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## **DOCTOR OF PHILOSOPHY**

### **Parental bereavement when a child with an intellectual disability dies**

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*Award date:*  
2008

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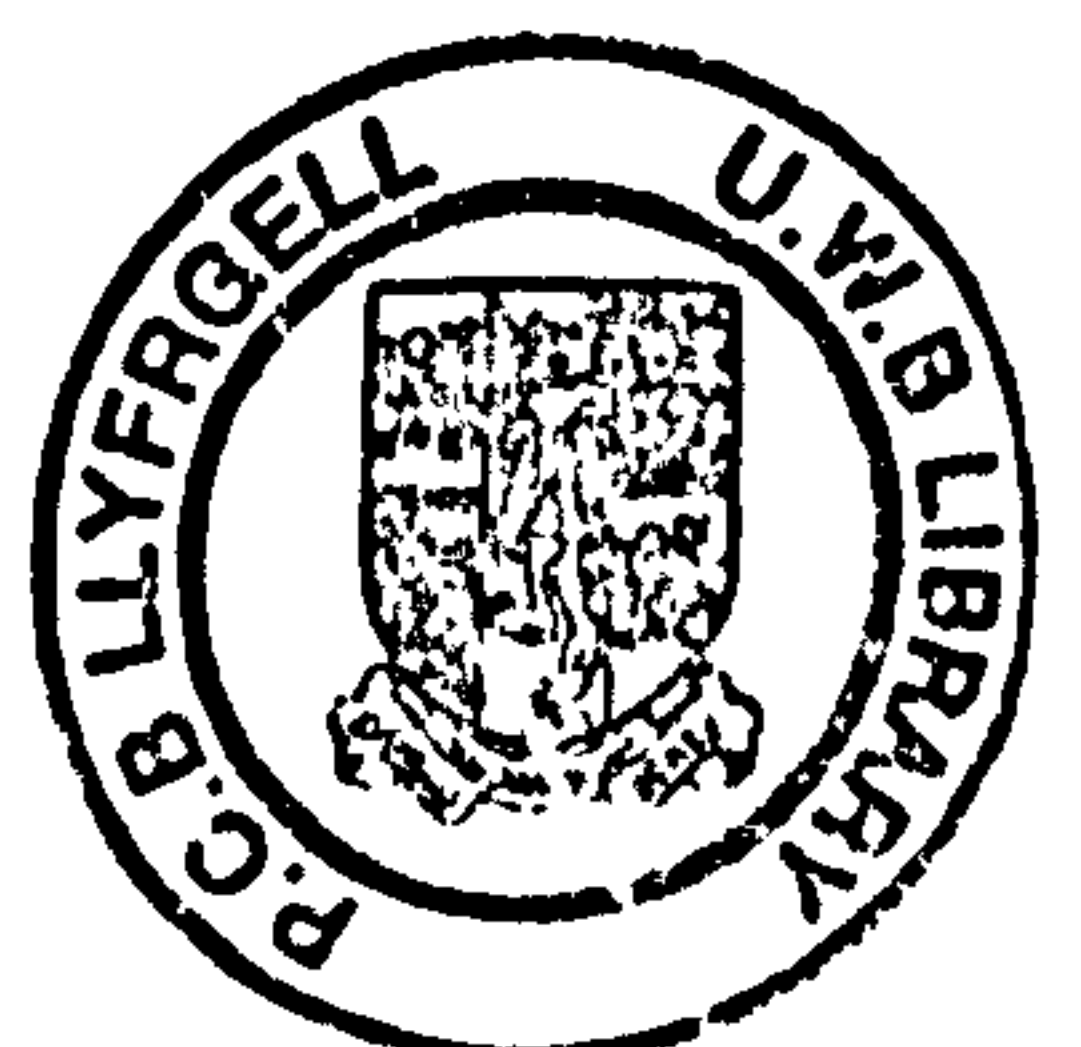
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**Parental Bereavement when a Child with an Intellectual  
Disability Dies.**

Deirdre Emer Reilly

A thesis submitted to the School of Psychology, Bangor University, in partial fulfilment of  
the requirements of the degree of Doctor of Philosophy.

February 2008



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## Acknowledgements

Thank you to the European Social Fund, Mencap Cymru, and Bangor University who funded the project. To Dewi Ward and Wayne Crocker who ensured this project got off the ground, and I received all the help I needed along the way. To my PhD committee for their guidance, Jaci Huws, Frances Vaughan and most importantly Richard Hastings whose commitment, support, and dedication have made me the envy of many.

Thanks to all IDDRG members past and present for their advice, company, friendship, and food, special thanks to Tracey and Matthew Lloyd who gave me their home when I most needed it. Thank you to all in the School of Psychology who have helped me out along the way. I would never have made it through the last three years without the many friends and flatmates, old and new, who were ever available with their words of encouragement, I hope some day and can repay my debt.

To my sisters and parents who ensured I made it this far and always knew the right time to call, I love you. To the extended Lillis clan who always gave me somewhere to go when I needed to get away, especially to Ciara who can always make things better.

And most of all my thanks go to all of the family members who participated in my research and shared their most cherished memories of their children, grandchildren, sisters and brothers. This thesis is dedicated to their memories and to mine:

Evan Lillis (1980 – 2003)

Mick Lillis (1912 – 1999)

Molly Reilly (1916 – 2007)

Níl libh na leigheas in aghaidh an bháis.

(There is no herb or cure for death, Irish proverb).



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## Summary

This thesis describes a series of studies investigating the experiences of parents whose child with an intellectual disability has died. Parental bereavement research has identified a range of symptoms exhibited in grief, and acknowledged that circumstances surrounding the loss may have an impact on symptoms exhibited. Little is known about the circumstances surrounding the death of a child with an intellectual disability. An extensive review of the literature (Chapter 2) uncovered a small number of qualitative studies with such an experience as their focus. Reports of disenfranchised grief, unsatisfactory healthcare, variations in coping strategies, and positive reflections characterised parents' accounts. The aim of the thesis was to use mixed methods to investigate the variety of parental experience when a child with an intellectual disability dies.

In Study 1 (Chapter 3) Interpretative Phenomenological Analysis (IPA) was used to inspect nine mothers' accounts of the parenting and bereavement experience. Five themes emerged: loss; benefit finding; coping; sources of support; and medical relationships. Similar analysis of interviews with six bereaved couples of children with Down syndrome and a congenital heart condition (CHC; Chapter 4) highlighted differences that result when couples are faced by a bereavement. Four themes were extracted from the data: "One disastrous diagnosis after another"; "We had to make a decision"; "We weren't really going through it together"; and Ripples from the child's life. Quantitative analysis of the experiences of 38 mothers whose child with Down syndrome and a CHC died (Chapter 5) uncovered intense grief reactions alongside high levels of positive perceptions. Mothers with higher grief scores used more active avoidant coping strategies. Regression analysis indicated that the use of active avoidant coping and holding positive perceptions accounted for a significant amount of variance in total grief scores. Findings of these three studies are discussed in terms of their contribution to the literature, implications for policy and practice, methodological and theoretical limitations, and potential avenues for future research.

**Chapter 1 –Parental Bereavement when a Child with Intellectual Disabilities Dies: An  
Introduction**



The term “intellectual disability” is defined by the DSM IV as the presence of significantly sub-average intellectual functioning associated with or resulting in impairments in adaptive behaviour, which occurs before the age of 18 (American Psychiatric Association (APA) 1994). Intellectual disability is classified according to four degrees of severity: mild; moderate; severe; and profound, and can be diagnosed in addition to the presence of another disorder (e.g. cerebral palsy). Disorders related to intellectual disability may result from predisposing hereditary factors (e.g. tuberous sclerosis), biological factors (e.g. Down syndrome due to trisomy 21), and a range of environmental influences, perinatal problems, general medical conditions acquired in infancy or childhood, and mental disorders (e.g. Autism) (APA, 1994). Individuals are also at risk of co morbid mental and physical illness (Van Schrojenstein Lantman-de Valk, 2005). Perhaps because of the increased comorbidity for other disorders, people with intellectual disability are also at risk of dying at a younger age than members of the typically developing population (Janicki, Dalton, Henderson, & Davidson, 1999). While the impact that raising a child with intellectual disability on their family has been the subject of much research (e.g. Hastings & Taunt, 2002; Hodapp, Ly, Fidler, & Ricci, 2001), the effect of the death of a child on parents of a child with an intellectual disability is an area that has received little research attention.

This thesis investigates the experiences of bereaved parents whose child with an intellectual disability has died. This introduction (Chapter 1) will provide information on a number of methodological issues that were considered prior to embarking on the research, set the methodological context within which the research was conducted, and explain some of the choices made with regard to methods used. An overview of methodology in bereavement research will be followed by a discussion of the use of qualitative methodology in



bereavement research, and finally an outline of potential ethical issues identified as relevant to the current research is provided. The introduction will conclude with a synopsis of each of the four experimental chapters.

### *An Introduction to Methodology in Bereavement Research*

Research investigating the effects of bereavement generally uses one of two strategies: the use of traditional quantitative research methods using standard measures of symptoms, or one of a variety of qualitative methods (Neimeyer & Hogan, 2001). In recent times, the necessity for using grief specific measures has been recognised by researchers, as grief and grieving phenomena have been acknowledged as symptoms in their own right and not simply equitable with affective symptoms such as depression (Prigerson et al., 1995a). The use of valid and reliable instruments measures such as the Inventory of Complicated Grief (ICG, Prigerson et al., 1995b), or the Revised Grief Experience Inventory (Lev, Munro, & McCorkle, 1993), allows for detailed measurement and facilitates the comparison of grief symptoms in large groups of differently bereaved people over time through statistical analysis (Neimeyer & Hogan, 2001). However, as in other research fields, the use of survey data collection has its limitations. Participants may feel obliged to exacerbate or exaggerate symptoms as a function of demand characteristics (Orne, 1962), and standard measures potentially miss unique and important differences displayed by particular groups or individuals (Stroebe, Folkman, Hansson, & Schut, 2006).

The domain of qualitative bereavement research, in some ways, overcomes these limitations. Qualitative methods, on the whole, have developed from social constructionist theories, which emphasise that there is no absolute truth but instead reality is constructed from experience (Murray & Chamberlain, 1999). Characteristics common to many

qualitative methods include prolonged contact with field situations; aiming to gain a systemic overview of the experience under investigation from an insider's perspective; themes may emerge from the data but the data itself remains in the original form; and many interpretations may be possible but some will be more relevant and compelling than others (Miles & Huberman, 1994). This focus on lived experience adds depth to the assessment of the grieving process, and are ideally suited to the detailed exploration of contributing factors in individual experiences (Neimeyer & Hogan, 2001). Additionally, the qualitative researcher may explore, in detail, variables of theoretical interest. Qualitative methods are therefore a tool useful in generating theory where little theory exists, and broadening general understanding as opposed to making causal inferences (Strauss & Corbin, 1998).

While qualitative research methods have distinct strengths to support their use in bereavement research such as an emphasis on meaning and coherence; minimising the researcher/participant power imbalance; focus on language as the key unit of data; and inherent reflexivity (Owens & Payne, 1999), they do not come without weaknesses. There are limits to the causal explanations that may be generated from qualitative descriptions of grief, the assessment of efficacy of intervention is difficult to demonstrate, identifying correlates of grief as measured by quantitative means is problematic, and the relevance of findings to broader populations is questionable (Neimeyer & Hogan, 2001; Owens & Payne, 1999; Stroebe, Stroebe, & Schut, 2003). In addition, issues of reliability, validity, and credibility can be of considerable concern (Stroebe et al., 2003). Chapter 3 provides an extensive evaluation of these limitations.

To overcome the weaknesses displayed by both qualitative and quantitative measures, there has been considerable advocacy in the realm of bereavement research for a move towards methodological pluralism (Neimeyer & Hogan, 2001). The central idea being that

any investigative procedure will have its weaknesses, and these failings may be complemented by another approach whose weaknesses are different (Owens & Payne, 1999). Specifically qualitative methods could bring novelty, scope and depth, to the subject area, which can then be refined and probed further by quantitative means (Neimeyer & Hogan, 2001; Owens & Payne, 1999; Parkes, 1985; Stroebe et al., 2003). Bryman (2006) outlines guidelines for integrating methods in research and postulates the need for a transparent research process that includes information as to which method is of priority; the function of the integration; and information on when the multi-strategy research formulation occurs. Research projects should also clearly indicate that quantitative and qualitative methods were geared towards answering specific and different research questions (Bryman, 2006).

### *Qualitative Methods in Bereavement Research*

A variety of qualitative methods have been used to investigate parental bereavement. Grounded theory methodology (Glaser & Strauss, 1967), posits that the data alone shapes the products and processes of research (Charmaz, 1995), and the researcher does not allow personal characteristics or theoretical views bias subsequent analysis. Schormans (2004) reports on themes extracted from interviews with foster carers whose child with a disability had died, which centred on the perceptions of their parental role, and the conceptualisation of their relationship with their child. Content analysis (Neuendorf, 2002), on the other hand allows the researcher to approach the data from a particular theoretical viewpoint and code data in light of categories suggested by the theory. Miles and Demi (1992) used content analysis to compare their model of sources and feelings of guilt in parents bereaved by accident, suicide, and chronic illness. They used patterns of guilt in different groups to generate theory in line with their conceptual model of grief. Limitations of both methods



include the reluctance to allow the data itself to guide the analysis process, and a lack of acknowledgement of the relationship between researcher and participant that may be particularly pertinent in sensitive research.

Focus groups (Morgan, 1993) allow the exploration of socially constructed realities within groups by holding group meetings with typically between six and twelve participants that are facilitated by a moderator, where individuals comment on broadly relevant questions. Research in Israel (Malkinson & Bar-Tur, 1999) facilitated the investigation of grief over time in a group of war-bereaved parents and explored how it is effected by the aging of the parent. Themes to emerge from the research indicated similarity in experience for many parents, and while the focus group approach may have encouraged more parents to become involved in the research, potentially important or different accounts may have gone unrecorded, as the authors report there were parents who remained silent in the group meetings (Malkinson & Bar-Tur, 1999). Similarly, in ethnographic research the objective of the research moves from understanding the experience of the individual to that of the group, though ethnographers are typically interested in matters of a social anthropological and cultural nature (Miles & Huberman, 1994). The use of a variety of data collection methods, sources, and participants, can give a vivid insight into the concept under scrutiny (e.g. Klass, 1997), however, data collection can be time consuming, un-standardised, and the focus on very small groups may have limited implications for theory and practice.

Interpretative Phenomenological Analysis (IPA, Smith, Jarman, & Osborn, 1999), is a qualitative research method whereby the participant is deemed an expert on their own experience, although the impact of the researcher and the context of the research is taken into consideration. It has yet to be utilised with bereaved parents. However, IPA has produced important findings with regard to parental experiences when their child has a life-limiting

condition (Davies, Davis, & Sibert, 2003), and has been used intensively in health psychology research (Brocki & Wearden, 2006). IPA aims to identify diversity as well as similarity across participants' accounts and therefore is suitable for undertaking an in-depth exploration of a small homogenous sample, but as with all qualitative methods relevance to a broader population will be limited (Owens & Payne, 1999). A full account of the background, theory, and practicalities of IPA is provided in Chapter 3.

One tool central to qualitative methods is the qualitative research interview (King, 1994). Qualitative interviews can vary greatly in their focus from broad to specific, but all share a common purpose, which is to “understand the world from the subject’s point of view, to unfold the meaning of people’s experiences, to uncover their lived world prior to scientific explanations” (Kvale, 1996, p. 1). Kvale proposes that the focus of an interview will depend on the role of the interviewer and proposes the metaphors of the interviewer as a miner and the interviewer as a traveller to differentiate between broad and narrow theoretical approaches to interviewing (Kvale, 1996). King (1994) proposes three broad categories of interview: the qualitative research interview; the structured interview; and the structured open-response (or semi-structured) interview. The type of interview to be used should follow a careful consideration of the nature of the research question to be addressed with regard to the specific aims of the research; prior knowledge of the subject area; and how much information participants will be likely to provide (King, 1994).

Despite their commonality, the use of interviews in qualitative psychology is currently much debated (Potter & Hepburn, 2005; Smith, Hollway, Mishler, Potter, & Hepburn, 2005). Potter and Hepburn (2005) propose that interviews are overused in qualitative psychology. They cite lack of descriptive information on the interview set-up and interviewer training among the issues that contribute to the major flaws of the method, and

propose that other methods should be used where possible. In response to these proposals Smith and colleagues (2005), argue that in idiographic studies interviews are still the most appropriate method. However, researchers do agree that the inclusion of additional information in published works detailing interview set-up; questions; researcher introductions and responses; and researcher perspective and training may improve the quality of interviews, as would a clear focus on the interview as an interaction between interviewer and interviewee (Hallway, 2005; Mishler, 2005; Potter & Hepburn, 2005; Smith et al., 2005).

### *Ethical Issues in Bereavement Research*

The interaction between researcher and participant is key in qualitative research but can put both parties at risk. Ethical consideration of potential risks to both researcher and participant should be undertaken regardless of research methodology; however, additional risk may arise in qualitative research as a result of characteristics of the method. Firstly, the interaction between interviewer and interviewee can give rise to problems of relationship boundaries (Dickson-Swift, James, Kippen, & Liamputtong, 2006). Risks associated with being a professional researcher may include uncertainty surrounding self-disclosure; emotional and physical impacts of research; feelings of vulnerability and guilt; and difficulty establishing an appropriate rapport with participants (Dickson-Swift et al., 2006; Dickson-Swift, James, Kippen, & Liamputtong, 2007; Dunn, 1991).

Secondly, both researcher and interviewee may encounter difficulty with research-therapy, and research-friendship boundaries (Dickson-Swift et al., 2006). Despite the non-therapeutic intentions of the qualitative interview, the interview often has therapeutic outcomes for both researcher and participant, and interviewers and counsellors require similar skills, therefore professional guidelines offered to counsellors with regard to, for



example, burnout and supervision, may also be informative in this area (Dickson-Swift et al., 2006; Hermansson, 1997).

Finally, risks additionally exist for third parties mentioned in transcripts who have not consented to participate in research. The limitations in confidentiality afforded by the use of pseudonyms are often not noted by research, and while some references to third parties in terms of normal activities or as a generalisation, may be ethically unproblematic. Potentially personal or damaging references should be avoided as should detailed descriptions that facilitate the recognition of both participants and individuals who have not consented to participate (Hadjistavropoulos & Smythe, 2001).

Elliott and colleagues (1999) describe clear, informative guidelines that are pertinent to reducing risks to participants and researchers when writing-up and publishing qualitative research. These include improved credibility checks, grounding in examples, and disclosing detail on the author's perspective. However, guidelines at the publications stage may come too late. Hadjistavropoulos and Smythe (2001) argue that such risks should be addressed prior to gaining ethical approval, and that such guidelines could be adopted from the American Psychological Association's (APA, 2002) guidelines on ethics for research and publication (APA, 2002, p. 1069), to be referred to by researchers and ethical committees.

In addition to risks posed by participation in qualitative research, risk to participants and researchers may result from the research topic. It is recognised that bereavement research may be intrusive and have the potential to cause emotional pain to participants, but this occurs in tandem with positive experiences reported by participants (Burnell & O'Keefe, 2004; Cook & Bosley, 1995; Dyregrov, 2004; Rosenblatt, 1995; Seamark, Gilbert, Lawrence, & Williams, 2000). Guidelines, such as those outlined by Parkes (1995), can ensure distress to participants is minimised, for example, by providing clear written information, not making

first contact via telephone, and providing descriptive results and feedback regardless of the outcomes of the research project. Parkes also makes suggestions for guidelines to protect researchers from harmful outcomes, including the use of good methodology, adequate training, and appropriate supervision (Parkes, 1995).

Undertaking qualitative research on sensitive topics such as bereavement requires reflection, imagination, preparation, and trust on the part of the researcher (Dickson-Swift et al., 2007; Johnson & Clarke, 2003; Lee, 1993), unfortunately, if this reflection and decision-making does happen, it often occurs too late in the research process to have sufficient impact on the design (Hadjistavropoulos & Smythe, 2001). In the design stages, considerable consideration should be given to the fact that sensitive research presents more than the minimum risk to participants and researchers and a number of ethical decisions will have to be made on issues such as recruitment; informed consent; assessment of actual and perceived risks, potential benefits of participation; and researcher qualifications (Cook, 2001). Additionally, ethical committees should be allowed access to complete information on a research study in order to aid a fair evaluation, which should also be unbiased by cultural or personal opinion (Cook, 2001; Hadjistavropoulos & Smythe, 2001).

### *The Present Thesis*

The primary aim of this thesis was to use both qualitative and quantitative methods to illustrate the experiences of parents whose child with an intellectual disability has died. Mixed methods were chosen to explore the area and while qualitative methods allow for the in-depth exploration of a relatively unknown phenomenon, quantitative measures allow the researchers to investigate participants' experiences in relation to each other, and in comparison with previously researched groups. The thesis is structured as four linked



chapters: one qualitative literature review; two qualitative studies; and one quantitative study. Each chapter focuses on issues related to the experiences of parents whose child with an intellectual disability has died, which has received little research attention to date.

The second chapter is a narrative literature review in which we identify previous research in the area, explore other related and potentially informative research areas, and illustrate potentially suitable methodology for future research studies. Topics raised as relevant to parents included parental experiences of disenfranchised grief, dissatisfaction with care post-loss, and un-met support needs after the death of a child with an intellectual disability. The review also indicated the absence of quantitative research with parents whose child with an intellectual disability has died, a lack of previous research in the UK relating to the individual experiences of mothers and fathers, and a deficiency of research into the potentially important experiences of specific groups of parents based on the diagnosis their child had received (e.g. Down syndrome).

The third and fourth chapters describe qualitative studies which explore the experiences of parents in the UK whose child with an intellectual disability has died, specifically their long lasting grief experiences. Telephone interviews were completed with all parents and the use of interpretative phenomenological analysis facilitated the telling of their complete story and allowed the parents' experiences to directly inform the research findings.

The third chapter was a qualitative analysis of the experiences of nine mothers whose child had died. The children had a variety of intellectual disability related conditions though mothers whose child had Down syndrome and died as a result of a congenital heart condition were excluded from the analysis because of the unique nature of their experience. Five themes emerged from the data: loss; benefit finding; coping; sources of support; and medical

relationships. In particular, continuing in their caring role by working within the world of intellectual disability following the death of their child was important to all of the mothers. Support from similarly bereaved parents was the most useful source of support, although it was not always readily accessible. The analysis indicated similarities and differences between the experiences of the mothers whose children had a variety of intellectual disabilities relate conditions. Suggestions for future research include the inclusion of the experiences of fathers and the in-depth analysis of experiences by condition.

In response to the lack of information on the experiences of bereaved fathers, the fourth chapter investigated the experiences of six married couples whose child with Down syndrome and a congenital heart condition had died. A qualitative analysis was carried out with regard to issues raised by couples in individual interviews and four themes emerged as descriptors of the experiences: “One disastrous diagnosis after another”; “We had to make a decision”; “We weren’t really going through it together”; and ripples from the child’s life. Research implications include the importance of looking beyond gender as a risk factor in bereavement research. The themes are discussed with regard to the implications for professionals dealing with couples; bereavement interventions for couples; the benefits and limitations of syndrome specific research; and the potential support needs for parents who do not have a supportive partner or family member throughout the life and death of their child with Down syndrome, particularly fathers, the individual experiences of whom have not been addressed in this thesis.

The fifth chapter describes a study which used quantitative methods to examine relationships between parent and child demographic variables and bereavement outcome variables for mothers whose child with Down syndrome and a congenital heart condition has died. Maternal grief, coping, and positive perceptions were assessed by

measures completed by 38 mothers. Statistical analyses found relationships between grief scores, active avoidance coping strategies, and positive perceptions relating to the life of their child with Down syndrome. Regression analyses indicated that active avoidance coping accounted for a considerable amount of the variance in grief scores. Implications and suggestions for interventions aimed specifically at this group are discussed. Also, findings suggest future research should undertake a more detailed investigation of the experiences of mothers to identify possible explanations for the lasting grief intensity they reported.

Chapter 6 (Conclusions) summarises the findings and implications of the above research and reflects on methodological and theoretical issues raised. Additionally, it examines the potential influence the current findings may have on service provision for bereaved parents, and the need for increased awareness amongst health professionals.

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**Chapter 3 – An IPA Analysis of the Experiences of Mothers whose Child with an Intellectual Disability has Died.**



### Abstract

Individuals with intellectual disability are at higher risk of premature death compared to individuals without intellectual disability, and therefore parents of people with intellectual disability are more likely to outlive their children. However, there has been relatively little research investigating the bereavement experiences of parents of deceased children with intellectual disability. Semi-structured interviews were used to explore the experiences of nine mothers whose child with intellectual disability had died. The transcripts were analysed qualitatively using Interpretative Phenomenological Analysis (IPA). Five themes emerged from the analysis: loss, benefit finding, coping, sources of support, and medical relationships. An analysis of the accounts indicated similarities and differences between the experiences of the mothers. In particular, continuing in their caring role by working within the world of intellectual disability following the death of their child was important to all of the mothers. Support from similarly bereaved parents was the most useful source of support, although it was not always readily accessible. Implications for theory and practice are described including suggestions that service providers should aim to maintain links with families after the death of their child, a parent-to-parent programme that enables parents to contact others in a similar situation might be beneficial, and further research into the functions of coping strategies and supports in loss would be worthwhile.

