parents who are living the similar phenomena



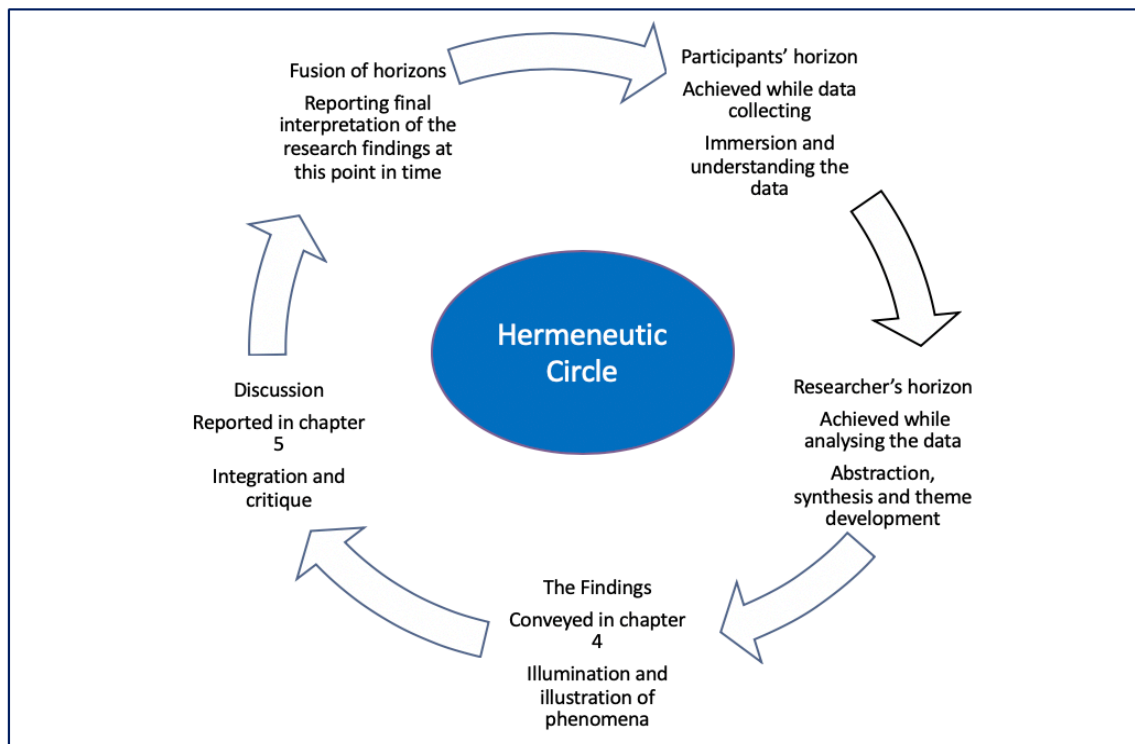
(section 4.8).

Seeking and sharing information from social support networks are common needs amongst parents who care for children with long term conditions as noted in Smith *et al.*'s (2015) review of the literature. This indicates that mothers not only need sufficient information and support from healthcare providers to help them make sense of the disruption of normality (during the diagnostic stage, decision-making to start GHT, management of GHT and future) but require continuous support and information provision to bridge gaps and assist them in re-establishing their new normal and adapting to it (section 4.8). They also need connections with other families who may have similar experiential knowledge or social support groups through charity organisations like those used to recruit some mothers, of which would be very beneficial (section 4.8). Information about the convenience of support groups and specialist networks occurred by chance rather than being afforded as an essential part of care delivery was likewise noted by (Smith *et al.* 2015). It should not be by chance that they discover them but established through the recommendations of their most trusted figure, their health provider. Healthcare providers can support parents by assisting them in using the internet in achieving such goals as noted by Laing (2014) who advised that the use of online information, mobile phone apps, leaflets and support groups can educate and support patients and their carer's at home, through the use of the internet and better help the outcomes of GHT. The next section will discuss the final step of the interpretation process and the reaching of the fusion of horizon according to Gadamer hermeneutic phenomenology.

### **5.5 The Temporary Interpretation is Achieved “Fusion of Horizons”**

Considering the findings and in keeping with the principles of hermeneutic phenomenology the theoretical work linking to the concepts of ‘uncertainty’, ‘stigma’ and ‘normalisation’ led to a wide volume of literature. The discussion chapter addressed the final phase (integration and critique) of the fourth step (gaining understanding through dialogue with text) in (Fleming *et al.* 2003, Ajjawi & Higgs 2007) analytical framework (see Chapter 3). This step involved a critique of the findings and the report of the final interpretation of the research findings by the researcher (see Figure 7).

Figure 7. Completing the Hermeneutic Circle (the Final Interpretation)

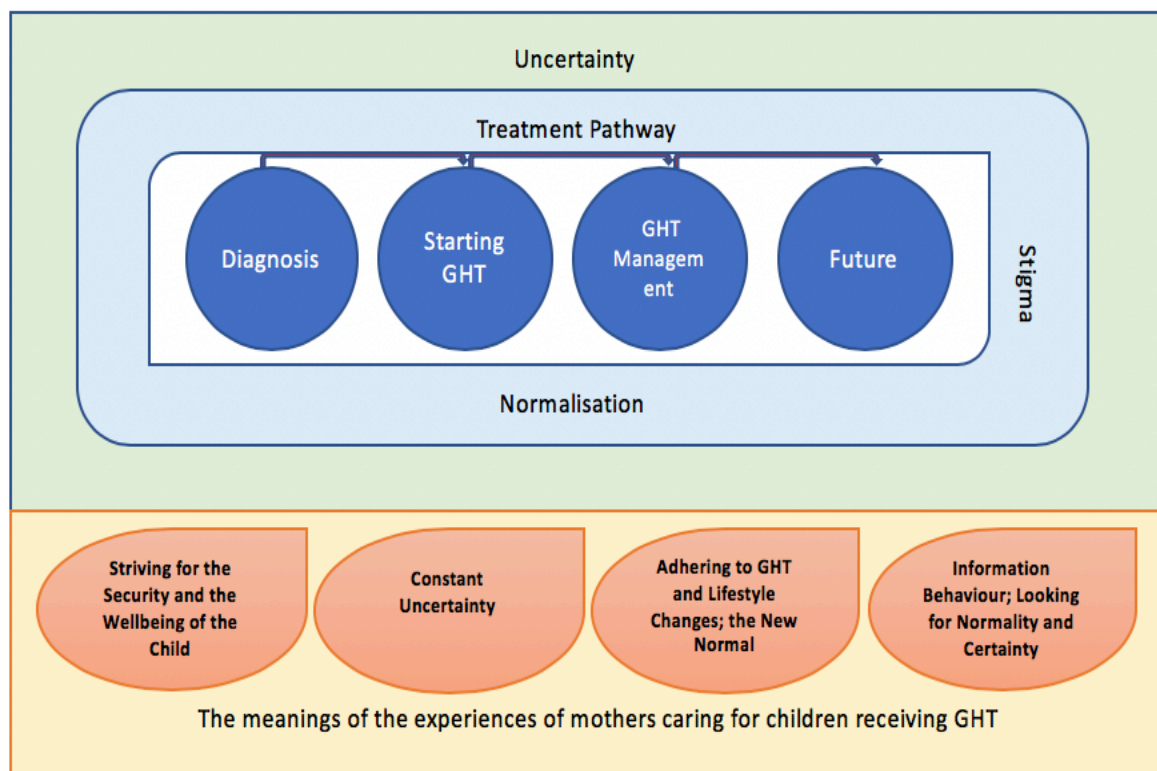


From this review, a theoretical framework supporting the concept of ‘uncertainty’, ‘stigma’ and ‘normalisation’ with both Mishel’s models of uncertainty in Illness (Mishel 1988), reconceptualised model of uncertainty in chronic illness (Mishel 1990) and Deatrick *et al.*’s (1999) five attributes of normalisation were considered as being of particular relevance. Following indepth hermeneutic analysis of the study findings the concepts were used as analytical lenses to examine the findings concerning the mothers’ experiences and therefore it became clear that the data were consistent with many aspects existing in both the theory related to uncertainty and attributes of normalisation. It also became evident that ‘social stigma uncertainty’ adds an extra dimension of uncertainty to the mothers’ experience that prevails all aspects of their experience.

The fusion of horizons and the hermeneutic circle play a vital role in hermeneutic philosophy, and Gadamer (1975) considers interpretation as a fusion of horizons, a dialectical interaction between the pre-understandings of the interpreter and the meaning of the text (Polkinghorne 1983). This process was helpful in the broadening of one’s horizon, as the pre-understandings held prior to starting the study have evolved. The analysis of the data resulted in a detailed description of the experiences of mothers

on a trajectory of illness and treatment pathway (reaching a diagnosis, starting GHT, managing GHT and the future) as illustrated in Figure 8. Further analysis led to a more advanced conceptual level illustrated by the concepts of ‘stigma’, ‘uncertainty’ and ‘normalisation’.

Figure 8. Initial Themes Evolving to Final Themes as they were Challenged by the Literature



Linking the concepts was considered appropriate as the over-arching concepts epitomised the mothers’ journeys. Consequently, combining the concepts together with the findings of this study after a series of steps shown in Figures (9-10) overleaf led to a final conceptual model as outlined in Figure 11. This model brings together the concepts and reflects the dimensions of diagnostic, treatment, future, and social stigma uncertainty, arising as a result of their child’s illness and need for GHT, together with the concept of seeking normality and reaching adaptation and a new reconstructed normal.

Figure 9. Uncertainty May be Ongoing or Evolving Overtime - Adapted from Mishel (1990)

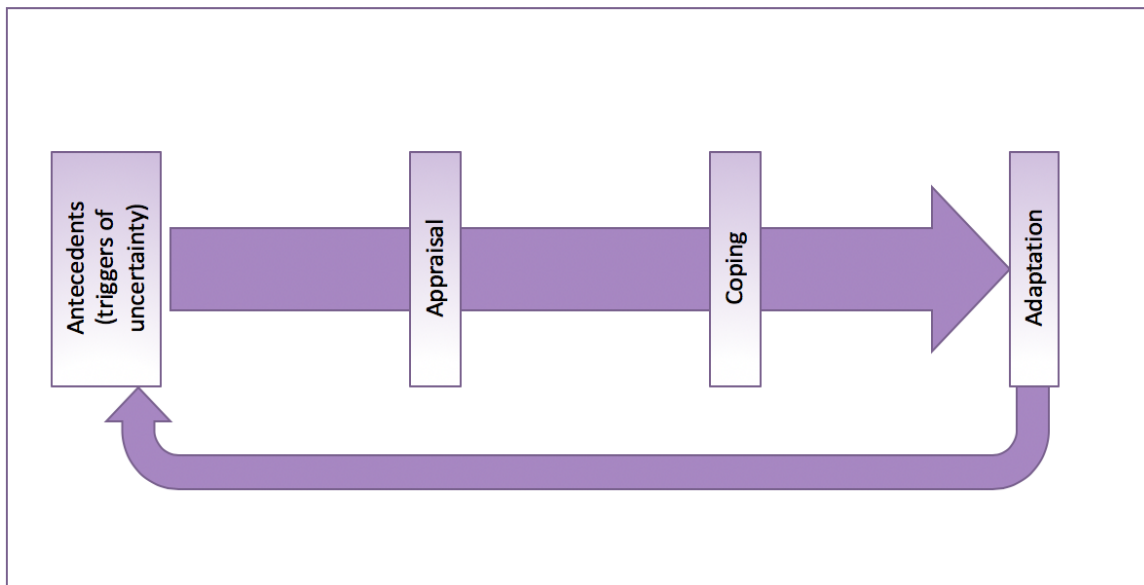
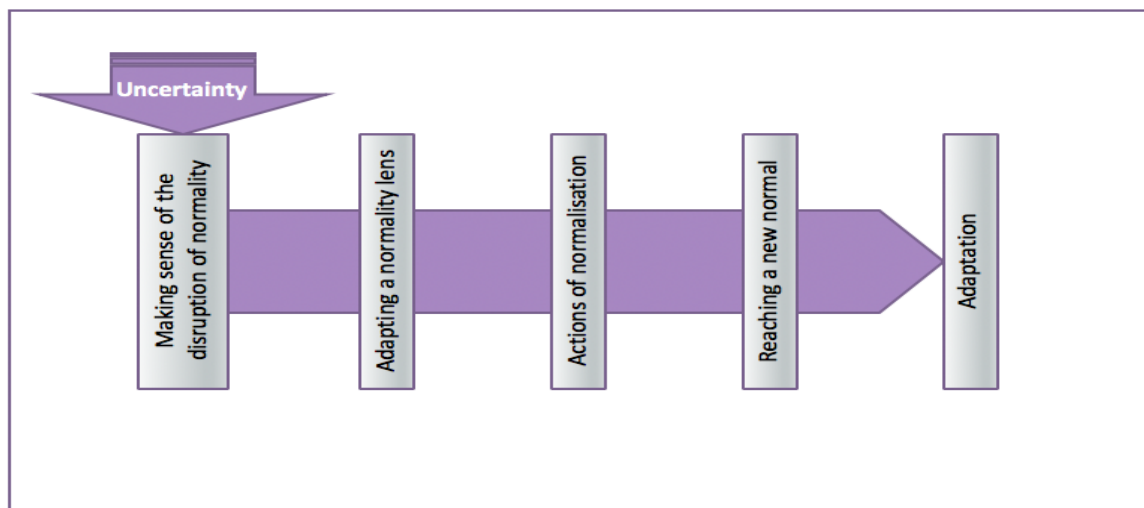


Figure 10. The Relationship Between Uncertainty and Reaching Normality Adapted from Deatrick *et al.* (1999)

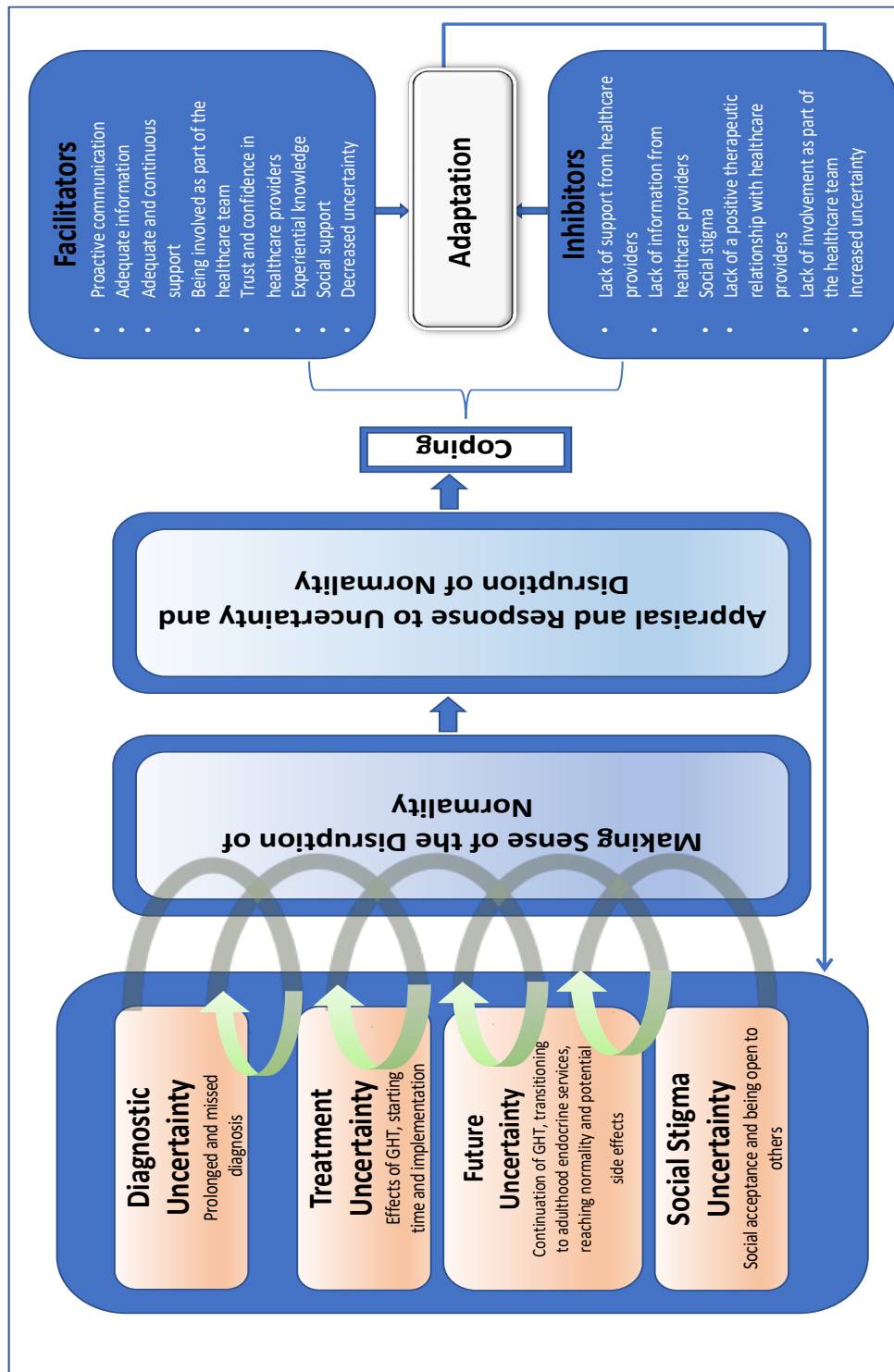


Normalisation may be a significant mediator of illness-related stressors such as treatment burdens and uncertainty on family outcomes (Knafl *et al.* 2010) and a drive for seeking information for those making sense of medical uncertainty (Genius 2012). Godbold's (2013) indepth analysis of sense-making revealed that people with compromised health bridged information gaps by familiarising themselves to shared

themes and thus to ideas of what is normal amongst a group of peers. New gaps in understanding source renewed search for normality and so this concept is not static, neither is uncertainty (Genuis & Bronstein 2017). Hinton & Kirk (2017) argued that parents come to rely on their uncertainty about childhood multiple sclerosis as a way to maintain hope; uncertainty allows for possible recovery and normality. Therefore, parents appeared to be held in a delicate balancing act; swapping the negative images of MS, against the uncertain, but more positive ideal of recovery and health. Similarly, Marini *et al.*'s (2016) study explored the experiences of care for growth hormone deficiency (GHD) for children and their parents in Italy and found that parents had continuous doubt about possible side effects of GHT and lived in a constant state of balancing between the desire to offer help for their child to reach normal height and get a 'normal child' and their doubts about the treatment. This reflects the state of living with constant uncertainty that can be adjusted to and can be the favoured state when other options are reflected as having negative outcomes (Mishel 1990).

