

**Parental Experiences of Secondary Dystonia and the Journey
through Deep Brain Stimulation Surgery**

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June, 2015

***Research submitted in partial fulfilment of the requirements for the degree of Doctor in
Clinical Psychology (DClinPsy), Royal Holloway, University of London***

Acknowledgements

I would like to thank Dr Tamsin Owen for her kindness, time and endless encouragement. As my internal and external supervisor, her advice and expertise has been invaluable to all aspects of this study.

I would like to thank Kieran and my family for all their love, understanding and emotional support throughout this process. Thank you also to my fellow trainees for keeping me company in this journey.

Finally I would like to thank the eight parents who generously agreed to take part in this study. I was touched by their positivity, resilience and unwavering love and commitment to their children. It was a privilege to hear their experiences, and I hope that this thesis will come to benefit them and others in their position.

Abstract

Secondary Dystonia is a heterogeneous movement disorder which profoundly impacts the lives of children and their families. Deep Brain Stimulation (DBS) Surgery is widely used in the treatment of childhood dystonia. However, motor improvements are more subtle in secondary dystonia and impairment measures have failed to capture the subjective meaning of post DBS change or the functional concerns of parents. Studies have largely ignored the psychological, social and emotional impact of secondary dystonia on children, and parents' experiences have been neglected.

This study aimed to move beyond a disability and impairment based conceptualisation of secondary dystonia to consider the lived experiences of parents and children with secondary dystonia. It also hoped to gain an insight into DBS decision making, the experiences of going through DBS surgery and the meaning of post surgery change. Semi-structured interviews were completed with eight parents of children with secondary dystonia who had undergone DBS surgery. Interpretative Phenomenological Analysis (Smith, Flowers & Larkin, 2009) was used to identify themes and connections across parents' accounts. Four superordinate themes emerged: 'a difficult life with disability', 'the meaning of disability and normality', 'an emotional and uncertain DBS journey' and 'the experience and perceptions of change'.

Findings highlight secondary dystonia to be a multifaceted socially bound phenomenon that significantly impacts the lives of children and parents. A defining feature was the lack of and search for control. Decision-making was experienced as a process fraught with uncertainty. This decision was the beginning of an emotional, uncertain and turbulent journey through DBS that tested the resilience of families. There was huge variability in DBS outcomes and the subjective meaning of change. Several research and clinical recommendations for healthcare professionals are suggested to meet the unique needs of this client group.

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Chapter 1: Introduction

1.1 Overview

Secondary Dystonia is a movement disorder, which profoundly impacts the lives of children, and their families. Dystonia impacts a child's physical functioning, as well their participation in activities of daily living, social functioning and psychological well-being. Children with secondary dystonia experience a greater severity of disability and have lower functioning capacity (Lin, Lumsden, Gimeno & Kaminska, 2014). These children are therefore dependent on parents for physical support, placing physical and emotional demands on them. Deep Brain Stimulation (DBS) is a neurosurgical procedure widely used in the treatment of childhood dystonia. However, motor improvements are more subtle in secondary dystonia (Lumsden et al., 2012), and impairment measures have failed to capture the subjective meaning of post DBS changes, or the functional priorities and concerns of parents (Gimeno, Tustin, Selway & Lin, 2012; Lumsden, Gimeo, Tustin, Kaminska & Lin, 2015).

The experiences and impact of parenting a child with dystonia has been neglected in the literature. Interpretative Phenomenological Approach (IPA) was chosen to capture the subjective meanings and narratives of parents of children with secondary dystonia, and privilege the experiences of decision-making, managing the DBS process and perceptions of change post surgery. Parents' narratives have also offered an invaluable insight into the previously ignored experiences of living with severe secondary dystonia and being dependent on an implanted device.

This chapter will begin by discussing the clinical features and impact of dystonia, DBS and summarise DBS surgery outcomes. Next, the concepts of disability and quality of life, the weaknesses of current disability measures and the limitations of an impairment focus will be outlined. Models of disability will then be put forward, to consider the wider systemic context relevant in childhood dystonia. A broad consideration will be given to the factors of parental coping, adaptation, resilience and carer burden that have emerged in the quantitative and

qualitative studies of chronic illness and disability. The context of DBS decision-making and hospital experience will be reviewed in light of wider literature on surgery decision-making and parental surgery experiences of children with chronic illness or disability. Finally, the existing qualitative studies exploring the impact of dystonia and DBS on adults and children will be discussed. Gaps in the literature will be considered, and the importance of capturing parental narratives and experiences will be summarised. Finally, the rationale for adopting a qualitative approach and research aims will be outlined.

1.2 Background Information

1.2.1 Defining Dystonia

Dystonia refers to a relatively common heterogeneous group of movement disorders that can affect both children and adults. It is estimated that there are currently approximately 70,000 individuals with dystonia in the UK (the Dystonia Society, May 2015). Childhood dystonia is thought to represent one of the most challenging clinical problems because of its heterogeneity, multiple aetiologies and differences to adult dystonia (Lin et al., 2014).

In adults and children, dystonia is characterised by involuntary sustained or intermittent muscle contractions, which frequently cause repetitive movements and/or abnormal postures (Sanger et al., 2010). More recently this definition has been broadened to capture how “dystonic movements are typically patterned, twisting and may be tremulous” and that “dystonia is often initiated or worsened by voluntary action and associated with overflow muscle action” (Albanese et al., 2013, p.866). Dystonic movements are characterised by an inability to inhibit unwanted movements because of abnormal activation patterns of groups of striatal neurons in the cerebellothalamocortical basal ganglia circuits (Mink, 2003). These movements can vary from slow twisting writhing movements (athetosis), to more rapid and jerky movements. Dystonia can be triggered and exacerbated when performing voluntary movements (e.g. walking, writing) or maintaining function, and can fluctuate in presence and severity over time (Volkman & Benecke, 2002).

There has been longstanding debate about the classification and aetiology of dystonia syndromes. Dystonia has historically been classified by aetiology, either as primary or secondary. Primary or idiopathic dystonia is a movement disorder of unknown exogenous cause, where dystonia is the only neurological feature (Phukan, Albanese, Gasser & Warner, 2011). In secondary or acquired dystonia, the dystonia develops secondary to other conditions such as cerebral palsy, neurometabolic, autoimmune, genetic and neurodegenerative conditions (Sanger et al., 2010). However, after a recent consensus update, it is now widely recommended (Albanese et al., 2013) that a broader classification system should be adopted which classifies dystonia along two axes; clinical characteristics (age at onset, body distribution, temporal pattern, associated features) and aetiology (nervous system pathology and whether dystonia is inherited or acquired). This broader classification system highlights the heterogeneity of this disorder, and the challenge for researchers investigating this complex disorder.

Dystonia can vary in distribution and affect the body in varying degrees. In focal dystonia, one body region is affected, commonly involving the trunk, upper or lower limbs. However, in generalised dystonia the trunk and at least two other sites are involved. Similarly, dystonia can vary in severity of gross motor function and functional limitations. Dystonia is further differentiated by whether the disease course is static or progressive, and can show daily and momentary variability in movements.

1.2.2 Childhood Dystonia

It is widely accepted by clinicians that the causes and associated features of dystonia are different in childhood and adulthood (Mink, 2013). Childhood dystonia is characterised by heterogeneity in clinical features, dystonia severity, age of onset, disease course and associated co-morbidities (Roubertie et al., 2002). In children, dystonia is often more generalised compared to adult onset dystonia (Egmond et al., 2014). The presence of childhood dystonia is further complicated by the impact of dystonia on the developing brain, growing musculoskeletal system and functional adaptation to increasing demands (Lin et al.,

2014). However, the impact of dystonia throughout childhood is not well described, and a clearer understanding of the non-motor features and quality of life could help inform treatment planning.

In childhood, secondary dystonia is more prevalent than primary dystonia (Roubertie et al., 2002). Dyskinetic cerebral palsy is the most common cause of secondary childhood dystonia (Lin et al., 2014). Cerebral Palsy (CP) is a clinical diagnosis that encompasses a group of disorders that cause impairment of movement and posture, following disturbance to the developing foetal or infant brain (Rosenbaum et al., 2007). Children with Dyskinetic CP experience choreoathetosis (combination of Irregular migrating contractions and, twisting and writhing) and dystonia (Rosenbaum et al., 2007).

Recently, a single NHS site retrospectively analysed the records of 279 children with dystonia (Lin et al., 2014). Secondary dystonia was more common than other subtypes (82.4%), with dystonic CP accounting for 53.7% of the sample. Furthermore children with secondary dystonia were shown to spend a higher proportion of life living with dystonia, experience a greater severity of disability and have lower functioning capacity. In two thirds of the sample, carers also perceived dystonia to be worsening over time. This study supports existing literature conclusions of the prevalence and severity of childhood secondary dystonia. A growing quantitative evidence base has focussed on defining and classifying motor functioning in dystonia. There is a need to understand the wider psychological and social experiences of secondary dystonia, and impact on family quality of life.

1.2.3 The Impact of Dystonia

Dystonia impairs intentional movement, causing physical disability, functional impairment and often pain. This understandably, to varying degrees, prevents children from leading 'normal lives', participating in daily activities and leads to dependence on family members. In children,

dystonia can also adversely impact growth and development, as well as participation in education and age-appropriate social and developmental activities.

There have been some quantitative studies investigating the wider social and psychological consequences of dystonia. The impact of dystonia on health-related quality of life is well established (Page, Butler & Jahanshahi, 2007), and individuals often also have cognitive and psychiatric co-morbidities which further influence their quality of life. It is common for individuals with dystonia to experience non-motor symptoms of alteration in mood, cognition and sleep (Kuyper, Parra, Aerts, Okun & Kluger 2011). Repetitive spasms and abnormal postures make activities such as play, and activities of daily living (washing, dressing or feeding) particularly difficult, potentially leading to feelings of depression, anxiety and isolation (Lim, 2007). Dystonia is also shown to have a negative impact on body image and self-esteem (Jahanshahi & Marsden, 1990). However, these studies have predominantly been focussed on adult samples of primary dystonia, and there is an urgent need to understand the unique social and psychological impact of childhood dystonia.

Secondary dystonia affects a child's physical appearance and often their ability to communicate. This will alter a child's ability to socially interact, and potentially their developing sense of self and health related quality of life. Indeed, in the adult literature perceived disfigurement, negative body concept and low self-esteem associated with dystonia have been shown to be major contributors to depression (Lewis, Butler, & Jahanshahi, 2008), and are associated with higher perception of stigma (Papathanasiou, MacDonald, Whurr & Jahanshahi, 2001). Furthermore, children with secondary dystonia (especially those who are more severely and functionally impaired) will be more dependent on parents and carers for support. This dependence on parents and carers for support has been shown to significantly impact on a child's ability to learn independent living skills, a sense of autonomy and the developing sense of self (Pugh, 2008). This dependence also places additional physical and emotional demands on parents. Undoubtedly, families affected by dystonia experience unique physical, social and emotional challenges (Bakowski, 2010), and assume roles beyond the normative activities of parenting.

1.2.4 Management of Dystonia

Treatment options for dystonia have increased dramatically (Jankovic, 2006) and there are now a number of physical and medical approaches available to help control involuntary movements and relieve associated pain and discomfort. Physiotherapy can help to maintain range of movement, improve posture and prevent shortening or weakening of affected muscles. Occupational therapy support can provide specialist equipment to enhance mobility and prevent longer term deformity. Medical management involves drug treatments and botulinum toxin injections; however these do not relieve the motor symptoms in all patients (Difranceso, Halpern, Hurtig, Baltuch & Heuer, 2012), and pharmacological management is commonly ineffective in generalised and multifocal dystonia (Halbig et al., 2005; Pretto, Dalvi, Kang & Penn, 2008). Furthermore, medication is often accompanied by unwanted and adverse side effects (Lumsden et al., 2012). Therefore, Deep Brain Stimulation (DBS) Surgery has become the treatment of choice for otherwise intractable dystonia (Pretto et al., 2008), and childhood dystonia is now being routinely treated with DBS.

1.2.5 Deep Brain Stimulation

DBS is a reversible 'non-lesioning' neurosurgical treatment (Halbig et al., 2005), widely used to help improve motor control and functioning in Parkinson's disease, essential tremor and dystonia (Diamond & Jankovic, 2005). Surgery involves inserting very fine stimulating electrodes into the Globus Pallidus region of the brain. These electrodes are connected to a battery pacemaker, which is usually implanted in the chest or abdomen, and stimulates the targeted brain areas. There is now a significant body of evidence of the effectiveness of DBS in improving motor functioning, functional abilities and quality of life in an adult population (Halbig et al., 2005; Mehrkens et al., 2007). Increasing evidence suggests DBS is also successful in reducing childhood dystonia, demonstrating significant improves on impairment focussed measures, such as the Burke Fahn Marsden Disability Rating Scale (Gimeno et al., 2012; Haridas et al., 2011; Leland-Albright, 2003; Lumsden et al., 2012).

However, consistently in the quantitative literature effectiveness of DBS varies dependent on dystonia aetiology. Secondary dystonias appear to be less responsive to DBS compared with primary dystonia (Elthahawy, Saint-Cyr, Giladi, Lang & Lozana, 2004; Lumsden et al., 2012). Improvements in motor scores have been shown to be more subtle and not as durable in childhood secondary dystonia (Lumsden et al., 2012). It is likely this reflects the variability of symptoms, co-morbidities and fixed neurological deficits associated with a heterogeneous secondary dystonia population (Timmerman et al., 2010; Vidailhet et al., 2009). Furthermore, in adults and children with primary dystonia, a longer duration of dystonic symptoms is associated with poorer outcome (Andrews, Aviles-Olmos, Hariz & Foltynie, 2010; Lumsden et al., 2012). Lumsden et al. (2012) concluded that children should be offered surgery at a young age to minimise proportion of life lived with dystonia and maximise responsiveness.

DBS Surgery involves a number of diagnostic investigations and functional assessments to assess a child's suitability. It is necessary for the child's head to be shaven before the surgery. The child is given a general anaesthetic and the surgery typically lasts approximately six hours. Recovery typically involves an approximately ten day inpatient stay followed by returns to the hospital for follow up appointments at one, two, three, six, nine and twelve months post implant. During these appointments medical staff adjust the settings of the device to try and achieve optimal results. The outline of the battery can be slightly visible under the skin, and it is also necessary to charge the battery daily. DBS undoubtedly involves a substantial commitment from families. Given the unique features of DBS, and the subtle outcomes reported for secondary dystonia, an understanding of the decision-making process, and subjective challenges of the DBS experience would help inform clinician understanding and preparation of families.

Measurement Limitations

Research in dystonia, has predominantly relied on impairment measures and Health Related Quality of Life (HRQL; Lim, 2007). However, HRQL measures have not been designed or validated for dystonia. There is also a growing consensus that impairment based measures,

such as the Burke Fahn Marsden Dystonia Rating Scale (BFMDRS; Burke et al., 1985) used to measure post DBS change, are not sensitive enough to detect, small but significant changes in a secondary dystonia population (Gimeno et al., 2012; Jahanshahi & Marsden, 1990; Lindeboom, De Hann, Aramideh, Brans & Speelman, 1996). In a study of six paediatric cases of secondary dystonia the BFMDRS did not show a clinically significant change, however families reported significant benefit in terms of individualised goals, activity limitations and daily life participation (Gimeno et al., 2012). Meaningful surgery outcome may be more related to improvements in care, comfort and quality of life, than motor performance.

Beyond this, research has investigated the functional priorities of children with dystonia and parents using the Canadian Occupational Performance Measure (Gimeno, Gordon, Tustin & Lin, 2012). Pain was the most important factor interfering with activities of daily living and participation in activities. Access to assistive technology, self-care and social participation were also identified as a key concern across differing levels of motor abilities. This group is limited due to its small sample size of 57 children, and the differences between parent and child reports were not explored. A very recent study built on these limitations, to capture the concerns of parents of 273 children with dystonia (Lumsden, et al., 2015). Similarly, concerns raised were pain, difficulties performing activities of daily living, hand use and seating difficulties.

There is clearly a need to look beyond disability and impairment measures, because these measures are failing to capture the subjective meaning of post DBS improvements across wider domains, and are not capturing parents' functional priorities. A qualitative approach could therefore help to capture the complexity of change in a secondary dystonia population, and account for the subjective perceptions of subtle motor changes.

1.3 Defining Constructs

1.3.1 Disability

The International Classification of Functioning, Health and Disability (ICF) considers disability to be an umbrella term referring to the impairments, activity limitations and restrictions experienced by individuals (WHO, 2006). Thus, disability is a complex phenomenon and considers how individuals live in a 'context' made up of personal and environmental factors. This definition accounts for the interaction between person's body and features of society (WHO, 2001).

Dystonia is primarily conceptualised as a movement disorder. However, due to the characteristics and features of dystonia it could be viewed as a disability in line with the ICF definition, but also as a chronic illness because of its long standing and potentially progressive nature. Therefore for the purposes of this introduction, relevant empirical and theoretical literature will be discussed and appraised related to chronic illness or disability (CID).

1.3.2 Quality of Life

Quality of life (QoL) is an important construct given the profound impact on multiple areas of functioning of dystonia. In recent years, supporting the ICF classification, there has been a shift from an objective medical conceptualisation, to a broader subjective definition of QoL and well-being incorporating perceptions of physical, social and emotional well-being. Health-related Quality of Life (HRQL) is frequently used in quantitative health research. It considers how individual's perceptions of the functional aspects of disease and treatment impact on psychological and social well-being (Wallander & Koot, 2001). As already discussed it is well established that dystonia lowers QoL, and DBS has the potential to improve QoL in the adult population (Diamond & Jankovic, 2005). There is a lack of research examining the wider

social, emotional and psychological consequences of childhood dystonia on QoL, and the processes and meaning underlying QoL changes following DBS have yet to be explored.

1.4 Models of Disability and Chronic illness

1.4.1 Medical vs. Social Model of Disability

The Medical Model conceptualises disability in the context of pathology, impairment and disease processes located within the individual. However, it has increasingly been argued this fails to fully acknowledge the role of systemic factors leading to the development of various social models considering disability as a social construct (Hughes & Patterson, 1997; Kelly, 2005; Thomas, 2002). Disability is viewed as created by society through negative attitudes, social and societal barriers, stigma and intuitional barriers and is a result of social construction. Much of the literature exploring childhood disability and its effects on the family are presented in the context of normality and impairment measures have dominated quantitative research. Professionals define objective physical, developmental and social criteria to measure difference from normality (Mayall, 1996). However, increasingly researchers have moved beyond an individualistic view of disability, to consider the narratives of parents and how they develop their own understanding of their child's disability (Landsman, 2005). Studies have demonstrated that mothers focus on the lived experience of disability and the personhood of their child (Green, 2003a, 2003b; Jenks, 2005; Kelly, 2005; Landsman, 1998). It therefore seems important to explore how parents understand and make sense of their child's dystonia, and the wider social impact and context of dystonia experienced by families.

1.4.2 Models of Coping, Adaptation and Adjustment to CID

Living with CID is often challenging for the individual and wider family. Life can take on a different meaning, and require significant adjustments psychologically and socially within the family.

1.4.2.1 Cognitive Coping Theory

Lazarus and Folkman's Seminal Cognitive Coping Theory (1984) has been influential in considering how individuals cope with and manage the stress resulting from CID. Stress is conceptualised as an interaction of personal and environmental characteristics. The environmental stimulus could be acute in the form of surgery or hospitalisation, or chronic in the case of living with a CID. Individuals appraise a potential stressful event through primary appraisals of environmental demands (threat, significance), and secondary appraisals of perceived resources to cope through controlling or changing the situation. Coping involves changing cognitive and behavioural efforts to reduce stressors, regulate emotions and gain control of one's immediate environment. (Lazarus & Folkman, 1984).

1.4.2.2 Systemic Models of Adjustment to Chronic Illness

The importance of parent and family factors has been highlighted in various conceptual models of how children adjust to chronic illness (Thompson & Gustafson 1999; Wallander & Varni, 1998). The Disability-Stress Coping Model (Wallander & Varni, 1992) proposed how condition parameters, social ecological factors, environment psychosocial stress, stress processing factors and interpersonal factors impact on psychological adjustment. Stress arises from a physical condition and functional limitations, within the context of general stress arising in life. The parameters of the CID condition (e.g. severity, visibility), functional dependence and interpersonal factors (e.g. temperament) are all considered to impact on adjustment. However, adjustment is also impacted by the social ecological context (e.g. family functioning, social support, parental adjustment).

Resilience has been viewed as the ability to withstand and rebound from crisis and adversity (De Castro & Moreno-Jiminez, 2007). Given the chronic, and often progressive nature of secondary dystonia, and the long term impact of having DBS implant, parents will encounter different challenges, requiring different coping strategies and responses over time (Rutter, 1987). Risk factors have been highlighted for an individual's ability to adapt to living with a

disability (Wallander & Varni, 1998). The factors relevant to an individual with dystonia include visibility of the disorder, unpredictability of outcome, presence of central nervous system difficulties, pain and impaired functional independence. These models and understanding move away from a problem focussed medical model of CID, to focus on a wider systemic framework, emphasising the psychological processes and patterns of resilience in families. Dystonia research needs to follow the CID literature to take a family-systems approach, and privilege family and contextual factors to understand parents' experiences, rather than simply relying on parents as a means of assessing the child's motor impairment and functioning.

1.5 Parenting a Child with CID

Dystonia affects the child and the family system, presenting parents with a unique set of challenges. However, no studies have sought to explore the experiences and adjustment of parents in their own right. Therefore the wealth of quantitative and qualitative literature capturing the experiences and impact of parenting a child with CID will be considered.

1.5.1 Family Functioning, Adaptation and Resilience

Families respond in different ways to the challenges of parenting a child with CID. It has been suggested that parents caring for a child with a chronic condition or disability assume four major caregiving responsibilities: managing the illness; identifying, accessing and co-ordinating resources; maintaining the family unit; and maintaining self (Sullivan-Bolyai, Sadler, Knafel, Gilliss & Ahmann, 2003). Additionally DeCastro and Piccinini (2002) found that changes to family relationships occur in the context of a chronic physical disorder.

There is growing evidence in chronic health that after an initial period of adjustment, parents focus on minimising disruption and creating a 'normal' life (Alexander, Renwick, Carnevale & Davis, 2012; Bedell, Cohn & Dumas, 2005, Glasscoe & Smith 2011). A number of studies have found that maternal or family adaptation to childhood disability is associated with better family functioning and parental psychological variables (e.g. optimism, internal locus of control

and problem solving strategies; Baker, Seltzer & Greenberg, 2011; Bourke-Taylor, Pallent, Law & Howie, 2012; Ekas, Lickenbrock & Whitman, 2010). Social support has long been identified as a stress buffering resource and studies have shown that parents with more social support show better adjustment to child illness (Plant & Saunders, 2007)

1.5.2 Impact on Parents

It has been well documented that caring for children with CID impacts on parent's physical, psychological and emotional well-being. Areas impacted include role ambiguity, caregiver burden, loss of privacy, marital relationship problems and poorer mental health and family functioning (Green 2007; Hewitt-Taylor, 2005). An area that has received a wealth of interest is parenting a child with CP. Mothers of children with CP have been found to experience increased parental stress, anxious and depressive symptoms and decreased psychological well-being (Barlow, Cullen-Powell, & Chesire, 2007; Cheshire, Barlow & Powell, 2010; Pousada et al., 2013; Rentinck, Ketelaar, Jongmans & Gorter, 2007).

1.5.3 Construct of Carer Burden

Within society, diagnosis of or living with disability is widely accepted as a negative experience. Subsequently the literature has predominantly focussed on the negative aspects of parenting a child with disability, and the construct of carer burden has been emphasised. Less attention has been paid to positive aspects of having a child with a disability. Green (2007) in a mixed methods approach demonstrated that burden of care was the result of socio-cultural constraints and not emotional distress. Furthermore, most mothers voiced benefits of having a child with a disability. Stigma was also found to contribute to carer stress in this study. Historically the literature has ignored the burden imposed by negative societal attitudes and inadequate support (Barnett, Clements, Kaplan-Estrin & Fialka, 2003; Leiter, Krauss, Anderson & Wells 2004; McKeever & Miller, 2004).

1.5.4 Qualitative Literature – Caring for a Child with Cerebral Palsy

The most common type of childhood dystonia is secondary dystonia as a result of CP. Unlike the dystonia population there is a wealth of qualitative studies considering the impact of caring for a child with CP on parents.

The QoL of parents and impact of caring for a child with CP have been explored qualitatively (Davis et al., 2010). Using a grounded theory framework, 37 parents of children and adolescents aged 3-18 years were interviewed. A number of parents reported that they had a good QoL, which they attributed to their personality, attitude and level of support received. Parents who felt their QoL was poor described a lack of social support and their time dominated by caregiving. Although caring had a negative impact on many different areas of parents' lives, positive impacts were reported. These included building new social support networks, drawing inspiration from the courage and resilience of their children, and their pleasure when children met goals or milestones.

A more recent qualitative study focussed exclusively on parents' experiences of caring for adolescents with severe CP (Burkhard, 2013). Utilising a phenomenological approach, eleven mothers were interviewed to capture their lived experiences. The main theme identified was 'a different life'. Parents showed acceptance of their difficult role and complete commitment to their child. Consistent with other studies, all mothers reported exhaustion following the physical and emotional demands of caregiving. Mothers described an individualised process of learning to balance multiple demands and trying to normalise and optimise family life. This supports similar findings by Bourke-Taylor, Howie and Law (2010) and consistent with Davis et al. (2010), mothers reported a positive impact on their maternal-efficacy and sensitivity to children. These conclusions were strengthened by theme validation, and homogeneity of sample selection.

However, the co-existence of features of spasticity, rigidity and dystonia in CP makes classification difficult (Sinkjaer, Toft, Larsen, Andreassen & Hansen, 1993; Wright,

Rosenbaum, Goldsmith, Law & Fehlings 2008). Applying existing findings from the CP literature is challenging because most studies predominantly involve cohorts of children with spastic presentations, and typically few or no dystonic children (Kuijper, van der Wilden, Ketelaar & Gorter, 2010). In the literature, prevalence of dystonic CP varies between 6.5-14.4% (Himmelman et al., 2009). It is therefore likely that the majority of children with CP in the literature do not meet criteria for dystonia unless clearly stated. Dystonia is a constantly changing state of muscle tone, and is likely to affect the manual and functional ability of children differently to other types of CP. Therefore, given the unique nature and heterogeneity of children with secondary dystonia, who have also undergone DBS surgery, further research is necessary to elucidate the lived experiences of caring for a child with secondary dystonia.

1.6 Surgery Decision-Making Literature

There is limited literature on decision-making for paediatric surgical procedures. Research has focussed on decision-making processes for non-elective and life threatening conditions (Daniel, Kent, Binney & Pagdin, 2005). In these studies respect for child's views, parental anxiety and expected QoL have been highlighted as important considerations in decision-making. However, in the context of elective procedures, such as DBS, because of absence of threat to life, social and psychological factors may be more important (Daniel et al., 2005). Children are often able to participate in decisions, and time pressure is less important. There are a growing number of quantitative and qualitative studies exploring decision-making for elective surgery.

In a recent review of studies, Dewar and Pieters (2015) explored individuals' decision to undergo elective neurosurgery for epilepsy. Decision-making was influenced by cognitive and emotional processes (Bonatti et al., 2009), as individuals weighed up treatment risks and benefits, in face of uncertain surgical outcomes. Fear of neurosurgery often delayed decision-making, and patients commonly held unrealistic expectations of surgical outcomes.

Most studies of parental decision-making have focussed on creating a descriptive model of how decisions are made. A grounded theory approach has been used to explore how parents undertake decision-making in normalising surgery for children with cleft lip palate (Nelson, Caress, Glenny & Kirk, 2012). Parent's main concern was their parental duty to do the right thing, and surgery was a way of facilitating child's social inclusion and helping them to reach full potential. Healthcare practitioner power and competency was also a feature shaping decision-making.

IPA has also been used previously for understanding decision-making in elective procedures. An IPA approach provides insight into how parents make sense of and experience decision-making. This is important because the concept of decision-making appears to be influenced by different psychological processes, and is highly context bound (Wirtz, Cribb & Barber, 2006). The decision to undertake DBS for families with secondary dystonia comprises a combination of unique factors: a lack of outcome certainty, a long term commitment to regular hospital follow up appointments and daily commitment to battery charging. It is undoubtedly a decision with significant consequences for the children and wider family. An understanding of the decision-making process, and factors that were important to families, would help clinicians better prepare and support families, and also potentially reduce decision-making times. This is important because shorter dystonia duration and younger age at surgery have been associated with better outcomes after DBS (Lumsden et al., 2012).

1.7 Hospital Experience and Psychological Impact of Surgery Literature

Surgery and hospitalisation are thought to be some of the most difficult experiences for children and parents. Two qualitative studies exploring the experiences of adults and children, who have undergone DBS for primary dystonia (Bakowski, 2010; Hariz, Limousin, Tisch, Jahanshahi & Fjellman-Wiklund, 2011) found DBS to be a significant life event, impacting the whole family and requiring practical, social and psychological adjustment. So far however, quantitative and qualitative research has neglected the psychological needs and experiences of parents, and children with secondary dystonia. The broader literature on surgery anxiety

and coping will be reviewed, studies exploring parental experiences of surgery for children with CID will then be considered, and finally how the qualitative DBS literature informs understanding of the experience of DBS will be extrapolated.

1.7.1 Parental Surgery Anxiety and Coping

A review of the literature demonstrates that parents experience intense stress, and feelings of helplessness when their child undergo surgery, and high levels of preoperative anxiety (Brennan, 1994; Cohen, Blount & Panapoulos, 1997; Fielding & Tam, 1990; Fielding, 1994; Kain, Mayes, O'Connor & Cicchetti, 1996).

In a study of 100 parents of children scheduled for elective surgery, parents were anxious about the surgery, anaesthesia, postoperative pain and being in hospital (Shirley, Thompson, Kenward, & Johnson, 1998). There is increasing evidence that anaesthesia is one of the most anxiety provoking factors for parents (Frank & Spencer 2005; Cagiran et al., 2014). It is clearly important to explore parent's experience of DBS surgery. These studies highlight the importance of taking a whole family approach to childhood surgery, and justify the necessity of considering the psychological needs and experiences of parents in their own right. Given the unique features of DBS surgery, a qualitative approach could help to increase understanding of the salient moments of surgery, and what factors contribute to parental anxiety.

