FROM GENERAL DEVELOPMENTAL DISABILITY 
TO 22Q11.2 DELETION SYNDROME: 
UNDERSTANDING PARENTAL EXPERIENCES 
A Mixed Methods Analysis 

“I was meant to have him on this journey, whatever this journey is going to be” 

Jane Goodwin, BPsych (Hons) 
Thesis submitted to the University of Newcastle 
for the degree of Doctor of Philosophy 
Submitted 6th February 2017
Declarations

Statement of Originality

The thesis contains no material which has been accepted for the award of any other degree or diploma in any university or other tertiary institution and, to the best of my knowledge and belief, contains no material previously published or written by another person, except where due reference has been made in the text. I give consent to the final version of my thesis being made available worldwide when deposited in the University’s Digital Repository**, subject to the provisions of the Copyright Act 1968.

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Signed,

Jane Goodwin
Statement of Authorship

I hereby certify that the work embodied in this thesis contains a published paper of which I am a joint author. I have included as part of the thesis a written statement, endorsed by my supervisor, attesting to my contribution to the joint publication. See below.

Signed,

Jane Goodwin

I attest that Research Higher Degree candidate Jane Goodwin has contributed to publications for which I am a co-author. For all publications, where applicable, Jane has:

• Contributed to the development of research questions
• Contributed to research design and methodology
• Contributed to the development of data collection tools
• Managed data collection procedures
• Conducted interviews
• Cleaned data
• Led all data analysis
• Led the writing of each manuscript

Signed,

Dr Linda Campbell
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Below are the details of thesis chapters that have been published or are under review for publication.

**Chapter 3**


**Chapter 5**


**Chapter 6**

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“Those who have a 'why' to live, can bear with almost any 'how'."

— Viktor E. Frankl, Man's Search for Meaning

« Les plus belles choses dans le monde ne peuvent pas être vu ou touché, ils se font sentir avec le cœur. »

— Le Petit Prince d'Antoine de Saint-Exupéry
Abstract

The negative psychological impact of having a child with a developmental disability has been well-recorded. Although positive psychological constructs (e.g., psychological growth) are beginning to receive more attention, they are relatively unexplored in parents who have a child with a developmental disability. The aim of this thesis is to describe the experience of parenting a child with a developmental disability, particularly the more positive aspects. One developmental disability that offers unique challenges and is poorly researched is 22q11.2 deletion syndrome (22q11DS). Therefore, this thesis specifically focuses on 22q11DS and uses a concurrent mixed methods design. Although children with 22q11DS have a characteristic neurocognitive and behavioural phenotype, the disabilities experienced by these children and the challenges faced by their parents can be extrapolated to the wide range of developmental disabilities.

Considering the wider experience of parents of children with a variety of developmental disorders, predictors of psychological growth were examined through a cross-sectional survey. Many participants experienced at least some psychological growth as measured by the Psychological Wellbeing - Post-Traumatic Changes Questionnaire (Joseph, Maltby, Wood, Stockton, & Hunt, 2011). A regression model demonstrated that increased: a) use of positive reappraisal as a coping mechanism; b) parental perceptions of coordinated and comprehensive health care; and c) child’s age were associated with greater psychological growth.

Given these findings, the thesis then sought to understand whether psychological growth despite potential distress was possible from parenting a child with a poorly understood and often tardily diagnosed developmental disability, 22q11.2 deletion syndrome (22q11DS). Semi-structured interviews informed by the methodology Interpretative Phenomenological Analysis (IPA) explored the experience of parenting a child with
22q11DS at two different stages of parenting: a) a young child with 22q11DS; and b) an adult child with 22q11DS.

As 22q11DS is a condition that has wide variability in presentation, with little knowledge about the impact it has on parents’ psychological wellbeing, IPA informed a detailed and open exploration of this phenomenon. Parents who had a young child with 22q11DS provided rich data that was interpreted as anticipatory traumatic distress, systemic stigma, confusion at professional smoke screens, and ‘not knowing’. However, they were able to redefine their unanswered fear, guilt, loss and grief in these early years through hope for the future and a perceived opportunity to become better people.

Similarly, for the interpreted experience of parents whose children were now adults with 22q11DS, stigma remained a constant distress, and feelings of guilt, loss and grief persisted throughout the years. Progressively, stigma undermined independence, friendships, and instinctual judgement. Ill-informed hierarchical structures experienced as layers of obstruction and ignorance pragmatically replaced hope and triggered angry advocacy for their child, which was used proficiently in educational, health and societal contexts. In time, looking backwards, they came to value their unique accomplishments collected on their journey with 22q11DS, and in turn, consciously valued authentic ‘self’. Psychological growth was viewed as part of the journey that juxtaposed ongoing distress. It was identified through a metamorphosis of empathy, humility, gratitude, and pride.

This thesis provides a unique contribution to the literature by delineating factors that contribute to psychological growth in parents who have a child with a developmental disability. Further, the under-explored journey of these participant parents who have a child with 22q11DS is uncovered, highlighting both negative and positive psychological outcomes. In conclusion, this thesis highlights a number of areas for further investigation and intervention. It demonstrates: a) the need for clarity in positive psychological research; b) that
positive psychological outcomes are realistic for parents whose children have a
developmental disability and should not be neglected; c) that hierarchical and medical
frameworks often poorly support the long-term biopsychosocial needs of families living with
lesser known developmental disabilities; and d) healthcare professionals are well-placed to
promote positive psychological outcomes.
Chapter 1

The experience of parenting a child with a developmental disability:

A review of the literature

1.0 Chapter statement

This chapter provides an overview of the literature surrounding the psychological impact of parenting a child with a developmental disability. This is not intended to be an exhaustive review of all research pertaining to parents who have a child with a developmental disability. Rather, it outlines relevant theories and literature to justify the research theme of this thesis. Each chapter contains further literature as appropriate to its particular topic.

This chapter begins by emphasising the potential for psychological distress in this population, including the risk of grief, burnout, and traumatic responses. However, these difficulties can be a platform for positive change. Therefore, in section 1.2 I highlight positive psychological theories and research that may provide insight into the phenomenon of positive psychological change in parents who have a child with a developmental disability. Although there is limited data in the population of interest, there is early evidence that positive psychological change is possible as a consequence of adverse experiences. Next, a narrative review of the literature surrounding positive psychological constructs as outcome variables in parents who have a child with a developmental disability reveals significant limitations in the current literature. The chapter concludes with a rationale for my thesis. The broad aim of this thesis is to provide insight into the experience of parenting a child with a developmental disability, particularly the more positive aspects. This original contribution to the literature will expand the current knowledge of a relatively unexplored phenomenon.
1.1 A Group at Risk

The term “developmental disability” encompasses a range of conditions that affect physical and/or mental ability. When a person has a developmental disability, they can experience challenges with language, learning, self-help, mobility, and independent living. This can be caused by genetic anomalies (e.g., Down syndrome), prenatal exposure to substances (e.g., foetal alcohol syndrome), and preterm birth. In this thesis, “developmental disability” refers to lifelong physical and intellectual impairment present from birth, with an emphasis on genetic syndromes.

Having a child with a developmental disability is often an unanticipated event, and most families have little preparation and have difficulty coping (Seligman & Darling, 1989). Parents thrust into this situation are exposed to emotional and physical stressors in their caregiving role. For example, as the affected child’s development does not follow the typical trajectory, parents worry about management of their child’s health and future (Heaman, 1995). Additionally, parents of children with developmental disabilities report stressors such as financial concerns, high demands on their time, feeling worn out, not being able to go out without their child, and the inability to spend time alone with their spouse (Heaman, 1995). Although each journey is different, parents who have a child with a developmental disability are likely to experience a mix of reactions related to the disability and associated stressors. Common themes in this experience are described below in terms of grief, distress, burnout, and traumatic distress.

1.1.1 Grief

Grief is common in parents who have a child who is acutely unwell or has a developmental disability (e.g., Obeidat, Bond, & Callister, 2009). Having a child with a developmental disability is often unexpected, and the initial shock is confounded by grief and chronic sorrow for the child that the parents expected or wanted (King et al., 2006). This is
ambiguous loss; that is, when a loved one is physically present but psychologically absent (Boss, 2010). The parent may grieve the “absent” child they had hoped for. Because the affected child is alive despite significant challenges, parents may experience pressure to feel “lucky”, and as such their grief about lost dreams for their child may go unrecognised. They also do not experience a clear loss such as death. Thus these parents do not necessarily have an opportunity to resolve their grief, which may become particularly salient when significant milestones are missed because of the developmental disability, as they reflect on the child they imagined they would have. Therefore, parents who have a child with a developmental disability are likely to experience some form of grief, particularly surrounding lost dreams.

1.1.2 Psychological Distress

Along with grief, parents who have a child with a developmental disability are vulnerable to other forms of psychological distress, such as depression. Although research assessments relating to distress are usually based on self-report measures rather than clinical tools (Bailey, Golden, Roberts, & Ford, 2007), there is a large amount of literature demonstrating that parents who have a child with a developmental disability are at increased risk of depressive symptoms. A meta-analysis of research of mothers who have a child with a developmental disability highlighted that they experience elevated depressive symptoms compared to mothers of typically developing children, and almost a third experience depression (Singer, 2006a). This is in keeping with a study of mothers who have a child with cerebral palsy, where 30% reported depressive symptoms (Manuel, Naughton, Balkrishnan, Smith, & Koman, 2003). These results are not surprising considering the grief parents experience and the stressors (including emotional and physical demands) parents face in their caregiving role. Section 1.1.3 outlines the impact of prolonged exposure to the stressors associated with being a parent of a child with a developmental disability (i.e., burnout).
1.1.3 Burnout

According to Selye’s (1976) stress model (also known as “General Adaptation Syndrome”), when presented with a stressor, people respond in a similar pattern. There are three stages: alarm, resistance and exhaustion. The initial reaction to a stressor is alarm, where the body releases stress hormones. This is an adaptive response and can mean, for instance, that parents of a child with a developmental disability are propelled to care for the child and fight for necessary medical support. Next, resistance is when the body’s physical functioning declines with sustained exposure to the stressor. If a stressor continues beyond this, the person’s resources are depleted and they move to the third stage; that is, exhaustion. As the stressors associated with caring for a child with a developmental disability are ongoing (and as such, likely to continue into the exhaustion phase), parents are physically and psychologically vulnerable because their adaptive energy has been sapped. They are susceptible to burnout, originally used to describe the consequences of severe stress and the inability to live up to high ideals experienced by people working in helping professions (Freudenbergner, 1974). Many carers, be they family or support persons, are reported to experience the effects of burnout, overwhelmed by the enormity of the task of caring (McCormack & Adams, 2015; McCormack & Joseph, 2012). Systemic trauma theory explains that in response to feelings of helplessness, burnout can include fatigue, listlessness, loss of empathy and oscillation between avoidance and over-engagement with the stressor (Figley, 1998). Similarly, constructivist self-development theory (McCann & Pearlman, 1992) provides insight into the distress carers experience. It blends object relations, self-psychology, and social cognition theories to explain how people adapt to traumatic events based on how their individual life history has shaped their coping, self-regulation, and self-esteem. Higher rates of clinical burnout are experienced by parents who care for children with chronic illnesses (36%) compared to parents of healthy children (20%; Lindström,
Åman, & Norberg, 2010). Therefore, the stressors related to being a carer for a child with a developmental disability means parents are at risk of exhaustion and thus burnout.

1.1.4 Trauma

Exposure to chronic stressors (e.g., constant worry about the child’s health; multiple or emergency trips to hospital) can also be regarded as traumatic for parents. That is, they are confronted by adverse events which in the early stage can initially shatter their worldview (Joseph & Linley, 2005); and they are chronically exposed to cumulative traumatic events during the course of long-term parenting. These adverse events can be experienced primarily or vicariously. Primary trauma is when someone experiences a threatening event that exceeds his or her ability to psychologically cope. In the context of having a child with a developmental disability, parents may be traumatised by having their world views shattered on learning that their child has been diagnosed with a genetic syndrome with lifelong consequences for them and their child. Vicarious traumatisation refers to schematic change, especially those schemas that are of personal significance (McCann & Pearlman, 1990). As a result, maladaptive schemas can develop as a form of self-protection (Figley, 1995).

Vicarious traumatisation defined professionals working with trauma narratives, but the concept of vicarious traumatisation can similarly apply to this group of parents. When individual caregivers feel overwhelmed, avoidance of further distress and social withdrawal are common forms of self-protection (Figley, 1995).

People affected by vicarious trauma report experiencing disrupted spirituality and/or perceived meaning and hope. For parents who have a child with a developmental disability, they may experience vicarious trauma because (for example) they see their child undergo painful medical interventions or continue to struggle to reach goals. Trauma symptoms in parents who have a child with chronic or critical illnesses such as cancer and accidental injury are well-recognised (e.g., Brocque, Hendrikz, & Kenardy, 2010; McCarthy, Ashley,
Lee, & Anderson, 2012). For instance, in a study of parents with a premature infant in a neonatal intensive care unit, both mothers and fathers experienced clinically significant traumatic distress persisting up to 6 months after the child was born, regardless of the severity of the individual child’s illness (Binder, Zeltzer, Simmons, Mirocha, & Pandya, 2011). Further, in an Australian study of parents who had a child that underwent cardiac surgery before the age of 3 months, evidence of experiencing traumatic distress was present in the majority of parents. That is, 83% of participants endorsed at least one trauma symptom (i.e., dissociation [e.g., feeling numb]; re-experiencing [e.g., dreams of the trauma]; avoidance [e.g., avoiding places that remind the person of the traumatic event]; and arousal [e.g., difficulty sleeping]) at a clinical level, and 27% reported trauma symptoms consistent with a diagnosis of acute stress disorder (Franich-Ray et al., 2013).

There are similarities for parents who have a child with a developmental disability and other clinical groups in terms of experienced events that could elicit primary traumatic distress (self) or vicarious traumatic distress (witnessing child’s distress), inclusive of the risk of re-traumatisation throughout the child’s life (e.g., continuous medical care, missed milestones) and anticipation of trauma. These events include but are not limited to shock at diagnosis, hospital stays, separation from the child, witnessing medical procedures, fear of disability and/or death, reduced quality of life, and impact on daily activities. In addition, developmental disabilities can involve both critical medical crises (e.g., heart surgery) and chronic illness (i.e., ongoing treatment). Therefore, it is likely that parents who have a child with a developmental disability also experience traumatic responses which could occur through chronicity of anticipation of threat, or experience of real threat: primary or vicarious.

1.1.5 Summary of risk

It is clear that stressors associated with having a child with a developmental disability place parents at risk of poor psychological outcomes. Initially, parents can experience shock
and grief at the loss of a child they hoped for or expected. The ongoing caregiving burden that follows can lead to burnout; and parents of a child with a developmental disability are more likely to have depressive symptoms compared to parents of typically developing children (Singer, 2006a). Further, exposure to a wide range of events may trigger traumatic responses particularly if they are cumulative over years. Despite all these risks, there is a growing body of evidence that suggests positive psychological change can occur as a result of struggles with adversity. These changes can include sense-making, positive perceptions, meaning-based coping, and psychological growth. Section 1.2 outlines the potential for positive psychological change in parents who have a child with a developmental disability.

1.2 Potential for Sense-Making

Psychology is the study of behaviour and related mental processes, and as such is neither positive nor negative (Wehmeyer, 2014). However, since World War II, psychology has become largely aligned with the medical model and healing illness. This virtually exclusive focus on psychopathology neglects the positive features that make life worth living (e.g., hope, spirituality, courage, perseverance); or fulfilled individuals who thrive in the face of adversity (Seligman & Csikszentmihalyi, 2014). In the 1960s, the Humanistic perspective called for a holistic approach to human existence, acknowledging the development of potential, motivation, life goals, and meaning (Wehmeyer, 2014). Despite this, there is little knowledge of the more positive aspects of parenting a child with a developmental disability. There is much to be learned from documenting flourishing parents (Seligman & Csikszentmihalyi, 2014). Aside from the fact the parental wellbeing is important for the parent themselves (and the family unit; e.g., Graungenard, Andersen, & Skov, 2011a); the literature suggests that a strength-based approach to disability is vital for caregivers’ positive outcomes (Lawton, Moss, Kleban, Glicksman, & Rovine, 1991). For example, when parents
who had a child with autism received a strength-based assessment from therapists, they showed improved affect, made more positive statements about their child, and also exhibited more physical affection toward their child compared to parents who received information about their child’s deficits (Steiner, 2011). Therefore, it is important to understand more about the positive side of parenting a child with a developmental disability.

People adapt to challenging situations in different ways. When existing schemas are used to manage a new situation assimilation occurs, whereas when individuals engage schematic processes to adapt to new information, accommodation occurs (Piaget, 1952). This has consequences in the context of trauma and distress for interpreting threat events, particularly for parents of children with disabilities. How this group of parents come to describe, explain, and interpret their experiences can vastly affect their reactions to their child, medical staff, role as parents, and others. From a social constructionist perspective, experiences and interactions with others shape realities. If parents are supported to accommodate their experiences positively, the life-changing experience of having a child with a developmental disability can provide a platform for positive change; whereas assimilation implies remaining at the previous level of functioning (i.e., without finding new meaning). Relevant theories and research which may provide insight into positive accommodation are outlined below.

1.2.1 Meaning-based coping

When faced with negative psychological states associated with significant stress, people may be consciously or unconsciously motivated to create positive psychological states in order to gain relief (Folkman, 1997). This is referred to as meaning-based coping. As parents of a child with a developmental disability are confronted by multiple stressors (e.g., as outlined in section 1.1.4), it is likely that some of them cope in this way. Indeed, research has demonstrated that parents can develop positive perceptions related to their affected child,
including tolerance and acceptance (Hastings & Taunt, 2002). They can also experience personal growth and improved relationships (Hastings & Taunt, 2002). Mothers have been found to cope by positively reframing their experiences; suggesting that their child is a source of happiness, strength, and growth (Hastings, Allen, McDermott, & Still, 2002). However, there is little research about when and how the parents’ experiences are positively accommodated into their worldview.

Furthermore, similar to meaning-based coping, Mishel’s (1990) Reconceptualised Uncertainty in Illness theory proposes that chronically ill people and their families must integrate the continuous uncertainty of illness into their lives by accommodating it into their worldview. This helps them to avoid a prolonged anticipatory state of distress, which occurs as a result of the unfulfilled expectation of predictability. Accommodating the illness can facilitate psychological shifts and adaptability for integrating uncertainty more positively into the individual’s belief system. Evidence for positive integration has been shown in parents who have a child with Asperger syndrome (Samios, Pakenham, & Sofronoff, 2009). Parents report changes such as spiritual growth (e.g., finding or growing in faith), greater understanding (e.g., empathy), and new possibilities (e.g., advocacy; Samios et al., 2009). Although not conceptualised in this way by the authors, these findings suggest that parents have reorganised their beliefs in order to gain relief from negative psychological states. For example, anxiety (i.e., the negative state) was associated with greater positive change (i.e., reorganisation of beliefs; Samios et al., 2009). Further research based in theory is required to examine the processes that lead to the use of this type of coping in parents who have a child with a developmental disability; and if certain negative psychological states are necessary and sufficient for meaning-based coping or psychological growth.
1.2.2 Posttraumatic growth

As mentioned previously (Section 1.1.4), parents who have a child with a developmental disability are at risk of experiencing traumatic distress. However, trauma can be a catalyst for positive psychological change or growth out of adversity; that is, posttraumatic growth (Joseph & Linley, 2005; Tedeschi & Calhoun, 1996). For this growth to occur, the trauma-related information must be integrated into the person’s worldview (Joseph & Linley, 2005). Although relatively unexplored in parents who have a child with a developmental disability, posttraumatic growth has been found in a similar population, namely, parents who have had a child in an intensive care unit. Eighty-eight percent of these parents reported a great degree of positive change as a result of the experience (Colville & Cream, 2009). Moderate levels of posttraumatic stress (compared to low or high levels) were more strongly correlated with posttraumatic growth (Colville & Cream, 2009). Therefore, there may be an optimum level of stress that encourages positive psychological change. If the stressor is not significant enough to shatter a person’s belief system, they may assimilate the information (i.e., return to their previous level of functioning), rather than accommodate it (i.e., change a schema to process the new information). If the event is too traumatic, a person may accommodate the information negatively (e.g., experience helplessness) and be unable to psychologically grow. Supporting this, Helgeson, Reynolds, and Tomich (2006) conducted a meta-analysis of benefit finding and growth. They found that benefit finding was related to less depression and more positive well-being, but also more intrusive and avoidant thoughts about the stressor. Again this indicates that a certain level of distress may be required to experience positive change, demonstrating the potential for meaning-based coping as a tool to gain some relief from negative psychological states (Folkman, 1997). Further, Helgeson et al. (2006) suggest that the time which has passed since the stressor was first experienced also affects benefit finding. However, this review involved a variety of traumatic situations; the
majority of which were limited to finite situations, such as experiences with natural disasters. Only three studies of the 87 studies included examined parents who have a child with a developmental disability (i.e., exposure to stressor/s with no foreseeable end point), and they all looked at stress-related growth only. Thus, although positive psychological change is possible after traumatic and stressful events, this phenomenon has been under-researched in parents who have a child with a developmental disability.

1.2.3 Summary of potential for sense-making

In conclusion, families of children with developmental disabilities have the opportunity to thrive as a result of their experiences and construct meaningful stories surrounding the journey with their child. For the purpose of this thesis, these positive changes are broadly termed “positive psychological outcomes”. Such positive psychological outcomes may include resilience (effective management of difficult situations and learning from experiences to become strengthened and more resourceful; Rolland & Walsh, 2006) and growth (positive change in psychological functioning after trauma or adversity, such as changing life values; Joseph, 2012). Although there is research demonstrating that positive psychological outcomes can and do exist, it is unclear which factors promote these positive psychological outcomes (Carroll, 2013).

1.3 Positive Psychological Outcomes: A Narrative Review

1.3.1 Background

Parental wellbeing is essential, not only for the parents themselves but for the other members of the family, including the child with the disability and their siblings (Graungaard et al., 2011a; Ormond & Seltzer, 2009). To examine how positive psychological outcomes can be fostered in this population, a narrative review was conducted. The aim was to identify predictors of positive psychological outcomes for parents of children with developmental
disabilities. That is, positive psychological constructs as dependent variables only. We examined quantitative studies pertaining to the analysis or promotion of positive psychological outcomes (e.g., resilience, posttraumatic growth), with or without comparison groups.

1.3.2 Methods

A database search was conducted. For the purpose of this review, it was required that studies quantitatively examined any factors (including but not limited to social support, marital satisfaction, and parental or child characteristics) contributing to positive psychological outcomes as a dependent variable (e.g., resilience, hardiness, and posttraumatic growth) in biological parents of children affected by any developmental disability (e.g., Down syndrome). An electronic search of the databases CINAHL, EMBASE, Medline, and PsycINFO was conducted in January 2015. Searches were carried out using terms related to the concepts of psychological wellbeing AND developmental disabilities AND parents, see table 1. All terms were mapped to exploded\(^1\) subject headings where applicable. See Appendix A for an example database search strategy.

\(^1\) When a search term is “exploded”, citations associated with that MeSH heading (or subheading) are retrieved along with more specific related terms/headings/subheadings (NIH US National Library of Medicine, 2015). That is, it expands the search.
### Table 1. Review search terms, mapped to exploded subject headings where applicable.

<table>
<thead>
<tr>
<th>Concepts</th>
<th>Search Terms</th>
</tr>
</thead>
<tbody>
<tr>
<td>Psychological wellbeing</td>
<td>resilience – psychological, resilience, psychological adaptation, benefit-finding, well-being, wellbeing, hardiness, coping, positive outcomes, adaptive behaviour, posttraumatic growth</td>
</tr>
<tr>
<td>Developmental disabilities</td>
<td>developmental delay, learning disability, mental retardation, intellectual development disorder, intellectual impairment, intellectual disability, deletion [chromosome], undiagnosed condition, rare disorder, rare disease, Down syndrome, Williams syndrome, Prader-Willi syndrome, Fragile X syndrome, Tuberous Sclerosis, 22q11 Deletion syndrome/DiGeorge syndrome, Autism</td>
</tr>
<tr>
<td>Parents</td>
<td>family relations, family conflict, parent child relations, intergenerational relations, mother child relations, father child relations, parenting, maternal behaviour, paternal behaviour, patient-family relations, human relation</td>
</tr>
</tbody>
</table>

Databases were searched from inception to January 2015. The systematic search was updated in August 2016 and no additional references were found. Studies were only included if they were written in English and contained original data (e.g., grey literature\(^2\), opinion pieces, and review papers were not included). As this was an examination of parental positive psychological outcomes, papers were excluded if they did not report positive psychological constructs (e.g., only depression) or did not relate to the parents (e.g., child or siblings only), or included positive psychological constructs as independent variables only (rather than outcome variables). Studies of children in the prenatal period, over the age of 18, or those adopted were not included in the current review. Research that involved parents affected by a developmental disorder themselves was excluded to avoid the impact of confounding variables on positive outcomes. Quality of life was not included as it is a well-established

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\(^2\) Grey literature refers to research that is a) unpublished or b) published in non-commercial form. For example, government reports, newsletters, and conference proceedings.
area that has already been the topic of many systematic reviews, and mood was excluded as it was considered too transient to be considered a positive psychological outcome.

The candidate and another researcher independently extracted data from each study into a form based on systematic review literature (Higgins & Green, 2011; Popay et al., 2006; Vandenbroucke et al., 2007). The first supervisor assisted where any disparities were identified.

1.3.3 Results

A total of 2,173 references were identified through the initial search (see Appendix B), 189 of which were duplicates. Through a review of the title and abstracts of the remaining 1,984 references, 1,896 were excluded because they did not relate to positive psychological constructs, parents, a child with developmental disorder (e.g., parenting whilst affected by a disability), or children living at home. The remaining 88 manuscripts were reviewed, with 74 excluded (51 = not examining positive constructs as dependent variables, 11 = quality of life, 6 = qualitative, 4 = “child” over 18 years, 1 = not parental outcomes, 1 = not original data). A total of 14 articles were included in the review, the results of which are presented in Table 2.
Table 2. Summary of Included Articles

<table>
<thead>
<tr>
<th>Study, location, design, and setting</th>
<th>Positive psychological outcomes</th>
<th>Findings</th>
</tr>
</thead>
<tbody>
<tr>
<td>(Ekas, Whitman, &amp; Shivers, 2009), USA Design: Cross-sectional survey Setting: Mail out questionnaire Child’s condition type: ASD</td>
<td>Outcome: 1. Child-related enjoyment 2. Life satisfaction 3. Psychological wellbeing 4. Optimism 5. Sense of control Predictors: Child age, number of additional children with Autism, religiosity (beliefs, activities, spirituality).</td>
<td>- Older child with ASD was associated with lower levels of child-related enjoyment, life satisfaction, well-being, sense of control, as well as higher levels of pessimism. - After controlling for religiosity and spirituality, child’s age was a significant negative predictor of child-related enjoyment. More than one child with ASD reduced sense of control. - Greater religious belief predicted increased maternal optimism. - Greater religious spirituality predicted increased maternal self-esteem, life satisfaction, positive affect, wellbeing and control of internal states. - More participation in religious activity was a significant predictor of lower maternal self-esteem, life satisfaction, wellbeing, positive affect, and control of internal states.</td>
</tr>
<tr>
<td>(Ekas, Lickenbrock, &amp; Whitman, 2010), USA Design: Cross-sectional survey Setting: Mail out questionnaire Child’s condition type: ASD</td>
<td>Outcome: 1. Life satisfaction 2. Psychological wellbeing Predictors: Optimism, friend support, partner support, family support</td>
<td>- Partner support was associated with increased life satisfaction and psychological wellbeing. - Optimism was associated with increased life satisfaction and psychological wellbeing.</td>
</tr>
<tr>
<td>(Fatima &amp; Suhail, 2010), PAK Design: Cross-sectional interview Setting: Interviews Child’s condition type: Down syndrome</td>
<td>Outcome: Life Satisfaction Predictors: sociodemographic factors, belief in just world, interaction between just world beliefs and group</td>
<td>- The more the mothers believed in a personal just world and in immanent justice, the more they were satisfied with their life. - Mothers of typically developing children were more satisfied with their life than mothers who had a child with Down syndrome.</td>
</tr>
<tr>
<td>(Frey, Fewell, &amp; Vadasy, 1989), USA Design: Repeated measures questionnaires &amp; interview Setting: Parents jointly interviewed at their home Child’s condition type: Down syndrome, Cerebral Palsy, other unspecified conditions</td>
<td>Outcome: Family Adjustment Predictors: Social network, religion, problem-solving ability, child’s communication competence and sex</td>
<td>- Greater social support, problem-solving ability, and child’s communication ability was linked to higher family adjustment for fathers. There was no change in this across the 2 years. - For mothers, having a female child and increased communication ability in their children was associated with greater family adjustment. There was no change in this across the 2 years.</td>
</tr>
<tr>
<td>(Friedrich, Cohen, &amp; Wittturner, 1987), USA Design: Cross-sectional survey Setting: Appears to be a mail out questionnaire, unclear. Child’s condition type: IQ ≤ 64, otherwise unspecified.</td>
<td>Outcome: Family relations Predictors: Coping resources, general/specific beliefs, child variables</td>
<td>- Positive family relations were predicted by greater marital satisfaction, greater internal locus of control, and less maternal depression.</td>
</tr>
<tr>
<td>(Greer, Grey, &amp; McClean, 2006), IRL Design: Cross-sectional survey Setting: Mail out questionnaire/meeting with researchers Child’s condition type: mild, moderate, severe or profound range of intellectual disability.</td>
<td>Outcome: Positive perceptions (happiness and fulfilment; personal growth and maturity; strength and family closeness) Predictors: Behavioural and cognitive coping strategies, mother and partner career status.</td>
<td>- Levels of happiness and fulfilment; and levels of personal growth were not predicted by coping strategies or parents’ career statuses. - Strength and family closeness was predicted by the coping strategy of mobilising the family to acquire and accept help.</td>
</tr>
<tr>
<td>(Hastings et al., 2005), UK Design: Cross-sectional survey. Setting: Telephone interview, mail out questionnaires, home visit Child’s condition type: ASD</td>
<td>Outcome: Positive perceptions of child Predictors: Child variables (behaviour problems, Autism symptoms, adaptive behaviour), paternal depression and anxiety, maternal depression and anxiety.</td>
<td>- Mothers’ positive perception scores were not predicted by child variables nor paternal anxiety and depression. - Fathers’ positive perceptions were predicted by maternal depression.</td>
</tr>
<tr>
<td>Reference</td>
<td>Country/Region</td>
<td>Design</td>
</tr>
<tr>
<td>-----------</td>
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</tr>
<tr>
<td>(Higgins, Bailey, &amp; Pearce, 2005), AUS</td>
<td>AUS</td>
<td>Cross-sectional survey</td>
</tr>
<tr>
<td>(Mak &amp; Ho, 2007), HK</td>
<td>HK</td>
<td>Cross-sectional survey</td>
</tr>
<tr>
<td>(Olsson &amp; Hwang, 2008), SWE</td>
<td>SWE</td>
<td>Cross-sectional survey</td>
</tr>
<tr>
<td>(Samios et al., 2009), AUS</td>
<td>AUS</td>
<td>Cross-sectional and longitudinal survey</td>
</tr>
<tr>
<td>(Samios, Pakenham, &amp; Sofronoff, 2012), AUS</td>
<td>AUS</td>
<td>Cross-sectional survey</td>
</tr>
<tr>
<td>(Trute, Bennetos, Worthington, Reddon, &amp; Moore, 2010), CAN</td>
<td>CAN</td>
<td>Cross-sectional telephone survey</td>
</tr>
<tr>
<td>(Xue, Ooh, &amp; Magiati, 2014), SGP</td>
<td>SGP</td>
<td>Cross-sectional survey</td>
</tr>
</tbody>
</table>

Note. Abbreviations/acronyms are as follows: Intellectual disability (ID), Autism Spectrum Disorders including Autism, Autism Spectrum Disorder, Asperger’s syndrome or pervasive developmental disorder not otherwise specified (ASD), United States of America (USA), Australia (AUS), Sweden (SWE), Ireland (IRL), Pakistan (PAK), United Kingdom (UK), Hong Kong (HK), Canada (CAN), Singapore (SGP).
**Study characteristics**

As seen in Table 2, the vast majority of studies were cross-sectional surveys, predominately mail out. They were conducted in the USA, Australia, Canada, Hong Kong, Ireland, Pakistan, Singapore, Sweden, and the United Kingdom. Autism Spectrum Disorders (ASD) or pervasive developmental disorder not otherwise specified were the most common diagnostic groupings. Four studies included a variety of developmental disorders within the one article (i.e. Frey et al., 1989; Mak & Ho, 2007; Olsson & Hwang, 2008; Trute et al., 2010), such as a combination of Down syndrome, cerebral palsy, and other unspecified conditions. Two studies (i.e. Friedrich et al., 1987; Greer et al., 2006) utilised only parents of children with intellectual disabilities, but it was unclear if there were also associated genetic or developmental conditions. Fatima and Suhail (2010) included only parents of children with Down syndrome. The studies by Ekas et al. (2009; 2010) reported data from the same sample. This was also the case for the two papers by Samios et al. (2009; 2012).