Although it can be debated that the three concepts are of particular relevance, considered in isolation neither sufficiently addressed or captured the variety of experiences described by the mothers. Significantly, by joining the concepts, this newly developed framework helps capture the 'experiences' of mothers caring for children commenced on GHT. Therefore, this newly developed framework, seen in Figure 11 overleaf, adds an original input to knowledge which is required when completing a doctoral study. It is also expected that this new framework will form the basis for healthcare professionals to gain a deeper understanding and contemplate and reflect upon the maternal and wider family needs when caring for children commenced on GHT. Specifically, it aims to facilitate recognition and interpretation of mothers' needs with respect to concepts of 'uncertainty', 'stigma' and 'normalisation' and identifies some of the facilitators and inhibitors that can have an impact on their coping and adaptation by anticipating some of these uncertainties and recognising the nature of making sense of the disruption of normality and seeking normality, professionals can help to minimise some of the stress and anxiety experienced by these mothers that can flare at any time during the treatment journey.

Figure 11. Final Framework: Fusion of Horizons



Furthermore, it provides a base for nurses and healthcare professionals to consider some of the nursing therapeutics that can be used to support mothers through this complex and prolonged journey. It will not always be possible to ease uncertainty linked to

lacking information, for example prognostic information or risks information, but working with mothers to alleviate other areas of uncertainty may prove important for long term adaptation. This new framework can guide professionals to the key areas for concern and will provide a model for reflection on the complexity of the mothers' journeys. Some of these key areas and clinical implications arising from the study will now be considered leading to the following section where recommendations are stated.

## **5.6 Recommendations**

The findings reflect mothers' experiences, although many of the recommendations could equally apply to fathers. Thus, in some recommendations the term 'parents' is used. However, a similar study to explore fathers' experiences of caring for children receiving GHT would corroborate this.

### **5.6.1 Clinical Practice**

It seemed that, mothers' knowingness and expert knowledge of their child and their needs appears not to be commonly accepted or recognised by service providers, consequently:

- Healthcare providers should recognise parents as integral members of the healthcare team by involving them in all decisions and information-sharing about their child's treatment.
- Proactive communication with parents can help reduce uncertainties and enhance their sense of satisfaction. In addition, professionals need to acknowledge mothers' contribution and responsibility for delivering GHT for their children.

It was evident that mothers' use of services was influenced by others' competence and unknowingness of their situation and a lack of trust in providers. Service providers appeared to lack a consistent and coordinated approach to service delivery, therefore:

- The importance of building a trusting relationship between parents and caregivers needs to be recognised.

Mothers' appeared to endure prolonged diagnostic periods. These delays were mostly due to long waiting periods for appointments and tests and missed diagnosis of the condition, hence:

- Professionals need to ensure correct measurement of growth in children and detecting growth failure especially general practitioners (GP) as they are the gatekeepers for referrals to paediatric endocrinology services.
- Policy makers need to ensure standardisation of measuring techniques, growth monitoring and referral guidelines in Ireland.
- The healthcare system and healthcare providers should aim to reduce unnecessary delays in reaching the diagnosis and acknowledge the stressfulness impact for parents.
- Genetic counsellors can also help parents who have children diagnosed with a rare genetic condition such as Prader-Willi syndrome (PWS) and Turner syndrome (TS) in easing some of their uncertainty by highlighting sources of social support available.
- Appropriate psychological support to manage emotional challenges associated with adapting to the child's diagnosis can be helpful.

When a child is finally diagnosed and referred to the paediatric endocrinology services it is important to:

- Recognise the stress of adopting the 'wait and see' approach before confirming a diagnosis or commencing the child on GHT for parents; supporting them with resources and information accordingly is critical.
- Healthcare providers can significantly help at this stage by giving enough support during the decision-making process to commence GHT (encourage shared decision-making in GHT).
- Mothers lacked information, and so, information and awareness of the demands placed on parents when deciding to commence their child on GHT and caring at home is required.
- Healthcare providers can equip parents with information such as clear guidelines, audio-visual material, and online resources verses written material.



- Recognising that written information may not suit all parents and giving them time to communicate face-to-face about initial uncertainties and worries they may have.
- professionals should provide the contact details of support organisations and connect parents with other families who have children receiving GHT.
- Healthcare providers should acknowledge the importance of experiential knowledge in helping mothers make an informed decision to commence their children on GHT.
- Healthcare providers should explain to parents the rationale for starting GHT and the suitability of its timing. This also suggests that policy makers should establish standardised guidelines for GH prescribing specifically for Ireland.
- Providing appropriate psychological support to manage emotional challenges associated with the commencement of GHT for both child and parents could be beneficial.
- Mothers lacked device choice in some cases. Therefore ensure choices are offered when choosing a GHT device that suits their child and parents' needs.

In relation to management of GHT, mothers appear to lack support in all aspects of their role in caring for their child receiving GHT and thus:

- Offering continuous support provided by GHT drug companies, not only at the beginning of the treatment but spread along the treatment journey is important.
- Contact initiated by the drug company nurse to the family should be continued along the treatment journey.
- Explanation of the rationale for changes issued to GHT management such as (storage guidelines) and support with implementing these changes should be offered.
- Endocrinology nurses or GHT drug company nurses should provide options on how to manage challenges associated with GHT administration at home and outside the home and then follow up with their adaptation.
- Endocrinology nurses or GHT drug company nurses need to pay particular attention to GHT administration challenges when one parent is the only person who is able to administer the injection and provide support accordingly such as encouraging other members of the family and encouraging the child to learn self-

administration. Offering techniques for parents to manage GHT administration, if they need to be away from the child, at their regular GHT administration time, such as in the case of going out late and needing a babysitter, should also be considered.

- Education and advice in the management of GHT must be realistic and built on care provided by parents at home.
- Connecting families with other support services such as psychologists and play therapists to support them in coping with emotional issues and GHT management issues is required.
- The paediatric endocrinology team should promote concordance to enhance adherence to GHT and recognise the importance of GHT being consistent with maintaining normalcy in the family life.
- Recognising the difficulties in organisational aspects of managing GHT and the need for a better coordinated approach between the endocrinology team, local pharmacy and GHT drug company to minimise the level of the burden associated with maintaining GHT supplies and reducing the multiple points of contact for parents.
- At paediatric endocrinology follow-up appointments, the team needs to give enough time to the parent and children to answer questions.
- It is necessary to be aware of the sensitivity of the stigma associated with growth failure and communicate with the child and family at the clinic accordingly. Using words like ‘tiny’, ‘small’ and ‘what’s wrong with the child?’ should be avoided. They ought to acknowledge the issues mothers face due to stigma and provide support and advice to help with emotional challenges for both child and parent.
- Making parents aware of the availability of charity support organisations and social media pages that accommodate other parents with similar situations as a source of experiential knowledge to assist them in creating a social constructed normality is key. This also brings attention to the need for additional charity support organisations and social media pages that are specific to Ireland to fit the Irish context.
- Signifying the child and parent of digital health apps that may help empower them with adherence information and education tools.

For future matters concerning GHT:

- Parents need to be informed of the possibility of the continuation or discontinuation of GHT and transitioning to adult health care services if needed as early as possible.
- Paediatric endocrinology teams need to facilitate smooth transitioning of both the child and parent into adult healthcare services.

In general:

- Healthcare providers should focus not only on providing practical support in managing GHT challenges but on the psychological support in managing emotional and psychosocial challenges faced by the family throughout their journey on GHT.
- Uncertainties can heighten at any point during the GHT regimen and healthcare providers need to support them at these points. If some of the uncertainties cannot be eliminated, then healthcare providers should help them in managing these uncertainties to facilitate adaptation. Managing uncertainty could be in the form of teaching parents the skills in problem solving and cognitive reframing; treatment related side effects, patient provider communication, resource management, education and information.

### **5.6.2 Nursing Education**

It is critical to ensure that standard and up-to-date child growth monitoring guidelines and measuring techniques are included in the content of children's nursing education programmes. Also, reinforce the importance of family-centred care and include theoretical instruction on the burden of care-giving for parents caring for a child with special needs. Raising nurses' awareness of parents needs in combination with lectures on , chronic conditions, adaptation, trust, supporting parents caregiving role, decision-making, advocacy, and teaching skills are essential.

### **5.6.3 Further Research**

The accuracy of growth measuring techniques used by the general practitioners (GP) signposts an unnecessary delay in referral to specialised services and receiving the appropriate medical treatment. So, further research is needed in this area. In addition, the notion that GHT is a taboo topic and is not easily disclosed to others suggests further research in GHT abuse and its psychosocial impact on children receiving GHT and their caregivers. The lack of public understanding and knowledge around growth disorders and GHT is an area worthy of further research to explore the public understanding about GHT and how it impacts on the adaptation of patients and their families to GHT. Although this study proposes that mothers were involved to some degree in the decisions to start their children on GHT, whether all felt the consultations and decision-making were at a suitable level and made with adequate support and information is unknown and so, this is another potential area for future research. Further research is required in approaches to tackle organisational issues such as (multiple points of contact for GHT supplies and needle disposing) to minimise associated GHT burden. Moreover, a longitudinal study exploring the impact of GHT on family function and emotional well-being would further the work of this study. Also, measuring the level of uncertainty associated with GHT and its management and its impact on family function and emotional well-being would further the work of this study. A similar study to explore fathers and family member's experiences of caring for children receiving GHT would add more depth to the mothers' experiences.

## **5.7 Limitations**

### **5.7.1 The Methodological Approach**

The original text by Gadamer is written in German which meant using translated sources of Gadamer as sources of information and others' interpretations of it. Geneallos (1998) states that hermeneutic interpretations of data are usually an approximation and are never final. The interpretation of the findings is grounded by hermeneutic phenomenological principles where many realities and interpretations are likely. The findings can consequently vary depending on the interpretation of the reader and their opinion of hermeneutic phenomenology. Moreover, the transcripts were not

returned to the mothers for validation as the theoretical view implemented that member-checking is unsuited in phenomenology (Webb 2003). Consequently, the findings are my interpretations of the mothers' experiences. Findings were verified by professional colleagues and experts caring for children with complex needs and they were found to represent mothers' experiences.

### **5.7.2 The Sample**

The intention was to recruit parents of children receiving GHT. However only mothers agreed to participate. Therefore, the findings are limited by the lack of fathers' perspective on caregiving for a child on GHT. Since mothers are usually the primary caregiver this may explain why they chose to participate.

The children represented a range of medical conditions with different indications for GHT. Growth hormone treatment was prescribed for impaired growth velocity caused by either insufficient GH production such as in growth hormone deficiency (GHD) or by an impaired response to physiological GH levels such as Turner's syndrome (TS), Russell Silvers syndrome (RSS) and Prader-Willi syndrome (PWS). The nature of the complex needs associated with PWS and the extra care needs accompanying their care made them slightly distinctive in their experiences. This is both a limitation and an advantage as the children had a range of disorders, which offers a perspective on experiences of GHT care needs, rather than focusing on the impact of a particular disease.

The mothers' experiences with GHT ranged from six months to eight years with the majority being less than three years. Mothers of children receiving GHT over many years may have reported different experiences, in that they may have had time to adapt and cope with it thereby needing less support, assistance and reassurance. This may be an assumption since five of the mothers had long-term experiences with GHT (up to eight years) and their accounts were similar to other mothers in the study.

The experiences of mothers who participated in the study may differ from those who did not agree to participate. Also, their experiences may not represent all mothers of

similar groups of children receiving GHT. Most of the mothers were professionals which may have made them more critical of the health services. As they encountered challenges, they may have been more assertive in seeking information to handle and take control of the situation. This may not apply to mothers with a lower level of literacy and reduced socioeconomic status who may face further difficulty negotiating healthcare encounters and seeking information. The study sample was limited to the Republic of Ireland (ROI), so the findings are not generalisable to mothers who live in other countries where healthcare provision and services may vary. However, the findings were mostly supported by data from studies conducted across the world.

### **5.7.3 Data Collection Methods**

In phenomenological methods, the accuracy of the reports given by the participants at the point of data collection is crucial (Polkinghorne 1988, Sutton & Austin 2015). This could be critiqued in that, interviewing produces retrospective recounts which is not the same as collecting the data when the event is happening. Retrospective reporting of experiences may be prejudiced by recall bias as mothers described their experiences over time. However, what they conveyed was seen by the mothers as part of their experiences and was as they remembered it. Polkinghorne (1989) proposes that any time deferment may assist a more comprehensive description as the chance to reflect on the experience has been offered. The use of multiple interviews may have allowed mothers to have several opportunities to give information and to add comments to the data over time. However, as previously discussed in the methodology chapter and due to the challenging recruitment phase, one-time interviews were chosen in addition to the diary recordings. The legitimacy of the choice to reduce the number of interviews and diaries was supported by the participating mothers as they confirmed having no more to say or add at the end of the first interview. However, with adding the diary, mothers had the opportunity to share untold information at the interview and share new events between interview and diary collection (two months). Diaries provided an extra opportunity for reflection and reconsideration, yet, they served mostly to confirm what was already said at the interview and no added information was gained.

The interview and diary data may not entirely echo and mirror the totality of the

mothers' experiences but only those that they chose to disclose, and the meaning attached to the experiences of these mothers. The interview process, however, did deliver a chance for an indepth exploration of the mothers' experiences which quantitative methods would not have captured. The diaries add to the realities of the data collection through interviews as they served to confirm what was interpreted from the interviews.

#### **5.7.4 Being a Parent and a Single Researcher**

Additionally, as previously highlighted in Chapter 3, the mothers knew I was a mother of a child receiving GHT which significantly helped gain their trust, but at times lead some mothers to ask about my experience with GHT. However, measures were put in places to minimise my experiences influencing theirs. Also, my personal experience of caring for a child receiving GHT may have influenced the interpretation of the data although I have attempted to remain transparent about my fore-knowledge and understanding (Gadamer 1975). However as Heidegger (1962) acknowledged, the impact of the researcher on the research process is fundamental to the philosophical underpinnings of hermeneutic phenomenology and therefore it can be reasoned that I was in keeping with the methodological approach selected.

The analysis was influenced by my perceptive and analytical abilities as I was the key instrument of data collection. For data analysis, I used Nvivo 11 and written documents to allow others to follow my decision trail. Data passages are embraced throughout in sufficient length for the reader to understand the context and my understanding of the findings. Bias is hard to control, but it's possible influence is a limitation. However, I have been open about my background and the factors that may have swayed my interpretation. I have embraced a systematic and self-conscious attitude in my description of the research processes used and how the findings arose. This has been conveyed in a way that might allow another researcher to analyse the data and reach similar conclusions. Using a framework suggests that the analysis is a stage that 'finishes the process', although analysis is continuing in this work even to the end point as writing and rewriting continues.

## 5.8 Conclusion

Due to increasing numbers of children needing GHT from a very young age in past decades, more parents are undertaking the responsibility for GHT administration at home (Van Dongen & Kaptein 2012, Kaptein 2013). Consequently, parents often must undertake complex care with regard to GHT administration, storage, medication adherence, and costs. With much of the previous research focusing on GHT pharmaceutical effects, administration and medication adherence, the aim of this study was to explore the experiences of parents of children commenced on GHT. This hermeneutic phenomenological study delivers an indepth understanding of the experiences of a small group of mothers which can aid to increase the sensitivity of healthcare professionals to the needs of mothers and guide future care and support for families.