Evidence suggests parental anxiety predicts child preoperative anxiety (Ahmed, Farrell, Parrish & Karla, 2011; Cagiran et al., 2014; Crawford, 2014; Davidson et al., 2006) In support of Lazarus and Folkman's (1984) cognitive coping model, the anxiety experienced by parents is related to the coping style adopted. There is a significant body of literature that suggests that coping strategies play an important role in how individuals respond to stressful situations and negative life events (Endler & Parker, 1990; Lazarus, 1993; Lazarus & Folkman, 1984; McCrae & Costa, 1986). Furthermore, parent's emotion focused coping styles have also been shown to predict higher levels of children's preoperative anxiety (Blount, Landolf-

Fritsche, Powers & Sturges, 1991; Crawford, 2014). Crawford (2014) has argued that how the parent-child dyad appraises stress during hospitalisation determines how well they will cope. An understanding of the contextual nature of parental stress during DBS surgery and hospitalisation is crucial. A qualitative approach could attend to DBS context and help gain a richer understanding of the cognitions, coping and emotions of parents during this stressful experience.

1.7.2 Parents' Experience of Surgery and Hospitalisation for Children with CID

Parents of children with CID experience unique challenges and stressors. It is likely that the experiences of these parents during surgery and hospitalisation are subjectively different and their experiences have been explored qualitatively.

The lived experiences of parents' of children with CP undergoing orthopaedic surgery have been explored (Iversen, Graue & Råheim, 2013). A hermeneutic phenomenological approach was used to analyse the semi-structured interviews of 12 parents of 9 children aged 8-16 years with CP of differing severities. The overriding theme arising from parents accounts was a feeling of vulnerability and helplessness. Parents experienced demanding challenges, in an unfamiliar situation. Parents experienced a loss of control, and felt continuously responsible. This was made worse by parents feeling tormented by the legitimacy of their decision to let the child have surgery. Exhaustion and fear dominated accounts, as parents attempted to be constantly available for the child whilst also being strong for the child's sake. In this study, rigour was established through agreement between researchers, and feedback from participants on preliminary themes.

The themes from Iversen et al. (2013) support conclusions from earlier described quantitative studies that parents often feel anxious, fearful and helpless when their child is hospitalised. These feelings are also apparent in other studies of children with disabilities during hospitalisation (Haines, 2005; Hopia, Paavilainen & Åstedt-Kurki, 2005). A relatively consistent picture also emerges from an investigation of parental pre and post-operative

stressors in spinal fusion surgery for adolescent scoliosis (Salisbury, LeMontagne, Hepworth & Cohen, 2007). The primary stressor pre and post-operatively was parental role loss. The possibility of poor surgical outcomes and uncertainty of recovery were also significant concerns pre-operatively, and concerns about pain postoperatively. However the conclusions were only from a sample of 92 parents, and are potentially limited in generalisability to wider CID populations.

The value of qualitative research in thoroughly exploring and capturing the views and experiences of parents during hospitalisation has been demonstrated. Although these studies are informative in providing an overview of the area, the experiences of parents of children with secondary dystonia are likely to be qualitatively different because of the uniqueness of the DBS procedure, particularly the meaning of brain surgery, lack of outcome certainty in secondary dystonia and the long term commitment to DBS appointments and charging the system. Research is therefore needed into parental experiences of DBS, and this is something that will be explored in this study.

1.7.3 Qualitative Studies exploring Experience of DBS and Living with Dystonia

Although parents' narratives have not been captured, qualitative studies have explored the DBS experience of adults and children with dystonia.

1.7.3.1 Adult Primary Dystonia

To date there has only been one qualitative study exploring adult perceptions of DBS surgery, and of living with, and being dependent on a technical device (Hariz et al., 2011). Thirteen adults with primary dystonia were interviewed using a grounded theory methodology.

Dystonia was shown to have a profound impact on almost all aspects of day-to-day life, and people described struggling with a disabling and disfiguring disorder. The decision to undergo DBS was viewed as the patient's own decision, and expectations were managed by staff.

DBS had a 'life changing effect' for individuals. DBS improved motor functioning, increased confidence and revealed participant's true potential. A demanding transition process from a limited life pre-operatively, to new life with possibilities and challenges was highlighted. A key theme identified was coping with new challenges post surgery. This involved experiencing negative and interfering side effects, adapting to a new body and concerns of the visibility of, and dependence on, an implanted battery.

Participants were selected using purposive sampling to represent different chronological age, age of disease onset, symptom severity and type of dystonia. This reflects the clinical heterogeneity of primary dystonia, and allows conclusions to be generalisable to the adult primary dystonia population. Theoretical saturation, independent coding and triangulation enhanced the credibility of conclusions.

1.7.3.2 Childhood Dystonia

The qualitative experiences of young people living with dystonia and the impact of DBS have also been explored (Bakowski, 2010). Seven young people aged between 13 and 19, with primary and secondary dystonia were interviewed and analysed using IPA.

Interpretation of master themes suggests living with dystonia significantly affects QoL, and tests the strength and resilience of young people and their families. Young people experienced anxiety about the future, sadness and frustration at a life missed. Dystonia is a unique and isolating experience, and brings up multiple feelings of difference for young people. All participants relayed feelings of fear of continuing to live with dystonia, of an unknown future, or of dying during neurosurgery. A process of adjusting and adapting following DBS was demonstrated in young people's accounts. All but one participant found that DBS exceeded their hopes for change. In agreement with current literature, the two participants with secondary dystonia experienced more modest post surgery change. These changes led to a renegotiation of life after surgery, and regaining of independence as a young person. Alongside positive change, young people also described living with a compromise of

continued limitations imposed by the DBS implant, and of movements not fully returned back to normal.

This study was limited because, contrary to the theoretical underpinnings of IPA, a heterogeneous group of children with primary and secondary dystonia were included. Most of the participants had primary dystonia, and only two children had secondary dystonia. It is likely the experiences of children with secondary dystonia are different, particularly given the reduced impact of DBS on secondary dystonia (Gimeno et al., 2012). Furthermore, by only including children who were able to communicate verbally, the study is only capturing the experiences and perceptions of a subset of the secondary dystonia population, and neglecting those with severe and additional difficulties.

1.7.3.3 Conclusions from the Qualitative Dystonia and DBS Literature

Key themes and shared conclusions are identifiable across the adult and child literature. Adults and young people described the long and difficult journey of living with dystonia, and the profound impact of dystonia on daily living and QoL. Dystonia also had a profound impact on the QoL of family members. A number of adjustments and coping strategies were required to cope with the multiple stressors and demands of living with or caring for an individual with dystonia. A demanding transition process to life after surgery was experienced by adults and children, with the need to adjust and cope with new physical and psychosocial challenges post-surgery. Family members also had a key role in supporting adjustment to post-surgery change and challenges.

In summary, conclusions from the qualitative CP literature of parental experiences, combined with the knowledge gained from adult and child dystonia studies is consistent with the profound impact of dystonia on families, and the important role parents play throughout a child's day-to-day life, and in times of adversity such as surgery. As it stands, no studies have explored parents' experiences of either caring for a child with dystonia, or of going through the DBS surgery process. Additionally no studies have focussed solely on the unique

experiences of children with secondary dystonia, who are potentially at increased risk of psychological distress due to the ongoing nature of difficulties.

1.8 Study Rationale

The literature described previously has identified many gaps and highlighted the need for further research in light of several important findings.

Firstly, the wider CID literature and qualitative dystonia studies demonstrated that parents experience multiple challenges caring for a child with a disability. However parental experiences and the impact of dystonia on parents have largely been neglected.

Secondly, quantitative and qualitative research has focussed predominantly on the experiences of individuals with primary dystonia. It is likely the experiences of children with secondary dystonia are different: the majority experience dystonia from birth, dystonia occurs secondary to another condition (e.g. CP) and DBS has a reduced impact. Children with secondary dystonia often have severe disabilities and communication impairments. Interviewing parents will allow the experiences of children with severe secondary dystonia to be partially captured. Research has been reliant on impairment and disability measures, and neglected the personal and subjective experiences of living with and parenting a child with secondary dystonia.

Thirdly, surgery and hospitalisation has been shown to be a stressful and anxiety provoking experience for parents. DBS is increasingly used in childhood dystonia, and the efficacy of DBS for secondary dystonia is currently the focus of much interest. It would be useful to understand what stages of the DBS process were challenging for parents, and how they managed the process.

Fourthly, DBS decision-making has never been a focus of research. A greater understanding of the relevant factors and processes, and psychological implications of making this decision

could help staff better support parents in this decision and potentially reduce decision-making times. Additionally, understanding the psychological, social and practical implications of adjusting to and being dependent on a technical device would help clinicians to appropriately prepare families.

Finally, quantitative measures are not sensitive enough to capture the subtleties of post DBS change, and measures are not capturing the functional priorities and concerns of parents. A greater understanding of the subjective meaning of change, and the impact of DBS in secondary dystonia, would aid informed decision-making.

1.9 Adopting a Qualitative Approach

Given the absence of research focusing on secondary dystonia and parents' experiences a qualitative approach with exploratory aims was chosen. It was important not to constrain or impose restrictions on the data through hypothesis testing quantitative approaches, to allow subjective meanings and salient idiographic experiences to emerge. A qualitative approach was also deemed appropriate to move beyond an impairment focus, to gain a wider perspective of the experiences and perspectives of parents, within a broader social, psychological and emotional framework. Furthermore, a qualitative approach privileges an insider perspective and views of participants are given primacy over researchers and healthcare professionals. This was important because impairment measures chosen by researchers and clinicians have been shown not be capturing the functional priorities and concerns of parents.

Given the wealth of studies demonstrating disability as a complex social phenomenon, to answer questions about parental experiences, the approach needs to be able to examine the complexity at multiple levels of meaning (Fiese & Bickham, 1998) and consider how experience is continuously impacted by social, cultural and interpersonal context. Phenomenological inquiry focuses on individual experience and how they perceive a phenomenon. The phenomena being explored are parenting a child with secondary dystonia

DBS. IPA (Smith, Flowers & Larkin, 2009) is grounded in phenomenological epistemology and therefore suitable for exploring the lived experiences of parents and how they make sense of these experiences (Holloway & Todres, 2003). The rationale for using IPA is extensively discussed in the methodology chapter.

1.10 Summary and Research Aims

This study aimed to increase understanding of the experiences of parents and children with secondary dystonia to enable services to meet the specific needs of this unique population. A greater insight into DBS decision-making and the experience of DBS was hoped for, to increase clinician awareness, to allow parents to be fully informed of likely DBS outcomes, to help families to develop realistic expectations of post surgery change and to help clinicians prepare families for the challenges of the DBS experience. A greater understanding of the decision-making process could help reduce decision-making times and improve clinical outcomes of the surgery (Lumsden et al., 2012). Furthermore, this understanding could inform the development of psychoeducational resources and any necessary clinical interventions.

This study will attempt to answer the following research questions:

1. What are the lived experiences of parents and children with secondary dystonia?
2. What influences DBS decision-making, and how do parents make sense of their decision?
3. How do parents experience and manage the DBS process?
4. What is the impact and meaning of post-surgery change for parents and children with secondary dystonia?

Chapter 2: Methods

This chapter will begin by outlining the study design, the researcher's epistemological position, the theoretical underpinnings of IPA, and why IPA was chosen as the qualitative approach. This will be followed by a description of the methods that were used to operationalise the research aims and ensure the research was rigorous, epistemologically valid and ethical. This will include sampling and recruitment, data collection, procedural and ethical considerations, and service-user involvement. Finally a description will be provided of data analysis, and efforts to ensure the validity and quality of this study will be outlined.

2.1 Research Design

A cross-sectional qualitative design was used in this research. Semi-structured interviews were completed with eight parents of children with secondary dystonia to retrospectively explore their experiences of living with dystonia and DBS.

2.2 Epistemological Position

Epistemology is concerned with the acquisition of knowledge, and considers how and what it is possible to know (Willig, 2008). This research assumes a hermeneutic phenomenological epistemology to exploring a person's lived experience from the perspective of participants themselves and through the meanings they make (Larkin & Thompson, 2012). This approach is therefore not concerned with existence of an external reality and is aligned with a critical realist assumption that there is correspondence between a person's account and their subjective reality. Phenomena are considered to exist and be examinable, but researchers can never be in direct contact with experience, and understandings are partial (Nightingale & Cromby, 1999).

Participants engage in sense-making in a linguistic, relational and cultural world, and researchers are attempting to enter the life world of participants. Researchers therefore engage in 'intersubjective meaning-making' to reflect on their own experiences and assumptions (Larkin & Thompson, 2012), and to 'go beyond the text' to interpret participant accounts in a broader social, cultural and theoretical context to make it more meaningful (Larkin, Watts & Clifton, 2006). Drawing on social constructionist ideas, language is not assumed to represent whole truths, but is influenced by the cultural and historical context, and researchers must situate experience in a social and cultural context. IPA is an experiential and phenomenological approach, accepting that experiences are meaning-rich and language used to describe them reflects experiences and reveals something about that meaning (Larkin et al., 2006; Lyons & Coye, 2007).

Similar to many IPA studies, this research adopts a stance aligned with critical realism, within a hermeneutic phenomenological position. IPA was chosen as the methodology which aligns with this epistemological position, and the exploratory aims of the study.

2.3 Interpretative Phenomenological Analysis – Theoretical Underpinnings

IPA is a qualitative approach used to explore individuals' lived experience and how they make sense of their major life experiences (Smith et al., 2009). IPA has broad theoretical underpinnings and has been informed by phenomenology, hermeneutics and idiography (Smith et al., 2009). These will be briefly discussed, because the philosophical underpinnings and epistemological position determined the researcher's approach to the study design, data collection and analysis.

2.3.1 Phenomenology

Phenomenology is the philosophical study of being and experience. IPA is grounded in the core ideas from a number of phenomenological philosophers: Husserl, Heidegger, Merleau-Ponty, and Sartre (Smith et al., 2009). Husserl originally considered the importance of

capturing everyday lived experiences. Husserl proposed the 'bracketing off' of past knowledge and assumptions to take a reflexive move towards seeing the essence of a phenomenon (Smith et al., 2009). Heidegger (1927, 1962 as cited in Smith et al., 2009) moved towards an interpretative position, arguing that individuals' experiences and perceptions are dependent on the context of their lived world, and one cannot separate from one's own position to access another's world completely. Therefore in IPA, interpretations are used to try and access participants' lived experiences and a researcher must reflect on their preconceptions and assumptions. Merleau-Ponty also considered the body as central to experience, and distinguished between the subjective body as it is lived and the objective body that is observable and known to others (Merleau-Ponty, 1962). IPA researchers therefore adopt a view that unique meanings are formed by an individual's relationship with a relational world.

2.3.2 Hermeneutics

Hermeneutics is the theory of interpretation, and underpins the interpretative role in IPA. The work of Heidegger, Schleiermacher and Gadamer are considered to have influenced IPA's emphasis on contextual meanings (Smith et al., 2009). Schleiermacher proposed that interpretation involves making sense of grammatical and psychological elements, and unique intentions and meaning must be considered in the wider context. In IPA, the researcher therefore offers insights which go beyond the claims of participants (Smith et al., 2009). Ricoeur (1970, as cited in Smith et al., 2009) proposed the hermeneutics of empathy and suspicion, and subsequently IPA researchers are trying to gain an 'insider's perspective' of what an experience is like, whilst assuming a curious and critical position (Smith et al., 2009). IPA involves a double hermeneutic as the researcher attempts to make sense of the participants' sense-making (Smith et al., 2009). Heidegger argued that the interpretative process is influenced by the researcher's experiences, assumptions and preconceptions. However, these 'foreunderstandings' may not arise until the researcher has started to engage with the participant accounts. Therefore, reflexivity and openness need to be continuing processes throughout an IPA analysis (Smith et al., 2009). An IPA analysis is also a dynamic

and iterative process, and involves moving between the part and the whole to embody the hermeneutic circle. This has been argued to create richer perspectives, and encourages the author to consider multiple and new interpretations.

2.3.3 Idiography

Idiography means being concerned with the particular and, in contrast to the nomothetic approach, is concerned with making claims regarding meaning at an individual level. In IPA an idiographic stance is embodied by a focus on the detail through systematic engagement with data and understanding the subjective meanings of particular people in particular contexts in a case-by-case analysis, before making general statements (Smith et al., 2009). IPA also involves small purposively selected samples, to privilege idiographic experiences of shared phenomenon.

2.4 Rationale for choosing IPA

IPA was chosen as the most appropriate approach for several reasons given the exploratory aims of this study:

This study hoped to explore the lived experiences of parents and children with secondary dystonia, and how they make sense of a major life experience (DBS Surgery). IPA allows for an idiographic exploration of parental experiences, but also a relational focus as the researcher is attempting to understand the sense parents make of their child's experience of dystonia. This approach acknowledges the social context of disability, and privileges the relational nature of secondary dystonia. Given the unique presentation of each child and heterogeneity of secondary dystonia, interpretation was deemed essential to make sense of these experiences and to situate those experiences within the complex social, cultural and professional systems in which children with secondary dystonia exist.

IPA has been widely used in health psychology, and within this field studies have successfully used IPA to explore parental decision-making, and experiences of caring for a child with chronic illness (Daniel et al., 2005; Glasscoe & Smith 2011). IPA was therefore appropriate to consider the meaning and experience of having a child with secondary dystonia and how parents make sense of their decision to undergo DBS.

Unlike quantitative measures, IPA allows insight into the process of decision-making and experience of hospitalisation. Health research has also focussed on a broader social understanding of illness, and considered the constructed nature of illness. IPA therefore allows exploration of the child's perceptions and experiences of dystonic movements, and the meaning assigned to them (Brocki & Wearden, 2006).

From a phenomenological perspective, IPA can also help research move beyond an objective and observable conception of dystonia, to consider subjective and lived experiences. This was particularly important because quantitative measures have failed to capture the subtleties of post DBS change reported by parents, suggesting that perceptions of surgery success was unique to families and perhaps based on the meaning they assign to these subtle motor improvements. IPA may therefore help to elucidate the psychological processes underlying this disparity between quantitative measures and parents self-report, and understand the variability of DBS secondary dystonia outcomes. (Brocki & Wearden, 2006). Parents are knowledgeable of, and experts in, the unique features of their child's dystonia. IPA was chosen to privilege this expert position, and fully attend to the voice of participants (Larkin et al., 2006).

IPA is also appropriate for under-researched areas, and able to capture the perspective of untold narratives of parents and children with secondary dystonia. By not holding pre-conceived hypotheses, participants' voices are unconstrained which allows new and often unanticipated features of a phenomenon to be revealed (Shaw, 2001). Importantly for a novice qualitative researcher, IPA offered clear guidelines, training and support groups to help ensure production of a high quality piece of research.

2.5 Excluding other Qualitative Approaches

The first stage of designing the study involved developing exploratory research aims and considering which qualitative approach would be most appropriate. This involved considering other methods before choosing IPA.

Thematic Analysis (Braun & Clarke, 2006) has tended to use larger samples and takes a nomothetic stance focussed on generalisability. The analysis is also focussed at a more descriptive level with less significance placed on researcher reflexivity. It was decided this approach would not produce a sufficiently deep or idiographic understanding of participant experience, and would not sufficiently attend to the role of social and cultural context through interpretations.

Discourse Analysis (Potter & Wetherell, 1995) focuses on the function of language, and how reality is constructed through language, rather than reflects it. This approach does not fit with the researcher's critical realist epistemological position, and although IPA acknowledges the role of language in understanding how participants make sense of their experience, this approach would not attend to the experiential and sense-making focus of the research aims.

Grounded Theory (Glaser & Strauss, 1967) is an inherently sociological method used to describe and generate theories of social processes. This approach adopts a positivist epistemology and uses theoretical sampling which ceases data collection when data saturation is achieved and new ideas stop emerging. IPA privileges idiography, and given the heterogeneity of secondary dystonia and variable impact of DBS it was viewed that participants could assign different meanings to the same phenomena (e.g. living with secondary dystonia, post surgery outcomes), and therefore required an approach that focussed on how participants make sense of particular phenomena. Grounded theory could have been used to form a model decision-making, but this study had a broader more exploratory focus on capturing previously unheard experiences of secondary dystonia rather than on developing a model of social process (Harper, 2012).

2.6 Sample Size

Eight parents of children with secondary dystonia were recruited from a specialist NHS tertiary children's hospital. A number of factors were considered when selecting a sample size. This sample size was chosen to be consistent with the principles of IPA: Recruitment of small samples is recommended in IPA to allow a greater depth of analysis to elucidate meaning and fully consider the context of individuals' experiences (Smith et al., 2009). A sample size of seven was used in an IPA study exploring children's experiences of dystonia (Bakowski, 2010). For Doctoral research, Smith et al., (2009) propose a sample of four to ten participants, and some have proposed smaller sample sizes (Reid, Flowers & Larkin, 2005; Smith, 2004). It was hoped that an analysis of eight accounts would achieve coherence and integration of the lived experiences of a group of parents, whilst also sufficiently preserving nuances and privileging the unique perceptions and meanings (Elliot, Fischer & Rennie, 1999). A further consideration was availability of participants. Due to the specialist nature of the target population and those eligible to undergo DBS, there was a limited number of potential participants. A sample of eight was deemed feasible within the time constraints for recruitment and analysis within a Clinical Psychology Doctorate.

2.7 Inclusion and Exclusion Criteria

In IPA it is important to obtain a 'fairly homogenous sample' (Smith et al, 2009). Homogeneity refers to the selection of participants who are able to provide a unique idiographic perspective of a particular phenomenon within a particular context. In this research, a homogenous sample was needed to gain insight into the lived experience of DBS, from the perspective of a parent of a child with secondary dystonia. To be able to examine the convergence and divergence within this shared experience (Smith et al, 2009), it was necessary to consider how participants may vary from each other, and what variation could be controlled for. A number of inclusion criteria were therefore agreed upon prior to commencing recruitment.

Parents of children who were patients at a tertiary children's hospital were eligible to participate if they met the following inclusion/exclusion criteria:

- *Their child had DBS surgery and was less than 17 years old at the time of surgery.*
- *Their child had a diagnosis of secondary static dystonia.* Children with secondary progressive dystonia deteriorate over time, and parents' experiences were anticipated to be significantly different given the progressive nature of the condition.
- *Their child had secondary dystonia that developed during infancy (birth to 2 years).* A later-onset means parents would have experienced parenting and family-life without the presence of dystonic movements. Their experiences and perspectives of post-surgery change were anticipated to be significantly different
- *They were the main carer (self-selected).* The main carer was likely to spend the most time with the child, be involved in physical care, and be aware of the subtleties of motor changes post DBS. During consultation, service-users suggested it was important to allow the family to choose who participates in the research and strongly felt fathers should not be excluded.
- *They were able to comprehend and speak English fluently.* Participants' perceptions, understandings and meaning are constructed through language, and therefore they needed to speak English fluently to avoid biases in interpretation. It was pre-established that during the recruitment period all potential participants were English speakers, and no one would have to be excluded.
- *The surgery took place between 12-24 months prior to the interview.* Ideally interviews take place as close as possible to the phenomenon of interest. However, it can take over a year to fine tune the electrodes to create the optimal settings. A recent study found the time to maximal motor improvement was 1.7 years in adults

with dystonic CP (Romito et al., 2014). In this research, it was important that participants were able to reflect on their experiences of surgery and have a greater understanding and perspective on the effects and impact of DBS. An upper limit of 24 months was set because service-users reported that as time passes it becomes “harder to remember life before the surgery” and recall the subtleties of change. It was hoped this upper limit would reduce bias and ensure participants could richly recall experiences.

Strict inclusion and exclusion criteria were required to create a fairly homogenous sample. However, the specificity of secondary dystonia and rarity of DBS surgery defined the boundaries of the population and created some variability within the sample. Following service-user consultation it was also thought important to capture positive and negative experiences, and so a decision was made not to exclude based on surgery complications. This sample was therefore reflective of the children who undergo DBS at the tertiary children’s hospital.

2.8 Sampling and Recruitment

Purposive sampling was employed to select a homogenous sample based on pre-determined characteristics, which could provide the researcher with access to a particular perspective of a phenomenon (Smith et al., 2009): Namely DBS and secondary dystonia. This non-probability sampling method was chosen to be consistent with the theoretical orientation of the IPA philosophy (Chapman & Smith, 2002). Families were invited to take part based on a consecutive sampling strategy. All families who were being followed up by the service and were eligible for participation based on the inclusion/exclusion criteria, were invited to take part in sequential order, based on the date of their follow-up appointment. To improve homogeneity only those attending their one year review were considered at the start of the recruitment phase.

Participants were recruited from a paediatric neurology department at a tertiary children's hospital. Recruitment took place over a six month period between July 2014 and January 2015. Recruitment ceased once all eight participants had been interviewed. Potential participants were identified by the external supervisor, in her capacity as Clinical Psychologist within the team. The external supervisor screened the neurology patient database for families who met inclusion/exclusion criteria. Parents were then approached by the external supervisor approximately four weeks before their follow-up appointment and were provided with an information sheet (Appendix 1) via post or email. If parents were interested in taking part and consented to be contacted, the researcher then contacted the families at least 24 hours after the receipt of the information sheet. The researcher explained what the study would involve, answered any questions and arranged a convenient interview time.

During the recruitment period, nine eligible families were identified. One family was excluded because they had already been involved in service-user involvement. Eight families were invited to take part, and all consented to participate. Two interviews were completed one month prior to the one-year hospital review to increase recruitment within the time constraints of a doctorate in clinical psychology.

2.9 Sample Characteristics

All parents and children were given a pseudonym after interview to provide anonymity. All eight participants identified themselves as a main carer: seven mothers and one father were interviewed. The demographic characteristics are provided in Table 1.

The children of the parents were between three and seventeen years of age at the time of surgery. Three of the children were male, and five were female. Despite fulfilling inclusion criteria of a diagnosis of secondary static dystonia, there was variability in dystonic aetiology. Of the eight children, six had a diagnosis of CP, one had an inherited genetic condition; and one diagnosis was unknown. All children fulfilled inclusion criteria as were born with or developed dystonia during child birth, or as a result of complications during birth or in the neo-

natal period. The children's motor and verbal capabilities varied. Motor ability was defined using the Gross Motor Function Classification System (GMFCS; Palisano et al., 1997) and communication ability using the Communication Function Classification System (CFCS; Hidecker et al., 2011). Two children experienced complications with their DBS system in the year post surgery. This demographic information and surgery information is summarised in Table 2.

Information was primarily gathered by the researcher from the interviews. However, the external supervisor collected diagnosis and surgery details from the hospital database.

Table 1: Participant (Parent) Demographics

| Parent Pseudonym | Gender | Ethnicity | Marital Status | Timing of Interview | Location of Interview | Length of Interview |
|-------------------------|---------------|------------------|-----------------------|--------------------------------|------------------------------|----------------------------|
| Julia | Mother | White British | Divorced | 1 Year Review | Clinic | 98 Minutes |
| Rachael | Mother | White British | Married | 1 Year Review | Clinic | 71 Minutes |
| Laura | Mother | White British | Married | 1 Year Review | Clinic | 89 Minutes |
| Linda | Mother | White British | Married | 1 Year Review | Clinic | 82 Minutes |
| Theresa | Mother | White British | Divorced | 1 Year Review | Clinic | 88 Minutes |
| Natasha | Mother | White British | Separated | 1 Month prior to 1 Year Review | Participant's Home | 62 Minutes |
| John | Father | White British | Married | One month prior 1 Year Review | Participant's Home | 111 Minutes |
| Caroline | Mother | White British | Married | One Year 11 Months Post DBS | Participant's Home | 129 Minutes |

Table 2: Child Demographics

| Parent Pseudonym | Child Pseudonym | Gender | Dystonia Subtype | Age at time of DBS surgery | CFCS Level* | GMFCS Level* | Complications DBS procedure |
|-------------------------|------------------------|---------------|-------------------------|-----------------------------------|--------------------|---------------------|---|
| Julia | Imogen | Female | Secondary Dystonia | 17 years old | I | IV | N/A |
| Rachael | Megan | Female | Secondary Dystonia | 16 years old | I | I | N/A |
| Laura | Wade | Male | Secondary Dystonia | 16 years old | I | II | N/A |
| Linda | Philip | Male | Secondary Dystonia | 14 years old | IV | IV | Removal & re-implantation of DBS System |
| Theresa | Charlotte | Female | Secondary Dystonia | 12 years old | I | V | N/A |
| Natasha | Ivy | Female | Secondary Dystonia | 3 years old | IV | V | N/A |
| John | Billy | Male | Secondary Dystonia | 9 years old | III | IV | N/A |
| Caroline | Emily | Female | Secondary Dystonia | 11 years old | III | IV | Lead Revision |

*Gross Motor Function Classification System (GMFCS; Palisano et al., 1997) – This is a five level physiotherapist rated classification system based on self-initiated movement and has an emphasis on sitting, walking and wheeled motor ability. Typically children at ‘level I’ can walk without restrictions, but have difficulties with more advance motor skills, whereas at ‘level V’ all areas of motor function is limited and children require assistive technology and physical assistance.

*Communication Function Classification System (CFCS; Hidecker et al., 2011) – CFCS scale ranges from ‘level I’ indicating minimal impact on communication, where as children at ‘level V’ struggle to communicate effectively and be understood by even familiar people.

Please Note: Diagnosis and cause of disability has not been included in this table and identified next to parent and child pseudonyms to maintain anonymity

2.10 Data Collection

2.10.1 Semi-Structured Interviews

Semi-structured interviews are recommended for data collection to elicit rich and detailed accounts of individuals lived experiences (Smith et al., 2009). Individual face-to-face interviews are the preferred method for collecting this data (Reid et al., 2005). Semi-structured interviews were selected to provide participants with an opportunity to tell their story, and open up a dialogue that was gently guided by the research questions (Smith et al., 2009). The researcher adopted an exploratory participant-led approach to explore what was meaningful for each participant. Additional prompt questions could then be used to inquire about interesting and unexpected areas (Robson, 2011). Interviews also provided access to interpersonal and non-verbal cues such as body language, which enhanced understanding and later interpretation.