**Reported positive outcomes**

The reviewed articles included a large variety of positive psychological outcomes. For clarity of reading, we have summarised our findings under the broad headings of adjustment, wellbeing, positive perceptions, and satisfaction. However, there was much variation in how these constructs were operationalised. This is further explored in the discussion.

**Adjustment**

Generally, psychological adjustment is conceptualised as positive mental health or the ability to cope effectively with environmental demands (Seaton, 2009). Several studies in this narrative review (i.e., Frey et al., 1989; Higgins et al., 2005; Samios et al., 2009, 2012; Trute et al., 2010; Xue et al., 2014) identified forms of adjustment as a
positive psychological outcome. Adjustment encompassed concepts such as family cohesion, expressiveness, amount of conflict (Frey et al., 1989), and family functioning (Trute et al., 2010; Xue et al., 2014). Similarly, Higgins et al.’s (2005) definition of adjustment included family adaptability, cohesion, and self-esteem. Both articles by Samios et al. (Samios et al., 2009, 2012) defined adjustment as satisfaction with life, positive affect, and health status; however, the 2012 study also included perceptions of relationships with their partner as an indication of adjustment.

Higgins et al.’s (2005) study found that coping strategies (i.e., self-esteem, optimism, and spousal support) did not predict adjustment. Yet positive appraisal and affective positivity (Trute et al., 2010), sense making – reframing (Samios et al., 2009), and social support (Frey et al., 1989) were related to better adjustment. In addition, greater family capabilities including coping, support and fewer demands (e.g., stress; Xue et al., 2014) was associated with adjustment. Each of these variables can be viewed as coping methods, in so far as they concern employing cognitive, affective, and social supports to help manage challenges.

Child factors also appear to have an effect on adjustment. Both mothers and fathers reported better family adjustment when the affected child had greater communication skills (Frey et al., 1989). Mothers also appeared to adjust more readily when their child with a disability was female (Frey et al., 1989). Fathers’ adjustment was also related to improved problem solving ability (Frey et al., 1989). However, other child and parent variables (i.e., child age, number of children with ASD, parent gender, marital status, and spiritual beliefs) did not predict adjustment (Samios et al., 2009).
Positive perceptions

Positive perceptions include child-related enjoyment (Ekas et al., 2009); strength and family closeness (Greer et al., 2006; Mak & Ho, 2007); and positive perceptions of the child with the developmental disability (Hastings et al., 2005). Similar to Adjustment, the results of the impact of coping mechanisms on positive perceptions are inconclusive although this is most likely linked to differing study methodologies. For instance, whilst coping strategies (along with career status) did not predict the positive perception outcomes of happiness and fulfilment, and personal growth (Greer et al., 2006), mobilising the family to acquire and accept help as a coping style did increase strength and family closeness (Greer et al., 2006). Mak and Ho (2007) also found that emotion-focused coping (along with caregiving stress and social support) did not predict mothers’ positive perceptions. Yet problem-focused coping and relationship-focused coping (along with higher family income) predicted greater positive perceptions. Both these studies used the same measure (Kansas Inventory of Parental Perceptions) to assess the outcome; however, the data was collected from culturally diverse samples in Ireland (Greer et al., 2006) and Hong Kong (Mak & Ho, 2007) which may explain the discrepancy in results.

In terms of child factors, the diagnosis of the child appears to be related to positive perceptions. For instance, Mak and Ho (2007) found that mothers of a child with ASD had fewer positive perceptions compared to mothers of children affected by intellectual disability. Similarly, having an older child with ASD was associated with lower levels of child-related enjoyment (Ekas et al., 2009). However, in another study, children’s behaviour problems, ASD symptoms, and adaptive behaviour were not predictive of mothers’ positive perceptions of their child (Hastings et al., 2005). A

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3 Positive perceptions related to having a child with a developmental disability. That is, the child is seen as the source of the outcome.
possible explanation for this discrepancy is age differences, in that the study by
Hastings et al. (2005) included parents with younger children (~3 years), whereas the
affected children in Ekas et al.’s (2009) were older (~9 years). The difficulties of
parents who have older children may not have been present yet in the younger sample.
Finally, the only predictor of fathers’ positive perceptions of their child was mothers’
depression (Hastings et al., 2005). That is, when mothers were less depressed, fathers
were more likely to report positive perceptions of their child.

Psychological wellbeing

Psychological wellbeing was identified as another positive outcome explored
among parents of a child with a developmental disability. The three papers reporting on
this outcome, described psychological wellbeing as: how the participant had been
feeling in the past 2 weeks (Olsson & Hwang, 2008); wellbeing in terms of personal
growth, environmental mastery, and purpose in life (Ekas et al., 2010; Ekas et al.,
2009); and perceived sense of control over life events (Ekas et al., 2009), which is
similar to how ‘adaption’ has been conceptualised in the literature. Better self-reported
wellbeing was associated with greater religious spirituality (Ekas et al., 2009) and
partner support (Ekas et al., 2010). Olsson and Hwang (2008) identified a number of
predictors of positive psychological wellbeing. For mothers, these included higher sense
of coherence, perceived positive impact of child, satisfaction with participation in life,
and self-rated health (Olsson & Hwang, 2008). For fathers, the only predictor was better
self-rated health (Olsson & Hwang, 2008). Religious spirituality was related to parents
having a greater sense of coherence (Ekas et al., 2009); and having better social
(primarily friend-based) support was associated with positive affect (Ekas et al., 2010).
Poorer psychological wellbeing and poorer sense of control was predicted by having an
older child with an ASD, and more participation in religious activity (Ekas et al., 2009). Greater economic hardship also resulted in poorer wellbeing (Olsson & Hwang, 2008).

**Life satisfaction**

Three papers reported on the final positive outcome of life satisfaction. Life satisfaction included reflective judgements about one’s own life (Ekas et al., 2010; Ekas et al., 2009; Fatima & Suhail, 2010), as well as optimism (Ekas et al., 2009). Greater life satisfaction was associated with better partner support and optimism (Ekas et al., 2010), stronger religious beliefs and religious spirituality (Ekas et al., 2009), and personal belief in a just world and immanent justice (Fatima & Suhail, 2010). However, more participation in religious activities reduced life satisfaction in the context of parenting a child with a disability (Ekas et al., 2009). In addition, optimism mediated the relationship between family support and life satisfaction (Ekas et al., 2010). In terms of child factors, poorer life satisfaction was associated with having an older child with ASD (Ekas et al., 2009); and having a child with Down syndrome, as opposed to a typically developing child (Fatima & Suhail, 2010).

**1.3.4 Discussion**

The aim of this narrative review was to identify factors involved in promoting positive outcomes for parents of children with developmental disabilities. It is evident that many families with children who have disabilities do better than expected and can thrive and grow because of their experiences. However, investigation of positive psychological outcomes in this population is a relatively new field of research. This poses difficulties for a narrative review, as the body of work is not yet large enough in parents of children with developmental disabilities to focus on a particular positive outcome (e.g., positive reinterpretation). Nonetheless, at this early stage, it is important
to examine the state of this field of research to identity problems as well as areas for exploration. Areas for improvement and opportunities for investigation are outlined below.

**Limitations of existing research**

*Sampling issues*

The narrative review highlighted a number of limitations within this field of research. Firstly, the majority of the research was conducted on families affected by ASD, as well as unnamed conditions or intellectual disabilities. However, parents may have different reactions depending on the neurocognitive and behavioural phenotype associated with a specific form of developmental disability. For example, in a study of parents with a child with Fragile X, ASD, or Down syndrome, the most consistent predictor of maternal outcomes was the child's behavioural symptoms (Abbeduto et al., 2004). It is thus worthwhile to explore disability type as related to positive psychological outcomes. Researchers must also be as specific as possible when reporting the diagnoses of their sample in order to provide context for interpretation of the results. Another issue in terms of sampling is that the vast majority has been conducted on convenience samples of mothers. It is important to widen the research effort and include the experiences by other family members such as fathers and siblings, as well as the person with the developmental disability. The interactions between these different family members could also provide insight on how the family system impacts psychological wellbeing. Although these studies are a strong foundation, there is a need to extend to participant pool in order to gain a holistic understanding of parenting a child with a developmental disability.
Research methods

Even though parental wellbeing is vital, not only for the parents themselves but for the others members of the family including the child with the disability and their siblings (Graungaard et al., 2011a; Orsmond & Seltzer, 2009), it is being neglected in research and intervention. The majority of studies examining positive psychological outcomes were cross-sectional surveys, which do not allow for thorough exploration of phenomena such as adaptation that may develop and change over time (e.g., Lazarus & Folkman, 1984). This is not necessarily a criticism of the researchers in the field: in fact, the use of primarily cross-sectional surveys demonstrates a good understanding of the caregiving burden this population experiences. It is also difficult for fulltime carers to fit research into their schedules.

Creative thinking is required to access and aid a representative population of parents who have a child with a developmental disability. The research (and potential interventions for promoting positive psychological outcomes) needs to be available in a format that is appropriate for these parents. For example, an online study may allow for more flexibility compared to a face-to-face research. As the child’s physical health is already a priority, future trials to promote positive psychological outcomes may need to be built into the child’s medical care. A feasibility study would prove useful in delineating perceived barriers and opportunities for such interventions. Therefore, there is a need to broaden the methods used for researching positive psychological outcomes in parents who have a child with a developmental disability. This would provide an evidence base for interventions to support parents that could be built into standard care.

Positive psychological concepts are not well-defined

Although we subsumed the results from the articles in this narrative review under headings (or “themes”) related to adjustment, wellbeing, positive perceptions, and
satisfaction; it was extremely challenging to find commonalities between the articles, despite the outwardly similar subject areas. This was partly due to overlapping or poorly operationalised definitions of many constructs. Therefore, it is difficult to draw firm conclusions regarding the overarching findings of these studies. For example, there were discrepancies regarding coping as a predictor of the various outcomes. Certain coping styles promoted positive outcomes (e.g., mobilising the family to acquire and accept help impacted strength and family closeness; Greer et al., 2006), whereas others seemed to have no such association (e.g., self-esteem, optimism, and spousal support did not affect adjustment; Higgins et al., 2005). The lack of a cohesive statement regarding coping as a resource to support positive psychological outcomes in these parents reflects the varied ways that ‘coping’ has been conceptualised in the literature. We suggest that researchers endeavour to base their definitions of key concepts like coping and psychological wellbeing on strong theoretical underpinnings and psychometrically valid scales rather than subjective definitions to enable more specific predictions and to ease interpretation to facilitate the design of interventions that can be used in clinical practice.

Related to this, it is important to note that seemingly important papers in the field have been excluded from this narrative review. This is because positive psychological constructs were used as predictor variables, rather than outcome variables (e.g., Lloyd & Hastings, [2009] investigated hope as a resilience factor; however their outcome variables were anxiety and depression). Or, more critically, many articles appeared to examine positive psychological outcomes; but in actual fact were only investigating the absence of negative outcomes (e.g., lack of depression). This is an important distinction, because there is evidence that these are independent concepts (Hastings & Taunt, 2002). Numerous studies were included until the finals stages of the
narrative review before being culled as a result of robust discussion between the research team, where the discrepancy between the absence of psychopathology and actual positive psychological outcomes was highlighted. It was decided that studies reporting the absence of psychopathology rather than actual positive psychological outcomes should not be included. One such example is Siman-Tov and Kaniel’s (2011) paper, which examined adjustment in mothers and fathers of children with autism. Adjustment was one of the broad themes that was included in our narrative review, and thus it appears as though their paper should have been included. Siman-Tov and Kaniel’s (2011) definition of adjustment included mental health; however, this was defined as an absence of major mental health conditions (e.g., depression and anxiety). The measurement tool they used for this variable (i.e., the Mental Health Scale) assessed psychological distress and physical health only. Thus, although this article may initially seem to examine positive psychological constructs, a thorough reading and critical reflection demonstrates that it is not the case. Hence, it is necessary for researchers to clarify whether they are examining lack of psychopathology or true positive psychological constructs, recognising that negative and positive psychological outcomes may not be on a continuum.

1.3.5 Narrative review conclusions

This narrative review has provided a snapshot of the state of the literature relating to positive psychological outcomes. It demonstrated that many parents (and families) of children with developmental disabilities are resilient and show positive growth subsequent to the birth or diagnosis of their child. In order to maximise outcomes for these families there is a need for clear definitions and robust studies examining the factors underlying positive psychological outcomes, including the impact of risk factors, culture, health care setting, and type of diagnosis. Parental psychological
health is an important factor in the child’s health and family functioning (Graungaard et al., 2011a) and thus policy must be built to promote it as standard care.

1.4 Rationale for this thesis

There is little research providing support to help parents adapt positively, above and beyond the absence of psychopathology. Yet this narrative review demonstrates that there is evidence that these outcomes are achievable, perhaps even realistic. Therefore, the aim of this thesis is to give an overview of the general experience of having a child with a developmental disability, with a particular emphasis on positive psychological constructs. Although psychology as a discipline has traditionally focused on psychopathology, there is much to be learned from people who adapt well and create meaning from situations they are thrust into and have little control over. This does not diminish the fact that these parents face many challenges and can experience a long and difficult journey; however, positive psychological processes do not necessarily need to be examined in the context of poor functioning. In fact, there is evidence that they can be quite independent (Hastings & Taunt, 2002; Helgeson et al., 2006).

1.4.1 22q11.2 deletion syndrome (22q11DS)

Although the information above relates to developmental disability in general, one developmental disability that offers unique challenges and is poorly researched is 22q11.2 deletion syndrome (22q11DS). Therefore, following on from the nomothetic study exploring potential predictors of psychological growth in parents who have a child with a developmental disability, two further interpretative phenomenological studies will explore the “lived” experience of parents who have a child with 22q11DS. Examining positive psychological outcomes as related to particular developmental disabilities can be particularly insightful, as the child’s symptoms and/or behaviour may
The impact on parental wellbeing (Abbeduto et al., 2004). The developmental disability 22q11DS has a unique advantage from a research perspective. That is, it is the most common microdeletion syndrome, occurring in approximately 1 in 4000 live births (Oskarsdottir, Vujic, & Fasth, 2004); with a recent study of high-risk pregnancies indicating a prevalence of 1 in 992 live births (Grati et al., 2015). However, there is low professional awareness of 22q11DS which means it is clinically under-recognised (Bassett et al., 2011), and many children experience a delay in diagnosis despite experiencing chronic symptoms (Goodwin et al., 2015). Therefore, examining the impact of 22q11DS on parents has distinct advantages: the results are likely to be generalisable to a large population (i.e., the many families affected by 22q11DS); yet may also provide insight into the journey families experience when they have a child who is affected by a rare developmental disability (e.g., as related to diagnostic issues & lack of awareness in professionals managing the child’s healthcare).

22q11DS occurs when there is a deletion from chromosome 22 at band q11.2 (Robin & Shprintzen, 2005). The condition is also known as velo-cardio-facial syndrome or DiGeorge sequence because it was described independently in different parts of the world before genetic testing technology revealed a single underlying syndrome (Shprintzen, 2008). More than 90% of 22q11DS cases are de novo (i.e., a spontaneous event from unaffected parents; McDonald-McGinn & Zackai, 2008). 22q11DS is associated with over 180 clinical features and has variable expressivity (McDonald-McGinn & Zackai, 2008). Most commonly, the syndrome is associated with developmental delay, characteristic facial features, congenital heart defects (e.g., tetralogy of Fallot), and palatal anomalies (McDonald-McGinn et al., 1999). Other features can include increased risk of infection (Jawad, McDonald-McGinn, Zackai, & Sullivan, 2001), neonatal hypocalcaemia (Kitsiou-Tzeli, Kolialexi, & Mavrou, 2005)
and recurrent otitis media (Dyce et al., 2002). The behavioural phenotype is characterised by executive dysfunction (Bish, Ferrante, McDonald-McGinn, Zackai, & Simon, 2005), attention deficits (Niklasson, Rasmussen, Oskarsdóttir, & Gillberg, 2005), social impairments (Shashi et al., 2012), and autism spectrum disorder features (Fine et al., 2005). People with 22q11DS are also at an increased risk of developing mood (Green et al., 2009) and psychotic disorders compared to the general population (Murphy, Jones, & Owen, 1999).

Developmental disability affects 1 in 50 children, with 1 in 200 having severe intellectual disability with an IQ< 50. Although a common presentation, the majority of developmental disability is due to a large number (~4000) of rare genetic conditions. 22q11DS provides an exceptional research opportunity and as such will be the focus of the qualitative aspect of this thesis. Although children with 22q11DS have a characteristic neurocognitive and behavioural phenotype, the disabilities experienced by these children and the challenges faced by their parents can be extrapolated to the wide range of individually rare genetic forms of developmental disability.

1.4.2 What this thesis adds

This thesis will provide a unique contribution to the literature by broadening the current knowledge of the experience of parenting a child with a developmental disability, especially the more positive aspects. It will seek to overcome the previously outlined limitations by: a) extending investigation to developmental disabilities outside of ASD (through exploration of parenting experiences related to developmental disabilities in general, as well as detailed examination of a specific syndrome [i.e., 22q11DS]); b) examining predictors of a specific positive psychological outcome (i.e., psychological growth); c) providing evidence of differential reactions across the child’s age and stage; and d) merging findings from quantitative and qualitative data.
Chapter 2
Mixed Methods: A Rationale

2.0 Chapter statement

This chapter presents an overview and explanation for the use of mixed methods in this thesis. It begins by providing a rationale for mixed methods research in general, and explaining why combining these seemingly opposite approaches can be fruitful. The pragmatic choice of including both quantitative and qualitative research is also delineated in the context of unexplored phenomena. The chapter explores the advantages of mixed methods research, emphasising the holistic nature of this approach and how mixed methods are utilised. It concludes with an outline of the research design and the broad aims of this thesis.

2.1 Rationale for the use of mixed methods

In the past twenty years, there has been a sharp increase in combining qualitative and quantitative methods to explore life-changing phenomena, particularly in health research (O'Cathain, Murphy, & Nicholl, 2007). In a purist form, qualitative research is viewed as the antithesis of quantitative work (Pope & Mays, 1995). That is, there are perceived differences between the two types of research in terms of ontology, epistemology, and methodology. However, this is arguably an unnecessary divide, as a combination of qualitative and quantitative research can draw upon the strengths and reduce the limitations of each method individually (Johnson & Onwuegbuzie, 2004). The preference for positivist methods in psychology has meant that research is often conducted in isolation from real-life events; and thus does not necessarily reflect individuals’ health and illness experiences (Crossley, 2000) nor interpretation of those experiences. Qualitative methods can bridge this gap by providing a rich, insider’s
perspective to a particular phenomenon; and thus can complement rather than oppose quantitative work. Mixed methods research as an expansive approach—that is, extending the breadth and range of inquiry—can bring meaning (qualitative) to numbers (quantitative). It is important to note that neither qualitative nor quantitative research is superior. Rather, the appropriate methods for the research question at hand (i.e., qualitative, quantitative, or mixed) should be utilised in order to shed light on the complexities of a particular phenomenon.

Pragmatism is the philosophy that underlies the majority of mixed methods research (Johnson, Onwuegbuzie, & Turner, 2007). It encompasses positivism and interpretivism, and highlights the complex nature of the social world along with the need for a holistic understanding of phenomena4. In practical terms, as applied in this thesis, pragmatism is utilising the methods most suitable for the research question. Improving the lives of families affected by 22q11DS requires research answering a range of complex questions relating to their experiences. Both qualitative and quantitative methods are necessary to answer these wide ranging questions and enable a more complete understanding of the experience of parents who have a child with a developmental disability, such as 22q11DS.

2.2 Types of mixed methods research

There are a variety of ways to conduct mixed methods research. Qualitative and quantitative data can be collected simultaneously (i.e., concurrent) or sequentially (Grbich, 2013). Usually, concurrent studies involve triangulation or embedded designs. Triangulation is the use of different methods to examine the same topic, with the intention of bringing the strengths of each method together (Creswell & Plano-Clark, 4 The philosophical and theoretical underpinnings related to the qualitative methodology are discussed in detail in chapter 5.
Quantitative and qualitative data complement each other, and are drawn together for comparison. In contrast, an embedded design seeks answers to separate questions, such as a qualitative follow-up of quantitative survey results (Creswell & Plano-Clark, 2007). Another difference is that an embedded design is based within one data set, with the other only providing secondary support. The second broad type of mixed methods research are sequential studies. They are generally two-phase designs. They can be explanatory (i.e., qualitative work helps to explain the initial quantitative results) or exploratory (i.e., results of the initial qualitative study inform the second, quantitative study) in nature (Creswell & Plano-Clark, 2007).

Triangulation is a useful approach because it is time efficient and allows for each type of data to be analysed with its respective technique (Creswell & Plano-Clark, 2007). For these reasons, as well as the ability to give equal weighting to the qualitative and quantitative aspects, a concurrent triangulation design was utilised in this thesis. Although triangulation involves the difficult task of merging potentially different results, the benefits for this particular research topic outweigh the test of being challenged as a researcher. An embedded design would not be appropriate for examining the research aims of this thesis (outlined in section 3.4), as separate questions were not being asked in each method. Even though explanatory and exploratory designs are advantageous for certain research questions (e.g., examining outliers and creating measurements, respectively), they require resources and time beyond the scope of this thesis. Therefore, a concurrent triangulation design was the most suitable for the research questions at hand.
2.3 Benefits of mixed methods research

As highlighted in the Chapter 1, positive psychological outcomes are difficult to delineate. Purely quantitative or qualitative methods are insufficient for understanding the full experience of parenting a child with 22q11DS due to the complexities of the relatively unexplored area of positive psychological outcomes. Mixed method designs are a comprehensive and holistic means of researching a particular phenomenon. Using only quantitative methods leaves research vulnerable to being driven by the researchers’ own agenda. It means that there is a risk of not reflecting the participants’ experiences accurately (e.g., selecting an unsuitable theory as the basis of the research); or succumbing to the confirmation bias\(^5\), and thus potentially omitting important phenomena (Johnson & Onwuegbuzie, 2004). On the other hand, if qualitative research is conducted in isolation, it may not be generalisable to the understanding or prediction of issues affecting the wider population (Johnson & Onwuegbuzie, 2004). Further, there are a multitude of benefits associated with mixed methods research, especially when it comes to examining issues related to illness and healthcare experiences (Pope & Mays, 1995). These broadly include: a) examination of novel research areas, because an exploratory approach is permissible (rather than the need for clear hypotheses); b) richer data, as a range of perspectives can be included (including disempowered groups); c) more confidence in research results, because multiple methods allow for a holistic examination of a phenomenon; and d) synthesis of theories through an assessment of similarities and differences in the results obtained from different methods (e.g., Jick, 1979; Johnson et al., 2007; Pope & Mays, 1995; Tashakkori & Teddlie, 2003). Therefore, a mixed methods approach was considered the most appropriate and

\(^5\) Confirmation bias is the tendency to interpret new evidence as confirmation of existing theories or preconceptions (Wason, 1960).
advantageous way of beginning to investigate the unchartered topic of the positive aspects of parenting a child with a developmental disability.

Additionally, much of the research surrounding health, illness, and disability has been conducted and reviewed by health professionals. This means that professionals’ perspectives have been given precedence over and above other stakeholders (e.g., parents of the person affected by the disability). Therefore, there may be unexplored issues that need to be addressed from a healthcare consumer viewpoint that are not yet recognised or validated by the current state of research. Using both qualitative and quantitative work allows for expansion of the perspectives included in the research. More specifically, the researcher can pursue their own agenda and hypotheses (e.g., as informed clinical experience or previous research) through quantitative research, whilst also gaining access to the views of the participants through qualitative methods (O’Cathain et al., 2007). This gives weight to the meanings, experiences, and views of a variety of stakeholders (Pope & Mays, 1995). Another advantage is that paradoxes between the two data sources can open up new ways of thinking about a particular topic and enable further theory conceptualisation (Rossman & Wilson, 1985).

2.4 Use of mixed methods in this thesis

In order to develop an experiential understanding of parents who have a child with a developmental disability (particularly 22q11DS) and explore the intricacies of positive psychological outcomes, a mixed methods approach was selected; with two qualitative studies and one quantitative study included in this thesis. A concurrent triangulation design was chosen because it allowed time efficient exploration of the research questions outlined below. The qualitative and quantitative aspects of the thesis were conducted in parallel and merged for final discussion, with equal weight given to
each study. Merging was done by noting convergence and divergence between the data
sources, and providing theoretical explanations for the same (see chapter 8). The
overarching research questions of this thesis are as follows. The synthesis of the
individual studies (both quantitative and qualitative) aims to provide insight on these
issues. The quantitative study (chapter 4) explores many types of developmental
disabilities (e.g., 22q11DS, Prader-Willi syndrome), whereas the qualitative studies
(chapters 5 & 6) draw on parents who have a child with 22q11DS only, due to the
syndrome’s unique features:

1. How do parents interpret their parenting experience of a child with a
developmental disability?
2. How do they perceive that phenomenon as impacting sense-making and their
own psychological wellbeing?
3. In what ways might the experience be different for parents of children at
different stages of life?
Chapter 3
Psychological Growth in Parents who have a Child with a Developmental Disability

3.0 Abstract

3.0.1 Background
The negative psychological impact of having a child with a developmental disability has been recorded throughout the literature (e.g., poorer mental health). Aspects of positive psychological outcomes such as resilience and growth are beginning to receive more attention, however little is known about the factors that promote positive outcomes in this population. Therefore, the aim of this exploratory study was to identify potential predictors of psychological growth in parents who have a child with a developmental disability.

3.0.2 Method
The sample comprised 432 parents of children with developmental disabilities. An online survey was utilised, with questions relating to the participant’s coping, social support, perceptions of family-centred services, and psychological growth.

3.0.3 Results
A regression model was built, demonstrating that increased use of positive reappraisal as a coping mechanism; parental perceptions of coordinated and comprehensive health care; and child’s age were associated with greater psychological growth. Greater discrepancy between ideal and actual practical social support along with use of escape avoidance as a coping mechanism were associated with less psychological growth.