By applying the philosophical approach built on principles of hermeneutic phenomenology this thesis has offered indepth findings and interpretation of the mothers' experiences of caring for children receiving GHT. Hermeneutic phenomenology was chosen as the research methodology as it fits the aim of exploring the 'lived experience' of the individual from the perspective of those who experience it first-hand (Matua & Van Der Wal 2015). By implementing the approach based on interpretive phenomenology guided by the work of Gadamer, the study aimed to discover the meaning and essence associated with the mothers' experiences of caring for their children receiving GHT. Gadamer (1975) encourages the researcher's pre-conceptions and fore-knowledge as valid components of the research and bringing together this fore-structure with the research findings, constructing a shared understanding of the phenomena in what he called a 'fusion of horizons'. Therefore, based on my current knowledge and experience of caring for a child on GHT it was deliberated that conducting the research grounded on the philosophical underpinnings of hermeneutic phenomenology stayed close to my aim to explore the 'life-world' of the mothers involved in caring for their child on GHT. However, as Gadamer also determined hermeneutics is an evolving process and reaching a conclusive interpretation is impossible, consequently the interpretation reached in this thesis is only one understanding constructed in appreciation that more are possible.



Using an analytical framework; a combination of Fleming *et al.* (2003) and Ajjawi & Higgs (2007) based on Gadamer's philosophical underpinnings for data analysis; the experiences of the mothers were explored. Throughout their child's illness and GHT pathway the concepts of 'uncertainty', 'stigma' and 'normalisation' that are central to the four major meanings of their lived experiences; (1) "It's the right thing to do" Striving for the security and the wellbeing of the child, (2) "Doubting yourself constantly" Constant uncertainty, (3) "But then you just get used to it I suppose" Adhering to GHT and lifestyle changes; the new normal, (4) "I hadn't been told anything about it" Information behaviour; looking for normality and certainty, reflected the mother's experiences. Gadamer (1975) comprehended hermeneutics to be a means of co-creation in which the researcher expands their understanding and gains new knowledge through a circle of reading, reflexivity and interpretation. By doing so, my goal was to expose additional meaning and gain a deeper understanding of the mothers' experiences. Accordingly, the findings were reviewed in relation to Mishel's uncertainty in illness model and reconceptualisation of uncertainty in chronic illness (1988, 1990) and Deatrick *et al.*'s (1999) five attributes of normalisation.

A model that incorporated all the findings relating to the three concepts was developed. This model may help raise healthcare professionals' awareness of the uncertainty, stigma and normalisation faced by mothers and help in the development of strategies to support parents and families.

A final quote from Michelle:

"She (the nurse) was lovely and she was very relaxed and very friendly. That was a huge thing for us because we were really nervous about it and she really put us at ease, and I think that first person that you come in contact with I think would make a huge difference to your experience with it (GHT). I just remember feeling very comforted and safe and she just seemed like she knew what she was doing, but at the same time she was professional, but friendly with it."

## References

Acerini C., Albanese A., Casey A., Denvir L., Jones J., Mathew V., Musson P. & Sparrow S. (2012) Initiating growth hormone therapy for children and adolescents. *British Journal of Nursing* **21**(18), 1091-1097.

Adler P. & Adler P. (1987) *Membership Roles in Field Research*. Newbury Park, CA: Sage.

Agwu J., Leishenring A. & Darnley I. (2004) Community growth monitoring in practice. *Archives of Disease in Childhood* **89**(4), 394-394.

Ahmed S.F., Blamires C. & Smith W. (2007) Facilitating and understanding the family's choice of injection device for growth hormone therapy by using conjoint analysis. *Archives of Disease in Childhood* **39**(2), 110.

Ajjawi R. & Higgs J. (2007) Using hermeneutic phenomenology to investigate how experienced practitioners learn to communicate clinical reasoning. *The Qualitative Report* **12**(4), 612-638.

Alaszewski A. (2006) *Using Diaries for Social Research*. Thousand Oak, London; New Delhi; Sage.

Andrews M. (2000) A parent's view of short stature. *Pediatric Annals* **29**(9), 582-584.

Andsager J.L. & Powers A. (2001) Framing women's health with a sense-making approach: Magazine coverage of breast cancer and implants. *Health Communication* **13**(2), 163-185.

Angen M.J. (2000) Evaluating interpretive inquiry: Reviewing the validity debate and opening the dialogue. *Qualitative Health Research* **10**(3), 378-395.

Annells M. (1996) Hermeneutic phenomenology: Philosophical perspectives and current use in nursing research. *Journal of Advanced Nursing* **23**(4), 705-713.

Antoniazzi F., Cavarzere P. & Gaudino R. (2015) Growth hormone and early treatment. *Minerva Endocrinologica* **40**(2), 129-143.

Audi L., Fernández-Cancio M., Camats N. & Carrascosa A. (2013) Growth hormone deficiency: an update. *Minerva Endocrinologica* **38**(1), 1-16.

August G.P., Lippe B.M., Blethen S.L., Rosenfeld R.G., Seelig S.A., Johanson A.J., Compton P.G., Frane J.W., McClellan B.H. & Sherman B.M. (1990) Growth hormone treatment in the United States: demographic and diagnostic features of 2331 children. *The Journal of Pediatrics* **116**(6), 899-903.

Austgard K. (2012) Doing it the Gadamerian way—using philosophical hermeneutics as a methodological approach in nursing science. *Scandinavian Journal of Caring Sciences* **26**(4), 829-834.

Barstow C. & Rerucha C. (2015) Evaluation of short and tall stature in children. *American Academy of Family and Physicians* **92**(1), 44-50.

Baxter L., Bryant J., Cave C.B. & Milne R. (2007) Recombinant growth hormone for children and adolescents with Turner syndrome. *Cochrane Database Systematic Reviews* (1), Cd003887.

Benson A., Lambert V., Gallagher P., Shahwan A. & Austin J.K. (2017) Parent perspectives of the challenging aspects of disclosing a child's epilepsy diagnosis to others: Why don't they tell? *Chronic Illness* **13**(1), 28-48.

Bernard R.S. & Cohen L.L. (2006) Parent anxiety and infant pain during pediatric immunizations. *Journal of Clinical Psychology in Medical Settings* **13**(3), 282-287.

Bhosle M., Klingman D., Aagren M., Wisniewski T. & Lee W.C. (2011) Human growth hormone treatment: synthesis of literature on product delivery systems and administration practices. *Journal for Specialists in Pediatric Nursing* **16**(1), 50-63.

Bolton G. (2018) *Reflective Practice: Writing and Professional Development*. SAGE publications Ltd, London, Thousand Oaks.

Bowlby J. (2005) *A Secure Base: Clinical Applications of Attachment Theory*. 393. Taylor & Francis, London and Newyork.

Bozzola M., Colle M., Halldin-Stenlid M., Larroque S. & Zignani M. (2011) Treatment adherence with the easypod™ growth hormone electronic auto-injector and patient acceptance: survey results from 824 children and their parents. *BMC Endocrine Disorders* **11**(1), 4.

Brod M., Højbjerg L., Alolga S.L., Beck J.F., Wilkinson L. & Rasmussen M.H. (2017) Understanding treatment burden for children treated for growth hormone deficiency. *The Patient-Patient-Centered Outcomes Research* **10**(5), 653-666.

BSPED (2015) *Shared Care Guidelines: Paediatric use of Recombinant human Growth Hormone (r-hGH, Somatropin)*. The British Society for Paediatric Endocrinology and Diabetes BSPED. Retrieved from [https://www.bsped.org.uk/media/1377/sharedcaregh\\_july2015.pdf](https://www.bsped.org.uk/media/1377/sharedcaregh_july2015.pdf) on 17th Februray 2018.

Bullinger M., Quitmann J., Power M., Herdman M., Mimoun E., DeBusk K., Feigerlova E., Lunde C., Dellenmark-Blom M. & Sanz D. (2013) Assessing the quality of life of health-referred children and adolescents with short stature: development and psychometric testing of the QoLISSY instrument. *Health and Quality of Life Outcomes* **11**(1), 76.

Caelli K. (2001) Engaging with phenomenology: Is it more of a challenge than it needs to be? *Qualitative Health Research* **11**(2), 273-281.

Cappa M., Iughetti L., Loche S., Maghnie M. & Vottero A. (2016) Efficacy and safety of growth hormone treatment in children with short stature: the Italian cohort of the GeNeSIS clinical study. *Journal of Endocrinological Investigation* **39**(6), 667-677.

Chaplin J.E., Kriström B., Jonsson B., Hägglöf B., Tuvemo T., Aronson A.S., Dahlgren J. & Albertsson-Wikland K. (2011) Improvements in behaviour and self-esteem following growth hormone treatment in short prepubertal children. *Hormone*

*Research in Paediatrics* **75**(4), 291-303.

Chatelain P. (1999) Trends in the diagnosis and treatment of short stature as revealed by KIGS. *Growth Hormone Therapy in KIGS* **10**, 11-20.

Chen C., Visootsak J., Dills S. & Graham Jr J.M. (2007) Prader-Willi syndrome: an update and review for the primary pediatrician. *Clinical Pediatrics* **46**(7), 580-591.

Chesler MA., Parry C. (2001) Gender roles and/or styles in crisis: an integrative analysis of the experiences of fathers of children with cancer. *Qualitative Health Research*, **11**(3):363.

Clayton M.F., Dean M. & Mishel M. (2018) Theories of Uncertainty in Illness. in *Middle Range Theory for Nursing*, 4<sup>th</sup> edn. (Smith M.J.& Liehr P.R.,eds), Spring Publication Compant, New York, pp. 49-82.

Cohen M.H. (1993) Diagnostic closure and the spread of uncertainty. *Issues in Comprehensive Pediatric Nursing* **16**(3), 135-146.

Cohen M.H. (1995) The triggers of heightened parental uncertainty in chronic, life-threatening childhood illness. *Qualitative Health Research* **5**(1), 63-77.

Cohen M.H. & Martinson I.M. (1988) Chronic uncertainty: its effect on parental appraisal of a child's health. *Journal of Pediatric Nursing* **3**(2), 89-96.

Cohen M.Z., Kahn D.L. & Steeves R.H. (2000) *Hermeneutic Phenomenological Research: A Practical Guide for Nurse Researchers*. SAGE Publications Ltd, Thousand Oaks, London & New Delhi.

Colaizzi P.F. (1978) Psychological research as the phenomenologist views it. In *Existential-Phenomenological Alternatives for Psychology*. (Valle R.S.& King M., eds), Oxford University Press, pp. 6.

Collin J. Whitehead, A & Walker J. (2016) Educating children and families about growth hormone deficiency and its management: part 2. *Nursing Children and Young People* **28**(2), 30.

Conrad P. & Potter D. (2004) Human growth hormone and the temptations of biomedical enhancement. *Sociology of Health & Illness* **26**(2), 184-215.

Converse M. (2012) Philosophy of phenomenology: how understanding aids research. *Nurse Researcher* **20**(1), 28-32.

Cook D., Yuen K., Biller B., Kemp S. & Vance M. (2009) American Association of Clinical Endocrinologists medical guidelines for clinical practice for growth hormone use in growth hormone-deficient adults and transition patients-2009 update. *Endocrine Practice* **15**(2), 1-29.

Corbin Dwyer S. & Buckle J. (2009) The space between: On being an insider—outsider in qualitative research. *International Journal of Qualitative Methods* **8**(8), 54-63.

Corti L. (1993) Using diaries in social research. *Social Research Update* **2**(2).

Cousounis P., Lipman T.H., Ginsburg K.R. & Grimberg A. (2013) Internet informs parents about growth hormone. *Hormone Research in Paediatrics* **80**(2), 86-91.

Cresswell J.W. (2003) *Research Design: Quantitative, Qualitative and Mixed Methods Approaches*. SAGE Publications Ltd, Inc, Thousand Oaks, California.

Crotty M. (2000) *Phenomenology and Nursing Research*. Elsevier Health Sciences, London.

Cunliffe A.L. (2009) Reflexivity, Learning and Reflexive Practice. In *The SAGE Handbook of Management Learning, Education and Development*, (Armstrong S.J. & Fukami C.V., eds). SAGE Publications Ltd, London, Thousand Oaks, New Delhi, Singapore, pp. 405-418.

Cutfield W.S., Derraik J.G., Gunn A.J., Reid K., Delany T., Robinson E. & Hofman P.L. (2011) Non-compliance with growth hormone treatment in children is common and impairs linear growth. *PloS One* **6**(1), E16223.

Danda V.S.R., Sreedevi P., Arun G. & Rao P.S. (2017) Growth hormone treatment in Turner's syndrome: A real world experience. *Indian Journal of Endocrinology and Metabolism* **21**(3), 378.

Daniel E., Kent G., Binney V. & Pagdin J. (2005) Trying to do my best as a mother: Decision-making in families of children undergoing elective surgical treatment for short stature. *British Journal of Health Psychology* **10**(1), 101-114.

Darendeliler F., Karagiannis G. & Wilton P. (2007) Headache, idiopathic intracranial hypertension and slipped capital femoral epiphysis during growth hormone treatment: a safety update from the KIGS database. *Hormone Research in Paediatrics* **68**(5), 41-47.

Deacon B. & Abramowitz J. (2006) Fear of needles and vasovagal reactions among phlebotomy patients. *Journal of Anxiety Disorders* **20**(7), 946-960.

Deal C.L., Tony M., Hoybye C., Allen D.B., Tauber M. & Christiansen J.S. (2013) GrowthHormone Research Society workshop summary: consensus guidelines for recombinant human growth hormone therapy in Prader-Willi syndrome. *Journal Clinical Endocrinology and Metabolism* **98**(6), E1072-1087.

Deatrick J.A., Knafl K.A. & Murphy-Moore C. (1999) Clarifying the concept of normalization. *Journal of Nursing Scholarship* **31**(3), 209-214.

Denscombe M. (2014) *The Good Research Guide: for Small-Scale Social Research Projects*, 4<sup>th</sup> edn. McGraw-Hill Education (UK), Berkshire.

Denzin N.K. & Lincoln Y.S. (1998) *The Landscape of Qualitative Research: Theory and Issues*. SAGE Publications Ltd, London.

Dervin B. & Frenette M. (2003) Sense-making methodology: communicating communicatively with campaign audiences. in *Sense-Making Methodology Reader: Selected Writings of Brenda Dervin*, (Dervin B. & Foreman-Wernet L.,eds). Hampton Press, NJ, Cresskill, pp.397.

De Sales T. (2003) Horizons revealed: From methodology to method. *International*

*Journal of Qualitative Methods* **2**(1), 1-17.

Desrosiers P., O'Brien F. & Blethen S. (2005) Patient outcomes in the GHMonitor: the effect of delivery device on compliance and growth. *Pediatric Endocrinology Reviews: PER* **2**, 327-331.

Diekelmann N.L. (1992) Learning-as-testing: A Heideggerian hermeneutical analysis of the lived experiences of students and teachers in nursing. *Advances in Nursing Science* **14**(3), 72-83.

DiVall S.A. & Radovick S. (2013) Growth hormone and treatment controversy; Long-term safety of rGH. *Current Pediatrics Reports* **1**(2), 128-132.

Dodgson J.E., Garwick A., Blozis S.A., Patterson J.M., Bennett F.C. & Blum R.W. (2000) Uncertainty in childhood chronic conditions and family distress in families of young children. *Journal of Family Nursing* **6**(3), 252-266.

Dowling M. (2007) From Husserl to van Manen. A review of different phenomenological approaches. *International Journal of Nursing Studies* **44**(1), 131-142.

Dowling M. & Cooney A. (2012) Research approaches related to phenomenology: negotiating a complex landscape. *Nurse Researcher* **20**(2), 21-27.

Drake A.J. & Kelnar C.J. (2006) The evaluation of growth and the identification of growth hormone deficiency. *Archives of Disease in Childhood-Education and Practice* **91**(3), 61- 67.

Dumas H., Panayiotopoulos P., Parker D. & Pongpaichana V. (2006) Understanding and meeting the needs of those using growth hormone injection devices. *BMC Endocrine Disorders* **6**, 5.

Dykens E.M., Roof E. & Hunt-Hawkins H. (2017) Cognitive and adaptive advantages of growth hormone treatment in children with Prader-willi syndrome. *Journal of Child Psychology and Psychiatry* **58**(1), 64-74.