2.10.2 Interview Schedule

An interview schedule (Appendix 2) was developed to guide the semi-structured interviews. The interview schedule was constructed through an iterative process of; familiarisation with the relevant literature, discussion with the supervisor, advice from the London IPA group and consultation with service-users. Published IPA guidance was also followed to ensure the interview schedule was informed by the epistemological position and theoretical framework adopted.

Importantly, the interview questions were open ended to encourage unbiased narrative and reflection. The initial warm up question 'tell me a little about your child' was asked as it was hoped to help build rapport because it was less personal, open to interpretation and service-user consultation suggested the child was easier to talk about. This was followed up by 'could you tell me about what life was like before DBS?'. As recommended by the IPA group, a broad introductory question allowed the participant to construct the parameters of the

conversation, and speak about what was personally meaningful for them. The interview questions were structured chronologically. It was felt this would help parents to reflect on experiences of change over time. This was important because during consultation service-users reported it was difficult to remember life before surgery and the subtleties of change.

As recommended by the Research Ethics Committee, the interview schedule was refined to try and manage length of interview. A pilot interview was completed to test out the interview schedule for sensitivity, clarity, flow and content of questions. After listening back to the interview, prompt questions asking directly for thoughts, feelings and examples situated in experience were added to the schedule. On reflection, this pilot interview helped the researcher be more familiar with the interview questions, feel more comfortable with silences, and have an experience of a research interview as a different conversation to a clinical session.

2.10.3 Interviewing Procedure

Participants were interviewed between July 2014 and January 2015. Five of the interviews were conducted by the researcher in a private clinic room at the hospital and three interviews were completed in a quiet room in participant's homes. As recommended by RHUL, the researcher attended the NHS Trust recruitment site's managing aggression and break away training, and followed the NHS lone-working protocol.

Interviews were scheduled to allow parents to speak freely without the presence of their child. Parents were offered a choice of when they would like to be interviewed. The Researcher suggested the interview took place during the child's neuropsychological assessment, to minimise disruption to the child and family. For home visits, interviews took place whilst children were at school/childcare.

Time was invested before the start of the interview to make the participant feel comfortable and build rapport. Before each interview the information sheet was discussed again, an

opportunity provided to answer any questions and the researcher emphasised key ethical features: participation was voluntary, right to withdraw at any time, confidentiality and anonymity practice. Participants signed a written consent form indicating they had read and understood the information sheet.

In line with a participant-led interview, the researcher was guided by how comfortable participants were in talking and how much they wanted to say. Consequently there was variation in interview length: Interviews ranged in length from 62 to 129 minutes. The interviews were audio recorded on a personal digital recorder. At the end of the interviews several parents reported how much they enjoyed sharing their experiences and that hospital appointments are understandably focussed on the child, often neglecting their experiences. During the course of the interviews, four parents became tearful. The researcher gave the participants time and checked they were happy to continue. None of the parents were distressed at the end of the interviews.

2.11 Procedural and Ethical Considerations

2.11.1 Ethical Approval

Following Peer Review by RHUL research subcommittee, this study received full ethical approval from the London-Bloomsbury Research Ethics Committee (Appendices 3 and 4). Approval was granted from the Research and Development Department of the NHS recruitment site (Appendix 5). The research was also approved by the Royal Holloway University of London Psychology Department Ethics Committee (Appendix 6).

The Research was carried out in accordance with the ethical guidelines set out by the British Psychological Society (2010). The relevant ethical issues were gaining informed consent, ensuring confidentiality and anonymity, managing distress and potential risk. The study was designed to minimise potential risks, and procedures were in place to manage ethical situations that may have arisen.

2.11.2 Informed Consent

Potential participants were provided with an information sheet, outlining without deception the purpose of the study and likely topic areas to be discussed during the interview. The information sheet also outlined in detail what participation would involve, and full ethical considerations. Information sheets were checked for readability and content, and approved by two parents during service-user consultation.

After receiving the information sheet, all participants were given time (at least 24 hours) to consider participation and provided with an opportunity to ask questions. Furthermore, interviews were arranged 2-4 weeks in advance, so participants were reminded at the start of the interview that they were under no obligation to take part, and that participation would not impact the current or subsequent care their child received. It was important parents understood that the research was not a routine part of the service, to ensure that participants were not taking part out of loyalty to the service after long term involvement with the DBS team.

Before the interview took place participants were also reminded of their right to withdraw and the potential use of verbatim quotes in published reports. All participants were over 18 years old and deemed to have capacity to provide informed consent. All participants completed a written consent form (Appendix 7) prior to participation in the interviews. The researcher countersigned the consent form. The participant kept a copy, and the researcher placed a copy in the site file stored securely at the NHS recruitment site.

2.11.3 Confidentiality and Anonymity

Participants were informed that all information collected was confidential, and of the limits to confidentiality, such as a disclosure of risk to themselves or the child. In this event, the researcher would be required to inform the team and other necessary parties in accordance with NHS site protocols.

All transcription was undertaken by the researcher, so that the raw data remained confidential. Confidentiality was protected by assigning a participant number to identify audio files, transcribed materials and personal information. All identifying information was removed and participants were given pseudonyms to protect their identity. The only identifiable information was the consent forms, which were stored separately at the research site.

All data collected was held according to the Data Protection Act (1998) and NHS confidentiality standards. All paper data was stored securely in a locked filing cabinet at the NHS site. All electronic data was stored on an encrypted storage device. Audio recordings were deleted after transcription and analysis.

2.11.4 Distress and Participant well-being

Brinkmann and Kvale (2008) suggest that qualitative studies are particularly vulnerable to ethical issues because human interaction affects researchers and participants. It was not anticipated that the interviews would cause any harm to participants, and service-user consultation suggested that some parents might find it of benefit psychologically to share their experiences and “reflect on the positives”. However it was possible that parents could become upset or distressed when talking about their child's health and the impact of DBS.

It was the researcher's responsibility to manage any distress during the interviews. A sensitive approach was adopted by the researcher, with a focus on initially building rapport and familiarity to help participants feel comfortable. Participants were reminded to share what they felt comfortable with, take their time, and that they did not have to answer all the questions. Although understandably some participants were upset at times, no-one refused to answer any questions or asked to stop the interview. After the interview, participants were debriefed about the research, and were provided with a debriefing sheet outlining further support options (Appendix 8). However, no participant needed a referral for further support.

As Brinkmann and Kvale (2008) suggest, interviews can also have an impact on the researcher. It was therefore important that the researcher had access to supervision throughout to reflect on the process and any personal reactions and feelings that arose during the interviews.

2.12 Service-User Involvement

As already referred to throughout this chapter, service-user involvement was conducted by the researcher at different stages of designing the study. This involved liaising with parents who had all been through the DBS process. Below is a summary of this, and the outcome and impact of service-user involvement on the study design and development of materials.

Stage 1 - Initially the researcher met two families who had undergone DBS, and shadowed clinic appointments, to hear participant stories and familiarise herself with the context of DBS. The researcher also consulted with the clinical team to hear their views of parents' concerns, and important areas to focus on.

Stage 2 - Consultation with three families to discuss exploratory research aims, interview schedule, procedural and ethical issues (see Appendix 9 for Consultation Interview Guide). This consultation informed many research decisions:

- Exploratory Aims: All parents felt it was paramount to talk about child's views and experiences. They described how the children were at the heart of this process, and would be unable to separate their experiences from the child's experiences. It was therefore deemed valid to focus on both parent's and children's experiences, and consider dystonia as a relational construct. Parents thought the research was important and could have useful clinical implications

- Interview Schedule: The main areas of the interview schedule were deemed acceptable. Parents wanted to be asked directly about children's experiences, and be asked directly about positive as well as negative experiences
- Procedure: Parents agreed it would be best to be interviewed as close to the one year follow up as possible. Parents wanted choice of when the interview took place during the review appointment. Parents strongly thought that the families should chose who takes part in the interview, that it should not be assumed who the 'main carer' was, and that fathers should not be excluded.
- Ethical Issues: The parents consulted welcomed the opportunity to share their experiences. Parents did not think it would be distressing talking about their experiences, and many thought it would be positive for them to discuss their experiences.

Stage 3 - Two parents provided feedback on the information, consent and debrief sheets. Overall these were positively received, and the only adjustment was to add a definition of secondary dystonia

Stage 4 - A pilot interview was completed with a father of a child with primary dystonia. As discussed this was important to enhance researcher competence and familiarity with the interview skill. The father felt questions were appropriate, sensitive and comprehensible. It also led to minor revision of the interview schedule based on feedback and researcher reflections (e.g. more experiential focus of prompt questions).

Stage 5 – Two families were shown the master table of themes, and reported that the themes represented their experiences of living with dystonia and DBS. No themes were considered to conflict with their experiences.

Stage 6 - Some parents have agreed to consult on the clinical outputs of this research (e.g. psychoeducational leaflets for parents). This will be completed after development of these materials.

2.13 Data Analysis

The interviews were analysed following the flexible IPA guidelines proposed by Smith et al. (2009) under guidance from the academic supervisor, and support from the London IPA peer support group. Interviews were digitally recorded and transcribed verbatim by the researcher. The process of listening and re-listening involved in transcription is an important element of engaging with the text (Tilley, 2003). IPA does not require prosodic aspects of communication to be recorded (Smith et al., 2009). However, non-verbal utterances (e.g. laughter, crying), pauses, significant volume/pitch changes were recorded (Smith & Osborn, 2003) to capture the vividness of conversation and strengthen interpretations. All transcripts were typed into a word document table with wide margins to allow exploratory coding and emergent themes to be noted.

Reading and Re-Reading: Each transcript was read and listened to several times. This enhanced familiarity, and allowed the researcher to begin to actively engage with the data and enter the participant's world. Richer and contradictory sections were highlighted, whilst remaining open to the development of new thoughts with each re-reading.

Initial Exploratory Coding: Initial notes were recorded in a line-by-line review of the data. These examined the semantic content and language used, and was recorded in the right-hand margin. The researcher recorded descriptive, linguistic and conceptual comments. Descriptive comments focussed on the content and subject of what was said. Linguistic comments recorded the use of language (e.g. tense, pronoun use) and non-verbal communication (e.g. hesitation). Conceptual comments are interpretive in nature, and required the researcher to question the underlying meaning of experience at a higher level of abstraction.

Developing Emergent Themes: Emergent themes were developed from the researcher's exploratory codes to capture the essence of what was important for each participant. These were recorded in the left-hand margin. Emergent themes were created to map connections and relationships between exploratory codes, and required a higher level of abstraction. The task was to capture participant experience and researcher interpretation within theme labels. It was essential at this stage, to remain close to the data and ensure notes were grounded in participant experience.

Clustering and Collapsing Emergent Themes: Emergent themes were then listed chronologically, and the researcher looked for connections to begin to organise clusters of related themes into mind-maps. Themes were further refined and taken to a higher level of abstraction, allowing some themes to be discarded. Clusters were given a descriptive label to capture the conceptual nature of themes within them. A summary table was created of clusters, themes and key quotes.

Moving to the next case: The previous four stages were repeated for the remaining seven transcripts. To retain an idiographic focus, the researcher tried to bracket ideas from previous cases and treat each transcript individually.

Cross-Case Analysis: Clusters were compared and contrasted across cases, looking for convergences and divergences within the data. The researcher re-organised clusters and created super-ordinate theme labels to capture a more abstracted and synthesised overall representation of participant experience. A master table of themes (Results Table 3) was created to capture the clusters nested within superordinate themes. The final task was to create a coherent narrative account of the findings, as presented in the results chapter.

2.14 Validity and Quality in IPA

General Guidelines have been developed to assess the validity and quality of a growing field of different qualitative methodologies (Elliot et al., 1999; Yardley 2000, 2008). Yardley's (2000) criteria and Elliot et al.'s (1999) recommendations have been followed throughout the different stages of this study to maintain quality and ensure the research is rigorous and reliable within the epistemological position and principles of IPA. Attempts to maintain quality following Yardley's criteria and apply these to IPA specifically (Smith, 2011) will be summarised below.

2.14.1 Sensitivity to Context

An extensive literature review was completed to increase understanding of theory and ensure the study was sensitive to the context of relevant literature. This is also an important pre-requisite on which to base the hermeneutic aspect of IPA, where added-value interpretation comes from an oversight of the whole (Smith et al., 2009). Sensitivity to socio-cultural context was demonstrated by liaising with the hospital clinical team, meeting families and observing goal setting and review appointments. This helped increase researcher understanding and enhance sensitivity to context in planning and implementation stages. Sensitivity to participant perspectives was of paramount importance and was fully attended to through service-user involvement, and a flexible and open interview to allow participants to convey what was important to them. An iterative and thorough data analysis ensured themes were grounded in participants' perspectives, and various verbatim extracts were used to support each theme in the narrative account, and appendices. The researcher also attended to divergences within the data, to ensure that nuanced experiences were also represented, and not excluded based on prevalence.

2.12.2 Commitment and Rigour

Commitment was shown by thoroughly engaging with the topic area, the researcher asking prompt questions during interviews to enhance the richness of phenomenological data collected, and a rigorous and thorough depth of analysis. Methodological competence was demonstrated through careful consideration of qualitative approach, to ensure the research paradigm and methodology was appropriate to answer the research questions. Furthermore, a pilot interview was completed to enhance researcher skill in conducting semi-structured interviews. The researcher attended an IPA support group, and consulted published IPA literature to ensure methodological competence in producing a high quality IPA study. This was also achieved by the researcher striving to achieve a deep and interpretative analysis grounded in idiographic engagement (Smith, 2011) by attending to and highlighting convergence and divergences. To achieve a rigorous IPA, as recommended by Smith (2011) extracts from at least half of the participants were represented in each theme (Table 3).

2.14.3 Transparency and Coherence

To provide a credibility check (Elliot et al., 1999) the first transcript was independently coded by the internal supervisor. This was vital to ensure the analysis was not limited to the researcher's perspective (Elliot et al., 1999). The internal supervisor and a fellow trainee clinical psychologist also read through the case-by-case themes, and considered overall fit with super-ordinate theme clusters and verbatim quotes. This was also important to ensure there were no overstatements, the themes were coherent and interpretations were grounded in participant accounts (Elliot et al., 1999). The internal supervisor was able to provide a credibility check to assess if the themes resonated with their clinical experience at the hospital. This process was important to enable the data to be considered from multiple perspectives, rather than a single point of view (Mays & Pope, 2000).

A paper trail was kept from the coded transcript analysis through to the development of themes to ensure transparency in linking raw transcript data to the final report. Consideration

was given to the reader's perspective, and a participant extract and theme list (Appendix 10) was included to allow the reader to follow the process, assess fit, and make sense of the study as a consistent whole (Yardley, 2008). By aligning with a hermeneutic phenomenologist position, it was acknowledged that researcher experience and 'foreunderstanding' was embedded in interpretations (Heidegger, 1962 as cited in Smith et al., 2009), and transparency and reflexivity are demonstrated (see owning one's own perspective and epistemological position).

A reflective journal was also kept from the beginning of the planning stage, and throughout the analysis (Appendix 11). This helped to promote reflexivity, and present transparency in how the researcher's position and assumptions may have impacted on the analysis (Meyrick, 2006). Reflections were also recorded before and after each interview. This helped the researcher reflect on preconceptions and thoughts that arouse, and to bracket off these ideas during the analysis.

2.14.5 Impact and Importance

Yardley (2000) argued that the value of research lies in its empirical relevance and impact on clinical practice. This research was thought to have huge importance given the growing number of childhood DBS surgery procedures completed, the inability of quantitative measures to capture the subjective meaning of change and the novelty of capturing parental narratives (discussed in introduction). Following analysis, the results were examined in relation to existing theories and literature, and focus given to how the results could inform clinical practice and DBS surgery preparation and adjustment support. To ensure dissemination and clinical value, the research will also be written up for publication and conference presentation, and be shared with the clinical team (see discussion).

2.14.6 Owing One's Perspective and Personal Reflexivity

Reflexivity is integral to the qualitative paradigm because it acknowledges how a researcher can never provide a truly objective view and the researcher's role in the double hermeneutic of interpretation. It was important to acknowledge how the researcher's own values and beliefs may impact on the analysis (Elliot et al., 1999), and reflect on the researcher's own position in relation to the phenomenon to consider how they may have shaped and influenced data collection and analysis (Willig, 2008). The researcher is a female white British Trainee Clinical Psychologist. She did not have children, and had no personal experience of being a parent. The researcher also has no personal experience of a movement disorder or chronic illness. However, her father was diagnosed with a chronic illness approximately ten years ago, which has involved frequent hospitalisations, surgeries and a long process of adjustment as a family.

Pre-training, the researcher also worked clinically with children with disabilities and their families in a variety of different settings (NHS, education services and respite social care), and during training the researcher has worked in a paediatric department. In these roles she witnessed the profound impact of CID on the different aspects of children's lives, and the strength of parents to manage and find ways of adjusting and learning to live with disability. In summary, the researcher has professionally witnessed and personally experienced the impact of chronic illness and hospitalisation/surgery stress on the family system, and she values the importance of research addressing the often unheard voice of the carers and relatives.

Her research interest in dystonia predominantly stemmed from an academic interest in health and disability contexts. The researcher had little knowledge or experience of childhood movements disorders prior to undertaking the research. The participants were aware that the researcher was not part of the clinical hospital team which, on reflection, allowed participants to speak openly regarding their positive and negative experiences of DBS. Being separate from the team and not having a child with dystonia, allowed the researcher to have a naive and curious stance, and to step into the lived world of participants. However, during the

interviews the researcher was surprised by the overwhelmingly positive accounts of experience and parental reactions. This highlighted how the researcher may have held implicit preconceptions and hidden assumptions (Coyle & Wright, 1996) of the negative aspects of parenting a child with a disability, perhaps reflecting the dominant narrative held in the empirical literature and wider societal discourses. These reflections were recorded in a reflective journal to allow the researcher to remain open and sensitive to times preconceived ideas may close down conversation. It is hoped that providing insight into the reflexive process, and the researcher's multiple positions and experiences, will demonstrate transparency in how these factors may have influenced the research, thus increasing its integrity and trustworthiness (Maso, 2003).

Chapter 3: Results

Four superordinate themes, comprised of sixteen subthemes, emerged from the analysis of participant accounts. These are presented in Table 3, and will be discussed in detail in this chapter. All theoretical discussion and integration with existing literature is presented in the discussion to allow participant experience to be privileged and presented in an unfolding manner as experienced by the researcher during the analysis.

Given the extensive nature of the data, themes were selected to capture the richness of data essential to participants' experience, and were not chosen based purely on prevalence (Smith & Osborn, 2003). This demonstrates the researcher's effort to capture the group experience as a whole, whilst also retaining the idiographic nature of the phenomenon. Given the explorative focus of IPA, the researcher also attempted to highlight interesting and novel themes, and unexpected aspects of participant experience. Themes were therefore chosen to elucidate research aims, and offer insight into areas previously neglected in the literature.

The researcher hoped to present a coherent multi-layered narrative account of participant experience; Rich descriptions of participant experience were embedded within verbatim extracts, and higher level analytic interpretations. The narrative was written to allow the reader to distinguish descriptive commentary, from more abstract and interrogative interpretations made by the researcher (Smith, 2004).

Extracts were selected from transcripts to display the meaning of each theme, demonstrate examples of interpretative analysis, and display the experience of different participants across themes. Table 3 presents the number of participants contributing to each theme. To help illustrate the richness of participant accounts, a summary table of additional extracts is provided in Appendix 12. By displaying quotes not included in the narrative, the researcher hoped to demonstrate the depth of participant experience pertaining to each subtheme, evidence the themes as grounded in the data and display transparency in the representation of themes across participants.

To maintain confidentiality and anonymity of participants, some quotes have been edited to remove identifiable information and pseudonyms have been used for parents and children. To support clarity and enhance readability, less relevant material has been removed as indicated by three dots '...'.

Table 3: Master Table of Themes

| Superordinate Theme | Sub-Theme | Parents contributing to the theme |
|---|---|--|
| A Difficult Life with Disability | Trapped Inside an Uncontrollable Body A Restricted Life – Child and Family Perspectives Emotional & Physical Demands of Parenting | 8 8 8 |
| The Meaning of Disability and Normality | The Impact of Social Comparisons and Feeling Different Treated Differently - The Social Construction of Disability Making the Choice to 'get on it with' and the Process of Acceptance Overcoming Disability and Taking Control | 8 6 8 8 |
| An Emotional and Uncertain DBS Journey | Facing the Uncertainty of Decision-Making Struggling with the Unknown and Unexpected Emotionally Overwhelmed by DBS and the Hospital Experience Coping with DBS - 'it's the people who get you through' | 8 7 8 8 |
| The Experience and Perceptions of Change | Hope and Realistic Expectations of Change Significance of Positive Changes – 'it's everything' Making Sense of 'Quieter' Changes than Expected Compromise of Change and an Ongoing Struggle A New Perspective Looking Forward | 8 8 4 8 8 |

3.1 A Difficult Life with Disability

This theme captures the physical, psychological and social impact of living with dystonia. It captures the systemic impact of dystonia on the lives of the whole system, and the physical and emotional demands of parenting. Control was central to the meaning of experiences, as children struggled to gain control over their bodies, and parents struggled with their inability to control the situation and make things better. Lives were perceived to be restricted, isolated and trapped in an unwanted life with disability. Overall therefore this theme highlights the experience of life as difficult for the child, parent and family system before DBS surgery.

3.1.1 Trapped inside an Uncontrollable Body

All parents described in detail a range of dystonic movements that physically impacted the everyday life of their child. There were descriptions of tightness, stiffness and shakiness of particular limbs. For Linda, this muscular tightness was so extreme it was as if the limbs were so out of the child's control they were locked and no amount of effort could release them:

When his leg goes so tight that you can't bend it, or like the knees go so together you can't even pull them apart (Linda)

For most these muscle contractions were intermittent, and exacerbated when trying to perform tasks or 'enhanced at high emotion'. In these children particular limbs were most commonly affected (e.g. arms, hands, legs). However, for other parents the dystonia impacted the whole body, with more sustained muscle contractions resulting in abnormal postures:

Her body was just stiff, so it was just a constant spasm and her muscles were really tight all the time. She had a really strong curve, she, like had a banana (Natasha)

Similarly, Caroline described how Emily would go 'as stiff as an ironing broad'. All parents described the variability and changeable nature of dystonia:

It's that element of unknown with Emily, I think (pause), what are her movements going to be like? It's very unpredictable with those dystonic movements (Caroline)

For most parents the struggle of dystonia appeared to lie in its unexpected nature and living in a constant state of unpredictability: For all parents these movements were 'involuntary' and the narratives were dominated by an overwhelming sense of a lack of control:

It's very hard because obviously you've got this amazing personality and this child in there that's trapped in this body that's not doing what she wants it to do, very difficult (Caroline)

This description evoked powerful imagery of being 'trapped', and emphasises the emotional reaction of a mother watching her child struggle. Julia echoed the words of Caroline, by comparing her daughter's dystonia to a 'ball and chain':

That arm could kick off, all the time. And, its just like a ball and chain really, you're never going to get rid of it (Julia)

This metaphor suggests something inescapable and imprisoning about dystonia, and is further symbolic of feeling 'trapped'. There was a sense of being unable to break free from a life controlled by disability.

Parents spoke also of their child's attempts to try and gain control of their body:

She would absolutely sweat buckets just trying to maintain that posture (Caroline)

Caroline provided a very visual image of her daughter's physical efforts to desperately try and overcome the power of dystonic spasms. Similarly Rachel conveyed through exaggerations and repetition the 'constant' struggle, and sense of an inescapable life with dystonia:

It's so (emphasise word) tight, so painful, and again, what she does because it's so tight and whatever position she's constantly having to rearrange it (gesturing controlling arm). So she can't concentrate on anything, cause she's constantly trying to put it in a position where its not stiff and its not painful
(Rachel)

Like Rachel, for many children dystonia caused pain. This was emotional and difficult for parents to watch and was best captured by Julia:

This guy said to Imogen, 'if there is anything, anything we could do to make things better for you what would it be'. She said 'chop my left arm off'. That's how much that spasm was bothering her **(Julia)**

In this evocative description, this extract highlights the extent to which uncontrollable spasms impact on the child, and offers a succinct depiction of an experience observed across most of the sample.

3.12 A Restricted Life – Child and Family perspectives

All parents described how dystonia limited and restricted different aspects of the lives of the whole system. For the parents, there was a stark disparity between the cognitive and physical capabilities of the child:

Obviously severely affected by cerebral palsy from a mobility point of view. She has athetoid and dystonic CP. Obviously urr, quadriplegic so basically full body. Urm, however she's got amazing cognitive ability **(Caroline)**

Parents talked of the positive aspects of their child's mental abilities, and Laura particularly took pride in explaining Wade got 'virtually full marks' with the assistance of a scribe. There was a sense of wanting to show their child as more than physically impaired, and focus on their strengths perhaps as a way of coping with the difficulties. For Rachel this disparity was impacting on the emotional wellbeing of her daughter:

She should be like an A star student head girl type girl, because that's, that's who she sees herself as. Because she doesn't have that ability to be an A star student, that really upsets her. (Rachel)

There was a sense that the disability was not only restricting the child, but impacting on the child's developing sense of self because she was acutely aware of her own limitations and disappointment at being unable to have the identity she aspired to hold.

For all the families, dystonia affected the child's ability to physically participate in activities of daily living. For Caroline however, dystonia had a larger impact:

She couldn't roll over, she couldn't eat, she couldn't talk, she just couldn't do anything (Natasha)

Parents spoke of the social impact of disability. Many described their child's lack of friends as the hardest thing, and a sense of being isolated either because of difficulties communicating or physical impairments, which was emotionally upsetting for parents. This social isolation was strongest in Laura's account:

because of his speech impediment he (pause) and he's told me this, so (pause), in quiet moments he tells me things (pause), urm because he's embarrassed about his speech, anybody who tries to be friendly he feels that they're pitying him, he, he doesn't feel that he's accepted for who he is (Laura)

In this poignant and emotional extract Laura was struggling to express her words in the interview. Wade's insight into his disability appeared to be contributing to feelings of depression, and there was a sense that Wade was developing a fragile sense of self, and one that was defined by disability. Wade's disability appeared to be having a negative psychological impact on his self esteem and confidence.

Parents spoke of how disability 'changes every aspect of your life'. Parents described feeling restricted themselves as a result of their child's dependence on them and a sense of being unable to do things (e.g. go on holiday, leave child, eat out as a couple). Laura described a feeling of isolation as a result of the difference created by disability:

Urm (pause) I suppose there's always a sense of isolation... It would be lovely to know somebody who's got a child similar to your own child. I know that's just about impossible (Laura)

Throughout most parents' accounts, there was a sense of isolation that paralleled the child's experience. Sadly for Laura, there was a sense of sadness having resigned herself to it being 'impossible' to find someone who would understand her experiences.

For Julia, she felt restricted by being 'tied' to her child:

Well you kind of tied to the house in effect, and to Imogen (Julia)

The language used implies a sense of parental duty, and a lack of choice. There is similarity between the child's experience of feeling trapped within their own bodies, which is echoed in Julia's account of being trapped by disability and tied to her child.

For the parents with other children, there was a sense of an overwhelming struggle to meet the different and sometimes competing needs of all their children and 'provide equal amounts

of time'. This struggle was strongest for Natasha, and succinctly captures the experience of other parents:

Its, me feeling like I'm being ripped one way and another, because I should be with my other kids as well. That is what I've found hardest (Natasha)

This evoked powerful imagery of being 'ripped', as if she was constantly splitting herself to meet the multiple demands of parenting. The language of 'ripped' could also describe her emotional state, and it is possible that this feeling of being torn creates inner conflict and pain at being unable to give her children everything.

All the parents described wider consequences on family functioning and changing relationships. For some, siblings took on caring responsibilities and a different role within the family. For others, their relationship broke down with their partner. This systemic impact was captured succinctly through John's unequivocal language:

Anybody who says it doesn't affect everybody is either lying to themselves or neglecting someone along the line because they're not aware it's affecting other people, because it has to (John)

3.1.3 Emotional and Physical Demands of Parenting

All parents described how every day tasks (e.g. getting dressed, washing) involved physical assistance and parental support. For Rachel there was a sense of holding responsibility, and the repetitiveness in her language ('I have to, I've got to') conveying perhaps how her duties feel like they were never ending:

Because, on a daily basis (sigh) if (sigh), just, you know, if she wants to put her hair up in a towel after she's washed her hair, I've got to help her, I have to

help her do her bra up, help her with sanitary towels when it's time of the month (Rachel)

For parents of children with more severe and generalised dystonia, there were frequent descriptions of the physical effort involved in daily care, and descriptions of how 'draining', 'exhausting' and 'time-consuming' the role. Julia described the physical effort involved in trying to help her daughter get dressed:

It used to be like pulling a nail, a big nail out of a brick wall. It was just oohh, such an effort (Julia)

This visual metaphor and language emphasises the daily struggle some parents would go through just to complete activities of daily living. All parents conveyed an unwavering dedication to their child. For Julia and Laura this involved the time and money to provide extra support sessions (e.g. hyperbaric oxygen, conductive education):

And the sacrifice driving up to Wales sort of three times a week, you know that's three days of driving up and down...that's a lot of time and money (Laura)

Laura used the word 'sacrifice' to describe her willingness to give up aspects of her own life, to provide the best QoL for her child.

For several of the parents, there was a sense of isolation through being alone in their responsibility for the child. Rachel described how 'it does all fall on me', Theresa explained how 'I did feel very much it was on my head', and Julia expressed the role as being 'all left to me'. For Julia, having carers helped to alleviate this responsibility:

It's just nice to just, not have, maybe the responsibility, just having a total chill out for, knowing you're not at the end of a bell type thing (Julia)

This metaphor encapsulates the sense of duty and being on call '24/7' that dominated many of the parents' narratives, and extends previous descriptions of being trapped and tied to the child. For Julia this weight of responsibility resulted in a physical injury to herself, because of her unwillingness to accept she needed more support physically.

