VOLUME 1

RESILIENCE IN PARENTS OF ADULTS WITH INTELLECTUAL DISABILITY

BY

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A thesis submitted to the University of Birmingham
for the degree of
DOCTORATE IN CLINICAL PSYCHOLOGY

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June 2015

Dedication

I would like to dedicate this doctorate to my parents, I could not have done this without you.

Acknowledgements

I would like to thank the following people - who without this thesis would not have been possible:

All the families who agreed to take part in the research study, for allowing me into their homes and taking the time to share their stories.

Chris Oliver and Beth Grunfeld for supervision and guidance throughout the research process.

Janet Carr for support with the recruitment of families, and for being an inspiration in terms of research in the field of Down syndrome.

The service-users and teams I have worked with across my placements during training, who have helped me to develop my clinical skills. A special thank you to the service-users who consented for me to write up and present their cases for the clinical component of the thesis.

Finally, a heartfelt thank you to my family, particularly my parents and Luc, for your ongoing love, support and belief in me.

Thesis overview

This thesis is submitted in partial fulfilment of the requirements of the degree of Doctorate of Clinical Psychology at the University of Birmingham. It comprises both research and clinical components of the course. All identifying information has been changed throughout to ensure participants' confidentiality.

Volume I of the thesis comprises the research component and contains two research papers. The first is a systematic literature review which examines quantitative research reporting psychosocial outcomes of caring for an adult child with intellectual disability. The second paper is an empirical qualitative study which explores the experience of caring for a child with Down syndrome across 50 years. The research papers are followed by an executive summary of the two papers for distribution of findings within the public domain.

Volume II comprises the clinical component and contains five Clinical Practice Reports (CPRs). The CPRs demonstrate clinical work completed on placements during the course of training. The reports include a cognitive-behavioural and psychodynamic formulation of a 66 year old lady with symptoms of anxiety and depression, a service evaluation examining the quality and effectiveness of a Home Treatment Team for older adults, a case study of a 26 year old man with symptoms of social anxiety – formulation and intervention based on a cognitive behavioural model of social anxiety, a single case experimental design of a 59 year old man with learning disabilities displaying challenging behaviour – utilising a cognitive analytic therapy model and finally a case study of a 13 year old girl experiencing recurrent abdominal pain – formulation and intervention based on a cognitive behavioural model of social anxiety.

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All personal details have been altered throughout each clinical practice report to ensure confidentiality is maintained.

LITERATURE REVIEW

Psychosocial outcomes of parents caring for an adult child with intellectual disability - a systematic review

Abstract

Objective: The aim of this review was to identify the psychosocial outcomes of parents caring for an adult child with intellectual disability. A secondary aim was to identify mediating variables impacting on outcomes for this ageing caregiving group.

Method: A systematic literature search was conducted using Medline, Psychinfo and Embase. Studies included used quantitative methodology and presented empirical findings on the psychosocial outcomes of family carers' of adult children with intellectual disability. Each study was rated against an established quality assessment tool.

Results: Twenty three studies met the inclusion criteria. Outcomes measured across the studies included: depression, overall health, mental health, physical health, stress, burden, quality of life, psychological wellbeing, negative affect, family problems and life satisfaction. Overall, findings indicated poorer psychological wellbeing and overall health when compared to parents of children without disabilities. There were a number of mediating variables identified: support, behavioural problems, employment, health, age, residence of adult child and additional caring responsibility.

Conclusions: Overall, the literature indicates that parents of adults with intellectual disability have poorer psychosocial outcomes when compared to non-caregiving populations. However, a range of mediating variables make it difficult to identify clear pathways. Further longitudinal research is needed to establish predictors of poor psychosocial outcomes in parents of adults with intellectual disability.

Introduction

Changes to the social care system over the last thirty years have seen individuals with intellectual disabilities move from living in segregated settings to the community (Ellison, White & Chapman, 2011). Many individuals with intellectual disabilities remain within the family home, often cared for by their parents (Cuskelly, 2006). Within families which have a child with a disability, traditional care roles are often taken on, with mothers usually fulfilling the primary caregiving role (Barnett & Boyce, 1995; Essex & Hong, 2005). Advances in medical care have also increased the life expectancy of individuals with intellectual disabilities, which means parents of individuals with intellectual disabilities can remain in this caregiving role well into old age and often until they are no longer able to care for their child due to their own age related ill-health or the death of the parent (Cuskelly, 2006).

The role of caring for a child with intellectual disabilities can place considerable demand on parents. Typically, children will develop increasing independence as they mature until as adults they no longer require the same level of input from their parents, but for individuals with intellectual disabilities their parents often remain in a perpetual caregiving role (Kropf & Kelly, 1995). Whether the child remains at home or in a community placement, parents often still provide considerable emotional and practical input to meet their child's needs (McDermott, Valentine, Anderson, Gallup & Thompson, 1997). The nature of this caregiving role means parents are often limited in terms of employment opportunities, with many parents never working full time (Parish, Seltzer, Greenberg & Floyd, 2004). This can have financial implications for families and can also lead to social isolation as social opportunities are restricted (Einam & Cuskelly, 2002). The

caring role can also be physically demanding as parents may need to provide daily physical assistance as well as managing potential behavioural problems.