- Eiser C., Vance Y., Glaser A., Galvin H., Horne B., Picton S., Stoner A. & Butler G. (2005) Growth hormone treatment and quality of life among survivors of childhood cancer. *Hormone Research in Paediatrics* **63**(6), 300-304.
- Erling A., Wiklund I. & Albertsson-Wikland K. (1994) Prepubertal children with short stature have a different perception of their well-being and stature than their parents. *Quality of Life Research* **3**(6), 425-429.
- Evans N., Wakefield C., McLoone J. & Cohn R. (2015) Familial diagnostic experiences in paediatric oncology. *British Journal of Cancer* **112**(1), 20.
- Faisal S., Blandford A. & Potts H.W. (2013) Making sense of personal health information: challenges for information visualization. *Health Informatics Journal* **19**(3), 198-217.
- Fex A., Flensner, G. Ek, A.C. & Söderhamn O. (2011) Living with an adult family member using advanced medical technology at home. *Nursing Inquiry* **18**(4), 336-347.
- Finlay L. (2009) Debating phenomenological research methods. *Phenomenology & Practice* **3**(1).
- Fisher B.G. & Acerini C.L. (2013) Understanding the growth hormone therapy adherence paradigm: a systematic review. *Hormone Research in Paediatrics* **79**(4), 189-196.
- Fleming V., Gaidys U. & Robb Y. (2003) Hermeneutic research in nursing: developing a Gadamerian-based research method. *Nursing Inquiry* **10**(2), 113-120.
- Foddy B. & Savulescu J. (2017) Ethics of performance enhancement in sport. *Ethics in Sport*, 307.
- Ford K. & Turner D. (2001) Stories seldom told: paediatric nurses' experiences of caring for hospitalized children with special needs and their families. *Journal of Advanced Nursing* **33**(3), 288-295.

Fuchs G.S., Mikkelsen S., Knudsen T.K. & Kappelgaard A.M. (2009) Ease of use and acceptability of a new pen device for the administration of growth hormone therapy in pediatric patients: an open-label, uncontrolled usability test. *Clinical Therapeutics* **31**(12), 2906-2914.

Gadamer H.G. (1975) *Truth and Method*. Sheed & Ward, London.

Gadamer H.G. (1990) The Universality of the Hermeneutical Problem. in *The Hermeneutic Tradition: From Ast to Ricoeur*, (Ormiston G.L. & Schrifft A.D.,eds), State University of New York Press, Albany, pp. 147-158.

Gadamer H.G., Weinsheimer J. & Marshall D.G. (2004) *EPZ Truth and Method*. Bloomsbury Publishing USA, New York.

Garganta M.D. & Bremer A.A. (2014) Clinical dilemmas in evaluating the short child. *Pediatric annals* **43**(8), 321-327.

Geanellos R. (1998) Hermeneutic philosophy. Part I: Implications of its use as methodology in interpretive nursing research. *Nursing inquiry* **5**(3), 154-163.

Geanellos R. (1999) Hermeneutic interviewing: An example of its development and use as research method. *Contemporary Nurse* **8**(2), 39-45.

Geanellos R. (2000) Exploring Ricoeur's hermeneutic theory of interpretation as a method of analysing research texts. *Nursing Inquiry* **7**(2), 112-119.

Geanellos R. (2005) Sustaining well-being and enabling recovery: the therapeutic effect of nurse friendliness on clients and nursing environments. *Contemporary Nurse* **19**(1-2), 242-252.

Genuis S.K. (2012) Constructing "sense" from evolving health information: a qualitative investigation of information seeking and sense making across sources. *Journal of the Association for Information Science and Technology* **63**(8), 1553-1566.

Genuis S.K. & Bronstein J. (2017) Looking for "normal": Sense making in the

context of health disruption. *Journal of the Association for Information Science and Technology* **68**(3), 750-761.

Germei E., Vallini I., Bianchetti M.G. & Schulz P.J. (2018) Reconstructing normality following the diagnosis of a childhood chronic disease: does “rare” make a difference? *European journal of pediatrics*, 1-7.

Gibbs G. (2002) *Qualitative Data Analysis: Explorations with NVivo (Understanding Social Research)*. Open University Press, Buckingham.

Giorgi A. (1989) Some theoretical and practical issues regarding the psychological phenomenological method. *Saybrook Review* **7**(2), 71-85.

Godbold N. (2013) *Patterns in Sense Making Interactions: How People Make Sense of Kidney Failure in Online Renal Discussion Groups*. PhD thesis, University of Sydney, Sydney.

Goffman E. (1963) *Stigma. Notes on the Management of Spoiled Identity*. Simon and Shuster. Inc, New York.

Gower C., Higgins A., Doherty N. & McCormack D. (2017) Understanding the experiences of fathers of children with congenital heart disease: An interpretative phenomenological analysis. *Journal of Health Psychology* **22**(11), 1447-1457.

Grassley J.S. & Nelms T.P. (2008) Understanding maternal breastfeeding confidence: a Gadamerian hermeneutic analysis of women's stories. *Health Care for Women International* **29**(8-9), 841-862.

Grbich C. (1998) *Qualitative Research in Health: An Introduction*. SAGE Publication Ltd, Thousand Oaks.

Green J., Darbyshire P., Adams A. & Jackson D. (2015) Balancing hope with reality: how neonatal nurses manage the uncertainty of caring for extremely premature babies. *Journal of Clinical Nursing* **24**(17-18), 2410-2418.

Grimberg A., Cousounis P., Cucchiara A.J., Lipman T.H. & Ginsburg K.R. (2015)

Parental concerns influencing decisions to seek medical care for a child's short stature. *Hormone Research in Paediatrics* **84**(5), 338-348.

Grootenhuis M.A. & Last B.F. (1997) Predictors of parental emotional adjustment to childhood cancer. *Psycho-Oncology* **6**(2), 115-128.

Growing Up in Ireland (2013) *Development From Birth to Three Years*. Stationery Office / Department of Children and Youth Affairs, Ireland, Dublin.

Guerriere D.N., McKeever P., Llewellyn-Thomas H. & Berall G. (2003) Mothers' decisions about gastrostomy tube insertion in children: factors contributing to uncertainty. *Developmental Medicine and Child Neurology* **45**(7), 470-476.

Hauffa B., Touraine P., Urquhart-Kelly T. & Koledova E. (2017) Managing transition in patients treated with growth hormone. *Frontiers in Endocrinology* **8**, 346.

Haug Fjone H., Ytterhus B. & Almvik A. (2009) How children with parents suffering from mental health distress search for 'normality' and avoid stigma: to be or not to be... is not the question. *Childhood* **16**(4), 461-477.

Haverkamp F., Johansson L., Dumas H., Langham S., Tauber M., Veimo D. & Chiarelli F. (2008) Observations of nonadherence to recombinant human growth hormone therapy in clinical practice. *Clinical Therapeutics* **30**(2), 307-316.

Hawkes C. & O'Connell S. (2016) A national survey on the diagnosis and treatment of paediatric growth hormone deficiency. *Irish Medical Journal* **109**(2), 359.

Heidegger M. (1962) *Being and Time*. (Macquarrie J, Robinson E, trs). Harper & Row, New York, NY.

Hekman S. (1983) From epistemology to ontology: Gadamer's hermeneutics and Wittgensteinian social science. *Human Studies* **6**(3), 205-224.

Hensley W.E. (1993) Height as a measure of success in academe. *Psychology: a Journal of Human behavior* **30**(1), 40-46.

Hensley W.E. (1994) Height as a basis for interpersonal attraction. *Adolescence* **29**(114), 469.

Hensley W.E. & Cooper R. (1987) Height and occupational success: a review and critique. *Psychological Reports* **60**(3), 843-849.

Hentinen M. & Kyngäs H. (1998) Factors associated with the adaptation of parents with a chronically ill child. *Journal of Clinical Nursing* **7**(4), 316-324.

Hermanussen M. & Bogin B. (2014) Auxology—an editorial. *Italian Journal of Pediatrics* **40** (8).

Hey-Hadavi J., Pleil A., Deeb L.C., Fuqua J.S., Silverman L.A., Reiner B., Newfield R., Rajcic N., Wajnrajch M.P. & Cara J.F. (2010) Ease of use and preference for a new disposable self-injection pen compared with a reusable pen for administering recombinant human growth hormone: A multicenter, 2-Month, single-arm, open-label clinical trial in patient-caregiver dyads. *Clinical Therapeutics* **32**(12), 2036-2047.

Hindmarsh P. (1996) Monitoring children's growth. *British Medical Journal*, 312:122.

Hinton D. & Kirk S. (2017) Living with uncertainty and hope: A qualitative study exploring parents' experiences of living with childhood multiple sclerosis. *Chronic Illness* **13**(2), 88-99.

Hintz R.L. (2004) Growth hormone: uses and abuses: It has anabolic effects, but its use in ageing and other conditions is not established. *BMJ: British Medical Journal* **328**(7445), 907.

Hoey H. (2014) *Growth Disorders in Childhood*. The Irish College of General Practitioners. Retrieved from [https://www.icgp.ie/assets/1/F74018BC-19B9-E185-833836772EA4ED0A\\_document/Article\\_1.pdf](https://www.icgp.ie/assets/1/F74018BC-19B9-E185-833836772EA4ED0A_document/Article_1.pdf) on 9th of December 2017.

Houk C.P. & Lee P.A. (2012) Early diagnosis and treatment referral of children born small for gestational age without catch-up growth are critical for optimal growth

outcomes. *International Journal of Pediatric Endocrinology* **2012**(1), 11.

Howe C.J., Ratcliffe S.J., Tuttle A., Dougherty S. & Lipman T.H. (2011) Needle anxiety in children with type 1 diabetes and their mothers. *MCN THE American Journal of Maternal Child Nursing* **36**(1), 25-31.

HSE (2012) *Training Program for Public Health Nurses and Doctors in Child Health Screening, Surveillance and Health Promotion*. Health Service Executive, Ireland, Dublin. Retrieved from <https://www.hse.ie/eng/services/publications/children/introduction-to-training-programme-for-public-health-nurses-and-doctors-in-child-health-screening-surveillance-and-health-promotion.pdf> on 26<sup>th</sup> of August 2018.

HSE (2018) *A National Model of Care for Paediatric Healthcare Services in Ireland. Chapter 23: Paediatric Endocrinology and Diabetes Care*. Health Service Executive, Ireland. Retrieved from <https://www.hse.ie/eng/services/publications/clinical-strategy-and-programmes/paediatric-endocrinology.pdf> on 26<sup>th</sup> of August 2018.

Husserl E. & Lauer Q. (1965) *Phenomenology and the Crisis of Philosophy. Philosophy as Rigorous Science and Philosophy and the Crisis of European Man*. Harper & Row, Publishers, New York.

Jensen R.B., Thankamony A., Day F., Scott R.A., Langenberg C., Kirk J., Donaldson M., Ivarsson S.-A., Söder O. & Roche E. (2015) Genetic markers of insulin sensitivity and insulin secretion are associated with spontaneous postnatal growth and response to growth hormone treatment in short SGA children: the North European SGA Study (NESGAS). *The Journal of Clinical Endocrinology & Metabolism* **100**(3), E503-E507.

Jensen R.B., Thankamony A., O'Connell S.M., Salgin B., Kirk J., Donaldson M., Ivarsson S.-A., Söder O., Roche E. & Hoey H. (2013) Baseline IGF-I levels determine insulin secretion and insulin sensitivity during the first year on growth hormone therapy in children born small for gestational age. Results from a North European Multicentre Study (NESGAS). *Hormone Research in Paediatrics* **80**(1),

38-46.

Jessop D.J. & Stein R.E. (1985) Uncertainty and its relation to the psychological and social correlates of chronic illness in children. *Social Science & Medicine* **20**(1), 993-999.

Johnston L.B. (2008) Individualization of growth hormone therapy. *Best Practice Research in Clinical Endocrinology and Metabolism* **22**(3), 517-524.

Kafle N.P. (2013) Hermeneutic phenomenological research method simplified. *Bodhi: An Interdisciplinary Journal* **5**(1), 181-200.

Kapoor R.R., Burke S., Sparrow S., Hughes I.A., Dunger D.B., Ong K.K. & Acerini C.L. (2008) Monitoring of concordance in growth hormone therapy. *Archives of Disease in Childhood* **93**(2), 147-148.

Kappelgaard A.M. & Laursen T. (2011) The benefits of growth hormone therapy in patients with Turner syndrome, Noonan syndrome and children born small for gestational age. *Growth Hormone IGF Research* **21**(6), 305-313.

Kaptein A.A. (2013) Transjecting growth hormone: continuous nightmare or controlled nuisance? Evaluation of a new needle-free device. *Patient Preference and Adherence* **7**, 703.

Karlsson K., Englund A.C., Enskar K. & Rydstrom I. (2014) Parents' perspectives on supporting children during needle-related medical procedures. *International Journal of Qualitative Studies on Health and Well-being* **9**, 23759.

Kastrup K., Christiansen J.S., Andersen J.K. & Ørskov H. (1983) Increased growth rate following transfer to daily sc administration from three weekly im injections of hGH in growth hormone deficient children. *Acta Endocrinologica* **104**(2), 148-152.

Keats D. (1999) *Interviewing: A Practical Guide for Students and Professionals*. UNSW Press, Sydney.

Kemp S.F. & Frindik J.P. (2011) Emerging options in growth hormone therapy: an update. *Drug Design Development and Therapy* **5**, 411-419.

- Kerr A.M. & Haas S.M. (2014) Parental uncertainty in illness: managing uncertainty surrounding an "orphan" illness. *Journal of Pediatric Nursing: Nursing Care of Children and Families* **29**(5), 393-400.
- Kim J.S., Sinacore J.M. & Pongracic J.A. (2005) Parental use of EpiPen for children with food allergies. *Journal of Allergy and Clinical Immunology* **116**(1), 164-168.
- Kirk J. (2009) Improving adherence to GH therapy with an electronic device: first experience with easyPod. *Pediatric Endocrinology Review* **6**(4), 549-552.
- Kirk J. (2012) Indications for growth hormone therapy in children. *Archives of Disease in Childhood* **97**(1), 63-68.
- Kirk J. & Clayton P. (2006) Specialist services and transitional care in paediatric endocrinology in the UK and Ireland. *Clinical Endocrinology* **65**(1), 59-63.
- Kirk S., Glendinning C. & Callery P. (2005) Parent or nurse? The experience of being the parent of a technology-dependent child. *Journal of Advanced Nursing* **51**(5), 456-464.
- Kleiman S. (2004) Phenomenology: to wonder and search for meanings. *Nurse researcher* **11**(4), 7-19.
- Knafl K.A., Darney B.G., Gallo A.M. & Angst D.B. (2010) Parental perceptions of the outcome and meaning of normalization. *Research in Nursing & Health* **33**(2), 87-98.
- Knafl K.A. & Deatrick J.A. (1986) How families manage chronic conditions: An analysis of the concept of normalization. *Research in Nursing & Health* **9**(3), 215-222.
- Koch G.A. (1993) *Towards Fourth Generation Evaluation: Listening to the Voices of Older Patients: a Hermeneutic Inquiry*. PhD thesis, University of Manchester, Manchester.
- Koch T. (1995) Interpretive approaches in nursing research: The influence of Husserl



and Heidegger. *Journal of Advanced Nursing* **21**(5), 827-836.

Kremidas D., Wisniewski T., Divino V.M., Bala K., Olsen M., Germak J., Aagren M., Holot N. & Lee W.C. (2013) Administration burden associated with recombinant human growth hormone treatment: perspectives of patients and caregivers. *Journal of Pediatric Nursing* **28**(1), 55-63.

Kvale S. (1983) The qualitative research interview: A phenomenological and a hermeneutical mode of understanding. *Journal of Phenomenological Psychology* **14**(2), 171.

Kvale S. (1997) *Interviews. An Introduction to Qualitative Research Interviewing*. SAGE Publications Ltd, Thousand Oaks.

Lagrou K., Froidecoeur C., Thomas M., Massa G., Beckers D., Craen M., De Beaufort C., Rooman R., Francois I. & Heinrichs C. (2008) Concerns, expectations and perception regarding stature, physical appearance and psychosocial functioning before and during high-dose growth hormone treatment of short pre-pubertal children born small for gestational age. *Hormone Research in Paediatrics* **69**(6), 334-342.

Lagrou K., Vanderfaellie J., Froidecoeur C., Thomas M., Massa G., Tenoutasse S., Craen M., Lebrethon M.C., Beckers D. & Francois I. (2007) Effect of 2 years of high-dose growth hormone therapy on cognitive and psychosocial development in short children born small for gestational age. *European Journal of Endocrinology* **156**(2), 195-201.

Laing P. (2014) Growth failure and hormone therapy. *British Journal of Nursing* **23**(6).

Lambert V. & Keogh D. (2015) Striving to live a normal life: A review of children and young people's experience of feeling different when living with a long term condition. *Journal of Pediatric Nursing: Nursing Care of Children and Families* **30**(1), 63-77.

Lamnek S. (1995) *Qualitative Sozialforschung*. Psychologie Verlags Union

Weinheim. Retrived from  
[http://www.ciando.com/img/books/extract/3621283625\\_lp.pdf](http://www.ciando.com/img/books/extract/3621283625_lp.pdf) 26<sup>th</sup> of August 2018.

Langham S. & Kirk J. (2011) National audit of patient choice in pediatric GH therapy. *Hormone Research Paediatric* **75**(2), 101-105.

Laverty S.M. (2008) Hermeneutic phenomenology and phenomenology: A comparison of historical and methodological considerations. *International Journal of Qualitative Methods* **2**(3), 21-35.

Lee R.L. & Lau V.W. (2013) An interpretive phenomenological study of chinese mothers' experiences of constant vigilance in caring for a hospitalized sick child. *Journal of Advanced Nursing* **69**(8), 1808-1818.