## Introduction

People with an intellectual disability have an increased risk of premature death compared with typically developing individuals (Janicki et al., 1999). While life expectancy for people with intellectual disability has increased greatly since the early twentieth century (Patja, Iivanainen, Vesala, Oksanen, & Ruoppila, 2000), people with Down syndrome, for example, may still die up to 17 years earlier than their peers (Janicki et al., 1999). In addition, limited life expectancy in intellectual disability is related to life-limiting conditions (e.g. Mucopolysaccharide disorders, Pastores et al., 2007); higher rates of medical complaints (e.g. congenital heart conditions, Hayes et al., 1997); and inadequate health care (Jopp & Keys, 2001; Mencap, 2007). As a result, people with intellectual disability are more likely to predecease their parents although formal data on the increased risk of this eventuality are not available.

Parents of children with intellectual disability face increased challenges and demands associated with their child's current and future medical, educational, and personal needs, (Reeves et al., 2006), and report more parenting stress and mental health problems than parents of typically developing individuals (Olsson & Hwang, 2001). These risks may have important repercussions post-loss. Parental bereavement research has shown that the loss of a child may result in wide ranging psychological implications for parents, including anxiety and depression (Kreicbergs et al., 2004; Leahy, 1993). Grief responses in parents who lose a typically developing child differ, for example, according to how the child dies (Dyregrov et al., 2003), and the age of the child (Moss et al., 1986; Rubin & Malkinson, 2001). Compared to parents of typically developing children, parents of individuals with intellectual disability

are already a psychologically at-risk group. However, although these parents are more likely to experience the death of a child, little is known about their bereavement experiences.

We located four previous studies reporting the bereavement experiences of parents of children with intellectual disability, three in the USA and one in the UK. The studies report on the double losses of disability and death that parents experience, and indicate that while these parents form a unique group, their losses are not always acknowledged, or understood by medical and bereavement professionals (Milo, 1997; Todd, 2007; Wood & Milo, 2001). This disenfranchisement of grief was particularly true for foster parents (Schormans, 2004). None of the studies found evidence for an effect of the child's age on grief, or of diminishing effects of grief over time. The potential for gender differences in coping between parents has been suggested (Milo, 1997; Wood & Milo, 2001), and Todd (2007), highlights the value of religious coping for some parents, but does not indicate whether this was gender related.

Completion of the Grief Experience Inventory (GEI: Sanders et al., 1978), indicated that grief profiles of the biological mothers and fathers did not differ significantly from bereaved parents who lost typically developing children (Milo, 1997; Wood & Milo, 2001). Schormans' (2004) grounded theory analysis of interviews with six bereaved foster mothers and two bereaved foster fathers indicated that birth and foster parents' experiences of losing a child with intellectual disability are analogous. Todd's (2007) account of qualitative interviews with the bereaved parents of 13 children with intellectual disability (five couples and eight mothers) in the UK concurs with findings from the USA on disenfranchised grief, the loss of identity, and positive outcomes for parents. The lack of information on the children's conditions and how they died in Todd's research limits the conclusions with regard to how diverse or similar the participants were, and how the circumstances of the deaths compare to those of the bereaved parents in other studies.



Further research in this field is needed to build on the limited number of studies and investigate whether findings thus far are more general patterns with practical implications for theory and practice. Differences in healthcare provision, education, and cultural views of disability generally, may result in very different outcomes for parents in the UK, in comparison with the USA. The aim of the present study is to focus on the experiences of mothers who lose a child with an intellectual disability in the UK, to provide a rich description of their bereavement experiences, and offer an interpretation of these experiences in terms of issues relevant to service providers, researchers, and medical professionals.

## Method

### *Methodological Approach*

Limited research exists in the area of parental bereavement and intellectual disability and, as this experience is potentially different to previously researched groups, qualitative methods are suited to deepening understanding of the event (Neimeyer & Hogan, 2001). Interpretative Phenomenological Analysis (IPA: Smith et al., 1999) is a qualitative approach whereby the participant is deemed an expert on their own experience, and methods of interpretative analysis are used to uncover the central themes from participants' accounts of particular events. These themes may be common across, or unique to participants, and IPA aims to reveal rather than suppress the diversity of participants' accounts (Jarrett, Payne, Turner, & Hillier, 1999). By allowing the bereaved participants to identify the symptoms, themes, practices, and outcomes, which characterise their loss, IPA captures how participants themselves make sense of their experiences.

IPA studies typically focus on small samples of between six and eight to allow in-depth examination of participant accounts, although participant numbers vary from one to

thirty (Brocki & Wearden, 2006). The use of a small, somewhat homogeneous group facilitates the analysis of patterns of similarity and difference in the group and helps avoid the loss of subtle differences between participants (Smith, Michie, Stephenson, & Quarrell, 2002). Validity is achieved through credibility checks carried out by multiple researchers, ensuring the themes are internally coherent and grounded in examples from the transcripts (Elliott et al., 1999). Additional tools which aid verification and consistency, and ensure the quality of results include: discussion with the research team; peer review; consultation with the participants on the accuracy of the findings; and the use of a reflexive journal (Carradice, Shankland, & Beail, 2002).

Within the IPA approach, it is recognised that the interviews are a product of an interaction between participant and researcher. The researcher's view will influence the emerging themes, and is necessary to make sense of the personal world of the participant through interpretative activity (Smith et al., 1999). In the current study, the primary researcher was a PhD student completing psychological research in bereavement and intellectual disability. She had received training in working with bereaved individuals and had access to clinical supervision throughout the study. A detailed audit of the analysis was conducted by a second researcher who had considerable expertise in IPA. A reflexive diary was kept by the researcher. This was consulted throughout the analysis and write-up process. The audit process resulted in some minor changes to themes and analysis such as how lists of relevant quotes were created and stored. The study was approved by the research ethics and governance committee of Bangor University's School of Psychology.

*Participants*

Participants were recruited as part of a larger project to interview bereaved individuals who had lost a family member with an intellectual disability. Information was sent to support groups, hospices, and service providers in the UK requesting that flyers be forwarded to relevant families (See Appendices A-F). Interested individuals were asked to contact the research team (see Appendices G and H). Parents who had lost a child at least 12 months previously were invited to participate. One parent who lost her child 10 months previously asked to take part, and was included in the research. No upper limit was placed on time since loss and no limits were placed on age of child or parent.

The participants were drawn from a group of 35 bereaved mothers who completed the interview (described below). In order to narrow the sample to a more homogenous group, interview transcripts for nine mothers were selected for analysis (Table 3.1). These mothers represented all of the interviewees whose children had not been in receipt of routine medical care from birth for a congenital condition, as was the case, for example, for children who had Down syndrome and a congenital heart condition (whose mothers were recruited through an associated support charity). The perspectives of these other mothers, sharing the experience of losing a child with Down syndrome and a congenital heart condition, will be the subject of a separate report.

Table 3.1 Characteristics of the Participating Mothers and their Children

Mother (age)	Child (age)	Time since death	Child's diagnosis	Cause of death
Helen (42)	Polly (30mths)	10 years	Down syndrome	Meningitis
Jennifer (47)	Josh (7yrs)	5 years	Down syndrome	Leukaemia
Rose (60)	Andy (18yrs)	3 years	Down syndrome	Liver failure
Joan (62) <sup>1</sup>	Nick (13yrs)	3 years	Profound and multiple intellectual disabilities	Hospital infection
Jackie (42)	Jake (14yrs)	2 years	Sanfillipo disease	Degenerative condition
Toni (52)	Michael (15yrs)	7 years	Global Developmental Delay	Epilepsy
Sandra (26)	Julie (23mths)	10 months	Cerebral palsy & intellectual disability	Feeding tube complications
Alice (40)	Joe (15.5yrs)	4 years	Down syndrome	Blood clot
Philippa (55)	Charlotte (15yrs)	2.5 years	Down syndrome	Septicaemia

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<sup>1</sup> Joan was a foster mother who also lost two other foster children 22 and 18 years ago. In her interview she talks predominantly about her most recent loss, Nick.



The ages of the mothers in the present sample ranged from 26 years to 62 years ( $M = 47.56$  years;  $SD = 11.60$ ). The children were aged between 23 months and 18 years ( $M = 10.64$  years;  $SD = 6.55$ ), at death, and the mean time since death was 4.20 years (range 10 months -10 years;  $SD = 2.79$ ). All mothers were biological mothers to the child who died with the exception of Joan who was an adoptive mother to Nick. Joan had also been a foster parent to two children who had died in the past, but spoke mainly of her most recent loss in the interview. Seven of the mothers were English, one was Scottish (living in England), and one was Welsh. Three had other children with intellectual disability and two of the mothers had given birth to additional children following the death of their child. Five of the mothers had separated or divorced from their child's father prior to the death. None of the mothers had divorced or separated from a partner since the death of their child. The mothers lost children with various intellectual disabilities, as described in Table 3.1. Two of the children died suddenly in their own homes, three died either in a hospice or at home with a palliative care team present, and four of the children died in hospital following a period of medical intervention.

### *Procedure*

The semi-structured interview was developed from questions used by Milo (1997) and the schedule was piloted with a small group of parents. The pilot resulted in some changes to the schedule. The interview schedule included subsidiary questions and prompts, and topics were arranged chronologically covering the entire experience of giving birth to, raising, and losing a child with intellectual disability (See Appendix J). Interviewees did not see a copy of the schedule. However, participants received information on the aims of the project and potential topics that would be raised during the interview (e.g., parenting a child with

intellectual disability, how their child died, what support they received). To facilitate participation by people across the UK, a decision was made to conduct all interviews over the telephone. Previous bereavement research carried out via telephone found that bereaved participants were more satisfied with the interview length than non-bereaved controls, and the majority rated the experience as not at all, or a little stressful (Taneja et al., 2007). In addition, although interviewing by telephone can reduce the interpretation of non-verbal cues that can provide a further context for analysis (Kvale, 1996), telephone interview has been found to be a useful method of qualitative data collection (Sturges & Hanrahan, 2004).

By beginning with general questions about the family and the child while they were alive, mothers became more relaxed, became familiar with the researcher, and were more at ease about telling their story. Allowing participants to tell their entire story has been highlighted as important in bereavement research (Burnell & O'Keefe, 2004; Dyregrov, 2004). The order in which the topics were discussed was dictated by the participant, and at the end of the interview mothers were given the opportunity to raise any additional topics. The majority of participants became tearful at some point during the interview, and when this happened the interviewer encouraged the mothers to take their time. None requested that the interview end prematurely. All of the interviews were one-time interviews carried out over the telephone, and lasted an average 61.33 minutes (range 40-111mins).

Once the participant had initiated contact, had been provided with an information pack by post, and had provided written consent by return post (See Appendix I), the researcher telephoned the participant to answer any queries, ascertain consent, and arrange a time to conduct the interview. The interviews were recorded using a Sony™ ICD10-MX20 digital recorder and during the interview demographic detail (See Appendix K), was recorded by the researcher. Any unanswered questions were asked at the end of the interview. After

the interview, the recorder was switched off and participants were given an opportunity to discuss the topics further. The majority of participants made additional comments at this time about the experience of taking part in the interview and asked how the data were to be used. Comments made at this point were not included in the results. Participants were sent a personal note thanking them for their participation and were provided with a list of relevant support contacts should they wish to discuss issues further (See Appendix O). As the recruitment process was confidential, the total number of parents who received information about the study is not known.

### *Data Analysis*

The interviews were fully transcribed and were not shared with participants. This topic is the subject of current debate amongst IPA researchers, but given the interpretative nature of the approach (see above) it is not applied in the majority of IPA research (Flowers, 2007). The interviewer listened to and read through the first interview several times to gain familiarity with the interview content. The interviewer then read through the transcribed data line by line noting preliminary interpretations in the left hand margin of the transcript. Further readings were repeated and emerging themes were noted in the right hand margin (See Appendix P). This process of reading and note taking continued until the interviewer was satisfied that all themes had been identified from the data and a separate list of themes and relevant verbatim quotes was created. Additionally, a master list of themes was drawn up and emerging sub-themes were arranged into groups under master theme headings. The process was repeated for each transcript and the master themes list was expanded. Following the analysis of each transcript, the second researcher read through the transcript, the list of themes and quotes, and the list of master themes to ensure the emerging themes were visibly



grounded in the data and none had been omitted. Discussion between the researchers resulted in minor changes to the arrangement of themes. The master theme list changed constantly throughout the analysis and write-up, with the addition and collapsing of themes into categories until a final list of five themes emerged. The themes are discussed below and illustrated with verbatim quotes from the interviews. For purposes of confidentiality, all names have been changed, however the researchers additionally made it clear in the information sheets provided prior to consent, that any data collected may be used in the creation of reports and articles in the form of quotes.

### Results

Following IPA analysis, themes identified in the interviews were grouped into five master themes: loss, benefit finding, coping, sources of support, and medical relationships. Each theme will be discussed and illustrated with verbatim extracts from the interviews. When providing extracts from the interviews the following transcript conventions are used:

... Short pause

(...) Words omitted to shorten quote

[text] explanatory information included by author

X - Initial of mother providing the quote

I - Comment by interviewer

#### *Theme 1. Loss*

Mothers typically described two stages of loss. Firstly, the loss they experienced with the birth and diagnosis of their child with intellectual disability, and secondly, and most relevant for this research, the loss that surrounded the terminal diagnosis and death of the child. This included, when possible, preparation for loss. With the exception of Rose's son



and Helen's daughter, the children had been undergoing medical treatment for some time and the mothers had, as much as possible, prepared for the possibility their child might die. Three of the children had been receiving palliative care. For Jackie, whose son Jake had a degenerative condition, the period of preparation was drawn out:

When we were first told the diagnosis all that you can remember really is the fact that it was terminal, and when you're first told that it's really difficult to get that out of your head. I mean it's like it's going to happen tomorrow. But gradually, as... I don't suppose you ever get used to the idea, but you learn to live with it and you realise it's not going to happen immediately (Jackie).

Participants also described making practical preparations that included finding out about and using hospice services, making funeral plans, discussing and planning the death itself (e.g., location, who might be present), discussing what might happen during and after the event (e.g., post mortem procedures) with professionals and family members, and spending as much time as possible with their child:

More than the others, when Nick was ill we actually made a conscious effort to go out and enjoy the world and we took him... you know, we flew him here, there and everywhere. We went to Disney Paris. I thought right, you're going to get a lot out of life boy (Joan).

Accepting, having control over, and understanding the situation was important for mothers, some of whom cited gender differences in their acceptance of and preparation for the death compared to their spouses:

S I always knew that it would happen, and my husband never did. He... once she'd come out of hospital he sort of thought, um, that they'd got it all wrong and

that she would just be okay (...) I always knew that this would happen, and I kind of always knew that that would be the way that I would find her one day.

I So in a way you were maybe coping?

S So in a way I was more prepared than he was (Sandra).

For some of the mothers, circumstances had forced them to prepare for the death of their child on more than one occasion. This in itself was stressful and traumatic, but it also made future acceptance of the child dying difficult. Also, despite having made arrangements, there were moments when mothers felt unprepared and not in control of events:

The rest I wouldn't do any of it differently, but this I would, was when the undertakers arrived to take his body from the house...And I didn't feel prepared for this I'm not sure if anyone could have prepared me but, I still... it was traumatic you know? (...) I wish I'd met them before, I wish they hadn't just come to take his body away (Toni).

The majority of the mothers were with their child when they died but for those who were not present, having been in control of their care made coming to terms with the death easier:

Actually he had died. It was rather sad, I wasn't actually with him but I was downstairs preparing. (...) we went up and he had actually stopped breathing and we realised that he'd died which was a shock then. (...) I wasn't crying. I knew it would happen and actually it was a relief because I'd just about decided he'd had enough (Jennifer).

Following the death of their child, grief symptoms reported by the mothers included anger, shock, disbelief, hopelessness, despair, guilt, vulnerability, and physical and mental

health impacts. Mothers spoke of the individual nature of the grief experience, and gender differences that made communication with spouses difficult.

We went to the group, a couple of groups, and he just didn't think that it was his cup of tea so I finished off the group on my own and that upset me so we rowed about that actually (...) And I think Barry thought it was a bit too heavy for him. I think it was a bit too honest and he couldn't handle...the honesty of it in front of other people (Jackie).

Mothers reported that their experience of bereavement was a unique one, set apart from their partners' experiences and from bereaved mothers of typically developing children. This was reflected in the support with which they surrounded themselves. Three of the mothers spoke of feeling that their child and their loss was undervalued by others because their child had an intellectual disability, including Joan, who was an adoptive mother to Nick and foster parent to Simon and Tracey, felt that her disenfranchisement was compounded further by people who did not understand or acknowledge her unique situation:

I remember when Simon died somebody actually said to Martin well it's not like losing your own. And you know, the other thing they say about children with disabilities is it was a blessed release wasn't it. And I want to go hang on. This was their life (...) this shows what you know about our children (Joan).

Feelings of grief did not disappear over time but did become more manageable:

I suppose, whereas when Joe first died, you know, I cried everyday. But then it went to sort of every other day and then you don't, you don't notice that that's happening but then you gradually think I haven't cried for a while about Joe now, and then you feel guilty about that because you think you should have. (Alice).

I feel um, I'm over the shock of his death, um... but, the grief doesn't change. The shock and the nearness does, but the grief doesn't change (Toni)

Further losses that accompanied the death of the child were the loss of the caregiving role, friendships, and support. The loss of the caregiving role was consistently discussed by all of the mothers. Having become full-time carers and developed a support network of friends in similar situations it was difficult to maintain or replace this activity and support source once their child died. Mothers talked about how they missed providing care to somebody and the world that came along with that:

You feel really, really isolated because the circles that you've been moving in with like the hospices, doctors, the nurses, school... it's like your whole world has collapsed. It's not just Jake you've lost; it's all of that as well. (Jackie)

The mothers were no longer members of that world and had a gap that needed to be filled. All of the mothers became involved in intellectual disability services following their loss and seven of the mothers now continue to maintain these links with the intellectual disability community through charity work, employment in special needs schools and services, caring for their other children with intellectual disability, and becoming foster parents for children with severe intellectual disabilities. This maintenance is often fuelled by a desire to pass on the skills their children taught them:

I don't want it to just go; I don't want to just waste it because there are so many people who wouldn't...who would never consider doing something like that and would never, ever look at a child like Julie because it would just be too scary.

Where it's not something that scares me any more (...) if I can help someone just get a break for a weekend or whatever, I'd really like to do that. (Sandra)



Keeping in touch with the intellectual disability community was important to all but some mothers found it easier than others to maintain links:

I worked as a classroom assistant in his primary school, just like a few hours a week, and then, after he died, even though he wasn't at that school anymore - he'd been left there four years. But after he died I just found it too difficult to go back there (...) I found it too difficult so I moved on and worked with older kids with special needs and then I left that because I just thought I can't do this anymore, I need to do something completely different (Alice).

### *Theme 2. Benefit Finding*

All nine mothers shared positive perceptions of parenting their child with ID and all reflected on some positive impacts of their death. With regard to the entire experience of raising and losing a child with intellectual disabilities, the mothers reflected on the opportunities their child opened up to them and others during their life. They also spoke of positive outcomes including education, priorities, inspiration, joy, pride, personal change, strength, and closer relationships with other family members. In terms of personal change, mothers reported character change for the better, and felt that they had become more mature, aware, insightful, caring, and empathic. For example:

Empathy: Following her son's death Rose was put on anti-depressants by her GP: "I think I'm more understanding to people with... to start with; someone with depression because I never thought there was such a thing as depression" (Rose).

Strength: But it doesn't worry me any more, you know; I'm quite happy... nothing really scares me to that extent as the thought of losing her, and that's... I've been there,

you know, and I've survived it, so it kind of makes you a much stronger person (Sandra).

Personal change: It's given a depth to my life I guess, and I think that's true of Michael before his death but just something around living with impairment. It made me really look at things differently, and it brought depth and much more reflection, and I would say that I grew up an awful lot through Michael (Toni).

The benefits that resulted from the child's life were also reported by some mothers to be found in siblings, other family members and the wider community:

My niece now, the one that used to come round every day, has got a job now in a, an adult... for adults with special needs, but only through Andy really, 'cos she found... she had so much joy out of Andy that she, she's finding now that she's, she's helping adults with special needs (Rose).

However, the mothers acknowledged that others often did not share their views of the benefits to be gained from people with intellectual disability:

To me they were perfect, but the rest of the world saw them as a little Mongol boy, the boy in the wheelchair, the little girl that couldn't talk. They never saw them as lovely, lovely human beings. And that... I think that's my sadness (Joan).

In addition to benefit found in parenting the mothers reflected positively on their loss and often viewed the entire experience as a positive one. Mothers reflected on the relief they felt now their child was free from illness; they were free from caregiving; and discussed a higher meaning associated with their experience. This higher meaning included a view that their child has lived and died for a reason:

But why she was taken away nobody will ever be able to understand because it, it's more spiritual. It's not just, it's not just something that's happened.

Everything happens for a reason and I'm sure that's how I dealt with it. Mostly was by saying this is why it happened because it's... It was a gift. Gifts don't last forever (Helen).

Although all reported positive outcomes, some found it more difficult to recognise these benefits in their experience. This was associated with needing time to see the benefits; a loss of spiritual belief; and describing the process as a waste of life. Sandra had experienced the most recent loss as her daughter aged 23 months had died ten months previously. In the interview she discussed the strength she possesses as a result of her experience, although she also mentions how she found it difficult to see any meaning in her daughter's short life:

She was full term. There was no reason for it to happen. So I think if you, if you know there's perhaps a chromosome abnormality or there's something that you can justify it, then it might be easier to reason and to, to carry on with. But there was nothing with her. And it was so totally unexpected. And then to fight every day and to lose her anyway, it kind of felt that there was no reason behind it; there was nothing...you couldn't sort of take anything from it (Sandra).

Although all discussed positive consequences, struggling to see the positive outcomes of the situation was reported by six of the mothers and was associated with their religious beliefs. Of the six mothers reporting a struggle to find meaning, one had always been an atheist and, although she has explored possibilities, has not changed her beliefs. Two had lost their Christian faith following the birth of their child, and three reported an increase to their Christian faith following the loss of their child. The three mothers who reported no struggle to find meaning reported having strong Christian faith, which remained largely unchanged throughout the birth, life and death of their child.



I never went through a stage that I thought, you know, why me or why, you know, why did God do this to me. Even though I wasn't a particularly religious person, every time Joe was in hospital and things weren't looking good, that's the first thing I'd do is say, please God, please don't let him die, please look after him. And, you know, you know, it... automatically you turn to it (Alice).

This complex relationship between finding meaning and personal beliefs could have important implications both pre- and post-loss not just in terms of mothers finding benefit in their experience but also with regard to sources of support and coping strategies. The issue of benefit finding also contributed to what mothers viewed as a paradox of loss and gain. Mothers felt this paradox has to be experienced to be appreciated and with increasing medical input into conception and termination people are choosing to avoid the experience. Mothers also discussed their feelings surrounding what they see as their ongoing campaign against the views of a society that undermines and ignores their bereavement experiences, and undervalues their children. There was a hope that their involvement in the present research might help to change these views in some way:

I believe very strongly that um, services should be different for disabled families, and I also believe around Michael's death has just taught me that we don't live with death, (...) and that's why I answered your thing, let's talk about this; illness, impairment and death, are things that affect us all. And if we can incorporate them in our life, we're going to be much better people (Toni).

### *Theme 3. Coping.*

Mothers used various coping strategies while their children were alive and after their death. The majority of the mothers talked of predominantly using activity-based problem-

focused, as opposed to emotion-focused, coping to gain control over their child's treatment in preparation for their death. Among the activities were furthering their education, using respite services, resolving conflicts with care providers, and planning the death. Although these activities may have incorporated an element of cognitive coping as the mothers had to first accept the inevitability of death to enable them to plan for it, only Jennifer spoke specifically of coping cognitively when preparing for her son's death:

And my way of surviving it was thinking well he could die and sort of facing that and then if he didn't die that was going to be great (Jennifer).

Two of the mothers coped spiritually through prayer in preparation for their child's death, whereas all of the mothers mentioned using spiritual or religious ways of coping after the loss of their child, including continued attachment to the deceased and belief in the afterlife. For Jennifer, formerly an atheist, her loss showed her the importance that belief in the afterlife holds for the bereaved and she began to explore her faith. She now finds comfort in the possibility that there is life after death, and that her son may have found release from the constraints of his disability:

I was definitely an atheist because I was going to die and it didn't matter but because it's not me, because it's my son that's the worst thing when it's your child (...) he had so much life I couldn't think of it being just turned off. I wanted to think that he was flying around; I used to think of him flying around in heaven. And the other big feeling I had (...) at last he's equal. He's the same as everyone else. (Jennifer).

Two of the mothers discussed having explored other beliefs and visited a clairvoyant and a spiritualist. Neither had a strong faith in what they had been told but both had been

comforted by the experience. This belief in doing what made them feel better regardless of the views of others was reiterated by mothers with reference to various types of coping.

And she did... she came out with a few things that, you know, just sort of... and even if it's crap, even if she was just clutching at straws and guessing at things, I don't actually care because I came out of there feeling better and feeling more hopeful... she made me feel better and that was what it was about (Alice).

After the death mothers tended to use a variety of emotion- and problem-focused coping strategies. It was important that mothers had the space and support to cope as they wanted to and sometimes this was at odds with the expectations of society and the needs of others. Mothers talked about issues surrounding disability with others and often experienced inappropriate replies:

Acquaintances have said, well, you know, it wasn't so bad, and, you must of known he was going to die and... but you don't expect the child to die suddenly like that, and because he was Down's they seem to think it wouldn't matter as much (Rose)

However, mothers tended not to talk about bereavement and death with others, feeling that it was not a topic for conversation. Having previously had their children and their parenting roles devalued may have influenced the decision to avoid talking about what they reported as taboo subjects. Mothers who reported experiencing devaluing attitudes also tended to report coping that was aimed at protecting others and masking their emotions:

Well I know from my own experience that it's not something that you would talk through to people unless they were professionals um because you don't want to upset anybody. You don't want to put people in a situation....My mother tends to

think that we must move on from these things. It's not healthy to talk about it  
(Helen)

Gender differences were also apparent in coping. Mothers reported that their husbands and sons coped differently to them and this made the emotion focused activities that mothers favoured, such as talking about feelings and looking at reminders, difficult:

I think it's hard... I think if I had daughters it would be easier but, because I've got sons, it's like they're like, you know I know not all men are the same but a lot of men are that they, you know, they try and sort of push it to one side and won't deal with it (Alice).