Feelings of increased responsibility and pressure for this group of parents can contribute towards high levels of stress (Hassall, Rose & McDonald, 2005). It is common for parents in a caring role to neglect their own health/personal needs and, in combination with high stress, this group of parents is at high risk of physical/mental distress (Seltzer, Greenberg, Floyd, Pettee, & Hong, 2001). A number of studies suggest that parents caring for a child with intellectual disability often show considerable resiliency and adapt well to the caregiving role (Carr, 2008; Krauss & Seltzer, 1993). Nevertheless, there is evidence which suggests parents have poorer psychosocial outcomes than parents with a child without disabilities (Cuskelly, 2006; Murphy, Christina, Caplin, & Young, 2007). Risdal and Singer (2004) for example, report a higher rate of marital problems amongst parents with a child with a disability than amongst parents of children without a disability.

There are contrasting hypotheses about the long term impact of caregiving. The adaptation model suggests that, as parents age, they adjust to the role and adapt and build on their resources for coping with the demands associated with caregiving and consequently the negative impact decreases (Lazarus & Folkman, 1984). From this model younger caregivers would be expected to report more burden than older caregivers (Hayden & Heller, 1997). Conversely, the cumulative stress model suggests that as parents' age and their own health declines, the impact and demands of prolonged caregiving become more difficult to cope with (Hoyert & Seltzer, 1992; Shearn & Todd, 1997).

There is variability in how parents respond to the role of caregiving for a child with intellectual disabilities with growing research interest in what may mediate the negative impact of this caregiving role (Minnes, Woodford & Passey, 2007; Saloviita, Italinna & Leinonen, 2003). Research to date has found that how able the individual is, the severity and frequency of behavioural problems, social support, coping style and health of the mother all play important roles in mediating the impact of this role (Essex, Seltzer & Krauss, 1999; Hayden & Goldman, 1996; Hong, Seltzer & Krauss, 2001; Kim, Greenberg, Seltzer & Krauss, 2003; Lecavalier, Leone & Wiltz, 2006; Pruchno & Meeks, 2004).

Whilst many studies have focussed on the wellbeing of parents caring for children or young adults with intellectual disabilities (Baker et al., 2003; Floyd & Gallagher, 1997; Weiss, 2002), there is far less research which examines the impact of caring for an adult child with intellectual disabilities over a prolonged period. This is clearly important given the changing nature of care and support noted above. The aim of this review is identify the psychosocial outcomes of parents caring for an adult child with intellectual disability. A secondary aim is to identify mediating variables that might impact on parental outcomes for this ageing caregiving group.

Method

Search strategy

Three electronic databases were used to carry out the literature search: Medline (1946 – July 2014), PsychINFO (1967 – week 5 2014) and EMBASE (1974 – July 2014). Four blocks of search terms were used, the terms in each block were combined using "OR" and then the final results of each block was combined using "AND". Block 1: exp parents; family; caregivers. Block 2: intellectual development disorder; exp developmental disabilities or specific language impairment; learning disorders or learning disabilities; "intellectual* disabilit*" or "intellectual* impair*". Block 3: adult offspring; daughters or sons; "adult child*" or "old* child*". Block 4: The term 'psychosocial outcome' is an umbrella term incorporating a broad range of outcomes: wellbeing; "quality of life"; mental health; health; coping behaviour; "resilience (psychological)"; exp major depression; exp stress; caregiver burden; exp emotional states; psychosocial factors; exp employment status; exp life experiences; coping or impact* or mood*. The titles and abstracts of these studies were examined and unsuitable studies excluded. Full texts were obtained of the remaining articles and examined against inclusion/exclusion criteria (see below). A backwards search was completed on the remaining articles by hand searching the reference list of each article for further relevant studies. Finally, the last step involved performing a forward search on the identified studies. Figure 1 shows an overview of the search strategy/exclusion process.

Inclusion/exclusion criteria

Inclusion criteria:

(a) Family carer

(b) Family carers aged 45 and over

(c) Adult child with intellectual disability

(d) Long term care at home

(e) Study assesses psychosocial outcome of parent

(f) Peer reviewed

Exclusion criteria:

(a) Not published in English

(b) Book chapters

(c) Review article

(d) Conference or dissertation abstract

(e) Qualitative studies

(f) Date: pre 2000

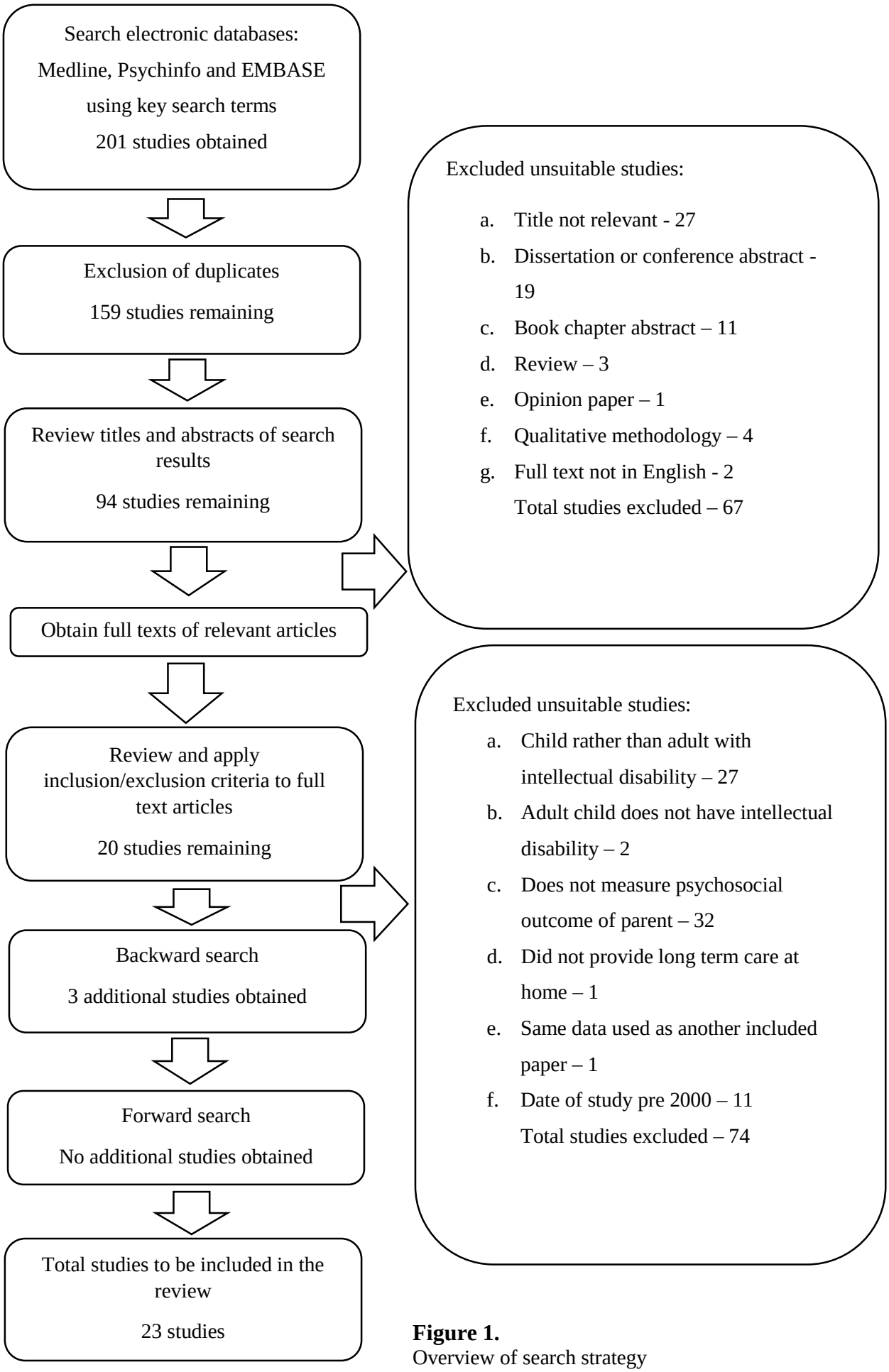

Figure 1.
Overview of search strategy

Methodological quality assessment

Each article was reviewed and assessed against an established quality assessment checklist for quantitative research, a copy of the checklist can be found in Appendix 1 (Kmet, Lee & Cook, 2004). This process was completed independently before the quality assessment results were reviewed and cross validated in research supervision. The checklist was adapted to fit the type of studies being reviewed, so only 11 out of the 14 items were used. Each study was scored on how well it met each item: 2 –fully met criteria; 1 – partially met criteria; 0 –did not meet criteria. A total sum was the aggregate of all scores. The total possible sum was 28 (total score of all items on checklist) minus 6 (3 n/a items). A summary score for each study was calculated. This was the total sum divided by the total possible sum, multiplied by 100 to give an overall percentage of quality. Studies were categorised into high quality (score of 17 and above), moderate quality (score of 11-16) or low quality (score of 10 or less). Appendix 2 provides a summary of the quality assessment.

Results

A total of 201 studies were identified. Duplicates were excluded leaving 159 studies. Exclusion/inclusion criteria were applied which reduced studies to 20. A further three studies were included following the backwards search; the forward search did not identify any further studies. A total of 23 studies were suitable to be included in the review and all 23 were assessed to be high quality quantitative studies (see Table 1).

Table 1 provides a summary of significant findings from the studies. Seventeen studies were cross-sectional design. For the six longitudinal studies, the follow-up period ranged from three to ten years, and the majority had two data collection points with one exception which had five data points. The majority of the studies were conducted in the United States of America (14), followed by United Kingdom (3), Israel (2), Taiwan (2), Australia (1) and Canada (1). Parental sample ages varied across studies. Using the sample means (as the majority of studies reported this) 11 studies examined the 45-64 age range, 16 the 65-84 age range and one the over-85 age range.

Table 1. Summary of studies

Authors (Country)	**Study aims**	**Study design**	**Sample - size, age**	**Control group**	**Psychosocial measures**	**Results/ Outcome**	**Quality rating**
Ben-Zur, H., Duvdevany, I., & Lury, L. (Israel)	Document the mental health and stress levels among mothers of children with intellectual disability (ID)	Cross-sectional Mothers whose adult child out of home placement vs mothers whose adult child living at home.	Mothers of adult child with ID - living in community placement n=50 Age 58.90 (9.76) Mothers of adult child with ID–living at home n=50 Age 62.44 (10.72)	None	Mental Health Inventory – short version. (MHI; Veit & Ware, 1983) Questionnaire on Resources and Stress (QRS-F; Friedrich et al. 1983)	**Mental health**: NS **Stress**: Community 26.08(9.74) Home 22.33 (8.76) ($t=2.02$; $p<0.05$)	High
Cairns, D., Brown, J., Tolson, D., & Darbyshire, C. (Scotland – UK)	Document the effects of prolonged caregiving on self-reported physical and mental health.	Cross-sectional	Parent of adults with ID, from 3 local authorities in Scotland. n=100 Group 1: Age 65-74 n=60 Group 2: Age 75-84 n=33 Group 3: Age 85+ n=7	UK norms 0 = worst possible health 100= best possible health 50 = average	Medical Outcome Study SF-36v2 (Ware et al. 2002) - generic measure of health status	**Physical Health** component summary (PCS): similar to UK norms. Group 1 55.31 (10.68) Group 2 50.26 (5.73) Group 3 41.25 (8.41) **Mental Health** component summary	High