3.0.4 Conclusions
It is realistic for parents who have a child with a developmental disability to experience positive psychological growth. This study provides a platform for future research.
3.1 Background

(Singer, 2006b) The negative impact of having a child with a developmental disability has been recorded throughout the literature in terms of poorer mental health and marital adaptation (e.g., Florian & Findler, 2001), depressive symptoms (Singer, 2006b), parenting stress (e.g., Davis & Carter, 2008), and somatic symptoms (e.g., Ha, Hing, Seltzer, & Greenberg, 2008). However, the more positive aspects of parenting a child with a developmental disability, such as psychological growth, are beginning to gain traction in the literature. Much of this research has been qualitative, without examination of factors that may affect these positive outcomes. Therefore, the aim of this study was to explore factors that may predict psychological growth in parents of children affected by a developmental disability. The limited research available in this field means that we have used psychological theory to inform our investigation and analyses.

Trauma is defined as an event that is unexpected, out of the ordinary, creates long-lasting problems, and substantially interrupts one’s personal narrative (Tedeschi & Calhoun, 1995). Parents who have a child with a developmental disability are likely to experience events that could elicit traumatic distress, inclusive of the risk of re-traumatisation throughout the child’s life. These events include but are not limited to shock at diagnosis, hospital stays, separation from the child, witnessing medical procedures, fear of disability and/or death, reduced quality of life, and impact on daily activities. In a study of parents with a premature infant in a neonatal intensive care unit, both mothers and fathers experienced clinically significant traumatic distress persisting up to 6 months after the child was born, regardless of the severity of the individual child’s illness (Binder et al., 2011). Further, in an Australian study of parents who had a child that underwent cardiac surgery before the age of 3 months, evidence of
experiencing traumatic distress was present in the majority of parents. That is, 83% of participants endorsed at least one trauma symptom (i.e., dissociation [e.g., feeling numb]; re-experiencing [e.g., dreams of the trauma]; avoidance [e.g., avoiding places that remind the person of the traumatic event]; and arousal [e.g., difficulty sleeping]) at a clinical level, and 27% reported trauma symptoms consistent with a diagnosis of acute stress disorder (Franich-Ray et al., 2013). Therefore, it is likely that parents who have a child with a developmental disability experience traumatic responses.

When faced with traumatic distress, people can succumb, survive with impairment (e.g., experience depression), recover (i.e. demonstrate resilience), or thrive (O'Leary & Ickovics, 1994). Thriving is also known as psychological growth and can include changes in self (e.g., new awareness of a possible authentic self), improved relationships (e.g., with fellow trauma survivors), changes in life philosophy (e.g., meaning and purpose in life), changes in priorities (e.g., appreciating the simple things), and enhanced spiritual beliefs (e.g., return to faith). Although parents who have a child with a developmental disability are at risk of experiencing traumatic distress, trauma can be a catalyst for positive psychological change or growth out of adversity; that is, posttraumatic growth (Joseph & Linley, 2005; Tedeschi & Calhoun, 1996). For this growth to occur, the trauma-related information must be integrated into the person’s worldview (Joseph & Linley, 2005). Although relatively unexplored in parents who have a child with a developmental disability, posttraumatic growth has been found in a similar population, namely, parents who have had a child in an intensive care unit. Eighty-eight percent of these parents reported a great degree of positive change as a result of the experience (Colville & Cream, 2009).

In order for clinicians to understand parental responses to a child’s developmental disability, delineation of the mix of emotions that result from such
challenges is necessary. Much is known about the potential adverse effects of parenting a child with a developmental disability (e.g., depressive symptoms). Factors these parents experience, such as socioeconomic disadvantage, child behaviour problems, and lack of support from services may explain the difference in psychological wellbeing between parents of children with developmental disability and those with typically developing children (see Hastings, 2016 for a review). However, a pathological focus has meant that the more positive aspects of the experience of raising a child with a developmental disability have been neglected. Although psychosocial distress is important to recognise and treat, a strength-based approach can make use of a person’s natural resources to improve their overall functioning (e.g., Wehmeyer, 2014).

Positive mental health is similar in mothers of children with developmental disability compared to mothers of other children, despite their risk of negative psychological outcomes (Hastings, 2016). Further, there is a relatively large body of work highlighting the positive gain parents can experience in relation to their child who has a developmental disability (e.g., Hastings et al., 2002). That is, parents perceive positive outcomes for themselves (e.g., growing as a person) and have positive perspectives about the impact the child has had on the family (e.g., making the family more tolerant). Parents have reported experiencing new perspectives on life, increased sensitivity, improved family dynamics, and increased confidence and assertiveness (Hastings & Taunt, 2002; Taunt & Hastings, 2002), yet the concept of psychological growth overall has received little research attention. These positive perceptions are important because they predict parental wellbeing (e.g., Baker, Blacher, & Olsson, 2005) which is essential, not only for the parents themselves but for the other members of the family, including the child with the disability and his/her siblings (Graunegaard, Andersen, & Skov, 2011b; Orsmond & Seltzer, 2009).
Generally speaking, psychological growth in people who have experienced traumas such as having cancer has been associated with higher socio-economic status (Cordova, Cunningham, Carlson, & Andrykowski, 2001), higher education levels (Sears, Stanton, & Danoff-Burg, 2003), time since diagnosis (Cordova et al., 2001), social support (Cadell, Regehr, & Hemsworth, 2003), and emotion-focused coping styles such as positive reappraisal (Urcuyo, Boyers, Carver, & Antoni, 2005). Indeed, Joseph and Linley’s organismic valuing theory (2005) purports that psychosocial factors influence how posttraumatic stress is managed and thus whether people experience psychological growth. If a person’s social environment is facilitative of their fundamental psychological needs of autonomy, competence, and relatedness, it is likely that they will experience psychological growth. There is evidence of the importance of these factors in parents who have a child with a developmental disability, although it has not been specifically explored from this perspective. For example, families with children with disabilities have been found to have better quality of life when their health care services have a family-centred approach (Davis & Gavidia-Payne, 2009). A family-centred approach is when care is sensitive to the individual family’s needs, wishes, and values (King, Rosenbaum, & King, 1995). Parents’ opinions of the care that they and their child receive may (positively and negatively) impact their perceptions of their child’s developmental disability (Davis & Gavidia-Payne, 2009; King, King, & Goffin, 1999). The time spent in contact with healthcare services means that it is likely to be part of their social environment (Davis & Gavidia-Payne, 2009), which can thus facilitate (or prevent) psychological growth. Related to this, social support is also known to be important for parental wellbeing; either by integrating parents into a larger social network, or protecting them from the negative impact of stressful events (Armstrong, Birnie-Lefcovitch, & Ungar, 2005). Access to social support predicts
parenting stress in families affected by disability better than aspects of child functioning (Smith, Oliver, & Innocenti, 2001). Therefore, it is likely that perceptions of healthcare and social support contribute to psychological growth.

Problem-focused coping, whereby an individual modifies themselves or the environment to manage a difficult person-environment relationship, is traditionally viewed as adaptive (Folkman & Lazarus, 1990). However, for parents of a child with a developmental disability, emotion-focused coping may prove more useful to manage the demands associated with a large caregiving burden. Emotion-focused coping functions as a tool to change either the meaning of, or attendance to, the stressful situation, even though the actual conditions have not changed (Folkman & Lazarus, 1990). Lazarus and Folkman's (1984) coping theory proposes that parents in this situation may use positive perceptions as a tool to manage their stressors. Indeed, the emotion-coping strategy of reframing (i.e., viewing an event in a more positive light; positive reappraisal) is a significant predictor of perceived positive gain (i.e., positive experiences associated with the child) in parents of children with a developmental disability (Minnes, Perry, & Weiss, 2015). Further, accommodative coping is related to fewer depressive symptoms, along with greater self-acceptance (Seltzer, Greenberg, Floyd, & Hong, 2004). There are many potential stressors associated with having a child with a developmental disability, such as hospital stays, separation from their child, witnessing medical procedures, fear of disability and/or death, reduced quality of life, and impact on daily activities. To our knowledge, there is no research specifically examining the effect of the child’s age on parents’ psychological growth. However, we know that more exposure to a stressor can also improve adaptation (Lazarus & Folkman, 1984). For example, parents who have an older child with autism have reported experiencing a declining degree of emotional distress as their child ages (Gray, 2006). Although not
specific to parents of children with developmental disability, benefit finding has been shown to be affected by the amount of time that had passed since the stressor onset (Helgeson et al., 2006). Hence we propose that psychological growth associated may increase with the child’s age.

Although positive psychological change is possible after traumatic and stressful events, this phenomenon has been under-researched in parents who have a child with a rare developmental disability. The current study aims to address this research gap. Joseph and Linley’s (2005) organismic valuing theory of traumatic growth is the most appropriate model to apply because a) becoming a parent of child with a developmental disability conceptually fits the definition of “trauma”, b) there is evidence that parents who have a child with a developmental disability are at increased risk of experiencing traumatic distress, and c) there is a validated measure to assess this type of psychological growth. Therefore, the aim of the current study was to explore participant and child characteristics that were associated with psychological growth in a sample of parents of children with developmental disabilities. We specifically recruited parents of children with rare genetic disabilities because much of the literature focuses on other health conditions or more common disabilities, such as autism. Further, a previous study has indicated that despite stress, anxiety, depression, and child characteristics, parents who have a child with a rare developmental disability can experience positive gains associated with their child (Griffith et al., 2011b). To the authors’ knowledge, there are no other quantitative papers that examine this type of psychological growth in parents who have a child with rare syndromes.

Based on Joseph and Linley’s organismic valuing theory (2005), it was anticipated that more positive social support (emotional and practical), positive reappraisal as a way of coping, and having access to services that were more family-
centred, would significantly contribute to a model of psychological growth. Further, we predicted that the child’s age would be associated with psychological growth because previous research has demonstrated that greater exposure to a stressor can improve adaptation (Lazarus & Folkman, 1984) and time since a medical diagnosis can impact psychological growth (Cordova et al., 2001). As our sample included participants who had a child without a diagnosis, the child’s age was included in the model as a proxy for diagnosis age.

3.2 Methods

3.2.1 Participants

Participants included 432 caregivers (92.6% parents) of children with developmental disabilities, aged between 19 and 79 years of age ($M = 42.59$). The majority were female (92.1%), married or in a de facto relationship (77.6%) and living with the child (88.4%). The modal education level (47%) was completion of a university Bachelor degree or above. Participants resided in a variety of countries, including those in North America (47.7%), Europe and the United Kingdom (23.8%), and Australia or New Zealand (22%).

The children’s diagnoses included 22q11.2 deletion syndrome (velo-cardio-facial syndrome [28%]), Prader-Willi syndrome (14.1%), Down syndrome (8.8%), tuberous sclerosis (7.2%), Williams syndrome (5.6%), Fragile X syndrome (4.9%), multiple congenital anomalies without a known diagnosis (7.4%), and other rare or very rare genetic syndromes (20.8%). Their mean age was 11.17 years (range: 1 month – 54 years).
3.2.2 Measures

The survey contained four measures, along with a demographic questionnaire to record factors such as the participants’ gender, age, education, income, and country of residence. The child’s age, developmental ability, and diagnosis were also collected.

Coping

The Ways of Coping Questionnaire (WAYS; Folkman & Lazarus, 1988) assessed the thoughts and actions used to cope with specific stressful encounters (i.e., situational coping); in this case, having a child with a developmental disorder. The scale consists of 66 items. Eight ways of coping are measured: confrontive coping (e.g., *I did something which I didn't think would work, but at least I was doing something*), distancing (e.g., *I went along with fate; sometimes I just have bad luck*), self-controlling (e.g., *I tried to keep my feelings to myself*), seeking social support (e.g., *I talked to someone to find out more about the situation*), accepting responsibility (e.g., *I criticized or lectured myself*), escape-avoidance (e.g., *I hoped a miracle would happen*), planful problem solving (e.g., *I just concentrated on what I had to do next - the next step*), and positive reappraisal (e.g., *I changed or grew as a person in a good way*). Respondents rated how often they used each way of coping, using a four-point Likert scale (where 0 = does not apply/not used, and 3 = used a great deal). Internal consistency estimates range between .61 and .79 (Folkman & Lazarus, 1988).

Social support

The perceived provision of social support was tested through the short form of the Significant Other Scale (SOS; Power, Champion, & Artis, 1988). Participants rated their actual and ideal levels of emotional (2 questions; e.g., *Can you lean on and turn to this person in times of difficulty?*) and practical support (2 questions; e.g., *Does he/she give you practical help?*) for up to seven important people in their life.
questionnaire has satisfactory reliability and validity (Power et al., 1988). Emotional and practical social support were calculated as a discrepancy between the participant’s self-rated ideal and actual support. Therefore, a higher score means the participant is experiencing greater inconsistency between the support they receive and the support they would ideally like. A lower score demonstrates that the support they are receiving is close to their ideal perception of support.

**Family-centred services**

The Measure of Processes of Care (MPOC-56; King et al., 1995) was utilised to evaluate the participants’ perceptions of the care they and their children receive from services. This measure consists of 56 items rated on a 7-point scale. Responses were recorded from 1 = ‘Never’ to 7 = ‘To a great extent’ (along with a “not applicable” option), to statements beginning with, *In the past year, to what extent do the people/centre who work with your child…*. Five domains of family-centred care were assessed: enabling and partnership (e.g., *make you feel like a partner in your child’s care*?), providing general information (e.g., *have information available in various forms, such as booklet, kit, video, etc.?*), providing specific information about the child (e.g., *provide you with written information about your child’s progress*), coordinated and comprehensive care for the child (e.g., *plan together so they are all working in the same direction*), and respectful and supportive care (e.g., *accept you and your family in a non-judgemental way*). Various studies have demonstrated that the MPOC-56 has good reliability (internal consistency and test-retest) and validity (King et al., 1995).

**Psychological growth**

Changes in psychological wellbeing were measured through the Psychological Wellbeing - Post-Traumatic Changes Questionnaire (PWB-PTCQ; Joseph et al., 2011), specifically designed to examine psychological growth from the perspective of the
organismic valuing theory (i.e. the theory informing this research). Importantly, it can predict subjective wellbeing over and above other measures of psychological growth (i.e. the CiOQ and PTGI; Joseph et al., 2011). On each of the 18 items, participants indicated how much they perceived themselves to have changed as a result of having a child with a developmental disability; from 1 = Much less so now to 5 = Much more so now. For example, I have a sense of purpose in life. Scores 54 or lower indicate no psychological growth, scores between 55 and 72 indicate some psychological growth, and scores 73 or higher indicate high levels of psychological growth (Joseph, 2011). This questionnaire has demonstrated good reliability and validity across both a sample of people that had experienced trauma (including natural disasters, cancer, rape, bereavement, sexual abuse) and the general population (Joseph et al., 2011). The Danish version has also recently been found to be valid and reliable (la Cour, Nielsen, Andersen, & Madsen, 2016). To our knowledge, it has not been validated on parents who have a child with a developmental previously. However, it is a versatile instrument that can be used to examine the impact of many different types of trauma (Joseph et al. 2011). Therefore, it is deemed to be the most suitable tool available to examine psychological growth in parents who have a child with a developmental disability.

### 3.2.3 Procedure

Following ethics approval from the University of Newcastle’s Human Research Ethics Committee, participants who had children with genetic developmental disabilities (e.g., an as yet undiagnosed syndrome, 22q11.2 deletion syndrome, Prader-Willi syndrome, Down syndrome, or other genetic/rare syndromes) were recruited to complete a survey. The survey was hosted by the online survey software SurveyMonkey and informed consent was implied through the first question of the survey where participants could decline to participate or choose to proceed. It was
disseminated through online support groups such as Facebook pages, websites, and blogs. One hundred and ninety groups from all over the world were identified via established networks, electronic searches, and word of mouth. Of the 190 groups contacted to post the research, 110 agreed, 8 refused, and 72 did not respond after 3 contact attempts.

3.2.4 Data Analysis

Whilst calculating a true response rate in the current study is not possible due to the design, 682 people began the survey and 432 reached the points of consent that allowed their data to be used for analysis, indicating a response rate of 63.34%. As the survey was lengthy, containing 262 questions, participants were given the option to complete it across two sittings. Thus, there were missing responses for the experimental variables related to coping (i.e., confrontive coping [3.47%], distancing [3.94%], self-controlling [4.86%], seeking social support [6.02%], accepting responsibility [3.94%], escape-avoidance [6.25%], planful problem solving [4.17%], and positive reappraisal [5.79%]), family-centred services (i.e., enabling and partnership [19.44%], providing general information [18.06%], providing specific information about the child [17.36%], coordinated and comprehensive care for the child and family [20.37%], and respectful and supportive care [18.06%]), social support (emotional [30.09%] and practical [28.94%]), and psychological growth (10.42%). Pairwise deletion was used, as the data appeared to be missing at random; although the measures towards the end of the survey tended to have fewer responses.

SPSS version 21 was used to analyse all data. We assessed the internal consistency of the standardised measures (or their subscales) within our sample and found acceptable levels for virtually all of them. That is, coping (confrontive coping [\(\alpha = .38\]), distancing [\(\alpha = .54\)], self-controlling [\(\alpha = .54\)], seeking social support [\(\alpha = .75\)],
accepting responsibility \( \alpha = .57 \), escape avoidance \( \alpha = .75 \), planful problem solving \( \alpha = .72 \), positive reappraisal \( \alpha = .79 \); practical social support \( \alpha = .88 \), emotional social support \( \alpha = .89 \); family-centred services (enabling partnership \( \alpha = .96 \), general information \( \alpha = .94 \), specific information \( \alpha = .83 \), coordinated & comprehensive care \( \alpha = .96 \), respectful & supportive care \( \alpha = .93 \)); and psychological growth \( \alpha = .94 \).

Only variables with acceptable alpha scores (acceptable \( \alpha = .60 \) [Hair et al., 1998]) were included in regression analysis. That is, confrontive coping, distancing, self-controlling, and accepting responsibility were excluded.

### 3.3 Results

Means and standard deviations for the experimental measures (i.e., social support, ways of coping, & family-centred services) are outlined in Table 3.
Table 3. Means and standard deviations for subscales of social support, ways of coping, and family-centred services measures

<table>
<thead>
<tr>
<th></th>
<th>Mean</th>
<th>Standard deviation</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Social Support</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Discrepancy between ideal</td>
<td>.79</td>
<td>.93</td>
</tr>
<tr>
<td>and actual emotional social</td>
<td></td>
<td></td>
</tr>
<tr>
<td>support</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Discrepancy between ideal</td>
<td>.90</td>
<td>.91</td>
</tr>
<tr>
<td>and actual practical social</td>
<td></td>
<td></td>
</tr>
<tr>
<td>support</td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>Ways of Coping</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Confrontive coping</td>
<td>3.78</td>
<td>2.26</td>
</tr>
<tr>
<td>Distancing</td>
<td>4.31</td>
<td>2.66</td>
</tr>
<tr>
<td>Self-controlling</td>
<td>7.06</td>
<td>3.42</td>
</tr>
<tr>
<td>Seeking social support</td>
<td>7.76</td>
<td>4.19</td>
</tr>
<tr>
<td>Accepting responsibility</td>
<td>1.54</td>
<td>1.91</td>
</tr>
<tr>
<td>Escape avoidance</td>
<td>6.23</td>
<td>4.57</td>
</tr>
<tr>
<td>Planful problem solving</td>
<td>7.27</td>
<td>3.68</td>
</tr>
<tr>
<td>Positive reappraisal</td>
<td>9.47</td>
<td>4.97</td>
</tr>
<tr>
<td><strong>Family-centred Services</strong></td>
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<td></td>
</tr>
<tr>
<td>Enabling partnership</td>
<td>3.92</td>
<td>1.65</td>
</tr>
<tr>
<td>General information</td>
<td>2.07</td>
<td>1.66</td>
</tr>
<tr>
<td>Specific information</td>
<td>3.39</td>
<td>1.78</td>
</tr>
<tr>
<td>Coordinated &amp; comprehensive</td>
<td>3.71</td>
<td>1.67</td>
</tr>
<tr>
<td>care</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Respectful &amp; supportive care</td>
<td>4.36</td>
<td>1.65</td>
</tr>
</tbody>
</table>

3.3.1 A comparison of parents of young and adult children

The sample of parents with children aged 17 years of age or younger included 317 parents, and there were 87 parents who had children aged 18 years and older. Parents of younger children were significantly younger (M = 39.64, SD = 7.93) compared to parents of adult children (M = 53.1, SD = 7.56), \( t(411) = -14.39, p = .0005 \), two-tailed. Whilst the proportion of males in both groups were low (4.6%), there was a significantly higher proportion of males in the sample of parents with older children (11.1% vs 2.8%), \( \chi^2 (1, N = 416) = 11.283, p = .002 \). Consequently, we included gender and age as control variables in our cross-age group analysis.
To explore the impact of the child’s age on parental functioning, a series of ANCOVAs were conducted with the subscales of coping, social support, family-centred services, plus psychological growth as the independent variables and age and gender as covariates (see Table 4). The child’s current age had a trend level effect on emotional discrepancy score, $t(1, 397) = 3.68, p = .056, \eta^2 = .03$. An inspection of the mean scores indicated that parents of younger children reported higher emotional social support discrepancy and practical social support discrepancy scores, indicating a greater inconsistency between the support they receive and the support they would ideally like to receive. The child’s age group had a significant effect on psychological growth scores, $t(1, 365) = 7.77, p = .006, \eta^2 = .021$. The mean scores indicated that parents of young children reported less psychological growth compared to parents of adult children. There was also an effect of the age group of the child on the accepting responsibility coping style, $t(1, 393) = 4.87, p = .028, \eta^2 = .012$. The mean scores indicated that parents of adult children were more likely to score higher on accepting responsibility. Finally, the age group of the child had a significant effect on parents reporting that they had received specific information from their health care providers, $t(1, 346) = 12.6, p = .005, \eta^2 = .036$. That is, parents of younger children perceived that they received more specific information. No other significant between-group differences were identified on variables of interest including coping and family-centred services.
<table>
<thead>
<tr>
<th></th>
<th>Child aged 17 years or younger</th>
<th>Child aged 18 years or older</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Mean</td>
<td>Standard deviation</td>
</tr>
<tr>
<td>Social Support</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Discrepancy between</td>
<td>.88</td>
<td>.85</td>
</tr>
<tr>
<td>ideal and actual</td>
<td></td>
<td></td>
</tr>
<tr>
<td>emotional social</td>
<td></td>
<td></td>
</tr>
<tr>
<td>support</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Discrepancy between</td>
<td>.98</td>
<td>.88</td>
</tr>
<tr>
<td>ideal and actual</td>
<td></td>
<td></td>
</tr>
<tr>
<td>practical social</td>
<td></td>
<td></td>
</tr>
<tr>
<td>support</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Ways of Coping</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Confrontive coping</td>
<td>3.73</td>
<td>2.26</td>
</tr>
<tr>
<td>Distancing</td>
<td>4.28</td>
<td>2.70</td>
</tr>
<tr>
<td>Self-controlling</td>
<td>7.03</td>
<td>3.39</td>
</tr>
<tr>
<td>Seeking social support</td>
<td>7.75</td>
<td>4.19</td>
</tr>
<tr>
<td>Accepting responsibility</td>
<td>1.48</td>
<td>1.76</td>
</tr>
<tr>
<td>Escape avoidance</td>
<td>6.47</td>
<td>4.57</td>
</tr>
<tr>
<td>Planful problem solving</td>
<td>7.20</td>
<td>3.61</td>
</tr>
<tr>
<td>Family-centred</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Services</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Enabling partnership</td>
<td>4.01</td>
<td>1.63</td>
</tr>
<tr>
<td>General information</td>
<td>2.09</td>
<td>1.58</td>
</tr>
<tr>
<td>Specific information</td>
<td>3.61</td>
<td>1.69</td>
</tr>
<tr>
<td>Coordinated &amp;</td>
<td>3.75</td>
<td>1.64</td>
</tr>
<tr>
<td>comprehensive care</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Respectful &amp; supportive care</td>
<td>4.41</td>
<td>1.62</td>
</tr>
<tr>
<td>Psychological</td>
<td>Psychological growth</td>
<td></td>
</tr>
<tr>
<td>growth</td>
<td>64.09</td>
<td>13.48</td>
</tr>
</tbody>
</table>

**3.3.2 Psychological Growth**

We tested our main hypothesis using a multiple regression analysis. As this was an exploratory study, initially, a univariate search was conducted with each variable hypothesised to be associated with Psychological Growth. That is, the discrepancy between ideal and actual social support (emotional & practical); ways of coping (seeking social support, escape avoidance, planful problem solving, & positive
reappraisal); family-centred services (enabling partnership, general information, specific information, coordinated & comprehensive care, & respectful & supportive care); participant factors (marital status, country of residence, education level, & annual income); and child factors (current age, age at diagnosis, IQ, developmental level as rated by the participant) were all included. The significant variables from the univariate search (i.e., emotional social support discrepancy, practical social support discrepancy, confrontive coping, distancing, seeking social support, escape avoidance, planful problem solving, positive reappraisal, enabling partnership, general information, specific information, coordinated & comprehensive care, & respectful & supportive care, & child’s current age) were then all added to the model. Non-significant variables were removed one at a time, based on the highest p-value.

A model was created, with positive reappraisal, coordinated and comprehensive care, practical social support discrepancy, escape avoidance, and child’s current age being significant. The residuals were examined for outliers, normality, constant variance, and non-random patterns. These assumptions were met. The main effects in the model were found to be independent. Potential covariates were reintroduced, yet they did not improve the variation in Psychological Growth as was explained by all of the five independent variables together, that is $R^2 = .405$ (see Table 5). It was found that for a one unit increase in Positive Reappraisal, Coordinated and Comprehensive Care, and Child’s Current Age, there was an increase in Psychological Growth by 1.25, 1.72, and .186 points (respectively) on average, controlling for all the other variables. For a one unit increase in Practical Social Support discrepancy and Escape Avoidance coping, there was a decrease in Psychological Growth by 1.91 and .59 points (respectively) on average. In order to explore how much of the total variance in the dependent variable was uniquely explained by each of the variables in the model, squared part correlation
coefficients were calculated. The variable with the strongest relationship to psychological growth was positive reappraisal, which explained 19.1% of the total variance. The next strongest variables were coordinated and comprehensive care (3.8%), escape avoidance coping (3.2%), practical social support discrepancy (1.3%), and the child’s current age (1.2%).

Table 5. Variables associated with Psychological Growth: unstandardised and standardised regression coefficients and 95% CIs

<table>
<thead>
<tr>
<th>Variable</th>
<th>Unstandardised coefficients</th>
<th>Standardised coefficients</th>
<th>95% CI</th>
</tr>
</thead>
<tbody>
<tr>
<td>Positive reappraisal</td>
<td>1.25**</td>
<td>.46</td>
<td>[.984, 1.517]</td>
</tr>
<tr>
<td>Coordinated and comprehensive care</td>
<td>1.71**</td>
<td>.21</td>
<td>[.901, 2.532]</td>
</tr>
<tr>
<td>Practical social support discrepancy</td>
<td>-1.91*</td>
<td>-.12</td>
<td>[-3.490, -.335]</td>
</tr>
<tr>
<td>Escape avoidance</td>
<td>-.59**</td>
<td>-.19</td>
<td>[-.895, -.283]</td>
</tr>
<tr>
<td>Child’s age</td>
<td>.186*</td>
<td>.11</td>
<td>[.025, .347]</td>
</tr>
</tbody>
</table>

\[ R^2 = .405 \]
\[ F(5, 266) = 36.26 \]

Note. CI = Confidence Interval. * \( p \leq .05 \). ** \( p < .01 \).

3.4 Discussion

Over the years there has been a focus on the negative outcomes, such as stress and mental health problems, associated with parenting a child with a developmental disability. However, increasingly reports emerge that many parents not only adapt well to having a child with a disability but indeed can thrive and grow as individuals. The recognition of resilience and psychological growth among parents at risk of poor psychological wellbeing is important. A better understanding of the underpinning buffering and protective mechanisms can lead to more targeted interventions, with a
positive transactional effects on the whole family system. In the current study we aimed to investigate psychological growth among parents of children with developmental disabilities and importantly, the factors associated with psychological growth in this cohort. We found that many of the participants had experienced at least some psychological growth since having their child. In terms of Joseph and Linley’s (2005) organismic valuing process theory, this demonstrates that many participants had integrated their experiences of having a child with a developmental disability positively into their worldview. That is, parents reported changing life values and developing strengths after having their child. For example, participants endorsed statements related to purpose in life, acceptance of self, and hope for the future as a result of their child’s developmental disability. Our model exploring variables related to psychological growth explained just over 40% of the variance in this sample. Greater positive reappraisal; coordinated and comprehensive care; and child’s age was associated with more psychological growth; and greater practical social support discrepancy and escape avoidance was associated with less psychological growth.

More specifically, we hypothesised that coping styles such as positive reappraisal would contribute to a model of psychological growth, which was supported in our sample. There is evidence to suggest that emotion-focused coping is a better strategy when faced with situations that cannot be changed through rational problem-solving (Collins, Baum, & Singer, 1983). This was evident in our sample, where greater use of positive reappraisal was associated with greater psychological growth. This is unsurprising because positive reappraisal refers to efforts to create meaning by focusing on personal growth. It is also unsurprising because there is likely an overlap between the two concepts. Positive reappraisal describes efforts to create positive meaning by focusing on personal growth and can include a religious dimension (Folkman &
Lazarus, 1988). Psychological growth in the context of this paper refers to positive psychological changes in self, relationships, life philosophy, priorities, and spiritual beliefs. However, it is not necessarily effortful like positive reappraisal. Future research can delineate the different ways positive reappraisal and psychological growth manifest in parents who have a child with a developmental disability.