Jennifer, who had used positive cognitive coping strategies before her son died continued to do so after Josh's death using humour and also positive reframing:

And I used to say to people the way I get through it was to turn it on its head and I just used to keep telling us, oh we're so lucky we've had Josh for six years because I believed that we were and that the best way of looking at it was that we were lucky we had him for that long (Jennifer).

In terms of cognitive coping, four of the nine mothers tended towards avoidance and did things that prevented them thinking about their grief, and experiencing future loss. Activities mentioned included gardening, employment, avoiding emotional occasions, changing family life, and focusing on other children:

I couldn't speak to my sister at all, even though we're quite close and we're only just sort of getting back on track now. Because I had to pull away from her for quite a long time because she was just a complete emotional wreck and I couldn't handle having her around me because I knew I needed to get on with things  
(Alice)



However, not all activity stopped the mothers thinking about the loss. Four of the mothers coped by involvement in activities that focused on their loss. For Joan, this centred on raising her other children with special needs and adopting another child. Other problem-focused coping activities included writing, caring for the grave, organising the funeral and memorials, going to university, charity work, and working in intellectual disability services:

I've carved a place out for myself that says, yes, I do belong. You will not tell me

I do not belong (Toni)

There was no typical pattern of coping strategies. However, mothers who used activity-oriented problem-focused strategies before their loss tended to continue to do so, with the exception of one mother who had been prevented from doing so by illness. Resources and sources of support available also impacted the coping strategies used.

#### *Theme 4. Support Sources.*

The support sources mothers listed as useful and available differed greatly pre- and post-loss. At the time of diagnosis, during the child's lifetime and towards the end of life, mothers relied mainly on formal sources of support like medical services, educational services, hospices, and intellectual disability groups. It was important that these services were flexible and offered to parents, and for the majority of mothers, this was the case. Mothers also received informal support from their family, friends, and other parents. Intellectual disability groups in particular were valuable in various ways: as a source of advice, information, respite, and friendship, and they also played a role in preparation:

The [named] Society was our main help at the time and they actually held different parents' groups, like family days out and things like that where you could meet other families with children with the same disease and similar disorders as well.

So that really, really helped because obviously... because Jake's disease is degenerative you could see children through the whole of the spectrum really and it was quite frightening seeing what your child was going to become but it helped as well (Jackie).

Following the death of their child, mothers found formal supports were no longer readily available. Although some had follow-up contact with, and became friends with, professionals they met through hospitals and services, most of the mothers found continued contact was difficult and discouraged by medical service providers:

I mean I have stayed friends with a lot of them but because professionally they're not really supposed to keep in touch after, like you're off so that's all now. You don't matter anymore. That's really hard (Jackie).

The mothers had lost their role as a caregiver and were therefore no longer members of support groups. They had nothing to share with the other mothers and there was no formal source of support to replace the support lost, such as a bereaved parent group or continued contact with the hospital. Six of the mothers sought or took up offers of counselling; three found it useful, but an equal number found it unhelpful. Three of the mothers were not offered any bereavement counselling. Communal memorial services held at hospices for example, when available, were also a source of support to some, but not all, mothers. Following the death of their son or daughter mothers had to rely on informal sources of support including their extended family, community, and friends, although two mothers did mention feeling unsupported by friends and family who either did not understand or were too emotionally connected:

You know, you need to talk to someone who's not going to judge you for saying how, you know, how I felt when she was born; that perhaps it would have been

better if she hadn't survived. I know that people who know me would be just horrified to hear me say something like that about one of my own children (Sandra).

Throughout the interviews it became clear that the mothers were grateful for the opportunity to discuss their grief and the topics of death and disability, and were hopeful that their participation may help others. For some it was the first time they had the opportunity to discuss the issues with someone not directly related to their experience, and some queried whether other mothers had expressed similar sentiments, trying to ascertain whether they were alone in their experience. Being a mother made the experience different and some mothers experienced a lack of support from their spouses post-loss. They explained that this was a result of gender differences in grief, coping, and a lack of awareness surrounding male bereavement needs:

I mean my husband wasn't offered any counselling at all; he just went back to work and, you know, as men tend to do, just carried on. (...) He was aggressive and, and, and angry with everyone, with the world in general, and he didn't, he didn't speak to anyone about it really. I mean he's better now and he's talked to me, but I think he didn't want to off load on me because he knew I was going through my own sort of hell (Sandra).

Other bereaved mothers became a valuable source of support as the only people who understood exactly how they felt. Friendships with bereaved mothers sometimes replaced previous friendships networks and support groups:

I And did you find it helpful being around other bereaved parents?

J Yeah, yeah. Because you don't have to explain what you're feeling. They know. And everybody means so well when they say oh... I mean what do you say

to somebody that's lost someone so... Words are never enough are they? And so whereas I understand that from other people, you don't have to worry about that with someone else who's been through it because they just know instinctively (...) And it helps a lot (Jackie)

Additionally, the mothers spoke of the importance of sibling support and awareness of the need for counselling for both siblings and medical staff:

And [my son] only went for a little while but [the counsellor] was brilliant in counselling because she actually went into the school and explained that all the things that Peter was doing were completely normal, that that was, you know, to be angry with the world was a completely normal thing, you know, for him to be feeling (Alice).

Suggestions from mothers as to what they felt would be most helpful included counselling from similarly bereaved people, services that work at the family's pace, practical advice on what to do following the loss, and training for professionals:

So I guess, what I have more to say is to people who are supporting families in that situation and then I think, get your acts together, lose your fear of talking about this, and find ways to support families that are based around what families need (...) and professionals need, just like families need emotional support, professionals need emotional support (Toni).

### *Theme 5. Medical Relationships*

During the terminal care period, it was important for all families that their child received adequate, appropriate care. Mothers appreciated honest care professionals who respected their wishes, respected their child, and gave them viable options of care.



Inaccuracies in diagnosis, inappropriate comments, and care that was perceived as inadequate added to stress felt by the mothers:

Because I was saying that there is something wrong with this child and no one, I was knowing even at that early stage, that unless his seizures were controlled, he would die, but no one was meeting me in that conversation (...) And because that, because I was finding difficulties in having that conversation with professionals I was feeling I couldn't really have it with my friends or family (...) so it was a pretty lonely place to be (Toni)

Mothers who stressed the importance of honesty, communication, and conflict resolution with health professionals were more likely to discuss their child's needs. They recognised the need to prepare the child for their death, psychologically, as well as physically. Mothers recognised the relief and equality their child would receive in death. When mothers were able to discuss this, professionals responded more positively to them:

And all I cared about was looking after him, and people could see that, and they just responded to that and so they lifted their game, and after Michael died, some of the professionals came and said you know, wow, thank you for letting me be part of that (Toni).

Unfortunately procedures were not always discussed openly. Respect for the child's body, access to their child's body, and compliance with families' wishes regarding the autopsy, funeral home, and funeral were among the topics raised as important to mothers but not always understood by medical professionals, thus hindering coping for some parents:

[I said] You, you shouldn't touch somebody's child without their permission. You shouldn't do operations without signatures, and don't you think that it's about time that you pulled yourself together and thought about how you were

dealing with people because people will get very hurt by you touching their babies without their permission. So that part did definitely spoil what could have been, I wouldn't say a nice night, but a night of calm just for me and Jenny which never happened in the end (Helen).

Continued relationships with medical or educational services were an important resource, which were not available to all mothers. Continued contact with medical professionals facilitated communication regarding service improvements and aided benefit finding for parents, for example, educating the relevant staff on the impact of carrying out an autopsy without the parents' permission:

They take them away. You're not allowed to see them and it's not done. It's not how it should be done. It is against the law. So I, I hope that something good actually does come out of this to make people aware of it (Helen).

### Discussion

The findings of the current study have much in common with previous studies investigating the experiences of families where a child with an intellectual disability has died (Milo, 1997; Schormans, 2004; Todd, 2007; Wood & Milo, 2001). Similarities include mothers feeling they had experienced an undervalued and unique loss; the importance of control and preparation; the loss of the caregiving role; positive outcomes for parents; changes to spirituality; and the importance of honest medical relationships. These findings also indicate similarities between the parenting and bereavement experience in the UK and the USA. Comparison with earlier research (e.g. Milo, 1997) suggests that the last ten years has seen little change in mothers' perceptions of the value assigned by society to individuals with intellectual disability, despite a growing evidence for the positive contributions a child

with an intellectual disability has on family life (Hastings, Beck, & Hill, 2005b). Current research attention to bereavement experiences denotes some change with regard to the disenfranchisement of this group (Todd, 2007). However, the continued failure to recognise the needs of bereaved parents who lose a child with intellectual disability reflects the ongoing disparity individuals with intellectual disability experience with regard to healthcare (Scheepers et al., 2005), and services generally (McNally, 2004). Until people with intellectual disability are truly valued by society, their families are likely to continue to be marginalised.

The major difference between the present study and previous research is the focus on the post-bereavement requirements of the group in question. The reality of premature death in people with intellectual disability has been illustrated extensively by research (e.g., Janicki et al., 1999), and it is therefore imperative that health professionals have a clear picture of the post-bereavement needs of parents who lose a child with intellectual disability, and how these needs can best be met. The current study suggests that the pattern of coping strategies used by the mothers is more complex than utilising cognitive coping strategies (Milo, 1997). It also suggests coping strategies are additionally related to access to activity, support resources, spiritual resources, and the use of avoidance strategies. Further research investigating these relationships and the role of personality in coping (DeLongis & Holtzman, 2005) could be worthwhile. Mothers highlighted, in particular, the value of continuing the work their child had started by seeking out a role for themselves in the world of intellectual disability. This maintenance of role is potentially a powerful coping strategy for parents and a potentially important implication for service providers who could employ the skills of this highly motivated group.



Mothers also repeatedly mentioned the strength and support they received from other bereaved mothers who had similarly lost children with intellectual disability. There was, however, no formal organisation to facilitate contact with other parents whose child with a similar intellectual disability had died. This type of parent to parent support has previously been found to be valuable to parents by facilitating connections between parents who are experiencing a similar set of circumstances (Ainbinder et al., 1998). Connections are usually made locally, but in cases of rare or exceptional circumstances, connections may be made at a national or international level. Parents who have benefited from this type of support network include parents of children with intellectual disability, physical disability, and chronic illness (Ainbinder et al., 1998; Kerr & McIntosh, 2000; Poyadue, 1993; Santelli, Turnbull, Marquis, & Lerner, 1997). A similar service for bereaved parents of children with intellectual disability could be provided through an existing intellectual disability service provider, and evaluation could indicate its usefulness on a wider scale.

A service of this nature could also facilitate continued contact with service providers, which mothers felt was lacking after the death of their child. In the present study, continued contact with intellectual disability organisations allowed mothers to continue to use their skills and occupy a caregiving role, whereas, post-bereavement contact with medical care providers allowed mothers to have input into service provision, give medical staff feedback on their experiences, and generally ensure that their experiences (positive and negative) might inform medical care in the future. A programme to facilitate ongoing contact between a paediatric oncology team and families who had lost a child with cancer has been described by Russo and Wong (2005), and could similarly be implemented with bereaved parents of children with intellectual disability. It is also possible, however, that an ongoing relationship of this type may indirectly facilitate continued contact with the dead child (e.g., visiting



places the child visited while alive, maintaining friendships made via the child). Continued attachment with the deceased of this kind is similar to religious coping (Stroebe, 2004), and could be an important part of religious/spiritual coping for parents. The potential negative, neutral, and positive impacts of religious coping are, however, unclear (Benore & Park, 2004; Stroebe, 2004), and further research investigating whether or not religious coping is useful, and who it may be useful for, is necessary.

The present study also suggests that there is a need for increased awareness of the importance of the place of death. Bereavement support is most often provided by hospice services (Russo & Wong, 2005) and although hospices may be best equipped to provide bereavement support to families, people with intellectual disability will not necessarily die in a hospice as some will die suddenly. Also, predicting length of survival for individuals with various diseases and conditions, which result in premature death, is difficult and consequently individuals and families have difficulty accessing appropriate concurrent life-prolonging and palliative care services (Graham & Robinson, 2005; Nelson et al., 2000), particularly child-focused models (Donnelly et al., 2005). It is therefore important that individuals with intellectual disability have improved access to hospice care.

In addition, other medical professionals who may encounter bereaved families should have an awareness of the bereavement needs of families of people with intellectual disability, in addition to their knowledge of bereavement in general. For example, medical professionals should be made more aware of the possibility of the provision of palliative care and the death of a child with intellectual disability occurring in a single parent family, and be aware of the additional social support implications this might have (Harwood, 2007). A recent review suggests the traditional view of the risk parenting a child with intellectual disability poses to the marital relationship may have been overestimated (Risdal & Singer, 2004). However,

given the experiences reported by the current sample, further research, both quantitative and qualitative, examining the specific marital experiences of families of seriously ill children with intellectual disability may be worthwhile.

While some may see the potential for generalisation from this study to be limited, IPA allows the researcher to gain a valuable in-depth insight into the experiences of a small group of participants. Previous researchers have reported difficulty recruiting participants (Todd, 2007), but that was not encountered in the current study. On reflection, the issue of the importance of involvement in research was raised frequently by the mothers, and signifies that these parents are a willing participatory group who should be consulted more often. The study also highlighted variability in experiences (e.g., place of death, length and type of illness, marital situation) that point to the possibility of extensive variety between the experiences of parents who lose a child with an intellectual disability, which have not been captured by this study. In particular, research with groups of parents of children with similar conditions and prognoses could be beneficial (e.g., Mucopolysaccharide disorders). Furthermore, given previous findings on gender, religious, and ethnic differences in the fields of parenting (Aranda & Knight, 1997; Pelchat, Lefebvre, & Levert, 2007) and bereavement (Abramovitch, 2000; Wijngaards-de Meij et al., 2005), research in the UK to examine the experiences of mothers and fathers individually, and the experiences of parents from a variety of ethnic backgrounds, could give additional insight in the topic area. The use of quantitative measures, found to be reliable in previous research, could allow comparison within and between groups of bereaved and non-bereaved parents.

Limitations of the present research include sampling issues. As with all bereavement research, those who are most vulnerable may choose not to take part (Dyregrov, 2004).

Concerns surrounding confidentiality, anonymity, and the qualitative nature of the research

may have made parents more reluctant to become involved, particularly parents of children with very rare conditions. We do not know how many families initially received information on the study, or whether any of the service providers failed to disseminate flyers in order to protect parents from what may be viewed as a traumatic experience. This protective concern has been found by other researchers (Todd, 2007). However, the strengths of the current study include clear parallels with previous findings in the parental bereavement and intellectual disability field.

In summary, mothers who experience the death of a child with intellectual disability reported a lack of acknowledgement of their loss, a need to continue in their former caregiving role, and a lack of support from both society generally, and more specifically from medical professionals. Recommendations described above may improve the services and support currently offered to this vulnerable group. Service providers must focus not only on developing services that better provide for the palliative and bereavement needs of people with intellectual disability, but also the needs of their families who are at risk of being misunderstood and unsupported during this complicated and painful ordeal.

**Chapter 4 – Couples’ Experiences of the Death of their Child with Down Syndrome and a  
Congenital Heart Condition.**



### Abstract

Individuals with Down syndrome are at increased risk of co-morbid health problems including congenital heart conditions (CHCs), and mortality is higher in people with Down syndrome and a CHC than those with Down syndrome and no CHC. Consequentially, parents of children with Down syndrome and a CHC are more likely to outlive their child. The bereavement experiences of these parents have not yet been investigated by researchers. Semi-structured interviews were used to explore the experiences of six couples whose child with Down syndrome and a CHC had died. The interviews were carried out with each parent separately and analysed qualitatively using Interpretative Phenomenological Analysis (IPA). Four themes emerged from the analysis: “One disastrous diagnosis after another”; “We had to make a decision”; “We weren’t really going through it together”; and Ripples from the Child’s Life. There was a high degree of similarity of experience within couples. Differences between couples lay in their experiences, or lack of experiences, coping and supporting each other as a couple. Practical implications include the importance of considering the needs of individual couples, and especially to include support for fathers.

### Introduction

There are several research reports focusing on the experience of parenting a child with Down syndrome. Some data are consistent with the notion of a “Down syndrome advantage”, whereby parents of children with Down syndrome report better outcomes than parents of children with other disabilities report. For example, parents of children with Down syndrome report less stress than parents of children with other disabilities including autism, Prader-Willi syndrome, Williams syndrome, Smith-Magenis syndrome, and cerebral palsy (Hodapp et al., 2001; Hodapp, Ricci, Ly, & Fidler, 2003; Most, Fidler, Laforce-Booth, & Kelly, 2006; Pisula, 2007; Stores, Stores, Fellows, & Buckley, 1998). Although this apparent advantage is much debated (Cahill & Glidden, 1996), the most recent research supports this hypothesis, with a variety of advantages found for families of children with Down syndrome including lower divorce rates when compared with families of children with other birth defects or no identified disability (Urbano & Hodapp, 2007), less family conflict and more mother-child closeness in comparison with families of children with fragile X syndrome (Lewis et al., 2006), and more readily available support for families in contrast with families of children with fragile X syndrome (Poehlmann, Clements, Abbeduto, & Farsad, 2005). However, two areas that have received little attention are the joint experiences of couples raising a child with Down syndrome, and parental experiences associated with particular life events relating to the child with Down syndrome. In the present paper, the event we focus on is the impact on the family when an individual with Down syndrome dies.

First, we consider the potentially different experiences of mothers and fathers raising children with Down syndrome. When compared within couples, mothers and fathers have been found to use different ways of coping and finding support, with mothers showing higher levels of seeking social support, suppression of competing activities, turning to religion, and

emotional expression, than fathers (Spangenberg & Theron, 2001; Sullivan, 2002). Couples also experience different sources of stress. While fathers' stress related to the child's diagnosis of Down syndrome, mothers' stress has been found to be related to parental role, involvement in childcare, and father's stress (Cuskelly, Jobling, Chant, Bower, & Hayes, 2002; Roach, Orsmond, & Barratt, 1999). Pelcat, Lefebvre, and Perrault's (2003) qualitative study of parenting in Down syndrome outlines the importance of the recognition of roles, communication, and expectations within families. Pelcat et al. (2003) suggest that fathers' expectations may be more specific and more difficult to fill than mothers'. For example, fathers felt a greater need for support from male members of their extended families (Pelchat et al., 2003). While there are similarities between families with and without a child with Down syndrome such as maternal reports of an unfair division of domestic labour when both parents worked outside the home, difficulties faced are made more arduous by some of the additional stressors when raising a child with Down syndrome. For example, the child's care needs may mean that a considerable amount of time is consumed arranging specialist appointments and services (Pelchat et al., 2003).

Although the challenges faced by families of a child with a disability across the lifespan are recognized (e.g., internal family factors - adolescent transitions, and external factors – service discontinuity), this has largely been confined to maternal report of the adolescent years (Schneider et al., 2006; Todd & Jones, 2005). Research into parenting and Down syndrome across the life span is incomplete in several areas of knowledge. For example, researchers have rarely addressed the changing needs of families across the life span, investigated the experiences of those accessing support services for non-English speaking families, or explored the experiences of fathers of children with Down syndrome. An additional risk faced by families of children with Down syndrome that has the potential

for a large impact on family life is the premature death of the child (whether as a child or a young adult). Although there are data suggesting that this risk is significant, researchers have not addressed the experiences of family members adjusting to this loss.

There is widespread recognition of the increased health risks faced by people with Down syndrome. Individuals are at increased risk of developing dementia (Coppus et al., 2006), leukaemia (Sullivan, Hussain, Glasson, & Bittles, 2007), congenital heart conditions (CHC) and epilepsy (Day, Strauss, Shavelle, & Reynolds, 2005), and consequently have considerable healthcare needs, which are not always met (Melville et al., 2005). Almost half of all children born with Down syndrome are also born with a CHC (Hayes et al., 1997). All CHCs are to a greater or lesser extent operable, but CHCs that remain partially remediated or uncorrected expose individuals to the possibility of further health complications. As a result, children with Down syndrome and a CHC under the age of 10 are admitted to hospital twice as often as children with Down syndrome who do not have such conditions (Frid, Anneren, Rasmussen, Sundelin, & Drott, 2002). Although mortality for infants with Down syndrome and CHC is reducing (Frid, Drott, Olausson, Sundelin, & Anneren, 2004), mortality within the Down syndrome population is higher in individuals with CHC (Yang, Rasmussen, & Friedman, 2002), and parents are therefore more likely to outlive their child than parents of children with other disabilities. Precise statistics as to the likelihood of this event have yet to be reported.

Parental bereavement research within the general population has shown that the loss of a child through illness may result in wide ranging psychological implications for parents, including anxiety and depression (Kreicbergs et al., 2004; Leahy, 1993). With regard to gender differences and grief, comparisons within bereaved couples have found higher maternal grief (Goodenough et al., 2004; Schwab, 1996; Wijngaards-de Meij et al., 2005).



However, others have questioned the accuracy of particular measures of grief (Lang & Gottlieb, 1993), suggesting that measures are not adept at measuring male grief. Alongside this, the reactions of a couple to the loss of their child will constitute much more than a collection of grief symptoms and may include problems in the marital relationship (Schwab, 1992). Parents may also lose support from their spouse and other sources (Rando, 1985), but may gain from their experience, including acquiring increased personal strength, insight, and closer familial relationships (Laakso & Paunonen-Ilmonen, 2001).

Thus far, the majority of research on parental bereavement in general has focused either on maternal loss experiences, or compared parents with reference to gender. Research specifically on couples' experiences of the death of a child is limited. Exploratory investigations found that mothers took more time than fathers to recover and return to work when their child died from cancer (Sirkiä et al., 2000). Kamm and Vandenberg (2001) found that positive attitudes within couples about open communication were related to higher grief in the early stages of loss, and lower grief in later stages. Moriarty, Carroll, and Cotroneo (1996), found high levels of hostility in bereaved couples and more grief symptoms in mothers. Although these studies give some insight into parental bereavement and couples' experiences, the experiences of parents whose child with an intellectual disability dies are potentially qualitatively different to parents whose typically developing child dies (Reilly, Hastings, Vaughan, & Huws, in press). For example, parents and foster parents whose child with an intellectual disability has died report disenfranchised grief, experience additional losses including the loss of their defining caregiving role, and report a lack of continued contact with the formerly supportive world of intellectual disability services (Milo, 1997; Schormans, 2004; Todd, 2007; Wood & Milo, 2001).

For parents whose child with Down syndrome and a CHC dies, various factors may result in a different grief experiences in comparison with bereaved parents whose child had Down syndrome but no CHC, or had a different intellectual disability condition. Among explanations offered for the “Down syndrome advantage” are the readily available support groups because of the relative commonality of the syndrome, an extensive research tradition resulting in fewer unknowns for parents, and positive child characteristics (Seltzer, Krauss, & Taunematsu, 1993). Parents of a child with Down syndrome and a CHC may have spent significantly more time in hospital with their child, or following a regime of medical care, in comparison with parents of children with Down syndrome without CHC (Frid et al., 2002). These issues may impact on the bereavement experiences and bereavement needs of parents of a child with Down syndrome and a CHC who has died.

The aim of the present study was to investigate, using qualitative methods, the experience of parenting a child with Down syndrome and a CHC from the perspective of couples. We examined the retrospective accounts of both partners in terms of how they construct their experience of their child’s birth, diagnosis, life, and death. We identify similarities and differences that may occur between and within couples, and how these ways of coping and sources of support might change over time.

## Method

### *Methodological Approach*

Given that limited knowledge exists in the area of parental bereavement and Down syndrome a qualitative approach such as Interpretative Phenomenological Analysis (IPA: Smith et al., 1999), which aims to illuminate diversity in experiences rather than suppress difference (Jarrett et al., 1999), is particularly suited to investigations in this area. IPA is a

qualitative approach whereby the participant is deemed an expert on their own experience, and methods of interpretative analysis are used to uncover the central themes from participants' accounts of particular events. The aims of the current research are based around the bereavement experiences of couples. By allowing both partners to individually identify the symptoms, themes, practices, and outcomes that characterize their loss, IPA captures how participants themselves make sense of their and their partners' experiences, and by carrying out separate interviews participants in this study are free to do so without prompting or pressure from their partner.

IPA methodology tends to involve small somewhat homogenous samples of between six and eight participants. Small sample sizes allows the in-depth examination of participant accounts and facilitates more readily the identification of similarity and difference between accounts (Smith et al., 2002), although participant numbers vary from one to thirty (Brocki & Wearden, 2006). Couples completing separate interviews have previously been used as informants in IPA studies, generally in the context of caregiving spousal relationships (Clare, 2003; Newton-John & de C. Williams, 2006), and qualitative research has recognized the richness of experience provided by the inclusion of both partners from a dyadic relationship (Forbat & Henderson, 2003). In IPA, credibility checks, carried out by multiple researchers, ensure the themes are internally coherent and grounded in examples from the transcripts (Elliott et al., 1999). Additional tools sometimes used in IPA include: discussion with the research team, peer review, consultation with the participants on the accuracy of the findings, and the use of a reflexive journal. These tools aid verification and consistency, and ensure the quality of results (Carradice et al., 2002). The practice of sharing transcripts with participants is a much debated topic in qualitative research (Forbat & Henderson, 2005). Given that the



analysis of the interviews was an interpretative process in this case, the researcher decided it was inappropriate to ask participants to comment on her interpretation of the interviews.