Table 1. Summary of studies

Authors (Country)	Study aims	Study design	Sample - size, age	Control group	Psychosocial measures	Results/ Outcome	Quality rating
						(MCS): below UK norms. Group 1 32.03 (9.48) Group 2 37.26 (7.02) Group 3 37.99 (9.00)	
Carr, J. (UK)	Document wellbeing of mothers who have a 40 year old adult child with Down syndrome (DS).	Longitudinal Follow up when child 11 years, 21 years, 30 years 35 years and 40 years.	n=18 mothers of adults with DS Age 59-87 (75.9)	n=16 mothers with non-disabled 40 year old Age 60-80 (69.6)	Malaise Inventory (Rutter et al. 1970)	NS	High
Chen, S. C., Ryan-Henry, S., Heller, T., & Chen, E. H (USA)	Document the physical and mental health of mothers who have a child with ID	Cross-sectional	n=108 mothers of adults with ID Mid-life n=33 Age 55-64 (60.2/2.9) Later-life n=75 Age 65-90 (74.4/6.3)	National norms for each age group	Medical Outcome Study SF-36 - generic measure of health status	**Physical health** component summary (PCS): Mid-life: Study group Norm 49.7(10.4) 45.0(11.6) t(195)=2.13,p=0.04 **Mental health** component summary (MCS): NS	High

Table 1. Summary of studies

Authors (Country)	**Study aims**	**Study design**	**Sample - size, age**	**Control group**	**Psychosocial measures**	**Results/ Outcome**	**Quality rating**
Chou, Y., Pu, C., Kroger, T., & Fu, L (Taiwan)	Document differences in quality of life/ related factors between mothers with different employment status, who care for an adult child with ID	Cross-sectional	n=302 mothers of adults with ID Age 52.30(5.9) Employed full time (F/T) – 37.4% Mean age 50.80 Employed part time (P/T) – 16.2% Mean age 51.40 Not employed (N/E) – 46.4% Mean age 53.80	Taiwanese general population (statistics not stated)	World Health Organisation Quality of Life (Yao, Chung, Yu, & Wang, 2002) EQ-5D scale (The EuroQol group, 1990)	WHO **QOL** : F/T 95.00 P/T 94.00 N/E 89.80 F = 6.44, P=0.01 For all groups lower **QOL** than general population (statistics not stated) **Health**: F/T 5.80 P/T 6.10 N/E 6.30 F = 5.68, P=0.01	High
Chou, Y. C., Lee, Y. C., Lin, L. C., Kroger, T., & Chang, A. N (Taiwan)	Document difference in QOL, level of social support, perception of having a family member with an ID, service utilization, and future plans, between younger	Cross-sectional	Older caregivers n=315 Parents – 77.4% Sibling – 5.4% Other – 17.2% Age 55-86 (66.8/8.1)	None	WHOQOL-BREF – Taiwan version (Yao, Chung, Yu, & Wang, 2002)	Age 55+ Younger than 55 Overall **QOL***** 84.6(14.6) 90.5(13.2) Physical health*** 22.3(4.8) 24.8(4.0) Psychological***	High

Table 1. Summary of studies

Authors (Country)	Study aims	Study design	Sample - size, age	Control group	Psychosocial measures	Results/ Outcome	Quality rating
	and older family carers.		Younger caregivers n=472 Parents 61% Sibling – 16.9% Other – 22% Age 16-54 (43.3/8.9)			17.3(3.7) 18.7(3.2) Social Relationships*** 12.5(2.4) 13.1(2.0) Environment* 26.7(5.0) 27.5(4.7) *p=0.05 **p=0.01 ***p=0.001	
Einam, M., & Cuskelly, M (Australia)	Document factors that influence employment opportunities for families with a child, who is entering adulthood, with multiple and severe disabilities.	Cross-sectional	n=25 families with adult child with multiple disabilities (aged 17-21 years) 2 parent families n=22 Mother only families n=3 Responding mothers n=25 mean age 49.8	Families whose son or daughter has no known disabilities, same age range as study group.	Malaise Inventory (Rutter et al. 1970)	Malaise Inventory Questionnaire (**depression**) Mother, Father (SG) 5.9(3.5), 3.6(3.0) Mother, Father (CG) 3.0(2.3), 2.8(2.3) Study group significantly more psychological symptoms than control group (U=497.5, p<0.01)	High