Lenhard W., Breitenbach E., Ebert H., Schindelbauer-Deutscher H.-J. & Henn W. (2005) Psychological benefit of diagnostic certainty for mothers of children with disabilities: lessons from Down syndrome. *American Journal of Medical Genetics Part A* **133**(2), 170-175.

Lester S. (1999) *An Introduction to Phenomenological Research*. Stan Lester Developments, Taunton.

Lincoln Y.S. & Guba E.G. (1985) Establishing trustworthiness. *Naturalistic inquiry* **289**, 331.

Lipinski S.E., Lipinski M.J., Biesecker L.G. & Biesecker B.B. (2006) Uncertainty and perceived personal control among parents of children with rare chromosome conditions: the role of genetic counseling. *American Journal of Medical Genetics* **142**, 232-240.

Léger J. (2017) How should we investigate children with growth failure? *Annales D'endocrinologie* **78**, 106-107.

Macnamara E. (2014) *The Role of Uncertainty in Coping Efficacy: The Experience of Parents of Children with Undiagnosed Medical Conditions*. Master Thesis, John Hopkins University, Baltimore, Maryland.

- Madeo A.C., O'brien K.E., Bernhardt B.A. & Biesecker B.B. (2012) Factors associated with perceived uncertainty among parents of children with undiagnosed medical conditions. *American Journal of Medical Genetics Part A* **158**(8), 1877-1884.
- Mak Y. & Elwyn G. (2003) Use of hermeneutic research in understanding the meaning of desire for euthanasia. *Palliative Medicine* **17**(5), 395-402.
- Marini M.G., Chesi P., Mazzanti L., Guazzarotti L., Toni T.D., Salerno M.C., Officioso A., Parpagnoli M., Angeletti C., Faienza M.F., Iezzi M.L., Aversa T. & Sacchetti C. (2016) Stories of experiences of care for growth hormone deficiency: the CRESCERE project. *Future Science OA* **2**(1), FSO82.
- Marks I. (1988) Blood-injury phobia: a review. *The American Journal of Psychiatry* **145**(10), 1207.
- Mason J. (2002) *Qualitative Researching*, 2<sup>nd</sup> edn. SAGE Publications Ltd, London.
- Matua G.A. & Van Der Wal D.M. (2015) Differentiating between descriptive and interpretive phenomenological research approaches. *Nurse Researcher* **22**(6), 22.
- McCloud C., Harrington A. & King L. (2012) Understanding people's experience of vitreo-retinal day surgery: a Gadamerian-guided study. *Journal of Advanced Nursing* **68**(1),94-103.
- McConnell-Henry T., DNE R. & Karen Francis R. (2011) Member checking and Heideggerian phenomenology: A redundant component. *Nurse Researcher* **18**(2), 28.
- McNeill T. (2004) Fathers' experience of parenting a child with juvenile rheumatoid arthritis. *Qualitative Health Research*, **14**(4):526–545.
- Meakins L., Ray L., Hegadoren K., Rogers L.G. & Rempel G.R. (2015) Parental vigilance in caring for their children with hypoplastic left heart syndrome. *Pediatric Nursing* **41**(1), 31.
- Meinhardt U., Eiholzer U., Seitz L., Bøgelund M. & Kappelgaard A.M. (2014)

Parent preference in Switzerland for easy-to-use attributes of growth hormone injection devices quantified by willingness to pay. *Expert Review of Medical Devices* **11**(1), 31-38.

Mihelicova M., Siegel Z., Evans M., Brown A. & Jason L. (2016) Caring for people with severe myalgic encephalomyelitis: An interpretative phenomenological analysis of parents' experiences. *Journal of Health Psychology* **21**(12), 2824-2837.

Mishel M.H. (1984) Perceived uncertainty and stress in illness. *Research in Nursing & Health* **7**(3), 163-171.

Mishel M.H. (1988) Uncertainty in illness. *Journal of Nursing Scholarship* **20**(4), 225-232.

Mishel M.H. (1990) Reconceptualization of the uncertainty in illness theory. *Journal of Nursing Scholarship* **22**(4), 256-262.

Mishel M.H. & Braden C.J. (1988) Finding meaning: Antecedents of uncertainty in illness. *Nursing Research* **37**(2), 98-103, 127.

Mishel M.H. (1997) Uncertainty in acute illness. *Annual Review of Nursing Research* **15**(1), 57-80.

Mitchell D.B. & Hauser-Cram P. (2008) The well-being of mothers of adolescents with developmental disabilities in relation to medical care utilization and satisfaction with health care. *Research in Developmental Disabilities* **29**(2), 97-112.

Moore W.V., Kaplan S. & Raiti S. (1987) Comparison of dose frequency of human growth hormone in treatment of organic and idiopathic hypopituitarism. *The Journal of Pediatrics* **110**(1), 144-148.

Moran D. (2000) *Introduction to Phenomenology*. Routledge, London.

Morse J.M. (1994) *Critical Issues in Qualitative Research Methods*. SAGE Publications Ltd, London.

Moustakas C. (1994) *Phenomenological Research Methods*. SAGE Publications Ltd, London.

Munhall P.L. (1994) *Revisioning Phenomenology: Nursing and Health Science Research*. National League of Nursing, New York.

Murray P., Dattani M. & Clayton P. (2016) Controversies in the diagnosis and management of growth hormone deficiency in childhood and adolescence. *Archives of Disease in Childhood* **101**(1), 96-100.

Nadeem M. & Roche E. (2014) Height in Turner syndrome: does growth hormone therapy have impact? *Irish Medical Journal (IMJ)* **107**(2), 60-1.

Naiki Y., Horikawa R. & Tanaka T. (2013) Assessment of psychosocial status among short-stature children with and without growth hormone therapy and their parents. *Clinical Pediatric Endocrinology* **22**(2), 25-32.

NICE (2010) *Human growth hormone (somatropin) for the treatment of growth failure in children/ Guidance*. The National Institute of Health and Care Excellence, UK, London.

Nicholl H. (2010) Diaries as a method of data collection in research: Honor Nicholl used diary recordings for a doctoral study on mothers' observations of caring for children with complex needs. *Paediatric Care* **22**(7), 16-20.

Nicholl H.M. (2008) *An Exploration of Mothers' Experiences in Caring for Children with Complex Needs*. PhD thesis, Trinity College, Dublin.

Nicholl H.M. & Begley C.M. (2012) Explicating caregiving by mothers of children with complex needs in Ireland: a phenomenological study. *Journal of Pediatric Nursing* **27**(6), 642-651.

Nicholls A.R., Cope E., Bailey R., Koenen K., Dumon D., Theodorou N.C., Chanal B., Saint Laurent D., Müller D. & Andrés M.P. (2017) Children's first experience of taking anabolic-androgenic steroids can occur before their 10th birthday: a

systematic review identifying 9 factors that predicted doping among young people. *Frontiers in Psychology* **8**, 1015.

Norgren S. (2009) Adherence remains a challenge for patients receiving growth hormone therapy. *Pediatric Endocrinology Reviews: PER* **6**, 545-548.

Nygaard C., & Clancy A. (2018) Using heroes, flying blind-A metasynthesis of parents' experiences of caring for children with special health-care needs at home. *Journal of Clinical Nursing* **27**(15-16), 3179-3196.

Oeffinger K.C., Mertens A.C., Sklar C.A., Kawashima T., Hudson M.M., Meadows A.T., Friedman D.L., Marina N., Hobbie W. & Kadan-Lottick N.S. (2006) Chronic health conditions in adult survivors of childhood cancer. *New England Journal of Medicine* **355**(15), 1572-1582.

Ong K., Beardsall K. & de Zegher F. (2005) Growth hormone therapy in short children born small for gestational age. *Early Human Development* **81**(12), 973-980.

Otero S.C., Eiser C., Wright N.P. & Butler G. (2013) Implications of parent and child quality of life assessments for decisions about growth hormone treatment in eligible children. *Child: Care, Health & Development* **39**(6), 782-788.

Parahoo K. (2014) *Nursing Research, Principles, process and Issues*, 3<sup>rd</sup> edn. Macmillan International Higher Education, New York.

Parvin M., Roche E., Costigan C. & Hoey H.M. (2004) Treatment outcome in Turner syndrome. *Irish Medical Journal* **97**(1), 12-14.

Pascoe E. (1996) The value to nursing research of Gadamer's hermeneutic philosophy. *Journal of Advanced Nursing* **24**(6), 1309-1314.

Penrod J. (2001) Refinement of the concept of uncertainty. *Journal of Advanced Nursing* **34**(2), 238-245.

Pfützner A., Hartmann K., Winter F., Fuchs G.S., Kappelgaard A.-M. & Rohrer T.R. (2010) Intuitiveness, ease of use, and preference of a prefilled growth hormone

injection pen: a noninterventional, randomized, open-label, crossover, comparative usability study of three delivery devices in growth hormone-treated pediatric patients. *Clinical Therapeutics* **32**(11), 1918-1934.

Polit D., Beck C. & Hungler B. (2001) *Essentials of Nursing Research: Methods, Appraisal and Utilisation*, 6th edn. Lippincott Williams & Wilkins, Philadelphia.

Polkinghorne D. (1983) *Methodology for the Human Sciences: Systems of Inquiry*. Suny Press, New York.

Polkinghorne D.E. (1988) *Narrative Knowing and the Human Sciences*. Suny Press, New York.

Polkinghorne D.E. (1989) Phenomenological Research Methods. In *Existential-Phenomenological Perspectives in Psychology*, (Valle R.S. & Halling S., eds). Springer, New York, pp. 41-60.

Pringle J., Hendry C. & McLafferty E. (2011) Phenomenological approaches: challenges and choices. *Nurse Researcher* **18**(2), 7-18.

Rankin D., Harden J., Noyes K., Waugh N., Barnard K. & Lawton J. (2015) Parents' experiences of managing their child's diabetes using an insulin pump: a qualitative study. *Diabetic Medicine* **32**(5), 627-634.

Raphael J.L., Zhang Y., Liu H. & Giardino A. (2010) Parenting stress in US families: Implications for paediatric healthcare utilization. *Child: Care, Health and Development* **36**(2), 216-224.

Rice V.H. (2012) *Handbook of Stress, Coping, and Health: Implications for Nursing Research, Theory, and Practice*. SAGE Publications Ltd, Thousand Oaks.

Rodgers B.L. (2005) *Developing Nursing Knowledge: Philosophical Traditions and Influences*. Lippincott Williams & Wilkins, Philadelphia.

Rohrer T.R., Horikawa R. & Kappelgaard A.-M. (2017) Growth hormone delivery devices: current features and potential for enhanced treatment adherence. *Expert Opinion on Drug Delivery* **14**(11), 1253-1264.

Rosenfeld R.G. & Bakker B. (2008) Compliance and persistence in pediatric and adult patients receiving growth hormone therapy. *Endocrine Practice* **14**(2), 143-154.

Ross J.L., Sandberg D.E., Rose S.R., Leschek E.W., Baron J., Chipman J.J., Cassorla F.G., Quigley C.A., Crowe B.J. & Roberts K. (2004) Psychological adaptation in children with idiopathic short stature treated with growth hormone or placebo. *The Journal of Clinical Endocrinology & Metabolism* **89**(10), 4873-4878.

Rubin R.R., Peyrot M., Metzinger C.P., Xu Y., Lippe B., McCormack L. & Aaron Davis D. (2011) An observational study to validate the Satisfaction Measure of the Injection of Growth Hormone Therapy (SMIGHTy) questionnaire. *Current Medical Research and Opinion* **27**(10), 2009-2017.

Saari A., Sankilampi U., Hannila M.-L., Saha M.-T., Mäkitie O. & Dunkel L. (2012) Screening of turner syndrome with novel auxological criteria facilitates early diagnosis. *The Journal of Clinical Endocrinology & Metabolism* **97**(11), E2125-E2132.

Salem J.B. (2009) *RSS/SGA A Comprehensive Guide*. Quadco Printing Inc., Chicago.

Sallfors C. & Hallberg L.R.-M. (2003) A Parental Perspective on Living with a Chronically Ill Child: A Qualitative Study. *Families, Systems, & Health* **21**(2), 193.

Salmela M., Aronen E. & Salanterä S. (2011) The experience of hospital-related fears of 4-to 6-year-old children. *Child: Care, Health and Development* **37**(5), 719-726.

Sangasubana N. (2011) How to Conduct Ethnographic Research. *The Qualitative Report*, **16**(2), 567-573. Retrieved from <https://nsuworks.nova.edu/tqr/vol16/iss2/14> 20<sup>th</sup> of September 2018.

Savage M.O. & Bang P. (2012) The variability of responses to growth hormone therapy in children with short stature. *Indian Journal of Endocrinology and Metabolism* **16**(2),178.



Schoppe-Sullivan S. (2019) Dads are more involved in parenting, yes, but moms still put in more work. *The Connection*. Retrived from [www. theconversation.com/dads-are-more-involved-in-parenting-yes-but-moms-still-put-in-more-work-72026](http://www.theconversation.com/dads-are-more-involved-in-parenting-yes-but-moms-still-put-in-more-work-72026) on 18 May 2019.

Schwandt T. (2000) Three Epistemological Stances for Qualitative Inquiry. In *Handbok of qualitative research*, (Denzin N.K. & Lincoln Y.S,eds). SAGE Publications Ltd, Thousand Oaks.

Sheppard L., Eiser C., Davies H., Carney S., Clarke S., Urquhart T., Ryder M., Stoner A., Wright N. & Butler G. (2006) The effects of growth hormone treatment on health-related quality of life in children. *Hormone Research in Paediatrics* **65**(5), 243-249.

Shih F.J. (1998) Triangulation in nursing research: issues of conceptual clarity and purpose. *Journal of Advanced Nursing* **28**(3), 631-641.

Siguero L., Aedo M., Moreno L. & Valverde M. (1995) Treatment with growth hormone. *Hormone Research in Paediatrics* **44**(3), 18-25.

Silva N., Bullinger M., Sommer R., Rohenkohl A., Witt S. & Quitmann J. (2018) Children's psychosocial functioning and parents' quality of life in paediatric short stature: The mediating role of caregiving stress. *Clinical Psychology & Psychotherapy* **25**(1), 107-118.

Silverman D. (2010) *Doing Qualitative Research*. SAGE Publications Ltd, London.

Smith D.W. (2013) *Phenomenology*. *The Stanford Encyclopedia of Philosophy*, Winter 2013 edn. Retrived from <https://plato.stanford.edu/entries/phenomenology/> 26<sup>th</sup> of August 2018.

Smith J., Cheater F. & Bekker H. (2015) Parents' experiences of living with a child with a long-term condition: a rapid structured review of the literature. *Health Expectations* **18**(4), 452-474.

Smith M.J. & Liehr P.R. (2014) *Middle Range Theory for Nursing*, 3rd edn. Springer Publishing Company, New York.

Smith P.J. & Werther G.A. (2005) Children with growth disorders. *Australian Family Physician* **34**(9).

Smith S.L., Hindmarsh P.C. & Brook C. (1993) Compliance with growth hormone treatment--are they getting it? *Archives of Disease in Childhood* **68**(1), 91-93.

Sommer R., Bullinger M., Chaplin J., Do J.k., Power M., Pleil A. & Quitmann J. (2017) Experiencing health-related quality of life in paediatric short stature—a cross-cultural analysis of statements from patients and parents. *Clinical Psychology & Psychotherapy* **24**(6), 1370-1376.

Speziale H. & Carpenter D. (2007) *Qualitative Research in Nursing: Advancing the Humanistic Imperative*, 4th edn. Lippincott Williams & Wilkins, Philadelphia.

Spoudeas H.A., Bajaj P. & Sommerford N. (2014) Maintaining persistence and adherence with subcutaneous growth-hormone therapy in children: comparing jet-delivery and needle-based devices. *Patient Preference and Adherence* **8**, 1255.

Stabler B., Siegel P.T., Clopper R.R., Stoppani C.E., Compton P.G. & Underwood L.E. (1998) Behavior change after growth hormone treatment of children with short stature. *The Journal of Pediatrics* **133**(3), 366-373.

Starke M. & Möller A. (2002) Parents' needs for knowledge concerning the medical diagnosis of their children. *Journal of Child Health Care* **6**(4), 245-257.

Stewart J.L. (2000) Uncertainty in childhood illness: A synthesis of the parent and child literature. *Research and Theory for Nursing Practice* **14**(4), 299.

Stewart J.L., Mishel M.H., Lynn M.R. & Terhorst L. (2010) Test of a conceptual model of uncertainty in children and adolescents with cancer. *Research in Nursing & Health* **33**(3), 179-191.

Stheneur C., Sznajder M., Taylor M. & Chevallier B. (2011) Experience of

adolescence in patients treated with GH during childhood. *Pediatric Endocrinology Reviews: PER* **9**(1), 431-440.

Stochholm K., Juul S., Juel K., Naeraa R.W. & Højbjerg Gravholt C. (2006) Prevalence, incidence, diagnostic delay, and mortality in Turner syndrome. *The Journal of Clinical Endocrinology & Metabolism* **91**(10), 3897-3902.