Parents experienced strong emotional reactions to witnessing their child struggling to control their body and manage the pain:

It's hard, it's hard for her, because her bodies fighting against her every step of the way and you just want to change that, and you can't (Theresa)

For Theresa just as the child was battling for control of their body she appeared to be struggling with inner conflict of being unable to 'fix things and make things better'.

These accounts all highlight the emotional impact dystonia had on parents. This theme was strongest for Caroline whose emotions were so overwhelming that she experienced psychological difficulties and reported feeling 'burnt out':

I've, I've recently, on a number of occasions had to go to my GP for depression and anxiety, it's very hard because I want to be able to fix things for her, and be in control, that's what mum's do.. and unfortunately we can't always do (Caroline)

Like Theresa and many other parents, the main difficulty seemed to lie in being unable to 'fix things' as opposed to the physical demands of parenting.

3.2 The Meaning of Disability and Normality

This theme encapsulates the multiple meanings of disability within an individual, family and societal context. Social comparisons, the negative reactions of others, and societal barriers created feelings of difference for children and parents. At times parents struggled to accept the impact and meaning of disability, but engaged in a process of adjustment choosing to 'get on with it' and take control of disability by attempting to provide the best quality of life for their child. Secondary dystonia was perceived as a socially bound phenomenon, and parents searched for normality by overcoming societal barriers and physical limitations of disability.

3.2.1 The Impact of Social Comparisons and Feeling Different

All parents described feelings of difference that exist at multiple levels within the family unit and the social context of disability. For all parents there was a process of social comparison which influenced their perceptions of self and the child. All parents described their child's insight of their physical limitations, and a sense of how disability contributed to feeling different:

He's self conscious about various things about the way he walks, the way he talks etc. So I think this, urm, he always likes all his limbs covered. He always wears long trousers, he always wears long shirts (Laura)

For Wade, this feeling of visible difference was so strong that he felt embarrassed and tried to hide his perceived physical difference. Many parents also perceived their child to be different because of their physical disability. For most parents this sense of physical difference was apparent from birth, and continued to exist throughout the child's life:

It was obvious from her fisting and various things as she started to develop, urm, that she had problems (Theresa):

*You can still see there something wrong with her (**Natasha**)*

This difference seemed to be created by a sense of being different to the perceived normality, which for Laura created an unbearable sadness at Wade not having a 'normal teenage experience':

*I'm sad for him for what he's missed and what, having three older ones, and they used to have friends coming...having a house full of teenagers... And that hasn't been Wade's experience. He hasn't had a normal teenage experience, his teenage experience is sitting in front of a computer (**Laura**)*

This emotional extract, captures the sense of loss felt by many, which appeared to be heightened by how different Wade's life has been in comparison to his siblings.

For six of the parents this feeling of difference extended beyond the child, to perceptions of themselves as different. For some this involved comparisons to other parents:

*Well I suppose emotionally, because you don't have time to yourself, like other parents (**Julia**)*

For some this created a sense of isolation as both a parent and a person. This was most apparent for Laura, who struggled to find support because of the uniqueness of dystonia:

*I went once. It doesn't work when you've got different disabilities, you know I don't have any thing in common with a child with severe autism, or hearing, deaf or blind. You can't, you can't do that, you can't generalise, cause we've all got very specific disabilities, for the children (**Laura**)*

Laura spoke of herself as having a specific disability, before correcting herself. This highlights how perhaps for Laura she also feels disabled at times, and this is something that impacts on her sense of self. For many this feeling of difference started at the point of diagnosis, recalling vividly how 'you think it will never ever happen to you'.

Additionally for many there was an awareness of having a different relationship with their child:

My relationship with (Siblings) is probably a normal father-son relationship, where as with Billy it's a bit more than that (John)

For Caroline this difference came from consciously taking on a different parenting role to try and be more than a parent to Emily and perhaps compensate for her lack of friends:

As parents we have to be able to (pause) kind of act not just as parents, but able to be a bit silly and say ok I don't really like One Direction but I will play it full blast in the car because it's the fun thing to do, because she needs that, (Caroline)

In contrast other parents struggled to detach themselves from their perceptions of the child as 'vulnerable', which resulted in an overprotective parenting style. Julia particularly struggled to let go and continued to 'cotton wool' Imogen:

It's hard sometimes to let go, when you've been so intense with somebody (Julia)

3.2.2 Treated Differently – The Social Construction of Disability

Throughout several accounts there appeared to be a barrier created by society's reaction to disability. For some, particularly Natasha and Caroline, this seemed to be a result of people who 'sit and stare' and were experienced as judgment and 'stigma':

She'd dribble everywhere and be wet through, and there shouldn't be this stigma when you are out and about with your child, and it shouldn't exist, but it does, but its hard (Caroline)

For Natasha and Caroline this misunderstanding was emotionally challenging because there was a sense that people only see the physical disability, and don't look beyond that to see the child's personality or appreciate what their child is cognitively capable of:

but it frustrated me because actually what I want people to see is what we see, we see such an amazing child here, its not about both those physical movements, its about her personality, its about who she is (Caroline)

They were doing like an up and down song, so her chair was being raised up and down... and some of the parents were laughing behind me, and I know they weren't laughing at her, but I found it really rude...they're laughing because they think its cute whatever, But actually how do you think she might feel if she hears that you're laughing (Natasha)

These extracts highlight how children were treated differently because of a visible difference, with little insight into the personality and ability hidden behind the disability.

In contrast, Laura's son experienced a more extreme negative reaction of bullying:

Some of the children there were quite unpleasant to him or talking about him, and I suppose he feels sometimes people talk behind his back about how he walks or how he talks (Laura)

In its most negative form, it appeared that society's reaction to disability has the power to isolate a child, and influence their developing sense of self and psychological well-being.

Throughout parents' accounts there were various examples of how societal reactions created a barrier to the child engaging in opportunities and experiences. This is captured most strongly in Julia's experience of her daughter not being allowed to go for a balloon trip:

When he saw her in a wheel chair he said 'oh she can't go'. I said 'well why not'. 'Oh she has to be able to do an emergency crouch'. But I had somebody with me. And I said 'well we could put her in the crouch'. And he wouldn't have it. Ah, I was absolutely devastated, devastated (Julia)

For Julia she experienced sadness and devastation at missing this 'once in a lifetime chance'. The power of Julia's emotional reaction was still so strong, that she switched to speaking in the present tense as if she were talking to the pilot in the interview room. Julia perceived the pilot's reaction as 'a bit of bias and maybe prejudice' for assuming 'she wasn't suitable' because of the wheelchair. There is again, that sense of misunderstanding a child's capabilities because of the visual appearance of disability.

Several of the parents also described the 'fight' to access services, and a sense of how disability was created by society's reaction:

The fact that you spend a large proportion of your life fighting for things for Billy because he's disabled, that actually makes him disabled (John)

All these experiences serve as reminders of how difference can be created by societal reactions to disability, and highlight disability as a socially constructed phenomenon. The meaning of disability as social constructed was eloquently captured by John who is also the father of triplets:

Almost being a triplet it should be a registered disability, it should be. No one ever describes me as the disabled child's father I am the triplets' father...occasionally I am Billy's dad but I'm not labelled by that... And they get that at school, where people just call them the boys or the triplets, and that's almost as much an affliction if you like, as you know Billy's disabilities (John)

For John being disabled wasn't about physical impairment, but about how you are viewed, labelled and treated by society. John was questioning the meaning of disability, and this underlines a difference between being different, and being treated differently.

Similar to his father, the meaning of disability as socially constructed was also reflected by Billy, in how he makes sense of his own disability.

he's quite sympathetic to people not understanding why he can't do things, or sometimes people can't understand him... as if its them that's got the problem, rather than sort of people don't understand me because I'm not, its they should be trying harder (John)

This extract locates the source of disability as a 'problem' in the perceptions and reactions of others.

3.2.3 Making the Choice to 'get on it with' and the Process of Acceptance

This theme captures the perceptions and reactions of all parents to having a disabled child, and the process of acceptance. Many spoke of their 'shock' at diagnosis and the cognitive

processing that accompanied their reaction. Several parents described a 'decision' or 'choice' between two response options:

When she was first diagnosed I was so low, really low, urm and I think from there there's two ways you can go, you can either go up or down, I after a couple of weeks of really moping and feeling depressed and down, I sort of gave myself a kick up the behind and said 'you know you've got to get on with this, and you've got to make the best of this' and that was it (Theresa)

For other parents, there was a choice between having a positive and negative attitude, and Linda choose to be positive and 'get on with it':

I think personally, you know, just get on with it, because if you start moaning with things, I think that's when your problems start, I'd rather just get on with it, get it done (Linda)

For these parents there was a sense of taking control over the situation. On reflection, I wonder if this choice may have enabled parents to move from a position of feeling helpless, to more empowered and able to start to accept and adjust to the presence of disability.

When considering the meaning of disability, several parents spoke of their parental 'responsibility', and there was a sense of an unwavering commitment to their child.

People say to me I don't know how you do it, I just do, I'm her mum, it's my job (Natasha)

It's not what you want for her, it's not what you want for you, but it is what it is so you've got to make the best of it (Theresa)

Unlike the choice in how to respond psychologically, there was no choice but to accept and 'keep going': All eight parents spoke of their acceptance of their parental role. However, there was a pervading feeling of sadness of it not being the life they would have chosen or wanted for themselves and their child.

For some this process of acceptance was almost automatic. For Linda, she described how it was the only life she has known:

Cause obviously it's not something, where if you said if your child's got cancer that's just been diagnosed, whereas when he's had since been born, don't know any different and there's nothing can do about it (Linda)

Where as for others, there was a sense of a process of adjustment which happens over time:

I just think it is something that you get used to, if you know what I mean. I think you just get used to your lot (Rachel)

However, for Caroline this process was not over, as she continues on a 'learning curve':

I don't think we've finished learning yet... the disability I don't think it stops here, I think it continues, you continue to learn about how to understand it, accept it if you can (Caroline)

For all parents there were occasional moments where they struggled with acceptance of the presence of disability in their lives, which seemed harder to accept than the reality of their parenting role, and thoughts turned to how life could have been:

But there are days that you go this shouldn't be happening, we shouldn't be brushing her teeth, we shouldn't have to dress her, yeah very difficult (Caroline)

However, parents seemed to balance these moments, by choosing to focus on how positives have come out of a 'negative', and there was a sense of overwhelming pride in parents' accounts of how their children have responded to having a disability:

Because obviously some of the time she's sitting, she's watching...She's a lot more empathetic, a lot more patient I suppose, she's amazing, she's incredible, she really is (crying) (Theresa)

It appears as if acceptance therefore, was not experienced as an absolute stage one reaches, but as a journey with 'ups and downs' along the way. Although this was never asked about directly, it was clearly important because all parents brought up 'acceptance' and wanted to share their individual stories of this process.

3.2.4 Overcoming Disability and Taking Control

Every parent spoke of the importance of providing their child with the same experiences and opportunities as every one else:

I don't want her to be stopped doing things because of her disability. I want her to be able to experience everything that everyone else can experience (Theresa)

There was a sense of not letting disability hold the child back, and of a responsibility to provide 'normal experiences'. For all parents this involved ensuring their child could participate in every day experiences. Several parents spoke of a need to overcome physical and societal barriers. For Julia, it appeared to be important to push the boundaries of what can be achieved, and to not be defined by the limitations of having a disability:

We've done things that a lot of parents wouldn't, that I think wouldn't do (pause) or wouldn't contemplate doing because someone had a disability. But I've never let that stop me at all (Julia)

Both Theresa and Julia spoke fondly of 'magical experiences'. There was a sense from their language of a feeling of disbelief and complete joy at giving their child happiness through swimming with dolphins or scuba diving. This was captured most strongly in Theresa's account:

I cried, I always cry, just her face, being able, for her to be able to do something that everybody else could do, you know, that experience, that she'd always wanted to do. She was squealing with delight, she was just, so happy, it was amazing (Theresa)

On reflection, it appears that these experiences meant more to parents because of the effort and barriers they overcame to achieve them, and because these children were often restricted by their disability and missed out on every day experiences. For Theresa, there was a sense of overcompensating to try and manage her own personal struggle with being unable to 'fix things':

I want to solve everything, and as a mum you want to wave a magic wand and make it all go away and you can't, so the next best thing is to try and do, give her a whole range of experiences (Theresa)

Like Theresa, for many parents there was a need to take control of disability. However, for Rachel, Linda and Laura this manifested itself by supporting their children to achieve their 'full potential' and 'be the best she can be'. For some, this involved providing assistive technology and equipment to maintain posture. For Laura, this involved hours of practice and perseverance:

We used to, at the bottom of our stairs, we'd have Wade sitting on the bottom step, and we used to practice, you know hours at a time, getting him to stand up (Laura)

Several parents became very proactive in searching for information and appeared to become an expert in disability. The strongest indicator of this was Laura, who returned to university to 'concentrate on disability modules':

Because I wanted to learn as much as I could, about disability and how, well what goes on. Urm (pause) and yes try and learn as much as possible about it, so as a parent you're clued up (Laura)

These different reactions appeared to be a way of coping within an uncertain situation, by trying to gain control of the uncontrollable.

What is striking about these accounts is the lengths parents went to provide for their children and the strength of parental love and devotion shines through. This is succinctly captured by Theresa:

My kids have always been the most important thing in my life, I'd do anything for them (Theresa)

Throughout this theme was the sense of parents seeking to take charge and overcome the barriers and limitations created by disability, to not allow their children to be defined by disability. This often came at the cost of sacrifice to parents own physical and emotional health. However, all parents were able to find the strength and take control, to move forward in their lives.

3.3 An Emotional and Uncertain DBS Journey

Parents experienced a range of emotional stressors and psychological struggles from starting to contemplate DBS and throughout their hospitalisation. The process of decision-making was fraught with uncertainty. This struggle with uncertainty continued throughout hospitalisation, and parents experienced extreme emotional reactions. Parents used emotion focussed coping strategies and sought support to cope. Overall DBS appeared to be experienced as an ongoing and turbulent journey.

3.3.1 Facing the Uncertainty of Decision-making

Deciding to have DBS was a very 'big decision' and a difficult process for all parents. For all parents this process was made up of different stages. Every parent spoke of considering the risks of surgery, and for many there was a fear of the child becoming 'more damaged':

*I thought that she, you know cause its brain surgery at the end of the day isn't it. I thought she might come out and she wouldn't be able to speak, she wouldn't be able to see, you know. I think my main fear was that they would do something else to make her more disabled (**Rachel**)*

Many parents described overwhelming fear because of the meaning of brain surgery. Although unable to articulate why, there was a sense that brain surgery was more of a risk than other surgery, and some parents didn't want to verbalise their fears:

*Because it was you know, we don't wanna, something as big as surgery, brain surgery, you, you don't want to think ok, you know what... It was a big decision to make (**Caroline**)*

There was a contrast in decision-making between parents whose children were more physically able and higher functioning, and those who were more severely restricted by disability:

Wade had a reasonable quality of life before... And urm the fact that before DBS he could walk, you know he had his intelligence and that sort of thing, and had reasonable speech so the idea that any of those could be affected in a bad way was probably one reason why we took a while to decide (Laura)

So ultimately in my mind set what have I got to lose... from the kind of physical point of view even if the surgery went wrong Ivy wasn't going to lose anything, because she couldn't do anything (Natasha)

It appeared that there was a sense of weighing up how much the child has to lose, and this impacted on the time to make a decision and parents struggle with the decision.

The lack of guarantee for a positive outcome or certainty of how the DBS would change the child's dystonia made the decision more difficult for every parent:

Urm (pause), and not knowing, what, you know nobody kind of knew what it was going to do for him, cause there's no real, its not that simple is it, its so complex (Linda)

This lack of certainty of what could be achieved was compounded because each child's disability was completely different, and because of a lack of understanding of how DBS impacts children with secondary dystonia:

Because it's secondary, there's a lot more questions, is it worth doing, you can't give me any definite answers... I don't know about anyone else, but for

me that was the biggest thing ever, I'm doing this but is it actually going to work

(Natasha)

However for all the parents there was a sense that the hope for a better life outweighed the risks, and the surgery was 'worth a chance' if it 'would give a glimmer of making life easier' for the child:

Hope of a positive outcome, was, overweighed any other objections I think

(Theresa)

In this process it was also important for parents to involve the child in the decision if the child was old enough, and for some to rely on professional advice which was perceived as 'expert':

It's her brain, it's us making that decision for her, she needs to have some say in it, as best she can **(Caroline)**

And he was really encouraging this was what Wade should do (pause) because you know the effects it had had on this girl, and (pause) yes he sort of put a strong case forward, so we decided ok **(Laura)**

These quotes highlight both the vulnerability of the child, but also the power of professionals in this decision.

It was clear for all parents that there was uncertainty in this decision. However, there was stark contrast in parents' ability to accept this uncertainty:

So there was no doubt that it was the wrong thing to do, even weighing up what risk there is. We didn't really have any doubt, and (pause) we've always said that you know we don't really know what the future is going to be for Billy
(John)

I've said to my husband, 'are we doing the right thing here, because I really, I really don't know', and I could have been persuaded one way or the other. Because, I just, I don't know, and still know, still don't know if I've made the right decision (Rachel)

Rachel's extract clearly demonstrated her struggle with the uncertainty of decision-making. Her switch to present tense, further demonstrated how she continues to be affected by this the burden of responsibility and difficulty accepting she made the right decision. The impact of uncertainty on the struggle of decision-making is clearly demonstrated by Linda's experiences:

But I don't think Philip could have done without the DBS knowing after the first time how it had really worked for him. I know the first decision probably was a bit, it was worrying, because it is difficult to understand everything, but the second time I would not have even, it were just definitely yes (Linda)

Sadly due to complications Philip needed to have the DBS taken out and re-implanted. This knowledge of how DBS could benefit Philip, provided Linda with a new perspective and certainty, and made the decision easier.

3.3.2 Struggling with the Unknown and Unexpected

Several of the parents found the DBS experience challenging, because of a struggle with the unknown and difficulty coping with the unexpected. Some parents experienced uncertainty of what would be involved in DBS:

I didn't know how long she'd be. You don't know how long someone will be recovering afterwards. She could get infection or temperature, or anything like that (Julia)

For others there were unexpected complications after the DBS procedure. This left Laura overwhelmed by emotion, and searching for an explanation:

*Yeah that was worrying, that was, yeah, that was upsetting, that was upsetting.
Urm (pause) cause I suppose I can't quite understand why, you know why, why
is he not recovering as quickly as other children do, or as quickly as we were
told that he would (Laura)*

Laura's emotions appeared connected to her struggle with not knowing what was happening, and how things were different to what she had been told to expect. This experience seemed particularly difficult for Laura, as she switched to the present tense during the interview, as if she was reliving the emotions. Similarly Linda experienced fear because Philip was very unwell and the medical team didn't understand what had happened:

*Well, it were, really frightening, because, nobody knew why, you know, at that
particular point, before they realised the wire had snapped, because I don't
think one has never snapped before, it (sigh) (Linda)*

Sadly for Linda she continued to live in a state of unknown because the DBS system was removed and Philip's dystonia deteriorated:

*because we didn't know what we were gonna expect, you know how well he
were gonna be, or, you know, he had a lot of things wrong with him, that he
didn't have before, so you know with this wire snapping, everything sort of
went, it (sigh) we didn't know what was going to happen basically, or where we
were going to get him to, and neither did the team, so that was were kind of
really a worrying time (Linda)*

In this striking extract Linda was struggling with many uncertainties and, there appears to be a link between emotions and uncertainty. This suggests how for some parents the uncertainty

continued beyond the hospitalisation procedure. For Linda, her uncertainty seemed greater because even the medical team were unable to find answers, and there was a sense of them entering unknown territory because a wire had 'never snapped before'. This highlights how DBS research and understanding is still in its infancy, and this can impact on parents' experiences and uncertainty.

Furthermore parents spoke of how it was easier to cope if you were prepared because you 'you know what to expect', and you can 'still yourself'. This attitude is evidenced by Linda's experience of the second DBS surgery:

*And I don't think I felt actually, as worried, because I just felt it was what he had to have, you know we'd worried the first time because of the unknown and not sure what to expect (**Linda**)*

By taking away the uncertainty and knowing what to expect, Linda's worry was absent.

However, not all parents experienced this struggle. In complete contrast John was able to embrace to uncertainty of DBS:

*So the whole being in hospital, the whole DBS thing was just another adventure, we don't know how its gonna pan out, you don't really know where its gonna take you,... but its in the lap of the gods as it were, as to what the outcome will be, so therefore it's an adventure (**John**)*

It appeared as though John's attitude of 'life's an adventure' helped him to accept the uncertainty and not be burdened by difficult emotions during DBS.

3.3.3 Emotionally Overwhelmed by DBS and the Hospital Experience

All parents spoke of a range of difficult emotions experienced on the day of the surgery and hospitalisation. All parents struggled with the fear and worry of brain surgery, particularly that something could go wrong leading to strong emotional reactions:

*Just the fear of the surgery, that something would go wrong... mean there's loads of fears, you try not to let them in (**Theresa**)*

*The morning of the surgery I remember being terrified, and nearly just saying I don't want to do it, I didn't want to do, just purely for the fear fact (**Theresa**)*

There was a sense of trying to control the worries, and that these thoughts were too unbearable to 'let in'. These thoughts were so terrifying that Teresa nearly changed her mind and several parents reported being unable to leave their child throughout the admission.

For many the anaesthetic was the most 'horrendous' and 'distressing' experience:

*Horrible, horrible feeling of (pause), at that point you want the distress to stop, but as soon as she's obviously put out it's a really horrible feeling to see your child kind of asleep but not with it, in that state, and obviously from an emotional point of view that's when we both kind of broke down (**Caroline**)*

For Caroline, her distress recalling this moment was so vivid that she was unable to finish her sentence, and there was a sense of not being able to find a word to articulate how she felt. It was apparent that Caroline needed to stay strong for Emily, and that the emotions became too much to cope with once she was put to sleep.

For others, this moment seemed to be distressing because of feeling alone, and unprepared for the day ahead:

And then to go off, and just be left. That, I think that's the worse thing, I kind of walked up the stairs and just thought 'oh my god, I've gone from that, to now what I am going to do for the rest of the day'. And I was on my own which I don't think helped (Rachel)

There was a sense of in this moment being overcome by the reality of what had just happened, and feeling at a loss of what to do next. This experience of waiting whilst the child was in surgery was particularly difficult for many parents. There was a sense of feeling helpless and powerless, and the only option was to wait:

It's that waiting, that hoping that you're gonna hear something, or not necessarily hear something. I think we went back up to the ward, I felt absolutely exhausted (Caroline)

For Julia, it appeared as though her worries were so intolerable to her, that she needed to keep busy as a way of avoiding her thoughts:

I had all the information and research, what to do and where to go, so that I never had a minute where I was lying there you know, worrying really. (Julia)

This avoidance of emotions was still evident in the interview, and there was a sense of minimising during the interview, as if the emotions were too painful to recall. There was also a cognitive dissonance between how Julia described herself as not stressed, and her behaviour of 'compensating' so she didn't 'really really really worry'.

The moment of recovery was also particularly important, and the overwhelming relief described by several parents. For Natasha this was accompanied by sheer joy:

I was just a mess, I was like, just wow (smile). It was like having her born all over again, that sense of, when she first came out and she cried, that my

baby's ok... It was that utter relief that she was ok. I know I took loads of pictures of her, and I didn't even text anyone straight away, I just wanted to enjoy it (Natasha)

This moment meant so much for Natasha, because unlike other parents she described thinking 'Ivy was going to die'. This experience was as significant and emotional as the day Ivy was born. This simile conjures up a feeling of a new beginning, further emphasised by her attempts to capture the happiness of this moment.

In contrast, for other parents the hospital experience was difficult because of disruption caused to their normal family life. It was important to try and manage the siblings' emotional reactions to DBS. Natasha felt her children 'picked up' on her feelings and this showed in their behaviour. However, she spoke of being unable 'to give them what they needed', and would have liked more support:

It is a lot, it's a huge amount for a family to deal with... they've struggled a lot, there's like no support for them at all, that's something that I've found more difficult than anything (Natasha)

3.3.4 Coping with DBS - 'it's the people who get you through'

This theme captures parents' positive coping strategies and the positive experience of care received by parents. The notion that a positive attitude was a choice was repeated throughout interviews, as many parents appeared to believe they had power over their attitude:

I think after I thought it was like giving myself a kick up the backside, you know if you're not going to be positive, you're not gonna cope, were not going to get anywhere (Linda)

Linda reflected on how the presence of negative thoughts shocked her, and she chose to be positive, believing a positive attitude would help get Philip to make progress. On reflection there were similarities between this coping frame of mind, and the coping frame of mind parents adopted in reaction to diagnosis. This choice seems to provide parents with a power over their thoughts, that helped them move on from adversity. For others, like John, earlier stressful hospital experiences served to almost buffer against stress, and gave him a different perspective and ability to cope:

I had six hours sat in a corridor not knowing if they were alive or dead, so nothing can top that. You know literally nothing can top that (John)

Some parents also appeared to have a decision between struggling with uncertainty, and accepting there was 'nought we could do about it' during the wait for the surgery:

There was nothing I could do to help the operation I was just redundant to that. So, I made it me time that day (Julia)

Julia's choice to accept her powerlessness, seemed to allow her to move out of this position of waiting helplessness, to taking control of her day and making it 'me time'. There were further examples of parents displaying control over their reactions. Parents displayed evidence of the cognitive ability to find something positive out of the DBS experience. For the parents with daughters the hair shaving was a pivotal movement, and although this was often a devastating experience for parents, they were able to make it a positive experience for the family, or treated it as an opportunity to raise money for charity. I wonder if these positive attitudes, helped to protect parents at times of adversity and challenge. Those parents who were able to see the positive, and demonstrate control over their attitude and reactions, appeared to have struggled less with the uncertainty and perhaps felt less helpless and out of control during the process.

Other strategies described as important were taking a break and getting out of the hospital environment. For parents there seemed to be something about 'escaping' the 'intense environment' and participating in different activities (e.g. going for a walk, out for dinner). For many it was very important to be busy and preoccupied as a way of coping. Julia planned in advance a series of activities to keep herself 'planned up and busy and occupied' on the day. This seemed to help provide a distraction and there was a sense for many of trying to avoid ruminative worry and 'not letting your brain do loop the loops'.

In contrast to not engaging in worrying thoughts, Natasha spoke of preparing herself for the loss of her daughter:

I prepared myself for the fact my child was going to die and I was going to go home without her (Natasha)

This reaction seemed to be a form of self-protection so that the reality of facing potential bereavement would be less difficult.

Everyone spoke of the importance of emotional support. For some, this involved meeting parents in similar situations which provided a normalising effect and helped parents not feel so alone:

actually you've just had the same operation as my daughter, I don't feel like so alone anymore (Natasha)

Others relied on partners and family, and there were many accounts of this support being invaluable to being able to cope.

The overwhelming positive of the experience for most parents was the support received from staff, and all were appreciative of the medical team at different stages of the process. Their

role was perceived as positive because of the many roles they played: emotional kindness and support, experts in care, always being available and taking over responsibility:

to have that personal a bit more of a personal approach, and to be that little bit more warm, a little more caring, to take the time, is massive because it makes you feel, it makes you feel that you're being looked after, it makes you feel that Emily's safe and the care that she is receiving is there (Caroline)

It seems these experienced help parents to have confidence in staff and feel looked after. Many parents appeared to have more confidence in the expertise of the specialist staff team, and Linda felt nurses had a better 'understanding of things' because of the complexity of disability. This allowed parents to hand over responsibility and reduced their anxiety, and always feel supported.

The role of staff was captured most succinctly by Theresa's description of 'it's the people who get you through'. There was a feeling that without staff support you would be alone and this could really break you:

I wouldn't have liked to have felt I was just on my own that could really break you (Linda)

Interestingly Philip experienced complications resulting in two further surgeries, and therefore Linda had the most contact with the team. Her experience of care, was viewed as overwhelmingly positive despite experiencing most set backs and the longest process.

There appeared to be a disparity between parents' experiences, as evidenced by the stark contrast between Linda positive experience of limitless support, and Rachel's experience of the 'facelessness' of care. What is clear however is the positive experiences of staff support and perceptions of the staff as helpful and caring, helped parents to cope better, and overcome the many challenges of the DBS experience. In contrast for Rachel this was

lacking, and one can hypothesise this contributed to her feeling alone and lacking confidence in staff and potentially feeling alone in her responsibility for the decision.

3.4 The Experience and Perception of Change

This theme captures how parents experienced change from the point of initially considering DBS, to their perceptions of change after DBS. Parents felt a strong sense of hope, but tried to have realistic expectations. All parents experienced some positive changes after DBS. However there was disparity between the 'life changing' impact of DBS, and disappointment of subtle changes. All parents sought to assign meaning to these changes, and evaluate if DBS was worth it, whilst negotiating to the new challenges and compromises of life with a DBS System.

3.4.1 Hope and Realistic Expectations of Change

Parents hoped for 'more control' and reductions in dystonic spasms:

her arm, which is (pause), the worst part of everything for her, would (pause) be significantly better...from being twisted and horrible, that it would be more relaxed and in a better steadier position, constantly. (Rachel)

It was hoped that these physical changes could lead to an improvement in participation in activities of daily living, and QoL. This could be in the form of better motor control so Wade could hold a cup, or improve Philip's walking, to an ability to concentrate better at school without needing to 'constantly struggle with those movements'.