Table 1. Summary of studies

Authors (Country)	Study aims	Study design	Sample - size, age	Control group	Psychosocial measures	Results/ Outcome	Quality rating
			Responding fathers n=13 mean age 51.1			Mothers in study group reported more symptoms than fathers in study group (U=91.0, p=0.06)	
Ghosh, S., Greenberg, J. S., & Seltzer, M. M (USA)	Document whether individuals with an adult child with DD or mental illness (MI), whose spouse develops an age related disability during the study period had poorer physical, social, psychological, and financial wellbeing than multiple comparison groups.	Longitudinal Surveyed 1992-1994 Follow up 2004-2006	Data was drawn from the Wisconsin Longitudinal Study (WLS) Parents in the WLS who had child with severe MI or DD were identified from the 2004-2006 survey n=107 parents child with a DD Age 64.01(4.63) n=120 parents child with severe MI Age 64.27(3.33)	In addition to caring for a spouse: n=1463 Drawn from the WLS – respondents who do not have a child with any disability Age 63.93(3.75)	Modified version of Ryff's Scale of Psychological Wellbeing (Ryff, 1989) The Center for Epidemiologic Studies Depression Scale (Radloff, 1977) Health Utilities Index – health related QOL (Feeny, Furlong, Boyle & Torrance, 2003)	Time point 2 results: ()=caring for spouse **Psychological wellbeing**: DD(YES) 4.62 DD(NO) 4.79 MI(YES) 4.69 MI(NO) 4.80 CG(YES) 4.76 CG(NO) 4.86 **Depression**: DD(YES) 8.03 DD(NO) 6.29 MI(YES) 8.24 MI(NO) 7.41 CG(YES) 6.75 CG(NO) 5.81 **Health** QOL: DD(YES) .81 DD(NO) .82 MI(YES) .77 MI(NO) .85	High

Table 1. Summary of studies

Authors (Country)	Study aims	Study design	Sample - size, age	Control group	Psychosocial measures	Results/ Outcome	Quality rating
						CG(YES) .84 CG(NO) .88	
Ha, J., Hong, J., Seltzer, M. M., & Greenberg, J. S (USA)	Document effects of having a disabled child on parents' physical and mental health, as compared to parents' of nondisabled children.	Cross-sectional	Sample/data was drawn from the MIDUS study (Study on Midlife in the United States; Brim et al, 2004). n=163 parents child with DD Age 53.79(12.30) N=133 parents child with MI Age 58.73(11.69)	n=1393 Parents with at least one living child – but no child with a disability or chronic mental health condition Age - 56.17(12.97)	Negative affect – sum of 6 items (Mroczek and Kolarz, 1998) Psychological wellbeing (Ryff, 1989) Somatic symptoms	Negative affect: DD: 10.20(4.32) MI: 10.03(4.08) CG: 8.83(3.29) **Psychological wellbeing**: DD: 224.38(38.0) MI: 226.08(38.49) CG: 231.50(34.77) **Somatic symptoms**: DD: 4.18(2.08) MI: 4.19(2.18) CG: 3.53(2.04) Controlling for sociodemographic characteristics, parents of a child with DD - significantly higher negative affect (β=.09, p<.001) marginally poorer psychological wellbeing (β=-.04, p<.10 and significantly more	High

Table 1. Summary of studies

Authors (Country)	**Study aims**	**Study design**	**Sample - size, age**	**Control group**	**Psychosocial measures**	**Results/ Outcome**	**Quality rating**
						somatic symptoms (β=.08, p<.01) than the control group.	
Hong, J., Seltzer, M., & Krauss, W (USA)	Document change in social support and its impact on psychological wellbeing in older mothers with a child with mental retardation.	Longitudinal Mothers first interviewed in 1988 and followed up 8 times at 18 month intervals. Psychological wellbeing measures introduced at the third follow up. Present study: data from the third and sixth interviews were analysed.	n=251 mothers with adult child with mental retardation, living at home Age (Time 3): Group 1 - Older middle age mothers (n=103) 58-65 (62.07) Group 2 Older mothers (n=148) 66-87 (72.10)	None	Ryff's Positive Psychological Functioning Scale: 2 subscales - the purpose in life and personal growth subscales (Ryff, 1989)	**Purpose in life**: Time 3 - Group 1 24.59(4.48) Group 2 23.61(4.69) Time 6 - Group 1 25.16(4.17) Group 2 23.87(4.75) **Personal growth** Time 3 - Group 1 25.96(3.43) Group 2 24.13(4.06) Time 6 - Group 1 25.68(3.51) Group 2 24.19(3.93)	High
Kim, H. W., Greenberg, J. S., Seltzer, M. M., & Krauss, M. W (USA)	Document whether processes of stability and change in maternal coping styles account for why caregiving	Longitudinal Sample drawn from two longitudinal studies. The study on mothers of	n=246 mothers of adults with ID Age 56-85 (66.6)	None	Zarit Burden Scale (Zarit et al. 1980) Center for Epidemiological Studies – Depression (Radloff, 1977)	Subjective **Burden**: ID (Time 1) 29.63(6.63) ID (Time 2) 28.43(6.92) MI(Time 1)37.09(10.25)	High

Table 1. Summary of studies

Authors (Country)	Study aims	Study design	Sample - size, age	Control group	Psychosocial measures	Results/ Outcome	Quality rating
	and other life challenges take a toll on the mental health of some mothers who are in the caregiving role, whereas others are able to maintain their psychological wellbeing over time.	adults with ID began in 1988, data collected at 18 month intervals until 2000. The study of mothers of adults with mental illness consisted of points of data collection between 1990-1995– separated by 36 months. For present study, data from second and fourth follow up, of mothers with an adult with ID, were used to match the timing and duration of the other study.	Mothers of adults with MI n=74 Age 55-78 (66.0)			MI (Time 2) 34.92(8.93) **Depression**: ID (Time 1) 9.55(7.78) ID (Time 2) 9.38(8.07) MI (Time 1) 11.22(7.94) MI (Time 2) 11.61(8.02)	