Interestingly, greater use of another type of emotion-focused coping (escape avoidance) was associated with less psychological growth suggesting that aspects of emotion-focused coping may have different roles. We know that following catastrophic trauma, escape avoidance is useful in the immediate aftermath of the traumatic event as a protective factor. However, as distress becomes more regulated, continued use of avoidant coping inhibits purposeful rumination and meaning making (Bryant & Harvey, 1995). This recognition has contributed to the belief that emotion-focused coping is less useful than problem-focused coping for wellbeing. Further research can unravel positive aspects of emotion-focused coping, shedding light on more specific ways in which it may be useful for coping (perhaps even growth) within populations in which situational changes are not possible. Adaptive coping for this population may differ from those who are caring for a typically developing child. Social support is worthy of consideration in the context of parenting a child with a developmental disability, as the Buffering model suggests that social support can protect people from potentially harmful effects of stressful events (e.g., Cobb, 1976). Although seeking social support as a way of coping did not significantly contribute to the model, the discrepancy between ideal and actual practical social support was associated with psychological growth. Emotional social support was not associated with psychological growth. It is possible that the participants in this study were generally satisfied with the support they receive, particularly parents who have an adult child. They may receive emotional
support from their partner, because the vast majority were married or in a de facto relationship. Certainly the degree of support available from a spouse is associated with satisfaction with family functioning (Snowdon, Cameron, & Dunham, 1994). However, the discrepancy the participants reported between their ideal and actual practical social support demonstrates the need to bridge the gap in terms of practical social support. This is especially true for parents who have a younger child, as they were significantly less satisfied than parents who had an older child. Social support could be improved by healthcare and disability professionals referring parents to local support services which can foster relationships as their child grows and their family’s needs change.

Parents who perceived their healthcare services as more coordinated and comprehensive reported greater psychological growth. This is expected due to the amount of time this group of parents spend in contact with a variety of health services as a result of their child’s ongoing support needs. It is logical that coordination between these services would be helpful for parents and impact their experiences positively. Davis and Gavidia-Payne (2009) also found that coordinated and comprehensive care promoted family quality of life. The other measures of family-centred services (i.e., enabling partnership, general information, specific information, & respectful & supportive care) were not associated with psychological growth in our study. This is not to say that these other aspects of family-centred care are unimportant. Rather, all aspects of family-centred services should be emphasised in order to empower parents and positively influence their wellbeing (e.g., Dunst, Trivette, & Hamby, 2007). Further, research has demonstrated that unless parents have positive experiences in their intervention programme, it is unlikely their quality of life will be improved, despite the amount of time spent engaged in the programme (Davis & Gavidia-Payne, 2009).
Therefore, the current study along with previous research highlights the importance of family-centred services for positive outcomes.

As hypothesised, we found that the child’s age was positively related to psychological growth. This may mean that psychological growth simply needs a period of time to develop; to allow the parent time to reflect. This is in keeping with Lazarus and Folkman’s (1984) coping theory which suggests that more experience with the stressor leads to greater adaption. Alternatively, psychological growth may be fostered once the immediate health concerns in early childhood are managed (e.g., heart defect repairs). The child’s age at diagnosis was not related to psychological growth, which makes sense in this coping context, as parents may have suspected there were problems with the child’s development well before a diagnosis was provided. Thus, they have been adapting to the ‘stressor’ regardless of a diagnosis.

The child’s developmental level was also not significantly associated with psychological growth in the model. Positive psychological outcomes are generally unaffected by the child’s symptom severity (Hastings, 2016). For example, research by Smith et al. (2001) supports our findings, as they demonstrated that factors such as social support predict parenting stress better than the child’s functioning. This may also be the reason the child’s estimated intellectual functioning did not contribute to the model. However, it is important to note that the responses in our study may have been biased because the parents rated their child’s developmental level. Many participants did not know their child’s IQ or had never had it tested. A clinical assessment of the child’s developmental ability may provide more insight. Surprisingly, participant factors such as their marital status, country of residence, education level, and annual income did not contribute to psychological growth. The relative homogeneity of these factors in the sample may have contributed to this.
To summarise, psychological growth was associated with greater positive reappraisal; coordinated and comprehensive care; and an older child. A bigger discrepancy between ideal and actual practical social support discrepancy and more use of escape avoidance as a way of coping was associated with less psychological growth. However, it is important to consider the complexities of family systems, including the large number of societal, individual, and child factors (including those unexplored in this study) that transactionally contribute to the development of psychological growth amongst parents. This study provides a starting point for further research and development of interventions, as well as areas for healthcare professionals to reflect on in their practice. It is necessary for clinicians to consider parents as an important factor in their child’s health. They must provide information for the family that encourages realistic expectations, both positive and negative (Kisler & McConachie, 2010). Professionals should avoid concentrating on pathology and weaknesses only. Strengths can also be acknowledged during appointments and assessments, and may have a positive effect on parents’ wellbeing. Simple changes to practice such as encouraging basic counselling skills; that is, active listening, good communication, and empathy, so that parents firstly feel ‘heard’ and then secondly, referring parents to relevant information about the syndrome and appropriate support services would be very beneficial. Therefore, healthcare professionals can help to provide the right environment for fostering psychological growth for parents.

3.4.1 Limitations

These results need to be considered alongside a number of methodological limitations. The online nature of the survey made it impossible to determine the response rate, or compare potential participants to those that completed the survey. This means the risk of bias in our sample may be particularly high, as people who adapted
well to their child or felt strongly about the topic may have been more likely to participate. However, care was taken to word the study advertisement neutrally as to not attract only positive parenting experiences. Also, our diagnostic groupings were too varied to provide a meaningful between-groups comparison. Some research has demonstrated that syndrome specific features may lead to different outcomes for parents (e.g., Abbeduto et al., 2004); although a recent study demonstrated that parental coping strategies were strongly related to parental positive and negative outcomes, rather than the child’s diagnosis (Minnes et al., 2015). As this study was exploratory in nature, it provides a platform for future research to examine the impact (if any) of different diagnoses and/or behavioural phenotypes on psychological growth in parents.

The large age range represented in our sample poses difficulties too. As mentioned previously, greater exposure to a stressor leads to better adaptation (Lazarus & Folkman, 1984). We found some differences between those who are parenting a child (i.e. 17 years or younger) compared to those with an adult child (i.e. 18 years or older), such as the discrepancy between ideal and actual social support. However, there are many ages and stages contained within these two groups. Given the developmental experience of parenting, and how perceptions change over time with experience, there are likely subtleties within these broad ranges that need to be explored further. We hope that our preliminary findings provide impetus for future research to examine the way the psychological growth may change with time and experience.

Another limitation is that the measure of psychological growth we used is designed to assess perceived changes in psychological wellbeing following traumatic events. Despite this, the PWB-PTCQ was the most appropriate measure for assessing psychological growth as is has been shown to demonstrate incremental validity over and above existing measures as a predictor of subjective wellbeing including coping (Joseph
et al, 2012). Further, a recent systematic review highlighted a high prevalence of acute and posttraumatic stress symptoms in parents who have a child with a serious illness (Woolf, Muscara, Anderson, & McCarthy, 2015). In the current study, even though we did not measure for traumatic distress, caring for a disabled child results in chronicity of stress (Davis & Carter, 2008); and positive changes despite such stress was evident for these participants. The results of our study highlight that the PWB-PTCQ may be useful to assess positive changes as co-existing with stress, regardless of whether trauma symptoms are present. Future studies could examine the impact of trauma and posttraumatic growth in this group.

3.4.2 Conclusions

This study provides insight into potential predictors of psychological growth in parents who have a child with a developmental disability. It highlights that psychological growth is achievable and indeed realistic for this group. The need for communication between different healthcare professions is evident in this sample, along with a need to bridge the gap between parents’ ideal and actual support. This may be helped by providing parents with genuine expectations of their experiences, both positive and negative. Finally, the way parents cope impacts their psychological growth, and it may be that specific aspects of emotion-based coping strategies are more useful when problem solving is irrelevant to the stressor. Future research should aim to provide interventions to foster social support and promote specific adaptive coping mechanisms (e.g., positive perceptions, such as reappraisal). This may be useful to facilitate parental psychological growth and thus wellbeing.
3.4.3 The next stage…

Given the findings of this survey, we sought to explore this phenomenon in the context of a single developmental disability, in order to provide greater insight into the potential for psychological growth despite distress. The impact 22q11DS has on parents is under-researched. However, the syndrome’s unique features (e.g., variable expressivity, often delayed diagnosis) provide an opportune avenue for study of the interpretations parents may have, both positive and negative. Qualitative methods can offer a rich, insider’s perspective, with the chance to explore a particular phenomenon on its own terms. Therefore, the next stage of this thesis is qualitative, in order to develop an experiential understanding of parents who have a child with 22q11DS and explore the intricacies of positive psychological outcomes. Next, the philosophical underpinnings of the qualitative methodology are provided, followed by two qualitative studies which examine the parental experience of having a child with 22q11DS at different stages of life.
Chapter 4

Qualitative Methodology: Interpretative Phenomenological Analysis (IPA)

4.0 Chapter Statement

When studying parental experiences of parenting a child with a disability and positive outcomes, it is difficult to capture a) the essence of such a phenomenon, and b) the subjective meaning-making individuals bring to these experiences through a nomothetic enquiry. Therefore, when exploring unique phenomena that are poorly researched or challenge established meta-theoretical viewpoints, a qualitative method can reveal the individual experience and inform the direction of nomothetic enquiry. Any researcher embarking on qualitative research needs to underpin their research question with the philosophical principles behind the chosen qualitative methodology, and be explicit about their own philosophical position or risk conducting their research from a positivist perspective. To gain insight into the experience of having a child with 22q11DS, interpretative phenomenology is useful as an investigative approach because it is concerned with subjective consciousness regarding an external reality.

“Interpretations of truth” agreed upon by consensus viewpoints are sought rather than “truth” itself (Spinelli, 2005). This chapter of the thesis will define the philosophical underpinnings and methodological approach of the studies in Chapters 5 and 6 which aim to explore the phenomenological, interpreted subjective reality of the parental experience of parenting a child with a disability.

4.1 Epistemology

A positivistic approach, highlighting measurement and experimentation has traditionally dominated psychology. This is because the mental health disciplines have aimed to be “scientific”, and as such have borrowed their research philosophy and
methods from the natural sciences (Joseph et al., 2009). Positivism seeks an objective, empirical, and systematic foundation of all knowledge (Bracken, 2002). A scientific understanding of the natural world involves removing the experiential or functional qualities (e.g., beauty, use) and this has been brought over into the behavioural sciences.

However, the experiential aspect of psychology has been largely neglected, despite its proposed importance (Smith, 1996). More recently in the 1980s and 1990s, the appropriateness and ability of nomothetic empiricism in resolving psychological issues has been questioned (Bracken & Thomas, 2001; Joseph et al., 2009), as researchers highlighted the importance of context, values, and partnership in understanding human experience (Bracken & Thomas, 2001). For example, the significance of events, such as having a child with a developmental disability, cannot be explained nomothetically (Bracken, 2002) because the lived experience is unique to each individual.

Qualitative research has the potential to fill this void, as it aims to understand the subjective experience of a phenomenon from the participant’s perspective rather than answer a researcher’s hypothetical questions (Joseph et al., 2009). However, an understanding of the philosophical foundations is essential, otherwise the researcher risks utilising qualitative methods through a nomothetic lens; that is, conducting this type of research from a cause and effect, generalizable positivist tradition. Importantly, including qualitative investigations in this thesis has the potential to offer an alternative viewpoint to the quantitative study of this thesis that is more aligned with the metatheoretical frameworks of the medical model (Joseph et al., 2009).

As I am interested in the underlying mechanisms that explain observable phenomena (Blaikie, 1991), my position in seeking a qualitative enquiry as part of this thesis aligns well with a critical realist position that emphasises that the social world is
objective, and comprises relations that are not accessible through direct observation (Blaikie, 1991). From a critical realist standpoint, discursive accounts of mentation are grounded in social practices (Parker, 2002) by examining the individual experience through language, analogy, describing, interpretation, metaphor, and meaning making.

4.2 Phenomenology

Critical realists seek to explore the individual meaning-making of unique phenomena. Phenomenology is a philosophical school of thought which aims to study and capture the human experience. The relationship between objective (how the world really is) and subjective (how we perceive the world) realities is examined through this approach. There are several leading figures in phenomenology, most notably Husserl (1859-1938), Heidegger (1889-1976), Merleau-Ponty (1908-1961), and Sartre (1905-1980). Husserl first highlighted the relationship between experience and perception versus reality (i.e., intentionality). This is the orientation or directedness of one’s consciousness; the basic interpreting process (Smith, Flowers, & Larkin, 2009; Spinelli, 2005). Each act of intentionality is comprised two aspects: noema (where the attention is directed or the what) and noesis (mode of experience or the how). For example, when a parent hears the diagnosis of their child, the noema would be the content of the medical professional’s speech and the noesis would be how they interpret the speech based on their individual biases, such as perceptions of those affected by developmental and medical issues. These elements together form the overall experience and thus how each person perceives and responds to the situation. Our experience influences our perception, yet this perception influences the way we view the experience. Husserl suggested it was important to extricate oneself from the activity and instead reflect self-consciously. This reduction requires the researcher to “bracket”, or set aside their
prejudgements. That is, the researcher must separate the natural and interpreted world by relying on intuition and searching for consistent meaning.

Husserl’s work was extended upon by several other philosophers, such as his student Heidegger. Whereas Husserl’s ideas were from the transcendental branch of phenomenology, Heidegger was more existential (and hermeneutic) in his approach. He suggested that the *dasein* or unique attribute of humans is our consciousness and knowledge “that we are” (Smith et al., 2009; Spinelli, 2005). However, making meaning of experiences must always be considered in this uniquely human context; that is, we are always being in relation to the world: objects, activities, other people, language, and culture (Smith et al., 2009). There are ontological (collective principles of being) and ontic (particular ways the ontological characteristics are manifested) aspects of existence. For example, the ontological feeling of empathy may be expressed ontically as a parent learning sign language to overcome their child’s speech difficulties. All interactions with the world are *dasein*; however, any reflection is through interpretation. Thus, according to Heidegger, one’s experience cannot be extricated from the interpretation of the phenomenon. Whereas Husserl was interested in going “back to the things themselves” (Smith et al., 2009, p. 12), Heidegger suggested this was actually examining “the thing itself as it appears to shows itself” (Smith et al., 2009, p. 24). In terms of hermeneutics this means there are both visible and concealed meanings in any discourse, and as such it is important to examine the context from which it has come (Smith et al., 2009; Spinelli, 2005).

Heidegger, along with Merleau-Ponty and Sartre helped to move phenomenology from Husserl’s abstract descriptions towards an interpretative viewpoint which in turn moves the understanding of the phenomenon from abstract describing to interpreting the interaction between self and the world. The concept of
examining the lived experience can be seen through Merleau-Ponty’s idea of body-subjects. He argued that we are embodied in the world; that is, the body is a tool for communicating with the world, not solely an object in the world (Smith et al., 2009). As one’s unique perspective shapes their knowledge, it is impossible to share another’s experience (yet it can be empathised with; Smith et al., 2009). Although the lived experience can never be fully encapsulated, it is still important to include it and reflect upon it. Sartre expanded on this, suggesting that nothingness (or things that are absent) are just as important as those that are present. They equally affect how one interprets the world (Smith et al., 2009). Sartre also agreed with the notion that reality is unable to be captured, as well as being beyond understanding (Spinelli, 2005). These contributions to phenomenology demonstrate how people are engaged in the world; and that the presence and/or absence of social relationships affect experience.

4.3 Hermeneutics

The second philosophical school that may prove useful in addressing questions surrounding the experience of parenting a child with a developmental disability is hermeneutics: the theory of interpretation. This involves finding the meaning in terms of when the text was produced, as well as the time and context of its uncovering. As previously mentioned, Heidegger was a proponent of the role of hermeneutics within phenomenology. Schleiermacher (1768 - 1834) and Gadamer (1900 - 2002) were also major hermeneutic theorists. Schleiermacher proposed the need to understand the author as well as the words themselves; that is, there are two ways to interpret a text: both grammatical (exact textual meaning) and psychological (author’s personal meaning; (Smith et al., 2009). Through a rich and thorough analysis, the interpreter can have a greater understanding of any given piece than the original author (Smith et al., 2009).
Further, the concept of the hermeneutic circle is of great importance. In order to understand a text, it is necessary to appreciate both the “part” and the “whole” as meaning can be made at many different levels in multiple ways. For example, a word must be interpreted in light of its sentence; the sentence must be read in relation to the paragraph, which must be considered with regard to the entire text.

Gadamer highlighted the “dance” between the new information a text brings to the reader, and the preconceptions the reader projects on to the text. These biases may only come to light during the interpretation process. As such, it is important for the qualitative researcher to practice reflexivity. That is, note must be taken of how the researcher has influenced the research, perhaps through their own experiences or cultural assumptions. Reflexivity is necessary for transparency, yet it is also an acknowledgement of the participants’ expertise in their own lives, as the research is a joint product of the researcher and participant. This is pertinent for the ensuing articles, which, myself being a woman without children, are most certainly a collaboration of the researcher and the researched (Ashworth, 2008). Once the researcher can engage in the spirit of openness: a double hermeneutic dance between the researcher and the researched, insight into the meaning of a discourse can become apparent (Smith et al., 2009).

4.4 Idiography

The final philosophical influence that could provide insight to the topic at hand is idiography. Contrary to popular methods in psychology, which tend to be nomothetic (i.e., drawing conclusions at the group level, for example group means and statistics); idiography relates to the detail of a particular case. This allows a rigorous, meaningful analysis of each participant’s story, which can then be explained in relation to others
interpreting the phenomenon. Nomothetic methods risk misrepresenting a given experience through categorising it before enough is known to do so (Galton, 1883). Idiography means that the researcher takes interest in the specific group’s experiences, rather than looking at population outcomes.

Symbolic interactionism draws together these different philosophies (i.e., phenomenology, hermeneutics, and idiography). Through this viewpoint, the social scientist should be concerned with individuals’ meaning-making of events as well as the idea that those meanings are only created through interpretation in and as a result of social interaction (Smith, 1996). Symbolic interactionism underpins a particular qualitative method, known as interpretative phenomenological analysis. This method helps to provide an account of how situated and related qualities of human understanding come about.

4.5 Interpretative Phenomenological Analysis

The interpretivist paradigm developed as a critique of positivism in the social sciences and sits well within qualitative research as it emphasises retaining the integrity of the phenomenon (Blaikie, 1991). That is, the subjectivity of experience is noted and valued over generalisability. Central to interpretivism is the idea of understanding human behaviour, rather than explaining it as is the focus of nomothetic research. Phenomenology emphasises focusing on the experience itself, rather than trying to categorise it based on biases or assumptions. As such, interpretivist qualitative methods can be useful for examining interpretations of the phenomenon of parenting a child with 22q11DS.

Interpretative Phenomenological Analysis (IPA) is a critical realist methodological approach, developed by Jonathan Smith as an alternative to social
cognition and discourse analysis in psychological research (Smith, 1996). It seeks to provide a detailed explanation of how people make sense of a major life experience in its own terms (Smith et al., 2009). As contact with an experience is always through interpretation, the researcher in IPA can assist its appearance (through interviewing) and help make sense of it (through analysis). A researcher’s access to the participant’s personal world is affected by their own conceptions, therefore a double hermeneutic is involved (Smith & Osborn, 2008). That is, the researcher is making sense of how the participant makes sense of their world. Although hermeneutic analysis was originally designed for historical texts, it is clear that the principles described earlier (see Section 4.3) are highly useful for IPA. Phenomenology and symbolic interactionism form the theoretical basis of IPA (Smith, 1996). IPA is the methodology utilised for the qualitative aspect of this thesis.

4.5.1 Why IPA?

Ultimately, IPA was chosen as the qualitative methodology for this thesis because it is used for exploring in detail how people make sense of a major life event. That is, it was a natural fit for exploring the phenomenon of parenting a child with 22q11DS. IPA has some overlap with other qualitative methodologies but it is unique because, whilst it stays grounded in the participants’ accounts, it can also extend beyond the participants’ own sense-making and conceptualisations through a combination of psychological, interpretative, and idiographic components. Another qualitative analysis approach that could have been used in this thesis is grounded theory (Glaser & Strauss, 2009). Grounded theory is popular in social scientific disciplines, partly because its highly structured approach appeals to the positivism that is ingrained in these fields (Smith et al., 2009). It samples on a large scale with the aim of providing a theoretical-and/or conceptual-level account of a particular phenomenon. In contrast, IPA seeks a
more detailed and nuanced analysis of the lived experience (Smith et al., 2009). Convergence and divergence between participants is emphasised. An in-depth exploration of parents who have a child with 22q11DS (as opposed to a grounded theory approach) is most suitable, considering the limited research in this field and keeping in mind that IPA is particularly useful when the topic at hand is complex, poorly understood, or previously explored (Smith et al., 2009). Therefore, it was the most suitable methodology for examining the ‘lived’ experience of parents who have a child with 22q11DS.

4.5.2 Conducting IPA

Utilisation of IPA is a commitment to exploring, describing, and interpreting the way in which participants make sense of a given phenomenon (Smith et al., 2009). It is a useful method for poorly explored/understood or complex issues because it explores the experience in its own terms. The subject of this thesis is one such example; parenting a child with a developmental disability creates a significant impact on one’s life, yet it is a complicated and little-examined topic; particularly the positive effect of the experience. Whilst psychological theory can help inform these phenomenological investigations, one must be open to “the data of experience” free from assumptions (Smith et al., 2009; Spinelli, 2005). Bracketing of relevant literature (i.e., suspending the influence of such biases) can be required to truly become immersed in the interpretive process.

IPA is intended to be adjusted and developed by its researchers, dependent on the question at hand (Smith, 2004). Through this flexible approach, IPA lends itself to in-depth, semi-structured interviewing; as it involves deep analysis of rich personal accounts (Smith, 2011; Smith & Osborn, 2008). Open-ended questions are utilised and can be grounded in relevant theory underpinning the research question. The researcher
remains quizzical, encouraging a dialogue of sense-making, yet often remaining silent in the interview to enable the production of rich discourse. Nevertheless, if necessary, the researcher must intervene and gently direct the participant back to the topic at hand. The double hermeneutic can be looked at in two ways. Firstly, as previously mentioned, IPA involves the researcher trying to make sense of how the participant is trying to make sense of their world (Smith & Eatough, 2007). Secondly, the researcher is both empathetic and critical. That is, an “insider’s perspective” of the phenomenon is sought: the researcher takes the participant’s side (Smith & Eatough, 2007; Smith & Osborn, 2008), acknowledging that they are the expert in their own life. However, they also pose questions throughout the interpretative activity which can allow for insight beyond the participant’s understanding of their own sense-making. IPA studies are usually associated with topics of significant importance to the participant, and as such are transformative for the individual. The questions can relate to “hot cognition”, which are issues that are urgent and emotive (Smith & Eatough, 2007). The participant can engage with the experience closely, despite the passing of time. “Cool cognition” can also be examined through IPA, which refers to more long-term reflection across the life course (Smith & Eatough, 2007). However, hot cognitions can produce richer transcripts, which make for better IPA. An open interviewing technique that funnels down to the essential enquiry, is essential for this. Although it is focused on in-depth exploration of the individual experience, good IPA can influence generalisations.

4.5.3 Analytic Process of IPA

Throughout the analytic process, the principle of epoché; or bracketing the influence of prejudices and biases is of upmost importance. The researcher must put themselves aside, which can be a difficult task. One must focus on the experience itself rather than try to categorise it, based on biases or assumptions. Traditionally, the
positivist nature of psychology leads the researcher to drive the course of the data. However, in IPA the researcher must be willing to abandon their expected path and go with the participant. This is central: the researcher must be open to engaging with the unexpected. This respect of the participant’s expertise in their own life can allow for a more intimate discourse. Yet, redirection may be required if the participant ventures too far beyond the scope of the research. Therefore, the analysis begins in the interview, where the researcher’s openness can determine the quality of the data. Although the focus should be the phenomenon under investigation rather than the participant’s or researcher’s preconceived biases (in terms of IPA), these preconceptions can be acknowledged, often becoming apparent during the interpretation process. It is unrealistic to assume that biases can be fully bracketed although consciousness of biases is an integral aspect of the double auditing between researchers. Acknowledging biases and stating those biases helps the reader recognise the unique insights that researchers bring to the double hermeneutic, interpretative process of the analysis.

Transcription is the next stage of the analysis. The researcher gains a greater understanding of the participant’s experience and as such can begin ruminating on divergent and convergent themes, identify discrepancies within and between participants, and discover the richest, “hot cognitions”. The analysis generally proceeds from particular to shared, and descriptive to interpretative (Smith, Jarman, & Osborn, 1999), although of course there is flexibility throughout. It is also often an iterative and inductive cycle (Smith et al., 2009). See Appendix C for a worked example. After the analysis, the reader should be left with a sense of what it is like to experience a particular phenomenon. The specific analytic technique for each study is discussed further in Chapters 5 and 6.
4.5.4 Validity in Qualitative Research

Qualitative research seeks validity through credibility. The intersubjective nature of qualitative research positions the researcher relative to their own biases and presuppositions. Therefore, these were clearly stated (see Chapter 7). The greatest threat to credibility in qualitative research is a lack of openness to the data, a lack of creative social enquiry, and a move away from the rigorous steps of the method and philosophical underpinnings of the study (Schwandt, 2015). We created an audit trail to account for the systematic examination at each level of analysis (e.g., tracking between researchers). This allowed for transparency of the findings and enhanced the quality and transferability.

4.6 Studies of this thesis

The survey revealed that psychological growth is possible and perhaps even likely in parents who have a child with a developmental disability. However, qualitative investigation is necessary to learn about the “lived” experience and untangle the potential for psychological growth despite distress. IPA is suitable for exploring rich descriptions of subjective interpretations of this experience. A specific developmental disability (i.e., 22q11DS) was chosen as the focus of the qualitative aspect of this thesis. As previously mentioned, 22q11DS is a common microdeletion syndrome. Therefore, an understanding of parents’ journeys with the condition is useful in providing insight into caring experiences with developmental disability. Yet, the syndrome is clinically under-recognised and parents are likely to experience challenges associated with rarer syndromes, such as lack of awareness in professionals managing the child’s healthcare. Thus, examining the impact of 22q11DS on parents is useful in the context of positive psychological outcomes in parents who have a child with a developmental disability.
The qualitative studies of this thesis are outlined below. The recruitment procedures, interview process, and recruitment strategies are discussed in more detail in the Method section of each study. As IPA is an exploratory rather than explanatory and descriptive investigation, the research questions are broad and open, and hypotheses are not tested. The overall aim of the studies was to learn about primary caregivers’ positive and negative experiences in coping with their child’s 22q11DS. We aimed to identify features of their lived experience; in particular, how have they made sense of their experience? The interview questions were broad and open-ended, centred around: a) how the primary caregiver made sense of their experience parenting a child with a developmental disability, b) their positive and negative experiences of support, c) if and how they have changed as a result of their child’s condition, and d) how they expect their future will be influenced by their child and related experiences. The interview schedule was the same across groups (1. Parents who have a young child with 22q11DS, and 2. Parents who have an adult child with 22q11DS).

The analysis was based on procedures described by Smith et al. (2009), and was consistent across both qualitative studies. Independent audit of the themes occurred to enhance the quality, transferability, and transparency. Each transcript was analysed one at a time by the independent auditors, with relevant items and psychological constructs noted in the margin. Next, these themes were listed in a table and grouped based on different aspects of the parent’s experience. The themes were listed and analysed together to identify emerging shared experiences. The two auditors independently looked for new themes and removed themes with little support. The final list was a summary of themes covering all transcripts, grouped into clusters under appropriate headings that were agreed upon by both auditors after robust discussion. Conclusions were drawn in terms of the similarities and differences between participants.
Every effort was made to bracket biases and presuppositions: assumptions and beliefs were discussed during supervision. Throughout the process, the transcripts and recordings were consistently referred back to, to ensure there was enough supporting evidence for the emerging themes and that they were staying true to the data. Despite these attempts, the interpretations will be impacted by my own unique perspective of the world, and my own experiences (discussed further in Chapter 7).

In the next two chapters, the research lens of this thesis narrows to consider the poorly researched phenomenological experience of 22q11DS. They seek the subjectively “lived” experience of parenting a child with 22q11DS. Both positive and negative interpretations were sought in these studies. Chapter 5 outlines the experience as related to having a young child with 22q11DS; then chapter 6 moves on to parents who have an adult child with 22q11DS.
Chapter 5

“He'll be missing a piece of his alphabet but don’t write him off”: The positive and negative ‘lived’ experience of parenting a young child with 22q11.2 deletion syndrome

5.0 Abstract

5.0.1 Background
The presentation of 22q11.2 deletion syndrome (22q11DS) is symptomatically variable presenting diagnostic challenges for paediatricians and anxious uncertainty in parents. The ‘lived’ experience of parents with a small child diagnosed with 22q11DS is unknown; particularly how they make sense, both positive and negative, of their role as parents.

5.0.2 Method
Informed by Interpretative Phenomenological Analysis, idiographic and unique interpretations were sought from parents. Semi-structured interviews with two fathers and four mothers of a young child with 22q11DS (ages 8 months – 3 years) provided the data set for transcription and thematic analysis.

5.0.3 Results
Four themes embodied the uncertainty and fear that at times threatened to overwhelm these participants prior to a diagnosis of 22q11DS. Simultaneously they experienced future trepidation, systemic stigma, confusion at professional smoke screens, and ‘not knowing’. Heightened fear is agonisingly packed with distress, grief, guilt and hope. Despite unrelenting distress, they actively defied themselves to reframe fear and negativity allowing unexpected intra and interpersonal growth from the adversity of 22q11DS.
5.0.4 Conclusion

This study provides a lens into the experiences of parents struggling to make sense of their journey with 22q11DS in the early parenting years. It highlights that the early days are fraught with uncertainty, challenges within health systems, stigma of uncertainty and ignorance, and positively, the metamorphosis of personal strengths that is possible. Healthcare professionals are encouraged to openly discuss their limited knowledge, seek expert guidance and refer to the guidelines for management of 22q11DS for guiding parents on the journey with 22q11DS.