In the children whose dystonia caused pain, parents hoped that the DBS would help alleviate some of the pain, and for many there was a sense of preventing future deterioration and providing best outcomes for the future:

I was worried, and she knew, losing range of movements, once those movements are lost, you can't get them back (Theresa)

Parents all spoke of having realistic expectations and not expecting 'miracles' or 'massive leaps'. Although having specific goals, some parents, specifically John and Caroline, were just hopeful that 'some improvement would be great' and that 'any positive that came out of DBS made it a worthwhile thing'. It appeared as though the parents of children with more severe disabilities and dystonia that affected their entire body were able to just hope for a more generic positive change to help improve the child's QoL:

To make the quality of her life, a little bit better. Urm and I think that's the point isn't it, its about improving her quality of life, even if its by a small amount, to make things a little bit easier for her (Caroline)

In contrast parents of children with less severe dystonia had more specific hopes and expectations of surgery (e.g. improvement in fine motor skills, reduction in spasm in one arm as described earlier). It is possible that for parents of more severely disabled children, a small change had the power to create a huge improvement to life.

but then the small things when you're dealing with a physically challenged child, the small things make a huge difference (Natasha)

Although all parents discussed the importance of 'being realistic in what you want to achieve', many spoke of 'wishing, hoping something's gonna be more than it is'. In contrast to other parents, Theresa initially 'thought it would switch off the muscle spasms, the tightness, the pulling, the flailing of the arms'. She described thinking 'it would be like Parkinson's, it would be a switch'. Theresa emotionally recalled the moment the staff explained the realistic outcomes of surgery:

We were both excited by it, the thought that we could make her life so much more, and improve her so much. And then for someone to say 'look actually its not going to be anywhere near what you're thinking' was really difficult, really really hard, I think we both cried (sigh), oh dear. But you know, its that bit of hope that someone can do, you'll take anything, but when your expectations so high, and then you know its slashed in half immediately, its really difficult, really difficult. (Theresa)

There was a sense of Theresa's desperation for change and of her hopes being shattered. Her choice of language of 'slashed' conjures images comparable to being physically assaulted and exposed, and I wonder if that was how she felt in that moment. I was struck by the power of medical professionals to drastically alter a person's life and expose and shatter their dreams. Theresa then embarked on a process of 'licking our wounds' and readjusting their expectations to set goals that 'seemed a bit trivial'. The initial expectations perhaps impacted on Theresa's perception of success.

3.4.2 Significance of Positive Changes – 'it's everything'

All parents experienced positive impacts of the DBS surgery. There were copious examples of changes to a child's movement disorder, therefore the focus of this theme will be on the meaning attributed to these changes, and how parents experienced these changes. Parents noticed improvements in posture, reduction in dystonic spasms and muscles were 'more relaxed' and 'much looser'. Parents described examples of how their children were not in as much pain and had more control of their whole body and particular limbs. As is evident in Rachel's extract these changes were hugely significant to families:

Probably one of the really big things is that she's not in half as much pain as she was before. Which is huge, and very significant (Rachel)

For many, these changes led to an improvement in QoL, more independence for the child, more participation in activities of daily living and positively meant children were not as heavily reliant on medication. In a vivid description Linda considered how Philip's QoL would have been without DBS:

I wouldn't like to think what his quality of life would have been without having it, because when he didn't have it was not good, at all, for him, well and for us for that matter (Linda)

Most parents described how their goals were achieved, and for some were surpassed. For many there were even unexpected changes. Five parents reported an improvement in their child's speech. There was a consensus of children 'speaking much more fluidly', 'so much clearer' and 'in Ivy's case 'vocalises a lot more':

was just like wow, this is, this is, and we were told at the time not to expect anything from a speech point of view, and it absolutely was, her, her words were so much clearer. So that was a massive shock to us, and an added bonus (Caroline)

As demonstrated by Caroline, this change was so unexpected because the medical team told parents this is something that is rarely impacted. There was a real sense of awe and amazement, and this conveyed the huge meaning of these changes for many families. There was a sense for many of DBS exceeding expectations, and the changes meaning more because the changes were so unexpected.

Rachel and Natasha also spoke of unexpected improvements in their daughter's mood and anxiety:

Because she's like a completely different child, although still very Ivy, her personality is a lot more obvious now she's not having to deal with 24-7 spasms, she sleeps better so she's happier (Natasha)

She likes the fact that she knows she's less anxious about things, um and urm, I think she feels a little more confident and capable if that sounds right (Rachel)

It seemed as though alleviating some of the spasms has helped Ivy to feel happier, and allowed Megan to feel more confident within herself. The visible nature of disability was particularly salient in Rachel's accounts, and it is possible that having more bodily control has helped Megan to feel more 'normal'. For many parents, there was something about a visible difference after DBS. Natasha talked about a paediatric doctor coming to see Ivy after DBS:

I think for me, that to me is a really positive outcome from having the DBS if someone whose known her from day dot, can see the change in her, its not been for nothing (Natasha)

The changes appeared to mean more if they were noticeable by others. Again this returns to the idea of the social context of disability, and the potential mediating impact the perception of others has on Natasha's perception of change.

Although all parents reported positive changes after DBS surgery, the strongest and highly emotive narratives of the significance and meaning of change were from John, Caroline, Natasha and Linda. For these families DBS appeared to have a wider impact than the motor abilities of the child and helped families to get 'more of a normal life'. Beyond this impact on family life, there was a remarkable sense of allowing the child to participate in family life and wider society by removing some of the barriers created by disability:

Instead of being sat in his wheel chair at the side of a hall, or in his walking frame just scooting about, he can now be part of things, he can now communicate (John)

Children were able to share their personality and 'share his sense of humour', and there was a sense of elation at DBS unlocking the true potential of the child:

You were starting to see 'wow Emily this is what you are capable of doing', this is amazing (Caroline)

There was an overwhelming sense of DBS helping children to live rather than simply existing in these narratives, and giving parents a greater connection with their child. The post DBS changes also appeared to be perceived as significant because of the meaning parents attributed to these changes. The changes were perceived as meaningful because it was the first time children were able to do things. In this emotive extract John captures the meaning behind the impact of DBS:

Phew (exhale), ecstatic, ecstatic (tearful), yeah yeah. Its just after the surgery, he gave me a kiss, and that's the first proper kiss he's ever been able to give anyone, sorry I get a bit (crying) (John)

John became so emotional explaining how much DBS has changed his life that he was unable to speak and started to cry. Similarly Natasha and Caroline were 'just amazed' when their daughters were able to eat solid food, exclaiming 'we never thought we'd see Ivy eat'. These changes also appeared to have more significance because they had been told their child would never be able to for example speak or eat solid food:

Its amazing, its like, and I say its like a new born baby, its not, its more than that because this is a kid that I got told wouldn't do it, and now she's had something to help her she is doing it (Natasha)

DBS surgery appeared to lead parents to drastically change their beliefs and assumptions of what their child was capable of. It seemed that the parents who were hoping for any small change, perceived the surgery more positively because the outcomes surpassed their expectations:

That's why I feel so positively about it because I was expecting to get nothing from it as well, but we got lots from it (Natasha)

3.4.3 Making Sense of 'Quieter' Changes than Expected

There was a feeling amongst some of the parents of disappointment at their hopes and goals not being achieved, and of how much dystonia continued to impact their children. Although Julia could see positive results from the DBS, her daughter had a different perspective:

But Imogen can't see any difference, well not saying any difference, she doesn't think it's made any difference and sometimes she thinks its worse. And now she says the right arm is doing it, which it wasn't before (Julia)

Julia seemed to be struggling with this, as she almost tried to convince herself that her daughter can see some difference. Some parents were unsure if there had been an improvement and were dependent on medical professional opinion. Many parents tried to make sense of the lack of change by attributing it to the unpredictable nature of dystonia which is impacted by different factors (e.g. growth spurt, hormonal changes, medication changes, stress):

*Also, being thirteen she's going through massive hormonal changes, um growing, you know spurts, and I think, we've also, she was on baclofen, well she is on on baclofen... we've tried also since then to cut back on that, so you know its very hard to gage exactly where you are **(Theresa)***

It felt like parents needed to find meaning, as a way of perhaps justifying to themselves their decision, and possibly helping them to cope with a disappointing outcome. Parents seemed to be searching for answers. Because of her daughter's disappointment, Julia engaged in a process of almost rationalising her daughter's perspective:

*But, its, it's so subtle, that if you got any spasm you've got spasm, whether it's big or small **(Julia)***

It appears as if this was understood by Julia in terms of her daughter still having a disability, and that small changes did not alter the fact she was disabled. In contrast, what seemed most important to Rachel was the visibility of disability:

*Probably one of the really big things is that she's not in half as much pain as she was before. Which is huge, and very significant. But again, its, its, its hidden you see *(nervous laughter)* isn't it. Not everybody knew about that anyway before. **(Rachel)***

She perceived the changes to be hidden, and this was almost enough to dismiss the significance of the positive changes of pain reduction. Like Natasha in the previous theme, it was important for Rachel that people noticed changes, because of her perception that people 'judge' her daughter by 'how she is walking and her arm':

For people who didn't know she was constantly in pain before, the fact that she's now not, they didn't know anyway. So they wouldn't see that as a

change, and not everybody um takes as much notice of speaking and that sort of thing, so they don't notice (Rachel)

It is possible that it was also important for people to notice, given the uncertainty Rachel continued to feel about if she made the right decision. People seeing a change, perhaps would have provided external validation that she made the right decision. Notably the parents with more able children (e.g. Rachel, Laura) appeared to be most disappointed with the outcome, and perhaps were expecting more specific changes that were not met.

3.4.4 Compromise of Change and an Ongoing Struggle

All parents described ongoing challenges of negotiating and adjusting to a life with a DBS System. Parents spoke of the difficulties they experienced throughout the gradual setting adjustment process. Many parents described 'waiting for a change', 'wanting things to be quicker', and the 'whole process being a lot slower than anticipated really'. Most struggled with the 'ups and downs' in dystonia presentation, until the medical team 'got the settings right':

Sometimes you go forward and then back a few steps, that then worries you, because you think 'oh gosh he were doing that and now he's gone back'
(Linda)

For many this lead to anxiety of if the child was on the correct settings, and even if the system was functioning correctly:

I mean we've had set backs as well, we've had issues, we've had times when she's been unwell and had dystonia which has been ridiculous and like I've actually checked to make sure the batteries still on (Natasha)

Parents appeared to experience a lack of certainty of how long changes could last, and that dystonia remains unpredictable in its nature. This narrative was most dominant in Laura and Caroline's accounts after complications with the DBS system. Caroline captured this shock and sadness experienced at this time:

Then obviously recently to know that she had to go under surgery again, was an absolute kick in the teeth to be honest. I think we were shocked because it had only been such a short amount of time, and something's gone wrong, its quite rare, that kind of it was one of those 'why us' (Caroline)

Additionally DBS often led to new difficulties and challenges in the child's dystonia:

And we've got different problems now to the ones we've got before...now her arms are often flailing to the side, still held quite high... (Theresa)

There was a sense for many of the compromise of change, because the dystonia improved in many ways, but this was accompanied by negative physical changes. Many parents also spoke of the negative physical changes because of scarring or the visibility of the DBS battery:

I suppose he has got poor body image if he wants to always cover his body up. So it's a bit of a shame that he's now got this scar that he is embarrassed about, urm, that hasn't faded (Laura)

Sadly there was a sense of DBS adding to the child's difficulties and negative self concept. The DBS may have created another layer of visible difference for these children, when particularly for Rachel she was hoping her child would look more 'normal' after the surgery. In contrast to the other parents, Rachel continued to question her decision and struggle with the responsibility of it. In this extract she seemed to be mourning the loss of her 'perfect child', and voiced a sense of regret:

Megan had the most beautiful white long blonde hair, and it hasn't come back the same. Its not like that, its different and its not as nice and that's terrible, terribly shallow...So (pause) that to me has completely changed her, cause she's not the same girl (Rachel)

Megan seems to have become a different child to Rachel, and although not finishing her sentence, she later described how she wouldn't make the same decision again. A further compromise involved the extra responsibilities of charging up which children found 'frustrating sometimes'. However, parents spoke of how they've 'made it a family routine' and 'a bit of relaxation time'.

Overall parents experienced ongoing challenges and compromises after DBS. Life was therefore still difficult for families, and even after DBS the child was still disabled and restricted by dystonia.

3.4.5 A New Perspective Looking Forward

All parents searched to assign meaning to their DBS experience, and evaluate whether DBS was worth it. All parents engaged in a cognitive process of considering what life would have been like without DBS:

I think if she hadn't had it, what would life be like now, I'd actually probably be struggling to look after her (Natasha)

This led the majority of parents to be overwhelmingly positive about DBS and exclaim 'I feel like it's been really worth it'. Others, like Theresa weighed up the balance of the positives and negatives, to reach the conclusion that 'the positives far outweigh the negatives'. Sadly Rachel and Laura had a different perspective of DBS. For Laura 'it was worth a go' because without trying they would have always been wondering 'whether it could help or not'. Rachel

was the only parent who voiced a sense of regret and that she wasn't sure she would 'make the same one again' with 'the benefit of hindsight'.

Regardless of outcome the DBS process significantly impacted all parents, who reflected on gaining a new perspective of themselves, their child and their future. DBS seemed to provide parents with a new perspective of their child. Parents described viewing their child as unique now 'she's bionic' and more mature and 'wordly-wise' after coming down to London. Many parents also described how DBS has made them proud of their child. Parents emotionally described feeling amazed at their child's 'strength and determination' in coping with the surgery. Parents were proud of their children for 'staying positive all the way through' and 'taking everything in her stride':

So proud of her, so amazed by how she absolutely deals with it (Caroline)

Parents also spoke of how they have changed as people and parents. Parents described being 'more tolerant' and becoming more of a 'realist'. For Natasha she gained a greater perspective on life:

Urm and I'd, like when if I moan now that things are bad, its urm, you think why because they're not anything like they used to be (Natasha)

Many parents spoke of becoming stronger throughout the process:

I think it's certainly makes you stronger, because you've not really got a choice. Urm, and I think it makes you cope a lot better, don't really know how to explain it. Urm, I just think it does make you a lot stronger person. (pause (Linda))

There was a sense of parents' self-identity changing as they reflected on becoming a stronger and 'better person'. It appears as if by overcoming the challenges and 'trauma' of DBS surgery and for some the initial diagnosis of disability, parents identified themselves as

stronger. This sense of strength seemed to have led to identification of themselves as experts in DBS and their own recovery process. There was a sense of personal growth after DBS, as many parents wanted to use their experiences to give back to other parents undergoing the surgery, and were in a unique position to share their experience of a parent of a child undergoing DBS:

*I've been through something that I can talk to people of all different ages of all walks of life and just try and do something to positive for them, through something that wasn't (**Natasha**)*

Parents also gained a new perspective of their future, and many talked about how the child wasn't on 'perfect setting for her' or 'anywhere near maximum'. This allowed parents to feeling 'excited' because 'there's more to look forward to'. For all there was a sense of being at the 'very beginning' and 'early on on the whole DBS Path', which lead to a sense of optimism and hope that 'there's going to be an ongoing impact for years to come'.

Chapter 4: Discussion

This qualitative study primarily explored the lived experiences of parents of children with secondary dystonia. Secondly, the interviews with parents were analysed to capture the experiences of children with secondary dystonia through the sense parents made of their child's experience. The aims of the study were to explore the following questions:

1. What are the lived experiences of parents and children with secondary dystonia?
2. What influences DBS decision-making, and how do parents make sense of their decision?
3. How do parents experience and manage the DBS process?
4. What is the impact and meaning of post-surgery change for parents and children with secondary dystonia?

In this final chapter, how the results answer the research aims will be discussed. These findings will be discussed in relation to existing literature, and how they inform psychological theory and understanding. Next, the strengths and limitations of the study will be considered, and the clinical and future research implications will be presented. Finally, personal reflections of the researcher and conclusions will be made.

Overview of Findings

The analysis revealed four superordinate themes:

1. A Difficult Life with Disability
2. The Meaning of Disability and Normality
3. An Emotional and Uncertain DBS Journey
4. The Experience and Perceptions of Change

4.1 Addressing the Research Questions

4.1.1 Question 1: What are the lived experiences of parents and children with secondary dystonia?

Life was experienced as difficult and different by parents and children. Dystonia exists within a relational context (parent-child), but also within a wider family and societal context. Social comparison and being treated differently at many different levels in society (negative reaction, stigma, and discrimination) contributed to how parents and children experienced self as 'different'. The concept of control was central to lived experience and there was a sense of parental conflict in the meaning of normality and difference. Overall this theme considers dystonia a multifaceted and complex phenomenon that significantly impacts the lives of children and parents.

4.1.1.1 An Objective and Observable Body

Parents' perceptions of their child's physical experiences of secondary dystonia were consistent with the extensive literature measuring the physical impairment of dystonia. Children experienced involuntary movements, which were exacerbated by emotion and voluntary action (Albanese et al., 2013; Sanger et al., 2010) and, for some, resulted in abnormal postures (Egmond et al., 2014). These movements varied in severity and there was diversity in which area of the body was affected (Roubertie et al., 2012).

4.1.1.2 Subjective Experience of Body

Parents perceived their child's subjective experience to be trapped inside an uncontrollable body. Children tried in vain to overcome the power of dystonic spasms and control these movements. However, dystonia was experienced as inescapable. It's unexpected, unpredictable and changeable nature was also a struggle for children. This insight adds to the existing understanding of secondary dystonia, and suggests that the physical features of

constantly changing muscle tone and unpredictable spasms were experienced negatively by children. This sense of being trapped and being unable to control an uncontrollable body seems to be unique to secondary dystonia, and has not been previously reported in the dystonia or physical disability literature. Children's struggles were also made worse when they had an the understanding of how their body should function resulted in feeling restricted and different. Consistent with the work of Merleau-Ponty (1962) sense making drew on the body as an objective and physical entity, with subjective meaning in relation to the child's perception of self.

4.1.1.3 Social and Emotional Impact on Children

Parents described how secondary dystonia had a significant impact on their child's social and emotional well-being. Children were unable to participate in activities of daily living and engage in age appropriate social activities, leading to social isolation. This is consistent with the child and adult literature demonstrating an impact on HRQL (Bakowki, 2010; Lim, 2007; Page et al., 2007). Children were aware of their limitations and being visibly different through a social comparison process with siblings and peers. Festinger (1954) proposed how individuals use comparisons to describe and evaluate their own situation. In dystonia, this perceived difference led to frustration and in some cases impacted on the psychological well-being of children. For more able children, this awareness seemed to impact on their developing sense of self as they and became tied to an identity defined by disability, leading to social withdrawal and feelings of depression. In adults, dystonia has shown to have a negative impact on body image and self-esteem (Jahanshahi & Marsden, 1990) which contributes to depression (Lewis et al., 2008). This is the first study to capture a similar experience in children with secondary dystonia, and adds to existing literature demonstrating how dystonia can have a negative social and psychological impact on children.

4.1.1.4 The Social Construction of disability

Beyond feeling different in comparison to others, the child's sense of difference was perceived by parents to be exacerbated by the negative reactions of others (staring, negative comments, bullying). This is consistent with findings in the adult dystonia literature that disfigurement and negative body image are associated with a higher perception of stigma (Papathanasiou et al., 2001). Parents experienced sadness that people cannot see the child's personality and cognitive abilities hidden behind the physical disability. Families also experienced negative societal barriers to participation in activities, which was very distressing for parents. These experiences serve to demonstrate the power of society and the significant impact that labelling, prejudice and assumption-making can have on a child's developing sense of self and ability to participate. Historically, literature has ignored the burden imposed by negative societal attitudes (Leiter et al., 2004; McKeever & Miller, 2004). However, this theme suggests secondary dystonia is a socially constructed phenomenon, and consideration of societal context is essential to understanding the lived experiences of children and parents.

4.1.1.5 The Lived Experience of Parents

Parents described the physical impact of disability through caring and assistance with everyday tasks. Socially, parents described feeling restricted by disability, and gave examples of being unable to leave the child, go on holiday and have time to themselves. Parents made practical, financial and social sacrifices, and there was a sense of parents own needs being either deferred or denied. This impact and response to parenting a child with disability is consistent with qualitative studies exploring experiences of caring for child with CP (Davis et al., 2010).

The concept of difference was also central to parent experience. The uniqueness of secondary dystonia contributed to social isolation, as parents perceived their experiences to be different to other parents of children with disabilities. Perceived difference therefore created a social barrier for parents, mirroring the isolated experience of children. Parents also

engaged in a social comparison process (Festinger, 1954) comparing their child to siblings and peers. This led to profound feelings of sadness and a sense of loss for how disability had stopped their children from leading a 'normal life'. For parents, difference was more than a felt sense as they assumed different parenting roles. There were many different nuanced accounts of parenting. For some parents this role was unrelenting and demanding, and experienced as being on duty all the time. These findings support the experiences of a different and difficult life parenting a child with CP (Burkard, 2013; Davis et al., 2010). Parents chose to put the needs of their child above their own. For some, this led to stress and psychological difficulties. This is consistent with the literature demonstrating mothers of children with CP experienced increased parental stress, and decreased psychological well-being (Cheshire et al., 2010; Pousada et al., 2013; Rentinck et al, 2007).

However, within the negative lived experiences of parents there were positives. Similar to studies of parenting a disabled child (Burkhard, 2013; Green, 2007) parents perceived positives coming out of the disability. Parents described a closer relationship with their child, closer family functioning and drew inspiration from the strength of the child. Parents' narratives were dominated by emotional moments of pride, joy and happiness recalling childrens' achievements and attitude to life. This supports the wider disability literature, as parents focussed on the personhood of their child rather than defining the child by disability and impairment (Green, 2003a; Jenks, 2005; Kelly, 2005).

4.1.1.6 The Meaning of Disability for Parents

A life with secondary dystonia was not the life parents would have chosen, and parents at times experienced sadness, frustration and a sense of loss. However, parents described making a decision/choice to get on with life and react positively. This choice seemed to help parents to take control, and start to accept and adjust to this unwanted life. The experience of parents was therefore consistent with features of Cognitive Adaptation Theory (Taylor, 1983) which proposed that adaptation following set backs or threatening events (e.g. CID) takes place through understanding why the event occurred, gaining mastery and control, and

enhancing the self by restoring self-esteem. Parents' efforts to gain control were a defining feature of secondary dystonia. Accepting that they could not alter or change their child's disability, parents shifted their focus onto something controllable: becoming an expert in disability, accessing specialist equipment and assistive technology, and physiotherapy to improve motor abilities. However, despite parents' relentless and often self-sacrificing efforts, their children struggled with an uncontrollable and often painful movement disorder. Parents struggled with their inability to 'fix' things, which was perceived as the role of a parent.

For parents, there was a sense of not wanting to be defined by disability or let physical impairment and societal barriers prevent children from having 'normal' experiences. Parents therefore adopted a strengths-focussed approach focussing on what the child can do. Miller (2000) proposed that individuals with CID engage in various social coping tasks to maintain self concept and function effectively. Parents' behavioural responses were in keeping with this theory. Proposed tasks were maintaining a sense of normalcy, adjusting to altered social relationships, dealing with role change, dealing with social stigma of disability and maintaining a feeling of being in control. These tasks neatly captured the challenges faced by parents. The concept of creating a 'normal life' has been well documented in the chronic health literature (Alexander et al., 2012; Bedell et al., 2005 & Glasscoce & Smith, 2011) and seemed central to parents' experiences.

Without exception, parents were accepting of their role and showed unwavering dedication and love to their child. However, parents at times struggled to accept the presence and impact of secondary dystonia on the child. The term acceptance does not capture the complex cognitive and emotional processes parents experienced. Adjustment to CID has been considered a fluctuating process over time (Wallander & Varni, 1998) and in this study adjustment and acceptance were experienced differently. For some this was automatic after diagnosis, or a process that developed over time. Others were still learning to accept, and continued to struggle with acceptance.

Overall, parents experienced conflict regarding the meaning of disability and normality. Parents went to great lengths to provide 'normal' experiences to not let their child be defined by disability, and to overcome the societal barriers and physical limitations. However, parents also accepted life to be different and described examples of adjusted parenting and family accommodation. Parents were also conflicted between loving the child for who they are regardless of disability, and wanting to 'fix' the child and remove secondary dystonia. This conflict seemed to be at the heart of parents' cognitive processing, decisions and behavioural reaction.

4.1.1.7 Understanding Secondary Dystonia

The child's experience of secondary dystonia fit within the Disability-Stress Coping Model (Wallander & Varni, 1992). The factors that seemed important in children's adjustment to secondary dystonia were: parental adjustment, parental attitude to disability, social support, (social ecological); severity of dystonia, visibility of difference (child condition); level of functional dependence on parents and disparity between cognitive and physical abilities; self concept and feelings of difference (inter-personal factors). A wider social constructionist perspective is missing from the Disability-Stress Coping Model. In secondary dystonia the meaning of difference and normality was central to experience, and social comparisons and negative wider societal reactions influenced children's perception of themselves.

4.1.2 Question 2: What influences DBS decision-making, and how do parents make sense of their decision?

For all parents the decision for their child to undergo DBS was viewed as significant, with life altering consequences for the child. Overall, previous research has not captured the complexity of decision-making involved in DBS. In health psychology, models such as the Theory of Planned Behaviour (Ajzen, 1991) and the Health Belief Model (Becker, 1974) have been proposed to explain health related behaviour and can be applied to explain health decision-making. The Health Belief Model considers how the perception of health threat and

perceived threat reduction influence behaviour, whereas the Theory of Planned Behaviour considers how attitudes towards the specific action, subjective norms regarding the action and perceived behavioural control influence behavioural intentions. However these models are not elaborate enough to explain the cognitive processing and meaning of parents making the decision for their child to undergo neurosurgery in this context.

This decision involved consideration of a number of factors, before deciding to go ahead with the surgery. This decision process was set in the context of their child being physically, functionally, psychologically and socially disadvantaged due to disability. The treatment context offered potential for long term benefit in reducing dystonic spasms, but had short term costs (hospitalisation, surgery), longer term costs (recovery, setting adjustment) and the consequences of their child being dependent on a technical device. Undoubtedly, the overall sense was that parents were trying to do their best for their children. DBS decision-making has never been explored, and this research aim provides a new understanding of the psychological processes and factors influencing DBS decision-making.

4.1.2.1 The Context of Disability & Hope for a Better Life

The driving motivator to consider DBS Surgery was the parental desire to give their child a better life. This decision was therefore set in the context of the child's physical difficulties and wider social and emotional experiences of dystonia discussed in question one. Parents had different hopes for surgery, some were physical (more control, reduction in spasm), others were functional (improve participation in activities) and some were about QoL (pain reduction). These hopes were consistent with the functional priorities of parents identified in the literature and the different priorities for higher and lower ability children (Lumsden et al., 2015). Beyond functional concerns, parents of more able children were also motivated by a desire for their child to have a more 'normal life' through participation in age appropriate activities, independence and looking visibly 'more normal'. This was the first study to focus on the visible aspect of dystonia and how feeling different, could motivate families to undertake DBS to try and achieve a sense of normality.

4.1.2.2 Parents' Attitude to Parenting a Child with Disability

This decision was located in the context of parent's belief system and experiences. Parents believed they must do everything to help their child achieve their full potential and provide the best opportunity in life. The DBS decision is therefore in keeping with how parents have reacted and coped with disability throughout the child's life. As already, discussed parents struggle with their inability to 'fix' the child and, as such, appear to go to any lengths to give their child a chance at a better life. Research has previously identified parents' concern about ensuring everything possible is done for their child (Henderson, 2008; Larson, 1998, Nelson et al., 2012). Notably, all hopes were about the child and parents never voiced their difficulties as a motivator to undergo surgery. This speaks to the parents' unwavering commitment to give their child a better life. This study offers new insight into the broader relational and social context in which DBS decision-making takes place, and is in line with previous studies of elective surgery where social, emotional and psychological factors were important in decision-making (Bonatti et al., 2009; Daniel et al., 2005; Dewar & Peters, 2005).

4.1.2.3 Uncertainty of Outcome and Potential Risks

All parents faced the dilemma of deciding whether the child should have surgery with its associated risks and no certainty of what impact DBS would have. Decision-making involved weighing up surgery risks and parents experienced fear that the surgery would go wrong causing more damage to the child. The meaning of neurosurgery was significant and has been under researched in the literature. In this study neurosurgery was perceived to be more risky than other types of surgeries, and resulted in greater decision-making burden.

Decision-making was influenced by the severity of child disability. Parents whose children were more physically able with high cognitive functioning perceived their children as having more to lose than parents of children who were severely impaired. This process impacted on the ease of decision, and consequently the length of time it took to make the decision. Parents of more able children appeared to agonise over this decision, and displayed

ambivalence as they often changed their minds, whereas parents of less able children were not tormented by the uncertainty of if they had made the right decision. This decision was also experienced as difficult because parents could be offered no certainty of DBS outcome. This uncertainty represented the main struggle for parents, and Mishel's Uncertainty in Illness theory (1988) conceptualises how unfamiliar procedures and potential change in health status, lead to increased uncertainty and distress.

Ultimately, all parents made the final decision by privileging the hope for a better life over all perceived risks. Parents believed the surgery was worth the chance. In terms of models of decision-making regret theory (Loomes & Sugden, 1987) proposed that in conditions involving risk people often make decisions, by weighing up consequences of a possible action with consequences of different decisions. It proposes that people are motivated to take action to avoid future regret. In this context, parents described feeling lucky to be offered DBS, and there was a sense of parents wanting to try all options, and find out conclusively if DBS could help their child to avoid regret.

4.1.2.4 Involving Children and Trusting Professionals

A key feature was listening to the views of the child and involving them throughout the decision-making process. In the literature there is diversity in how far children are involved in surgery decisions. Some studies have shown children are rarely involved, and parents and professionals role in involving children is essential (Coyne, 2008). In DBS, because it is an elective surgery and given the long term impact of being dependent on a technical device, parents sought to involve children as much as possible considering their age and cognitive abilities. Children attended all the appointments and were involved in discussions from the beginning. However, parents also held a protective role in keeping positive and minimising risks to try alleviate children's worries and concerns.

Another important factor was the trust and value parents placed in professional opinion. Professionals seemed to hold a position of power in influencing parents to go ahead with the

surgery. Healthcare professional power and competency has previously been shown to influence parents' decision-making (Nelson et al., 2012). It seems that for elective surgery, when professionals can't guarantee positive outcome because of the heterogeneity of secondary dystonia, parents engaged in a long process of weighing up perceived benefits and costs as a family, and were overly reliant on professionals in the face of this uncertainty.