An underlying principle of IPA is the influence that the researcher has on the interview process, for example, developing a relationship with the interviewee, guiding the direction of the interview, and interpreting the data. This influence is not only present but necessary to make sense of the personal world of the participant through interpretative activity (Smith et al., 1999). Additionally, an audit of themes, in this case conducted by a second researcher with considerable experience with IPA and qualitative data, ensures findings are reliably grounded in the data. The audit process resulted in some minor changes to themes, and analysis. To adhere to recommendations on conducting research with bereaved participants (Parkes, 1995), the researcher received training in working with bereaved individuals, kept a reflexive diary, and had access to clinical supervision throughout the study. Further guidelines outlined by Parkes (1995), which informed the study's development included: providing a clear written explanation of the project to obtain informed consent, and providing public access to findings regardless of the outcomes of the research. The study was granted ethical approval by the University ethics committee. All names have been changed to preserve participant identity, however information sheets clearly indicated that any data provided to the research team would be used in publications to arise from the project (See Appendices E and F).

### *Participants*

Recruitment was facilitated by the Down's Heart Group, a charity in the UK that offers support and information regarding heart conditions associated with Down syndrome. The charity forwarded information about the study to potential participants on behalf of the



research team (See Appendices A-F). All family members were invited to take part (See Appendices G and H). Although there was a minimum period since loss for inclusion purposes (the individual had died at least 12 months previously), no upper limit was placed on time since loss and no limits were placed on age of child or parent.

Table 4.1 Characteristics of the Participating Couples and their Children

Couple (age)	Child (age at death)	Time since death	Child’s diagnosis	Cause of death
Couple 1 June (60) Miles (65)	Pete (18)	10yrs	Down syndrome, AVSD <sup>1</sup> .	Septicaemia
Couple 2 Marilyn (62) Simon (76)	Gill (14)	7.5yrs	Down syndrome, Tetralogy of Fallot <sup>2</sup> .	Respiratory arrest
Couple 3 Diane (45) Gary (47)	Anthony (6)	8yrs	Down syndrome, Inoperable non- specific CHC	Heart failure
Couple 4 Jane (56) Carl (52)	Joanne (3)	19yrs	Down syndrome, Tetralogy of Fallot <sup>2</sup> .	Heart failure
Couple 5 Geraldine (42) Thomas (42)	Laura (2)	12yrs	Down syndrome, AVSD <sup>1</sup> and pulmonary atresia <sup>3</sup>	Contracted a viral infection following surgery.

<sup>1</sup> Atrioventricular septal defect: A hole in the heart wall between the top (receiving) chambers, and a hole between the bottom (pumping) chambers of the heart.

<sup>2</sup> Four problems: A narrowing between the right pumping chamber and the pulmonary artery; a ventricular septal defect; a thickening of the right pumping chamber; and an abnormal flow of unoxygenated blood around the body.

<sup>3</sup> The valve between the heart and the lungs is not formed.

Six married couples (see Table 4.1 for description), whose child with Down syndrome and a CHC had died, completed separate interviews (described below) with the researcher. The mothers' ages ranged from 43 years to 62 years ( $M = 53.33$  years;  $SD = 7.79$ ). The fathers' ages ranged from 43 years to 76 years ( $M = 56.33$  years;  $SD = 12.23$ ). The children were aged between 2 years and 18 years ( $M = 9.08$  years;  $SD = 6.33$ ), at death, and the mean time since death was 10.83 years (range 8 -19 years;  $SD = 4.06$ ). All parents were biological parents to the child who died, three of the couples had a daughter who died, and three had a son. All of the parents were English, with the exception of one Australian father, and one mother who came from New Zealand. All of the children were born in England, and all of the families were living in England when their child died. None of the couples had other children with disabilities, and only one couple (Geraldine and Thomas, couple 5) had given birth to additional children following the death of their child. Three of the children died in hospital following a period of medical intervention, and three were receiving palliative care when they died (one in a hospice, two at home).

### *Procedure*

The semi-structured interview was developed from questions used by Milo (1997), and to investigate the research aims in an unbiased manner the parents were interviewed over the telephone separately, in the order most convenient to the couple. The interview schedule was piloted with one couple whose interviews have not been included in this analysis. The pilot study resulted in some changes to the schedule. The interview schedule included extensive subsidiary questions and prompts should the parent find it difficult to discuss a particular area. The interview followed a chronological schedule covering the entire experience of giving birth to, raising, and losing a child with Down syndrome and a CHC.

Information was provided to the participants on the aims of the project and potential topics that would be raised during the interview (e.g., parenting a child with Down syndrome, how their child died, what support they received), but interviewees did not see a copy of the schedule (See Appendix J). By beginning with general questions about the family and the child while they were alive, parents became more relaxed, became familiar with the researcher, and were more at ease about telling their story. Research with bereaved individuals has highlighted the opportunity of telling ones' entire story as an important outcome of participation (Beck & Konnert, 2007; Burnell & O'Keefe, 2004; Dyregrov, 2004). The participant dictated the order in which the topics were discussed, and at the end of the interview participants were given the opportunity to raise any additional topics. All of the interviews were one-time interviews carried out over the telephone, and lasted 58 minutes on average (range 36 - 92mins). Carrying out interviews over the telephone removed geographical limits on participation, and has been found to be a useful method of data collection (Sturges & Hanrahan, 2004; Taneja et al., 2007). Also, bereaved participants have been found to rate telephone interviews as less stressful than non-bereaved controls (Taneja et al., 2007). The interview questions elicited strong emotions in participants, and the majority of participants became tearful at some point of the interview. The researcher was sensitive to individual needs and encouraged the parents to take their time. No participant requested that the interview end prematurely.

Written consent was provided by each participant following receipt of an information pack by mail (See Appendix I). In the majority of cases, one contact number was provided for both partners and on receipt of the consent form the researcher telephoned the initially available partner to answer any queries, ascertain consent, and arrange a time to conduct the interview. Permission to record the conversation was confirmed prior to the start of each



interview, and the interviews were recorded using a Sony™ ICD10-MX20 digital recorder. The researcher recorded demographic detail (See Appendix K), during the interview, and any unanswered questions were asked at the end of the interview. Afterward, the recorder was switched off and participants were given an opportunity to discuss the topics further. The majority of participants made additional comments at this time about the experience of taking part in the interview, and asked how the data were to be used. Comments made at this point were not included in the results. This point in the interview often provided an opportunity to arrange an interview with their partner, or arrange a time at which to make contact with the partner, which was usually within a few days. The interviewer did not review a participant's interview prior to conducting the interview with their partner. In the interview with the partner, the interviewer explained that their partner's comments would not inform the interview content or schedule, and where possible the interviewer avoided allowing her prior knowledge to impact on the interview. Participants were sent a joint letter thanking them for their participation, and were provided with a list of relevant support contacts should they wish to discuss issues further (See Appendix O). As the recruitment process was confidential, the total number of parents who received information about the study is not known.

### *Data Analysis*

All interviews were fully transcribed. To begin analysis the interviewer listened to, and read through the first transcript a number of times to familiarize herself with the interview content. The interviewer then began the process of making preliminary notes on the left hand margin of the page of the transcript while reading the transcript line by line. This process was repeated several times and emerging themes were noted in the right hand margin (See Appendix P). This process of reading and note taking continued until the interviewer



was satisfied that all themes had been identified from the data. A list of themes relating to the couples' experiences was drawn up from the notes and a list of relevant verbatim quotes corresponding to these themes was created. The process was repeated for each transcript, with the interviews of each mother and father pair analysed in turn. Each emerging theme was added to the list of themes and a master list of themes developed as emerging sub-themes were arranged into groups under master theme headings. For purposes of audit and validation, the second researcher read through each transcript, the list of themes and quotes, and the list of master themes, ensuring the emerging themes were visibly grounded in the data and none had been omitted. Discussion between the researchers resulted in minor changes to the arrangement of themes. The master theme list changed constantly throughout the analysis and write-up, with the addition and collapsing of themes into categories until a final list of four themes emerged. The themes are discussed below and illustrated with verbatim quotes from the interviews. For purposes of confidentiality, all names have been changed. When providing extracts from the interviews the following transcript conventions are used:

... Short pause

(...) Words omitted to shorten quote

[text] explanatory information included by author

## Results

Four themes emerged from an analysis of interviews with parents: "One disastrous diagnosis after another"; "We had to make a decision"; "We weren't really going through it together"; and Ripples from the Child's Life. Each theme will be discussed and illustrated with verbatim extracts from the interviews.

*Theme 1: "One disastrous diagnosis after another".*

None of the parents interviewed were aware their child had Down syndrome prior to the birth, and receiving a diagnosis of Down syndrome at, or soon after, the child's birth was difficult for all. This difficulty was quickly overshadowed by being informed about the seriousness of their child's heart condition, which occurred within 12 weeks of the birth in all but one case (where the heart condition was not diagnosed until the child was 18 months old).

Thomas recognized that there was no-one to blame for the trauma of the dual diagnosis, but it was difficult nonetheless:

I felt that the staff were actually very sensitive, but just by definition it seemed to be one, sort of, disastrous diagnosis after another. (Thomas, couple 5)

Where possible, the couples coped with the difficult situation together:

I was with her and the doctor came in and told us that, that she had Down's. He told us a little bit about the condition and but I don't think we were taking a lot in. And then he left and, you know, we sort of looked at each other and talked a little bit and Jane said, do you want to keep her. And there was no hesitation I wouldn't have even thought of it if she hadn't of asked. (Carl, couple 4)

Circumstances, however, did not always facilitate this preferred way of coping. For example Jane and Thomas described how complications with the birth had separated them from their respective partners:

And I felt very sorry for him because the first day I was really not with it. I kept phasing in and out so I'd wake up and then drop off and wake up and drop off, so it was a good day, I think, he was left completely on his own - well I wasn't functioning properly. (Jane, couple 4)

And I think what I remember is that feeling of being, sort of... you know, because my wife and I, together, are very strong, but when we're not together, I think it's, it's more difficult for each of us, so that, that whole experience, for both of us, we had to cope with that first 24 hours on our own, from the time the ambulance left to the, to the time that we were able to get my wife discharged the next morning.

(Thomas, couple 5)

The impact of the diagnosis of a potentially life- threatening heart condition that came after the diagnosis of Down syndrome was of considerably more concern to all parents, who recalled that the heart condition and related health problems became the focus of their joint worries and the diagnosis of Down syndrome became a secondary issue:

I have to say that in all of this that the heart thing took over completely from the Down's thing. The Down's thing just was an incidental, because we were so worried about losing her. (Jane, couple 4)

[The paediatrician] just said he thought he'd got a really serious heart problem. And so then things just progressed from there, and we had to go and see a specialist and everything. So that was really more of a shock, in the end, really, than having a child with Down's syndrome really. (Helen, couple 6)

Only two of the mothers worked (part-time) while their child was alive, and the others worked at home providing full-time care, however, responsibilities for achieving medical care were shared between partners:

And his condition deteriorated really quite badly, and we got the distinct impression from junior doctors that they would have let him die. We did have to

bring quite a lot of pressure to actually get him taken into intensive care. (Miles, couple 1)

Achieving medical care was not always straightforward and some parents discussed how they had to “kick up a stink” (Steve) to ensure their child received the medical attention they deserved:

I took him to the doctor, no, no, he's fine, he's just got a cold. And I did for four consecutive Mondays in that March and on the fourth time the doctor suggested it was me that had the problem, not Anthony. And he said really, there is nothing wrong with this child; I think the problem is you. I came away absolutely gutted and (...) took him to see a different doctor on the fifth Monday of that month and the doctor took one look at him and said he's got an enlarged liver, he's in heart failure. (Diane, couple 3)

What we found out was that it [examination of the heart] should have been done. I think they've changed all the rules...., [we] kicked up a stink, and they do it as a standard practice now when the kid is born. (Steve, couple 6)

The CHC additionally resulted in intense ongoing medical care of varied length in each family, meaning that parents spent a lot of time looking after their child, and visiting hospitals. This took its toll on both partners and the marital relationship.

Agreement as to the reality of this was high amongst couples:

Hospital was really hard. I'd never been in hospital before, and then I was never out of hospital. Carl doesn't like hospitals. It's hard to take, but I got used to it. (Jane couple 4)



Well, she was really difficult to look after because of the pain. She basically cried an awful lot, you know. It was frustrating not to be able to do anything for her, you know. I don't think in the first 12 months the two of us had a meal together. It was just holding her all the time. Trying to get her to sleep at night would take hours. It was hard going. (Carl, couple 4)

In summary, the experience of receiving the CHC diagnosis, and the subsequent care needs of their child with Down syndrome resulted in a life-long caring role for all mothers and fathers, which was fraught with difficulty but shared between parents. This joint responsibility took its toll on mothers and fathers and their marital relationship.

*Theme 2: "We had to make a decision"*

As a result of the involvement in care required of these parents, they had to make decisions jointly throughout their child's life surrounding their medical treatment, for example, whether their child should undergo a risky procedure. Such decisions were always agreed on jointly but a lack of information and time made decisions difficult:

When we went to see them about the heart, they said, she'd had a one in, a two in three chance of surviving the operation. And that was what I remember. And I said, terrible gamble, you know, there's a third chance that she'd die on the operating table. And it put us in a terrible state. So we, you know, we didn't know what to do or think, really. We had to make a decision there and I remember saying, it's like putting us in the position of God. (Simon, couple 2)

In addition to choices surrounding treatment, parents had to make decisions during their child's life, and following their death, on issues of parenting, education, autopsies, seeing the body, burial and funeral arrangements, and their own future. All decisions were

generally made jointly and parents tended to talk about the importance of making decisions together or at least having someone else to share the decision-making responsibilities:

But I think to a large extent we took it all in our stride because we had each other as well and I wasn't coping with it as a single parent or, you know, with a partner who just wasn't there or wasn't supportive. We faccd everything together as a couple so, you know, it simplified it. So, in that sense, we've been very lucky.  
(Geraldine, couple 5)

I think it was important to me. I still believe this, and I have done ever since, to have two people, not, not... even if it's not, you know, your, your partner, I think it's vital to have two people there. (Thomas, couple 5)

Sharing responsibility meant couples could share positive reflections on the experience:

Thomas- Geraldine was with her when she died, although it was during the night and, and Geraldine was asleep. But I, but I believe, overall, it was probably better that Geraldine was there, not, not that she wasn't there, if you see what I mean.

Interviewer - So you think it benefited her more to be there than to not have been?

Thomas- My belief is that the regret of not being there, for Geraldine, would've been more destructive. You know, whereas I think, with me, I, I, I felt that, you know, Geraldine was there so it, sort of, reassured me to think she was there.

(Thomas, couple 5)

However, couples also shared regrets surrounding decisions made such as having more children and medical procedures:

No, because we'd both said that we didn't want a post mortem. In retrospect I wish there had been. I mean, they say that he died because there was heart and lung failure, but I don't know for sure really. You know, nobody really knew why he was in hospital or anything. I just wish now that there had been a post mortem, but at the time you don't want to go through that. (Helen, couple 6).

When he died I said, there's no way you're doing an autopsy on him. There was no way they were going to cut him open, but I wish they had have done it now. (Steve, couple 6)

Making decisions was difficult but facilitated by good communication between partners while their child was alive. After death, however, grief created a new barrier between the couples and they reported increased difficulties with regard to coping and decision-making.

*Theme 3: "We weren't really going through it together".*

The couples formed two distinct groups with regard to whether they coped with their grief together as a couple, or separately. Although the partners were interviewed individually, two of the couples spoke of difficulty comforting each other in their grief, gender differences in grieving causing problems, and additional losses that may have confounded the grief experience:

I suppose at the time what made it worse was that we were both going through it, we weren't really going through it together; we were going through it apart and rubbing up against each other. (Carl, couple 4)

I was foul, really foul. I didn't want to be there. I wanted to be back in my old house, with my old friends. Because I knew no one up here, apart from family and they were saying don't get upset. And it was a complete nightmare. (Jane, couple 4)

Well, when we got people coming round they were basically more interested in, or I thought they were more interested in how Helen felt. You know, friends, and everything .... it just seemed to me I was left on the outside, you know, like, because when he died people kept knocking on the door, and how's Helen, and they didn't seem to bother with me. (Steve, couple 6)

Both of these couples discussed additional losses including financial worries following the death, illness, the impact of grief on siblings, and lost support networks. However, it was the topic of having more children after their child had died that became a key focus of the interviews with both mothers. In addition to their child dying, Helen and Jane lost a caregiving role they had expected would continue for years to come, and this was made more acute by an inability to have more children and recover this role:

I was sterilized when my youngest was two (...) I thought it was my responsibility to have it done (...) when Billy died, I really was angry about that, because even though we were [age] when Billy died, it would have been nice to have the choice. I know you can't replace your child or anything, but it was my way of life, bringing up children. (Helen, couple 6)

...and realizing that you're not going to have any more children as well, that was really really difficult. Because I wanted to try straight away, but Carl didn't. I don't know how things would have worked out, had I got pregnant straightaway, but it



would have had to have been straightaway, because it was too late later on. I don't really know. You can do the, if only, forever. (Jane, couple 4)

These additional losses were potentially responsible for increased communication difficulties and problems in the marital relationships:

So the menopause thing merged into the grieving, and they kind of fed on each other, probably. Talking about it now, I don't know why Carl is here, actually. Why he didn't clear off with [our daughter] I don't know. (Jane, couple 4)

In contrast, the other four couples talked about how everyone experiences grief differently, and that this diversity could not be attributed to gender difference alone. These coping differences required sacrifice and patience, and it was the belief of four of the couples that tackling their grief jointly had brought them closer:

Well, we've always been very good friends and we've just... we spent the time together just talking, you know, about Laura, about what we were going to do. I think Thomas found it harder to relate to other people or have contact with other people during that time (...) I'm sort of more easygoing about that sort thing, but my main priority, my main concern then, was, you know, what Thomas needed (...) And I think, looking back now, I think that it helped, you know. It helped because I think we talked about a lot of things then that, had we both been working and we both dashed back to work after a month or whatever, I don't think we would ever have talked about. (Geraldine, couple 5)

Coping with their grief was not an easy task and one parent highlighted the difference that being bereaved as a couple made to his experience by comparison with a previous loss:

But, having lost parents, there's a huge difference between losing a parent and losing a child, because if you lose a parent your wife isn't bereaved in the same

way, if it's your parent. They may have been fond of the person, but they can actually be involved as a slightly outside person to help you through it, but when you're a couple who both lose a child, it's almost impossible to be able to do anything without being aware of the impact on the other person. You know, even, for example, and it's, it's not an issue, but I was just trying to give you a clear example, even taking part in this research. As it turned out, we both were keen to do it, but if one was keen to do it but the other one would be upset by it, even the thought of the partner doing it, it, it is different because you are bereaved as a couple, and that's true all the way through. (Thomas, couple 5)

The death had changed the lives of both partners thus they had to rebuild their lives and readjust together not separately:

So I think it was something which mostly we coped with, Miles and myself, and then increasingly [our son], as we felt able talk about it, and readjusted, in a sense, to the loss of David, and being a three rather than a four. (June, couple 1)

By sharing their emotions and supporting one another the partners were less dependent on external sources of support and were the main source of support for each other:

But we helped one another, you know. I mean, because, you know... I knew that Marilyn had, you know, had lost Gill and she knew that I'd lost Gill, so we helped one another. Well, we didn't think or consider it as help, you just, you just fall on one another's shoulders and sob. (Simon, couple 2)

This was in contrast to Jane and Carl (couple four) and Helen and Steve (couple six) who had found their grief difficult to cope with jointly. These couples highlighted the need for support from other sources: family, friends, doctors, other bereaved parents; counselling, and Down syndrome support groups:

So she (...) had no real support. It wasn't until after about a year when we finally found a house and moved in and she got a doctor who said, well, what counselling did you get. The answer was none. She finally got some help. I really regret that we did that. (Carl, couple 4)

You can't try to do it on your own, you can't. We got into the Down's club, at [place], and the Down's Heart, they've been really good. (Steve, couple 6)

The majority of parents were not offered bereavement counselling when their child died. Of the two mothers who did receive formal counselling, Jane found it useful, and Geraldine found hers less so. Geraldine's husband Thomas requested, but didn't receive any, counselling through his local GP surgery and consequently felt let down by services:

And Thomas, unfortunately, had a worse experience and never really got any counselling at all. And I felt he was let down; he was let down. It often takes a lot of courage to ask for help, to get to the point of asking for help and then, when you do, then he really had to sort of fight to get some help and then never did. And then that in itself creates its own stress so he was let down and it wasn't fair. (Geraldine, couple 5)

Of the couples who described sharing their grief, some talked of acting as one another's counsellors and found this to be a useful way of coping:

And I do believe this, I mean, I know, you know, it's a bit of a, sort of, odd way of looking at it, but if you have a very close relationship with your partner, which, which I have, and we are very close friends as well, then it may not be the same, but in a certain sense you have got that in your life. It may not be as formal but,

you know, in a certain way you do, sort of, talk through everything gradually anyway. (Thomas, couple 5)

However, comments such as those made by Helen below, outline the need for counselling is one that is unmet in some cases:

I work in a special school now (...) and actually about six months ago we had a training day where a few people came in from the (...) hospice thing in [place], some of our children go there, and they did a talk on bereavement and things, and they were saying what they do and what counselling they give and that. I did, I did feel like perhaps I wished I'd had, but it was never offered to me. (Helen, couple 6)

#### *Theme 4: Ripples from the Child's Life*

Although the death of their child had brought negative repercussions including financial worries, physical and mental ill-health, breakdown in familial communication, and lifestyle changes, parents discussed the positive ripples that had spread out from their child's life touching both family members and outsiders:

David's life touched on lots of other people, and a surprising number of people have been influenced by David. (...) There are friends who've looked after David, who've subsequently gone into special education, into clinical psychology, specialising in learning disability. He's obviously influenced June's current work, so all these ripples have gone out from David's life, and that helps to make sense of... sense of David's life. (Miles, couple 1)

The parents explained a variety of additional positive outcomes of their child's life including relief, career change, personal strength, confidence, patience, sensitivity, and



perspective. These positive outcomes, were for most parents a result of the entire cyclic experience:

I perhaps have realized that I am a stronger person and the things that I have been through, if someone else was telling me about them, I'd be thinking, my goodness. But somehow you do... there's like an inner strength that rises up and you do, as I say, you find it within you to cope with these things. And so I guess I look back and I think, gosh, you know, we have been through a lot and we have come out the other side, you know. (Diane, couple 3)

However, some of the fathers differed from their partners and from each other with regard to how they viewed the impacts of their child's lives. Thomas stressed that the change for the better that he and his family experienced was an outcome of his daughter being born not dying:

I believe it was Laura being born that changed and not Laura dying, although obviously, when she died it devastated us. But I think our lives changed when Laura was born. (Thomas, couple 5)

Although he did go on to recognize the positive impact that her death also had:

I think we're the only couple I've ever met who are so heavily involved together. I was heavily involved with the play school as well, and it isn't, like, just a token appearance, you know, where you think, well, I'll have a day off work and go. It's, it's, like, you can live that life as much as, as you can. And I think that's possibly something to do with Laura dying. It makes you unapologetic for, for making the most of every opportunity. (Thomas, couple 5)

Also, two of the other fathers expressed an opinion that the experience of raising and losing their child had confirmed who they were, and maintained the positive outlook they already had:

Interviewer - Has your experience affected your outlook on life?

Miles - No, I don't think so, really. I've always been quite pragmatic about most things, so I always start from where we are, not where we might have been. I've always had quite a positive outlook on the future, and I don't think it's significantly changed that. (Miles, couple 1)

I think yeah, that. I think it's confirmed who I was, really. I don't think it's changed me that much. But, you know, hopefully, it deepened me or something and made me aware, more aware, that life is very fragile, but I think I knew that anyway.

(Simon, couple 2)

Most importantly for the couples, however, was the impact the experience of raising and losing a child with Down syndrome and a CHC had on their marital relationship:

I mean, you often hear of families with handicapped children sort of broken up as a result of that child being born, or dying, or just the sheer difficulty of living. But in our case I think it brought us even closer. (Marilyn, couple 2)

I think that pain like that either blows you apart or pulls you together. I don't think... I mean, I don't... I'd hate to be too philosophical about this, but I don't think you could be unchanged by it. I'd say, yeah, we're definitely closer. But I mean you hear of people who are blown apart by it, you know. They can't take it. The man walks out. You know, it's too, it's too much for him. (Simon, couple 2)

A positive outcome had been achieved despite tribulations along the way and even the couples that had encountered some difficulty in coping with grief as a couple reported this:

It's not something that wouldn't affect anything. So yes it affected us for all the good and all the better or all the worse. I don't think it changed it probably. It changed both of us the way we think of each other and think of life and things I suppose. I mean I became much less driven by work after that. The only thing that matters now is Sal and Jane. (Carl, couple 4)

### Discussion

The findings of the current study give insight into an area of parenting a child with Down syndrome that has been largely overlooked by researchers. Qualitative and quantitative methods have been used to investigate parental experiences of parenting a child with Down syndrome across the life span, but this research on the topic of the death of a child with Down syndrome and a congenital heart condition (CHC) is the first of its kind. The interviews with couples highlighted issues which are uniquely pertinent to this group, namely: receiving multiple diagnoses of Down syndrome and a CHC, the more considerable threat carried by the CHC diagnosis, and a role in decision-making surrounding medical treatment such as whether or not their child should undergo heart surgery. Parents recognized the positive outcomes or ripples that have resulted from their child's life in a similar way to those described in previous research with bereaved biological and foster parents whose child with an intellectual disability died (Milo, 1997; Schormans, 2004; Todd, 2007). Also reported were difficulties similar to those found in previous intellectual disability and bereavement research with regard to their own and their partner's coping needs (Milo, 1997; Wood & Milo, 2001).