Sullivan-Bolyai S., Deatrck J., Gruppuso P., Tamborlane W. & Grey M. (2003) Constant vigilance: Mothers' work parenting young children with type 1 diabetes. *Journal of Pediatric Nursing: Nursing Care of Children and Families* **18**(1), 21-29.

Sullivan-Bolyai S., Knafl K., Tamborlane W. & Grey M. (2004) Parents' reflections on managing their children's diabetes with insulin pumps. *Journal of Nursing Scholarship* **36**(4), 316-323.

Surkan P.J., Ettinger A.K., Hock R.S., Ahmed S., Strobino D.M. & Minkovitz C.S. (2014) Early maternal depressive symptoms and child growth trajectories: a longitudinal analysis of a nationally representative US birth cohort. *BMC Pediatrics* **14**(1), 185.

Sutton J. & Austin Z. (2015) Qualitative research: data collection, analysis, and management. *The Canadian Journal of Hospital Pharmacy* **68**(3), 226.

Sävendahl L., Maes M., Albertsson-Wikland K., Borgström B., Carel J.-C., Henrard S., Speybroeck N., Thomas M., Zandwijken G. & Hokken-Koelega A. (2012) Long-term mortality and causes of death in isolated GHD, ISS, and SGA patients treated with recombinant growth hormone during childhood in Belgium, The Netherlands, and Sweden: preliminary report of 3 countries participating in the EU SAGhE study. *The Journal of Clinical Endocrinology & Metabolism* **97**(2), 213-217.

Taback S. & Van Vliet G. (1999) Managing the Short Stature of Turner Syndrome: An Evidence-Based Approach to the Suggestion of Growth Hormone Supplementation. In *Current Indications for Growth Hormone Therapy*, 2<sup>nd</sup> edn, (Hindmarsh P.C.,eds). Karger Publishers, pp. 102-117.

Tauber M., Jaquet D., Jesuran-Perelroizen M., Petrus M., Bertrand A.M., Coutant R.

& Group N.F.S. (2013) User assessment of Norditropin NordiFlex®, a new prefilled growth hormone pen: a Phase IV multicenter prospective study. *Patient Preference and Adherence* **7**, 455.

Thomlinson E.H. (2002) The lived experience of families of children who are failing to thrive. *Journal of Advanced Nursing* **39**(6), 537-545.

Thomson R. & Holland J. (2005) 'Thanks for the memory': memory books as a methodological resource in biographical research. *Qualitative Research* **5**(2), 201-219.

Tiwaree C., Kantawng S., Wonghongkul T. & Lertwatthanawilat W. (2016) Predicting factors of uncertainty in illness among children with leukemia undergoing chemotherapy. *Pacific Rim International Journal of Nursing Research* **20**(1), 33-44.

Trevino K.M., Fasciano K. & Prigerson H.G. (2013) Patient-oncologist alliance, psychosocial well-being, and treatment adherence among young adults with advanced cancer. *Journal of Clinical Oncology* **31**(13), 1683.

Vaismoradi M., Salsali M. & Ahmadi F. (2011) Nurses' experiences of uncertainty in clinical practice: a descriptive study. *Journal of Advanced Nursing* **67**(5), 991-999.

Valandra V. (2012) Reflexivity and professional use of self in research: A doctoral student's journey. *Journal of Ethnographic & Qualitative Research* **6**(4), 204-220.

Van der Zalm J.E. & Bergum V. (2000) Hermeneutic-phenomenology: providing living knowledge for nursing practice. *Journal of Advanced Nursing* **31**(1), 211-218.

Van Dongen N. & Kaptein A.A. (2012) Parents' views on growth hormone treatment for their children: psychosocial issues. *Patient Preference and Adherence* **6**, 547-553.

Van Manen M. (1977) Linking ways of knowing with ways of being practical. *Curriculum Inquiry* **6**(3), 205-228.

Van Manen M. (1984) " *Doing*" *Phenomenological Research and Writing: An Introduction*. Publication Services for Dept. of Secondary Education, Faculty of Education, University of Alberta.

Van Manen M. (2016) *Researching Lived Experience: Human Science for an Action Sensitive Pedagogy*, 2<sup>nd</sup> edn. Routledge, New York.

Van Oers H.A., Haverman L., Limperg P.F., Van Dijk-Lokkart E.M., Maurice-Stam H. & Grootenhuis M.A. (2014) Anxiety and depression in mothers and fathers of a chronically ill child. *Maternal and Child Health Journal* **18**(8), 1993-2002.

Van Pareren Y.K., Duivenvoorden H.J., Slijper F.M., Koot H.M., Drop S.L. & de Muinck Keizer-Schrama S.M. (2005) Psychosocial functioning after discontinuation of long-term growth hormone treatment in girls with Turner syndrome. *Hormone Research in Paediatrics* **63**(5), 238-244.

Villimez C., Eisenberg N. & Carroll J.L. (1986) Sex differences in the relation of children's height and weight to academic performance and others' attributions of competence. *Sex Roles* **15**(11-12), 667-681.

Walters A.J. (1995) A hermeneutic study of the experiences of relatives of critically ill patients. *Journal of Advanced Nursing* **22**(5), 998-1005.

Webb C. (2003) Editor's note: Introduction to guidelines on reporting qualitative research. *Journal of Advanced Nursing* **42**(6), 544-545.

Wellington J.J., Bathmaker A.-M., Hunt C., McCulloch G. & Sikes P. (2005) *Succeeding with your Doctorate*. SAGE Publications Ltd, London.

Whitehead H.M., Hadden D. & Carson D. (1989) The Northern Ireland experience of growth hormone therapy for short stature. *The Ulster Medical Journal* **58**(2), 153.

Whitehead L. (2004) Enhancing the quality of hermeneutic research: Decision trail. *Journal of Advanced Nursing* **45**(5), 512-518.

Wickramasuriya B., Casey A., Akhtar S., Zia R., Ehtisham S., Barrett T., Shaw N. & Kirk J. (2006) Factors determining patient choice of device for GH therapy.

*Hormone Research in Paediatrics* **65**(1), 18-22.

Wild R. (1988) Somatonorm post-marketing surveillance study (UK and Ireland, 1986-1987): a preliminary report. *Acta Paediatrica* **77**(s343), 89-94.

Wimpenny P. & Gass J. (2000) Interviewing in phenomenology and grounded theory: is there a difference? *Journal of Advanced Nursing* **31**(6), 1485-1492.

Wojnar D.M. & Swanson K.M. (2007) Phenomenology: an exploration. *Journal of Holistic Nursing* **25**(3), 172-180.

Woodgate R.L. & Degner L.F. (2002) "Nothing is carved in stone!": uncertainty in children with cancer and their families. *European Journal of Oncology Nursing* **6**(4), 191-202.

Woolley S.L. (2011) The limits of parental responsibility regarding medical treatment decisions. *Archives of Disease in Childhood* **96**(11), 1060-1065.

Wright C.M. (2000) Identification and management of failure to thrive: a community perspective. *Archives of Disease in Childhood* **82**(1), 5-9.

Wright S., Yelland M., Heathcote K., Ng S.-K. & Wright G. (2009) Fear of needles: nature and prevalence in general practice. *Australian family physician* **38**(3), 172.

Wysocki T., Gavin L. (2004) Psychometric properties of a new measure of fathers' involvement in the management of pediatric chronic diseases. *Journal of Pediatric Psychology*, **29**(3):231.

## Appendices

### Appendix 1- Participant Information Leaflet

#### Information leaflet

You are being invited to take part in a research study led by Rasha Alsaigh at Trinity College Dublin that will explore parents' experiences of caring for children receiving Growth hormone treatment (GHT).

#### Who am I?

I am Rasha Alsaigh (lead investigator), a PhD student at the School of Nursing and Midwifery, Trinity College. I am completing a Doctoral degree and to do so I need to undertake a study in nursing. Professor. Imelda Coyne who works at the School of Nursing and Midwifery in Trinity College Dublin will supervise this study.

#### Why this letter?

This letter is an invitation for parents who have children receiving GHT to contribute to this research study.

#### What is the study about?

Nothing is known about parents' experiences of caring for a child receiving (GHT). The aim of the study is to explore what this is like for parents.

Some questions you may have about the study:

- What do I have to do to be included in the study?

You need to be a parent (age 18 and above) of a child between the ages of 2-18 years who is currently taking a daily Growth Hormone injection at home for at least six-months. You will need to be able to communicate in verbal and written English.

- What do I have to do in this study?

It will involve an interview with myself, which will take between 30-60 minutes maximum. I will record the interview and if you are happy I may ask to meet with you again for another

interview. I will also ask you to write an account of your experiences and this will take no more than 30 minutes.

- Where will an interview be held?

An Interview will be held at your home or any other suitable venue at your convenience.

What are the benefits of the study?

There will be no direct benefit for you or your child from helping in this study. However, it is hoped that information from the study will help nurses and other professionals to promote better understanding of the needs of parents of children receiving (GHT). No fees will be paid to you for contributing to this study.

What are the risks?

It is unlikely for adverse outcomes to occur but discomfort or distress from sharing personal information could be an issue at an interview. If this happens steps are put in place to support you.

Do I have to participate?

There is no obligation to participate. If you chose to participate, I would appreciate it if you contribute fully in the interview and diary writing. However, if you feel at any time during the study that you wish to withdraw, you could do that by letting me know. By making this decision, you will not be penalised and will not give up any benefits that you had before entering the study. This will not influence your child's care.

What will happen to your information if you wish to withdraw before completing the interviews and written diaries?

I will be using the collected information unless you don't wish me to do so. If that is the case your data will be destroyed.

Stopping the study:

You understand that the investigator may withdraw your participation in the study at any time without your consent.



Is the information confidential?

All means will be taken to ensure that the information provided by you will be kept confidential throughout the study. Documented information will be kept secured. Tape recording will also be kept in a locked drawer. Gathered information from the study will not be used in any other study and will not be shared with anyone other than myself and supervisor. After the study is finished, your Information will not be used in future unrelated studies without future specific permission being obtained and all information and documents will be destroyed after 5 years. You will be offered a transcript copy of your interview and a copy of your diary records.

In the unlikely event that issues related to child protection arise, I am required by the guidelines of the Children First National Guidance to report this. If this occurs, I will advise you before taking action.

What will happen with the information?

The study will be published in a peer-reviewed journal and copy of the summary will be sent to you if you like.

Approval:

This study has Research Ethics Committee approval from The School of Nursing & Midwifery, Trinity College Dublin.

Contact Information of Lead Investigator:

Rasha Alsaigh. PhD student,  
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## Appendix 2- Informed Consent Form

### Informed Consent Form

**Title of the study:** Parents' experiences of caring for children receiving Growth hormone treatment (GHT).

**Lead Investigator:** Rasha Alsaigh RN, BSN, MSc. PhD student (Full Time) at the School of Nursing and Midwifery, Trinity College, Dublin. Tel: 0851997724 Email: [alsaighr@tcd.ie](mailto:alsaighr@tcd.ie)

**This lead investigator is academically supervised by:** Professor Imelda Coyne, Professor of Children's Nursing in the School of Nursing & Midwifery, Trinity College Dublin, Ireland. Tel: +353 1 896 4071 Email: [coynei@tcd.ie](mailto:coynei@tcd.ie)

**Background:** Nothing is known about the parents' experiences caring for a child receiving GHT. The aim is to explore what this is like for parent/parents.

**Participation will involve:** You have agreed to take part in at least one audio recorded interview. This will take between 30- 60 minutes to complete and will be held in a location convenient to you. The researcher will need to interview you at least once, however if another interview is needed, there will be a time gap between the interviews. You may also write an account of your experiences and this will take no more than 30 minutes to complete. All information collected in this study will be treated confidentially and your identity and that of your child will remain confidential at all times and in any related publications. Your information will not be used in future unrelated studies without future specific permission being obtained from you. You will be offered a transcript copy of your interviews and a copy of your diary records.

**Declaration of Consent:** I have read, or had read to me, the information leaflet for this project and I understand the contents. I have had the opportunity to ask questions and all my questions have been answered to my satisfaction. I freely and voluntarily agree to be part of this research

study, though without prejudice to my legal and ethical rights. I understand that I may withdraw from the study at any time and I have received a copy of this agreement.

Participant's Name (Block Letters): \_\_\_\_\_

Contact Details: \_\_\_\_\_

Participant's Signatur: \_\_\_\_\_ Date: \_\_\_\_\_

**Statement of Investigators responsibility:** I have explained the nature and purpose of this research study, the procedures to be undertaken and any risks that may be involved. I have offered to answer any questions and fully answered such questions. I believe that the participant understands my explanation and has freely given informed consent.

Lead Investigator's Signature: \_\_\_\_\_ Date: \_\_\_\_\_

You will receive a copy of this form.

## Appendix 3- Introductory Letter to the Study

### An Introductory Letter to Whom it may Concern

Dear xxxxxx ,

My name is Rasha Alsaigh and at the moment I am undertaking a PhD in the School of Nursing and Midwifery at Trinity College Dublin under the supervision of Professor Imelda Coyne. I have great interest in children who are taking Growth hormone treatment (GHT) and I am studying the “Lived Experience of Mothers who have children on GHT”. Nothing is known about the mother’s experience having a child on Growth Hormone Therapy and the aim of the study is to explore this experience in order to get rich and significant information of its nature. In this qualitative study using a phenomenological approach, I am planning to selectively recruit mothers of children who are between the ages of 2-18 years and who receive a daily subcutaneous GH injection for at least six months. Data will be collected using in depth interviews and diary records. The study will take up to three years and I am hoping to start recruiting in early 2015 following ethical approval from the School of Nursing and Midwifery at Trinity College Dublin and then from your respected organisation. I would really appreciate it if I could meet with you wherever and whenever convenient to discuss this further and to get your support for the study. If you have any questions please contact myself or Professor. Imelda Coyne who will be happy to answer any questions.

Best Regards,

---

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## Appendix 4- Diary

### Diary

#### **“Parents’ Experiences Caring for Children Receiving Growth Hormone Treatment “**

**Name:**

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—

**Date Completed:**

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—

**Important notes:**

- All written information in this diary will be kept confidential.
- I will make sure to remove all references to named people in the diary when I analyse the data.
- It is best to fill your diary soon after the experience so you could reduce the chance of forgetting important information.
- You are not restricted to a number of pages.
- You are welcome to write whatever you want to include in your experience. For example, you could write about your thoughts, feelings and actions that accompany your experience.
- When you complete the diary and would like to forward it to me, please use the envelope provided.
- The diary should be completed by \_\_\_\_\_, if you have completed it before than please just send it in.
- I will contact you \_\_\_\_\_ weeks from now to follow up with you and see how you are getting on with writing the diary.
- If you have any questions about writing this diary please don't hesitate to ask me.

I hope you find participating in this study interesting and thank you once again for your valuable contribution to this study.

Best Wishes,

Rasha Alsaigh.  
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# Appendix 5- Pre-understandings Prior to Starting Data Collection

My pre-understandings

Sources of my Knowledge

1. From my personal experience

1) Before GHT:

- Son was IUGR and was born in the USA SGA.
- Did not thrive.
- Had other problems and health issues.
- Diagnosed at the age of 6 months with RSS.
- We were informed that he would go on GHT at the age of 2 as a treatment plan.

2) Starting GHT:

- Moved to Saudi before starting GHT.
- He started GHT at the very first endocrinology check-up he had in Saudi at the age of 20 months.
- Un expected age to start GHT.
- Didn't get proper explanation and teaching to prepare me for treatment at home.
- No choices of devices were provided.
- No follow up on how treatment was going for me at home.
- We had to pay for 50% of the drug cost every month for nearly 2 years.
- We then discovered we could get it for free through our employers "by chance".
- I mainly give it and sometimes my husband does too.
- My son likes me giving it to him and not his dad.
- No one else gives the injections to him.
- He doesn't want to do it himself.
- Grandparents will not try to learn how to do it "dislike the idea"
- Questioned by family if I should give it or does he really need it.
- Grandfather thinks its cruel.
- His dad and I think that it's for his own good. We want to give him the chance to grow so he would have a better future.
- Son was okay taking them.
- Most days it hurts and some days it bleeds and bruises.
- Constantly asks me till when he will be taking this.
- I don't know that exact answer to that just when he stops growing.
- Sometimes I would have had to inject him twice because I would have forgotten to calibrate the injection after sticking the needle or the last of the vial did not deliver the full dose so a new vial would have to be inserted and injected with the rest of the dose.
- I can't give the injection anywhere other than his backside because his arms and legs are too skinny and it's too painful for him.
- Traveling is hard because we have forgotten the whole cooler with the three-month supply in the airport and trying to get prescription and dispensing a two three-month supply with a high-tech medication is hard, and keeping it cool in the cooler for more than 8 hours (traveling to Saudi Arabia).