Table 1. Summary of studies

Authors (Country)	**Study aims**	**Study design**	**Sample - size, age**	**Control group**	**Psychosocial measures**	**Results/ Outcome**	**Quality rating**
Magana, S., Seltzer, M. M., Krauss, M. W., Rubert, M., & Szapocznik, J (USA)	Document how problems in the family might influence the stress process for two groups of Latinos caring for an adult child with mental retardation.	Cross-sectional	Two group of mothers all with adult children with mental retardation: Group 1: Puerto Rican mothers from Massachusetts n=44 Age 63.6 (8.5) Group 2: Cuban American mothers from Miami Dade County n=49 Age 63.7 (8.3)	None	Center for Epidemiologic Studies Depression Scale (CES-D) (Radloff, 1977) Zarit Burden Scale Questionnaire on resources and stress –F (Freidrich, Greeberg & Crnic, 1983) – used to measure family problems	**Depression** symptoms: NS Burden: Group 1 6.2 (4.9) Group 2 16.7 (11.6) (t (93) = 49, p = .000) Family problems: Group 1 66 (3.5) Group 2 101 (2.8) (t (93) = 6.2, p = .000)	High
Magana, S., Seltzer, M. M., & Krauss, M. W (USA)	Document factors that account for the differences in depressive symptoms between Puerto Rican mothers and Non-Latina white mothers of	Cross-sectional	Two groups of mothers all with adult children with mental retardation: Group 1: Puerto Rican mothers from	None	Center for Epidemiologic Studies Depression Scale(CES-D) (Radloff, 1977) Questionnaire on resources and	**Depression** symptoms: Group 1 17.9 (12.3) Group 2 9.8 (7.8) (t= 5.0, p< .001) **Family problems**: Group 1 6.5 (4.1) Group 2 4.4 (4.0)	High

Table 1. Summary of studies

Authors (Country)	Study aims	Study design	Sample - size, age	Control group	Psychosocial measures	Results/ Outcome	Quality rating
	adults with mental retardation		Massachusetts n=66 Age 57.4 (8.8) Group 2: Non-Latina white mothers from Massachusetts n=161 Age 68 (6.5)		stress –F (Freidrich, Greeberg & Crnic, 1983) – used to measure family problems	(t = -3.7, p<.001)	
Miltiades, H. B., & Pruchno, R (USA)	To document the association between race and religious coping on caregiving appraisals for mothers who live with adult child with mental retardation	Cross-sectional	Data drawn from a larger study involving interviews with 996 women who had an adult child with mental retardation. n=142 Age 50-84 (64.18, 7.42) Group 1 - n=71 black mothers	None	Subjective burden – measured by a 9 item scale developed by Lawton et al. (1989) Health – 4 item scale ranging from poor (1) to excellent (4)	**Burden** : NS **Health**: Group 1 8.70 (2.11) Group 2 9.66 (1.90) Being black associated with lower self-rated health (r= 0.23, p<.01)	High

Table 1. Summary of studies

Authors (Country)	Study aims	Study design	Sample - size, age	Control group	Psychosocial measures	Results/ Outcome	Quality rating
			Group 2 - n=71 white mothers				
Minnes, P. M., & Woodford, L. M (Canada)	Document the psychological wellbeing of older parents in relation to stressors, resources and perceptions of stress	Cross-sectional	Parents (primary caregivers) of adults with DD living in urban and rural locations in eastern and central Ontario n=80 (mothers n=71, Fathers n=9) Age 50-88 years (65.7)	Norms (stats not stated)	The Centre for Epidemiological Studies – Depression scale (Radloff, 1977) Family Stress and Coping Interview (Nachshen, Woodford & Minnes, 2003)	**Depression**: NS **Family stress** and coping: NS	High
Pruchno, R. A., & Meeks, S (USA)	Document the effects of caregiving stressors within the context of health-related events	Cross-sectional	Sample from a larger study of 1081 women and 301 men who had an adult child with a DD. n=932	None	Depressed Affect Subscale of the Center for Epidemiology Studies Index for Depression (Radloff, 1977)	**Depression** symptoms: Low Stress group: 1.69 (2.70) High Stress group: 3.40 (3.87) t = 7.83, p<.01	High

Table 1. Summary of studies

Authors (Country)	**Study aims**	**Study design**	**Sample - size, age**	**Control group**	**Psychosocial measures**	**Results/ Outcome**	**Quality rating**
			Age 50-91 (66.3/8.4) Low health stress: n=447 High health stress: n=485				
Rimmerman, A., & Muraver, M (Israel)	Document the impact on mothers of an adult child with ID living at home or out of home – on undesired life events, life satisfaction and wellbeing	Cross-sectional	Mothers of adults with moderate ID. Identified by the Ministry of Labor and Welfare, all adult children participating in sheltered workshop programmes. n=160 (53%) Adult child living at home - n=93 Age 68.48/6.25 Adult child living out of home - n=67 Age 68.74/5.48	None	Affect Balance Scale (Bradburn, 1969) - wellbeing Life Satisfaction Index (Neugarten, Havighurst and Tobin, 1961)	**Wellbeing** and life satisfaction: NS	High