Specifically, the current study allowed both partners to express their opinions and the analysis of accounts generally revealed similar views within couples, and issues that potentially confounded the grief experience, such as an inability to have more children, or a lack of open communication with one's partner. Such issues may be closely related to marital satisfaction, a construct which is at the centre of research into cognitions in marriage and marital distress (Fincham, 1994). Parents in the current study discussed the importance of joint decision making with a partner, the different experience that comes from being bereaved as a couple, and an awareness of the potential for marital discord following bereavement. Marital deterioration and breakdowns in communication were reported by some parents, and previous research has reported these as a potential outcome in some marriages following the death of a child alongside marriage breakdown, a decrease in sexual intimacy, and conflict and anger (Oliver, 1999).

Differences in grief symptoms according to gender previously found by bereavement researchers have been questioned as potentially no more than a function of grief measures insensitive to male ways of grieving (Lang & Gottlieb, 1993). Although some couples in this study reported differences in grieving, which they attributed to gender differences, the majority of parents felt that the differences in grief could not be explained by gender differences alone. Also, in reporting benefits found in their experience, some parents reported positive outcomes in terms of their marital relationship. Although these views seemed to be reflected by both partners, further research is required. Quantitative research with both mothers and fathers of children with Down syndrome and a CHC could investigate grief experience, marital outcomes, and their contribution to adaptation in parental bereavement.

The current research raised issues not yet fully attended to by intellectual disability researchers one of which is the reality of receiving a second, potentially fatal, diagnosis for a



child already diagnosed with Down syndrome. Parents discussed matters surrounding the timing of the diagnosis, how the diagnosis was a second loss, and how the threat posed by diagnosis of Down syndrome came second to the physical health needs of their child.

Previous research in Down syndrome and other disabilities has illustrated the diversity of attributions of diagnosis that parents may hold and the diverse outcomes attributions of blame may have for parents (Dale, Jahoda, & Knott, 2006; Hall, Bobrow, & Marteau, 1997).

Research with parents of children with Down syndrome has investigated parents' experiences of the diagnosis itself, and Hedov and colleagues (2002) illustrated findings of dissatisfaction with the timing of the diagnosis, insufficient support and information, and too much negative information given to parents. Research in other areas of intellectual disability has confirmed this need for clear information and communication on the part of healthcare professionals involved in diagnosis (Graungaard & Skov, 2007). In this particular situation, parents experience a double diagnosis that brings with it decision-making responsibilities, but may also result in attributions of blame. The impact of both may make coping more difficult and add to parents' emotional responses.

Guilt, anger, and hostility have been investigated by bereavement researchers. Post-bereavement perceptions of inadequate healthcare have been found to be related to higher guilt in parents whose child died from cancer (Surkan et al., 2006), parents who experienced a sudden death were more likely to feel anger towards the child who died (Drenovsky, 1994), and couples who have experienced the sudden death of a child have shown high hostility (Moriarty, Carroll, & Cotroneo, 1996). In the current sample feelings of anger and guilt may have been complicated by the parents' involvement in the decision making process, and may be further confounded by experiences of disenfranchised grief (Attig, 2004), whereby others do not view their grief as valid or appropriate because their child had an intellectual disability

(Milo, 1997; Reilly et al., in press; Wood & Milo, 2001). Guilt, anger, and hostility were not explicitly explored in the current study but are potentially important topics for future research.

In the realm of bereavement research it has been suggested that qualitative methods may be used to explore novel subject areas, which can be refined and probed further by quantitative means (Neimeyer & Hogan, 2001; Owens & Payne, 1999; Parkes, 1985; Stroebe et al., 2003). Areas uncovered in the current study that have potential for further exploration include the use of additional outcome measures such as those investigating guilt, anger, and hostility. Research has already identified bereavement-related-depression and complicated grief as distinct from depressive symptoms (Prigerson et al., 1995a). Other grief symptoms could be similarly scrutinised and may provide indicators of the long-term implications of pre-bereavement experiences such as the process of diagnosis, and decision-making. Such constructs may also be useful in the development of more up-to-date methods of assessment of grief symptoms and outcomes (Neimeyer & Hogan, 2001).

In terms of bereavement interventions for parents of children with Down syndrome, the current research and sample has the potential to inform practice and research in this area. The effectiveness of traditional bereavement intervention has been scrutinised and found to be lacking (Jordan & Neimeyer, 2003; Rowa-Dewar, 2002). Following a review of intervention evaluations, Jordan and Neimeyer (2003) concluded that the paucity of intervention efficacy may be a result of a lack of specificity in research design regarding duration, timing, precision, and group composition. In particular, the lack of attention paid to moderator variables such as gender, time since death, and risk of complicated grief status must be addressed to achieve accurate reports of intervention efficacy. Any intervention with bereaved parents of children with intellectual disabilities should take these variables into

account and the practice of comparing outcomes by group (e.g., mothers of children with Down syndrome, mothers of children with Mucopolysaccharide disorders), and time since death, should be considered in future research. Within those parental bereavement intervention studies deemed methodologically sound, interventions have been found to be most useful for mothers and those who are the most distressed (Murphy et al., 1998; Murray, Terry, Vance, Battistutta, & Connolly, 2000). However, given the depth of experience described by fathers in this study it is imperative that interventions are tailored to paternal needs also. Suitable interventions for this group could be developed with reference to previous studies where extensive detail on procedure is provided (e.g. Murphy et al., 1998).

Given the individuality of the grief experience and the suggestion from the current study that not all parents will seek or require intervention after their loss, bereavement counsellors should tailor their services to suit the needs of the individual, or couple who need assistance. By focusing more on treatments that have been found effective and applying them in a flexible manner therapists can develop a model which best fits their clients' needs (Beutler, 2000). Interventions should also be informed by experience, and more extensive formal assessment of the client's marital relationship, personality, confounding losses, and coping style may help those providing bereavement intervention make informed judgments, in a move away from the "one size fits all" approach (Jordan & Neimeyer, 2003). Parents in the current and previous studies reported the benefits of support from similarly bereaved parents (Milo, 1997). Service providers could become involved in bereavement intervention by facilitating support groups for parents. Additionally, couples who report communication breakdowns and marital problems may benefit from a cognitive intervention based on changing cognitions or teaching new behaviours in marital relationships (cf. Fincham, 1994).



The current research provides an in-depth insight into the experiences of bereaved couples whose child with Down syndrome and a CHC has died and provides a starting point for future research. However, we do not know how many couples chose not to take part, and therefore it is unknown how representative these couples are of the wider group of similarly bereaved parents. Also, the putative effect of the “Down syndrome advantage” is unknown. To what extent might the relative commonality, characteristics of the child, or stereotypical views of the wider public of people with Down syndrome impact on the availability of support for parents? Research involving groups of parents whose children with other and more infrequent intellectual disability related conditions have died, would help illuminate the similarities and difference in experience.

In conclusion, although small in scale, this qualitative study identifies some important areas for future research and clinical practice. It highlights the importance of the process of diagnosis and decision-making, and suggests that improved and increased awareness on the part of healthcare professionals could have valuable implications for parents. As seen in previous research the study encountered positive perceptions on the part of parents. However, the current study additionally uncovered perceptions of how the bereavement experience may differ as a result of being part of a couple, and having a child with both physical and intellectual disabilities. The marital relationship may be impacted by positive and negative influences throughout the bereavement experience and healthcare and bereavement professionals should be aware of both the individual needs of each parent, and the mutual support partners can provide to each other.



**Chapter 5 – Study 3: Down Syndrome and Mortality Related to Congenital Heart Condition:  
Maternal Experiences of Bereavement.**

### Abstract

Individuals with Down syndrome and a congenital heart condition have considerable health needs, and as a result their parents may be more likely to experience the death of a child. Measures of maternal grief, coping, and positive perceptions were completed by 38 mothers whose child with Down syndrome and a congenital heart condition had died. Mothers reported symptoms of grief up to 23 years after the death of their child, and mothers' reports of grief were unrelated to the age of the mother or child, or the child's gender. Mothers who had higher grief scores reported more frequent use of active avoidance coping strategies, and had more positive perceptions relating to the life of their child with Down syndrome. Regression analyses indicated that active avoidance coping accounted for a considerable amount of the variance in grief scores. Findings are discussed in terms of possible explanations for grief intensity, and implications and suggestions for interventions aimed specifically at this group.

### Introduction

Traditionally, researchers investigating the impact that raising a child with intellectual disability has on parents have compared individuals with intellectual disability of mixed aetiology with parents of typically developing children (Dyson, 1991). More recently, however, research has become more phenotypic in focus, investigating how characteristics of particular syndromes may relate to certain responses in parents (Abbeduto et al., 2004; Dykens & Kasari, 1997). When compared to parents of typically developing children, parents of children with Down syndrome may experience more stress (Hodapp et al., 2001), and greater anxiety and depression (Spangenberg & Theron, 2001). However, when parents of children with Down syndrome are compared with parents of children with autism, psychiatric disorders, and other conditions (e.g., Williams syndrome), they have been shown to report less stress (Stores et al., 1998), less depression (Abbeduto et al., 2004), greater satisfaction with social support (Seltzer et al., 1993), less pessimism (Abbeduto et al., 2004; Fidler, Hodapp, & Dykens, 2000), and less family conflict (e.g., Lewis et al., 2006).

Families of people with Down syndrome experience various challenges throughout the life-span including difficulties accessing adequate healthcare services (Melville et al., 2005). This difficulty occurs despite the widespread recognition that people with Down syndrome are at increased risk of developing dementia (Coppus et al., 2006), leukaemia (Sullivan et al., 2007), congenital heart conditions (CHC) and epilepsy (Day et al., 2005), and consequently have considerable healthcare needs. For example, children with Down syndrome and a CHC under the age of 10 years have twice as many hospital admissions as children with Down syndrome who do not have such conditions (Frid et al., 2002). CHCs occur in almost 50% of newborns with Down syndrome. Atrioventricular septal defect

(AVSD), patent ductus arteriosus, ventricular septal defect (VSD), atrial septal defect (ASD), and Tetralogy of Fallot are the CHCs most commonly seen in individuals with Down syndrome. All of these problems are to a greater or lesser extent operable, but if not completely corrected they can leave individuals at risk of further health complications and hospital admissions (Frid et al., 2002; Hayes et al., 1997). Consequently, mortality within the Down syndrome population is higher in individuals with CHC (Yang et al., 2002), and parents of individuals with Down syndrome and CHC may therefore, outlive their child. Precise statistics as to the likelihood of this event have yet to be reported.

The death of a child typically results in grief that is longer lasting, more intense, and more painful than any other loss (Leahy, 1993; Rando, 1985; Rubin & Malkinson, 2001). Parental bereavement research has shown that the death of a child may result in wide ranging psychological implications for parents, including anxiety and depression (Kreicbergs et al., 2004; Leahy, 1993), and physical implications such as increased risk of cardiovascular disease (Li et al., 2002). Grief responses in parents who lose a typically developing child vary, for example, according to how the child dies (Dyregrov et al., 2003), the age of the child at death (Moss et al., 1986; Rubin & Malkinson, 2001), and the availability of support (Davies et al., 2004). However, little is known about the impact of child characteristics, such as having an intellectual disability, on parental bereavement.

Four qualitative studies (three in the USA, and one in the UK), have investigated the bereavement experiences of parents of children with intellectual disability (Reilly et al., in press). All of these studies report on the double losses (loss of the perfect of child at birth and the actual death of their child), that parents experience, and indicate that while these parents form a unique group in terms of their experiences, their losses are not always acknowledged by medical and bereavement professionals (Milo, 1997; Schormans, 2004; Todd, 2007;



Wood & Milo, 2001). Issues highlighted by the studies include ways of coping; the impact of grief symptoms; the availability and use of support; relationships with medical professionals; and positive outcomes. In two of the studies (Milo, 1997; Wood & Milo, 2001), completion of the Grief Experience Inventory (GEI: Sanders et al., 1978) indicated that grief profiles of the participants did not differ significantly from those of other bereaved parents. However, the absence of other quantitative measures, lack of specificity or lack of information on intellectual disability related conditions, and the small sample sizes in all four studies, lead to difficulties comparing these experiences to previous findings on parental bereavement, and findings on coping and adjustment in parents of children with Down syndrome.

We investigated whether relationships exist between maternal grief outcomes, coping processes, personal resources, and characteristics of the loss. Based on previous research we expected reports of grief intensity to reduce as time since death increased (Kreicbergs et al., 2004). Research investigating the impact of avoidance and problem focused coping on grief typically suggests that avoidance behaviours may be related to poor long-term adjustment for bereaved individuals, while problem focused coping and seeking social support are beneficial (Bonanno, Papa, Lalande, Zhang, & Noll, 2005; Hays, Kasl, & Jacobs, 1994). In the present study, we explored the relationship between avoidant and problem-focused coping strategies, and reports of grief intensity for mothers of children with Down syndrome and CHCs.

Previous research also suggests that disability-related positive perceptions may act as a coping mechanism for mothers of children with an intellectual disability (Hastings & Taunt, 2002). Therefore, we explored whether positive perceptions serve a similar function for bereaved mothers.

## Method

### *Participants*

The sample consisted of 38 mothers whose child with Down syndrome and a CHC had died. The diagnosis of Down syndrome and CHC was confirmed by maternal report only. Participant characteristics are described in Table 5.1. All were the primary caregiver and were biological mothers to the deceased, with the exception of one biological grandmother, and one adoptive mother. The majority (95%) of children were living at home when they died and in 63% of cases a daughter had died. All of the mothers were living in the UK when their child died and 95% ( $n = 36$ ) were British. The remaining participants were from Russia and New Zealand. The majority of participants reported a Christian family background, but current religious affiliations were not explored in any detail. Sixty-six per-cent of the mothers were in paid full- or part-time employment, and 16% had other children with special needs at the time of data collection. The CHC was not specified or unknown in 44% of cases, but the breakdown for specified CHCs where known is summarised in Table 5.1.

Table 5.1 Maternal Age, Child Age, Time Since Death (in years), and Rates of Specific Congenital Heart Conditions.

	Range	M	SD
Age of mother	28-79	51.41	10.43
Age of child at death	0-32	8.61	9.37
Time since death	1-23	10.30	6.37

Congenital heart condition	N	%
CHC not specified	17	44
Atrioventricular septal defect (AVSD) <sup>1</sup>	13	34
Tetralogy of Fallot <sup>2</sup>	3	8
Mitral stenosis <sup>4</sup>	1	3
AVSD and pulmonary atresia <sup>4</sup>	1	3
AVSD and Tetralogy of Fallot	1	3
Ebstein's anomaly <sup>5</sup>	1	3
Ventricular septal defect <sup>5</sup>	1	3

<sup>1</sup> A hole in the heart wall between the top (receiving) chambers, and a hole between the bottom (pumping) chambers of the heart.

<sup>2</sup> Four problems: A narrowing between the right pumping chamber and the pulmonary artery; a ventricular septal defect; a thickening of the right pumping chamber; and an abnormal flow of unoxygenated blood around the body.

<sup>4</sup> A narrowing or obstruction of the valve between left receiving and pumping chambers.

<sup>4</sup> The valve between the heart and the lungs is not formed.

<sup>5</sup> The valve between right receiving and pumping chambers is abnormally formed.

<sup>5</sup> A hole between the bottom (pumping) chambers of the heart.

### *Measures*

Mothers completed measures designed to assess grief, coping, and positive perceptions of their experiences of raising a child with Down syndrome (See Appendices L - N). Mothers also completed a demographic questionnaire (See Appendix K), which assessed the characteristics described above. With the exception of the Brief Cope (Carver, 1997), which assessed coping strategies used in response to the death of their child on an ongoing basis since the death, the measures required mothers to report their current experiences (or in the case of positive perceptions, current perceptions relating to the past).

Maternal grief intensity was measured using the Revised Grief Experience Inventory (RGEI; Lev et al., 1993), a 22-item self-report scale of grief symptomatology and intensity (See Appendix L). The RGEI was developed to improve on the 132 item Grief Experiences Inventory (GEI; Sanders et al., 1978), whose dichotomous yes/no items tended to yield little variability amongst the responses of bereaved individuals. The revised version assesses a variety of grief related experiences, and a factor analysis (Lev et al., 1993) has indicated four subscales: existential concerns (six items; e.g., “I feel lost and helpless”); depression (six items; e.g., “I cry easily”); guilt (three items; e.g., “I frequently experience angry feelings”); and physical distress (seven items; e.g., “My arms and legs feel heavy”). A total RGEI score is used as an indicator of overall grief intensity. Participants are asked to rate their agreement with items on a six-point Likert-type scale ranging from slight agreement to strong disagreement. Good reliability has been demonstrated, with parents of children without intellectual disabilities, for both the complete scale (Matthews & Marwit, 2004), and the subscales (Lev et al., 1993). In the present sample Cronbach’s alpha coefficient for the full RGEI scale was .97. Internal consistency for the subscales was also high: .94 for existential concerns, .90 for depression, .79 for guilt, and .91 for and physical distress.



Mothers' coping strategies were measured using subscales of Carver's (1997) brief form of the COPE inventory (See Appendix M). A factor analysis (Hastings et al., 2005a) has shown that the Brief COPE captures four coping dimensions for parents of children with developmental disabilities: problem-focused, active avoidance; religious/denial; and positive coping. Two of these (problem-focused and active avoidance) were used in the present study (See Appendix M). Hastings and colleagues (2005a) found good reliability for both the active avoidance coping and problem-focused coping subscales for mothers of children with developmental disabilities (.78 and .82, respectively). Cronbach's alpha values for the subscales in the current study were .73 (active avoidance) and .71 (problem-focused).

Maternal perceptions of positive contributions made by the child during their lifetime were assessed using a revised version of the Positive Contributions Scale from the Kansas Inventory for Positive Perceptions (PCS; Behr, Murphy, & Summers, 1992). The PCS is a 50-item scale that measures the extent to which the child with intellectual disabilities is viewed as having a number of positive characteristics (e.g., fun to be around), and has had a positive impact on the parent and the family (e.g., improved the social network). The PCS total score has been found to have good reliability ( $\alpha = .92$ ) with mothers of children with intellectual disabilities (Hastings et al., 2005b). With the permission of the Beach Centre at the University of Kansas, the scale was revised for use with bereaved parents (See Appendix N). Mothers were asked to answer according to whether they strongly disagreed; disagreed; agreed; or strongly agreed with statements that completed a stem, (e.g., "Having been a parent of a child with an intellectual disability is – the reason my life has better structure"). Mothers were asked to think only of their child with Down syndrome who had died when answering each statement. Five items were removed, as they could not be amended as items relevant for bereaved parents, (e.g., "I have someone who shares responsibility for doing

several tasks around the house”). Cronbach’s alpha for the remaining 45 items in the present sample of mothers was .96.

### *Procedure*

Participants were recruited through the Down’s Heart Group, a UK charity that offers support and information regarding heart conditions associated with Down syndrome. The charity forwarded information about the study to potential participants on behalf of the research team (See Appendices A – F). All family members were invited to take part (See Appendices G and H). However, 75% of the positive responses came from mothers. The number of fathers, sisters, and grandmothers who also replied was too small to make meaningful sub-comparisons. Consequently, only mothers’ responses were analysed. Demographic information (e.g., age, number of children in the family) was collected either over the telephone by the researcher, or via a form completed by the mother and returned by post (See Appendix K). Due to the confidential nature of the recruitment process, the total number of mothers who received information about the study is not known. The rate of return for questionnaires from those who initially expressed an interest in taking part was 86%. The study was designed with particular reference to published guidelines on conducting research with bereaved participants (Parkes, 1995), the key aspects of which include: providing a clear written explanation of the project to obtain informed consent; providing public access to findings regardless of the outcomes of the research; financial incentives being limited to the reimbursement of expenses incurred (although this was not relevant to the current research); researchers being suitably trained in the support of bereaved persons and supervised appropriately; and the potential for stress during data collection being minimised.

Results

Overall RGEI grief scores for mothers in the present sample ( $M = 75.59, SD = 32.66$ ) were compared to those of bereaved parents of children without disabilities in previous research (Lev et al., 1993; Robinson & Marwit, 2006). One sample t-tests found no significant differences between the mean score for mothers in the present study and these other two samples,  $t(37) = .478, p > .05$ , and  $t(37) = -.066, p > .05$  (two tailed) respectively, suggesting that the present sample of mothers reported no less or more symptoms of bereavement. The main data analysis then proceeded through two stages. First, Pearson’s correlations were used to explore associations between all demographic, child, and maternal report variables (Table 5.2). Second, linear regressions were conducted with the total RGEI and subscale scores as dependent variables and all significant correlates as predictor variables (Table 5.3).

Table 5.2. Correlates of Maternal Grief

Measure		BCAA	BCPF	PCS	Time
1	RGEI Total Score	.508***	-.054	.337*	.302
2	RGEI Existential Concerns	.440**	-.096	.384*	.275
3	RGEI Depression	.539***	-.056	.378*	.255
4	RGEI Guilt	.585***	.007	.232	.255
5	RGEI Physical Distress	.425**	-.028	.238	.343*

*Note.* RGEI, Revised Grief Experience Inventory, BCAA, Brief Cope Active Avoidance; BCPF, Brief Cope Problem Focused, PCS, Positive Contributions Scale; Time, Time since death  
\* $p < 0.05$ ; \*\*  $p < 0.01$ ; \*\*\*  $p < 0.001$ .

Associations between study variables are shown in Table 5.2. All grief outcome scores and potential predictor variables (coping, positive perceptions) are included in 5.2; time since death was the only demographic or background variable significantly associated with maternal grief and is therefore included in the table. No other demographic variables had statistically significant relationships with grief scores. The results in Table 5.2 indicate a positive correlation between positive perceptions of raising a child with Down syndrome and overall grief scores such that higher grief scores related to more positive perceptions. Also, the existential concerns and depression subscales of the RGEI both correlated positively with positive perceptions, and there was a positive correlation between total grief intensity and active-avoidance coping. Further, all four RGEI subscales scores also correlate with active avoidance coping. There were no statistically significant associations between RGEI scores and problem focused coping.

We carried out exploratory regression analyses for four RGEI domains, where there was more than one correlate, to establish the independent contribution of putative correlates of grief. Scores on the guilt subscale were not analysed further as only one initial correlation (with active avoidance coping) was found. For total RGEI grief, active avoidance coping and positive perceptions were entered into the regression model. Overall, a significant percentage of the variance was explained by the regression model, although positive perception scores were not found to be a significant independent predictor of grief. For both the existential concerns and depression subscales of the RGEI, active avoidance coping and positive perceptions were again entered into the regression model and a significant percentage of the variance in existential concerns scores and depression scores was explained. Active avoidance coping and positive perceptions were found to be significant independent predictors in both models. Finally, for scores on the physical distress subscale, active



avoidance coping and time since death were entered into the regression model and accounted for a significant percentage of the variance in physical distress scores. Both active avoidance coping and time since death were found to be significant independent predictors of physical distress.

Table 5.3 Regression Analysis of Maternal Grief.

Dependent Variable	Predictor Variable	$\beta$	$p$
Total RGEI scores <sup>1</sup>	Active Avoidance Coping	.576	.001
	Positive Perceptions	.203	.098
RGEI Existential Concerns <sup>2</sup>	Active Avoidance Coping	.425	.006
	Positive Perceptions	.301	.045
RGEI Depression <sup>3</sup>	Active Avoidance Coping	.541	.000
	Positive Perceptions	.272	.048
RGEI Physical symptoms <sup>4</sup>	Active Avoidance Coping	.480	.001
	Time since death	.407	.006

<sup>1</sup>( $R^2 = .37, F(2, 36) = 10.06, p < .001$ ), <sup>2</sup>( $R^2 = .32, F(2, 36) = 8.02, p = .001$ ), <sup>3</sup>( $R^2 = .43, F(2, 36) = 12.54, p < .001$ ), <sup>4</sup>( $R^2 = .34, F(2, 37) = 9.16, p = .001$ ).

### Discussion

Maternal grief scores on the RGEI were similar to those reported for bereaved primary caregivers of children in previous research (Lev et al., 1993; Robinson & Marwit, 2006). The Lev et al. (1993) sample included both mothers and fathers, and fails to include detail on time since loss, while the Robinson and Marwit sample was comprised of mothers only but, similar to the present sample, the time since the death of the child varied considerably from approximately 3 months to 31 years (Robinson & Marwit, 2006). Unlike findings from the wider bereavement literature (Kreicbergs et al., 2004), the results of the present study did not find evidence of a decrease in grief intensity over time. Rather, a significant positive relationship between time since death and physical distress was found. The grief experiences of this group of mothers may be particularly long lasting. However, alternative explanations include the possibility that grief was decreasing in an atypical way not examined by the measure, that grief was of a higher intensity to begin with, or that physical distress was increasing over time. The physical distress sub-scale includes items related to energy, appetite, sleep, and general health all of which may be characteristic of normal aging (Birren & Schaie, 2006). Thus, it may be the case that this sub-scale measures more than the process of grief.

Positive perceptions of parenting a child with Down syndrome and a CHC who has died correlated positively with overall grief scores. Just as parents of living children with Down syndrome hold positive perceptions of raising their child (Carr, 2005; Poehlmann et al., 2005), so too do mothers of children with Down syndrome and a CHC who have died. While they were not found to be an independent predictor of overall RGEI grief scores, positive perceptions were found to be independent predictors of scores on both the existential concerns and depression subscales of the RGEI. Thus, mothers who held the most positive

perceptions of their child with Down syndrome and their impact on themselves and their family seemed to experience the most intense grief. This is the first time that this relationship has been explored in ID research and it clearly requires replication. In addition, the mechanisms explaining this relationship along with studies taking multiple measures over time would be fruitful avenues for future research.