- Missing doses because of sleepovers at grandparents or waiting for a prescription refill after running out of them, and sometimes leaving the pen at home at hospital stays and it not being provided at the hospital.
- Have sometimes-conflicting feelings if we have chosen the right thing to do? Are we not exposing him to potential side effects in the future? Is it really helping him grow? Is it worth it in the end?
- Worry about side effects.
- Two follow ups a year.
- I have to nearly reassure him daily that he is taking it for his own good and it's helping him grow.
- Worry about the continuation of the treatment when we go back to Saudi.
- I would like to consider changing the device to try have him do it himself as the one I'm using is very heavy and chunky for his hand to manage easily but worry if the same device would not be available in Saudi when we are back.
- I give most of the injections when he is sleeping because it's painful for him when awake and he like it like that.
- GHT gives me an answer for people who question his size.
- Gives me confidence and a bit of control that I'm doing something for his problem.
- He is still the smallest and it is hard to see him not catching up with the rest of the class.

2. As a researcher

What do I know about the topic?

What I have in the literature review chapter mentions this.

- I have completed a systematic review for my MSc thesis about the effect of GHT on the height of children with RSS.
- I have knowledge about the psychosocial effects of being small and short.
- I have knowledge about the physical effects of GHT.
- The different childhood disorders that may need GHT.
- GHT and the quality of life of the child.
- Compliance or adherence is a hot topic for GHT.

3. From being a nurse

- Worked with sick children.
- Parents play an important role in the care of their sick child.
- The importance of involving younger children in their health care.
- Parents information needs.
- Family centered care.



## Appendix 6- Example of a Reflective Diary Log for One of the Interviews

Before the interview:

I have thought about what I would expect from this interview. Knowing that she is a mother of a child with PWS, I had some expected issues to hear in the interview.

- Getting the child on GHT (delay)
- GHT not being a big issue
- Child having more complex needs.
- Happy with the GHT results.

I am a bit nervous about the interview as it has been a three-month gap from the last conducted interview. I am hoping this interview goes smooth and the café is not too noisy.

Meeting the mother:

We met at the café and she was very friendly with me. She did say that she dismissed me entering the coffee shop as she didn't think I looked like I was old enough to be doing a PhD! I took this as a complement and thought about this as this might have been a barrier for me during recruitment. Parents may not take me seriously because I didn't look mature enough. Bear in mind I tried to dress formally and not look casual.

Interview:

We tried finding the quietest spot to start the interview. I explained as usual the information leaflet, consent, and any questions to be asked. She was happy enough to sign and told me she didn't have any questions to ask as she knew what to expect because she has two boys in college and they were preparing her for the meeting and what I might ask her!

I asked the general question and she told her story.

She did seem to be very saddened of the diagnosis and the regret of not starting the GHT sooner for her daughter. She is very worried about her daughter's health specially being obese and wants to do anything to help her. I could see that she is eager to see GHT results and after a year on GHT, it failed her expectations. She did mention that the Dr did say this is not a magic wand and needs to wait for results but she is disappointed and hopes to see better results in the near future. She hated to see her daughter injecting herself and they hurt sometimes for no obvious benefit in her opinion.

Her daughter is the youngest of 4 children being the only daughter in the family. She is very special to mother and I could see the love and adoration in her eyes when she talked about her so dearly. I can sense her sadness and feeling of regret not putting her on GHT from the start because she had to make that decision and she was reluctant from the start. She mentioned not knowing enough about GHT and not getting properly informed about it and that if she was better prepared and maybe met another parent that would have facilitated her decision and her daughter would have been on it sooner. Her daughter is facing obesity and hopes GHT would help control this. She said that the last visit she is so used to having the nurse say she has put on weight and that was expected but she didn't and she is relating this to the effect of GHT, which is a good thing now.

I gave her the diary and asked her to write what she wants and she said she will and found this very useful in case she remembers something else.

This mother expressed how it is hard for parents and a study like this is much needed to prepare them. She thought this interview is therapeutic in a way as its good that someone is listening to her.

## Appendix 7- Example of the Initial Preliminary Interpretation of Texts to Facilitate Coding

<p>Transcript 1: 160425_0005</p> <p>Code: PMA1</p> <p>Pseudo name: Francis</p> <p>Date of Interview: 25/4/2016</p> <p>So (Francis) tell me about your son:</p> <p>(K) is 11, 12 in October. He was full term bang on the day. He was 8 pound 10 a lovely big baby and never had any problems with him <b>he wasn't a great feeder</b> he kind of you know you could only get a sertant amount in to him and if you went too far and then he'd puke so amm <b>it wasn't until he was around 2</b> that he was always small but <b>I was a big baby too and small so my mother always said amm oh you were a big baby born but you never grew so I just put that down too...</b> he had a few little problems like he had low hearing he'd amm he had to get circumcise he was amm which was very his skin was very tight he had problems with his hips so <b>they were all problems in the early years</b> and then it was only when he'd start kinda play school he was small and then he started ... amm <b>primary school ...he was really small</b> and <b>girls used to swing him around</b> you know things that you really resolve like you know kind of obvious he was small and then where he goes to school girls they amm .... he goes there till first class then they more over to an all boys school its mixed up till first class and it was then he went see when he was in it was blue they wore a blue uniform it was not obvious that he was so small but <b>when he went to the boys school it changed to grey then it was people knew how old he was and then it was obvious because he is very small that's when so .... I had brought him to the doctor</b> and they had said amm <b>I was referred to xxxx they said it was just constitutional short stature and they said we</b></p>	<p>9/1/2017</p> <p>Pre-diagnosis stage:</p> <p>Early years health problem manifestation could be present.</p> <p>2 the catch-up growth period</p> <p>Assuming that he was small because his parents were small (trying to justify).</p> <p>Starting school and wearing a uniform brings the attention to how small he is for his age.</p> <p>Juvenile treatment (treating him as a doll or a baby because he is small)</p> <p>Getting diagnoses:</p> <p>She sought medical attention and they referred him to OLCHC, they diagnosed him with constitutional SS (missed diagnosed)! after a bone age X ray</p>
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## Appendix 8- Example of Open Codes in a Sample Interview

<p>Transcript 1: 160425_0005</p> <p>Code: PMA1</p> <p>Pseudo name: Francis</p> <p>Date of Interview: 25/4/2016</p> <p>So (Francis) tell me about your son:</p> <p>(K) is 11, 12 in October. He was full term bang on the day. He was 8 pound 10 a lovely big baby and never had any problems with him he wasn't a great feeder he kind of you know you could only get a sertant amount in to him and if you went too far and then he'd puke so amm it wasn't until he was around 2 that he was always small but I was a big baby too and small so my mother always said amm oh you were a big baby born but you never grew so I just put that down too.... he had a few little problems like he had low hearing he'd amm he had to get circumeise he was amm which was very his skin was very tight he had problems with his hips so they were all problems in the early years and then it was only when he'd start kinda play school he was small and then he started ... amm primary school ...he was really small and girls used to swing him around you know things that you really resolve like you know kind of obvious he was small and then where he goes to school girls they amm .... he goes there till first class then they more over to an all boys school its mixed up till first class and it was then he went see when he was in it was blue they wore a blue uniform it was not obvious that he was so small but when he went to the boys school it changed to grey then it was people knew how old he was and then it was obvious because he is very small that's when so .... I had brought him to the doctor and they had said amm I was referred to ++++ they said it was just constitutional short stature and they said we</p>	<p>26/1/2017</p> <p>Prefect baby</p> <p>Size</p> <p>Not a great feeder</p> <p>Vomiting</p> <p>Recognising a problem</p> <p>Mother was small</p> <p>Justifying size</p> <p>Health problem in early years</p> <p>Noticing small size at school</p> <p>Girls swinging him around</p> <p>Size more noticeable due to New School uniform colour.</p> <p>Brought to doctor.</p> <p>Referred to hospital.</p> <p>Diagnosis of CSS.</p>
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## Appendix 9- Codes to Categories Done Manually

Code	Category
Period of treatment (treatment duration)	(Treatment duration)
Age starting GHT (starting GHT)	Period of treatment
	(Starting GHT)
Not straightforward (starting GHT)	Age starting GHT
Starting GHT	Not straightforward
	Parents wanting to start GHT
Parents wanting to start GHT (delay) (starting GHT)	Delays in starting GHT
GHT)	Delays in starting GHT
Delays in starting GHT (delay) (starting GHT)	Frustrating wait to start GHT
Waiting for test (delay) (waiting)	Starting GHT
Sleep apnea testing (testing)	Start from the beginning (mothers feeling starting GHT)
Waiting for tonsillectomy (operation) (waiting)	GHT)
(delay starting GHT) (early years health problems)	Only thing you can really do (mothers feeling starting GHT)
Frustrating wait to start GHT (waiting) (delay starting GHT)	Very eager to start GHT
Starting GHT (starting GHT)	Start GHT Quickly (mothers feeling starting GHT)
No difficulties with GHT (taking GHT)	Starting GHT at 2 (guidelines)
Very straightforward (taking GHT)	Before becoming over weight (guidelines)
Endocrinology clinic (hospital visits) (follow ups)	Number of testing (testing)
Following up intervals (follow ups)	Had parents well briefed in Dublin (preparation)
When ENS rings (ENS support)	What would be involved with GHT (preparation)
Instruct to increase dosing (ENS Support)	Checking suitability to GHT
	Mother not remembering problems with tastings (testing)

## Appendix 10- Codes to Categories Using NVivo 11

▶ Starting GHT	21	393	24 Apr 2017 at 16:20	RS	16 Jul 2017 at 18:36	RS	●
▶ Support	22	361	24 Apr 2017 at 15:42	RS	13 Jul 2017 at 12:52	RS	●
▶ Being Small	15	267	25 Apr 2017 at 15:07	RS	13 Jul 2017 at 12:42	RS	●
▶ Getting Diagnosed	17	209	25 Apr 2017 at 12:10	RS	16 Jul 2017 at 18:34	RS	●
▶ Endocrinology Clinic Follo...	19	201	26 Apr 2017 at 13:01	RS	16 Jul 2017 at 18:38	RS	●
▶ Prior to GHT	16	198	25 Apr 2017 at 15:58	RS	11 Jul 2017 at 12:41	RS	●
▶ Positive Effects of GHT	21	156	24 Apr 2017 at 15:14	RS	16 Jul 2017 at 18:51	RS	●
▶ Being on GHT	19	151	25 Apr 2017 at 16:05	RS	15 Jul 2017 at 13:37	RS	●
▶ The GHT Device	16	142	24 Apr 2017 at 16:00	RS	13 Jul 2017 at 12:56	RS	●
▶ Seeking Information	15	135	24 Apr 2017 at 13:16	RS	13 Jul 2017 at 12:59	RS	●
▶ Opening Up to Others Ab...	18	131	24 Apr 2017 at 14:50	RS	16 Jul 2017 at 18:50	RS	●
▶ Negative Effects of GHT	19	125	24 Apr 2017 at 15:31	RS	16 Jul 2017 at 18:52	RS	●
▶ Compliance to GHT	16	95	19 Apr 2017 at 01:56	RS	15 Jul 2017 at 13:36	RS	●
▶ Recieving the Diagnosis	16	91	25 Apr 2017 at 15:56	RS	15 Jul 2017 at 12:53	RS	●
▶ The Future	16	75	25 Apr 2017 at 15:30	RS	16 Jul 2017 at 18:33	RS	●
▶ Traveling with GHT	14	49	25 Apr 2017 at 14:54	RS	16 Jul 2017 at 18:29	RS	●
▶ Stopping GHT	17	48	25 Apr 2017 at 16:07	RS	16 Jul 2017 at 18:52	RS	●
▶ Storage	12	41	25 Apr 2017 at 14:38	RS	13 Jul 2017 at 11:50	RS	●
▶ Cost	10	39	24 Apr 2017 at 12:48	RS	13 Jul 2017 at 12:53	RS	●
▶ Dose adjustment	6	12	15 May 2017 at 11:40	RS	5 Jul 2017 at 17:10	RS	●

## Appendix 11- Example of Finding Subcategories

Traveling with GHT

Internals\Interviews\interview 14 Loren

Reference 3: 1.40% coverage

Now, when he goes to Cubs and when he goes on hikes with the Cubs, we have to follow to give the injection because they wouldn't give an injection and he's not able to self-inject because he doesn't have the strength in his hands to press the plunger. So that will be the next-, is when he's doing it himself. Although then they'll be doing certain deals to-, is he actually doing them or are they getting flushed down the toilet or into his sister or into a teddy? It's too important for us to leave him responsible for it. (parents giving the injections)

Note 6-10-2017: (change in routine)

I can see here that when they are leaving the home environment and entering a different environment outside the home like traveling in a car or a plane or staying at a hotel or boat, they are faced with much hassle to adapt to the change of routine. They will have to do think a head and prepare from the needs for starting the journey (ask for letter, remembering the supply needed, choosing the suitable holiday accommodation to suit storage recruitments) , going on the journey (questioned and delays at checkpoints, carrying sharps, the distance and length of the journey to suit storage recruitments), staying at the accommodation (the availability of proper storage facilities, being exposed to others, remembering to give injection).

Internals\Interviews\Interview 7 Bernadette + Barry

1 reference coded, 2.02% coverage

Reference 1: 2.02% coverage

Yes. We went to Cork in the summer. Now, we only went for four days, but we were a bit-, because initially, you were supposed to keep it in the fridge every day, but the lady from the Saizen company said you can leave it out of the fridge for up to seven days. So that was fine because initially we thought, how are we ever going to go anywhere? We're going to need to go someplace with a fridge, but no we were fine. We just administered it as normal. (worry about traveling and storage)

Internals\Interviews\Interview 1 Francis

18 references coded, 1.14% coverage

References 1-4: 0.17% coverage

I says am how you getting on he said grand he said nearly forgot the injection. I had to run up and get it out of the fridge (difficulties when traveling) (possibility of forgetting to give the injection)

References 5-6: 0.14% coverage

So he was ooh did I cause he said he had to run to what ever the reception was and get it back off them. (difficulties when traveling) (possibility of forgetting to give the injection) (leaving it at reception in inconvenient) (can cause forgetting the injection)

References 7-8: 0.04% coverage

they were trying to remember.

References 9-10: 0.11% coverage

[son] might have even forgotten him self because he was enjoying himself so. (possibility of forgetting to give the injection)

Reference 11: 0.28% coverage

you have to bring it in an they give you a little container and you use ice packs you know I'm sure it be like bringing baby's' food away or what ever you'd you know you would have to keep it refrigerated

References 12-13: 0.10% coverage

any place we have stayed in were very good you know they would mind it (finding storage facilities but preface to having a personal fridge in the room)

References 14-18: 0.30% coverage

you are always kind of worried would anything happen to it or you know cause it GH. But they would not know what GH so you'd be afraid would it get nicked you know what I mean or something because it's such a big. (worry about storage)

Difficulties traveling:

Note 6-10-2017: (change in routine)

I can see here that when they are leaving the home environment and entering a different environment outside the home like traveling in a car or a plane or staying at a hotel or boat, they are faced with much hassle to adapt to the change of routine. They will have to do think a head and prepare from the needs for starting the journey (ask for letter, remembering the supply needed, choosing the suitable holiday accommodation to suit storage recruitments), going on the journey (questioned and delays at checkpoints, carrying sharps, the distance and length of the journey to suit storage recruitments), staying at the accommodation (the availability of proper storage facilities, being exposed to others, remembering to give injection).

Many mothers were able to adapt to this and are ok with the preparations that go with traveling with GHT.

Many mothers don't have the experience of traveling long distances with GHT and only know what to do if needed because they were fully informed of that.