Key words: Velo-cardio-facial syndrome, IPA, anticipatory trauma, psychological growth
5.1 Introduction

The ongoing care required by a child diagnosed with a developmental disability, creates chronic levels of stress for many parents (e.g., Stuart & McGrew, 2009). Initial diagnostic uncertainty and subsequent prognosis is often exacerbated by the demands on family life from chronic or acute medical issues that may arise associated with the particular disability (Stewart & Mishel, 2000). The psychological wellbeing of parents of children with 22q11.2 deletion syndrome (22q11DS), a poorly understood syndrome, is unknown. Given the wide variability of symptoms possible in the presentation of 22q11DS, parents are likely to experience varying degrees of psychological distress pre and post diagnosis (Bales, Zaleski, & McPherson, 2010) while seeking and receiving a diagnosis, and raising and caring for their child. Therefore, this phenomenological study explores the subjective interpretations of parents who are experiencing the early years of caring for a child with 22q11DS. In particular, it seeks both positive and negative interpretations of their ‘lived’ experiences parenting a child aged less than 3 years old with 22q11DS.

The family social system can be disrupted or incapacitated by family crises (Burr, 1973). Crises will occur when stress reaches the peak at which a family can no longer cope (Figley, 1998). The most important predictor of parental stress in parents who have a child with an intellectual disability is parental negative attribution of their child’s behavioural problems or social acceptance (Saloviita, Itälinna, & Leinonen, 2003). Negative appraisal is associated with higher individual, marital, and family burden in parents with a child with autism spectrum disorder (Stuart & McGrew, 2009). Conversely, psychological wellbeing is promoted through positive and problem-focused coping (Pozo, Sarriá, & Brioso, 2014). Should families endure a crisis state for a long period of time (as may be experienced when a child has a developmental disability),
they are extremely vulnerable to burnout even though a crisis is considered to be temporary (Maslach & Jackson, 1982).

Burnout can include oscillation between avoidance and over engagement, listlessness, fatigue, and loss of empathy and those exposed to chronic illness and care of others are particularly susceptible to burnout (Figley, 1996). Studies indicate that parents of children with disabilities experience significantly elevated levels of clinical burnout (38%) compared to parents of healthy children (20%; Lindström et al., 2010). Importantly, and despite experienced distress, current research recognises that struggles with adversity can provide a platform for positive change and stress-related growth personally, socially, or in terms of coping (Park, Cohen, & Murch, 1996). For example, Cheshire, Barlow, & Powell (2010) found that in parenting a child with cerebral palsy conscious engagement in positive reinterpretation by parents helped them find meaning, reduce depression and stress (Cheshire, Barlow, & Powell, 2010).

The developmental disability 22q11DS (also known as velo-cardio-facial syndrome [VCFS]) is a unique condition with large inter- and intra-familial symptomatic variability (Shprintzen, 2008). More than 180 features are associated with the syndrome including characteristic facial features, congenital heart defects and palatal anomalies (McDonald-McGinn et al., 1999). 22q11DS can manifest through intellectual disability and/or learning problems, with specific cognitive impairments in executive dysfunction (Bish et al., 2005), attention deficits (Niklasson et al., 2005), and social impairments (Shashi et al., 2012). Compared to the general population, people with 22q11DS are at greater risk of experiencing autism spectrum disorders (Fine et al., 2005), anxiety disorders (Fung et al., 2010), mood disorders (Green et al., 2009), and psychotic disorders (Murphy et al., 1999). Despite 22q11DS occurring in 1 in 4000 live births (Oskarsdottir et al., 2004), little is known about the subjective experiences of
parenting a child with this syndrome nor the impact on the immediate family functioning. The uncertainty that typically accompanies the diagnostic process and also the prognosis may be heightened in this population as compared to parents with children who are diagnosed very early with a more ‘predictable’ developmental disability. Therefore, it is important to learn more about parents’ lived experience as related to 22q11DS. Mishel’s (1990) reconceptualization of the uncertainty in illness theory explains uncertainty as the inability to find meaning in illness-related events. Parents who have a child with intellectual disability of unknown origin have elevated emotional strain and regret compared to parents with a known disability or no disability (Lenhard, Breitenbach, Ebert, Schindelhauer-Deutscher, & Henn, 2005). Parents who perceive less personal control over their child’s condition experience greater uncertainty, ambiguity, and lack of clarity (Madeo, O’Brien, Bernhardt, & Biesecker, 2012). Conversely, flexibility that can occur through uncertainty in parenting a child with a chronic medical issue can facilitate psychological shifts and adaptability for integrating uncertainty more positively into the individual’s worldview (Mishel & Clayton, 2008). To our knowledge, there is no qualitative literature related to parents of very young children at the beginning of their care journey. However, a study of parents of adults with rare genetic intellectual disabilities has highlighted the uncertainty they feel in terms of their child’s syndrome (Griffith et al., 2011a). It is unclear how this relates to meaning making. As yet, there is little to inform systems of care on the experience of families with 22q11DS, particularly when the child is very young.

Therefore, the aim of this study was to explore the ‘lived’ interpreted experiences of parenting a young child with 22q11DS. That is: a) how the participant made sense of parenting a child with 22q11DS; b) if and how they have changed and; c) how they expect their future will be influenced by these experiences. Interpretative
Phenomenological Analysis (IPA; Smith, 1996) as a qualitative methodology is underpinned by phenomenology, double hermeneutics, and symbolic interactionism and is therefore suitable for exploring rich descriptions of the subjectively ‘lived’ experience as recounted by parents in the early years of parenting a young child with 22q11DS.

5.2 Method

5.2.1 Participants

Participants of this phenomenological qualitative study were two fathers and four mothers (including two married couples) who had a young child with de novo 22q11DS. The parents’ ages ranged from 29 to 42 and the children’s ages ranged from 8 months to 3 years. Participants were Australians and recruited via a study information pamphlet through online forums and one health care setting. Data collection, using semi-structured interviews concerning the phenomenon under investigation, were conducted at a time and place of the participants’ choosing. Demographic characteristics of participants and their children are outlined in Table 6. Pseudonyms are used to protect the participants’ confidentiality.
Table 6. Participant and child characteristics

<table>
<thead>
<tr>
<th>Participant</th>
<th>Gender</th>
<th>Age</th>
<th>Marital status</th>
<th>Child</th>
<th>Gender</th>
<th>Age</th>
<th>Age of diagnosis</th>
<th>Developmental ability*</th>
</tr>
</thead>
<tbody>
<tr>
<td>‘Stephanie’</td>
<td>F</td>
<td>42</td>
<td>Divorced</td>
<td>M</td>
<td></td>
<td>21 months</td>
<td>9 months</td>
<td>3</td>
</tr>
<tr>
<td>‘Elizabeth’</td>
<td>F</td>
<td>29</td>
<td>Married</td>
<td>M</td>
<td></td>
<td>8 months</td>
<td>1 day</td>
<td>6</td>
</tr>
<tr>
<td>‘Eric’</td>
<td>M</td>
<td></td>
<td>Not provided</td>
<td>M</td>
<td></td>
<td>3 years</td>
<td>9 months</td>
<td>3</td>
</tr>
<tr>
<td>‘Deborah’</td>
<td>F</td>
<td>36</td>
<td>Married</td>
<td>M</td>
<td></td>
<td>3 years</td>
<td>9 months</td>
<td>3</td>
</tr>
<tr>
<td>‘David’</td>
<td>M</td>
<td>42</td>
<td>Married</td>
<td>M</td>
<td></td>
<td>2 years</td>
<td>8 months</td>
<td>5</td>
</tr>
<tr>
<td>‘Frances’</td>
<td>F</td>
<td>36</td>
<td>Married</td>
<td>M</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

* is the child’s developmental ability as rated by the parents on a scale of 1 -7, where 1 = severely delayed, 7 = not delayed at all.

5.2.2 Procedure

Following university ethical approval, interested participants who met the study criteria; that is, a parent of a child with 22q11DS aged 3 years or younger, were sent the information letter, consent form, and demographic questionnaire. Signed consent forms and completed demographic questionnaires were collected at the time of the interview. The candidate conducted three semi-structured interviews via telephone, and three at participants’ homes. As a phenomenological investigation, the interview sought to investigate both positive and negative interpretations of the ‘lived’ experience of parenting a child with 22q11DS, through tunnelling down to the phenomenon of interest (Smith, 1996). The use of semi-structured interviews allowed flexibility for the interviewer and participant to engage in double hermeneutics, that is, a reiterative, interpretative exploration with the interviewer striving to make meaning of the participant’s interpretation of experience (Smith, 1996). One couple were interviewed.
together (David and Deborah) but were analysed as individual cases. The interviews were digitally audio-recorded and lasted between 47 minutes and 1.5 hours. Participants were reimbursed for their time with a $20 gift card.

5.2.3 Analytic strategy

Interpretative Phenomenological Analysis (IPA) sits within a critical realism perspective, methodologically seeking to understand how individuals socially construct, understand and interpret their world, particularly poorly understood phenomenon (Smith, 1996; Smith et al., 2009). Access to another’s world depends on the researcher’s preconceptions; and as such, the researcher takes an active role in the dynamic process of analysis. In this way, IPA is connected to hermeneutics. In IPA, a double hermeneutic facilitates interpretation (Smith & Osborn, 2008). That is, the researcher strives to make sense of the participant making sense of their world. From a sociological perspective, symbolic interactionism is also integral to IPA, given that all communication is symbolic and based upon interaction and meaning. IPA seeks the emphasis placed on meanings constructed by individuals within their social and personal world. This interface between natural and social worlds positions IPA firmly within the critical realist perspective and allows the researcher to describe the way the participants’ worlds are socially constructed and interpreted.

As this study aims to combine hermeneutics, interpretation and phenomenology, IPA is well suited as a method to explore, describe, and interpret the unique phenomenon under investigation, that is, the ‘lived’ experience of having a young child with 22q11DS. The interpretivist paradigm emphasises retaining the integrity of the phenomenon (Finlay, 2009), however Smith (2004) values the input of the researcher and does not advocate overuse of bracketing, though awareness of biases should be consciously sought. Phenomenology emphasises focusing on the experience itself,
rather than trying to categorise it based on biases or assumptions. IPA seeks to provide a
detailed explanation of how people make sense of a major life experience, in its own
terms (Smith et al., 2009). Subjectivity of experience is noted and valued over
generalisability.

IPA is intended to be adjusted and developed by its researchers, dependent on
the question at hand (Smith, 2004). Through this flexible approach, IPA lends itself to
deep analysis of rich personal accounts (Smith, 2011; Smith & Osborn, 2008). The
researcher is both empathetic and critical. That is, an “insider’s perspective” of the
phenomenon is sought: the researcher takes the participant’s side (Smith & Eatough,
2007; Smith & Osborn, 2008), acknowledging that they are the expert in their own life.
However, they also pose questions throughout the interpretative activity which can
allow for insight beyond the participant’s understanding of their own sense-making.
IPA studies are usually associated with topics of significant importance to the
participant, and as such are transformative for the individual.

5.2.4 Analytic procedure

The analysis was based on procedures described by Smith, Flowers, and Larkin
(2009), see Table 7. Interviews were transcribed verbatim and analysed one at a time.
All data sets were de-identified with a pseudonym. The first stage of analysis involved
re-listening to the interview and re-reading the transcript. Next, relevant items and
psychological constructs were noted in the margin of the transcript as emergent themes.
Higher order themes and subthemes themes were listed in a table and grouped based on
different aspects of the parent’s experience. This procedure was carried out by the
candidate and the second supervisor independently. The subjective interpretation of
each gradually proceeded from descriptive to interpretative then particular to shared
(Smith et al., 1999). It was also an iterative and inductive cycle (Smith et al., 2009).
Finally, a narrative analytic account was used to link theory to themes generated through pertinent verbatim extracts from transcript. Conclusions were drawn in terms of the similarities and differences among participants. Throughout the process, the researchers consistently referred back to the transcripts and recordings to ensure they were staying true to the data.

Table 7. Stages of Interpretative Phenomenological Analysis

<table>
<thead>
<tr>
<th>Stage</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Re-listening, transcription, reading and re-reading.</td>
</tr>
<tr>
<td>2</td>
<td>Developing emergent themes through independent interpretation (e.g., noting).</td>
</tr>
<tr>
<td>3</td>
<td>Credibility established through robust researcher discussion.</td>
</tr>
<tr>
<td>4</td>
<td>Repeating stages 1 – 3 for the other five cases.</td>
</tr>
<tr>
<td>5</td>
<td>Searching for connections across emergent themes, identifying convergence and divergence.</td>
</tr>
<tr>
<td>7</td>
<td>Reviewing transcripts to validate interpretations in the results.</td>
</tr>
</tbody>
</table>

5.2.5 Trustworthiness, credibility, and transparency

Reliability and validity rigor in qualitative inquiry are regularly deliberated through the terms trustworthiness, credibility, and dependability. Guba and Lincoln (Guba, 1981; Guba & Lincoln, 1982; Guba & Lincoln, 1989) spoke of "trustworthiness" as providing rigor in qualitative research and encouraged qualitative researchers to evaluate their research post hoc. Their work remains seminal and germane however, verification in qualitative research, through a continual process of “checking,
confirming, making sure, and being certain” (p. 17, Morse, Barrett, Mayan, Olson, & Spiers, 2002) is now encouraged. A step by step verification throughout assures design quality, that is, right choice of method insuring within-design consistency and analytic proficiency for transparency (Teddlie & Tashakkori, 2009).

Interpretative qualitative research seeks human sense making rather than human experiential narratives (Denzin & Lincoln, 2011). Truth is thus irrelevant in seeking subjective interpretations of a phenomenon. Saturation, often the concern of grounded theory, is not a rigor concern in IPA which is seeking thematic representation that may be either convergent (across all interviews) or divergent (within one interview; Smith et al., 2009). Importantly rigor in IPA relies on investigator responsiveness that advocates purposive sampling of a small homogenous group, expertise for funnelling down to the research question, and adherence to a double hermeneutic, reiterative, investigative style of interviewing (Smith et al., 2009).

Inter-rater reliability is variably considered important or not important (Armstrong, Gosling, Weinman, & Marteau, 1997). The strict analytic guidelines of IPA safeguard final thematic representation where no theme is included that did not emerge through independent analysis and rigorous debate resulting in unique, rich and data substantiated themes in the final inter-rater consensus. Homogeneity guards against loss of interpretation through generalisation of experience. In IPA, validity and reliability are assured through trustworthy and steadfast adherence to the step by step protocols of auditing and analysis (Trochim, 2000).

5.2.6 Trustworthiness and Credibility

Therefore, the researchers sought to enhance trustworthiness and credibility of the findings rigorously at each level of analysis. The candidate and second supervisor independently conducted the initial thematic data audit prior to any discussion. Only
when each had completely and independently analysed the data did joint discuss begin concerning interpretations supported by rich thematic evidence. The first supervisor brought another level of rigor to the analysis debating differences and similarities. An audit trail throughout the process (from transcripts to tracking between the researchers) allowed for transparency of findings and enhanced the quality and transferability.

5.2.7 Researchers’ perspectives

Qualitative, interpretative studies are open to researchers’ own preconceptions and biases when interpreting data. The candidate and her supervisors are current researchers in disability and trauma within family life. As such, the researchers remained conscious of the need to bracket biases to guard against forcing the data. Independent audits, the audit trail, and reflection through discussion and write-up were utilised in an attempt to limit the impact of such prejudices. Simultaneously, it is important to acknowledge the unique insights from researchers’ experiences during the process of interpretative qualitative research.

5.3 Results

Four major themes emerged: (1) Agony of uncertainty; (2) Systemic stigma; (3) The pain and the gain; and (4) Making sense of THIS life/Growing me. These four themes embodied the uncertainty and fear that at times threatened to overwhelm these participants as related to a diagnosis of 22q11DS. Simultaneously they experienced future trepidation, systemic stigma, confusion at professional smoke screens, and ‘not knowing’. Heightened fear is agonisingly packed with distress, grief, guilt and hope. Despite unrelenting distress, they actively defied themselves to reframe fear and negativity allowing unexpected intra and interpersonal growth from the adversity of 22q11DS.
Table 8. Summary of superordinate themes

<table>
<thead>
<tr>
<th>Themes</th>
<th>Description</th>
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<tbody>
<tr>
<td><strong>Agony of uncertainty</strong></td>
<td>Participants’ lives are invaded by grief and sadness, particularly prior to the diagnosis when they are the only one searching for an answer to their child’s symptoms. Anxious uncertainty about how the syndrome is/will manifest is constant. Paradoxically, not knowing brings hope for the future.</td>
</tr>
<tr>
<td><strong>Systemic stigma</strong></td>
<td>Healthcare services increase feelings of uncertainty and frustration. Participants must fight to get the support their child needs.</td>
</tr>
<tr>
<td><strong>The pain and the gain</strong></td>
<td>Conflicting emotions exist simultaneously within participants. They feel loss, fear and distress; while at the same time experiencing gratitude, empathy, and hope.</td>
</tr>
<tr>
<td><strong>Making sense of THIS life/Growing me</strong></td>
<td>Participants actively search for meaning surrounding their child’s 22q11DS. They view their challenges as an opportunity to grow, finding new love and meaning in their lives.</td>
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</table>

5.3.1 Agony of uncertainty

*Grief/sadness/loss of expected role*

Prior to the diagnosis, participants experience doubt about their child’s symptoms. Suspicions invade their lives, and active imaginations conjure up worst-case scenarios; bringing distress and grief:

*I felt like I was going insane but I knew there was something that wasn’t right, and I was dreaming up terrible things ...We couldn’t think of anything to ask other than would he have reduced life expectancy?* [Deborah]

Participants describe being voiceless victims. Their instincts are dismissed by healthcare professionals who perceive their training to outweigh parenting knowledge. A sense of helplessness arises:

*They didn’t want to listen to me. They just... It was appalling.* [Stephanie]

Parents are stuck as the lone searcher. They cannot shake the feeling that something is being missed. Frances in particular was disturbed by premonitions:
The number 22 just kept going round and round my head every night. Honestly. It was like 22, 22...like, I know he’s got it. [Frances]

In contrast, Elizabeth did not have these suspicions; which brought about shock and sadness after her seemingly healthy child was born. Robbed of the choice and control over her child and her own life, she feels let down and misled:

It was like doomsday to me when he said it... I know that if it had been picked up, I would have chosen to abort, and so life would look very different for us at this point. [Elizabeth]

Although the diagnosis should have been picked up prenatally, careless work meant it was missed. Elizabeth feels as though she is missing out on the life she has worked so hard to build for herself. She must deal with the consequences of someone else’s mistake for the rest of her life.

In limbo/what is this about?

This theme highlights the uncertainty that emerges from having a young child with such a variable syndrome. Participants wonder how the diagnosis will reveal itself as their child grows. They find it difficult to enjoy the moment as they are caught up in continuous contemplation of what the syndrome is about:

What the fuck’s going to happen? I have no idea what VCFS really means... because there’s so many varying factors ... So that’s where I’m stuck ... I just don’t know. I’m in limbo. [Stephanie]

The ambiguity of 22q11DS brings about an anxious need to prepare for any medical, physical, or emotional issues that may arise. But how can one anticipate the unexpected? The enormity of battling an unknown enemy is an overwhelming responsibility. Restless questioning and investigation ensue:

What are his needs going to be? What sort of support will he need? Will he need to go to a special school, will he go to mainstream ...how will he cope? Will he have anxiety? Is he autistic? Does he have ADHD? Will he develop a mental illness later in life? Will he
be able to function in society, and will he be happy?! Will he... be healthy? ... What
curveball am I going to get thrown? [Frances]

Interestingly, Eric was the only participant who avoided this rumination. He did
not feel the need to learn about the syndrome, which saves him from the tormented
enquiry. He decides to face obstacles when they come instead of worrying about what
may or may not happen in the future:

_We didn’t delve too much into the literature and things that were given to us... We know_
there will be some challenges moving forward as he develops, but at this stage it hasn’t
really changed our lives at all [Eric]

Mixed up amongst fear of the unknown is a cautious hope. The participants dare
to dream that their child will be one who is less affected than most with 22q11DS. They
still dream for a normal or even exceptional life for their child:

_He’ll be missing a piece of his alphabet but don’t write him off:_ [Deborah]

**What is my child, what is the syndrome?**

Participants also question how well they know their child and in knowing, how
best to care. It is a constant cycle of questioning and uncertainty:

_He’s not doing the signs of what I’ve been told and what I’ve looked up... is it really_
VCFS doing that, or that because he had such a bad birth?! I’m going back around in
circles again... [Stephanie].

They are unsure which joys and challenges are simply part of who their child is,
or because of the 22q11DS. They wonder what their child would be like without the
deletion:

_That’s always something that I struggle with ... with the medical things ... with the_
feeding issues ... with the sleeping, we know that that is linked into the syndrome. But_
behaviour I know that they can have ASD, ADHD, anxiety. But what is normal boy, two-
year-old behaviour anyway? So it’s very hard to pinpoint what is... normal or not
[Frances].
**Cacophony of fears**

Due to the uncertainty, the participants are constantly haunted by a cacophony of fears. They grieve for themselves and the life they dreamed of having. They plead for reason in the chaos, needing an answer to the eternal question “why me?” The perceived injustice of the situations is present and overpowering, especially for Frances who has another child with health issues:

*It’s very heart-breaking (crying). I found out I’d lost a lot of time with my other child, stressing and worrying about her up until she had her surgeries, so I didn’t get to enjoy her as a baby (crying). And I didn’t bond to him as well… and then I felt that (sobs) why has it happened again, to me?* [Frances]

David speaks to these feelings, describing the sadness that can catch him unawares. An active campaign of hope and positivity is necessary to avoid wallowing in grief:

*If you can’t look forward, then it becomes a really, really bad time. I think the black dog will come and sit with you and you’ll just continue to dwell in your own self-pity.* [David]

The worry never leaves these parents, expanding into guilt about whether they caused the syndrome; despite knowing logically the deletions are de novo. David exemplifies this, ruminating over whether the medical treatment he was receiving at conception played a role:

*But there’s still – did I cause that? Did my actions cause that? We had no other choice, but I think maybe it’s my fault that we ended up in this situation.* [David]

**5.3.2 Systemic Stigma**

Adding to the frustration of uncertainty is the systemic stigma that pervades the participants’ experiences with healthcare services. Doubting their own instincts, the parents feel insecure and apologetic:
I almost felt like I had Munchausen’s (laughs) in and out, in and out, in and out. The child would vomit and then they’d get there and the child wouldn’t vomit anymore.

[Deborah]

Participants begin to take ownership of their concerns once they realise the seriousness of their child’s condition. They feel blocked at every turn and are constantly fighting for support. Parents comment on a mismatch between their intentions and the way they are perceived. It appears as though they are being difficult and disruptive, when they are simply doing what they feel is necessary to get their child the appropriate care:

I look like this nasty, snarly, foul-tempered, foul-mouthed woman with a baby on her hip... [Stephanie]

The parents perceive that the services created the monster that the services then dread dealing with. Parents must shout to be heard, which is frustrating and exhausting:

You get tired of pushing things. You get tired of when you’re walking in and see them rolling their eyes ... you think I’m a bitch because I’m trying to get my son better.

[Stephanie]

Stephanie in particular experiences extreme frustration at the authoritative ignorance. Her own expertise as a parent is ignored:

He read the MRI wrong! ... He came to the conclusion that my son was going to be violent, aggressive, non-communicative. [Stephanie]

She believes she knows more about her child than the counterfeit experts providing care. She cannot understand why healthcare professionals do not have the same emotional investment in her child as she does; finding them as a group to be cold, uncaring, and lacking in the practical skills she perceives to be necessary to help her son:

She didn’t look in his mouth to see if he’s got a cleft palate ... all they’re worried about is the money! [Stephanie]
5.3.3 The pain and the gain

Conflicting emotions exist simultaneously within the participants. For example, Frances feels she was swindled out of the mothering experience she desired after already having had a child with medical issues prior to the one with 22q11DS:

*I haven’t again had that experience of a normal baby... you know? Where everything is ok...so...I felt almost, sort of robbed of that? Of experiencing that...so yea (stops crying)...I felt that. But I also felt that, you know, I had to try to look at it from all angles and then see, well my daughter has prepared me.* [Frances]

At the same time, she is grateful for this prior learning that equipped her to be an advocate for her affected son. She recognises the experience that stole something precious from her was actually an opportunity for learning:

*So we’d gone through all of this with her... it almost prepared me for him, and what I was going to have to deal with ... I’d sort of become medicalised with her ... That’s why I’d been onto it. Because I think that’s what prepared me.* [Frances]

Although they experience loss, fear, and distress; participants actively search for meaning as a result of these feelings and challenges. They acknowledge while they are still unsure of what the significance of the situation is, they are certain a meaning will reveal itself eventually:

*I suppose that’s where I’m at, just trying to find the positive... I don’t know yet...* [Frances]

The pain and the gain is extended through a newfound empathy. The loss of normality in the participants’ lives brings about an appreciation for other people’s struggles. They channel their pain into empathy, care, and advocacy:

*I don’t look at disability the same way. So I think that that’s a really positive thing. You know, I want to try to advocate for children or people with disability. That never would have happened if it wasn’t for him, and it’s just seeing the world through different eyes.* [Frances]
As a health professional, Elizabeth particularly felt camaraderie with similar families; almost as if she is now a member of an exclusive club. She perceives she has a better understanding of and connection with her clients as a result of her son’s 22q11DS:

*I’ve tapped into this amazing secret world that not many people know about, which I can now use to help others, in a much better way. So I feel much better equipped to do the work that I’m already doing.* [Elizabeth]

Stephanie experiences a similar positive reappraisal of her situation. Despite the losses she has faced, there is a redefining of gratitude. She experiences glimmers of hope amongst the chaos of care:

*I’ve lost a lifestyle that I loved ... I’ve now gained a lifestyle that I was never going to ever have. So there are benefits.* [Stephanie]

### 5.3.4 Making sense of THIS life/Growing me

**Building relationships**

Similarly, participants search for meaning in relation to 22q11DS. They hypothesise whether the beginnings of growth they have experienced is the hidden purpose of the situation they have been thrust into. Redefining love is a salient feature of change. They realise their priority is now their child/ren and start to move to a more selfless, patient, and accepting existence. Frances speaks of the new type of love that has entered her life:

*You’ve really got to try to find this deep love to get through the challenging times (laughing)! And that’s a positive thing, like really sort of delving deep within yourself to try to find patience and acceptance, and all these things that if he hadn’t come into my life, well...I wouldn’t have known on the level of depth that I feel and have gone through.* [Frances]

This openness to love extends beyond their child. Family relationships are built for the better, with the affected child uniting the team towards a common goal. Old
wounds begin to heal; with previous conflicts no longer have the significance they once did:

Me and my father don’t have a great relationship unfortunately. It’s got better. But … he would give his kidney for my son… my son is everything to him now… You have to make the best of a bad situation. Not saying this is a bad situation, but I’m reconnecting with family … we’re close. [Stephanie]

**Opportunity not a burden**

Instead of viewing the challenges of parenting a child with 22q11DS as a burden, the parents decide it is an opportunity to become a better person. Whilst disappointment and regret are common emotions, parents do not lose sight of the lessons they perceive they need to learn:

In my trial of life, it provides a great opportunity … I get my moments when I ask why...but then I think this has been a really good opportunity to be able to love someone anyway. [David]

Some participants emphasise the spiritual aspect of this ‘opportunity’, as a chance to grow in their religious faith. Deborah knows that God gave her a child with 22q11DS because she had the resilience and education to manage. As such, she is accepting of her life path:

I think it’s because it’s part of God’s plan. I think that is the only simple answer … For me personally I can’t see that there’s been any harm come to it, I can only see good. [Deborah]

Deborah also perceives that even though God gave her struggles, he is there providing the resources for her to handle each situation. Her method of coping through faith gives her strength and happiness:

There’s so many things we look back and think, gee that was God’s providence in his life or our lives, working together. Other people might look back and think it was fate or whatever, but I think I look back and I see God’s hand in all of it. [Deborah]
Other participants did not speak of their journey as a spiritual one, however they engage in reflection on the meaning of their experiences. Their previous ideas of their purpose in life are challenged:

*He was a miracle. He was meant to be. I was meant to have him on this journey, whatever this journey is going to be.* [Stephanie]

They see their purpose has been redefined, but cannot quite figure out what their new role is. Their new significance is a blurry image that slowly gains focus with time:

*I’m still finding my feet, I have no idea what some days what I’m doing. However, when it does come to needing to get stuff done, I feel like I know exactly what I need to ask for...well if that’s the one good thing that has come out of it, I’ll gladly take it!* [Elizabeth]

**Counting blessings**

Gratitude is ever present throughout the participants’ accounts. They count their blessings and focus on what they do have rather than what is lacking:

*We’re very aware of how lucky we are to have him. At the moment and how well he’s doing – but yea, we love him to bits. Like I said, I think if we had found out sooner he might not be here with us today and now that he is here with us we feel very blessed with having him.* [Elizabeth]

Despite their child’s differences, they are thankful for the gift of a child and cannot imagine their life without them:

*I feel incredibly lucky that I’ve had the parenting training, the disability training, the medical training I’ve had...are all culminating beautifully at the moment, and I really feel for the parents that don’t have the background I do, because I think navigating through the system would be 100 times harder.* [Elizabeth]

This positive assessment also manifests as hope. The child’s young age, coupled with a diagnosis that is a “life sentence, not a death sentence” [Deborah] gives the participants hope that their child may live a normal life. Further, they see their role as that of a parent rather than a carer for a disabled child. Although they may struggle
along the way, they recognise things could be worse, and are optimistic about the future:

*He’s not dependent on you 365 days a year, 24 hours a day. He’s not going to a nursing home, you know when we die or something like that, it’s just a small little hiccup in the genetic pool.* [David]

### 5.4 Discussion

This qualitative study identified four major themes representative of these participants’ experiences parenting a child under three years old with 22q11DS: a) **Agony of uncertainty** - a dichotomous experience of grief and wonder; b) **Systemic stigma** - invalidation and stigmatisation within healthcare services that invalidated parents on their search for a diagnosis; c) **The pain and the gain** - simultaneous experiences of conflicting negative and positive emotions; and d) **Making sense of THIS life/growing me** – purposeful rumination allowing psychological growth despite distress.