One possible explanation for the positive relationship found between positive perceptions and grief may lie in a lack of support following loss. Intellectual disability research has linked adequate, helpful support, and positive perceptions of raising a child with intellectual disability such that helpfulness of support predicts positive perceptions (Hastings, Allen, McDermott, & Still, 2002). Bereaved mothers of children with an intellectual disability report a loss of role and support from intellectual disability organizations following the death of their child (Milo, 1997; Todd, 2007). It is possible that in this study those who reported the most positive perceptions, were also those who felt most supported prior to the death. Consequentially, this loss of support may have been particularly acute. The necessity of a clearer understanding of the pre- and post-loss needs of families of children with complex health needs has been highlighted (Emond & Eaton, 2004), but further investigations into the needs of bereaved parents of children with Down syndrome is necessary to create a clearer understanding of the experiences of this group.

We also found significant independent positive relationships between all of the RGEI dimensions and the use of active avoidance coping strategies by mothers to cope with their loss. The active avoidance coping subscale is comprised of strategies that reflect attempts to avoid the stressor (i.e., bereavement and grief) or escape from its effects, and includes items related to substance use; behavioural disengagement; self-blame; venting of emotions; and distraction. Given that the increased use of active avoidance strategies to cope with their

child's death is related to higher current grief scores, avoidance coping appears to be maladaptive for bereaved mothers. Previous research has suggested that active avoidance coping may be maladaptive for mothers of children with Down syndrome (Spangenberg & Theron, 2001); and bereaved individuals (Bonanno et al., 2005). Additionally, Fraley and Bonanno (2004), propose that a relationship between attachment and anxiety may be responsible for the risks posed by avoidance coping: fearful-avoidant attachment patterns combined with high anxiety are associated with elevated symptoms of grief and distress (Fraley & Bonanno, 2004). Parents of children with Down syndrome have been found to show elevated anxiety (Spangenberg & Theron, 2001). Therefore, future research could investigate whether elevated anxiety puts parents at greater risk when their child dies, and whether interventions aimed at reducing anxiety and avoidance coping strategies have significant adaptive outcomes for parents.

The Dual Process Model of coping with bereavement defines two types of stressors that bereaved people encounter: loss-oriented and restoration-oriented stressors (Stroebe & Schut, 1999). While loss-oriented stressors are thought to predict grief outcomes in the short-term, restoration-oriented stressors are secondary consequences of bereavement (e.g., loss of support networks), which are thought to predict long-term, less reversible, bereavement outcomes (Stroebe et al., 2006). Although the bereavement experiences of mothers whose child with Down syndrome has died have not been investigated until now, a small number of qualitative studies have examined the experiences of mothers whose child with an intellectual disability dies (Milo, 1997; Todd, 2007). Role in the world of intellectual disability and the role as a caregiver are among the outcomes of parenting a child with intellectual disability that parents view positively. The loss of these roles when the child with intellectual disability dies (Milo, 1997; Todd, 2007), may act as a source of restoration-oriented stress and also



bring with them more existential concerns. The use of active avoidance strategies in relation to these existential concerns may contribute to the continuing intensely experienced grief. The current findings highlight the importance of addressing existential concerns in addition to other outcomes of grief, such as helping people find new direction in their lives as suggested by Parkes (1998).

A primary limitation of the current research is that the data were obtained exclusively by self-report. Self-report measures may not capture the full complexity of the grief experience. In particular, a retrospective measure of coping with loss may be influenced by inaccurate recall and by the participants' generalized beliefs about coping (Stone et al., 1998). Cultural differences have also been found for coping (Bonanno et al., 2005), and bereavement experiences (Parkes, Laungani, & Young, 1996). The current findings, however, reflect the experiences of a small, British, largely Christian sample and this should be borne in mind. The likelihood of giving birth to a child with Down syndrome increases with age (Crane & Morris, 2006), and while research in the area of intellectual disability has not typically found age of the parent to be predictive of psychological outcomes (within typical limits), this issue may warrant further attention in bereavement research. Due to the confidential nature of the recruitment process it is unknown how many mothers originally received information on the study but decided not to take part. In addition, the experiences of mothers cannot be presumed to mirror those of other family members. Focusing on parents whose child with Down syndrome and a CHC has died has given the researchers access to a relatively large, and previously un-researched sample; the first quantitative study of parental bereavement experiences in the ID field. Unfortunately, there remain many bereaved families of children with other types of intellectual disability, who have yet to be invited to take part

in research. The experiences of these families, however similar or different to the experiences of families of children with Down syndrome, are of equal importance, and also deserve study.

Bereavement experiences of mothers of children with Down syndrome and CHCs are long lasting, potentially intense, and related to the strategies they use to cope with the loss. Concurrent to these lengthy grief reactions, mothers view their experiences of having raised a child with Down syndrome positively. Increased insight into positive perceptions, avoidance coping strategies and their relatedness to symptoms of grief could aid in the development of bereavement support services. Knowledge from related areas, including family intellectual disability research and palliative care, could inform developments in this area of study. For example, guidelines developed by paediatric palliative care research may provide a foundation for the development of policy on pain management, decision making, and family oriented care (Donnelly et al., 2005).

In terms of practical implications, the findings of this study suggest that interventions encouraging parents to acknowledge their emotions, and reinvent their life roles may be beneficial. Emotional disclosure and the reinterpretation of meaning are the main functions of bereavement counselling and therapy, and research has found them to be particularly beneficial for individuals who display complicated grief (Stroebe, Schut, & Stroebe, 2005). Risks factors for complicated grief include aspects such as experiencing the death of one's child concurrent to other parenting and personal stresses (Stroebe & Schut, 2001). Thus, grief counselling and grief therapy (e.g. Worden, 2002), may be suitable for use with bereaved parents of children with ID. Further, education on the experiences of bereaved parents of children with intellectual disabilities is necessary to make professionals mindful of the possibility that family members who lose a child with intellectual disability will experience positive as well as negative reactions. Priorities for professionals may include developing a

support system whereby parents might maintain their role in the world of intellectual disability; explore ways in which parents with strong positive perceptions might be supported in their grief; and additionally be aware that this support may be needed for a considerable length of time.

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**Chapter 6 - Discussion**



Parental bereavement and intellectual disability is a complex experience, appreciably exclusive, and somewhat unmapped. This thesis has begun to unravel the intricacies of this particular parenting experience in families of children with an intellectual disability. First, a thorough review of the literature (Chapter 2) identified similarities between the bereavement experiences of parents of children with intellectual disabilities and parents of typically developing children: the lasting nature of grief experiences, range of grief symptoms, and potential for benefit finding in their loss. Bereavement experiences, however, differed noticeably when considering issues such as preparation, the availability of formal bereavement support, disenfranchised grief, and multiple losses. The literature review presented substantial evidence to warrant the expansion of investigations in the field. This discussion will summarise and discuss the findings of three empirical studies (Chapters 3, 4, and 5), evaluate them in the light of previous research, and deliberate on the implications this body of work carries for theory, research, and practice in intellectual disability. This discussion will also address how our findings may instigate change and inspire further research in this area.

### *Qualitative Research in Parental Bereavement and Intellectual Disability*

Given the likely high levels of variability that exist between participating families, qualitative methods were deemed particularly suitable for undertaking exploratory investigations of groups of bereaved parents. Chapter 3 illustrates themes to emerge from an investigation of the experiences of mothers whose child with one of a variety of intellectual disabilities died. Specific maternal experiences of bereavement previously identified in research and replicated in this thesis included: losses experienced both at birth and death, dissatisfaction with medical care and medical professionals, disenfranchised grief, loss of

formal support from the intellectual disability community, a lack of specific bereavement support, benefit finding in loss, and positive perceptions of the entire experience (Milo, 1997; Schormans, 2004; Todd, 2007; Wood & Milo, 2001). Although the account of only one adoptive parent contributed to our study, it illuminated a multiplicity of sources of disenfranchised grief in agreement with Schormans' (2004) account. While biological parents of children who died reported feeling their grief was disenfranchised because their child had a disability, foster parents felt their foster status resulted in outsiders deeming their grief illegitimate (Schormans, 2004). This highlights the individuality of experience and supports the argument for a high degree of specificity in future research that will be elaborated on later in this discussion.

Additional themes revealed through the qualitative methods employed in this thesis provided a new perspective on coping mechanisms in bereaved parents, when compared with previous findings. Milo (1997) and Wood and Milo (2001) describe how clear patterns of coping strategies emerged differentially from their research with mothers and fathers. Mothers favoured cognitive coping strategies (Milo, 1997), while fathers tended towards emotional stoicism and activity coping (Wood & Milo, 2001). Alternatively, both qualitative studies in this thesis (Chapters 3 and 4), uncovered considerable variation in coping strategies used by mothers and fathers, both before and after the death of their child. Parents used a range of problem-focused, spiritual, avoidant, and cognitive coping strategies, while joint activity, mutual support, and communication, were described as most helpful for many couples. Additionally, coping strategies favoured by individuals were not consistent across the pre- to post-loss period. While they changed according to personal preference, coping choices were also a response to resources and the needs of others, most notably their spouses. This variability in coping strategies is in line with recent interpretation of diversity in

bereavement experience (Wortman & Silver, 2001) and suggests that the coping literature may need to re-examine the basis on which coping strategies are explored, researched, and categorised. By grouping people into categories according to their coping style at one point in time, valuable information on how ways of coping change; what influences the ways people choose to cope; and the relative impacts of these changes may be lost. Future research into coping, not only in the area of bereavement, should consider the potential merits of a more flexible approach.

The research described in Chapter 4 is the first of its kind to explicitly investigate the bereavement experiences of parents whose child with Down syndrome and a congenital heart condition dies, and the first to investigate married couples' experiences of the death of a child with an intellectual disability. Issues particularly relevant to the dual diagnosis of Down syndrome and a congenital heart condition included the trauma of receiving a second diagnosis of a heart condition at an already difficult time for parents; and the difficulties associated with achieving medical care and palliative care for the child. Caregiving responsibilities have been found to be associated with higher stress in parents of children with Down syndrome, and parents of children with chronic conditions (Kuster & Merkle, 2004; Roach et al., 1999). It is unknown to what extent responsibilities for making care-related decisions have an impact on the grief experiences of parents, but future research on this topic utilising quantitative methods and standardised outcome measures may shed additional light on the subject.

The distinct characteristics of couples interviewed for the study reported in Chapter 4 reinforce the complexities involved in sharing such a significant experience between partners, which have yet to be disclosed by researchers. While Todd (2007), includes partners in his account of intellectual disability and parental bereavement, the dimension of a shared



experience is not explicitly included in his analysis of accounts. Our study identified the significance of couples' joint experiences of the life and death of their child. Themes which added to the findings in one-parent studies were: the welcome support that can be provided by a partner when making decisions, the reality of decision-making and coping as a couple becoming more difficult with bereavement, the need to attend to individual and not gender-specific needs in grief, and the positive impact the loss may have on the marital relationship. Similar to the positive outcomes such as personal strength, and empathy reported by parents, couples shared descriptions of greater closeness to partners as a result of sharing this most personal of experiences. Although these findings emerge from a small group of distinct cases, the data are significant for those providing bereavement interventions.

### *Quantitative Research in Parental Bereavement and Intellectual Disability*

While qualitative methods are suitable for an in-depth exploration of individual experience, larger scale quantitative research is necessary to provide detail on a wider scale and provide comparison with previously researched samples. The mothers in Study 3 (Chapter 5) reported grief symptoms at levels similar to previously researched groups (Lev et al., 1993; Robinson & Marwit, 2006) on the Revised Grief Experience Inventory (RGEI, Lev et al., 1993). However, the grief experiences of the mothers in our current cross-sectional study did not reflect previous findings of a reduction in grief intensity over time (Kreicbergs et al., 2004). Possible explanations could be considered in future research, which might also further investigate the relationship between positive perceptions of the child and grief intensity.

Just as parenting research has found positive perceptions in parents raising a child with Down syndrome (Carr, 2005; Poehlmann et al., 2005), so too did the mothers in the



current study. However, an investigation of potential correlations between support following loss, contact with intellectual disability services, or parenting satisfaction could shed further light on this finding. In addition, we found evidence of the maladaptive nature of active avoidant coping that has also been shown both in the parenting in Down syndrome (Spangenberg & Theron, 2001), and bereavement literatures (Bonanno et al., 2005). Specific information on possible relatedness of coping strategies to the likelihood of one accessing bereavement interventions, or seeking social support could illuminate such findings further and should be a consideration for further research, and the development of other, favoured, interventions.

The use of a mixed method approach to the topic of bereavement and intellectual disability is one that was a necessary route to facilitate the comprehension of a sensitive topic, while simultaneously allowing for comparison with prior research in the parallel fields of bereavement and intellectual disability. This explicit variability in approach and method has been proposed as a fitting process of advancement of knowledge in a specific area, whereby creativity of method and the use of novel or adapted methods of investigation may inspire other researchers (Elliott, Fischer, & Rennie, 1999). This thesis has illustrated that research need not be exclusively qualitative or quantitative in design and that a combination of approaches has considerable advantages for researchers and participants.

The use of a quantitative measure of positive perceptions, for example, clarified their importance for mothers, and although few mothers reported coping through avoidance when interviewed, their responses to the more anonymous questionnaires indicated that some mothers did utilise active avoidant coping strategies and these strategies accounted for a considerable amount of variance in intensity of grief reactions. Additionally the use of qualitative method gave the researcher inside knowledge of an emotional experience in the

life of a bereaved parent, one that may have been difficult, if not impossible, to access with other methods. The relationship that developed between participant and researcher, although not anticipated by the research, was one of trust and honesty. Comments regarding how the parent had not spoken about such matters to anyone before often followed the disclosure of particularly personal details. Participants were in all cases grateful for the opportunity to contribute to the research and such remarks reiterated the importance of investigating such an emotionally sensitive topic

### *Theoretical Implications*

One particular overriding finding of the thesis was the positivity of the bereaved parents. The positive outcomes and perceptions they reported despite the traumatic events of illness and loss they have been endured were consistent, shared by mothers and fathers alike, and were present despite reports of high levels of physical and mental distress. Such findings add significantly to the nascent literature on positive perceptions in parenting and intellectual disabilities. The literature has suggested that positive perceptions may increase over time (Poehlmann et al., 2005), while others have found that while they it didn't increase, positive impact was stable over time (Blacher & Baker, 2007). Although we have no comparison of perceptions pre-loss, the high levels reported by mothers suggest the positive impact of having a child with Down syndrome is enduring. The simplest explanation is that the mothers valued their child; the more they miss them after their death.

Alternatively, the higher grief scores seen alongside these high levels of positive perceptions may result from a tendency to focus on the positive aspects of the lost relationship, while an individual who has turned a corner and no longer feels the need to centre their life on their dead child may also be open to other possibilities, such as a more

accurate recollection of their child's life (Parkes, 1998). Additionally, the positive perceptions could be related to the loss of the considerable support parents receive while their child is alive, with the most supported parents reporting the most positive perceptions. Mothers spoke of the considerable impact of the loss of formal support and this loss may have been complicit in the high grief scores found in mothers with the highest positive perception scores.

Couples also mentioned these positive perceptions of their child, in addition to positive implications for their marital relationships. Marriage break-up and a reduction in marital quality is an area that has received much attention by bereavement researchers (Lang & Gottlieb, 1993; Murray et al., 2000), and while the buffering effects of a strong marital relationship have been reported, the positive outcomes, and the interpersonal context in which the individual grieves are often neglected (Oliver, 1999; Ponzetti, 1992). The bereavement literature is lacking empirical research into reports of positive outcomes in couples. Whether this is specifically symptomatic of the death of a child with intellectual disabilities or a more widely experienced outcome is unknown and knowledge on this and the mechanisms that support it could be of considerable utility to bereavement theory. Findings on stability of positive perceptions over time in the area of intellectual disability research (Blacher & Baker, 2007), suggest that an investigation of perceptions in bereaved parents over time might be a valid direction for research.

Also of significance to bereavement theory is the disenfranchised grief reported by parents. Disenfranchised grief occurs when the loss is not recognised as a significant one (Attig, 2004), and has been reported by both foster/adoptive and birth mothers and fathers. The extent to which disenfranchisement puts parents at additional risk of distress post-loss is unknown, but it is reasonable to assume that it both limits opportunities to discuss the loss



and decreases the availability of support. While it is important to change societal attitudes to ensure the legitimacy of death in intellectual disability is recognised, it is also essential that the needs of families for whom change comes too late also receive attention. Ways in which those delivering bereavement care, medical professionals, and other involved parties such as funeral directors can support parents struggling to find hope and significance in their loss and thus work through experiences of disenfranchisement have been described (Attig, 2004; Doka, 1988), however, an investigation of the long-term impact of disenfranchisement on this group could be beneficial to parents and informative for service providers.

The important role social support has in families of children with intellectual disabilities is well known (White & Hastings, 2004), the role of social support in bereavement however, is less clear. Support from family members and friends may not be forthcoming when family members are similarly affected by the loss, or when disenfranchised grief results in a failure to recognise that support is required (Rubin & Malkinson, 2001). The current research was limited by the lack of an appropriate measure appraising support received, and the development of a scale to measure and evaluate support received in bereavement is an important topic for future study. Mothers spoke about the loss of formal support sources as an additional significant loss following the death of their child. Potential explanations for the withdrawal of support include the lack of funding or resources on the part on intellectual disability services; reluctance on the part of services and staff to confront the sensitive subject of bereavement; bereavement support services failing to recognise the importance of the loss; or again a lack of resources on the part of bereavement services. This lack of knowledge and theory needs to be addressed.



*Implications for Policy and Practice*

The depth and length of the interviews which formed the basis for the results presented in this thesis highlighted the existence of a knowledgeable group of individuals, eager to participate in research and keen that change should occur. It seems fitting, therefore, that bereaved parents, where possible, should be involved in policy and healthcare improvements. The qualitative aspects of this thesis highlighted a desire for continued involvement in services by parents who maintained a variety of roles in special education, respite care, foster care, and intellectual disability services. Achieving this role was not always easy for parents and were this path of involvement for parents more easy to negotiate, it could rectify the loss of contact with services that parents report; provide a wealth of experience for service providers; and provide an invaluable resource for parents who may be facing the reality of preparing for the death of their child.

Children's hospices currently attend to the palliative care needs of many families of children with intellectual disabilities and conditions that result in a shortened life. Over the years palliative care, preparation, and bereavement guidelines have developed in line with demand and hospices hold a considerable expertise in this area (Association of Children's Hospices; ACH, 2005). However, hospice services are not always available to families, not all children experience a lengthy period of illness, and parents may decline services offered (Russo & Wong, 2005). As a result, not all families will have the bereavement expertise of a children's hospice at their disposal. In addition, due to a greater understanding of rare disorders, more treatment options, and improvements in access to healthcare, children with conditions that formerly resulted in death in childhood are now surviving into adulthood, where palliative expertise has yet to catch up.

Palliative care services and expertise are necessary but lacking in adult intellectual disability services (Tuffrey-Wijne, 2003; Tuffrey-Wijne, Hogg, & Curfs, 2007). In particular, the high incidence of Alzheimer's disease in adults with Down syndrome (Holland, 2000), illustrates that it is imperative that specific services develop for individuals and their families in line with the proposed improvements in care services for individuals with dementia without Down syndrome (Ballard et al., 2001). One potentially useful example from dementia care is the provision of Admiral Nurses (Dugdale, 2000). Admiral nurses attend in particular to the needs of caregivers of persons with dementia as the disease progresses and provide bereavement support after the death of the person with dementia. With appropriate training, such a service could be particularly helpful with pre- and post-loss adjustment, decision making, and negotiating care in families of individuals with intellectual disability and dementia (Keady, Ashcroft-Simpson, Halligan, & Williams, 2007).

This thesis has also broached the subject of pre-loss experiences and the impact of preparation for death in families where a child with an intellectual disability. Findings from the area of chronic illness has suggested that preparation and involvement in care may impact on the post-loss symptoms of grief (Rando, 1983), and findings from the current and previous research highlight the importance of honest, open communication with professionals (Wood & Milo, 2001). Changes to training and education of medical professionals, for example medical students (Harwood, 2007), could raise awareness and aid with the development of concrete guidelines for communicating terminal diagnoses and making difficult decisions. Such training and exposure could also improve healthcare professionals' skills in dealing with people with intellectual disabilities themselves. Findings have underscored the reality that a lack of confidence and fear on the part of secondary healthcare staff is a considerable barrier to healthcare for people with intellectual disability (Sowney & Barr, 2006).

The question of who should provide bereavement interventions for this neglected group is a critical point for discussion. Given the potential for disenfranchisement amongst these parents, it is crucial that bereavement services are aware of the multi-faceted experience parents endure. Meanwhile, the understanding incorporated into intellectual disability services suggested that they are potentially suited to such a supportive role. The importance of support from other similarly bereaved parents, and continued contact with their child's services after death was important to mothers interviewed, and a service facilitated by intellectual disability providers may be suitable for parents. However, expertise in bereavement and the resources necessary to provide additional services may elude intellectual disability providers. Within paediatric oncology services, attempts have been made to maintain similarly desired links between staff and families and have resulted in promising results for parents and positive feedback on their efforts for professionals (Russo & Wong, 2005). The use of the parent-to-parent framework (Ainbinder et al., 1998) is also a possibility. This type of support puts parents in contact with families in a similar situation and they provide each other with mutual support, usually via the telephone. The use of this method is strengthened by evidence of the suitability of telephone support for bereaved parents (Nair, Goodenough, & Cohn, 2006).

The reality of the bereavement experience in families where a child with an intellectual disability dies is that support is not forthcoming, nor in many cases is it available when sought. Reasons why this is the case, and suggestions for the logistics of delivering support have been discussed above, but the precise content of a potentially useful bereavement intervention for these parents must also be attended to. Evaluations of traditional bereavement counselling interventions suggest they are effective for only the most distressed and at risk individuals. Bereaved parents are generally assumed to be amongst



those at most risk, and such grief counselling may therefore be suitable for in this case (Worden, 2002). Other potential useful interventions include couples therapy such as Emotional Focused Therapy (EFT; Johnson, 2003), which focuses on confronting emotions rather than changing behaviours. It is a potentially useful tool for couples experiencing difficulties understanding each other's grief experiences and communicating their emotions.

Furthermore, given the relationship between active avoidance and grief, an intervention which targets avoidance and instead encourages acceptance such as Acceptance and Commitment Therapy (ACT; Hayes, Luoma, Bond, Masuda, & Lillis, 2006), may be suitable for use in this area. ACT therapies have not been widely used with bereaved populations, however, their use with parents of children with disabilities has indicated improvements in parental reports of depression and psychological distress (Blackledge & Hayes, 2006). The further exploration of the use of ACT and other interventions with bereaved parents and couples should evaluate their potential for use for with this group. Interventions are most effective when applied flexibly to each individual situation (Beutler, 2000), and an open approach on the part of the therapist whereby a combination of approaches are applied depending on the requirements of parents and families and couples should be considered.

To return to the issue of study design, participants reported that the interview afforded them an opportunity to discuss matters that had often been a closed subject for parents. They appreciated the opportunity to tell their story, and reflected positively on the cathartic nature of the experience that they hoped would help others. Such reactions to involvement in research have been found by other bereavement researchers (Burnell & O'Keefe, 2004; Dyregrov, 2004), but may be particularly relevant in the case of parents whose child with intellectual disability has died who also report experiencing exclusion from the world of



intellectual disability. Some, but not all parents had the opportunity to discuss the death with their child's doctors after their death. Writing therapies have previously been associated with positive outcomes for individuals with PTSD, rheumatoid arthritis, and asthma (Exline, Smyth, Gregory, Hockemeyer, & Tulloch, 2005; Stone, Smyth, Kaell, & Hurewitz, 2000). Finding benefit through sharing ones story and in particular using a form of writing therapies may be suitable for bereaved parents (Niederhoffer & Pennebaker, 2002).

### *Methodological and Theoretical Issues*

Despite the considerable contributions this thesis makes to the bereavement and intellectual disability literature, two types of limitations exist within the work: limitations posed by methodological issues and limitations to the theoretical implications of the findings.

While the small samples used in qualitative research limit the generalisability of findings, assumptions can be bolstered by in-depth analysis, themes firmly grounded in the data, and in this case the use of mixed methods of evaluation to additionally illustrate outcomes quantitatively. One significant limitation with qualitative research as employed in this thesis, however, is a product of the risks inherent in qualitative research. Members of a vulnerable population such as bereaved parents may have been reluctant to participate. The personal nature of the interview, its' length, and the emotion often elicited from participants together with potential risks to confidentiality may mean, as has been suggested by others (Dyregrov, 2004), that the most vulnerable parents may not have taken part. Therefore, there may be parents for whom positive perceptions are not easily accessible, or continued involvement in the world of intellectual disability is not desired. This shortcoming should be carefully considered when putting our findings into practice.

In terms of the participants who agreed to take part, the majority were female, of similar ethnic background, and had a child with a similar intellectual disability. Limitations posed by participant characteristics are common complaints in research (Drotar, 1994; Hatchett, Holmes, Duran, & Davis, 2000), and future research might address such limitations by investigating ways of encouraging more ethnic minorities, family members other than mothers, and those with more rare disorders to take part. Researchers must first demonstrate how and why the experiences of these minorities are of value to research and service provision. Additionally, researchers could investigate alternative pathways for recruitment. In the present research, participants were recruited predominantly through syndrome support groups. By identifying alternative recruitment settings a more representative group may be identified. This would allow the in-depth examination of fathers' experiences and investigate whether cultural differences such as religious beliefs and familial support result in different bereavement outcomes for parents.