4.1.3 Question 3: How do parent's experience and manage the DBS process?

Parents experienced emotional stressors and psychological struggles during the DBS surgery and hospitalisation, and the ongoing challenge of negotiating and adjusting to a life with a DBS system. Overall DBS was experienced as an ongoing and turbulent journey which tested the resilience of parents. Parents' experience of the DBS process captured the following qualities: struggle with uncertainty, an emotional experience, coping and support, and ongoing challenges and compromise. Parental experiences of DBS have never been explored, and this research question offers new understanding of the psychological processes and challenges of DBS.

4.1.3.1 Struggle with uncertainty

Central to parents' experiences was a struggle with the unknown, unexpected and unfamiliar medical procedures and hospital environment. This led to feelings of uncertainty, which were emotionally distressing and difficult to cope with. Mishel (1988) proposed a theory of uncertainty to explain how individuals process illness-related stimuli concerning themselves or another person. Event familiarity was a core component of this theory, and suggests that in face of new procedures and new-illness related experiences (e.g. during DBS) uncertainty is heightened (Mishel, 1988). During DBS, parents were experiencing many layers of uncertainty: uncertainty of outcome, chance of potential risk, unfamiliar environment, unknown child recovery process, and unexpected complications. This led parents to feel very distressed and anxious. This is consistent with literature demonstrating that high levels of uncertainty have been associated with emotional distress, anxiety and depression (Wright,

Afari & Zautra, 2009). Event congruence (correspondence between expectations and reality) was another core component of Mishel's (1988) theory, and further contextualises parents' experiences. Parents' distress was heightened when: they felt unprepared, the child's recovery/reaction to DBS was different than expected, the expected standard of care was not met, or medical professionals were unable to provide answers or explanations for unforeseen complications.

Parents' distress can further be contextualised by Becker's (1999) Theory of Disruption. This Theory proposed that people experience inner chaos when expectations are not met, leading to a re-evaluation of the self and world. This re-evaluation was present in parents who were more distressed by uncertainty, lost confidence in their decision and questioned professional competence. Parents' experiences of uncertainty, helplessness and vulnerability, were consistent with wider paediatric surgery (Iverson et al., 2013). Notably, some parents were able to embrace and accept uncertainty. This seemed to occur when parents perceived situations as a challenge or 'adventure' rather than a threat (McCrae, 1984) leading to more positive appraisals and coping.

4.1.3.2 An Emotional Experience

Parents experienced a range of negative and positive emotions in response to the multiple challenges and stressors of DBS and hospitalisation. In the paediatric literature parents experience intense stress, helplessness and high levels of preoperative anxiety (Brennan, 1994; Kain et al., 1996). Similarly, worry and fear predominated parents' accounts. The anaesthesia was particularly distressing, supporting increasing evidence that anaesthesia is one of the most anxiety provoking factors for parents ((Frank & Spencer 2005; Cagiran et al., 2014). In DBS, this moment was perceived as distressing because parents felt alone, unprepared for the surgery wait and overcome by the reality of their decision and saying goodbye to the child.

During surgery parents relinquished control, leading parents to feel powerless and helpless, and many were faced with the fear of potential loss of their child. Loss of control has been related to increased anxiety and depression in the literature (Lachman, Neupert & Agrigoroaei, 2010), and supports findings of parents' experiences during CP paediatric surgery (Iverson et al., 2013). This overwhelming worry was experienced as too unbearable, so that parents tried to control worry through distraction and escape-avoidance strategies (e.g. leaving the hospital on the day of the surgery, keeping busy).

For other parents the greatest challenge was being away from their children and normal family life, and the lack of support for sibling anxiety. Consideration has not been given to siblings in the DBS or dystonia literature, however from a family systems perspective anxiety experienced by the child with secondary dystonia and parents will affect all other members (Dallos & Steadman, 2006).

4.1.3.3 Coping and Support

Parents used a variety of coping strategies to manage their negative experiences, cope with uncertainty and manage the challenges and stresses of DBS. Research has found that choice of coping strategy depends on controllability of the situation (Conway & Terry, 1992). People tend to use task-oriented coping strategies when they feel they can have an impact on the problem whereas they tend to use emotion-oriented coping when they feel that they must tolerate the stressor (Endler & Parker, 1990; Folkman & Lazarus, 1980). During DBS, parents relied on a range of emotion-focussed coping strategies to regulate their emotional distress. These strategies involved keeping busy, distraction and planning activities to avoid rumination. Parents also sought emotional support and were dependent on their partner and family members. For parents who perceived higher risks, and beliefs their child could die, psychological distancing seemed to be way of protecting oneself from potential loss.

There was variability in parents' experience of stress and psychological distress. For some parents, it appeared that previous difficult life experiences (e.g. child birth, diagnosis of

disability, hospitalisations) may have enhanced parents' resilience to cope with DBS. These ideas are further contextualised by the concept of psychological preparedness (Janoff-Bulman, 2004). This theory proposed that a person can be prepared for future traumatic events by experiencing stress and adversity. Perhaps these parents were able to have a positive attitude towards surgery, which helped act as a psychological buffer and perceive events as less threatening. These ideas are in keeping with the Theory of Hardiness (Kobasa, Madoi, Pucetti & Zola, 1985). Parents who considered the surgery as a challenge, and embraced the demands seemed to cope better. For example, parents spoke of a choice in making the head shaving a positive family moment, and how they spent the day of the surgery. This perception of having a choice seemed to enhance parents' feelings of control. This was in contrast to parents who approached the surgery more passively, appraised it as a significant threat and felt powerless.

Parents who seemed to struggle most with the surgery also perceived a lack of support from staff, and were in hospital alone. Social support has been identified in the literature as an important stress buffering resource (Plant & Saunders, 2007). Therefore emotional support seemed to be essential to promote positive coping and adjustment during DBS. The nursing and medical team had a strong influence on the experience of parents. Professional kindness, competency, compassion and an individualised approach helped parents to trust and have confidence in staff, which allowed them to take breaks and escape the hospital environment.

4.1.3.4 Ongoing challenges and compromise

After the surgery and hospitalisation, parents and children embarked on a process of adjusting and negotiating to having DBS. Previously in the adult and child qualitative dystonia studies (Bakowski, 2010; Hariz et al., 2011) a key challenge was adjusting post surgery. This involved experiencing negative side effects and concerns about the visibility of, and ongoing dependence on, an implanted battery. Parents reported similar concerns and challenges: concerns were the extra responsibility of charging, visible scarring and visibility of battery. The visible impact of DBS was significant for families where children were acutely aware of

difference, and DBS was perceived to add to their negative self concept. Other concerns were reported by parents that have not previously been documented in the literature. Parents particularly struggled with the wait for change, anxiety of if the system was working correctly and how change was not linear but perceived as going backwards and forwards. Parents also struggled with the presence of new and different dystonic movements, and uncertainty about how long changes will last. This suggests parents shared concerns with their children, but had an additional layer of worry and uncertainty.

4.1.4 Question 4: What is the impact and meaning of post-surgery change for parents and children with secondary dystonia?

Overall, DBS leads to positive changes for all families however there was variability in the extent and meaning of change. For some DBS was life altering, whereas for others families changes were perceived as more subtle. This research question also considered the novel area of how parents make sense of changes and the subjective meaning assigned to changes. This research question offers a unique focus on parental experiences of DBS change, and hopes to elucidate the experience of change for children through the eyes of their parents.

4.1.4.1 Impact of DBS

All parents described positive changes in secondary dystonia after DBS. Consistent with the child secondary dystonia literature (Gimeno et al., 2012;Lumsden et al., 2012) parents reported reduction in spasms, more relaxed and looser movements, more bodily control, improvements in posture and less pain. This led to improvement in QoL, independence, activities of daily living and children were less reliant on medication. For some parents, the changes were more subtle and parents were unsure of the degree of dystonia improvement, however these parents described changes in ease of care, manual handling, transfers and participation in activities. This supports findings of Gimeo et al. (2012) that DBS can lead to

significant benefit in terms of individualised goals and participation in activities of daily life, that were not being recorded by impairment focussed measures.

For some parents DBS also led to reported improvements in children's sleep, mood, anxiety, confidence, and perceptions of increased bodily control helping the child to feel more normal. For many parents, there was a positive and unexpected change in children's speech. Parents perceived children's speech to be clearer, more fluid and more vocal. These positive changes have not been reported in previous quantitative studies of DBS change in children. However, these novel findings were essential to parents' experience, who appraised these changes as significant because of increasing their child's ability to communicate and participate in family life. This meant children could share their personality and sense of humour, and there was a sense of DBS helping to unlock their full potential. Clearly impairment based measures are not capturing the wider impact and significance of DBS change, and are too focussed on physical ability and impairment, supporting conclusions by Gimeno et al. (2012).

4.1.4.2 The Meaning of Change and Sense-making of Parents

There was disparity in parents' accounts of the extent and significance of changes, and the meaning of change was clearly a nuanced and subjective experience. For some, there was joy at the 'life changing' impact of DBS, and DBS was perceived to have the greatest impact because of an improvement in family functioning and normality, and allowing the child to participate by removing a barrier created by disability. All parents engaged in a process of sense-making. It appeared as though changes were perceived as more meaningful if expectations were exceeded, changes were unexpected, and the child was able to do something they had never done before or were told would never do. It was also important that changes were visible and noticeable to others. These factors suggest the importance of capturing the subjective experiences of parents to evaluate DBS success and inform parents decision-making.

Sadly, other parents perceived changes to be more subtle leading to disappointment in DBS outcome, and were dependent on medical professionals to know if there had been motor changes. Through social comparison (Festinger, 1954) with previous body functioning, parents tried to cope with this uncertainty to evaluate change in light of perceived changes. These parents were disappointed because specific goals were not met, children struggled to see a change and the changes weren't visible. These parents engaged in cognitive processing to make sense of more subtle and 'quieter' changes to try and find answers and rationalise the lack of change.

This study demonstrated a huge variability across DBS outcomes of a heterogeneous group of children with secondary dystonia. This variability in DBS outcome has been recently reported in a study of adolescents and adults with dystonic CP (Romito et al., 2014). Consistent with this research, magnitude of improvement was unevenly distributed across the sample but in some cases DBS had the potential for remarkable change (e.g. 50% reduction in BFMDRS). Similarly, our study demonstrated that DBS had the potential to create significant changes for families, which is different to previous child secondary dystonia studies reporting modest and subtle changes (Bakowski, 2010; Gimeno et al., 2012; Lumsden et al., 2012). However, contrary to existing research, it appears as though the parents who appraised DBS change most significantly were parents of children with more severe disabilities. Parents of more able children appeared to have more specific goals and perhaps higher expectations, leading to feelings of disappointment. Parents with severely disabled children seemed to accept that any change would be positive, helping them to experience joy and elation at the changes.

4.1.4.3 A New Perspective

Parents spoke of gaining a new perspective of themselves, their child and their future and for many there was a sense of developing a stronger identity after the challenges of DBS. This is consistent with the literature on post traumatic growth that suggests that positive changes and psychological growth can come out of traumatic experiences (Carver, 1998). Consistent with

Tedeschi and Calhoun's (1995) model of post-traumatic growth, parents experienced perceived changes in self (confidence, stronger, experts in DBS) and developed a closer relationship with their children. There was a sense of overcoming a shared adversity with the child, which strengthened the parent-child bond. Parents were inspired by the courage and resilience of their children, and experienced overwhelming pride in their children. This is consistent with previous reports of parents of children with CP (Davis et al., 2010). Undoubtedly DBS was a life altering experience, with significant affects on the children and parents.

4.2 Critical Evaluation

4.2.1 Strengths

The main strength of this study lies in the collection of powerful and rich narratives that have yielded many insights into the lived experiences of secondary dystonia and DBS. As described in the methods, various steps were followed to ensure validity of this qualitative research and production of a high quality IPA. The researcher identified and targeted a clinically important gap in the literature. Service-user consultation was well planned and consequently meaningfully shaped the study design, data collection and data analysis. An important strength was the use of credibility checks and reflexivity to maintain quality and validity of final themes (Yardley, 2008).

In this study every parent approached agreed to take part, this reduces recruitment bias of people volunteering to share overly positive or negative experiences. Careful consideration was given to inclusion criteria and adoption of a sampling strategy to ensure homogeneity within the sample. All parents experienced the same phenomenological experience of parenting a child with secondary dystonia and undergoing DBS and all children had secondary static dystonia. This homogeneity strengthens confidence in drawing conclusions that apply to this unique group of parents. There was however variability in parent gender, and child characteristics (child age, gender, primary diagnosis, severity of dystonia and

communication abilities). Given the small size of the population some variability in sample was necessary, and the sample characteristics of the children were deemed to be representative of the population of children cared for at the hospital site (T. Owen, personal communication, April 9, 2015).

4.2.2. Limitations

As previously discussed the sample isn't truly homogenous because there was variation in child characteristics (age, medical condition, functioning and co-morbidities) and success of DBS surgery. Given heterogeneity of child characteristics it was difficult to ascertain what experiences were unique to secondary dystonia, and the themes are therefore representative of children who have secondary dystonia and another diagnosis e.g. CP. Furthermore the introduction of sample heterogeneity makes it more challenging to analyse the patterns of convergence and divergence within the group and to ensure the claims are representative of the shared and lived experiences of DBS and not attributable to sample variation. There also was variability in parent characteristics as only one father was interviewed, and although not selected for, all parents identified as white British. It is therefore likely that the themes are representative of mother's experiences from one cultural group. All participants were recruited from one hospital site, reducing generalisability of conclusions. However, IPA does not seek generalisability and is interested in documenting existence of patterns in life not measuring incidence. In terms of making claims, IPA can shed light on existing nomothetic research and have 'theoretical transferability (Smith et al., 2009).

In this study the researcher was attempting through a double hermeneutic to gain access to parents' lived experience, whilst also attempting to understand childrens' experiences through the parents' eyes, suggesting a triple hermeneutic was invoked to understand secondary dystonia as a relational experience. Studies have shown parents not to be valuable informants of their childs' well-being (Havermans Vreys, Proesmans & De Boeck, 2006), and at best the researcher was only provided a partial view of children's experiences.

4.3 Research Implications

The exploratory nature of the study and critical evaluation of the study, highlight research areas which warrant further investigation:

- Exploration of the experiences and differences between parents of children with different primary diagnoses co-occurring with secondary dystonia. Although beyond the scope of this research, qualitatively there was a sense that experiences may differ between genetic conditions and CP due to the origins of aetiology.
- Exploring the experiences and differences of children with different impairment severities of secondary dystonia. As indicated by these findings their experiences are likely to be subjectively different, the functional DBS priorities are different (Lumsden et al., 2015) and child may respond differently to DBS (Gimeo et al., 2012).
- This research raised questions of whether parents' experiences of caring for a child with secondary dystonia is similar for mothers and fathers.
- DBS was experienced by parents as a demanding and ongoing process, experiencing multiple and different stressors in this time. A prospective longitudinal follow up of parents could help identify at risk families, and understand how experiences change over time.
- It would also be important to investigate the experiences of children with secondary dystonia experiences in their own right, given the limitations of accessing experiences through a triple hermeneutic, and level of psychological distress and functional dependence.

4.4 Clinical Implications

This study increased understanding of the experiences and meaning of living with secondary dystonia and undergoing DBS. Subsequently it has a number of clinical implications relevant to the clinical team and hospital setting where DBS is offered:

4.4.1 Clinical Assessment and Support

- Clinicians need to routinely offer assessment of psychological well-being and QoL of parents and children. Assessment should move beyond an impairment focussed conceptualisation of secondary dystonia, to consider the wider personhood of the child, and the emotional and social impact of secondary dystonia.
- When difficulties are identified, a family approach could be helping in supporting adjustment at a family level (Kazak, 1989).
- This study suggests that visible difference was important in children's developing self concept, and children may need psychological support to help their adjustment and acceptance.

4.4.2 Decision-making Support

Managing uncertainty was the prominent struggle for parents, and clinicians have a responsibility to ensure parents can make an informed decision with all the relevant information:

- In the face of uncertainty of DBS outcome, parents need to be provided with the latest outcome evidence for secondary dystonia to ensure informed decision-making.
- Clinicians need to be clear and informative about likelihood of DBS changes and support families to develop realistic expectations of change.

- Clear information and recent photographs should be provided of the location of DBS implant, stitching in the head, scarring after surgery, and the visibility of the battery pack under the skin.
- This study suggested parents of more able children (lower GMFCS scores) and children where there was a disparity between cognitive and physical functioning, struggled more with uncertainty and the responsibility of decision-making, perceiving there to be more to lose. Clinicians' should be aware that parents who were more ambivalent in their decision may be more vulnerable to experience distress during and after the surgery.
- Decision-making ambivalence could make parents vulnerable, and place professionals in a powerful position. Professionals should be mindful of this, and ensure parents are provided the time and information to reach their own decision.

4.4.3 Surgery Preparation and DBS Support

There was variability in how parents experienced the DBS surgery and hospitalisation. Therefore Clinician's could identify at risk families, and adopt a stepped care approach to providing different levels of support tailored to the individualised needs of parents and children:

Psychoeducation:

Parents should receive more preparation to help reduce their uncertainty and anxiety. As a clinical output of this research, psychoeducational leaflets are being produced summarising parents' experiences of surgery and outcomes to help parents' decision-making, to feel more prepared and to normalise their emotional reactions. These will include details of:

- Emotional reactions to salient moments e.g. anaesthesia, saying goodbye, waiting during the surgery, recovery.

- The hospital environment, recovery process, potential complications, and where to do/what to do on the day of the surgery.
- Recommended strategies parents have found helpful e.g. seeking support, keeping busy, taking breaks, planning activities on day of surgery.
- What to expect after the surgery, e.g. scarring, time to achieve optimal setting level.

Clinical Practice:

- Clinicians' need to be aware of the wider context of a child's experience of secondary dystonia and take a systemic approach to preparing and supporting families through DBS.
- Information will also be shared with the clinical team regarding unique DBS outcomes, the subjective meaning of change, stressors and challenges of the experience, and what factors helped parents to manage.
- Clinicians need to evaluate the use of quantitative measures to capture DBS change, measures of disability (e.g. BFMDRS) and impairment (GMFCS) are not capturing the subtleties and subjective meaning of change experienced by families in this study. Furthermore improvements in speech, psychological well-being, sleep and confidence are not being captured. Increased understanding could help clinician's to provide more certainty of likely DBS outcomes to reduce parental anxiety and enhance informed consent.

Psychological Support:

- Parents identified to be struggling could benefit from psychological support during and after DBS. Acceptance and commitment therapy, mindfulness based approaches or cognitive behavioural therapy could help parents to cope with uncertainty.
- Longer term follow up of at risk families would be beneficial to support families adjustment to life after surgery.
- Greater sibling support and psychoeducation could be provided to alleviate sibling anxiety.

- Support groups could be established, or parents could be assigned a 'parent buddy' who had been through the process to help normalise emotional reactions.

These recommendations have been made in the hope of improving the quality of care and clinical support provided to families. Nationally and internationally, most DBS services are medically-led, and the psychological support and clinician follow-up options recommended in this study are not currently available. Within the service where the study was conducted, a clinical psychologist is part of the team. However, the remit of the role at the time of completing this research was focussed on the neuropsychological assessment of children pre and post DBS surgery. Currently families do receive support managing expectations through goal-setting with an Occupational Therapist, and are given details of a parent-led facebook group. However, the clinical team at the research site do not formally evaluate the well-being of parents, and psychological interventions and support groups are not available.

This study highlights the importance of a multi-disciplinary team approach in DBS services to ensure the psychological needs and well-being of children and parents are being considered from the moment DBS is introduced as a surgical option. Following completion and dissemination of the findings to the clinical team at the research site, service changes have been implemented. The clinical psychologist role has been expanded to support families through the decision-making process and offer psychological follow-up to families. Furthermore, as a team, greater emphasis has been placed on supporting families to develop realistic expectations and to enhance informed decision-making by sharing latest outcome evidence.

4.5 Personal Reflections

I feel that I embarked on my own uncertain and turbulent journey with IPA. I was struck by the parallel between my uncertainty and self doubt if I was doing the analysis right, and the uncertainty of participants. Through keeping a reflective journal, and attempting to embody the hermeneutic circle, I became more aware of implicit assumptions and my own values and

belief system. Looking back I realise many reflections were focussed on the meaning of difference at a family, social, psychological and societal level. Little consideration was given to cultural values, and I wonder if this was because the families came from the same cultural background as myself, and therefore unconsciously there was an assumption of holding similar beliefs and culture somehow seemed less relevant

Most importantly, I was surprised by participants' willingness and openness to share their stories with a stranger. I felt privileged to hear the many heart warming and heart breaking experiences of parents' and their childrens' journey with secondary dystonia and DBS.. I shared parents' joy as they vividly described the life altering impact of DBS, and shared the sadness of parents' disappointment with subtle changes. My interviews were very long, and parents spoke of never sharing their experiences. On reflection, it shocked and worried me that parents can intensely engage with medical services and go almost unnoticed. This parallels the findings and implications of this study, that secondary dystonia should be considered within a systemic framework. This research experience will have undoubtedly changed and shaped my own attitudes and perceptions of disability. It has validated for me the importance of working systemically and thinking about the needs of parents, which is often neglected in paediatric settings where a medical model often dominates.

4.6 Conclusions

The results of this study provide a rich insight into the experiences of parents and children of living with secondary dystonia and going through DBS. Eight parents were interviewed and transcripts were analysed using IPA. Consistent with the CID literature, parents and children experienced a difficult and different life with secondary dystonia. The concept of control was central to experience as children struggled to control an uncontrollable body, and parents struggled with their inability to make things better and sought to take control of disability. Overall, secondary dystonia was considered a multifaceted phenomenon, which exists within a relational, social and societal context, and had wider impacts on the physical, social and emotional functioning of children and parents.

This was the first study to capture the psychological processes involved in DBS decision-making. Parents were motivated by a hope for a better life and parental duty. This was weighed up against consideration of risks, what the child had to lose, and uncertainty of DBS outcome. Parents experienced many challenges and psychological struggles during DBS and hospitalisation testing their resilience and coping. Families continued to adjust to a new life and to being dependent on a technical device. There was variability in objective DBS outcomes and the subjective meaning of change. For some, DBS was perceived as life changing, whereas other families experienced disappointment and tried to rationalise a lack of change. Inherent to the idiographic nature of IPA, clinical recommendations have been made at the clinical team and hospital level to help support adjustment to secondary dystonia and DBS.

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Appendix 1: Participant Information Sheet



Participant Information Sheet Version 1.0 26.02.2014

Parents' experiences of secondary dystonia and the journey through Deep Brain Stimulation Surgery

We would like to invite you to take part in our psychology research study. This study hopes to gain a greater understanding of parental experiences of caring for a child with secondary dystonia before, during and after Deep Brain Stimulation Surgery (DBS). Before you decide whether to take part, it is important that you understand why the research is being done and what this study will involve for you. Please take time to read the following information carefully and discuss it with relatives, friends or members of your clinical team if you wish. One of our team will go through the information sheet with you and answer any questions you have.

- Part 1 tells you the purpose of the study and what will happen to you if you take part.
- Part 2 gives you more detailed information about the conduct of the study.

Please take your time to decide whether or not you wish to take part. Please ask if there is anything that is not clear, or you would like more information (See contact details at the end of this information sheet).

Part 1

What is the purpose of the study?

DBS surgery has some success in the management of secondary childhood dystonia (dystonia which occurs alongside other conditions e.g. cerebral palsy). However, little is known about the psychological, social and practical implications of DBS on the children and parents. The purpose of this study is to gain an understanding of parental experiences of caring for a child with secondary dystonia before DBS, making the decision to undertake DBS, managing the DBS process and the impact of DBS on the child and family. It is hoped this information will help staff to better prepare children and families for surgery, manage parents' expectations and support families' adjustment to life after surgery.

Who is organising and conducting the research?

The research is being supervised by Dr Tamsin Owen, clinical psychologist, within the Service. The study is being carried out by Allana Austin, who is a trainee clinical psychologist at Royal Holloway, University of London.

Why have I been invited?

We are inviting the parents of children with secondary dystonia, who have undertaken DBS surgery at the _____ Hospital in the last 2 years and are currently being followed up by the clinical team.

We are interviewing you because we are interested in hearing about the views and experiences of parents, but also your perspective of how the DBS affected your child's movements and quality of life. We are not interviewing the children, because research has already explored the views of children with dystonia, and we are also hoping to capture the experiences of some children who are not able to communicate verbally.

Do I have to take part?

No. It is up to you to decide whether or not to take part. If you decide you would like to take part, we will describe the study in more detail and go through this information sheet. If you agree to take part, we will discuss this information sheet in more detail and then ask you to sign a consent form to show you have agreed to take part. You are free to withdraw at any time, without giving a reason. Withdrawal or non-participation will not affect the standard of care your child receives or any future treatment in any way.

What will happen to me if I take part?

The researcher, Allana Austin, will contact you to discuss the study and answer any questions you may have. If you are willing to take part, an interview will be scheduled to take place at the _____ Hospital when you attend for your child's review appointment. This appointment can take place at a time suitable for you. If no preference we will suggest completing the interview during your child's neuropsychological assessment to minimise any disruption to your child. Alternatively, the researcher could visit you at home to complete the interview.

The interview will last approximately 60-90 minutes. The length of the interview will vary depending on how much you feel you wish to say.

We will ask you about:

- Your experiences as a parent of a child with secondary dystonia
- The impact of secondary dystonia before surgery
- The decision to undertake DBS: your hopes, fears and expectations
- What it was like going through the DBS process: stressors and support
- The impact of DBS on you and your child

There are no right or wrong answers, and you are free to decline to answer any question you do not feel happy to answer. Interviews will be completed in a private room, and with your consent we will audio record the interview for later analysis.

In summary taking part in this research, involves meeting a researcher once, to take part in an interview about your experiences as a parent of a child with secondary dystonia.

Expenses and payments

Taking part in this study is voluntary and you will not be paid for your participation.

What are the possible disadvantages or risks of taking part?

We do not anticipate that there will be any disadvantages to taking part, except for the inconvenience of making time for the interview. However, it is possible that some topics discussed concerning the stresses of caring and your child's health may be upsetting. At no point will you have to answer anything you don't want to, and if you become distressed at any time, you can take a break or stop the interview. If you do feel you would like to speak with someone after the interview you will be able to meet with Dr Tamsin Owen, clinical psychologist within the team or one of the parent counsellors to discuss this further.

What are the possible benefits of taking part?

We cannot promise the study will help you, but we hope the valuable information we get from this study will help to improve the experiences of children and families undertaking Deep Brain Stimulation Surgery. Additionally, some parents have reported wanting an opportunity to talk about their experiences in detail.

What if there is a problem?

Any complaint about the way you have been dealt with during the study or any possible harm you might have suffered will be addressed. The detailed information on this is given in part 2.

Will my taking part in the study be kept confidential?

Yes. We will follow ethical and legal practice, and all information about you and your child will be handled in confidence. The details are included in part 2.

This completes Part 1 of the Information Sheet. If the information in Part 1 has interested you and you are considering participation, please continue to read the additional information in Part 2 before making any decision.

Part 2

What will happen if I don't want to carry on with the study?

You are free to withdraw at any point, without giving a reason. Refusal or withdrawal of consent will not affect the current or future care your child receives at the

Hospital. You have the right to withdraw consent after it has been given, and to ask that your own data, including recordings, be destroyed. The researcher will give

you her contact number so even after the interview you can let her know if you have changed your mind, or you would like parts of the interview taken out. Any data that you do not want included will be destroyed.

What if there is a problem?

If you have a concern about any aspect of this study, you should ask to speak to either Allana Austin (researcher) or Dr Tamsin Owen (clinical psychologist), who will do their best to answer your questions (contact details are provided at the end of the information sheet). If you remain unhappy and wish to complain formally about any aspect of the way you have been approached or treated during the course of this study, you may contact Dr [redacted], consultant paediatric neurologist, of your clinical team. You can write to him at [redacted].

In the event that something does go wrong and you are harmed during the research and this is due to someone's negligence then you may have grounds for legal action for compensation, but you have to pay your own legal costs. Royal Holloway, University of London, is providing negligent and non-negligent indemnity cover for this research. The normal NHS complaints mechanisms will still be available to you.

Will my taking part in this study be kept confidential?

Yes, we will follow ethical and legal guidelines, and all information about you and our child will be kept strictly confidential and known only to the researchers.

All data collected during the course of the study will be held according to the Data Protection Act (1998). The audio-recording of the interview and transcripts of the interview will be given an identification number. This means that only the researcher will know whose data belongs to whom. The interview will be anonymous, and any identifiable information will be deleted. Your name and your child's name will not be disclosed to anyone else, and neither will you be identified in any report or publication. Some of your comments may be directly quoted when the research is written up, however this will be completely anonymous.

All anonymised paper data will be stored securely in a locked filing cabinet that only Allana Austin or Dr Tamsin Owen will have access to. Similarly, electronic audio recordings and interview transcripts will be saved on an encrypted electronic storage device. On completion of the research, all of the interview tapes will be deleted, and transcripts of the interviews will be stored at [redacted] for up to 5 years. Signed consent forms will be stored securely at Royal Holloway University, and destroyed after 2 years.

If any person in the study tells us that they or someone else is being harmed, or the researcher is concerned about risk of harm, then she is legally obliged to share this information with the appropriate clinicians (e.g. clinician from [redacted] Service, your GP). The researcher will always try and discuss these concerns with you first. We respect confidentiality, but cannot keep it a secret if anyone is, or is at risk, of being seriously harmed.

What will happen to the results of the research study?

The results of the study will be written up as part of a Doctorate in Clinical Psychology. The results may also be published in a journal or presented at a conference. We will also offer you a summary of the findings.

Who has reviewed the study?

All research in the NHS is looked at by an independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed and given favourable opinion by London-Bloomsbury Research Ethics Committee. The study has also been approved by the Research and Development Department and the Research Ethics Committee at Royal Holloway, University of London.

Further information and contact details

If you would like further information about taking part, please do not hesitate to contact Dr Tamsin Owen in the first instance. Contact details are below.

Dr Tamsin Owen, clinical psychologist
Email: tamsin.owen@.nhs.uk or by phoning

Thank you for considering taking part or taking time to read this sheet. If you decide to take part you will be given a copy of the information sheet and a signed consent form to keep.