This study highlights the ever-present and ongoing shadow of uncertainty experienced by these participants in parenting a young child with 22q11DS. Felt as being in purgatory without answers, not knowing leads to reflection about what life will bring for themselves and their child in the future. Similar to first responders who experience *anticipation of trauma* (van der Kolk, McFarlane, & Weisaeth, 1996), these parents are unable to live in the moment, and anxiously prepare for symptoms and events that may never appear. This anticipation creates a hypervigilance to potential traumatic events, evoking raised expectation of threat that can negatively impact decision-making (Papazoglou, 2013). Fear is constant, and grief about what could have been if it were not for the 22q11DS is not easily subdued.
The compounded impact of unavoidable stressors (e.g., difficulty with healthcare services) leaves these parents psychologically vulnerable. This is similar to a study of parents who have adult children with rare syndromes. Parents struggled with healthcare services and had to fight to get the support their child needed (Griffith et al., 2011a). However, all of the participants of this study recognised their own fragility to stress, and as such took active steps to embrace the positive aspects of their experiences.

Contradictory emotions (e.g., shame and pride) were experienced simultaneously. Participants work to bring about purposeful rumination to positively reframe their struggles. The opportunity for the co-existence of distress and psychological growth is welcomed with participants embracing 22q11DS as an opportunity for growing in their spirituality, feeling humbled, and uniting as a family. This mix of emotions has been shown in other studies of families affected by disability. For example, Kearney and Griffin (2001) found that parents experienced anguish and sorrow alongside hope, love, strength, and joy. A meta-analysis of benefit-finding after trauma showed that higher levels of benefit-finding are associated with more intrusive and avoidant thoughts about the stressor (Helgeson et al., 2006); again, highlighting the complex and conflicting emotions of parents with children with 22q11DS.

Despite being early in their journey with 22q11DS, the risk of burnout is recognised as very real for these parents. All showed a willingness to actively utilise their experiences with 22q11DS as a springboard for psychological wellbeing. This supports Folkman’s (1997) theory of meaning-based coping, where negative psychological states associated with significant stress may motivate people (either consciously or unconsciously) to create positive psychological states in order to gain relief. Hastings and Taunt (2002) have also suggested the usefulness of conceptualising positive perceptions as a style of coping in parents who have a child with a
developmental disability. This type of positive reappraisal has been demonstrated in other studies where a parent has a child with a severe disability (Graungaard et al., 2011a). Parents turned their experiences into resources such as engaging with hope. As yet, it is unclear which factors promote this positive coping; and how it is integrated with the conflicting emotions participants described.

The uncertainty that has dominated these parents’ experiences also align with Mischel’s (1990) reconceptualized uncertainty in illness theory. The amount of time since a stressor has passed has been shown to influence benefit-finding (Helgeson et al., 2006). However, parents who have a child with 22q11DS experience a range of stressors, not limited to one event. The syndrome can manifest in both chronic and critical ways, with uncertainty plaguing parents throughout their child’s life. These participants showed a desire to integrate the continuous uncertainty into their lives by reorganising their beliefs to avoid an chronic anticipatory state of distress associated with expectation of predictability (Mishel & Clayton, 2008).

In particular, Mischel (1990) notes that while reorganising, people can turn uncertainty from an aversive experience to an opportunity. This was present in our sample, where participants consciously referred to their child and the associated challenges as an opportunity rather than a burden. Ironically, the uncertainty brought hope for a happy future. Further, despite the children’s young age, participants were already describing the beginnings of growth (e.g., building relationships, gratitude) which is the desired outcome in Mischel’s (1990) theory. Longitudinal studies would be useful to examine if and how parents resolve the uncertainty of 22q11DS, and which factors promote specific outcomes, such as psychological growth.
5.4.1 Limitations

One potential limitation is that two sets of couples were included in the sample. The close relationship may have influenced their interpretations of their experiences, meaning that they provided relatively similar data. For example, Deborah and David were interviewed together and were in agreement about the impact their child with 22q11DS had on their lives. Their accounts complemented and supplemented each other, and they were very much committed to positively reinterpreting their journey together. Eric and Elizabeth had slightly different perceptions. Elizabeth was open about her frustration with the situation that had been thrust upon her, even though she deeply loves her son. Eric seemed more accepting (or perhaps avoidant), did not seem to have engaged in sense-making related to his son’s 22q11DS like his wife had. This does not necessarily indicate different interpretations. It may be an effect of gender or rapport with the interviewer. It is important to consider the study’s findings within the context of these participants and the study limitations. However, as a qualitative study, rather than seeking to generalise findings to all parents who have a child with 22q11DS, we sought detailed insights of this specific homogenous sample of parents with children under the age of 3. Due to the double hermeneutics employed in IPA, the researchers’ biases could have impacted on the study both positively and negatively. However, we took steps (e.g., audit trail, robust discussion) to attend to credibility and worthiness of the study at every step of the analytic process. Therefore, despite the limitations, this study provides a valuable contribution to the knowledge of 22q11DS and a basis for further research into the experiences of parents and families throughout the life of a child with 22q11DS including how they manage continued syndromic uncertainty and/or diagnosis of associated features (e.g., autism) at different stages of the child’s development.
5.4.2 Conclusions

This study provides unique and rich descriptions of the subjectively lived experience of parents of a young child with 22q11DS and how those interpretations are intrinsically linked to distress, anticipation of traumatic stress, and the potential for psychological growth. Purposeful rumination allowed them to positively reappraise their early experiences with 22q11DS, and develop strategies for redefining challenges as opportunities rather than burdens. According to Mischel’s (1990) theory, healthcare providers are inimitably connected to an individual’s interpretation of health care experiences such as having a child with 22q11DS. Therefore, healthcare professionals are well-placed to facilitate positive coping processes and validation of parental efforts through acknowledging any limited knowledge of 22q11DS, and collaboratively supporting parents according to the guidelines for managing 22q11DS (Bassett et al., 2011).

5.5 The next study

This phenomenological study has provided insight into parenting a young child with 22q11DS. In the early years, there is much uncertainty regarding the syndrome’s trajectory. As the child ages, there may be different joys and challenges on the journey, which may impact on the parents’ subjective interpretations. Therefore, Chapter 6 will explore the experience of parenting an adult child with 22q11DS.
Chapter 6

“You Don’t Know Until You Get There”: The Positive and Negative “Lived” Experience of Parenting an Adult Child with 22q11.2 Deletion Syndrome

Copyright © 2016 American Psychological Association. Reproduced with permission. The official citation that should be used in referencing this material is Goodwin, J., McCormack, L., & Campbell, L. E. (2016). “You Don’t Know Until You Get There”: The Positive and Negative “Lived” Experience of Parenting an Adult Child With 22q11.2 Deletion Syndrome. *Health Psychology*. Advance online publication. http://dx.doi.org/10.1037/hea0000415. This article may not exactly replicate the authoritative document published in the APA journal. It is not the copy of record. No further reproduction or distribution is permitted without written permission from the American Psychological Association.

6.0 Abstract

6.0.1 Objectives

22q.11.2 deletion syndrome, a complex phenotype associated with more than 180 features, presents complex challenges for parents including gaining an accurate diagnosis. Poorly researched, this phenomenological study sought the ‘lived’ interpretations of parents supporting an adult child with 22q11DS.

6.0.2 Method

Interpretative Phenomenological Analysis informed a detailed and open exploration of parenting a child through to adult life with 22q11DS. Using reiterative in-depth semi-structured interviews eight parents (two male, six female) of adult children with 22q11DS were individually interviewed providing the data set for transcription and thematic analysis.

6.0.3 Results

*Losing ‘I’ Finding ‘self’, overarched six subordinate themes that emerged from participants’ articulated descriptions of psychological distress and psychological growth. Distress in parenting a child with 22q11DS was experienced through stigma,
loss, grief, and guilt. Progressively, stigma undermined independence, friendships, and
instinctual judgement. Often isolated, ill-informed hierarchical structures experienced as
layers of obstruction and ignorance, triggered angry advocacy for their child. Diagnosis
brought opposing relief and grief. In time they came to value their unique
‘accomplishments’, gleaned on their journey with 22q11DS, and in turn, consciously
valued authentic ‘self’ expressed through empathy, humility, gratitude, and pride.

6.0.4 Conclusion

Unrelenting distress through societal, educational, and health care invalidation
persisted for decades for all participants. Conversely, distress facilitated psychological
growth for redefining ‘self’ and role as parents over time. Building on this
phenomenological cameo, future research can educate against the plight of 22q11DS
families. It can enlighten health care professionals in buffering against associated
stigma, blame, and self-doubt, and in fostering psychological wellbeing.

Key words: Velo-cardio-facial syndrome; 22q11DS; IPA; traumatic distress;
psychological growth
6.1 Introduction

Little is known of the experience of parenting a child with the developmental disability 22q11.2 deletion syndrome (22q11DS; also known as velo-cardio-facial syndrome) despite a prevalence of 1 in 4000 live births (Oskarsdottir et al., 2004). 22q11DS has a complex phenotype associated with more than 180 features including (most typically) characteristic facial features, congenital heart defects, and abnormalities of the palate (McDonald-McGinn et al., 1999). Parenting a child with any developmental disability presents unpredictable challenges (Carroll, 2013; Rolland & Walsh, 2006) likely to increase the carer’s risk of mental health problems such as depressive symptoms and anxiety (Hartling et al., 2014; Miodraga & Hodapp, 2010; Singer, 2006a). However, the experience of parents caring for a child in adult life with 22q11DS, despite the large inter- and intra-familial symptomatic variability that generates a distinctive group of features in each child, is poorly understood (Shprintzen, 2008). Additionally, and despite the unique variability in 22q11DS, there is no research that highlights the individual articulated account of parenting a child with 22q11DS into adult life. Therefore, this study aims to explore the ‘lived’ experience of parenting a child with 22q1.2DS into adult life. It explores both positive and negative subjective interpretations of the unique phenomenon from the parents’ perspective.

The behavioural phenotype of 22q11DS is characterised by intellectual disability and/or learning problems and specific cognitive impairments including executive dysfunction (Bish et al., 2005), attention deficits (Niklasson et al., 2005), and social impairment (Shashi et al., 2012). Comorbidity is high with ASD (Fine et al., 2005), anxiety disorders (Fung et al., 2010), mood (Green et al., 2009), and psychotic disorders compared with the general population (Murphy et al., 1999). In adult life, a higher rate of unemployment compared to those without disabilities (Sanford et al., 2011)
frequently inhibit financial independence creating relational and financial complications for both older parents and adult child.

Due to the variable nature of 22q11DS, poor public awareness, and lack of awareness of the syndrome among many health professionals, parents of children and adults with 22q11DS are poorly supported in their complex lives. Largely ignored in mainstream psychological research, inferences can only be made on their likely experience of disenfranchised grief, or grief/loss that is not recognised or validated by others (Doka, 1989), a plethora of sad emotions on receiving the diagnosis (as reported in Hallberg, Óskarsdóttir, & Klingberg, 2010) and sorrow for the child they desired or expected. Their own life dreams invariably and necessarily are placed on hold or adjusted as they assume a lifetime of care for a child who is unlikely to reach total independence. For some, traumatic responses to health-related events that threaten the life of their child may be cumulative and complex (Franich-Ray et al., 2013). When chronic illness and disability is part of a parenting experience there is likely shock at the diagnosis, long hospital stays forcing separation from the child, fear of disability and/or death, and reduced quality of life.

Trauma symptoms in parents who have a child with chronic or critical illnesses such as cancer and accidental injury are well-recognised (e.g., Brocque et al., 2010; McCarthy et al., 2012). For instance, 83% of parents whose child underwent cardiac surgery before the age of 3 months, exhibited evidence of experiencing at least one trauma response at a clinical level (Franich-Ray et al., 2013). Conversely, if medical treatment and hospitalization is experienced as traumatic by the child, difficulties can arise relationally with parents viewed as unintended accomplices of the traumatic event (Stuber & Shemesh, 2006). Therefore, it is possible that parents who have a child with
22q11DS also experience traumatic responses from both primary and vicarious exposure.

Despite psychological risks, there is a growing body of research that highlights the possibility of positive psychological changes as a result of a struggle with adversity (Joseph & Linley, 2008). With the rise of positive psychology in the early 1990s, the construct of growth out of adversity, or posttraumatic growth, has come to define distress as a catalyst for positive psychological change (Joseph & Linley, 2005; Tedeschi & Calhoun, 1996). Growth out of adversity is defined as a positive change in psychological functioning after trauma, such as developing strengths, changing life values and beliefs, and accepting personal limitations (Joseph, 2012).

Joseph and Linley’s (2005) organismic valuing process theory, purports that a traumatic event can shatter an individual’s former worldview causing traumatic distress. However, psychological growth out of such adversity will only occur if the individual is able to integrate the new trauma-related information into a new world view (Joseph & Linley, 2005). Additionally, if the social environment can provide the human needs of autonomy, competence, and relatedness, then growth will be promoted (Ryan & Deci, 2000). However, complicating recovery, integration of the trauma-related information can take a positive or negative pathway occurring in one of three ways: a) assimilation of the experience, returning to pre-trauma baseline but remaining vulnerable to future traumas (e.g. I’m invincible), b) negative accommodation of the experience causing psychopathology (e.g. bad things happen to me), or c) positive accommodation of the experience leading to growth (e.g. I can learn from this).

Psychological growth as a result of parenting a child with a developmental disability has been recorded in the literature. For example, families have been found to adapt and thrive in the face of adversity, constructing meaningful stories surrounding
the journey with their child (e.g., Green, 2002; King et al., 2006; Rolland & Walsh, 2006). Although parents may grieve the child they expected, they can discover new pathways of happiness or spirituality as a result of having a child with a developmental disability (e.g., King et al., 2006; Myers, Mackintosh, & Goin-Kochel, 2009; Retzaff, 2007). No research highlights whether parents experience these life-changing epiphanies as a function of having a child with 22q11DS.

This interpretative, phenomenological study explores subjective interpretations of parents who have raised a child with 22q11DS into adult life. It seeks both positive and negative ‘lived’ interpreted experiences of experiencing this unique disability from a parental perspective. Interpretative Phenomenological Analysis (IPA; Smith, 1996) underpinned by phenomenology, double hermeneutics, and symbolic interactionism, is a suitable qualitative methodology for this uniquely ‘lived’ experience (Smith, 2004) as it seeks idiographic meaning making of a diverse and often confusing phenomenon for parents and health professionals alike. As such, this study explored parental sense making of 22q11DS from the perspective of: a) parenting; b) experience of support; c) perception of ‘self’ change over time, and d) expectations of their future influenced by their child’s disability and related experiences.

6.2 Method

6.2.1 Participants

Eight parents (two male, six female) of an adult child with 22q11DS were recruited from a supporting foundation. Participants formed an homogenous, purposive sample relating to the unique phenomenon under investigation (Smith & Osborn, 2008). Inclusion criteria sought participants who did not have 22q11DS themselves. The demographic characteristics of participants and their children are outlined in Table 9.
Pseudonyms are used to protect the participants’ confidentiality. All eight participants were interviewed individually with two sets of couples contained in the sample. George and Gabriella are married to each other, as are Max and Maria.

Table 9. Participant and child characteristics

<table>
<thead>
<tr>
<th>Participant</th>
<th>Gender</th>
<th>Age</th>
<th>Marital status</th>
<th>Child</th>
<th>Gender</th>
<th>Age of diagnosis</th>
<th>Developmental ability*</th>
<th>IQ</th>
</tr>
</thead>
<tbody>
<tr>
<td>‘Anna’</td>
<td>F</td>
<td>54</td>
<td>Married</td>
<td></td>
<td>M</td>
<td>24</td>
<td>10 months</td>
<td>2</td>
</tr>
<tr>
<td>‘Gabriella’</td>
<td>F</td>
<td>63</td>
<td>Married</td>
<td></td>
<td>F</td>
<td>28</td>
<td>8 years</td>
<td>5</td>
</tr>
<tr>
<td>‘George’</td>
<td>M</td>
<td>63</td>
<td>Married</td>
<td></td>
<td>F</td>
<td>23</td>
<td>3 years</td>
<td>5</td>
</tr>
<tr>
<td>‘Maria’</td>
<td>F</td>
<td>58</td>
<td>Married</td>
<td></td>
<td>F</td>
<td>23</td>
<td>3 years</td>
<td>5</td>
</tr>
<tr>
<td>‘Max’</td>
<td>M</td>
<td>62</td>
<td>Married</td>
<td></td>
<td>F</td>
<td>23</td>
<td>3 years</td>
<td>5</td>
</tr>
<tr>
<td>‘Sandra’</td>
<td>F</td>
<td>55</td>
<td>Married</td>
<td></td>
<td>M</td>
<td>29</td>
<td>7 years</td>
<td>4</td>
</tr>
<tr>
<td>‘Tracy’</td>
<td>F</td>
<td>58</td>
<td>Married</td>
<td></td>
<td>M</td>
<td>25</td>
<td>3.5 years</td>
<td>5</td>
</tr>
<tr>
<td>‘Wendy’</td>
<td>F</td>
<td>57</td>
<td>Married</td>
<td></td>
<td>F</td>
<td>21</td>
<td>2 years</td>
<td>5</td>
</tr>
</tbody>
</table>

Note. * is the child’s developmental ability as rated by the parents on a scale of 1 -7, where 1 = severely delayed, 7 = not delayed at all.

6.2.2 Procedure

Following university human ethics clearance, recruitment occurred through an online support group. Additionally, letters were sent to parents who had participated in previous research at the University conducting this study and had consented to be notified regarding future studies. Potential participants were screened for eligibility following contact with the researcher. Prior to the interview, study materials (i.e., participant information statement, consent form, and outline of the semi-structured interview) were sent to the participants.
Data was collected through open-ended, semi-structured interview questions that allowed for reiterative sense-making of the participants’ rich, personal accounts (Smith, 2011; Smith & Osborn, 2008). Interviews were conducted one-on-one at the participant’s home, with the exception of two interviews completed via telephone at the request of the participants. All interviews were digitally audio-recorded and ranged in duration from 52 to 155 minutes. Participants were reimbursed for their time with a $20 gift card.

6.2.3 Epistemology

The philosophical underpinnings and methodological approach of the current study were based on phenomenology, symbolic interactionism, and critical realism. That is, we aimed to capture the constructed meaning around the phenomenon of having an adult child with 22q11DS by engaging the participants in reflective interpretation of their experience (Blaikie, 1991). Meaning-making is formed through interactions. As such, symbolic interactionism refers to the participants’ sense-making which is created as a result of social interaction, modified through individual interpretations (Smith, 1996). Further, as environments (and thus interpretations) vary, participants attribute their own subjective meanings to the experience of having an adult child with 22q11DS. By using a hermeneutic exploration and taking a critical realist stance, a researcher is able to capture both the objective and relative truths of participants. However, as the researchers’ access to the participants’ personal world is affected by their own conceptions, a double hermeneutic is involved. That is, the researcher strives to make sense of the participant making sense (Smith & Osborn, 2008).

6.2.4 Analysis

Rigour in qualitative research demands ongoing verification undertaken through step by step guidelines to ensure trustworthiness and validity. Thus, transparency
occurred through conducting procedures as described by Smith et al. (2009). Thus, interviews were audio-recorded and transcribed verbatim by the candidate. Independent auditing was conducted by the candidate and the second supervisor each developing an audit trail from reading and re-reading transcripts, noting relevant items and psychological constructs in the margin. The subjective interpretation of each researcher gradually proceeded from descriptive to interpretative (Smith et al., 1999). The candidate and her second supervisor then engaged in robust discussion to identify and agree on relevant convergent and divergent themes supported by rich data both within and across the data set. A summary of higher order themes and subthemes were grouped into clusters under appropriate headings. Finally, a narrative analytic account was used to link theory to themes generated through pertinent verbatim extracts from transcript. Saturation is not sought in IPA given the focus on divergent (one data set) and convergent (all data sets) rich themes (Smith, 1996). Conclusions were drawn in terms of the similarities and differences among participants (see Table 10).

Table 10. Stages of Interpretative Phenomenological Analysis

<table>
<thead>
<tr>
<th>Stage</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Re-listening, transcription, reading and re-reading.</td>
</tr>
<tr>
<td>2</td>
<td>Developing emergent themes through independent interpretation (e.g., noting).</td>
</tr>
<tr>
<td>3</td>
<td>Credibility established through robust researcher discussion.</td>
</tr>
<tr>
<td>4</td>
<td>Repeating stages 1 – 3 for the other seven cases.</td>
</tr>
<tr>
<td>5</td>
<td>Searching for connections across emergent themes, identifying convergence and divergence.</td>
</tr>
<tr>
<td>7</td>
<td>Reviewing transcripts to validate interpretations in the results.</td>
</tr>
</tbody>
</table>
6.2.5 Credibility

As an interpretative phenomenological analysis, this study sought validity through credibility, and reliability through dependability. Therefore, as the intersubjective nature of qualitative research positions the researcher relative to their own biases and presuppositions these need to be stated. Furthermore, the greatest threat to credibility in qualitative research is a lack of openness to the data, a lack of creative social enquiry, and a move away from the rigorous steps of the method and philosophical underpinnings of the study (Schwandt, 2015). Rather than a post-hoc assessment of worthiness, the audit trail accounted for the systematic examination at each level of analysis (e.g., transcripts, independent audits, meetings, notes, tracking between the candidate and second supervisor). This allowed for transparency of the findings and enhanced the quality and transferability. The discussions provided the conduit for commonalities and differences in the researchers’ individual interpretations to emerge. Throughout the process, the candidate and her supervisor consistently referred to transcripts and recordings, with rich data providing supporting evidence for the emerging themes and the double hermeneutic process.

6.2.6 Researchers’ perspectives

The candidate and her supervisors are current researchers in disability and trauma within family life. Each researcher was conscious of the need to bracket biases to guard against forcing the data into preconceived interpretations surrounding disability. However, the candidate and her supervisors were similarly conscious of the importance of knowledge and experience in this field of enquiry for engaging with the unexpected through independent audits, the audit trail, and reflection through discussion and write-up at all stages.
Table 11. Summary of themes

<table>
<thead>
<tr>
<th>Subordinate themes</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>Stigma and a double-edged sword</td>
<td>Stigma invades the participants’ lives because the ‘not knowing’ leaves them the target of judgement, or victims of indifferent professionals. The diagnosis is a double-edged sword of relief and grief.</td>
</tr>
<tr>
<td>Where is ‘I’</td>
<td>Participants lose themselves in the management of their child’s health, behaviour, and needs. There is no respite from this role, and as such relationships change.</td>
</tr>
<tr>
<td>Conflicting loss, grief, and guilt</td>
<td>The participants cautiously reflect on the life that could have been without 22q11DS and mourn each milestone that should have passed. Although logically they know their child’s deletions are de novo, they wonder if they did something to cause it.</td>
</tr>
<tr>
<td>Angry advocacy</td>
<td>Participants battle against hierarchical structures suspicious of their actions and intent. They must fight the layers of obstruction and lack of awareness of the syndrome to receive the care their child needs.</td>
</tr>
<tr>
<td>Pragmatic acceptance</td>
<td>There is an uneasy peace with the ambiguity of their child’s future. Participants re-evaluate their expectations and learn to celebrate success for their child.</td>
</tr>
<tr>
<td>Finding authenticity</td>
<td>Psychological growth is experienced through conscious engagement with empathy, humility, gratitude and pride.</td>
</tr>
</tbody>
</table>

6.3 Results

One superordinate theme: Losing ‘I’; Finding ‘self’; overarches six subordinate themes: (1) Stigma and a double-edged sword; (2) Where is ‘I’; (3) Conflicting loss, grief, and guilt; (4) Angry advocacy; (5) Pragmatic acceptance; and (6) Finding authenticity. These themes describe the isolation and stigma seeping into the participants’ lives as they progressively lose their own independence, friendships, and instinctual judgement. Battling against hierarchical structures suspicious of their actions and intent, angry advocacy pushes back against layers of obstruction and lack of awareness. Diagnosis is a double-edged sword of relief and grief demanding they co-journey an unchartered pathway with sometimes indifferent professionals. Finding ‘self’ comes through a pragmatic re-evaluation of ‘accomplishments’ that allows an authentic
and positive psychological shift in their whole-of-life interpretation. As such, psychological growth is experienced through conscious engagement with empathy, humility, gratitude and pride.

6.3.1 Stigma and a double-edged sword

This theme explores the stigmatisation experienced as hovering throughout their child’s life. Self-questioning and blame associated with why their child has this disability remains a lurking burden on these participants. As a consequence, guilt is a well-rehearsed response to perceived judgement by health professionals, particularly prior to the diagnosis. The ‘not knowing’ leaves them with the perception that they are the target of judgement; unfairly ladened with confusion and guilt:

They were very critical as to what I did or what I ate during my pregnancy or what medication I took ... they make you feel really uncomfortable. [Tracy]

The stigma is widespread, inclusive of educational settings, where these participants struggle at numerous levels to educate against ignorance in staff and other parents. They sense little understanding or empathy of the journey they are on. It is exhausting and irksome. Again, without open communication, suspicions of judgement leave them without a voice:

I don’t know whether they thought the syndrome could be caught like a disease... You’re at school but you’re on the perimeter ... we were just singled out and singled out on so many occasions. [Tracy]

For some, the obvious signs of disability bring open support. For those whose children do not immediately appear to be disabled, fear of invalidation are never far from the surface. There is the sense that these participants oscillate between the relief of their child being not quite disabled enough, and the frustration of ‘not quite there’ for support and assistance:
That’s the difficulty – that she’s not quite there, but she’s not down there either.