While the homogeneity of intellectual disability related syndromes seen in a large proportion of the current sample lends strength to the study findings, it omits the parents' experiences with a child who has a more rare condition that may also result in premature death, for example, tuberous sclerosis, or Lesch-Nyhan syndrome. An account written by parents of a child with a rare disorder, describes difficulties accessing intellectual disability services and achieving a programme of education for their son with Niemann-Pick disease. Professionals knew little about their son's disorder and failed to appreciate the immediacy of their needs brought about by the non-linear and often sudden nature of their son's decline (Wray & Wray, 2004). It stands to reason that parents in such a situation additionally receive little recognition of the significance of their loss. In comparison with parents of children with Down syndrome, an extensively researched and to a large extent understood syndrome,

parents of a child with a rare disorder may have unmet and unspecified needs, which have also been overlooked within this thesis.

Theoretically, the implications of the thesis may also be limited by variables unexamined by the research. Variables such as quality and levels of formal and informal support received, religious convictions, and attributions of blame may elucidate findings further. For example, the use of a measure examining religious coping strategies (Anderson et al., 2005), might explore the relationship between mothers' religious beliefs and finding meaning in their experience. All of the outcomes measures and data in the thesis relied on self-report and were retrospective in nature. The potential inaccuracy of the use of retrospective measures to examine coping, for example, has been noted (Stone et al., 1998). Alternatives to self-report measures include practitioner reports, observational data, or in the case of research with couples, data from multiple informants may be particularly applicable. Multiple evaluation of intervention effectiveness could also avoid response bias on the part of participants or therapists.

Additionally, the data were collected cross-sectionally. Collection of data at more than one time point would indicate greater reliability of findings, but was not employed within our research. Similarly, longitudinal follow-up would provide information on the lasting nature of the high levels of grief symptoms found for these parents. Follow-up of participants may indicate whether the intense grief experiences of the parents involved in the research were lasting over time or diminished in a manner similar to other bereaved parents (Kreicbergs et al., 2004). Any further research with this group should endeavour to replicate methods used. Quantitative methods would allow direct comparison with previous findings and qualitative interviews could explore parents' views on their grief over time.



This thesis attends only to the experiences of parents, and predominantly to those of mothers. It is important that future investigations consider the experiences of fathers to the same depth. However, the experiences of siblings are also worthy of considerable research attention. Substantial changes occur to family life when a child in the family dies, and siblings can be negatively impacted by the loss (Sirkiä et al., 2000). Impacts and changes may be considerably diversified when the child has significant additional care needs (Rodger & Tooth, 2004). Siblings may have made arrangements for future care, or been a carer for their brother or sister with intellectual disability. The relief this loss of responsibility may bring could be confusing and difficult to come to terms with. Such experiences may result in guilt and anger for siblings. The findings of this thesis should not be assumed to generalise to other family members, and sibling bereavement experiences unquestionably warrant considerable and specific research attention.

### *Implications for Future Research.*

Future research could potentially build on the investigations set in motion by this thesis. The experiences of fathers whose child with an intellectual disability has died has been the subject of research in the USA (Wood & Milo, 2001), and similar methods to the current study should explicitly investigate the experiences of fathers in the UK including: their needs pre- and post-loss, their preferred coping strategies, experiences of support, satisfaction with support, and positive perceptions, amongst other issues. Replication of quantitative investigations could assess paternal grief and other bereavement outcomes, and compare with mothers and previously researched groups of fathers to ascertain the longevity and intensity of their grief experiences. Recruiting fathers proved difficult in the current study where parents were largely recruited through support groups, but many fathers reported voluntary



continued involvement in special education, and this could be a potential alternative setting for recruitment. In addition, research could include reports from siblings and other informal caregivers to improve the services provided both before and after the death of a child with intellectual disability.

This thesis both combined and separated the experiences of parents with regard to type of intellectual disability. While parents reported a great deal of similarity despite different diagnoses for their children, parents of children with Down syndrome and a congenital heart condition reported unique experiences of a double diagnosis soon after birth and a battle for healthcare which resulted in their child's diagnosis of Down syndrome being of less importance due to a potentially fatal heart condition. Future research should consider investigating specifically the experiences of families by diagnosis. For example, families of children with a degenerative condition may have additional requirements not common to other disorders as their child experiences significant developmental decline, rather than growth over time.

There exists also a group of parents who never receive a diagnosis for their child. Support groups such as Syndromes Without a Name (SWAN) aim to promote the welfare and health needs of children who suffer from undiagnosed conditions, and their families. The lack of diagnosis makes access to services and claiming benefits difficult, and parents find themselves having to prove again and again that their child's need is genuine (Rosenthal, Biesecker, & Biesecker, 2001). Parents also have little or no indication of life expectancy and where the undiagnosed disorder results in premature death for that child it is reasonable to assume that the family will experience some form of disenfranchised grief. Well-trained and appropriately located bereavement support workers could highlight and explain the potential needs of these families to medical professionals.

Medical professionals need to also be made aware of the importance of a clear and accurate early diagnosis for parents, when a child is born with an intellectual disability. A large-scale evaluation of both parent and professional experiences of diagnoses could inform parents as to the reasons behind the barriers to a prompt diagnosis, such as medical expertise. Similarly professionals may benefit from an insight into the experiences of families coming to terms with the considerable health needs of their young child. Future research should additionally endeavour to distribute information jointly between psychological and medical research traditions.

This thesis has discussed interventions that may be potentially useful to this bereaved parent group. Parent-to-parent support has been found to be useful by parents of children with intellectual disability, physical disability, and chronic illness (Ainbinder et al., 1998; Kerr & McIntosh, 2000; Poyadue, 1993; Santelli et al., 1997). Research into the effectiveness of parent-to-parent programmes, however, points to the need for adequate programme management to enhance match-making and follow-up efforts (Ainbinder et al., 1998). Thus, any potential parent-to-parent programme for bereaved parents would, in addition to requiring expertise in supporting parents of children with intellectual disability, financial input and administration resources, require ongoing evaluation to assess its effectiveness for this group.

In a similar vein, the provision of bereavement counselling alone is not enough. The effectiveness of each intervention should be evaluated to ensure it is worthwhile, and to contribute to the knowledge base (Beutler, 2000). Thus, counsellors can avoid implementing ineffectual interventions. Parents are amongst the groups identified as at high risk of post-bereavement distress (Stroebe & Schut, 2001) and counselling should be available, accessible, and flexible for all those at high risk (Jordan & Neimeyer, 2003). An ongoing

audit of rates of uptake, characteristics of those who avail of services, and satisfaction with interventions could inform those providing the facility as to how the service might be improved, if necessary.

### *Conclusion*

To conclude, the experiences of families who lose a family member with an intellectual disability are of considerable importance given the significant health risks that accompany bereavement responses (Stroebe, Hansson, Stroebe, & Schut, 2001b). This thesis has illustrated the lasting and intense nature of maternal grief reactions; explored the potential influence yielded by coping strategies; investigated the impact of positive perceptions and their relatedness to support; and considered the differential experience of partners experiencing a bereavement as an equally bereaved couple. The weight of the grief experience, though individually exclusive, can be alleviated to some extent by suitable support, honest communication, and recognition of their loss. For all of the parents the positive reflections of their child's life and death were a benefit found in their loss. A benefit unfortunately not always addressed or acknowledged by others.



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Appendix A

Letter to Services English Version



The Wales Study of Bereavement in Families of Individuals with a Learning Disability.

Astudiaeth Profedigaeth mewn Teuluoedd Pobl ag Anabledd Dysgu yng Nghymru



Dear Sir or Madam,

We are conducting research into the effects of bereavement on families who have lost a child (at any age) with an intellectual disability. We wish to talk to as many parents, older siblings and grandparents as possible across the U.K. and Ireland. The data collected will be used to develop an intervention for health staff to train them on how to support bereaved or at risk families. We hoped that you might be able to pass on an introduction to parents or other family members through your organisation. We wish to talk to families who have lost a child with a learning disability at least 12 months ago and at any age. The research is funded by the European Social Fund and is a collaboration between Mencap and the University of Wales, Bangor. The study has undergone a full ethical review by the ethical research committee in the university.

If you are happy to let families know about our research, information about the study (written for families) is enclosed. We are asking organisations to send out information on our behalf. Families are then asked to make direct contact with us. We would provide you with all of the required copies of information and envelopes for distribution, and pay for any postage costs incurred. Until families make contact with us they would remain anonymous to the research team. We are not asking you to pass on any details about families to us.

I will telephone your organisation in the next 7-10 days to follow up this letter and to answer any questions that you may have. I would be happy to speak to your secretary if you can leave an appropriate message with them. You are welcome to contact me directly if you wish:

e-mail: [d.reilly@bangor.ac.uk](mailto:d.reilly@bangor.ac.uk) or telephone +44 (0) 1248 351151 ext.8706

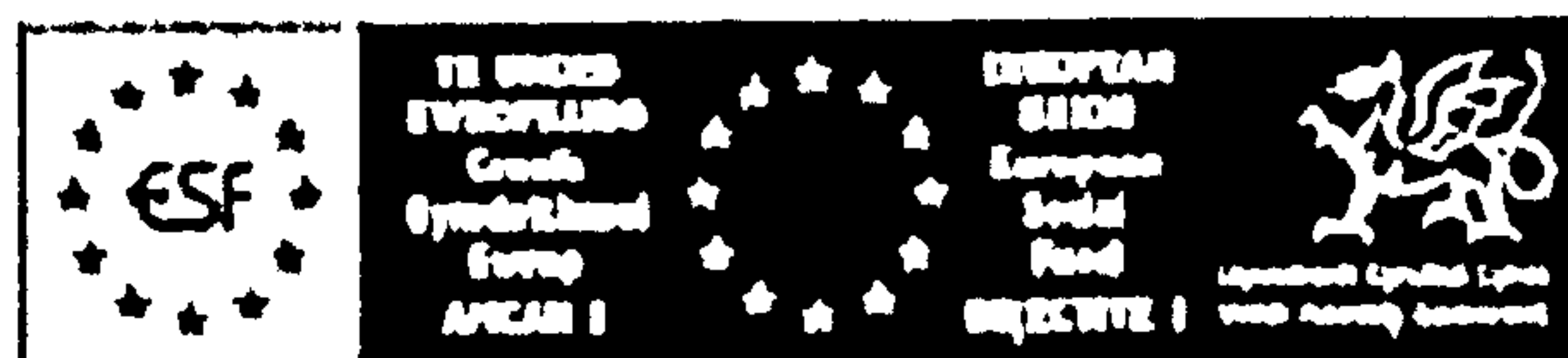
Thank you for your time, and we very much hope that you will be able to help us.

Yours sincerely,

Deirdre Reilly.

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Appendix B

Letter to Services Welsh Version





The Wales Study of Bereavement in Families of Individuals with a Learning Disability.

Astudiaeth Profedigaeth mewn Teuluoedd Pobl ag Anabledd Dysgu yng Nghymru



Annwyl Syr neu Fadam,

Rydym yn gwneud gwaith ymchwil i effeithiau profedigaeth ar deuluoedd sydd wedi colli plentyn (am unrhyw heneiddia) ag anabledd deallusol. Hoffem siarad â chynifer o rieni, brudyr neu chworydd hyn a theidiau a neiniau ag y bo modd ar draws y DU ac Iwerddon. Bydd y data a fydd yn cael ei gasglu yn cael ei ddefnyddio i ddatblygu ymyriad ar gyfer staff iechyd i'w hyfforddi ar sut i gynorthwyo teuluoedd sydd wedi cael Ein gobaith ni yw y gellwch ein helpu i gysylltu â rhieni neu aelodau eraill o'r teulu trwy eich sefydliad. Hoffem siarad â theuluoedd sydd wedi colli plentyn ag anabledd dysgu o leiaf 12 mis yn ôl ac o unrhyw oed. Mae'r ymchwil yn cael ei ariannu gan Gronfa Gymdeithasol Ewrop ac mae ar y cyd rhwng Mencap a Phrifysgol Cymru, Bangor. Mae'r astudiaeth wedi cael adolygiad maesegol llawn gan y pwyllgor ymchwil foesegol yn y brifysgol.

Os ydych yn foddion rhoi gwybod i deuluoedd am ein hymchwil, amgacir gwybodaeth am yr astudiaeth (a ysgrifennwyd ar gyfer teuluoedd). Rydym yn gofyn i sefydliadau anfon gwybodaeth allan ar ein rhan. Gofynnir i deuluoedd gysylltu'n uniongyrchol â ni wedyn. Byddem yn darparu'r holl gopïau o wybodaeth sydd eu hangen ynghyd ag amlenni i'w dosbarthu ac yn talu am unrhyw gostau postio. Hyd nes y bydd teuluoedd yn cysylltu â ni, ni fydd y tîm ymchwil yn gwybod pwy ydya nhw. Nid ydym yn gofyn i chi roi unrhyw fanylion am deuluoedd i ni.

Byddaf yn ffonio'ch sefydliad yn y 7-10 diwrnod nesaf i drafod cynnwys y llythyr hwn ymhellach ac i ateb unrhyw gwestiynau a all fod gennych. Byddwn yn hapus i siarad â'ch ysgrifennydd os gellwch udae neges addas i ni gydag ef neu hi. Cweso i chi gysylltu'n uniongyrchol â mi os dymunwch:

e-bost: d.reilly@bangor.ac.uk neu ffon 44 (0) 1248 351151 ext.8706

Diolch yn fawr i chi am eich amser, a gobeithio'n fawr iawn y byddwch yn gallu ein helpu.

Yn gywir,

Deirdre Reilly

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Appendix C

Letter to Families English Version



The Wales Study of Bereavement in Families of Individuals with a Learning Disability.

Astudiaeth Profedigaeth mewn Teuluoedd Pobl ag Anabledd Dysgu yng Nghymru



Dear Sir/ Madam,

I am contacting you to ask for our help with The Wales Study of Bereavement in Families of Individuals with a Learning Disability.

Our research aims to understand the experience of parenting, growing up with and then losing a child with a learning disability.

By listening to the experiences of people like yourself and others in your family we hope to understand more about what happens to people in this situation and about the services and support that are essential and appropriate at this time. The information we gather will be used to develop a training package for professionals and service providers so that they can better support families in the future.

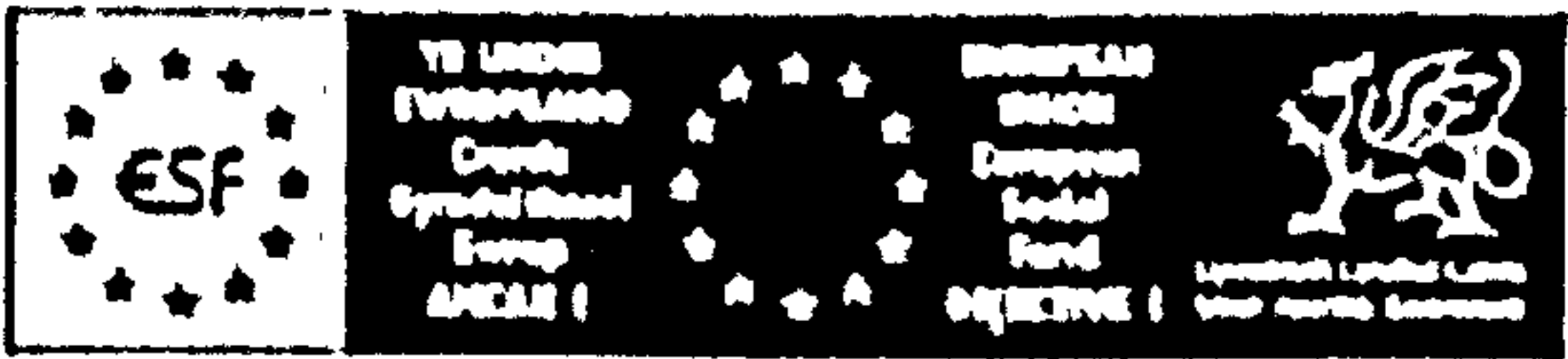
In order for our research to reflect a representative range of situations and experience, we would like to talk to as many families as possible. If you would like to contribute to the research please complete the attached form and return it to the address below.

We recognise that the subject of our research is likely to be painful for you to talk about at times. We will try to be as sensitive to your needs as possible and you would be under no pressure to continue with your participation if you were uncomfortable or reluctant. Attached to this letter is some more information about the study.

Thank you for your time. If you have any questions please contact:

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Appendix D

Letter to Families Welsh Version



The Wales Study of Bereavement in Families of Individuals with a Learning Disability.

Astudiaeth Profedigaeth mewn Teuluoedd Pobl ag Anabledd Dysgu yng Nghymru



Annwyl Syr/Fadam,

Rydw i'n cysylltu â chi i ofyn am eich help gydag Astudiaeth o Brifedigaeth yn Nheuluoedd Pobl ag Anabledd Dysgu yng Nghymru.

Bwriad ein hymchwil yw deall y profiad o fod yn rhiant, magu plentyn ag anabledd dysgu ac yna ei golli, am unrhyw heneiddia.

Trwy wrando ar bruliadau pobl fel chi ac eraill yn eich teulu ein gohanti yw y byddwn yn deall mwy am yr hyn sy'n digwydd i bobl yn y sefyllfa hon ac am y gwasanaethau a'r cymorth sydd yn angenrheidiol ac yn briodol ar y pryd. Bydd y wybodaeth byddwn yn ei chasglu yn cael ei defnyddio i ddatblygu pecyn hyfforddi i weithwyr proffesiynol a darparwyr gwasanaeth fel y gallan nhw gynorthwyo teuluoedd yn well yn y dyfodol.

Er mwyn sicrhau bod ein hymchwil yn adlewyrchu amrywiaeth teg o sefyllfannedd a phrofiad, hoffem siarad â chynifer o deuluoedd â phosib. Os hoffech gyfrannu at yr ymchwil llenwch y ffurflen amgacedig a'i hanfon yn ôl i'r cyfeiriad isod.

Rydym yn sylweddoli ei bod yn bosib iawn y bydd maes ein hymchwil yn un y bydd yn boenus i chi siarad amdano ar brydiau. Byddwn ni'n ceisio bod mor sensitif i'ch anghenion â phosib ac ni fydddech o dan unrhyw bwysau i barhau i gymryd rhan pe baech yn teimlo'n anghyfforddus neu'n anfodlon. Amgair mwy o wybodaeth am yr astudiaeth yn y llythyr hwn. Diolch i chi am eich amser. Os oes gennych chi unrhyw gwestiynau cysylltwch â:

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Appendix E

Information Sheet for Families English Version

### **Information Sheet for Families**

#### **1. Study Title**

**The Wales Study of Bereavement in Families of Individuals with a Learning Disability.**

#### **2. Research Team**

**Deirdre Reilly, Research Student**

**Richard Hastings, Professor of Psychology**

**Frances Vaughan, Clinical Psychologist.**

**Jaci Huws, Lecturer, School of Nursing.**

**The research is a collaboration between the University of Wales, Bangor and Mencap Cymru. Mencap Cymru are also part funding the research along with the European Social Fund (ESF).**

#### **3. What is the purpose of the study?**

**We are interested in how families cope with bereavement following the death of a child, including adult children, with a learning disability. We want to look at the circumstances surrounding their loss and their feelings about their experience. There has been little research in this area in the past but it is much needed. If service providers are to develop palliative and bereavement care services for families it is imperative that parents and other family members are involved and have the opportunity to aid the development of these services. We are seeking responses from all over the UK and Ireland so that we can get a good idea of the various services available in different areas.**

#### **4. Invitation to participate**

**We wish to identify parents, grandparents and siblings over the age of 18 who have lost a family member with a learning disability and who live in the UK or Ireland and are asking participants to fill a set of questionnaires on their experiences. Please read the remainder of this information sheet carefully and complete the form attached if you are interested in helping us with this research. If anything is unclear, or you would just like more information before you decide, please fill in the Initial Contact Form and we will 'phone you to discuss the project further and answer any questions you may have.**



We will try to contact each family only once. If you have already taken part in the study, thank you for your time, there is no need to reply a second time.

#### **5. What are the benefits of taking part?**

The main benefits of this research relate to improving the knowledge that we have about bereaved families and the services that they require. This research project has already investigated the experiences of parents through qualitative telephone interviews and these questionnaires are designed to strengthen the accounts detailed by those parents. Service providers say that they lack knowledge on the requirements of families at this time, and your participation will help inform and promote developments in this area.

We plan to keep participants up-to-date with the project's progress through a website which will include links to relevant organisations, a newsletter that we will post to participants, and a facility to request full copies of research publications associated with the project.

#### **6. What are the risks of taking part?**

We do not believe that you are at risk of any harm from taking part in this study. Parents who have taken part in bereavement research previously report that the process was very positive, with none regretting having participated. Although 75% of interviewees in one study reported the experience was painful to some extent they also highlighted their positive experiences, which included a hope that they might help others.

All information you provide will be treated as strictly confidential and will be kept securely locked in a filing cabinet without your names attached. Only the researcher will have access to questionnaires, which will be destroyed once the study has been completed.

#### **7. Do we have to take part?**

It is up to you to decide whether you want to take part. If you do decide to take part, or want further information, please return the Initial Contact Form to the address provided. If more than one member of your family wishes to take part please provide us with their contact details and we will provide them with an information pack and contact you all

individually. You may keep this information sheet for your records. You are still free to withdraw from the research at any time and without giving a reason.

#### **8. What will happen to me if I take part?**

After you have returned the Initial Contact form, we will go through the following contact process.

- i. We will telephone you to answer any further questions if you have requested this.
- ii. We will post you consent form and a set of questionnaires, which will ask a few more questions about you, your family and your child, sibling or grandchild. The questionnaires will ask for information on your supports, ways of coping and adjustment to the death.
- iii. We will send you information on the results of the study and give you the address of the project website. We will also provide you with a list organisations who can offer advice and support.

#### **9. What do I have to do now?**

If, having considered this information, you would like to discuss the study further, or you would like to take part, then please return the Initial Contact Form. If you decide not to take part, please discard this letter. You do not need to make contact with us.

All the information that you give us will be treated as strictly confidential, and will be kept securely locked in a filing cabinet without your names attached. The information that you provide will not be used in any way that would identify you as an individual or be discussed with any other member of your family under any circumstances.

**10. Further details.**

If you wish to contact the research team our details are below:

Deirdre Reilly: Email: [d.reilly@bangor.ac.uk](mailto:d.reilly@bangor.ac.uk) or Phone +441248 351151 ext 8706

School of Psychology,  
University of Wales, Bangor,  
Adeliad Brigantia,  
Penrallt Rd.  
Bangor,  
Gwynedd,  
LL57 2AS

If you have any complaints about the way that this research is being conducted you are welcome to address unresolved concerns to:

Dr. E. Charles Leek,  
Deputy Head of School,  
School of Psychology,  
University of Wales, Bangor,  
Adeliad Brigantia,  
Penrallt Rd.  
Bangor,  
Gwynedd,  
LL57 2AS

Appendix F

Information for Families Welsh Version



## **Taflen Wybodaeth i Deuluoedd**

### **11. Teitl yr Astudiaeth**

Astudiaeth o Brofedigaeth yn Nheuluoedd Pobl ag Anabledd Dysgu yng Nghymru.

### **12. Tîm Ymchwil**

Deirdre Reilly, Myfyriwr Ymchwil

Richard Hastings, Athro Seicoleg

Frances Vaughan, Seicolegydd Clinigol.

Mae'r ymchwil hon ar y cyd rhwng Prifysgol Cymru, Bangor a Mencap Cymru. Mae Mencap Cymru'n ariannu'r ymchwil yn rhannol hefyd ar y cyd â Chronfa Gymdeithasol Ewrop (ESF).

### **13. Beth yw pwrpas yr astudiaeth?**

Mae gennym ddiddordeb mewn darganfod sut mae teuluoedd yn ymdopi â phrofedigaeth yn dilyn marwolaeth plentyn ag anabledd dysgu. Dymunwn nodi'r cymorth a'r gwasanaethau a dderbyniwyd ganddynt, cyn, yn ystod ac ar ôl eu profedigaeth a sut y gall y rhain effeithio ar brofiadau teuluoedd yn dilyn colled. Ychydig o ymchwil a fu yn y maes hwn ond mae gwir angen amdano. Er mwyn i ddarparwyr gwasanaeth ddatblygu gwasanaethau gofal lliniarol a phrofedigaeth i deuluoedd mae'n hollbwysig bod rhieni ac aelodau eraill o'r teulu yn cymryd rhan yn y gwaith a'u bod yn cael y cyfle i helpu i ddatblygu'r gwasanaethau hyn. Rydym ni eisiau siarad â theuluoedd o bob cwr o'r DU ac Iwerddon fel y gallwn ni gael syniad da o'r gwahanol wasanaethau sydd ar gael mewn gwahanol ardaloedd.

### **14. Gwahoddiad i gymryd rhan**

Rydym ni eisiau gwybod am rieni, neiniau a theidiau a brodyr neu chwiorydd dros 18 oed sydd wedi colli aelod o'r teulu ag anabledd dysgu ac sy'n byw yn y DU neu Iwerddon. Darllenwch weddill y daflen wybodaeth hon yn ofalus a llenwch y ffurflen amgaeedig os hoffech ein helpu yn yr ymchwil. Os yw rhywbeth yn aneglur, neu os hoffech chi gael mwy o wybodaeth cyn i chi benderfynu, llenwch y Ffurflen Cyswllt Cychwynnol ac fe wnawn eich ffonio i drafod y project ymhellach ac ateb unrhyw gwestiynau posib.

Ceisiwn gysylltu â phob teulu unwaith yn unig. Ond gan y byddwn yn dosbarthu gwybodaeth trwy gyfrwng nifer o sefydliadau, efallai byddwch yn derbyn mwy nag un copi o'r daflen wybodaeth hon ac o'r gwahoddiad i gymryd rhan yn yr astudiaeth. Ymddiheurwn am hyn os bydd yn digwydd: dim ond unwaith y mac'n rhaid i chi ymateb.