Table 1. Summary of studies

Authors (Country)	**Study aims**	**Study design**	**Sample - size, age**	**Control group**	**Psychosocial measures**	**Results/ Outcome**	**Quality rating**
Seltzer, M. M., Floyd, F., Song, J., Greenberg, J., & Hong, J (USA)	Document long term outcomes of parents of adults with intellectual and developmental disabilities – regarding parental attainment, social participation, psychological functioning and health.	Longitudinal Comparisons made at two life stages – midlife and early old age.	Sample drawn from the Wisconsin Longitudinal Study. Parents who had a child with an intellectual or developmental disability (living at home) n=113 Parents who had a child with an intellectual or developmental disability (not living at home) n=107 Age	Parents who did not have a child with any form of disability n=1042	The Center for Epidemiologic Studies Depression Scale (Radloff, 1977) Modified version of Ryff's Scale of Psychological Wellbeing (Ryff, 1989) Health Utilities Index – health related QOL (Feeny, Furlong, Boyle & Torrance, 2003) only measured at second time point	By early years of old age, parents with co-residing children with disabilities had higher levels of **depression** than the comparison group (F = 4.66, .01) (d = .30) **Psychological wellbeing**: NS **Health**: In midlife parents of co-residing children with disabilities more likely to be overweight (F = 3.44, p .05) and have greater likelihood of cardiovascular impairments (F = 5.19, p .01). By early old age - evidence of more	High

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			Mid-life – early 50s, Early old age mid 60s			pervasive **health** impacts. Both groups, in early years of old age, had significantly poorer health QOL than the comparison group (F = 10.72, .001)	
Walden, S., Pistrang, N., & Joyce, T (UK)	Document the quality of life of parents of adults with ID and what factors impact on quality of life and experiences of caregiving	Cross-sectional	Parents of adults with ID using UK respite care services. n=62 (76%) (n=58 female, n= 4 male) Age 60 years/10.3	Data from published US and Irish studies.	Scales of Psychological Wellbeing (Ryff, 1989) Short Form of the Questionnaire on Resources and Stress (Friedrich et al, 1983) Symptom Checklist-90 Revised (Derogatis, 1994)	Current study, Previous Studies **Psychological wellbeing**: *Environmental mastery* 61.18 (9.60), 66.6 (11.6)** *Personal growth* 58.75 (9.05), 72.1 (7.5)*** *Purpose in life* 59.28 (9.46), 69.1 (9.5)*** Parenting **stress:** NS **Depression**	High

Table 1. Summary of studies

Authors (Country)	Study aims	Study design	Sample - size, age	Control group	Psychosocial measures	Results/ Outcome	Quality rating
						0.69 (0.64), 0.46 (0.52)** **p<0.01 ***p<0.001 **Psychological wellbeing** is significantly lower than that reported in a previous study of US midlife mothers with adult children without a disability (Ryff et al. 1994) Parenting **stress** appears higher in current study compared to US samples of parents of adults with ID (Seltzer & Krauss, 1989), and also compared to parents in the Republic of Ireland (Seltzer et al. 1995), it is similar level to that reported in a sample of parents in Northern Ireland.	

Table 1. Summary of studies

Authors (Country)	Study aims	Study design	Sample - size, age	Control group	Psychosocial measures	Results/ Outcome	Quality rating
						Parents of adults with ID reported higher levels of **depression** compared to normative US sample of adult women (Derogatis, 1994).	
Yamaki, K., Hsieh, K., & Heller, T (USA)	Document the health status of middle and older age female family caregivers of adults with intellectual and developmental disabilities.	Cross-sectional	Female family caregivers of adults with intellectual and developmental disabilities, in Illinois. n=206 Middle age n=130 Age 40-59 years (52.7/4.5) Older age n=76 Age 60 years+ (68.9/7.2) Majority were mothers	Population data from 2005-2006 Illinois Behavioural Risk Factor Surveillance System – this is an annual state level random telephone survey on the health and health risk behaviours conducted by the Illinois Department of Public Health.	Health Related Quality of Life (Moriarty, Kobau, Zack, & Zahran, 2005) Objective measure of health – Asked if they had ever been told by a health professional that they had: arthritis, asthma, diabetes, angina or coronary heart disease, heart attack, stroke, high blood pressure, high blood cholesterol, and/or osteoporosis.	Family caregivers of an adult child with ID generally tended to rate their **overall health** more favourably than the general population Older caregivers reported significantly more days in which their **physical health** was not good than the middle age caregiver group: 4.31 days, 2.18 days ($t = 22.11$, $p<.05$) Older caregivers reported twice as many **mentally**	High

Table 1. Summary of studies

Authors (Country)	Study aims	Study design	Sample - size, age	Control group	Psychosocial measures	Results/ Outcome	Quality rating
			(86.3%), sisters (10.8%) and a few aunts, nieces or grandmothers.		BMI calculated from respondents height and weight	**unhealthy** days when compared to same age group in general population (4.65 days, 2.3 days, 95% CI = 1.90-2.74) Compared with women in general population, caregivers reported significantly higher prevalence of arthritis, high blood pressure, obesity and activity limitations across the two age groups.	

Psychosocial outcomes

In terms of measuring outcome, there was variation across the studies. There were 11 psychosocial outcomes measured in total across the 23 studies. A table summarising how many studies measured each outcome can be found in Appendix 3. Outcomes included: depression, overall health, mental health, physical health, stress, burden, quality of life, psychological wellbeing, negative affect, family problems and life satisfaction. For the purpose of this review only the key findings from the outcome measures examined most frequently will be reported.