[Gabriella]

Participants watch hopelessly as their children slip between the cracks of much needed support. Feeling embattled, these participants continue to promote their own perceptions of their child’s needs. Stigma and suspected disbelief from professionals inherently burdens them throughout the journey:

For really disabled people I think there’s possibly stuff but for someone like him who just looks fully functioning ... it’s hard. Every two years he has to have a letter that says he’s got a disability ... It’s genetic (laughs) ... it’s not going to go away. [Sandra]

Prior to the diagnosis, participants are unsure whether to trust their instincts, and oscillate between judgement and concern about their child’s development. Mostly they experience a sense of relief when the label of 22q11DS is given. The diagnosis provides validation for the parents’ concerns. It relieves some guilt, and initially gives the parents new-found confidence in their intuitive skills:

It was quite an event because we knew what she had... There was an answer as to why. [Gabriella]

However, diagnosis proves a double-edged sword as initial relief is replaced with sorrow that the child has a syndrome that brings life adversity. Hope that would not give way to doubt prior to diagnosis is lost:

It’s nice to have a diagnosis. But then you look at the list of signs and symptoms and you think what sort of diagnosis is that?! [Sandra]

For many, a diagnosis of 22q11DS is unchartered territory where concern and uncertainty, variation in symptomatology, and a medical model that promotes experts must be confronted. Powerlessness, head butting, and incompetence become the new modus operandi as they struggle to have their parenting knowledge validated among medical professionals unfamiliar with the syndrome:
You really felt like you were hitting your head on a brick wall. We’ve been there, done that, it doesn’t work. Doctors don’t like you to tell them that… [Tracy]

6.3.2 Where is ‘I’?

These parents lose themselves in their child as they become the managers of their child’s health, behaviour, and extensive needs. Most are running a family and working. Feeling all-consumed and exhausted, they crave respite that never comes as formal support is lacking:

You’ve just got to keep on looking after your family. You don’t get time out, you don’t get any super answers, or anything like that. [Wendy]

There are few variations on a theme for these participants. Whether it is the exhaustion or the sense that this is unending, they perceive themselves as caught in a lifelong career of care for children who are unlikely to attain independence. Ultimatum are sometimes the only way to coerce assistance in their exhausted state of worry:

We were so tired … we said (to the doctor), “Either you give her the tablets, or we’re going to take them!” [Maria]

Participants openly express fear for their child’s fate once they can no longer provide all the necessary care. For them, it is not a matter of indispensability but of commitment and love. Guilt is never far from their musings on who will care when they are no longer alive. They are unable to imagine others offering the care their child requires:

What’s going to happen? It’s a bloody great concern, great concern…It’s a big ask to say look, if something goes wrong, it’s your job. [Max]

Throughout the journey, the participants’ relationships change. The parents’ self is entangled with their child, and their child’s pain is transferred to them. There is disappointment at the perceived disinterest directed at their child from once valued friends:
Adults wouldn’t take time to talk to her…and that I found…very disappointing. No compassion even in my close friends. [Maria]

Lack of knowledge about the syndrome from professionals is felt and interpreted as a personal insult. Anna perceives judgement from others, which causes a struggle with self-value:

_It kind of makes me feel like…we’re worthless…that’s how it feels, when they don’t know about it - makes me feel like I’m just rubbish._ [Anna]

**6.3.3 Conflicting Loss, Grief, and Guilt**

This theme captures the life participants expected prior to having a child with 22q11DS. They see the potential trajectories of their life, and feel sorrow and grief for what could have been. Although generally happy with the path they have been given, there are cautious reflections on the ‘other’ life, which would have been a simpler and more naïve existence without the extreme challenges they have experienced. Participants struggle against self-pity with each milestone that should pass, when reflecting on the losses and unendingness that 22q11DS brings:

_There’s all those things that change…no wedding, no grandchildren…live with him for the rest of your life…_ [Sandra]

The parents do not only grieve for themselves, they dream of how they could change their child’s fate:

_I think, if you just had these few bits of chromosome, a few bits of DNA, you’d probably be something really extraordinary. And it’s sad._ [Max]

Female participants continue to mourn their desired mothering experience remembering the cloak and dagger around having a sick child. Pain still appears raw as they reflect on the child’s birth, and their inability to celebrate their new motherhood:

_That’s supposed to be a really joyous time…We couldn’t get the baby baptised, and our friends couldn’t see her … we had this phantom baby._ [Maria]
Birth memories of loss remain poignant, even though their child is now an adult. This is a different commitment to the one they imagined. Rather than a journey of positive surprises, they care for a uniquely sick baby, and have the burden of unknowing. Isolated and alone, social connections start to dwindle:

*That’s where the social isolation started ... You join a 1% club, with 1% of people with a sick baby.* [Maria]

Like any adult, the children with 22q11DS have hopes for their future. Participants speak of bringing their child back to reality when dreams exceed the child’s capabilities. While trying to spare their child from the pain of failure, participants are forced to play the ‘bad guy’ and remind the child of their limits. Conflict is common and rather than stretching the umbilical cord of independence, these participants are trapped into reining in their child and crushing dreams:

*She doesn’t understand that she doesn’t actually have the ability... so we've had a lot of tears.* [Wendy]

Participants recognise that they are engaging their child in a charade of independence, adjusting tasks to fit their child’s competence. They seem trapped in the need to give their child the illusion that they are living a normal adult life. Conflict is evident in the participants as they recognise their collusion with ‘normality’ on the one hand, and self-criticism on the other, because they are not being true to their goals of teaching their children to work around their challenges:

*She has got her own business ... but it’s hardly flourishing...She’s running it in our house so whenever she’s got a client we’ve got to be as quiet as a mouse.* [George]

Similarly, the confusion of protection is complicated by the surreptitious guilt that nags them: *did we do something wrong?* Despite knowing their child’s deletions are de novo, the participants are not able to disengage from self-responsibility:

*It’s probably just something that weighs on your conscience ... I’ve often wondered about it ... if we could’ve avoided all this had we known better.* [George]
Child challenges because of their 22q11DS, magnifies parental pain. Guilt turns inward and self-blame is like a volcano simmering under the surface:

*Did we do something wrong ... I just feel like, why couldn’t it be me, why couldn’t I have had all the problems and not my son ... it just makes you feel like a failure.* [Anna]

Participants compensate for their guilt by enabling their child to achieve which seems to be a strategy for reducing their own distress by guarding against situations that could upset the child. As their child grows and seeks more independence, they speak of the struggle to relinquish control and let the child make their own mistakes. Should they take the easy path knowing they will be called on to pick up the pieces when things do not go to plan, or should they stick to the higher goals of learning from mistakes?:

*We’ve probably always tried to take the protective role but now that she’s older it’s not working... but I think really afterwards she’s realised that – well you were right.* [Gabriella]

### 6.3.4 Angry advocacy

Throughout the journey of having a child with 22q11DS, participants are constantly thwarted in their attempts to manage their child’s condition, particularly by the experts treating them. Frustration is never far from the surface ever ready to defend against the professional that excludes them or dismisses their insight:

*This woman came into the house and said, “Get your cardiologist to tell you what he’s not telling you...she’s got VCFS.”* [Maria]

The lack of empathy from professionals coupled with uncertainty surrounding the condition leaves participants feeling powerless. Max feels blocked at each turn and senses the indifference to his questions, creating more questions and concerns:

*There’s a huge range of symptoms...so where are we? Dunno ... What’s the prognosis? Dunno. What causes it? Dunno ... What do we do now? Dunno. What happens in the long term? Well, a few develop schizophrenia ... So how can we tell? You can’t. You don’t know until you get there.* [Max]
Anna finds the obstruction particularly difficult, as it compounds the guilt she feels. She lost time that could have been used to help her son. Anna shifts between exasperation with medical professionals’ secrecy regarding their lack of understanding about 22q11DS, and anger with them for not informing her more:

*That’s what hurts. They knew nothing about the syndrome and they were treating him …

*Why couldn’t they tell us that’s going to happen? [Anna]*

**6.3.5 Pragmatic acceptance**

A diagnosis of 22q11DS often brings more questions than answers. Initially, participants search for answers. They then learn that questioning is futile, and become resigned to the uncertainty that casts a shadow on their life. Participants avoid feeling victimised by asking themselves, ‘why not me?’:

*If there’s 1/100 births that has a heart condition, then why shouldn’t it be us? Someone’s got to make up those numbers, so…it was us! [Wendy]*

Now their children are adults, participants feel a lot of the difficulties (e.g., struggling through school) are behind them, and have made an uneasy peace with the ambiguity of their child’s future. They recognise that they are not in control of the overall journey and seem to consider anxiety as fruitless and a waste of time:

*There’ll be a few more bumps…I’m not too concerned, although I really have no idea what the future brings, so… definitely say a lot of Hail Marys…it’s got me this far, I’m sure it’ll get me through a few more years. [Tracy]*

Re-evaluation of their life values and expectations emerges as a direct reflection of having a child with a disability. Dreams they had for their expected child are now abandoned and there is a momentary collision with disappointment and sadness about missed opportunities:

*That’s what changes the most - is your expectation of life … and that’s disappointing.*

*[Sandra]*
However, their child’s version of success is considered more openly, and delight is embraced for what they have achieved. Accomplishments of any type trigger a fierce pride in these participants as they accept their child’s life path may deviate from the traditional. Value and successes worthy of celebration are adjusted:

*I would not stop talking about our kids ... I wouldn’t cover it up; I wouldn’t hide it.*

[Sandra]

**6.3.6 Finding authenticity**

Participants’ knowledge about the syndrome and themselves appears to have been a dual journey of growth. The long search for answers for themselves has been replaced by a wish to pass on their own knowledge and experience. When opportunities for altruism arise, participants volunteer. They hope they can make the journey easier for others. The participants know how difficult it can be to have an adult child with a developmental disorder, and express frustration when others are perceived as not advocating for equality when they have the potential. In particular, George is angered by this:

*I’ve recognised the difficulties that people with disabilities have in making their way in life ... When I could do stuff I did...because I was the only person who seemed to have any commitment to it.* [George]

The participants are redefining their identity through a recognition that everybody in the family has benefitted from the child with 22q11DS. There is a sense of belonging. Again, participants see the path life they could have taken without a child with 22q11DS. Instead of mourning what could have been, they are grateful they have become the parent they are:

*I’ve had the big transformation. I’m really pleased with that because...looking back...I don’t like the mother I would have been.* [Maria]

Other participants make sense of the diagnosis because they ‘know’ they have innate gifts that enable them to cope with the immense strain of having a child with a
developmental disorder. They philosophically engage with being given what we can bear and belief they were ‘given’ their child because they are strong and positive. Perseverance when times are tough is perceived as an existential gift:

*God’s given the hard troubles to me because he knew I wouldn’t pike out.* [Anna]

### 6.4 Discussion

This study highlighted the unremitting burden experienced by these participants in parenting a child with 22q11DS. Overarching these results is the inevitable intrusion of their child’s extensive needs. Their narratives define the journey as one of *Losing ‘I’* and ultimately *Finding ‘self’*. The subthemes expose the cumulative stressors experienced by these participants that continually change and often become more threatening as their child ages. Physical and mental exhaustion is felt as a constant in their lives. Stigma, in particular, brings isolation to these once independent adults. Progressively they lose their adult friendships, and lack trust in their instinctual judgement as they are doubted and questioned over their child’s presentation to professionals. Though they find advocacy skills to battle hierarchical structures, anger is a part of that armoury that pushes back against layers of obstruction and lack of awareness.

For these participants, eventual diagnosis was a mix of relief and grief as they entered an unchartered pathway with often indifferent professionals and abdicated from the imagined parenting experience they had expected. Each participant spoke of the creative challenge to redefine their lives as they confronted an uncertain future for their adult child with little societal assistance. Finding ‘self’ comes through a pragmatic re-evaluation of theirs and their child’s ‘accomplishments’ that allows an authentic and positive psychological shift in their whole-of-life interpretation. As such, the flexibility
to redefine self brings a conscious engagement with psychological wellbeing mirrored in the growthful domains of empathy, humility, gratitude and pride.

In contrast to previous research highlighting the potential for trauma in parents with ill children (e.g., Franich-Ray et al., 2013), these participants do not directly express being traumatised by their parenting of a child with 22q11DS. However, hypervigilance was noted as cumulative on earlier disappointment and it stretched their creative risk assessments to offset potentially traumatising events. Instead of trauma, participants speak of the burden and frustration of snowballing adversity throughout their child’s life, which is described as feeling stigmatised, shamed and self-doubting. Grief and loss are palpable in these participants as their child misses milestones and is actively excluded. These are similar to the feelings expressed in a qualitative study of mothers who had multiple births, with at least one of the children affected by disability (Bolch, Davis, Umstad, & Fisher, 2012). For example, contrasting their own experiences with mothers who had healthy children brought about significant distress (Bolch et al., 2012).

The current study demonstrates the parents’ disenfranchised grief (Doka, 1989) which has also been reported in parents of children with special needs (Bolch et al., 2012) and people who have received genetic testing results for themselves and their families (Sobel & Cowan, 2003). Participants experienced sorrow and loss throughout their child’s life and were unprepared for the stigmatisation of disability and the aloneness such stigma brought. The grief surrounding their child and related experiences cannot be openly acknowledged or publicly mourned because they are not recognised or validated due to this ever-present stigma (Doka, 1989) and the low awareness of this disability in society. Medical professionals and educators contributed to this; either through ignorance or a dismissive attitude towards parents’ concerns.
These findings are similar to mothers of teenagers with developmental disabilities. Even years later, mothers described the diagnosis period as particularly salient because of the interactions with healthcare professionals who were often unwilling to listen to the mothers’ concerns (Todd & Jones, 2003). This is a clear avenue for intervention, as both medical professionals and educators are well-placed to normalise the reactions to having a child with 22q11DS. Parents who have a child with 22q11DS experience complex and conflicting emotions. Reducing institutional stigma can provide these parents with an avenue to seek support and thus lessen the social isolation described.

These interviews were in many ways alive with shifts in perception and interpretation occurring throughout. There was a sense that their future was evolving before them with uncertainty. As they spoke of that future in which they would no longer be able to care for their child, there was a profound sadness and sense of exhaustion as they planned extensively without societal support for the future care of their adult child. The anticipatory loss and trauma casts a shadow over their everyday lives as it does for many individuals who live their daily life in anticipation of adversity (McCormack, White, & Cuenca, 2016). However, as they reflected on the journey, they began to interpret the stigma, ignorance, loss of relationships, and battles through layers of obstruction as providing the springboard for psychological growth through reconnecting with their empathetic and grateful self, and honouring pride in their journey. Redefining meaning in their experiences brought reconciliation with past belief systems to combat helplessness, frustration, and stigma; comparable to people who received predictive genetic testing for Huntington Disease (Sobel & Cowan, 2003).

The psychological growth experienced by these participants is similar to previous research surrounding positive aspects of having a child with a developmental disability; where parents have reported finding new perspectives on life, increased
sensitivity, opportunities to learn, improved family dynamics, and increased confidence and assertiveness (Hastings & Taunt, 2002). For example, parents with a child affected by autism or Down syndrome have been found to positively adapt by examining and adapting their views surrounding their child and their parenting role, even though grief is still real for the dreams that are no longer attainable (King et al., 2006). Viewing these positive perceptions (or ‘psychological growth out of adversity’ as we have conceptualised it) as a coping resource is valuable in terms of helping parents to adapt (Hastings & Taunt, 2002). As evidenced by the current study, conflicting interpretations can be experienced simultaneously (e.g., loss and gain).

Time can be an important factor in adjusting positively to parenting a child with a disability (Krauss & Seltzer, 1993). For example, although not defined as psychological growth, early studies also found that older mothers of developmentally disabled adults had similar or better outcomes on measures of depression, life satisfaction, parenting stress, and social isolation compared to a) older women not in a caregiving role, b) women who were caregivers for older adults and c) young mothers who had a child with an intellectual disability (Krauss & Seltzer, 1993). Taylor and Seltzer (2011) also found that the mother-child relationship improved while children with autism were in high school. The fact that the participants in the current study all had adult children may have contributed to the more positive outcomes these parents experienced, such as recognising their innate gifts as carers. As parents, they have experienced grief, loss, and guilt throughout their child’s life; all of which may have caused self-reflection and thus psychological growth. Future research should aim to delineate which factors are best predictive of these positive outcomes, and how best they can be promoted and supported in families affected by disability.
6.4.1 Limitations

One potential limitation is that two sets of couples were included in the sample. The close relationship may have influenced their interpretations of their experiences, meaning that they could have provided relatively similar data. For example, George and Gabriella shared similar frustrations and joys. The differences were greater than the similarities for Max and Maria. Max openly stated he believed his wife had found positive meaning whereas he could only see the negative impact of their daughter’s 22q11DS diagnosis. However, as a qualitative study this research does not seek to generalise, nor seek cause and effect. Double hermeneutics played an important role in the analytic process of this unique phenomenon, therefore the researchers’ experiences and biases may have impacted both positively and negatively on their interpretations. In an attempt to enhance the study’s credibility, the auditors vigorously adhered to an ongoing audit trail inclusive of multiple robust discussions. Although the sample may not be representative of all parents who have an adult child with 22q11DS as there are many factors impacting these relationships, the use of IPA has sought to provide hypotheses for future and larger research through an in-depth exploration of a homogenous group’s subjective interpretations that have experienced parenting a child with 22q11DS. Therefore, the findings contribute to the body of knowledge surrounding 22q11DS by highlighting both positive and negative interpreted impacts on these parents, particularly as their children moved into adulthood.

6.4.2 Conclusions

This study draws attention to the many struggles faced by parents of an adult child with 22q11DS and the potential for psychological growth, especially their ability to redefine ‘self’ and embrace an authentic way of living with 22q11DS. Importantly, the coexistence of distress and psychological growth is a major consideration for
therapists, health care workers, and support personnel to work in more creative ways with families caring for adult children with disabilities. Growthful domains of empathy, humility, gratitude and pride are likely triggers for psychological wellbeing despite ongoing distress and loss. Of interest is that the challenges from health care services and feelings of guilt appeared to trigger growth in these participants. This provides a basis for further qualitative and quantitative research and for aims of therapeutic support.

It is important that medical professionals acknowledge any gaps in their own knowledge when presented with unusual medical phenomenon. In doing so, opportunities for open dialogue can occur assisting rapport for supporting parents with realistic expectations. Supporting parents on a life-long journey of care is a relational challenge for many medical personnel. Though positive and negative outcomes are part of the journey with 22q11DS, the attitude of the health professional can impact parents’ reactions to their child’s disability. Family relations, parental age, years since diagnosis, and expectations on siblings are considerations for future research, support, care policies and programs, and factors for consideration in larger quantitative research.

Clinicians can access recently published guidelines for managing adults with 22q11DS (Fung et al., 2015) guiding them to provide appropriate care for the person affected by 22q11DS, and for managing and validating parents’ concerns for their child. Following the guidelines is imperative during the affected person’s adolescence and adulthood, where complications such as schizophrenia can arise (Murphy et al., 1999). Medical professionals also have the opportunity to offset stigma, blame, and self-doubt, the common legacies of disability for many families. There is guidance available for clinicians that can aid in this process, with advice on communicating effectively with parents and families from diagnosis and throughout their journey (see Kisler & McConachie, 2010). This can encourage new and diverse interpretations of their
complex experiences, and subsequently support psychological growth out of the adversity of 22q11DS.
Chapter 7

Critical reflections of conducting interpretative phenomenological research

7.0 Chapter Statement

Following the nomothetic enquiry into predictors of psychological growth in parents with a child with a developmental disability, the phenomenological investigation of this thesis sought the impact of a child with 22q11DS on parents’ individual lives. Description, clarification, and examination of meaning-making from such complex experiences was sought. The aim was not to categorise the participants’ responses from a positivist, medical model perspective. Rather, we sought the subjective “lived” experiences of these particular individuals. In doing this type of research, it is necessary to consider the role the researcher plays in the collection, selection, and interpretation of data. That is, it is important to have reflexive practice. Reflexivity requires a thoughtful, self-aware analysis of the intersubjective dynamics of the research process; based on the researcher’s biases and social positioning (Finlay, 2002). Throughout this chapter I reflect on my personal experiences with IPA. McCormack (2010) created the PHENOMENA guidelines for conducting qualitative research which are useful for reflexive practice. I use this tool as a basis for my reflections. That is, issues related to Project size; Honest rationale for using IPA; Equity and relational challenges; Neutrality; Obsolete interviews; Monitoring biases; Environmental challenges; Non-reciprocal person-centred stance; and Anxiety control are discussed. I note examples from my interviews and reflect on personal biases that may have affected my interpretations.
7.1 Project size and time constraints

The project size and time constraints were not particularly significant for conducting this thesis project. From a phenomenological perspective, I was aiming to capture the individual’s interpretations at the time of the interview, rather than an overview of all experiences (Smith et al., 2009). Therefore, a single session was appropriate, as participants’ interpretations could change between multiple sessions (or indeed as a result of the interview). Certain techniques also helped to focus on exploring the phenomenon at hand. For example, I received demographic information about the participant and their child prior to the interview, which helped me to avoid the trap of asking questions to clarify my own understanding about irrelevant details (e.g., “how old was your child at this stage?”). Also, each participant received the interview topics the day before, which meant they had already had the opportunity to consider the interview questions.

The project time constraints did mean that the data for both participant groups (i.e. parents of children with a) young children with 22q11DS and b) adult children with 22q11DS) was collected simultaneously. The data from parents with adult children was then analysed before the data from parents with young children. Upon reflection, I may have had conscious and unconscious rumination about the interviews, the participants, and initial perceptions about potential themes that impacted my interview questions and analysis. However, it was necessary to be pragmatic and complete the studies in this fashion. I am not aware of any specific instances where my previous knowledge impacted my interpretations, but continuous reflection and referral to the recordings and transcripts ensured the themes stayed true to the data, regardless of any similarities and differences between participants. In fact, the previous analysis was beneficial in that I
felt more at ease with the process of IPA and had some insight into the parents’ emotional journey, even though it was at a different stage of their child’s life.

### 7.2 Honest assessment of personal rationale for choosing IPA

As previously mentioned (see Chapter 4), qualitative researchers must clearly separate out the reason for asking questions through a qualitative paradigm as opposed to a positivist paradigm. I struggled to justify my reasoning at first because the philosophical underpinnings for IPA seemed innate: it was obvious to me that this is the way psychological research should be approached. That is, I assumed that phenomena such as the experience of having a child with a developmental disability should be investigated from the participant’s perspective instead of having a researcher project a hypothesis onto the phenomenon (Joseph et al., 2009). It was also my understanding that the researcher is always interpreting the participants’ accounts through their own worldview (i.e., the double hermeneutic), and that one’s experience cannot be separated from the interpretation of the same. Perhaps my own education, guided by many philosophically learned people, had blinded me to the norm of nomothetic investigation in psychological research. Delving into the literature helped me to verbalise my critical realist philosophical stance, and identify my natural leaning towards phenomenology. This became particularly apparent when reflecting on positivist assumptions that are not necessarily suited to the topic of this thesis (e.g., the need for objectivity; Bracken, 2002). This again emphasises the importance of a good philosophical understanding of the research methods employed. Further, the need for continuous reflection of the rationale for IPA is necessary to stay true to its philosophy and practice.
7.3 Equity and relational challenges

I did not have prior relationships with any of the participants, which meant that previous power imbalances were not part of the intrapersonal space between researcher and research during the data collection. However, as a social critique, the roles of “researcher” and “participant” can denote an unequal relationship. I did not have any particular relational challenges, yet there were occasions of shifting researcher–participant positions. For example, Anna oscillated between being comfortable with her expertise and deferring to me as the “expert”:

You have to go through those experiences. I’m really very upset with doctors who learn off the books. They have no idea! ... I don’t know what happens with uni, book work, assignments, whatever? What do you do? [Anna]

In an attempt to deconstruct my authority, I emphasised the participants’ rights and made it clear that I was there to learn from them. The recruitment design (i.e., potential participants were required to initiate contact with the researchers) may have also accounted for the lack of overt inequality, as it could have attracted people who were less inclined to be intimidated by potential power imbalances.

7.4 Neutrality and unexpected dynamics

There may have been a mismatch of researcher and participant expectations regarding the interview. For example, participants could anticipate a counselling session rather than a research interview. To avoid this, I took the time to explain my position as a research psychologist and not as a clinical psychologist. To further assist in their understanding of my role, I explained that I could refer them to a number of services if required, but I was not qualified to provide counselling or advice. These
ground rules I believe, helped to keep the interview focused on the research questions, limiting any expectations of counselling or ‘expert’ to be pleased.

7.5 Obsolete interviews

In qualitative research, there may be interviews that do not provide the richness required for analysis; either due to the interviewer’s techniques or lack of response from the participant. As such, the interview offers little insight into the phenomenon under investigation and may be removed from analysis. I was fortunate enough to have participants who enthusiastically engaged in the interpretative process and offered rich insights into their sense-making with little prompting. As a result, I included all conducted interviews in analysis. One interview did offer analytic challenges (Eric) that I initially believed would provide neither convergent nor divergent thematic content. At first, it seemed as though his child and related experiences had not created the need for reflection and did not rate as particularly significant for him. Excerpts from his interview highlight the difficulty I had drawing out thoughtful responses from him:

Jane: Was there anything you found challenging in terms of support?
Eric: Not really.

…

Jane: Do you think you’ve changed as a result of [your child] having 22q11DS?
Eric: Not really. They say everything happens for a reason but not really.

In this case, guidance from a supervisor experienced in IPA (LM) was invaluable in drawing my attention to the richer aspects of Eric’s interview (instead of focusing only on what was lacking), resulting in interpretations of avoidance and feeling “in limbo”. For example, the quote below shows that Eric is expecting future
difficulties, but due to the nature of 22q11DS they are impossible to predict while his son is young. Therefore, he avoids thinking about what may be:

*It hasn’t really affected our lives at this stage. We know that there will be some challenges moving forward as he develops, but at this stage it hasn’t really changed our lives at all.* [Eric]

Eric’s interview taught me the importance of robust discussion with more experienced researchers, as well as exploration of covert meanings in a participant’s discourse. What one participant does not say can be just as revealing as another’s rich self-interpretation. In this case, Eric’s resistance to respond exposed his avoidance but also his pragmatism about what was to come but not impacting currently. Without guidance it would have been easy to dismiss Eric’s interview.

### 7.6 Monitoring personal biases and pre-suppositions

A salient issue for me throughout each stage of the qualitative enquiry was monitoring my personal thoughts. Without this conscious effort, my biases could easily manifest as poor and/or judgemental interviewing techniques, or blind me throughout the analysis. When conducting IPA, the researcher must attempt to “bracket” their biases in order to avoid leading the participant based on their own agenda. Below I have outlined some of my own biases that may or may not have impacted on my interviewing technique and analysis.

I conducted the interviews at an interesting time in terms of disability support in Australia. The introduction of a new government’s policies meant that funding support in general was reduced for people affected by disability, and payments were proposed for previously free healthcare visits. Unsurprisingly, these proposed changes invoked fear and suspicion in many of those who would be affected by it. As the participants’
accounts of their child were virtually inextricable from their healthcare experiences, these issues arose in several interviews, particularly in parents who had an adult child with 22q11DS. This required me to constantly monitor my own emotions, as this was also a distressing time for me. It was difficult to remain neutral and not get caught up in my own thoughts, or become involved in a worried discussion about politics and the state of Australian healthcare. For example, when Sandra commented on these issues and the fact her son had to re-prove his disability (i.e., “every two years he has to have a letter that says he’s got a disability ... It’s genetic ...it’s not going to go away”), I was furious as it echoed my experiences. Around the time of Sandra’s interview, I had been involved with a meeting regarding government support for my sister, where she was required to somehow prove her disability was actually disabling. I had to be mindful in order to remain present and focus on the participant’s stories instead of my own. Even though this may have affected the interviews, my knowledge and experiences were also valuable. For example, my life role as a carer meant it felt natural to empathise with each of the participants and be on their side, as is necessary in IPA (Smith et al., 2009). According to Maslow (1966, p. 45), “there is no substitute for experience, none at all”, which in this context means that my own experiences gave me the insight and ability to see the humanness in the participants’ accounts, rather than simply a psychologically theoretical understanding.

When it came to analysis, it was difficult at times to reflect deeply on the participants’ accounts. By delving into their thoughts, I had to confront my own experiences and biases I did not realise I had during the interviews. A potential bias that was particularly apparent during analysis was my perception of traumatic distress in the participants who had an adult child with 22q11DS. Although not something I had (at least consciously) considered, one of my earliest interviews was with Max, whose quote
still sticks with me: “it was as traumatic as hell... It was awful.” This led me to think in terms of trauma symptoms in other participants’ accounts; that is, intrusio
e(e.g., “The words, the number 22 just kept going round and round my head every night.
Honestly. It was like 22, 22...” [Frances]); avoidance (e.g., “we had all the information but never delved into it” [Eric]); and hyperarousal (e.g., “She cancelled my appointment... There is no care... Why the fuck aren’t they helping me?!” [Stephanie]).

However, because I assumed traumatic distress was apparent (and perhaps had some discomfort referring to someone’s child and related experiences as “trauma”), I didn’t explicitly explore it in the interviews. This meant that the data did not have enough evidence to justify the inclusion of traumatic distress as a theme in the manuscript. However, the benefit of IPA is that topics that arise outside the scope of the current study can be noted and explored later. A review of the literature revealed that trauma symptoms are indeed possible in parents who have a child with a chronic illness (see Section 1.1.4), yet there is little literature related to specific disabilities such as 22q11DS. Reflection on biases such as this can identify gaps in research and thus be beneficial. I was also particularly cognisant of my interest in psychological growth throughout the interviews and analysis. Whilst I prompted participants to reflect on positive change in their lives, I was careful to encourage a balanced account by exploring negative changes too. I was very aware that whilst I had an academic interest in psychological growth, it may not exist in the participants I interviewed (or at all in this population). The use of independent analysis, followed by robust discussion with a supervisor experienced in IPA (LM) ensured I did not force the data into my preconceived ideas. I am currently involved in a survey examining trauma and psychological growth in parents who have a child with a developmental disability. Again, this is an area where my biases may have proved fruitful. Biases (i.e., previous
experiences and interests) are not always a limitation if there is good reflexive practice in place.

7.7 Environmental challenges

Being in Australia poses unique challenges for conducting qualitative research. The size of the country coupled with a relatively sparse population means that face-to-face interviews are not always possible. Although face-to-face interviews are considered ideal for promoting rapport (Smith et al., 2009), pragmatism had to prevail: if I was required to travel more than 8 hours to conduct an interview I did it by telephone instead, resulting in 5 telephone interviews. However, two of these participants were in relatively easy reach (i.e., less than 2 hours’ drive each way) but chose to be interviewed on the telephone for convenience. Giving participants the choice of location I believe gave them a sense of control whilst sharing potentially distressing experiences. Although as the researcher I encountered some travelling difficulties with the participant’s chosen option for interview, the priority was always to make the participant feel safe.

Another environmental challenge I encountered related to interruptions. For example, when conducting an interview at Deborah’s house, she had two children at home, one of whom very much wanted to be involved in the interview. He repeated her answers, spoke over her, and made lots of noise. As a mother, she was adept at carrying on despite the disruptions, but it was challenging for me to stay focused on the interview without rudely ignoring the young boy who so desperately wanted some attention. It created difficulties with transcription too, requiring much listening and re-listening to decipher the participant’s account over the child’s voice. Despite this issue, the challenging environment also provided an opportunity. Deborah’s husband (David)
came home during the interview and sat in for a while, before eventually contributing himself. He provided rich, insightful data which complemented his wife’s story; and was happy to sign a consent form to be included in the final analysis. As a couple, they were at ease sharing their experiences; and I believe more was gained from the interview having them together compared to interviewing Deborah by herself separately. Further, David’s presence had a calming influence on their child, who reduced his interruptions.

7.8 Non-reciprocal person-centred stance

The semi-structured interview is an interesting interaction between the researcher and participant. Each are entering into the “relationship” for different types of profit. The interviewer collects data; the interviewee has a chance to be heard and validated. The researcher must be flexible and avoid projecting their biases or judgement onto the situation (see Section 7.6). They need to remember the purpose of the double hermeneutic, and try to make sense of how the participant makes sense of their experiences. Personally, avoiding overt judgement was less of a challenge for me than maintaining control of my biases. As a young researcher with no children, it was easy to be on the participant’s side throughout the interview, as I could not imagine how I would react in their position. In fact, most of the participants in the studies of this thesis held the power in the interviews, as the expert in their own lives. Again, maintaining good reflexive practice was necessary to avoid causing harm to the participants; all of whom were vulnerable as they shared their emotional stories. Similarly, rigour and credibility required that robust discussion continued with my supervisors throughout the interpretation and write-up of this thesis to minimise contamination of biases.
7.9 Anxiety control and hidden agenda

The anxiety of the researcher is another area for reflection. Conducting an interview on a sensitive topic means the researcher must be ethically rigorous, empathetic, sensitive, and encouraging; while maintaining control of their biases and the research protocol. This is a lot to remember for a novice researcher. Researchers can be too rigid in their approach, sticking to a “script” instead of following the participant’s lead. Rigidity was not a major issue for me; rather, my concern about doing things wrong made me too flexible. As such, I tended to be hesitant about interrupting and guiding the interview which resulted in transcripts that were heavy on narrative and therefore long to gain the rich interpretation. However, experience helped reduce this anxiety and after the first few nervous interviews I was able to relax and enjoy the process. I also learned as I progressed through my interviews that the stage participants were at with processing their child’s developmental disability was almost as important as my interviewing ability.
8.0 Chapter Statement

This chapter offers a brief summary of the quantitative and qualitative results of this thesis by discussing the thesis questions: in what ways is the experience of parenting a child with a developmental disability different at different stages of life; how do parents interpret these experiences; and how do they perceive that phenomenon as impacting sense-making and their own psychological wellbeing? The chapter (and thesis) concludes with recommendations for future research and practice.