#### **15. Beth yw manteision cymryd rhan?**

Prif fanteision yr ymchwil hOn fydd gwella'r wybodaeth sydd gennym am deuluoedd sydd wedi cael profedigaeth a'r gwasanaethau sydd eu hangen arnynt. Trwy roi cyfle i deuluoedd i ddweud eu hanes yn llawn bydd gennym gorff o wybodaeth y gallwn ei ddefnyddio wedyn i gyfarwyddo a hyfforddi gweithwyr proffesiynol i gynorthwyo teuluoedd ar yr adeg hon a helpu unigolion i sicrhau canlyniadau cadarnhaol lle bo hynny'n bosib. Mae darparwyr gwasanaeth yn dweud nad oes ganddynt ddigon o wybodaeth am anghenion teuluoedd ar yr adeg hon, a bydd eich cyfraniad chi'n helpu i lywio a hybu datblygiadau yn y maes hwn,

Ein cynllun yw rhoi'r newyddion diweddaraf i'r rhai sy'n cymryd rhan am gynnydd y project trwy wefan a fydd yn cynnwys cysylltiadau â sefydliadau perthnasol, cylchlythyr y byddwn yn ei anfon at y rhai sy'n cymryd rhan, a chysleuster i ofyn am gopïau llawn o gyhoeddiadau ymchwil sy'n gysylltiedig â'r project.

#### **16. Beth yw risgiau cymryd rhan?**

Nid ydym o'r farn y byddwch mewn perygl o gael unrhyw niwed os byddwch yn cymryd rhan yn yr astudiaeth hon. Mae rhieni sydd wedi cymryd rhan mewn ymchwil ar brofedigaeth o'r blaen yn adrodd bod y broses wedi bod yn gadarnhaol iawn, ac nid oes neb yn edifar eu bod wedi cymryd rhan. Er bod 75% o'r rhai a gafodd eu cyfweld mewn un astudiaeth yn adrodd bod y profiad yn boenus i ryw raddau roeddent hefyd yn tynnu sylw at eu profiadau cadarnhaol. Roedd y rhain yn gysylltiedig â chael dweud eu hanes yn llawn, fformat y cyfweiliad, a'r gobaith y bydden nhw'n gallu helpu eraill.

Bydd yr holl wybodaeth y byddwch yn ei darparu yn cael ei thrin yn gwbl gyfrinachol a bydd yn cael ei chadw'n ddiogel dan glo mewn cwpwrdd ffeilio heb fod eich enwau arll. Dim ond yr ymchwilydd fydd yn cael mynd at dapiau'r cyfweiliadau a'r holiaduron. Bydd y rhain yn cael eu dinistrio unwaith y bydd yr astudiacth wedi dod i ben.

### **17. A oes rhaid i ni gymryd rhan?**

Chi sydd i benderfynu a ydych am gymryd rhan ai peidio. Os byddwch yn penderfynu cymryd rhan, neu os hoffech gael mwy o wybodaeth llofnodwch y Ffurflen Cyswllt Cychwynnol a'i hanfon yn ôl atom.. Os yw mwy nag un aelod o'ch teulu'n dymuno cymryd rhan rhowch eu manylion cyswllt i ni a byddwn yn rhoi pecyn gwybodaeth iddyn nhw a chysylltu â chi i gyd yn unigol. Gellwch gadw'r daflen wybodaeth hon ar gyfer eich cofnodion. Rydych yn dal yn rhydd i dynnu'n ôl o'r ymchwil ar unrhyw adeg a heb roi rheswm.

### **18. Beth fydd yn digwydd i mi os byddaf yn cymryd rhan?**

Ar ôl i chi anfon y Ffurflen Cyswllt Cychwynnol yn ôl, byddwn yn dilyn y broses gysylltu ganlynol.

- i. Byddwn yn eich ffonio i ateb unrhyw gwestiynau pellach a threfnu amser i'ch ffonio a chynnal cyfweiliad dros y ffôn sy'n gyfleus i chi.
- ii. Byddwn yn anfon ffurflen ganiatâd i'w llofnodi i ddweud eich bod yn fodlon cymryd rhan yn yr ymchwil hon. Rhaid i chi lenwi hon a'i hanfon yn ôl cyn i'r cyfweiliad gael ei gynnal.
- iii. Yn ystod y cyfweiliad dros y ffôn byddwn yn gofyn cwestiynau amdanoch chi, eich teulu a'ch plentyn, ŵyr/wyres neu frawd/chwaer sydd wedi marw. Byddwn yn gofyn yn benodol am eich teimladau trwy'r broses, pa wasanaethau a chymorth a ddefnyddiwyd gennych, sut yr ydych wedi ymdopi, beth allai fod wedi gwneud y broses yn haws, sut yr ydych wedi gwneud synnwyr o'ch colled ac a ydych wedi ennill unrhyw brofiadau cadarnhaol o'r broses gyfan.



- iv. Ar ôl gorffen y cyfweiliad bydd yn anfon set o holiaduron atoch, a fydd yn gofyn ychydig rhagor o gwestiynau amdanoch chi, eich teulu a'ch plentyn, eich brawd/chwaer neu'ch wŷr/wyres. Bydd yr holiaduron yn gofyn am wybodaeth am y pethau a fu'n gymorth i chi, sut y llwyddasoch i ymdopi a sut yr ydych wedi addasu i'r golled.
- v. Byddwn yn anfon gwybodaeth atoch am ganlyniadau'r astudiaeth ac yn rhoi cyfeiriad gwefan y project i chi. Byddwn hefyd yn rhoi rhestr i chi o sefydliadau sy'n gallu cynnig cyngor a chymorth.
- vi. Byddwn yn defnyddio'r wybodaeth y byddwn yn ei chasglu i ddatblygu pecyn cymorth i staff sy'n gweithio mewn gwasanaethau anabledd dysgu a gwasanaethau meddygol, yn rhoi gwybod iddynt am y pethau a fu'n gymorth i deuluoedd a'r hyn gallan nhw ei wneud i helpu teuluoedd ar yr adeg anodd hon. Mae'n bosib y bydd y wybodaeth a gynhyrchir yn cynnwys dyfyniadau byr a manylion cyfyngedig am brofiadau neilltuol. Ni fyddwn yn enwi unigolion, oni bai fod disgrifiadau helaeth yn cael eu defnyddio. Mewn achos felly byddwn yn cysylltu cto â theuluoedd i ofyn caniatâd.

### **19. Beth mae'n rhaid i mi ei wneud yn awr?**

Ar ôl i chi ystyried y wybodaeth hon, os hoffech drafod yr astudiaeth ymhellach, neu os hoffech gymryd rhan, yna anfonwch y Ffurflen Cyswllt Cychwynnol. Os penderfynwch beidio â chymryd rhan, taflwch y llythyr hwn. Nid oes rhaid i chi gysylltu â ni.

Bydd yr holl wybodaeth y byddwch yn ei rhoi i ni yn cael ei thrin yn gwbl gyfrinachol a bydd yn cael ei chadw'n ddiogel dan glo mewn cwpwrdd ffeilio heb fod eich enwau arni. Ni fydd y wybodaeth y byddwch yn ei darparu yn cael ei defnyddio mewn unrhyw ffordd a allai ddangos pwy ydych chi. Ni fydd eich cyfweiliad yn cael ei drafod gydag unrhyw aelod arall o'ch teulu o dan unrhyw amgylchiadau.



**20. Manylion pellach.**

Os ydych yn dymuno cysylltu â'r tîm ymchwil mac ein manylion isod:

Deirdre Reilly: [d.reilly@bangor.ac.uk](mailto:d.reilly@bangor.ac.uk) neu Ffôn +441248 351151 est 8706

Ysgol Seicoleg

Prifysgol Cymru, Bangor,

Adeliad Brigantia

Ffordd Penallt,

Bangor, Gwynedd

LL57 2AS

Os oes gennych chi unrhyw gŵynion am y ffordd y mae'r ymchwil hon yn cael ei chynnal croeso i chi anfon llythyr yn sôn am unrhyw bryderon heb eu datrys at:

Dr. E. Charles Leek,

Dirprwy Bennaeth yr Ysgol

Ysgol Seicoleg,

Prifysgol Cymru, Bangor

Adeilad Brigantia,

Ffordd Penrallt,

Bangor, Gwynedd,

LL57 2AS

Appendix G

Initial Contact Form English Version

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## Initial Contact Form

**Please read the following, place a tick in the appropriate boxes, and return the form to the address provided.**

- ☐ I would like more information before I decide to take part in the study. *Please complete the information below.*
- ☐ I would like to complete the questionnaires. *Please complete the information below.*

**Please tell us whether any other members of your family (for example parent, sibling or grandparent) are willing to participate in the research. We will contact them.**

**Other members of my family would like more information on the research:**

- ☐ Yes ☐ No *If yes, please include their contact details below or overleaf*

**Your Name (please print) and relationship to the deceased:**

**Your contact address & telephone number:**

**Telephone**

**Other family members' contact details:**

**Telephone** \_\_\_\_\_

### Other family members' contact details

**Telephone**

**Please tell us which days and what times of day are best to contact you and them:**

***Return this sheet to me at the university and I will contact you. Thank you for your time, Deirdre Reilly.***

Appendix H

Initial Contact Form Welsh Version



**Ffurflen Cyswllt Cychwynnol**

**Astudiaeth o Brofedigaeth yn Nheuluocdd Unigolion ag Anabledd Dysgu yng Nghymru.**

Darllenwch y canlynol, ticiwch y blychau priodol, yna anfonwch y ffurflen yn ôl l'r cyfeririad a ddarperir.

- ☐ Hoffwn gael mwy o wybodaeth cyn i mi benderfynu cymryd rhan yr astudiaeth.  
*Cwblhewch y wybodaeth isod.*
- ☐ Hoffwn gymryd rhan yr astudiaeth. *Cwblhewch y wybodaeth isod.*

Dywedwch wrthym a fyddai unrhyw aelodau eraill o'ch teulu (er enghraifft rhiant, brawd/chwaer neu daid/nain) yn fodlon cymryd rhan yn yr ymchwil efallai. Fe wnawn anfon pecyn gwybodaeth atynt.

Byddai aelodau eraill o'm teulu yn hoffi cael mwy o wybodaeth am yr ymchwil.

- ☐ Byddent
- ☐ Na fyddent. *Os byddent, nodwch eu manylion cyswllt isod neu dros y dudalen*

Eich Enw (printiwch) a'ch perthynas â'r sawl sydd wedi marw:

Eich cyfeiriad cyswllt a'ch rhif ffôn:

Cod post Ffôn

Manylion cyswllt aelodau eraill o'r teulu:

Cod post Ffôn

Manylion cyswllt aelodau eraill o'r teulu:

Cod post Ffôn

Dywedwch wrthym ba ddiwrnodau a pha adegau o'r dydd fyddai orau i gysylltu â chi ac â nhw:

*Anfonwch y dudalen hon yn ôl ataf i'r brifysgol a byddaf yn eich ffonio. Diolch yn fawr i chi am eich amser.*

*Deirdre Reilly.*

Appendix I

Consent Form



The Wales Study of Bereavement in Families of Individuals with a Learning Disability.

Astudiaeth Profedigueth mewn Teuluoedd Pobl ag Anabledd Dysgu yng Nghymru



Participant identification number:

Consent Form to Participate In Study

Name of researchers: Deirdre Reilly supervised by Prof. Richard Hastings; Jael Huws; and Dr. Frances Vaughan

Full information on the nature of the research, procedures, benefits and harms are detailed in the Information for Families Leaflet

Please complete the following and delete as necessary:

- Have you read the Information for Families Leaflet?YES/NO
- Have you had an opportunity to ask questions and discuss this study?YES/NO
- Have you received enough information about this study?YES/NO
- Do you understand that you are free to withdraw from this study:  
.....at any time  
.....without giving a reason for withdrawingYES/NO

I am willing to participate in this study. YES/NO

Signature\_\_\_\_\_

Date \_\_\_\_\_

Name in block letters \_\_\_\_\_

Address \_\_\_\_\_

Postcode \_\_\_\_\_Tel. No. \_\_\_\_\_

Researcher Signature\_\_\_\_\_Date \_\_\_\_\_

Ysgol Seicoleg  
Prifysgol Cymru Bangor  
Ardedol Brigantia, Ffordd Penallt  
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Appendix J

Interview Schedule



### Introduction

- Tell me about your family
  - How many people live in your home
  - How many children do you have
  - What are their names
  - What ages are your children
  - Do any of your children have a disability
  - Tell me about this disability
- Tell me about your child
  - What was your child like
  - How did you get on together
- \*Tell me about a special memory (if parent is having trouble talking on subject)
- \*Was there a particular activity your child especially enjoyed?
- Tell me about your child's disability
  - When was your child diagnosed

### Loss

- Tell me about the death of your child
- What age was your child when he/she died?
- How long ago did they die?
- How did your child die?

### If expected:

- How long before death did you know your child was going to die?
  - How/where/from whom did you receive the news?
  - What were the problems identified?
  - Was the information easy to understand/accurate/
  - What medical services did you use prior to your child's death
  - Did you find these adequate
  - What were the ongoing care requirements of your child in the lead up to their death?

- Do you feel the diagnosis could have been made earlier
- How did you cope at this time?
  - What support were you offered
  - What support did you find helpful
  - How was this a help to you during this time?
  - How did you deal with your feelings/situation
  - What might have made things easier at this time
- Tell me about the time between diagnosis and death
  - How did you feel
  - What was your relationship with your child like
  - What was your relationship with your family like?
  - Was there anything you did as a family that helped during this period
  - What support did you have at this time?
  - Who helped you out
  - How did you find this helpful?
  - Was there anything that may have made this time easier?
- Tell me about the time just before the death of your child
  - What helped you during this time
  - What were the milestones you had to overcome
  - What would have been helpful during this time
- Did you feel involved in the decisions surrounding your child's death
  - How did it feel to be involved/ uninvolved
  - Was the family included in decisions surrounding treatments,
- Were the health professionals adequately trained to meet your child's needs
  - Was there anything more that could have been done to make your child comfortable?
  - Did you feel welcome in hospital clinic etc
  - Is there anything else you would like to say about this?
- Were you prepared for the loss of your child?
  - Did you prepare yourself for the loss of your child?

- If so- what did you do to prepare for the loss of your child?
  - Was there anything you feel now you should have done at the time
  - Did you prepare your family/children for the loss?
  - If so -how did you prepared your children for the death of their brother/sister
- **If sudden or relatively sudden:**
  - How long before your child died were you informed?
  - Who told you?
  - What problems were identified?
  - Did you receive adequate/ comprehensible information
  - Were there any factors that you feel could have been identified earlier?
  - What medical services did your child receive before dying?
  - How would you evaluate these services?
  - Is there anything else you would like to say about this?
- **Did you have any opportunity to prepare for the death of your child?**
  - Prepared Questions above
- **Tell me about the time immediately after the death of your child**
  - How did you feel at this time
  - What did you do to cope at this time
- **What did you find helpful following the death of your child?**
  - Who helped you at this time
  - How did you find help
  - What made things easier
  - Why did this make things easier?
  - How did you use this resource
  - Did you get support from the medical community?
  - Was this support readily available?
  - Did you receive professional grief counselling?
  - Did you find this useful?
  - Were you still in contact with service providers
  - Did you find this helpful?

- What was your relationship with your family/spouse like immediately following the death of your child?
  - How did you cope as a couple
  - Did the death affect your relationship?
  - Was there anything that might have helped you as a couple
  - Is there anything you did that you found particularly helpful?

### *Time Since Loss*

- In the time since losing your child have things changed?
  - Do you still rely on support from professionals or services?
- How are you coping now?
  - What do you do to make things easier for yourself?
  - Do you find it hard to get through each day
  - Do you think of your child a lot/ how do you cope with this
  - Is there anything those around you could do to make things easier for you?
  - Has the loss of your child affected your mental health or health generally?
- Has your child's death affected your relationship with your family?
  - How do you cope as a family?
  - Is there anything you do as a family that you find helpful?
  - Do you talk about \_\_\_ at home
  - Did the death of your child change your plans for your family's future?
  - Are you closer as a result of \_' s death?
  - How are things different since \_\_\_ died?
  - Has your marital situation changed since the death of \_\_\_
- How did your child's death affect other family members?
  - How did \_\_\_'s siblings respond to the loss of their brother/sister
  - Is there any support they have received that they found useful?
  - Is there anything that could make things easier for them now?
  - How has your spouse been affected by the loss?
  - Is there any support your spouse found particularly useful



- Has your relationship with your spouse changed since the death of \_\_\_\_
  - What do you and your spouse find helped you as a couple
- How has your child's death affected your relationships with friends, acquaintances or professionals?
  - We may have touched on this earlier... would you like to add anything
  - Were there particular people who affected your life more than others
  - Is there perhaps one person who has been of great assistance to you throughout this process?
  - Has your view of others/professionals changed because of \_\_\_\_'s death?
  - Did you receive adequate information about your child's prognosis

### Reflection

- Do you feel you have been able to make sense of the death?
- Sometimes people who lose a loved one find some positive aspect in the experience (e.g. learning something about self). Have you found anything positive in this experience?

Further questions if respondents don't reply to above

- Have your experiences changed your outlook
  - Has your child affected the way you live your life?
  - Has your child's death affected your view of the world?
- How has your child's death affected your sense of priorities?
  - Has your outlook on life changed?
  - Have you changed anything in your life/career
  - Do you think about your family's future more?
  - Was your child an inspiration to improve your skills/situation
- How has the loss of your child affected your sense of identity
  - Have you changed as a person
  - Do you see a change in yourself from the beginning of the process
- How do you think now about your child's life?
  - Has your child's death affected your spirituality?
  - Are you any more or less religious

- Do you see a meaning in the life and death of your child

**Interview End**

- What would be most helpful or healing now?
  - Is there anything that might make things easier
  - How do you think you can move on
- What advice would you give parents in a similar situation

**Suggestions for parents finding it difficult to think positively of the experience:**

- Do you feel you have grown as a person
- Have your perspectives on life changed as a result of your experience
- Have you developed new relationships or have old ones been strengthened because of your experience?

**Additional filler questions may be required to expand some answers.**

**Examples are:**

- We have talked about this already is there anything you'd like to add?
- You mentioned \_\_\_\_, could you tell me more about this
- Would you like to add anything to that comment?

Appendix K

Demographic questionnaire

*The Wales Study of Bereavement in Families of Individuals with a Learning Disability*

*The following questions ask for background information about you, your child who died, and your family. Please tick the appropriate boxes or write in the spaces provided.*

These questions are about you:

1. Are you male or female? ☐ Male ☐ Female

2. What was your age in years on your last birthday? \_\_\_\_\_

3. What is your current marital status?

Married, and living with spouse..... ☐

Living with partner..... ☐

Divorced/Separated/Single and NOT living with a partner..... ☐

4. What is your ethnic background? Please tick

Welsh	English	Scottish
African	Afro-Caribbean	Bangladeshi
Chinese	Indian	Irish
Other European	Pakistani	Other .

5. Do you currently have a job outside the home? ☐ Yes ☐ No

*If no, please go to question 7, otherwise please answer following questions.*

What is your current job/occupation? (Please give a job title and a very brief description of your main duties).

\_\_\_\_\_

\_\_\_\_\_

\_\_\_\_\_

6. Is your job/occupation full or part-time? ☐ Full-time ☐ Part-time



7. If you are living with your spouse/partner, do they currently have a job outside the home? Yes ☐ No ☐

*If no, please go to question 9, otherwise please answer following questions.*

What is your spouse/partner's current job/occupation? (Please give a job title and a very brief description of their main duties).

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8. Is this job/occupation full or part-time? ☐ Full-time ☐ Part-time

The following questions are about your child:

9. How long ago did your child die? \_\_\_\_\_years \_\_\_\_\_months

10. Was your child male or female? ☐ Male ☐ Female

11. What age was your child when they died? \_\_\_\_\_years \_\_\_\_\_months

12. What was your relationship to your child who died?

Biological parent ☐ Step-parent ☐  
Foster-parent ☐ Adoptive parent ☐

Other\_\_\_\_\_.

13. Did your child live with you? Yes ☐ No ☐

If no, where did your child live? \_\_\_\_\_.

14. In total how many people currently live in your house? \_\_\_\_ Adults \_\_\_\_ Children

If there are other children living in the house how are they related to your child who died (e.g. biological brother, step brother), do they have a learning disability, and how old are they?  
*Please list ALL children*

.....  
.....  
.....  
.....

**15. Tick the boxes below to indicate any diagnoses/conditions that applied to your child who died.**  
Please tick all that apply

- Learning Disability (“Intellectual Disability”)..... ☐
- Down Syndrome..... ☐
- Congenital Heart Condition..... ☐
- Epilepsy..... ☐
- Other syndrome (please specify) \_\_\_\_\_

**16. Did your child with special needs have any other health problems Yes ☐ No ☐ not already mentioned?**

If yes, then please specify \_\_\_\_\_

**17. What was the cause of death? Please describe in the space below**

\_\_\_\_\_

\_\_\_\_\_

\_\_\_\_\_

\_\_\_\_\_

\_\_\_\_\_

**18. Was the death sudden or expected?      Sudden ☐      Unexpected ☐**

Appendix L

Revised Grief Experienced Inventory

PAGE/PAGES  
EXCLUDED  
UNDER  
INSTRUCTION  
FROM  
UNIVERSITY



Appendix M

Active Avoidance and Problem-Focused subscales of the Brief Cope

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EXCLUDED  
UNDER  
INSTRUCTION  
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Appendix N

Positive Contributions Scale (Kansas Inventory of Positive Perceptions)

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Appendix O

Excerpt from the Booklet of Contacts for Families

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Appendix P

Expanded explanation of the process of transcription.

The process of analysis began at the start of data collection and continued until the write up of the interviews and corresponding themes was complete. Each interview was carried out by the primary researcher with one interviewee at a time over the telephone and recorded onto a digital voice recorder. The researcher kept a reflexive diary while carrying out interviews to note any additional thoughts on the interview, notes on interruptions, and thoughts to arise from the experience. The length of interviews varied and the order in which topics were covered was led predominantly by the parent, although the interviews generally took the form of a story through the child's life, death, and the time since the death. The interviews were emotional for all of the parents and most cried at some stage although none requested that the interview be ended prematurely and at no point did the process become an overly emotional experience for the researcher.

Using the reflexive diary allowed the researcher and her supervisors to monitor the impact this role. Reflective notes made on this issue included comments as to the time parents took when talking about the moment of death. While the questions asked were very general e.g. "Could you tell me about when he/she died?" the responses were always very detailed and specific and parents often described at length the time preceding the death and the death itself. The reflexive diary also served as a memory aid for the researcher by indicating general issues such as tone of the interview and reasons behind interruptions and gaps in interviews.

Once all the interviews were complete the interviews were transcribed by an external source. This method of not reviewing and analysing transcripts until all the interviews had been completed ensured the process of analysis did not impact on the style or content of the interview. At the beginning of the qualitative analysis process itself the researcher listened to the audio file of the interview twice or three times while reading the typed transcript and



noting any relevant issues from the reflexive diary. This ensured the researcher was fully familiarised with the interview. The transcripts were then printed landscape style on A4 paper. The interviews were double-spaced and line numbers were added to ease the finding of quotes at later stages of the analysis. Then, the process of analysis generally involved two readings of the transcripts with the researcher noting thoughts and topics of interest in the left hand margin. Subsequent readings aimed to combine thoughts in the left hand margin into emerging themes, which were noted in the right hand margin. Once all emerging themes had been noted on the transcript in the right hand margin a list of these themes was typed separately and a catalogue of the page number, line number, and actual text of the quote for each occurrence of a theme was created in a computer spreadsheet. The broader theme titles were added to a separate spreadsheet for comparison with the other interviews.

The researcher then began the process again for the second interview. Once the themes had been noted on the right hand side a catalogue of themes and quotes was created. These broad themes were added to those noted in the first interview and a complete list of master themes began to develop. This process continued for each interview until a catalogue of themes and excerpts had been created for each interview and the master list of themes contained all the themes to emerge from the interviews. This list of master themes developed gradually as themes merged and became more or less visible throughout the process of analysis.

Meanwhile, a second researcher with considerable expertise in IPA methods and analysis read a copy of each interview while viewing the catalogue of themes and excerpts and the list of master themes. After reading each interview both researchers discussed the relevance and accuracy of the themes. This process of discussion resulted in changes to the interpretation of the interviews, for example, the process of analysis began to focus more on

the individual experiences of the parents as opposed to the more general experience of childhood illness and disability. Queries also often arose between researchers as to the intention behind particular comments; these queries were resolved by listening to the audio of the interview or referring to the reflexive diary.

Once all interviews had been analysed by both researchers they analysed the list of master themes further. Themes were combined and condensed until both researchers agreed the list was an accurate reflection of the interviews with parents. The themes were then written up in the style of the results section of a journal article. All names were changed and references that could make parents identifiable, such as references to particular hospitals, were removed. Each theme was illustrated by a number of examples from interviews and a discussion of the similarities and difference between accounts. In parallel to this process the themes continued to develop and combine into a small number of descriptive categories. Discussion between researchers and input from supervisors ensured that the themes gradually became more coherent and only the most suitable quotes were used to illustrate themes, this strengthened the discussion points raised and displayed how themes were visibly grounded in the data.

The product of this process of analysis will be feedback services as an accessible report currently in preparation in conjunction with Mencap Cymru. It is planned the themes will inform staff training considerations and influence the availability and format of bereavement support for parents of a child with an intellectual disability, who may be in need of support. These final parts of the project are currently underway.