Appendix 2: Interview Schedule

SEMI-STRUCTURED INTERVIEW SCHEDULE

Interview Process

- 1) Introductions
- 2) Interview process and Confidentiality
 - Review information sheets – Opportunity to ask questions
 - Consent Forms
- 3) Warm up questions
- 4) Interview Schedule
- 5) Debrief and endings

Introductions, Information about the interview process and confidentiality

- The participant will be reminded about the reason for the study, the length of time it is expected to take, can take a break and their right to end the interview or opt-out at any time.
- The issue of confidentiality will also be clearly explained.
- Explanation of the interview e.g. want to hear the details of your experiences- you are the expert, I'm not going to say much, there are no right or wrong answers, take your time answering
- Study predominantly about experiences of parent – experiences of having a child with dystonia go through DBS

N.B. Example Experiential Follow-up questions to use throughout interview

- What does it mean to you that...?
- How did that make you feel at the time?
- What were you thinking at the time?
- What is that like for you as a parent...?

Warm up questions

How was your journey here?

Can you tell me a little about your child?

1) Can you tell me about what life was like before DBS?

Prompt: Can you tell me about your experiences as a parent of a child with secondary dystonia before the DBS procedure? How did your child's movement difficulties affect them? Can you tell me about any difficulties you have faced? Can you tell me about any positive aspects?

2) Can you tell me about how these experiences have impacted on you?

Prompt: Affect on relationship with your child/partner, affect how you see yourself, impact on the rest of the family?

3) Can you tell me about how you have managed these experiences?

Prompt: What helps you? How do you look after yourself?

4) Can you tell me about the experiences that led you to the point where you were considering DBS Surgery?

5) Can you tell me about your experience of how you made the decision to undertake DBS?

Prompt: What factors influenced your decision? What helped you to decide? What was your child's view of the DBS surgery? Looking back is there anything that would have helped in this decision?

Prompt: Before the surgery what were your expectations, hopes and fears?

6) Can you tell me about your experiences as a parent of the DBS Surgery process (e.g. the surgery, follow up appointments)?

Prompt: Tell me about any positive or negative experiences? Tell me about any demands on you as a parent throughout this process?

7) How have your experiences of the DBS process impacted on you?

Prompt: Tell me about how the DBS process impacted on your child/relationships/work/family/view of self? What were the demands on you as a parent?

8) Can you tell me about how you managed and coped with your experience of DBS?

Prompt: Tell me about any sources of stress/challenges/support? What factors made the process easier for you to cope with? (Was there anything that made the surgery harder for you to cope with?)

9) What have your experiences been since the DBS Surgery?

Prompt: How has life changed? What impact has DBS had on your child? What affect have those changes had on your life? Has going through DBS changed or influenced you as a person in anyway?

10) Looking back on the DBS process, was having the surgery worth it?

Prompt: Tell me about any positive changes? Tell me about any negative effects? Has your child met their/your goals for surgery? How does the changes compare to your expectations/hopes?

12) What would you say to other families who are considering DBS?

Prompt: Looking back is there anything that would have helped you through the process? What could be offered by services that you might find helpful or supportive? What things were important to you and your child throughout the DBS process?

Space to reflect on the experience and ask questions

Example Questions:

Was there anything you expected me to ask, that I didn't?

Is there anything that I haven't asked you that you would like to tell me about?

What has it been like discussing these issues today?

Do you have any questions you would like to ask me?

Debrief

We have now reached the end of the interview. I would like to thank you for taking the time to talk to me. I have very much valued hearing your thoughts and experiences.

If distressed: If you would like to talk to Dr Tamsin Owen about anything? If you aren't sure now you can always contact her later on.

Contacting me: Tell the participant that they can contact me by telephone or email (on information sheet) if they think of anything they would like to ask me, or if they are worried about anything related to the interview

Report summary: Ask the participant if they would like to receive a copy of the main findings from the research

Thank the participant for taking part.

END

Appendix 3: NHS Research Ethics Favourable Opinion Letter with Comments



Health Research Authority

National Research Ethics Service

NRES Committee London - Bloomsbury

HRA NRES Centre Manchester

Barlow House 3rd Floor

4 Minshull Street

Manchester

M1 3DZ

Telephone: 0161 625 7815

Facsimile: 0161 625 7299

16 April 2014

Miss Allana E L Austin
Trainee Clinical Psychologist
Camden and Islington NHS Foundation Trust
Dept Clinical Psychology, Bowyer Building
Royal Holloway
University of London
Egham Hill, Egham
Surrey
TW20 0EX

Dear Miss Austin

Study title: Parents' experiences of secondary dystonia and the journey through Deep Brain Stimulation Surgery

REC reference: 14/LO/0455

IRAS project ID: 149049

The Research Ethics Committee reviewed the above application at the meeting held on 02 April 2014. Thank you for attending to discuss the application, together with Dr Tamsin Owen.

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details, unless you expressly withhold permission to do so. Publication will be no earlier than three months from the date of this favourable opinion letter. Should you wish to provide a substitute contact point, require further information, or wish to withhold permission to publish, please contact the REC Manager Dr Ashley Totenhofer, nrescommittee.london-bloomsbury@nhs.net.

Ethical opinion

- **Social or scientific value; scientific design and conduct of the study**

The committee stated it is important to not make the interview too long.

You agreed.

The committee noted that there may be home visits for the interviews and queried if there was a lone worker policy.

You stated you would follow the trust lone worker policy and you have had the appropriate training, such as break-away training.

The committee commented that they had concerns that due to the small sample size and the potential diversity in the social and economic backgrounds of the families the results may not be useful.

You stated that the philosophical assumption of IPA is that the sample is as homogeneous as possible. If family's backgrounds are different it still may be possible to draw parallels across them.

Dr Owen stated that due to the nature of qualitative research and the methodology underpinning IPA this is the recommended sample size. There will be a different richness of data than with other methodologies.

The committee queried why are non-English speakers excluded.

Dr Owen stated that of the families coming up for review during the study time-frame all of them have English as a first language.

The committee accepted this and stated that this may not be the case elsewhere.

The committee queried if travel expenses would be paid.

You stated they would not as all the interviews would either be at their clinic appointment or in their home.

- **Informed consent process and the adequacy and completeness of research participant information**

The committee noted the Participant Information Sheet is quite in-depth and queried if it had been checked for readability.

You stated it had been.

The committee noted that the Participant Information Sheet mentioned that the study may improve the approach to surgery and queried how this would be achieved.

You stated that they won't come to any conclusions that are relevant to all children but they may find some conclusions to children coming for surgery at this hospital.

- **Independent review**

The committee noted that the supplied peer review recommended some refinements to the interview schedule and queried if these had taken place already.

You stated that to look at how the surgery had changed the family's lives it is important to discuss events before and after the surgery. In order to not make the interviews too long there will be some refinement to the interview questions but you are not sure what these will be at the moment. You stated you plan to carry out some more pilot interviews and to attend an IPA meeting to discuss this.

- **Suitability of supporting information**

The committee stated that it is normal to notify participants GP as a courtesy and if they

choose to do this the Participant Information Sheet and Consent Form would need to be amended.

You stated you were unsure about doing this as the child is the patient and you are interviewing the parent.

- **Other general comments**

The committee noted that this was a clear and nicely written application.

The members of the Committee present gave a favourable ethical opinion of the above research on the basis described in the application form, protocol and supporting documentation, subject to the conditions specified below.

Ethical review of research sites

NHS Sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see “Conditions of the favourable opinion” below).

Conditions of the favourable opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.

1. Please confirm that the Trust Lone-worker policy will be followed.
2. The committee would recommend producing a GP letter to notify a participant's GP that they are taking part in the study. If this is done then the Participant Information Sheet and Consent Form will need to be amended to reflect this and this would need to be submitted to the REC for approval. This is a recommendation not a condition of the favourable opinion.

You should notify the REC in writing once all conditions have been met (except for site approvals from host organisations) and provide copies of any revised documentation with updated version numbers. The REC will acknowledge receipt and provide a final list of the approved documentation for the study, which can be made available to host organisations to facilitate their permission for the study. Failure to provide the final versions to the REC may cause delay in obtaining permissions.

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission (“R&D approval”) should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements.

Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at <http://www.rdforum.nhs.uk>.

Where a NHS organisation’s role in the study is limited to identifying and referring potential participants to research sites (“participant identification centre”), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of approvals from host organisations

Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publically accessible database within 6 weeks of recruitment of the first participant (for medical device studies, within the timeline determined by the current registration and publication trees).

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g. when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non-clinical trials this is not currently mandatory.

If a sponsor wishes to contest the need for registration they should contact Catherine Blewett (catherineblewett@nhs.net), the HRA does not, however, expect exceptions to be made. Guidance on where to register is provided within IRAS.

It is responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Approved documents

The documents reviewed and approved at the meeting were:

| <i>Document</i> | <i>Version</i> | <i>Date</i> |
|--|-----------------------------|-------------------|
| Evidence of insurance or indemnity | Gallagher Heath | 02 September 2013 |
| Interview Schedules/Topic Guides | 1.0 | 05 February 2014 |
| Investigator CV | Allana Austin | 05 February 2014 |
| Investigator CV | Tamsin Owen | 05 February 2014 |
| Other: Debrief Form | 1.0 | 05 February 2014 |
| Other: NHS to NHS Proforma | | |
| Participant Consent Form | 1.0 | 05 February 2014 |
| Participant Information Sheet | 1.0 | 26 February 2014 |
| Protocol | 1.0 | 18 February 2014 |
| REC application | 3.5 | 26 February 2014 |
| Referees or other scientific critique report | Peer Review Approval Letter | 17 January 2014 |

Membership of the Committee

The members of the Ethics Committee who were present at the meeting are listed on the attached sheet.

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Reporting requirements

The attached document "After ethical review – guidance for researchers" gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The NRES website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

Feedback

You are invited to give your view of the service that you have received from the National Research Ethics Service and the application procedure. If you wish to make your views known please use the feedback form available on the website.

Further information is available at National Research Ethics Service website > After Review

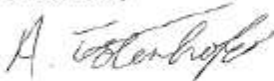
14/LO/0455

Please quote this number on all correspondence

We are pleased to welcome researchers and R & D staff at our NRES committee members' training days – see details at <http://www.hra.nhs.uk/hra-training/>

With the Committee's best wishes for the success of this project.

Yours sincerely



Signed on behalf of:
Dr Joe Brierley
Chair

Email: nrescommittee.london-bloomsbury@nhs.net

Enclosures: *List of names and professions of members who were present at the meeting and those who submitted written comments*

"After ethical review – guidance for researchers"

Copy to: Dr Andy Macleod – Royal Holloway University of London
Ms XXX XXX NHS Trust
Dr Tamsin Owen – Royal Holloway University of London

Appendix 4: NHS Ethical Approval Letter



Health Research Authority

National Research Ethics Service

NRES Committee London - Bloomsbury

HRA NRES Centre Manchester
Barlow House 3rd Floor
4 Minshull Street
Manchester
M1 3DZ

Telephone: 0161 625 7815
Facsimile: 0161 625 7299

16 April 2014

Miss Allana E L Austin
Trainee Clinical Psychologist
Camden and Islington NHS Foundation Trust
Dept Clinical Psychology, Bowyer Building
Royal Holloway
University of London
Egham Hill, Egham
Surrey
TW20 0EX

Dear Miss Austin

Study title: Parents' experiences of secondary dystonia and the journey through Deep Brain Stimulation Surgery

REC reference: 14/LO/0455

IRAS project ID: 149049

Thank you for your letter of 6th May 2014. I can confirm the REC has received the documents listed below and that these comply with the approval conditions detailed in our letter dated 16 April 2014

Documents received

The documents received were as follows:

| Document | Version | Date |
|-----------------|---------|-------------|
| Covering Letter | | 08 May 2014 |

Approved documents

The final list of approved documentation for the study is therefore as follows:

| Document | Version | Date |
|------------------------------------|-----------------|-------------------|
| Covering Letter | | 08 May 2014 |
| Evidence of insurance or indemnity | Gallagher Heath | 02 September 2013 |
| Interview Schedules/Topic Guides | 1.0 | 05 February 2014 |
| Investigator CV | Allana Austin | 05 February 2014 |
| Investigator CV | Tamsin Owen | 05 February 2014 |
| Other: Debrief Form | 1.0 | 05 February 2014 |
| Other: NHS to NHS Proforma | | |
| Participant Consent Form | 1.0 | 05 February 2014 |

Appendix 5: NHS Site R&D Approval Email

R&D Approval - Parents experiences of secondary dystonia and DBS

< @ .nhs.uk >
Wed 18/06/2014 11:11

To: Owen Tamsin <Tamsin.Owen@ .nhs.uk>;
Cc: Austin, Allana (2012);

Dear Dr Tamsin Owen

Title: Parents' experiences of secondary dystonia and the journey through Deep Brain Stimulation Surgery
REC Ref: 14/LO/0455
PI: Dr Tamsin Owen
CI: Ms Allana Austin
Sponsor: Royal Holloway University of London

Thank you for submitting your study to R&D Department I am delighted to inform you that NHS Permission has been issued for the above study. We have prepared a site file that will include the R&D approval letter and we will need to meet and explain your responsibilities as an investigator in order to remain compliant under the Research Governance Framework.

Please let me know when would be convenient to meet either at your office or at the R&D offices, Hospital.

As you may be aware, the Trust is working to achieve the national and local ambition of:
→ 80% studies recruiting their first participant within 30 days
→ 80% of studies recruiting the agreed number of participants within the planned study duration

For your study, the targets are recruiting your first participant by **18/07/2014** and recruiting **8 participants** in total by **02/03/2015**. If you are not able to meet these targets please do contact me to discuss an extension to the end date or other options.

You will need to send by email a monthly report of the recruitment numbers to the studies i.e. the numbers of participants recruited to your studies every month. This reporting is now a Department of Health requirement and the Trust is tasked with gathering data on every active study taking place at the organisation.

The accrual notification should be sent to: R&Drecruitment@ .nhs.uk

Stating:

1. The R&D number (RJ112/N) number given to you by the R&D department
2. The REC REF number
3. The Month and year
4. And the number recruited to the study for that month

If you have any queries throughout your project, please do not hesitate to contact me. Meanwhile, may I wish you success in your project.

Kind regards,

R&D Facilitator

Address:

Appendix 6: Email Approval from RHUL Departmental Ethics Committee

2014/065 Ethics Form Approved

Psychology-Webmaster@rhul.ac.uk

Thu 05/06/2014 13:50

Inbox

To: nxjt001@rhul.ac.uk; Owen, Tamsin;

Cc: PSY-EthicsAdmin@rhul.ac.uk; Leman, Patrick; Lock, Annette; umjt001@rhul.ac.uk;

Application Details: View the form click [here](#) Revise the form click [here](#)

Applicant Name: **Allana Austin**

Application title: **Parents' experiences of secondary dystonia and the journey through Deep Brain Stimulation Surgery**

Appendix 7: Consent Form



CONSENT FORM FOR RESEARCH STUDY

Parents' experiences of secondary dystonia and the journey through Deep Brain Stimulation Surgery

Version 1.0 (05.02.14)

Name of Researcher: Allana Austin (Trainee Clinical Psychologist)

| | Please initial to confirm |
|--|---------------------------|
| I confirm that I have read and understand the information sheet dated 26.02.14 (Version 1.0) for the above study. | |
| I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily. | |
| I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason, without my or my child's medical care or legal rights being affected. | |
| I understand that relevant sections of any of my child's medical notes may be passed onto the researcher by a clinician in the team, and that data collected during the study may be looked at by responsible individuals from Royal Holloway University, from regulatory authorities or from the NHS Trust, where it is relevant to my taking part in this research. I give my permission for these individuals to have access to specific information from my child's records. | |
| I consent to an audio recording of the interview being made and understand that it will be stored securely and destroyed after the purpose of the research is complete. | |
| I am aware and understand that the researcher, Allana Austin, may publish direct quotations said by me during the interview, but that these will be anonymised. | |
| I agree to take part in the above study | |

Name of Participant Date Signature

Name of Person taking consent
(if different from researcher) Date Signature

Researcher Date Signature

When complete, 1 copy for participant: 1 copy for researcher site file

Appendix 8: Debrief Sheet



PARTICIPANT DEBRIEF FORM (Version 1.0 05.02.14)

Parents' experiences of secondary dystonia and the journey through Deep Brain Stimulation Surgery

Thank you for taking part in the above research study

Purpose of the research study

The purpose of this study is to explore how parents experience Deep Brain Stimulation Surgery. In particular: parental experiences of caring for a child with secondary dystonia before DBS; making the decision to undertake DBS; managing the DBS process and the impact of DBS on the child and family. This information is important, because we hope it will help staff to better prepare children and families for surgery, manage parents' expectations and support families' adjustment to life after surgery.

Procedure

The study involves interviewing a small number of participants to gain an in-depth understanding of their experiences. Detailed questions and discussions are necessary to help the researcher to collect information about parents' individual experiences and perceptions of DBS and the personal meanings attributed to these experiences.

Support

If you were upset or distressed by participating in this study or participation has given you a reason to feel concerned or worried, we encourage you to discuss this further with:

- Dr Tamsin Owen, clinical psychologist within the _____ Team (_____)
- If you request you can be referred to the _____ parent counsellors

If you enjoyed talking and sharing your experiences there are online DBS and Dystonia support groups and blogs created by parents who have also been through the process:

- 'The _____ Hospital Dystonia Support Group' on Facebook
- <http://www.mydbstory.webs.com/>

Appendix 9: Service User Consultation Interview Guide

Service User Consultation – Methodology/Research Questions/Practicalities Consultation before Ethics and formal Semi-Structured Interviews

Introduce the aims and purpose of the project

E.g. Explore experiences, views and perceptions of parents of children with secondary dystonia going through the DBS surgery process
Parents views never been heard – interested in their journey through the process, often changes noticed in parents that are not captured by quantitative measures, could help lead to improvements in guidance/information/care provided by clinical team

Research Questions/aims/conceptual basis

Initial Opening questions:

- What do you think would be important to ask a parent of a child with dystonia who has - been through the DBS process?
- What do you think would be the important areas to consider?
- Anything else you would like to share about your experiences?

Discuss key areas of Interview Schedule:

- 1) Impact of dystonia on child, parents and quality of life of the family
- 2) Decision Making - Expectations, hopes and fears of the surgery
- 3) DBS Process
- 4) Impact of DBS on child (inc. motor changes) and parents

Prompt Questions:

- What is missing? Are all the important issues considered?
- Do you think it is important to ask about positive and negative experiences?
- Do you think it is important to try and capture the child's views explicitly through the parents?
- Do you think it is important to ask directly about support received from the clinical team, in making the decision and post surgery? What was useful, not useful?
- Would you feel comfortable to discuss impact on you as a carer and any experiences of burden of caring for a child with dystonia, or the added burden of going through this process?

Design/Procedure

- Who would be the best person to interview?
Should we specify who e.g. based on physical care/input into child's care?
- How would you like to be approached about taking part in the study?
E.g. the staff team discuss it with you, give you an information sheet and then I contact by telephone to discuss
- How long after receive information should we wait to contact you?

Practical

- When do you think would be best to interview parents in the DBS process?
Planning on interviewing parents at their 1 year follow up appointment
How much able to recall life before DBS as time goes on?
Balance with fine tuning of electrodes
- When do you think would be the best time to have this interview during the 2 day review?
Planning on interviewing parents when child is doing the neuro-psych interview?
Planning on having interview after the COPM when reminded of goals/watched videos?
- Interviews would usually last hour- hour and half depending on the person
Does this seem like a reasonable amount of time?

Ethics

- How would you have felt about talking to a Trainee Clinical Psychologist about the impact of dystonia and DBS?
- Would discussing the process be difficult emotionally?

Implications

- If you were going through the process again – what would be useful to know from this study?
- How do you think we could use the information gathered?

Future Involvement

- Would you be willing to be consulted again regarding e.g. development of materials, interpretation of findings and implementation?

Any final Questions about the study?

Thank you for speaking with me

Appendix 10: Participant Coding Extract and Emergent Themes for John's Transcript

| Initial Emerging Themes | Original Transcript (P= Participant; I= Interviewer) | Exploratory Comments (Descriptive, <i>Linguistic</i> , <u>Conceptual</u>) |
|---|---|---|
| <p>Positives coming out of disability</p> <p>Dystonia exists in wider family system</p> <p>Positives coming out of negative</p> <p>Raising positive awareness of disability</p> <p>Acceptance of disability</p> <p>Struggle with reality of disability sometimes – ongoing adjustment</p> | <p><i>I: And are there other ways Billy's disability has had an impact, in perhaps a difficult or positive way?</i></p> <p>P: There's a lot of positive, a lot of positive. It's a positive in a sense because (pause) because of Billy's disabilities we are probably a closer knit family. Urm (pause) not by any specific effort but by the nature of the beast as it were, because everybody helps each other. On a normal evening Billy would have gone to the toilet down here, and he would have put his pyjamas on in here, and we don't ever tell the boys they've got to do it, but (mother) and I will call in from the kitchen 'who's gonna do Billy's pyjamas tonight' and one of them will always volunteer, where if Billy didn't have his disabilities that wouldn't happen, urm so it's a positive out of the negative. Urm yeah we'd all like it if Billy didn't need help and one of the biggest positives is because of the way the triplets are with Billy all their friends are also that way. The triplets friends will come to tea, and there's no big deal about the fact that Billy has to be feed or anything, so if you like there's a wider circle of good will going on. A positive out of negative.</p> <p><i>I: What does that mean to you when you see one of the triplets helping Billy?</i></p> <p>P: I'm always very proud of them for accepting the whole situation. They all have their moments, they're 8. They all have their moments where I don't want to, and as bad as this sounds there are days when I don't want to do it, yeah, and there are days when (mother) don't, we are all human. So I would expect that out of them. I am very proud of them for doing what they do, and helping like they help. Sometimes you do think to yourself they shouldn't be doing, but the reality of the situation is that there may be a time in the future when they have to do it. I wouldn't say that they miss</p> | <p><i>Choosing to talk about positives, Repetition emphasising a lot</i></p> <p><i>Disability meaning closer knit family</i></p> <p><i>Nature of the beast, basic character of something – helping each other</i></p> <p>Siblings positive reaction to helping</p> <p>Positives coming out of negative</p> <p><u>Important to share positives, focussing on the positives, important to think about family context of disability</u></p> <p>Siblings understanding of disability - Helping others to not see disability as a big deal</p> <p>Proud of siblings for accepting situation</p> <p>Days don't want to perform caring tasks</p> <p><u>Guilt at having these feelings – meaning of parental role- shouldn't feel that way?</u></p> <p>Considering how life should have been</p> <p>Preparing for the future – reality of disability</p> <p>Try to ensure siblings don't miss out</p> |

| | | |
|-----------------------------------|---|--|
| Adjusting in family context | out on things, necessarily, because we try and find ways to make it all work, but they don't have the scope that a lot of other kids their age have, but the fact that they're triplets means that they possibility wouldn't have had that scope anyway. You know, there's not a bottomless pit of money, so I don't know, it's hard to say whether life would actually be much different if Billy was just any other child, it's really difficult to say. | <u>Family Adjusting to disability</u> |
| Questioning meaning of disability | <i>I: Could you say a bit more about that?</i> | Questioning impact of disability |
| Social construction of disability | P: Almost being a triplet it should be a registered disability, it should be. No one ever describes me as the disabled child's father I am the triplets father, and I use that, and I use that quite a lot when I explain to people, because Billy's Billy, Billy has his personality, he's one of the favourite kids at school the girls fight over helping him in school, you know he's just Billy. So (pause) Billy is Billy. So no one sort of, yeah ok, so occasionally I am Billy's dad but I'm not labelled by that. But I will always be the triplets dad, most people wouldn't even know what my name is, just the triplets dad, and they get that at school, where people just call them the boys or the triplets, and that's almost as much an affliction if you like, as you know Billy's disabilities. So we're quite a strange family really when you start thinking about it, better not think to hard. | Being a triplet could be registered charity Disability determined by how described, labelled, perceived by others <u>Disability not about physical disability – but how viewed by others and reaction of others?</u> |
| | <i>I: So thinking about disability, what does it mean to you that Billy has a disability?</i> | <u>Sense of disability meaning you lose your identity – identity defined by that label – John even started to define himself by label – internalised social construct?</u> |
| Child's perception of disability | P: First of all I've always been thankful that he was born with a disability rather than incur disability in life later on. Dr (name) laughs at this, but, until probably two years ago, urr, bit longer than that now probably, nearly 3 years ago, we used to say that in Billy's mind we're all, there's something wrong with us, yeah, he, sometimes you catch him looking at his brothers running about and doing foolish things, and it was almost like he was watching them thinking poor souls, and it really was, and I really believe that he was looking at everybody else thinking poor things, why do you put yourselves through that, and he would conduct himself in that sort of fashion. And then he had a really bad seizure, and it was almost like a switch had been turned. For the first, probably for the first six years of Billy's life, I don't really think, there was any significance in the fact he was | Difference between being born with and acquiring disability |
| Child's awareness of difference | | Child's perception of disability – others perceived as different, and pitying others |
| Changing nature of disability | | Seizure changed everything – <i>switch turned on/off, emphasise dramatic change - contrast between time points – disability suddenly mattered, child vulnerable, changing nature of disability</i> |
| Social construction of disability | | Before disability had no meaning to child's life Changing meaning of disability – perception of becoming more disabled with age Disabled because of changing age related |

| | | |
|---|--|---|
| <p>Same child regardless of disability</p> <p>Making adjustments and move on</p> <p>Social construction of disability</p> | <p>disabled, he was just Billy. As he's got older (pause), all those things that start with happen with education and everything else, and the fact that you spend a large proportion of your life fighting for things for Billy because he's disabled, that actually makes him disabled, he's still Billy and (pause) you just get on with life urm.</p> <p><i>I: Could you say what you mean by that?</i></p> <p><i>P: he's still Billy and (pause) you just get on with life urm. Its like having an old car, you don't not go out in it because its an old car, and it might break down, doesn't stop you driving to work every day or what ever you're doing in your old car yeah, you just get on with it, and you deal with the fact its an old car, and put a blanket over the engine in the winter so it will start, you just get on with it, you deal with it, its other peoples (pause) other peoples and the system that actually make Billy disabled. Does that make any sense?</i></p> | <p>activities - Fighting to access services <u>Is it lack of access to adjustment and societal reaction that mean Billy is now considered disabled?</u></p> <p>Same child with disability - <u>sense everyone treating him differently is the problem</u></p> <p>Get on with it, made adjustments, deal with it, Reactions of others/system create disability</p> |
|---|--|---|

List of Emergent Themes from John's whole Transcript

| Superordinate Themes | Emergent Themes | Page (Line Number) |
|--|--|---|
| A special but different relationship | Special connection | 6(17-18);10(10-12); 10(13-15); 10(22-33); 10(35-37); 11(3-4) |
| | Similarities between father-son | 10(17-18); 10(19-21) |
| | Providing opportunities | 1(16-21); 1(24-25); 16(9-13); 16(19-23) |
| | Treating child differently | 4(7-11); 4(19-20); 13(33-35) |
| Meaning of disability | Child's insight into difference | 3(19-23); 3(24-28); 4(1-5); 5(14-20) |
| | Questioning meaning of disability | 4(21-22); 6(36-38); 14(31-33); 15(24-26) |
| | Same but different | 4(11-15); 6(39-40); 10(32-33) |
| | Billy's just Billy – Where's the disability? | 15(25026); 16(3-8) |
| | Social construction of disability | 3(26-28); 15(1-3); 15(6-11); 15(26-30); 15(30-35) |
| | Impacting the family system | 9(13-15); 9(22-28); 13(17-21); 13(26-27); 13(28-32); 14(1-7); 14(29-31); 14(23-24); 14(27-30) |
| A difficult life before and after DBS | The negative impact of physical | 2(18-21); 2(29-33); 2(34-37); 2(37-38); 4(1-5); 15(23-25) |

| | | |
|---|--|--|
| | disability | |
| | An emotional parenting role | 1(13-15); 2(22-24); 3(8-10); 3(14-17); 6(3-4); 6(6-7); 10(23-26); 13(19-20) 13(15-16); 14(22-25) |
| | Vulnerable Child | 2(6-8); 2(21-22); 3(11-12); 6(8-10) |
| | Fragility of DBS System | 18(21-22); 6(10-12) |
| | Compromise of DBS | 17(20-21); 17(21-24); 17(25-26); 18(29-31) |
| | Systemic strain during DBS | 9(11-15); 9(19-21); 9(26-29); 9(33-35) |
| A positive attitude | Proud of child | 1(11-12); 3(17-18); 6(22-23) |
| | Positives coming out of disability | 14(8-9); 14(10); 14(14-16); 14(19-21); 14(22-23) |
| | Accepting uncertainty of life | 11(16-18); 13(6-9) |
| | Accepting unexpected life | 12(28-32); 12(36-37) |
| | Embracing unknown future | 6(34-35); 11(11-12) |
| | Life an adventure | 6(35-38); 11(12) |
| | Adventure of DBS | 6(20-22); 11(7); 11(9-10); 11(23-29) |
| | Certainty of DBS decision | 6(15-19); 6(32-34); 6(40); 16(14-18) |
| | Positive hospital experience | 7(3-4); 7(15); 7(15-17); 7(20-23); 9(11-12) |
| | Hospital experience better than expected | 7(18-19); 7(35-37); 8(2-5); 8(8-9); 8(13-14) |
| | Positive experience of nursing care | 8(1-2); 8(11-12); 8(25-27); 8(29) |
| | Positive coping during DBS | 7(7-12); 9(14-15); 9(33-34); 10(2-4); |
| | Adversity increasing resilience | 10(5-6); 10(7-9); 11(5-7) |
| | The life-altering impact of DBS | Emotional meaning of change |
| Removing barriers created by disability | | 5(4-8); 5(14-17); 5(23-25); 5(30-34); 6(5-6); 17(8-10); 17(17-19) |
| Providing normality of life | | 5(7-8); 5(9-10); 16(28-29) |
| Unexpected changes | | 4(27-29); 16(26-29) |
| Reducing carer distress | | 17(1-16) |
| A new perspective of child | | 21(24-25); 3(17) |

Appendix 11: Extracts from Reflective Journal

1st September 2014: Reflections before interview 4

I have now completed three interviews, and I am very aware that this is my half way interview, and I am now feeling pressured to ensure I am capturing rich data. The previous interview with Laura was a more challenging interview in terms of how guarded Laura was in expressing experiential and emotive accounts. I think this had led me to feel more pressured to ensure this data is rich in idiographic experience. I therefore listened back to interview 3 to reflect on my developing interviewing style. This helped me to move away from a self-critical position focused on what I had been doing wrong in the interview, and to accept that parents will be different in their ability to express emotions, and open up to a stranger. It also reaffirmed the importance of asking experiential questions (e.g. tell me more, what were you thinking at the time) to ensure interviews are grounded in phenomenological experience. On reflection I think I experienced Laura's interview overly critically, because there was a stark contrast between the highly emotive accounts of Julia and Rachel, and Laura's more reserved and factually focussed narrative. I am curious as to what the next interview will be like and which experiences will be most salient.