8.1 Synthesis of findings

8.1.1 In what ways is the experience of parenting a child with a developmental disability different at different stages of life?

The experience of parenting a child with a developmental disability changes with the child’s age and stage. The quantitative analysis in this thesis revealed that the child’s age was predictive of psychological growth. For a one unit increase in the child’s age, there was an increase in psychological growth by .186 points on average, controlling for all the other predictors. That is, parents who had an older child were more likely to experience this positive psychological outcome. Greater exposure to a stressor is associated with benefit-finding (Helgeson et al., 2006). Previous research has demonstrated that the child’s age may be a factor in parental positive psychological outcomes. For example, child-related enjoyment is associated with the child’s age in families affected by ASD (Ekas et al., 2009). These results make sense within the context of meaning-based coping. Parents who have an older child are likely to have
had prolonged and significant exposure to stressors, forcing them to seek meaning in order to gain relief (Folkman, 1997).

Indeed, these findings were supported by the qualitative data of this thesis. Although there were similarities in the accounts of participants who a) had a young child with 22q11DS, and b) had an adult child with 22q11DS (e.g., both had experienced some form of psychological growth), there were notable differences which are potentially due to the child’s age. Parents who had a young child described their constant fear. They discussed the agony of uncertainty; experienced through grief, guilt, and apprehension about the future. It is not surprising that parents who had a young child were so uncertain, as 22q11DS is a highly variable disability (Shprintzen, 2008).

In contrast, parents who had an adult child with 22q11DS did not comment on this uncertainty. Although this group are likely to experience some uncertainty (particularly surrounding the onset of psychosis), the significant issues for them related to grief surrounding milestones that their child will probably miss (e.g., marriage).

8.1.2 How do parents interpret these experiences?

The qualitative component of thesis highlighted the mix of reactions parents displayed in response to having a child with a developmental disability. Seemingly conflicting emotions (e.g., shame and pride) existed simultaneously, and it was a constant “tug of war” between the positives and negatives of the experience. The stressors associated with this type of caring role meant that parents were psychologically vulnerable; yet they actively aimed to positively reframe their experiences and find meaning. A clear example of contradictory emotions came from the parents who were now parenting an adult child with 22q11DS. They reflected that when they received the diagnosis, they felt grief and relief at the same time. They remembered that they grieved for the child they had hoped for, yet were grateful that
there was an explanation for all their child’s issues. Those who were parenting a young child with 22q11DS were differently conflicted. They described feeling psychologically fragile, yet demonstrated hopeful resilience in purposefully reframing their struggles in a positive light.

8.1.3 How do parents perceive the phenomenon as impacting sense-making and their own psychological wellbeing?

The uncertainty described by participants in all studies of this thesis is an area that appears to have impacted positive psychological outcomes. Parents who had a preschool child with 22q11DS feared for the future because 22q11DS has such a varied trajectory, and it is not possible to predict the child’s prognosis at a young age. These parents constantly anticipated trauma (van der Kolk et al., 1996). That is, they were hypervigilant to any symptoms their child displayed. Mischel’s (1990) Reconceptualized Uncertainty in Illness Theory is helpful in interpreting these parents’ experiences. Chronically ill people and their families may struggle to find meaning in illness-related events. However, if they accommodated the new information into their schematic framework positively, they were able to move forward gaining relief from constantly anticipating the distress of “not knowing”. Parents who had a young child with 22q11DS showed the beginnings of this positive accommodation (i.e., psychological growth), by actively working to view their child and the associated challenges as an opportunity rather than a burden. The positive integration of uncertainty was also present in parents who had an adult child with 22q11DS. Even though these parents worried for the future, they described positive change in themselves, including empathy and gratitude. Further, the quantitative study provided some interesting insight into the role uncertainty plays in psychological growth. The item “I feel I am in control of my life” was endorsed least by participants. Despite this,
many of the participants were shown to have experienced psychological growth according to Joseph’s (2011) PWB-PTCQ scoring guidelines. Therefore, the uncertainty of having a child with a developmental disability (e.g., 22q11DS) can provide a platform for positive psychological change.

8.2 Insights from the mixed method design

The mixed method design of this thesis has provided a comprehensive means of researching the phenomenon of parenting a child with a developmental disability. As positive psychological outcomes in this population are under-researched, a mixed methods design was valuable for exploration from the general to the specific. Purely quantitative or qualitative methods would have been insufficient for understanding the full experience of parenting a child with 22q11DS due to the complexities of this unexplored phenomena. The quantitative aspect provided generalisability to the experience of psychological growth. An online survey with questions about coping, social support, perceptions of family-centred services, and psychological growth was utilised. Four hundred and thirty-two parents of children with developmental disabilities participated and many experienced psychological growth as predicted by greater: a) use of positive reappraisal coping; b) perceived coordinated and comprehensive health services; and c) child’s age. A bigger discrepancy between ideal and actual practical social support, and more use of escape avoidance coping predicted less psychological growth. Therefore, it was concluded that psychological growth is a realistic outcome for parents who have a child with a developmental disability. The large sample size meant that we were confident in these findings.

The qualitative studies also supported the findings of psychological growth in parents who had a child with a developmental disability. This was an encouraging
result, as qualitative research allowed for the participants to honestly reflect on their experiences. Healthcare professionals’ perspectives have tended to drive research surrounding disability and parents’ experiences. The qualitative studies have therefore provided rich insight through semi-structured interviews where the focus remains on the interpretation of the participant rather than being researcher driven through hypotheses. The participant was permitted to direct the research towards the pertinent issues for them regarding this phenomenon, and the researcher using a double hermeneutic approach strove to make sense of the participant making sense of their unique experiences.

Although the quantitative study captures the factors promoting psychological growth well, it did not capture the rollercoaster of conflicting emotions associated with parenting a child with a developmental disability that participants in the qualitative studies described. Nor did it highlight the nuances of psychological growth as a process rather than an outcome, because it was limited by the researchers’ hypotheses (as well as constraints such as questionnaire length). The qualitative studies were informed by IPA and provided a detailed exploration of the experience of parenting a) a young child with 22q11DS, and b) an adult child with 22q11DS. Chapter 5 reported on six parents who had a young child. Despite the negativity that is encapsulated in the four themes of constant uncertainty, guilt, and fear for the future; one theme depicted their experience of hope. Thus, distress, and possibly traumatic distress, actively journeyed with them on a daily basis. Co-existing with this distress, they engaged with their experiences to actively reframe fear and negativity, allowing unexpected psychological growth from their challenges. Therefore, in the early parenting years, these parents interpreted their struggle with the uncertainty of their child’s prognosis as the catalyst to positively reframe their journey.
The second qualitative study of this thesis related to eight parents who had an adult child with 22q11DS. The overarching theme Losing ‘I’ Finding ‘self’ related to six subordinate themes. The participants described distress; experienced through loss, grief, and guilt. Stigma undermined their instincts, and they angrily fought against the layers of obstruction to advocate for their child. However, they came to appreciate their child’s version of success, and in turn, consciously valued authenticity such as empathy, gratitude, and pride. These participants had experienced decades of distress which resulted in different interpretations compared to parents who had younger children. Participants who had younger children were hopeful for the future, whereas parents who had adult children had become hardened and relied on their own strength to move forward positively.

8.3 Thesis contributions

Both quantitative and qualitative findings are major contributions to the literature on parenting a child with a developmental disability, as previous research has tended to focus exclusively on psychopathology. There is now an understanding of the mix of emotions at different stages of the parenting journey, and different expectations and interpretations that impact on the predictors of psychological growth and as such, growth domains that emerge. Parents who have a child with a developmental disability have anecdotally reported experiencing psychological growth (as outlined in the quantitative study) in the past, such as finding new perspectives on life, increased sensitivity, opportunities to learn, improved family dynamics, and increased confidence and assertiveness (Hastings & Taunt, 2002). The mix of emotions highlighted in the qualitative work of this thesis are similar to previous research, where parents with a child affected by autism or Down syndrome felt major grief but also positively changed.
their views surrounding their child and their parenting role (King et al., 2006). The positive psychological change experienced by the participants in this thesis support Folkman’s (1997) theory of meaning-based coping. The negative psychological states associated with significant stress motivated the parents (both consciously and unconsciously) to create positive psychological states in order to gain relief. This is also in keeping with Mischel’s (1990) Reconceptualized Uncertainty in Illness Theory, which proposes that people integrate continuous uncertainty into their lives by reorganising their beliefs. This avoids a chronic anticipatory state of distress associated with expectation of predictability (Mishel & Clayton, 2008). Parents who had an older child may have had the time and experience to integrate the uncertainty of 22q11DS into their lives.

Importantly, this thesis adds to the literature by providing insight into positive psychological outcomes in parents despite any distress associated with having a child with a developmental disability. Positive psychological outcomes as well as psychopathology, are an important area of investigation in parents/families who often spend many decades caring for their children when independent milestones are not able to be attained. By identifying factors that contribute to psychological growth, an area that was previously unexplored in this population, those working to support parents can help to facilitate realistic expectations to positively reframe many aspects of their lifelong journey with disability. Clear definitions of the positive psychological constructs we assessed were provided as related to theoretical models, including use of a standardised measure for the quantitative aspect of the thesis. This thesis has provided a greater understanding of the journey parents face with 22q11DS, which has received little research attention. The parents’ feelings about their child’s condition changed over time, particularly as related to positive psychological outcomes. Further, the research of
this thesis highlights that both positive and negative psychological outcomes can exist simultaneously, emphasising the need for a holistic approach in research and practice. Recommendations for future directions are outlined in section 8.4.

8.4 Implications for future research

8.4.1 Definition of concepts

This thesis highlights areas for further investigation and intervention in terms of positive psychological outcomes in parents who have a child with the developmental disability 22q11DS in particular. Most importantly, it demonstrates that positive psychological outcomes are realistic for parents who care for a child with a developmental disability and should no longer be neglected in research and practice, irrespective of the type of disability.

When continuing to research these phenomena, investigators must reflect on the related philosophical and theoretical underpinnings that guide their research. When researchers are explicit about their methodological underpinnings, qualitative and quantitative methods can complement one another and offer alternative viewpoints particularly within a medical model. As such, qualitative methods ask questions that provide alternative ways to understand subjective distress that avoids the medicalisation of that distress. Researchers must also be clear regarding the type of positive psychological construct/s they are exploring. Much confusion arises from lack of clear definitions and the tendency to view distress through an illness lens. Furthermore, clear paradigmatic stance will influence how the researcher constructs their enquiry; that is, a) absence of negative psychological outcomes (e.g., depressive symptoms); or b) presence of positive psychological outcomes (e.g., posttraumatic growth)? Each are worthy topics of investigation, but are potentially independent constructs (Hastings &
Taunt, 2002); though more recent evidence suggests a curvilinear relationship between growth and distress (Joseph, Murphy, & Regel, 2012).

The use of measurements also reflects a nomothetic approach to the construct being examined and often within the framework of disability condones an illness perspective. Depending on the researcher’s question, they have their place, but the challenge is to constantly and critically challenge our intent and whose perspective we are seeking; particularly when doing a mixed method approach. However, each can richly inform the other. For example, the Mental Health Scale assesses psychological distress (or lack of), yet mental health is not merely the absence of disease or infirmity (World Health Organization, 1946). Justification of the measures used will help to differentiate between the psychological constructs under examination. Reliable scales may need to be developed in order to adequately assess the positive psychological construct under investigation. Thus, it is important to aim for clarity in the literature surrounding positive psychological outcomes.

8.4.2 Research directions

More specific to developmental disability, future research should examine both positive and negative psychological outcomes, and aim to untangle the relationship between these coexisting (yet seemingly conflicting) interpretations parents described. At present, distress and growth may be independent constructs (Hastings & Taunt, 2002), or have a curvilinear relationship (Helgeson et al., 2006). Positive psychological outcomes may also be affected by the syndrome’s phenotype and/or the child’s age, which are factors to be considered in further investigation. Parents who have an older child with a developmental disability experienced more psychological growth, and parents who had an adult child with 22q11DS did not seem as preoccupied with uncertainty compared to parents who had a young child with 22q11DS. This may be due
to the fact there is simply less uncertainty regarding prognosis as the child ages (see Fung et al., 2015); however, Mischel’s (1990) theory also implies that parents who have an older child have managed to accommodate the uncertainty within their worldview because they have had greater experience with it. It is unclear whether it is the child’s developmental stage per say that causes these differences. Rather, it may mean that more exposure to the “stressor” improves adaptation (Lazarus & Folkman, 1984), which coincides with the child’s age. Longitudinal research incorporating the middle period of childhood (and transitions) is required to determine if there are critical periods in the child’s life that promote positive integration of uncertainty and lead to psychological growth in parents.

Another avenue for further research is how much distress (if any) is necessary to promote positive psychological outcomes in this group of parents. It is known that struggles with adversity can be a catalyst for positive change (Joseph & Linley, 2008), however little is known about which particular challenges may promote positive psychological outcomes in parents who have a child with a developmental disability, and how these interact with personal factors. Simply having a child with a developmental disability places parents at risk of experiencing distressing and/or traumatic events (e.g., witnessing the child endure painful medical procedures). Although there is evidence that trauma symptoms are possible in parents who have an unwell child (Franich-Ray et al., 2013), neither primary nor vicarious trauma was specifically examined in this thesis. Future research should investigate whether the psychological growth experienced by the participants of this thesis was a result of trauma that shattered their worldview due to the child’s developmental disability (i.e., posttraumatic growth), or a result of the stressors associated with parenting in general (stress-related growth).
A comparative study with parents who have a typically developing child could help determine if the difficulties associated with caring for a child with a developmental disability promotes psychological growth above and beyond the challenges related to parenting in general. Therefore, stressors and/or trauma and their interaction with psychological growth require more investigation. Too much or too little trauma can negate positive change (Colville & Cream, 2009). Untangling the seemingly conflicting (yet co-existing emotions) would be helpful in understanding the complexities of parenting a child with a developmental disability.

### 8.4.3 Healthcare services and interventions

The parents’ experiences of their child were virtually inextricable from their interactions with healthcare services. Participants reported breakdowns in communication and care when in contact with health services. Research is required with relevant stakeholders (including healthcare professionals) to examine the contributing factors. Healthcare professionals may not receive adequate education in communicating with families, meaning that training in empathetic practice may be required. Or they may be too overburdened to provide the necessary care, which has important implications for resource provision. The close interaction between families and health services means that healthcare professionals are well-placed to promote parental wellbeing and positive psychological change and thus research is needed to see how this can best be done.

This thesis has identified multiple avenues for targeted interventions. This could be as simple as turning the qualitative results into an easy-read book for parents who receive a diagnosis of 22q11DS for their child, outlining the experiences of parents who have been in the same position. Both negative and positive experiences could be incorporated to give parents a balanced view of what the experience will be like for
them. Although there is uncertainty about the syndrome’s trajectory when the child is young, being exposed to and becoming familiar with the emotional journey may help to alleviate some uncertainty. Knowing other parents have experienced similar emotions and managed despite their distress could reduce the feelings of being alone that were described by participants. In the initial stages, measures of negative and positive psychological outcomes could be compared between those who do and do not receive the book.

The time around diagnosis can be particularly distressing for parents (Goodwin et al., 2015) and as such is an opportune moment to promote the beginnings of psychological growth. Another area for intervention is healthcare services, as they are clearly very important in parents’ perceptions. Thus, it is necessary to examine how strategies could be developed to promote family-centred services (particularly coordinated and comprehensive care) that are person-centred and listen to the individual’s perspective. For example, it may be helpful to have a dedicated nurse experienced in 22q11DS to coordinate the child’s care, rather than leaving potentially burnt-out parents to navigate the complex healthcare systems. The acceptability and feasibility of such an approach is another area for future research.

8.5 Recommendations for parents and professionals

8.5.1 Parents as partners in child’s care

It is recommended that healthcare professionals collaborate with families regarding management of the child’s condition. Parents’ instincts were often correct about their child’s health and development, particularly with those who had 22q11DS. In fact, many children had a delayed diagnosis because of the time parents had to spend pushing indifferent professionals for testing. Although the apparent hypervigilance to
symptoms may be frustrating for healthcare experts, it is necessary to empathise with parents and take their concerns seriously. Information provision may also help with parents’ anxious uncertainty. For 22q11DS, the practical guidelines for management of the syndrome (Bassett et al., 2011; Fung et al., 2015) can be used in partnership with parents to provide education on what they might expect at different ages and stages. This can facilitate appropriate interactions between parent and professional, and serve as a gentle tool to guide the conversation to the most relevant issues for the child at that specific time. This kind of understanding about the syndrome will give parents a sense of control as related to their child’s syndrome and its management. Therefore, an initial step in offsetting stigma and validating parents’ concerns is respecting parents and treating them as equals in their child’s care.

Related to this, parents can take comfort in the fact that their instincts about their child are often accurate. This should give them the confidence to persevere with major concerns and ensure their child receives the appropriate support, despite the layers of obstruction in educational and healthcare settings. Families who have a child with a developmental disability need respectful support; otherwise, as the studies of this thesis highlighted, the risk of burnout brought on by constant caregiving and advocacy inhibits parental self-care. Although it is not always possible to have structured respite, parents can take advantage of support groups, disability services, and available social supports. Becoming part of these communities (e.g., support groups) has additional benefits, such as reducing feelings of isolation.

8.5.2 A strength-based approach

Despite its limitations, this thesis provides areas for healthcare and educational professionals to reflect on in their practice particularly their perspective on expert versus therapeutic partnerships. It is necessary for clinicians to consider parents as an
important factor in their child’s health. They must provide information for the family that encourages realistic expectations, both positive and negative (Kisler & McConachie, 2010). In fact, this study supports a collaborative, strength-based approach as likely to be more beneficial for parents’ wellbeing than the traditional emphasis on pathology (Steiner, 2011). As such, professionals should avoid concentrating on pathology and weaknesses only. Strengths can also be acknowledged during appointments and assessments, and may have a positive effect on parents’ wellbeing. Simple changes to practice such as encouraging basic counselling skills; that is, active listening, good communication, and empathy, so that parents firstly feel ‘heard’ and then secondly, referring parents to relevant information about the syndrome and appropriate support services would be very beneficial. Therefore, healthcare and educational professionals can help to provide the right environment for fostering positive psychological outcomes for parents.

Parents can also make use of the results of this thesis to begin positively reappraising their journeys. For example, they can use a diary to reflect on their experiences. A useful exercise would be to actively imagine their life with and without their child who is affected by the developmental disability, noting both positive and negative changes. Further, support groups provide a great opportunity for peer learning. Parents who have a child with a developmental disability that feel they have experienced positive psychological outcomes could describe their experience to other group members. The interview schedule used in this thesis can be used as a basis for the discussion (e.g., for you, what was/is unhelpful in managing with your child’s condition? For you, what was/is helpful in managing with your child’s condition?). Both these activities may promote purposeful rumination, resulting in conscious meaning-based coping (Folkman, 1997).
8.5.3 A lifespan approach

Challenges and joys associated with their child’s developmental disability will change for parents throughout the child’s life. Early on, there may be urgent fears about immediate health concerns (e.g., repairing heart defects, feeding), whereas as the child’s psychological health may become more pertinent as they reach adulthood. Of course changes with the child’s development are expected for any parent; however, a child with a developmental disability may not be able to live independently and thus the impact may be more significant to this group of parents because their role as primary caregiver is extended beyond what is typically expected.

Due to the changing emotional journey that parents are likely to experience, it is recommended that healthcare professionals endeavour to “check in” with families throughout the child’s development. The PWB-PTCQ (along with other measures of psychological wellbeing) is a useful self-report clinical tool designed to assess psychological functioning following adversity and guide parents in ways they may have changed. Combining clinical judgement with self-report measures provides professionals an open and collaborative way to work with parents which can be tailored to their individual needs. Information and resources in line with the child’s age and stage can continue to be provided even if parents do not specifically request it.

Healthcare professionals can use Bassett et al.’s (2011) guidelines for practical management of 22q11DS to anticipate any problems their clients (as a family) may face. These guidelines, along with Fung et al.’s (2015) guidelines for adults with 22q11DS are a useful tool for when the child leaves paediatric services. The participants commented on the limited support available at this time. Healthcare and educational staff should engage with families well before the child is due to leave their services as an adult to allow for care-planning and the family’s adjustment. Attention to this big
change in health (and educational) supports and how it may impact the parents as primary caregivers is vital, especially when considering the impact health services have on parents’ positive psychological outcomes.

8.5.4 A word of warning

This thesis has focused on the more positive psychological aspects of parenting a child with a developmental disability. As much research talks in categorisation of distress, the emphasis on the potential to grow in these studies does not mean that only seeing the positive side of the experience is a realistic or even worthwhile pursuit. Indeed, being overly optimistic (for example) can actually be maladaptive (Peterson & Vaidya, 2003). Neither is the content of this thesis meant to diminish the very real challenges these parents face in their caregiving role. Rather, it should be clear that both negative and positive psychological outcomes are normal for parents who have a child with a developmental disability and as such, each are worthy of consideration in research and practice. Hardship does not mean that parents are only at risk of negative psychological outcomes. In fact, the struggles they face can be embraced as an opportunity for positive psychological change if the appropriate supports are in place to facilitate psychological growth. This thesis provides an idea of what those supports may be (e.g., coordinated and comprehensive healthcare for the child, practical social support, coping style), but more research is required to identify if unexplored factors (e.g., the child’s diagnosis type) are also implicated. Finally, I do not claim to capture everyone’s experience or the entire experience of having a child with a developmental disability in this thesis. However, the findings have provided insight, and hopefully will motivate positive change in healthcare settings. Contributing to research can be beneficial both individually and for the affected population. Therefore, when feasible,
parents should also consider participating in research related to their wellbeing and their child’s syndrome.

8.6 Conclusion

In conclusion, the quantitative findings of this thesis found that many participants experienced psychological growth which was predicted by greater use of positive reappraisal coping, coordinated and comprehensive healthcare services, and the child’s age; along with less use of escape avoidance coping and a larger discrepancy between ideal and actual practical social support. However, the qualitative studies highlighted those stressors experienced by these parents that were likely to facilitate psychological growth. Anticipatory traumatic distress, systemic stigma, and ‘not knowing’ were overarching themes identified by parents early in their journey. Funnelling down to associated feelings of fear, guilt, loss and grief in these early years hope for the future and a perceived opportunity to become better people provided the platform for growth.

Parents whose children were now adults also interpreted their distress through stigma, guilt, and grief. Layers of professional obstruction and ignorance over time pragmatically replaced hope and triggered angry advocacy for their child, seen as a positive for change. As such, psychological growth was viewed as part of the journey that juxtaposed ongoing distress. It was identified through a metamorphosis of empathy, humility, gratitude, and pride.

This research provides a unique contribution to the literature by extending our understanding of psychological functioning in parents who have a child with a developmental disability beyond psychopathology and into positive changes across the spectrum of self, other, and society. The mixed methods design provided insight into the
differential reactions across the child’s age and stage. The investigation was also distinctive in that under-researched disabilities such as 22q11DS were included. The research highlights that positive psychological outcomes are achievable and realistic. The results provide a platform for further research and potential interventions to promote positive psychological outcomes in parents who have a child with a developmental disability.

It is recommended that clinicians working with families affected by developmental disabilities consult Kisler and McConachie’s (2010) article regarding communication with parents. It provides helpful advice for diagnostic communication, as well as facilitating good relationships between families and healthcare professionals. Professionals working with families affected by 22q11DS should refer to the global overview of 22q11DS (see McDonald-McGinn et al., 2015), as well as the practical guidelines for management of the condition (Bassett et al., 2011; Fung et al., 2015) in order to familiarise themselves with potential concerns families may face and how to manage them.

This thesis has provided insight into the integral role healthcare and educational professionals played in these parents’ experiences, and indeed the family experience. Those working within these systems have the potential to provide opportunities for psychological wellbeing in parents through active listening, validation, and a collaborative style of care management using a person centred approach. The development of systemic training and educational material is a future aim from this research. Finally, this thesis highlights that theories of growth can be applied to cumulative and complex situations where there is little reprieve from anticipatory trauma as experienced by those on a life-long journey with disability.
### Appendices

**Appendix A: Search strategies for PsycINFO database**

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<tr>
<td>37</td>
<td>12 and 31 and 36</td>
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<tr>
<td>38</td>
<td>limit 37 to english language</td>
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Appendix B: Flowchart of study selection process

1. Records identified through database searching
   \( (n = 2173) \)

2. Records after duplicates removed
   \( (n = 1984) \)

3. Records screened
   \( (n = 83) \)

4. Full-text references excluded, with reasons
   \( (n = 74) \)
   - Not examining positive constructs as outcomes \( (n = 51) \)
   - Quality of life \( (n = 11) \)
   - Qualitative \( (n = 6) \)
   - Child > 18 \( (n = 4) \)
   - Not parental outcomes \( (n = 1) \)
   - Not original data \( (n = 1) \)

5. Articles included in qualitative synthesis
   \( (n = 14) \)
## Appendix C: Worked example of IPA

<table>
<thead>
<tr>
<th>Emergent themes</th>
<th>Original Transcript</th>
<th>Exploratory comments</th>
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<tbody>
<tr>
<td>Distress, lone journey</td>
<td>It’s socially isolating having a kid with a disability. [My daughter] wouldn’t enunciate very well, she didn’t have a cleft palate, but she might’ve had submucous, we never really found out…um…so she wasn’t speaking very clearly, and she wasn’t…for the first, until she was about 10 or 12…she was fixated on anime characters and things like that. So she didn’t have a general conversation. She had a conversation around what she was interested in. So adults wouldn’t take time to talk to her…and that- that I found…very disappointing, you know. No compassion in – even my close friends. You know, most of them wouldn’t bother with [my daughter]. Jane: How did you make sense of that? Maria: Um…you’ve gotta fight the big fights and let the little ones go…so just, you know. Our job was to protect K and to help her achieve everything that she could achieve. So generally, we’d gone from being very, very social people to not being very social at all. And that’s – I</td>
<td>Feeling alone, isolated \Feeling alone, isolated \Impact on relationships</td>
</tr>
<tr>
<td>Fighting ignorance</td>
<td></td>
<td>Why don’t you value my child? \Noticing shift in priorities \Upsetting \Lack of understanding from friends – ignorant to challenges? \Discovering what true friendship is? \Letting go. Conserving energy. \Resilience?</td>
</tr>
<tr>
<td>Positive reframing</td>
<td>think a good thing. Because it’s been…it’s created a strong family, [my husband] and I both travelled a lot before we had kids, and when we had kids we decided not to travel – well, not at all really. And I think our intention was to focus on family, from relatively high flyers to um…you know, stop (both laugh) and have a family type thing. And I think that’s meant we’re a very close knit family, so that’s a big benefit. The kids, both of us are here – both of us have always been here in the morning and the afternoon. So, [my husband] was always home for dinner, and you know. Mum and dad is always around, and we’ve got 3 beautiful kids. And we’ve always had the expectation for them all to – to rise up and use the gifts that God’s given them. And they’ve all done that. And we had no trouble with teenage years, or any drugs or alcohol, or…it’s been a breeze. And I think that focus on the family is beautiful.</td>
<td></td>
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<tr>
<td>Change in expectations</td>
<td>Changing values</td>
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<td>– finding new meaning</td>
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<td>Changing values</td>
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<tr>
<td>Comparison between “other” life and current life</td>
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<td>Focus on relationships</td>
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<td>Improved relationships</td>
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<td>Accentuating the positive</td>
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<td>Forgetting negatives? Choosing to focus on positives?</td>
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Appendix D: Published paper - “You Don’t Know Until You Get There”: The Positive and Negative “Lived” Experience of Parenting an Adult Child with 22q11.2 Deletion Syndrome

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Health Psychology

“You Don’t Know Until You Get There”: The Positive and Negative “Lived” Experience of Parenting an Adult Child With 22q11.2 Deletion Syndrome

Jane Goodwin, Lynne McCormack, and Linda E. Campbell


CITATION


Article removed due to copyright restrictions
Appendix E: Conference proceedings

Oral presentations (*presenter)

1. Rudd, K., Goodwin, J., Campbell, L. E.* ‘Same, Same but Different: The importance of social support for fathers of kids with developmental disabilities’ VCFS 22q11 Foundation Conference. Sydney, Australia (2016).


3. Goodwin, J., Alam, S.*, Campbell, L. E. “‘At the end of the day it is more important that he stays happy’: An interpretative phenomenological analysis of people who have a sibling with 22q11.2 deletion syndrome’ VCFS 22q11 Foundation Conference. Sydney, Australia (2016).

4. Goodwin, J., McCormack, L., Campbell, L. E.* “‘You don’t know until you get there”: The positive and negative ‘lived’ experience of parenting an adult child with 22q11.2 deletion syndrome’ IASSIDD 15th World Congress. Melbourne, Australia (2016).

5. Rudd, K.*, Goodwin, J., Campbell, L. E. Same, Same but Different: The importance of social support for fathers of kids with developmental disabilities’ IASSIDD 15th World Congress. Melbourne, Australia (2016).


**Poster presentations (**presenter**)


cerebral palsy’ YPAGne Young people’s voices: shaping the future of research and healthcare. Newcastle Upon Tyne, UK (2016).


**Additional presentations (*presenter)**


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