Difficulties

Internals\Interviews\interview 14 Loren

3 references coded, 4.72% coverage

Reference 1: 1.24% coverage

**Travelling with the medication is a nightmare as well.** If the letter from the doctor isn't specific enough-, now, luckily for us I was stopped once and when I explained what it was for and showed her my letters, they checked into it and they agreed, but you're not allowed to bring sharps on the plane, but I don't have an alternative because they can't travel in the language because if I haven't got it I can't get it. So that's a logistical nightmare as well. **(difficulties traveling) (the need for a letter) (not able to carry needles) (airport or airlines restrictions) (check points)**

Reference 2: 2.08% coverage

We travel every year, but my thoughts on it is there should be a specific-, for everybody, not just for growth hormones, but for anyone who uses sharps, there should be a specific travel case that a carrier would have that's deemed safe and secure by airports throughout the world and then we'd be allowed free without-, now, they don't charge, free travel through so we don't have the hassle of explanations because all my children have autism and if we're stopped to discuss these little injections, these little syringes that I'm carrying. I have to, I don't have a choice. It's just another logistical disaster, not really thought-out. Then it's probably because there's a very small percentage of the population that have to do this so, we're not priority. **(difficulties traveling) (being stopped for sharps and to explain) (make traveling even more difficult for this mothers)**

Internals\Interviews\Interview 10 Louise

2 references coded, 3.38% coverage

Reference 1: 1.61% coverage

The travelling, extra precautions have to be taken just in terms of making sure that we bring the freezer container, making sure that it's refrigerated again as soon as we arrive at whatever destination, making sure that the staff knows which needle goes in the fridge and what goes in the freezer, but it's all perfectly manageable **(difficulties traveling due to storage)**

Reference 2: 1.77% coverage

Once or twice we've gone away for the weekend and brought the medicine, but forgotten the needles and we might phone the Pfizer representative. The nurse who is in New Ross and she's great as well and she might fax a request through to whatever local chemist we're going to in the locality we're going to with a request for the needles. So she's been very obliging as well. **(difficulties traveling due to forgetting something) (getting support for it).**

## Appendix 12- Core Categories and Subcategories

### Initial manifestations

- Prenatal problems, prematurity and stay at the NICU or SCBU
- Delayed milestones
- Feeding issues
- Growth deterioration
- Being small

### Seeking medical attention and support

- Multiple hospital visits and follow-ups including testing
- Surgeries
- Other medical treatment needed (devices and medication)
- Initial diagnosis or no diagnosis at all

Need for supportive therapies (dietician, speech therapy and OT)

### Prior to starting GHT

- Prenatal problems, prematurity and stay at the NICU or SCBU
- Accompanied health conditions
- Delayed milestones
- Feeding issues
- Multiple hospital visits and follow-ups including testing
- Surgeries
- Other medical treatment needed (devices and medication)
- Initial diagnosis or no diagnosis at all
- Need for supportive therapies (dietician, speech therapy and OT)
- Growth deterioration

Being small

- Parent noting child being small
- Self-Reassurance about size
- Size being an issue of concern
- Others reaction to child's size
- Protecting the child

### Getting Diagnosed

Reaching to the point of diagnosis

- Early Manifestations
- Delays reaching to final diagnoses
  1. The initial diagnosis of CSS and being reassured that the child would catch up.
  2. Parents being small.
  3. HP (GP) dismissing mothers concern of child's growth and not measuring growth properly.
  4. The way Endo handles mother's concern as serious or not (listening).
- Diagnostic tests
  - Difficulties faced by parents:
    1. Hard (scary) experience.
    2. Delay getting the test done (MRI)
    3. Difficulties remembering medical terms.
    4. Worry about MRI results (tumour).
    5. Not having conclusive test results to accept the diagnosis.

Receiving the diagnosis

1. State of receiving the diagnosis

2. No closure after receiving the diagnosis
3. Information about the diagnosis: (shared with seeking information code)
4. Manner in which diagnosis was delivered
5. Coping with the diagnosis
6. Medical professionals overlooking other medical issues after reaching current diagnosis
7. GHT not resolving all the initial manifestations????

#### Starting Treatment

- Making the decision to start GHT
1. Motives (Being aware of the availability of GHT being an option of treatment)
  2. Facilitators
- Difficulties in making the decision to start:
1. Not having an exclusive test result to reassure the need for GHT
  2. Doubting the need for GHT
  3. Not trusting the consultant
  4. Not knowing till when the child will be taking GHT
  5. Not having enough time to process the decision to start GHT
  6. Not enough explanation to help make the decision and support
- Timing of starting GHT
1. Happy
  2. Wanting it sooner
- Involving the child in DM
  - Emotions and worries starting injections

#### Living with GHT or managing GHT

##### The Injections

- Challenges
1. Emotional
  2. Technical and physical
- Facilitators
1. Dealing with phases of child not wanting GH injections

##### Device

- Choices (given different choices)
- Advantages of the devices (also having good support)
- Disadvantages (also lacking the support)
- Working with what they have (this also is getting used to the device eventually and feeling comfortable with it in the long run)
- Support (lacking” information and contact”, getting support” information and close contact “and reaching the supplies)
- At the start (can be with working with the device) (getting used to it eventually)
- Hopes for changes

##### Storage of GHT

- Inconvenience of storage in and out of home
- Change of storage guidelines
- No problems with storage

##### Traveling with GHT

- Difficulties
- Trying to adapt to traveling needs (well instructed, being cautious, having preferences, finding alternatives)

##### Compliance or adherence to GHT

##### Facilitators

- Training other family members for back up
- Parents giving the injection on camps.
- Parent giving the injection before leaving the child.



- Child or other members of the family reminding parents of the injection.
- Child growing older and accepting the injection more. (dealing with difficulties)
- Not skipping for any reason like illness or hospital stays.
- Giving it in his sleep. (dealing with difficulties)
- Reducing the pain during injecting. (dealing with difficulties)
- Making child feel guilty for refusing the injection. (dealing with difficulties)
- Having a set routine.
- Parent taking it seriously (know the child needs it, feel responsible to give it)
- Child self-injecting.
- Child's personality.
- History track on the device.
- Parent following up on child who is self-injecting.

#### Difficulties

- Child not self-injecting
- Traveling or sleep overs
- Remembering and forgetting
- Concealing
- Finding others who are willing to inject
- Trusting others to inject
- Technical issues
- Not seeing positive results

#### Reasons for missing injections

- Traveling
- Running out of GHT (bank holiday weekend)
- Sleepovers
- Having permission
- Forgetting
- Not trusting others to inject

#### Reaction after missing a dose:

- Making up for it during the week
- Letting it go "He is not going to die he can miss a few"
- Its ok "Having permission" or device not allowing to give the dose the next morning
- Give it in the morning

#### Cost

- Sense of appreciation (comparing countries)
- Consequences of the high cost
- Have Medical Card
- Trying for a Medical Card
- Issues with cost

#### Endocrine Clinic Follow ups

##### Patient-physician relationship:

- Communication
- Trust

##### Hospital visits

- Routine (frequency, attendance)
- Blood tests (hard, waiting on results, upping dose) (adjusting the dose)
- Location

Chosen service (public or private, choice of hospital, being under more than one hospital)

#### Dose adjustment

- Increasing the dose
- Reducing the dose

#### Positive effects of GHT

- Positive effects noted by parent
- Positive effects noted by Endo
- Seeing results help with coping
- Expectations
- Needed support
- Uncertainty around the positive effects of GHT

#### Negative Effects of GHT

- Uncertainty about SE
- Triggering the thought of SE
- Worries about SE
- Coping with SE
- Not having SE
- support needed
- Source of Information

#### Information needs

- Lack of information
- Seeking information
- 1. Type of information
- 2. Sources of information (reliable or not)
- 3. Importance
- Avoiding information

#### Opening up to others

##### Isolation

- Reasons for not going into much details
- 1. People not understanding
- 2. Having to justify why
- Circumstances of telling
- 1. Making it normal
- 2. Not being ashamed of it
- 3. Opening up only when needed
- 4. Justifying
- 5. Being judged (stigma)

##### The future (uncertainty)/Quality of life

#### Stopping GHT

##### Uncertainty and unknowingness

- Length of treatment
- Continuity of the treatment
- Potentially stopping GHT due to SE
- consequences of stopping the GHT
- Life time treatment

## Appendix 13- Synthesis and Initial Theme Development

Theme 1: Life before GHT (uncertainty)

- The start of the journey (Rocky or smooth)
- Noting the problem (I knew something was going on)

### **Initial manifestations**

- Prenatal problems, prematurity and stay at the NICU or SCBU
- Delayed milestones
- Feeding issues
- Growth deterioration
- Being small

### **Seeking medical attention and support**

- Multiple hospital visits and follow-ups including testing
- Surgeries
- Other medical treatment needed (devices and medication)
- Initial diagnosis or no diagnosis at all

Need for supportive therapies (dietician, speech therapy and OT)

- Getting diagnosed and receiving the diagnosis

### **Getting Diagnosed**

Reaching to the point of diagnosis

- Early Manifestations

Delays reaching to final diagnoses

The initial diagnosis of CSS and being reassured that the child would catch up.

Parents being small.

HP (GP) dismissing mothers concern of child's growth and not measuring growth properly.

The way Endo handles mother's concern as serious or not (listening).

- Diagnostic tests

Difficulties faced by parents:

Hard (scary) experience.

Delay getting the test done (MRI)

Difficulties remembering medical terms.

Worry about MRI results (tumour).

Not having conclusive test results to accept the diagnosis.

Receiving the diagnosis

State of receiving the diagnosis

No closure after receiving the diagnosis

Information about the diagnosis: (shared with seeking information code)

Manner in which diagnosis was delivered

Coping with the diagnosis

Medical professionals overlooking other medical issues after reaching current diagnosis

GHT not resolving all the initial manifestations????

- Becoming aware of GHT (knowing and not knowing about GHT)

Theme 2: The journey of starting GHT (not so strait forward)

(uncertainty)

dealing with the unknown and worrying

the only thing you can really do is growth hormone

I just think the chance is there to help him and what do you do, but do it?

- Making the decision

- Side effects
- Positive effects
- Hopes
- Communication and lack of information
- Cost

### **Starting Treatment**

- Making the decision to start GHT

Motives (Being aware of the availability of GHT being an option of treatment)

Facilitators

- Difficulties in making the decision to start:

Not having an exclusive test result to reassure the need for GHT

Doubting the need for GHT

Not trusting the consultant

Not knowing till when the child will be taking GHT

Not having enough time to process the decision to start GHT

Not enough explanation to help make the decision and support

- Timing of starting GHT

Happy

Wanting it sooner (not happy)

- Involving the child in DM
- Emotions and worries starting injections

Theme 3: Adjusting to life with GHT

### **In the home environment**

- Starting of injection
- Technical Issues
- Adherence
- The device
- Storage
- Cost

### **Outside of the home environment**

- Hospital visits
- Technical issues (travel and storage)
- Opening up to others (they don't understand)
- Reaching out to others for support (they don't understand and stigma)

### **Living with GHT or managing GHT**

#### **The Injections**

- Challenges

Emotional

Technical and physical

- Facilitators

Dealing with phases of child not wanting GH injections

#### **Device**

- Choices (given different choices)
- Advantages of the devices (also having good support)
- Disadvantages (also lacking the support)
- Working with what they have (this also is getting used to the device eventually and feeling comfortable with it in the long run)
- Support (lacking "information and contact", getting support "information and close contact" and reaching the supplies)

- At the start (can be with working with the device) (getting used to it eventually)
- Hopes for changes

#### **Storage of GHT**

- Inconvenience of storage in and out of home
- Change of storage guidelines
- No problems with storage

#### **Traveling with GHT**

- Difficulties
- Trying to adapt to traveling needs (well instructed, being cautious, having preferences, finding alternatives)

#### **Compliance or adherence to GHT**

##### Facilitators

- Training other family members for back up
- Parents accompanying the child at camps.
- Parent giving the injection before leaving the child.
- Child or other members of the family reminding parents of the injection.
- Child growing older and accepting the injection more. (dealing with difficulties)
- Not skipping for any reason like illness or hospital stays.
- Giving it in his sleep. (dealing with difficulties)
- Reducing the pain during injecting. (dealing with difficulties)
- Making child feel guilty for refusing the injection. (dealing with difficulties)
- Having a set routine.
- Parent taking it seriously (know the child needs it, feel responsible to give it)
- Child self-injecting.
- Child's personality.
- History track on the device.
- Parent following up on child who is self-injecting.

##### Difficulties

- Child not self-injecting
- Traveling or sleep overs
- Remembering and forgetting
- Concealing
- Finding others who are willing to inject
- Trusting others to inject
- Technical issues
- Not seeing positive results

##### Reasons for missing injections

- Traveling
- Running out of GHT (bank holiday weekend)
- Sleepovers
- Having permission from the Endo.
- Forgetting
- Not trusting others to inject

##### Reaction after missing a dose:

- Making up for it during the week
- Letting it go "He is not going to die he can miss a few"
- Its ok "Having permission" or device not allowing to give the dose the next morning
- Give it in the morning

#### **Seeing the positive effects of GHT**

- Positive effects noted by parent

- Positive effects noted by Endo
- Seeing results help with coping
- Expectations
- Needed support
- Uncertainty around the positive effects of GHT

#### **Worrying about negative Effects of GHT**

- Uncertainty about SE
- Triggering the thought of SE
- Worries about SE
- Coping with SE
- Not having SE
- support needed
- Source of Information

#### **Cost**

- Sense of appreciation (comparing countries)
- Consequences of the high cost
- Have Medical Card
- Trying for a Medical Card
- Issues with cost

#### **Endocrine Clinic Follow ups**

Patient-physician relationship:

- Communication
- Trust

Hospital visits

- Routine (frequency, attendance)
- Blood tests (hard, waiting on results, upping dose) (adjusting the dose)
- Location

Chosen service (public or private, choice of hospital, being under more than one hospital)

#### **Dose adjustment**

- Increasing the dose
- Reducing the dose

#### **Opening up to others (isolation)**

Isolation

- Reasons for not going into much details

People not understanding

Having to justify why

- Circumstances of telling

Making it normal

Not being ashamed of it

Opening up only when needed

Justifying

Being judged (stigma)

Theme 4: Life with or without GHT “unknowingness”

Future uncertainty

- The continuity of treatment
- The continuity of care
- Potential side effects

**The future /Quality of life**

### **Stopping GHT**

Uncertainty and unknowingness

- Length of treatment
- Continuity of the treatment
- Potentially stopping GHT due to SE
- consequences of stopping the GHT
- Life time treatment

Theme 5: Information and support needs

Seeking information

Lack of information

Lack of support

#### **➤ Information and Support needs**

##### **Information needs**

- Lack of information
- Seeking information

Type of information

Sources of information (reliable or not)

Importance

- Avoiding information

## Appendix 14- Example of a Reflexive Log

A day at the clinic 23/5/16:

Early morning:

“I am facing a lot of difficulties recruiting and the gatekeepers will not distribute the packs for me so they have asked me to go to the waiting area and call out the names they marked on the patients list that they can remember as patients on GHT! I had to stand in a huge room full of patients attending different clinics how awkward!!! I am also very self-conscious about how I look “wearing a hijab” and I am worried that people are going to miss judge me and may feel threatened by my appearance. I am always considering this as potentially problem in terms of gaining potential participant’s trust at their first encounter with myself”.

Mid-day:

“I came across a mother with a child and I asked her if she was the parent of ++? She smiled and said yes. Then I started to explain to her who I was and what I am doing and she agreed to take the study pack and to call her back in a week to follow up on her decision to participate. I am very glad she took the pack and I feel like I am finally getting the packs distributed.”

At the interview 13/9/16:

“This mother asked me if there were many parents participating in the study. I told her I have not been getting many at the time and that I was having some difficulties getting the information packs around and finding parents. As I was mentioning that I have posters and others distributing the packs to try increase the chances of getting more parents to participate, she said she only wanted to participate after meeting me personally. She said if one of the staff at the clinic had handed her the pack she wouldn’t have participated. She said if she hadn’t met me that day and heard about the study personally from myself, she probably would have never considered participating! It was only because she had met me and talked with me that she felt comfortable making the decision to participate”.

After thoughts:

“Before this incident, I would have felt that being at the clinic and meeting the parents to give them the information packs was a bad idea!! I didn’t really have the confidence as well. But I didn’t have a choice really as the gatekeepers didn’t leave me with another choice. They gave me the permission to talk to the parents and go ahead with distributing the packs. But after hearing this participant’s comment, my confidence attending the clinics and approaching other parents has definitely boosted. I also have lower expectation of my



appearance negatively influencing further recruitment.”

## Appendix 15- Examples of Annotations in NVivo

That feeling of, I could hear American parents who are putting their kids on medication so they'd bulk up so that they can make the football team or something. I really feared that I was in that category, even though I know I wasn't, but I sort of still feared it and when you started-, we went back about a year later and she said... and she doesn't en... ages with us but she said something along the lines of, where he started on the percentile, he ne... hormone. So yes he absolutely needed it." a... still felt-, even a month into it, I still felt like him perfect. I still had that fear and I more... other thing that they explained to us is that it... wellbeing and bone density and all those, ... suppose it's when they started to describe that kind of thing, the wellbeing thing, that's what made me think back to when he was a baby then, to think if he was running on 85% energy whereas, another baby was running on 100% energy, is that why he was a bit sluggish, is that why the feeding didn't work out? Is that why he needed speech therapy? We'll never know. We don't know.

Created: 17 May 2017

By: RS

this stigma about GHT!!!! could this effect how mothers see GHT and starting on GHT ??

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get your support if you needed it other than the healthcare:

R: I suppose we haven't really needed support. The bits that have been hard we've just figured out. His GP is absolutely lovely, but he felt that if the issue was severe enough to refer him

Created: 17 May 2017

By: RS

having a strong justification to follow up on it and start GHT this i have seen with interview 4

his level of expertise. So if we ever went back to him second... ng to figure out, the impression was very much, if that is what... hat's the best advice you can take so there's no point in me... , so I suppose he's very nice, but I suppose not ready for a... ecialised of course isn't it. I guess that's about it then really... other really.

Delete

about your experience?

R: The most traumatic thing of the whole experience was-, I don't know how to articulate it. Just that if he'd a broken leg, of course you'd go to the doctor and get a cast for it, but if somebody's eyes were blue and you'd rather they'd be brown, you wouldn't go to the doctor about that and in as much as that's a continuum where his level of severity was and whether it was appropriate to follow up on it and whether it wasn't. That was the hardest thing. Especially because I and my husband felt differently about it. That was the hardest thing I would say.