I think I am growing in my ability to adopt an interviewer position and with each interview am finding I need the interview schedule less. I wondered if this will help me in my next interview to use the interview schedule flexibly. This may be important for Linda, because I am aware through discussion with Tamsin that this family experienced complications with the DBS setting which resulted in removal and subsequent re-implantation. Linda will therefore be drawing on DBS experiences from multiple time frames, which will mean my linear interview structure, may not map onto her experience. With that in mind I have been re-reading the interview schedule and IPA guidelines on completing qualitative interviews, to allow me to respond flexibly to Linda's experience.

2nd September 2014: Reflections after Interview 4

As anticipated the interview was challenging because of multiple time frames. However, I think the interview went well in terms of Linda's ability to share her experiences. I was struck by the striking disparity between Philip's successful DBS outcomes, and the deterioration in Philip's functioning following infection and removal of the system. Linda spoke emotively of the meaning of changes and the challenges of DBS. I left the interview feeling inspired by the potential for DBS change, which I think mirrored Linda's striking sense of optimism and strength in the face of adversity. Interestingly Linda described herself as the 'main carer if you like', and did not appear to identify with this label. This made me consider how none of the

parents so far have identified themselves as a carer, and I wondered about the societal discourse of this label and the meaning it holds for parents. The term carer burden is also frequently documented in the literature. However, although Linda's and other parent's' interviews have described considerable stress and parenting demands, they have never used the word burden to describe their experiences.

This was the first interview I completed about a child who has significant communication difficulties. I was aware of Philip's dystonic severity prior to completing the interview, and was expecting this to be a significant feature of the child and parent's experience. However, during the interview the mother did not bring up Philip's communication. This highlighted how I held assumptions about the importance of verbal language and communication as central to experience. However, the narrative that emerged in the mother's account was one of being attuned to the needs of her child, and having their own way of communicating. This highlights the importance of reflection in increasing my awareness of assumptions that I may not have been aware of prior to completing the interview.

Appendix 12: Summary Table of Themes and Additional Extracts

| Subtheme | Participant | Quote |
|--|-------------|--|
| Superordinate Theme 1: A Difficult Life with Disability | | |
| Trapped Inside an Uncontrollable Body | Julia | She'd be in the toilet and her arm would wack against the wall (gesturing) and you'd hear the noise and the shoulder dislocating |
| | Rachel | The main disability she has is her arm. She has no function in it and it's, it does weird things. And its urm, it upsets her because it does weird things when she's not expecting it and it might hit someone in the face or catch hold of hair, or do stuff like that |
| | Laura | You need to be able to, to do really. Just go and get yourself a drink and he can't, or it's very, very difficult, because you know the shakiness of his hand and things like that. I mean mobility wise because he can, he can walk, he gets tired. So he can't walk far before he gets tired |
| | Linda | If it like you said right 'we'll go out in car', even when you say something, its like him getting excited or anxious, well I don't know about anxious cause I don't think he really, or if he gets upset, but mainly it's if you're gonna do something, if you're going to walk with him, or say 'right come on we're going to school', or put your shoes on, straight away the tenseness, you know he goes quite tight, pretty much most of the time |
| | Theresa | She tends to pull to the right, she also, urm, her tone is very variable, its not always the same, sometimes, well mostly shes stiff, her hands are stiff, her arms are stiff, urm and as she grew and her limbs became longer and urm, it became worse because it was all exacerbated, and every time she has a growth spurt everything becomes worse, becomes tighter, pulls more |
| | Natasha | The only way she could communicate was by crying, it was horrible |
| | John | When he was very small obviously he didn't walk, so his development was well behind his age. He'd walk a little way, fall, so that impacted. He's never really been able to feed himself, there's been spells where he could sort of do finger food, depending on what it was, what the texture was |
| | Caroline | She, obviously her extensions were much more enhanced through either excitement, or upset, so at high emotion, and very difficult to control, and if she had a tantrum trying to get her in to a safe zone was really difficult |
| A Restricted Life – Child and Family Perspectives | Julia | When they were little girls would come in and play with dolls, because Imogen was able to do that. But now she's older they want to go out and about and do stuff. If its not accessible, they're not going to hang around Imogen's house to do stuff. So that has been a big negative, that's emotionally upsetting for me |
| | Rachel | We can't as a family go out on a really long dog walk together, because we have to take all her needs into consideration. I know that sounds really, really urm selfish |

| | | |
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| | Laura | But he hasn't had any friends out of school. I think he uses social media, so there's that sort of friendship. But he hasn't had any friends. He's at Sixth Form College, which is a very good sociable sixth form college from what everybody else says. But he doesn't, hasn't got any friends and I haven't got to know any parents or anything like that |
| | Linda | I know sometimes (name of husband) Wade say it doesn't stop us doing anything, it probably does sometimes (pause), because sometimes it's easier not to do things that are difficult. I suppose that's quite obvious as well, because if he didn't have dystonia you would do a lot more things, just sometimes things just seem that much harder |
| | Theresa | I mean it affects everybody hugely, absolutely hugely urm, just because everything takes so long, feeding for instance, you know dinner time, you can't have a quick snack, you have to prepare it, and then you have to feed her, and she can't do it herself, so you can't do anything else |
| | Natasha | my relationship ended sort of like three months ago, because I couldn't cope with it, I couldn't cope with a relationship, trying to deal with Ivy, my other two kids, it was when Ivy was in hospital for like 10 days, I just I wasn't, I couldn't do. Not because, I said to him, its not fair for me to try and pretend this relationships ok when I don't have time for it, and I'm not going to be with someone when I don't have time for them |
| | John | they used to play football on Saturdays and Sundays, but they decided they didn't want to do it anymore, because it wasn't fair on Billy to sit out in the cold watching them to play football, or one of us couldn't go as well so then we weren't together. So it does affect everybody, Billy's disabilities affect everybody |
| | Caroline | I think the hardest thing is maybe friends, she's got no one really urm age appropriate for her that she can play with, urm. So that's hard |
| Emotional & Physical Demands of Parenting | Julia | Getting dressed. I mean especially her shoes, her toes would curl up, and it could take half an hour to get one shoe on, and you're on your knees and sweats pouring off you, its really hard going and I would know that she couldn't, her dad would scream at her sometimes for things, but I thought she can't bloody help that you know |
| | Rachel | Fortunately I am incredibly organised and I always haven been (nervous laughter). So I think from that point of view it's just been another thing that I've had to organise, but it does all fall on me |
| | Laura | But that sort of thing, the freedom to sort of, not worry. I mean, we are reluctant to leave him for more than a few hours on his own |
| | Linda | But I think with Philip its pretty much you seem to do one thing, and then you have to do another thing, and then after you do that there's another thing because your really having to do everything, whether its dress, feed, toileting |
| | Theresa | Its hard, its hard for her, because her bodies fighting against her every step of the way and you just want to change that, and you can't |
| | Natasha | Urm, and then yeah so I suppose leaving them a lot to be able to go up to the appointments. Again, appointment on Friday, but it's their last day at school, its (name of sibling) birthday, but I've still got to |

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| | | go to London, I've still got to go for an appointment |
| | John | Which has been really really, as a parent really difficult to deal with, because you see him going up and achieving more and then by no fault of his own or anyone else's, he's back to next to nothing, and that's really hard to deal with, really hard, its horrible |
| | Caroline | Or 'god its taking me forever just to get her up in the morning', because I've got to get her strip down, get a paddle, contend with those movements and get her dressed, and into a wheelchair, and everything's very time consuming, and again it just becomes very stress related, you become very fatigued, we become absolutely exhausted, which again then adds to the anxiety as well. It all kind of, I always say it kind of all wraps itself around each other and becomes one big mess |
| Superordinate Theme 2: The Meaning of Disability and Normality | | |
| The Impact of Social Comparisons and Feeling Different | Julia | But the spasms, the effort trying to do it, makes her spasms go bad. Its, she would never be able to do normal swimming |
| | Rachel | I, I know, I've got to think about it more because my son is three years younger, and is so capable. And, and it just highlights all the things that she can't do really. Urm (pause) and so, it's a huge impact. We have to think about things all the time |
| | Laura | I think he finds it very frustrating, very frustrating, urm (pause) not being able to do what everybody else, or just about everybody else that he meets takes for granted |
| | Linda | I just find it strange, I think because I see people at home and they are wittering about something, and I just think, I suppose that's life, you just think if you ever had anything that, you wouldn't have a clue |
| | Theresa | I suppose. You know she's grown up with it, and so she's used to it. It doesn't mean, when she was younger she used to say 'I wish I could walk, I wish I could run, I wish I could play with other children' you know |
| | Natasha | couldn't relate to any of them, as such purely because Ivy was so young, and also most of them are primary dystonias as opposed to secondary |
| | John | And he had quite a mature understanding of his disability. He's car mad, absolutely car mad, but he knows, the reality of ever being a racing driver are quire remote, but he'll tell you he'd need a special car. So he's got that understanding that you know he's not gonna walk out, jump in a car and just drive it, he's gonna have to have adaptations |
| | Caroline | Urm, it's, its made even more apparent since we had (brother), She doesn't get, she doesn't appear to get depressed or low about it, but she will say 'well (brothers) doing this, why can't I?', 'well (brothers) allowed to do that, why can't I do that?' |
| | Julia | I went up on the thing, and I cried the whole balloon ride, people put their arms round me, and I just glared at that pilot the whole time and just called him everything under the sun, I don't know. Oh I was just, what a chance to do something like that and he's ruined it. I was really really upset. I mean I, the actual experience of the trip was great. You were going over these fields, almost touching the top of |

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| Treated Differently - The Social Construction of Disability | | the grace and back up again. And she would have just loved that. And we will never do it again. Because were not, once in a lifetime chance. So that was that. |
| | Laura | he'd actually gone away on a, a Christian camp weekend and apparently some of the children there, who are children the same age, were quite unpleasant to him or, I'm sure they didn't mean it, but talking about him and I suppose he feels sometimes people talk behind his back about how he walks or how he talks and that sort of thing |
| | Theresa | It felt exactly the same as she did, I wanted to cuddle it, and cry, it was just amazing, amazing, but for her to be actually able to do that, for those people to be able to say, because a lot of places you go and they go 'oh you cant do that', you know 'you can't, that wont be possible' |
| | Natasha | Its ignorant people...Like when she had the NG tube before, I'd take her shopping and people would be like staring and someone said to me is that an oxygen tube, and you're thinking 'nice that your asking, but no' |
| | John | he's still Billy and (pause) you just get on with life urm. Its like having an old car, you don't not go out in it because its an old car, and it might break down, doesn't stop you driving to work every day or what ever you're doing in your old car yeah, you just get on with it, and you deal with the fact its an old car, and put a blanket over the engine in the winter so it will start, you just get on with it, you deal with it, its other peoples (pause) other peoples and the system that actually make Billy disabled |
| | Caroline | So for us, it can be a battle, people stare, you know we get that, (husband) likes to take Emily out in her power chair or her walker, especially her walker, round Sainsbury's. Our local Sainsbury's is brilliant, because we're regular customers, they're used to seeing Emily, so again its Emily, they are absolutely fine, its all 'hi Emily' its lovely. But he takes her in her walker in rush hour, and basically it's busy, and you can imagine you've got some people who think its great to see her whizzing round, she's disappearing down an aisle, so you might see her disappearing, or some people sit and stare at her kind of thing, but that's the point isn't it it's a big thing for us, its about kind of sod it why shouldn't she be able to run up and down Sainsbury's if he wants to. So yeah its, its (pause) there's that stigma there, and it will always be there |
| Making the Choice to 'get on it with' and the Process of Acceptance | Julia | Well, I'm just wondering, because it doesn't always happen at birth. It can happen after birth can't it. Kind of guessing that the yeast infection might have kicked in and caused some damage. But, I don't think anyone can really put their hand on their heart and say that so (pause), not worth dwelling on it really, if somebody said 'yes that's what did it, and we'll pay you compensation' because it was their fault, it wouldn't make her any different would it (pause) |
| | Rachel | You can't spend the whole rest of your life under the duvet can you? You have to kind of make a decision to get on |
| | Laura | Family and friends were very sort of, admiring of it and whatever. I mean last year, at the same time that urm Will was having, due to have the surgery, and we had the wait for surgery, my eldest was in Afghanistan as well, people would say 'oohh you know you've got one son there, and one son doing this', yes but you know that's just the way it is |

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| | Linda | I wouldn't, I don't really find it, I wouldn't say I find it difficult, because its been, I've had to do it since Philip were born, so its kind of, I don't know you just get on with it really |
| | Theresa | mean its devastating, its devastating for anybody, urm and it yeah, it its changes your life completely, its changed my life 100 times over, and if I could change it tomorrow I would, but you know you can't so you've just got to make the best of it |
| | Natasha | I took me, it did take a while for me to actually really believe that no its not my fault, I didn't know I had it, and even if I did I'd still have had her, I'd have just been more aware and I'd have been able to do more to help her, but I didn't know so how can I beat myself up, for something I didn't know |
| | John | There's a lot of positive, a lot of positive. It's a positive in a sense because (pause) because of Billy's disabilities we are probably a closer knit family |
| | Caroline | I think its becoming more apparent as Emily gets older, urm, that that we need to take more of a break. I think its something that we've learned as Emily is getting now older, more grown up, that things aren't going to change in a massive dramatic way for her, and actually me and (husband) need to have a little bit of time to ourselves |
| Overcoming Disability and Taking Control | Julia | have just done lots of things with her, and tried to really stimulate her as much as possible. We've travelled a lot, gone lots of places, she goes skiing, been doing that for about 9 years now, travels a lot, used to take her swimming and horse riding |
| | Rachel | It's just trying to be one step ahead of all the things that might happen in any scenario really and just trying to think about her constantly. Even like, if she went for a sandwich downstairs, can she open up a packet, you know all of those things. So all the tiny little things that you don't really think about it, because you take them for granted, suddenly are in your life and you have to make time and plan for them really |
| | Laura | we just had to be proactive really, we don't really feel we had any help or support from anybody really particularly. Urm (pause) so we took it into our own hands really, and urm (pause) he started going to Conductive Education |
| | Linda | I think, it is very demanding, and you kind of never stop, but I don't know if I think, certainly have to be positive, because if not, I don't know. I don't, (pause, sigh) |
| | Theresa | She's always had corrective seating, she's been in a line board since she was a tiny tiny baby, she has a standing frame, all those type of things, to try and make the best of her posture |
| | Natasha | Urm, she goes to school five days a week, very much keep her normal, by doing what a normal kid her age would be |
| | John | The desire for Billy to fulfil his full potential, whatever that might be, is quite important to us, not because we're pushy parents or because we expect miraculous things out of any of them, but by the nature of Billy to push himself as far as he pushes himself |
| | Caroline | Its really important that she gets to experience a rollercoaster, its that feel that you get, from the adrenaline, the rush, that urm, she gets to go on a trampoline or on a swing, its important for her learning as well, so that she's got something to relate to... for us, these are all the steps any child |

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| | | would take in their learning |
| Superordinate Theme 3: An Emotional and Uncertain DBS Journey | | |
| Facing the Uncertainty of Decision Making | Julia | she was up for it from the start, I kind of was as well, but obviously you have reservations knowing that if there's a slight slip or, knowing she could be more damaged, but you just have to (pause) give it a shot and just hope it works ok |
| | Rachel | I just, I just had to base that on the fact that, these people were experts and they had done it before and that they were saying you know it's not a significant risk. So that's all you can go by really isn't it, yeah, mmhh. |
| | Laura | And yes it was scary the thought of Wade having the surgery because the fact that urm (pause), yes he's got a very good IQ and urr his disability while it is what it is (laughter) the fact that he could be made worse was, was a worry, was a worry |
| | Linda | The surgery's the surgery ain't it. I don't think so, its not like anything comes out set in stone, it's different for everybody |
| | Theresa | but we moved on and we decided it was worth trying because I think in life if you try something and either you don't like it or it doesn't do what you anticipate, you've tried, but if you don't try you never know, and I think regret or looking back on things and saying 'I wish I had' is far more painful, more difficult than not trying them at all. |
| | Natasha | Very scary the mention of brain surgery on a child is scary, my biggest thing that obviously sat in my head as biggest fear as a mum, was if anything goes wrong its my fault, because I have chosen to let her have the operation, and would I be able to forgive myself if anything went wrong |
| | John | anything that would give a glimmer of making life easier for him in the future had to be considered |
| | Caroline | think that was a big concern, that what if we'd gone through this and it all has to come out again because its rejected, well I don't know what, we didn't, I suppose we didn't have any answers for that, we had to just risk it, and hope that everything would heal, and nothing bad would happen |
| Struggling with the Unknown and Unexpected | Julia | Well, we were trying to put things on hold, thinking like we won't go on holiday this year because we might be going down, and putting other things on hold you know. But there was no, it was just a long process that dragged out you know |
| | Rachel | because I don't know what I was expecting, but to have this huge scar and it is massive. And also, I think as well, because when we see scars in our lives, we see a nice tidy scar, don't we, I think. This one had almost Frankenstein stitches, do you know what I mean, which Megan had as well, but it wasn't so bad in real life as it was in that photo, the picture. So I think that was probably shock, actually about, 'oh Christ this is really going to happen and that's what she'll look like'. |
| | Laura | So that was a worrying time for me, thinking well he should be eating and drinking and getting up and about. So yes it was a little bit longer than we'd been led to believe |
| | Linda | Obviously then he got in for the surgery to take it out and then re, put it all back in. So that was a really |

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| | | worrying time, we didn't know what kind of to expect, and it weren't like having it done the first time, where you decide to have it done and you kind of prepare yourself. |
| | Theresa | It was just huge relief, relief that it wasn't due to the surgery or the DBS. And I think if I'd been pre-warned then it would have been alright, and id have been able to say to her, or I would, we probably would have talked about it before hand, 'don't worry this is only a side effect of the anaesthetic, this is nothing to do with the DBS'. You know it's a worry gone, because its not even entered your mind. So urm you know that was one thing that was very very strong in my mind and recollection, you know that feeling, was awful, awful |
| | Natasha | genuinely thought Ivy was going to die. In my head my child was going to die, and I couldn't, how could I say that to people. I certainly, I didn't even tell Dr (name) or anyone, told them afterwards, yeah you know I thought you were going to tell me that she wasn't here, because I could laugh about it then, but it was a genuine fear that my child was going to die. How I got over that, and allowed her to still have the surgery, I don't know, I don't know. |
| | John | I was expecting Billy to be poorly. Urm (pause) I was expecting it to be very sort of custodial being in a, a hospitals an institution, but if I wanted to go downstairs and get a coffee, I would just say to one of the nurses 'I'm going to leave Billy I've put the sides up on the bed, can you just keep an eye on him, and it was never 'phhh but yeah', it was 'yep of course', |
| Emotionally Overwhelmed by DBS and the Hospital Experience | Julia | Well I think they might perceive me, to be, well I probably did have a bit of tension in me. But I think I was trying to do lots and lots of things, to compensate for the, you know, just keep busy, and probably was rushing about like this, and probably looking stressed |
| | Rachel | We went (pause), we went down in the morning, and fortunately this nice nurse came along right as she was about to go in, and said 'why hasn't she got her stockings on', Oh everyone had forgotten about those. So I'm thinking 'oh my god, forgot about those, bloody hell, what else have you forgotten'. Do you know what I mean, Ahh (squeal), like that |
| | Laura | Yes it was very worrying, and coming into hospital and urm (pause). Yeah I mean it is scary, anybody having brain surgery |
| | Linda | But that were really a tough time...I think it was just cause he wasn't eating, he was like on drips, and he just, we'd not really seen that. So that was not good, and seeing him, seeing him not, he was pretty much doped up most of the time |
| | Theresa | I think the emotional side of things is much harder, much harder, cause you don't know how you're going to feel, and I'm sure I react and Charlotte reacts different to all the other people that have it, but I think, I think there should be more on how emotionally you may feel...like before the surgery you'll probably feel nervous and scared...and this is quite normal |
| | Natasha | It was really special, I felt like, It was like she had just been born, a really special, a really special moment |
| | John | Relief, relief obviously, you'd be a fool not to feel relief |
| | Caroline | its always horrendous anyway to put, because obviously we go down with Emily when she's had to be |

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| | | put out, and its quite a distressing moment. So that was always a concern (pause) that was a worry. Urm, because we knew that that was gonna have to happen again, so yeah, that was probably one of the biggest worries as well and how Emily was feeling at that moment |
| Coping with DBS - 'it's the people who get you through' | Julia | It's a long journey, for a short time, so just make the most of it, and don't make it hard, make it enjoyable; because it was an opportunity to do stuff you wouldn't be doing at home |
| | Rachel | So on the Saturday I did, I left her, I went out with my son and we had dinner and we had a wander about. And I felt a lot better when we came in on the Sunday. I'd got away |
| | Laura | Urm, yes it would have been quite nice to physically have met some families, before hand, who had gone through it. And to urm yes have that, have that support on a face to face basis |
| | Linda | They just took over, did everything. Which to me was great, because I wouldn't have wanted, I'd have rather that happened at that time, everything had changed, he were on that many different medication, I wouldn't have a clue anyway |
| | Theresa | I think the main thing that helped me cope was all the nurses are just amazing, and the team. You know everyones so kind and so lovely and you know that makes up for a whole multitude of stuff |
| | Natasha | When we're at the local hospital, I'm not so keen, I don't tend to leave her, but if we're at the Hospital. I'm quite happy to disappear and leave her because I know she's in the best of hands she can be, so actually being there isn't a problem |
| | John | It was an intense period in the hospital, but it wasn't unique, which I think for some people it would be possibly first experience of that, so although it wasn't unique, the environment was unique and almost an adventure |
| | Caroline | And we chose to do it, because we felt again it was really important to make it a family moment, so it was a case of ok we'll make it into a family moment, we'll take pictures before hand, and family pictures before hand, and we did all her make up, and we all got dressed up, and we made it a bit of an event, and then basically we were like ok you ready, and that initial chopping of the hair, you're like oh my god, and Emily's absolutely creasing up in the mirror |
| Superordinate Theme 4: The Experience and Perceptions of Change | | |
| Hope and Realistic Expectations of Change | Julia | Well she thought it would make the spasms less. I don't think she thought they would go away completely, but, they may get a good bit less, and that would be a big plus for her |
| | Rachel | And I think we had very realistic expectations as well we weren't hoping for her to suddenly be able to write with her left hand (nervous laughter) |
| | Laura | Our hopes and really I think, the reason we went ahead with it was you know...the same as for any parent, for Wade to be able to live independently, you know on his own, without needing any help from anyone else to do |
| | Linda | They're probably always more than what you say, to whether it be the team or anybody |

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| | Theresa | So urm (pause), giving her the best possible outcome long term, was my most important aim and obviously to alleviate discomfort and pain, because as a mum its awful to see and you know you want to help |
| | Natasha | I know one of them was her personal care, like with changing, that, we hit that at the three month |
| | John | Listen to everything that all the professionals say, but keep a balance on what's realistic in your day to day life, and don't expect too much. Then whatever happens it's a bonus. Keep it realistic |
| | Caroline | we hoped her arms would release a bit more because she was also scratching the living day lights out of herself from these movements (scratching chest), so we hoped this kind of accidental self-harming would calm down, and the hands would be a little bit more relaxed |
| Significance of Positive Changes – 'it's everything' | Julia | She must have been bad to have to ask for that, that might have been what we'd have to have happen, been on something, or some other drugs to help, where as she's not now |
| | Rachel | because she used to go and see somebody to help her with the pain and stuff all the time, and now she probably goes once every 6 weeks and that's when she's done something different that's upset her muscles rather than the actual dystonic pain. So that's really good |
| | Laura | Yes. Urm (pause), I think it's helped a bit. Urm (pause) I think he is less shaky, on the right hand side. Urm (pause) yeah I think he can, yeah, hold a drink better. I think he can drink from a cup half full, not full (pause), better |
| | Linda | It just feels like we're just getting, bit back (pause) back a bit, to a bit more of a normal life, I don't know what normal is, but in our kind of, just getting back to what we used to do |
| | Theresa | And the difference she was initially was really quite pronounced, because she'd gone from very fisted very tight arms, to them being quite lose and her hands being opened. And so that side of things was really quick, and amazing |
| | Natasha | she eats which is something she could never do before, if ever you put a spoon into her mouth she just couldn't open her mouth because the dystonia was just closing it shut, So yeah small things, but then the small things when you're dealing with a physically challenged child, the small things make a huge difference, like being able to sit her on my lap without dropping her because she's so stiff, its special like being able to change her, again before her legs were very side swept and very close together, very scissored, and now she keeps her legs open, the only time she keeps her legs open is when she's misbehaving when I'm trying to change her. And she, in the last couple of weeks started rolling over again, which is a huge huge thing |
| | John | On a personal level things like him giving me a kiss before I go to work in the morning, yeah, being able to wish me a happy birthday, being able to tell you what he wants to eat, they're all really big things. It's everything |
| | Caroline | it was a massive thing to see that she wasn't getting caught up so much, and getting stuck and tight, and we started to notice, I think the, the biggest thing was, she started to, she was picking things up, so I think there had been a flannel on the side which she had managed to pick it up with her hands, drive into the hallway, and drop it into a boot (smile), which was just the control for her to do that, just |

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| | | doesn't happen |
| Making Sense of 'Quieter' Changes than Expected | Julia | The way she's gone through, growing and stuff now, there's all different things that can impact how things are isn't it |
| | Rachel | I think, I think, you know he's disappointed about the arm, I think we all are. I think that's been really upsetting for us, that actually after everything (exaggerate word everything), so far, we haven't seen any benefit |
| | Laura | we haven't had the miraculous results, which we weren't expecting, but I think we'd hoped that the results would perhaps be, we'll know when he does the tests today and tomorrow whether there have been improvements, and I think there probably have been slightly improvements, but nothing major |
| | Theresa | No, not particularly. I think things, have probably lessened, like maybe the toe curling has lessened, but every time she has a growth spurt things go do-lally anyway, so |
| Compromise of Change and an Ongoing Struggle | Julia | And I said Imogen thinks since she's last had her adjustment on her DBS that things have got worse, and things aren't right. So she's obviously worried that maybe she should come back and get it changed |
| | Rachel | She doesn't like, her batteries in her tummy, she doesn't like it, she doesn't like it, She thinks everyone can see it, when actually I don't think you can |
| | Laura | I think he finds it frustrating sometimes, I mean well charge up, having to charge up |
| | Linda | And the other is, we do it, probably been doing it yesterday, you're kind of wanting things, to be quicker, to see something, but its not, you know we've learnt that it don't work like that (laughter), and it it does take a long time, |
| | Theresa | Every time they turn her up, the stimulator up, she has an initial honey moon period where she's much better and then it beds in and goes back a bit, that's quite difficult cause you think 'yay, no', you know 'yay, no' the whole time |
| | Natasha | everyone in general is happier, life is I wouldn't say is necessarily easier because there is always going to be issues in life, especially there's always, life's never easy |
| | John | It can still be a challenge don't get me wrong, but nowhere near. You have spells, he caught a virus a few weeks ago, urr maybe six weeks ago, and really knocked him back, urr (pause) and so you're, you're having to deal with aspects of the past over again, and when you've had such good results and things drop back a bit, it it can be quite difficult to deal with |
| | Caroline | We're able now to take a bit more of a breathe, but because she's still very very needy and very full on, she's extremely disabled, but there are areas she's improving so for us that's just a bonus |
| A New Perspective Looking Forward | Julia | She's been through a big procedure, and she's stayed positive all the way through really. So I think that's made her a better person. You know. I think, nothing puts her off, and having done that, I think that's reinforced that even more. I think that's what I'm trying to say |
| | Rachel | I am probably even more tolerant with her, because I can't believe what she's gone through, do you know what I mean. I think that urm (pause), yeah I have, I've spent a lot more time with her, helping |

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| | | her with coursework, and trying to help her really, be the best she can be, do you know what I mean, just by sort of yeah being with her and helping |
| | Laura | But we are very lucky that he can walk, so really it's the fine motor skills, that's, that's what he needs to be independent |
| | Linda | I do sometimes think back, I don't know how I got through, you know, thinking back. I certainly wouldn't want to go there, and wouldn't want anybody else too. Yeah, thinking back I do think, that I'm stronger |
| | Theresa | you can't turn them up to full potential initially because obviously the brains got to get used to them, so I'm still hoping there's going to be an ongoing impact for years to come |
| | Natasha | so you don't still, well now I don't think much about how bad it was, because things are good now. Urm and I'd, like when if I moan now that things are bad, its urm, you think why because they're not anything like they used to be |
| | John | We now know how much he understood of what was going on around him, but couldn't participate because he couldn't communicate... We now know because he can speak Spanish, Hmm (laughter), which is quite weird because we never knew how much he was understanding of English, Spanish, or anything. So this last year has been quite an eye opener for us |
| | Caroline | its, been a huge thing to me and Col, it has made things easier, its made us proud of our daughter, its something that we can say to other people look now what she's doing, like I said it's a feeling that, there was such a noticeable difference when things went wrong and it was quite distressing for us |