Psychosocial outcome: Depression

Ten studies examined depression as an outcome. Seven of these studies used The Center for Epidemiological Studies Depression Scale (Radloff, 1977). Two studies used the Malaise Inventory (Rutter, Tizard, & Whitmore, 1970) and one the Symptom Checklist-90 revised (Derogatis, 1994). Six of the studies found significant findings. One study found differences, but had not analysed for significance. Three of the ten studies did not find any significant findings.

Two studies found higher levels of depression in parents of adults with intellectual disability when compared with parents of adults without disability (Einam & Cuskelly, 2002; Seltzer, Floyd, Song, Greenberg & Hong, 2011), and one study when comparing parents of adults with intellectual disability to the general population (Walden, Pistrang & Joyce, 2000). Four studies examined mediating factors within the population of parents of adults with intellectual disability and found greater levels of depression in parents who were older, had a spouse develop a disability, had higher health stress and, in Puerto Rican mothers, when compared to non-Latina white mothers (Ghosh, Greenberg & Seltzer, 2012;

Magana, Seltzer et al., 2011; Seltzer & Krauss, 2004; Pruchno & Meeks, 2004). One study examined differences between caring groups, and found parents of adults with mental illness had higher level of depression than parents of adults with intellectual disability (Kim, Greenberg, Seltzer & Krauss, 2003)

One study compared mothers of adults with Down syndrome to mothers of adults without disabilities, and did not find any significant difference between the two groups (Carr, 2008). Magana, Seltzer, Krauss, Rubert & Szapocznik (2002) found no difference in depressive symptoms between Puerto Rican mothers of an adult child with intellectual disability and Cuban American mothers of an adult child with intellectual disability. Finally, one study found that older parents of adults with developmental disabilities had depression scores in the normal range when compared to norms of the general population (Minnes & Woodford, 2004)

All the papers examining depression were high quality. However, there were a number of limitations identified across the studies that may impact on generalisability of findings. These included: small sample size, issues with recruitment of sample (e.g. not documenting how recruited and use of convenience sampling), lack of ethnic diversity within sample, sample referred to as parents but mainly consisted of mothers, and differences in level of disability of the adult child (Einam & Cuskelly 2002; Ghosh et al., 2012; Kim et al., 2003; Magana et al., 2004; Pruchno & Meeks, 2004; Seltzer et al., 2011; Walden et al., 2000).

Summary of Depression:

Although the studies examining depression were all of high quality it was difficult to draw firm conclusions. There were contrasting findings for whether parents of adults

with intellectual disabilities have poorer outcomes than parents of adults without disabilities. At midlife one study reported that parents of adults with intellectual disabilities have higher rates of depression than parents of adult children without disability (Einam & Cuskelly, 2002), whereas another study by Seltzer et al. (2011) found no significant differences for this age group when compared to parents with an adult child without disabilities. For the older age group Seltzer et al. (2011) report higher levels of depression in parents of adults with intellectual disabilities than the comparison group of parents of adults without disabilities, a finding supported by a study by Walden (2000). However, Minnes and Woodford (2004) report scores in the normal range for older age parents when compared to norms of the general population. Comparing to other caring groups, one study found, using the same measure, that parents of adults with mental illness have higher levels of depression than parents of adults with intellectual disabilities (Kim et al., 2003).

Psychosocial outcome: Psychological wellbeing

Psychological wellbeing incorporates a number of factors including autonomy, balance of negative/positive affect, life satisfaction and purpose in life. Six studies examined psychological wellbeing as an outcome. Five of these studies used Ryff's Scales of Psychological Wellbeing (Ryff, 1989) and one study used the Affect Balance Scale (Bradburn, 1969). Three studies found significantly poorer psychological wellbeing in the sample group, one study found differences but did not analyse for significance and two of the studies did not find any significant differences.

Two studies found poorer psychological wellbeing in parents of adults with intellectual disability when compared to parents of adults without disability (Ha, Hong, Seltzer & Greenberg, 2008; Walden et al., 2000). Two studies examined mediating factors within parents of adults with intellectual disability and found poorer psychological wellbeing in parents who had a spouse develop a disability and older parents of adults with intellectual disability had poorer psychological wellbeing than younger parents of adults with intellectual disability (Ghosh et al., 2012; Hong, Seltzer & Krauss, 2001).

One study reported no significant difference in mothers' psychological wellbeing based on whether the adult with intellectual disability child lived in home or out of home (Rimmerman & Muraver, 2001). Seltzer et al. (2011) found no significant difference, at midlife or early old age, in reported psychological wellbeing between parents of adults with intellectual and developmental disabilities and parents of adults without disabilities – regardless of whether adult child with disability lived in home or out of home.

All the papers examining psychological wellbeing were high quality. However, there were a number of limitations identified across the studies that may impact on generalisability of findings. These limitations included: a lack of ethnic diversity within sample, use of convenience sampling, and sample referred to as parents but mainly consisted of mother (Ghosh et al., 2012; Hong et al., 2001; Walden et al., 2000).

Summary of Psychological Wellbeing:

These results provide initial evidence that parents of adults with intellectual disabilities may have poorer psychological wellbeing than parents of adults without disabilities (Ha et al., 2008; Walden et al., 2000). There was evidence indicating that wellbeing decreases from midlife to old age for this caring group (Hong et al., 2001).

However, one study provided contrasting evidence, with no significant findings between the study group and a control group (Seltzer et al., 2011).

Psychosocial outcome: Health

Overall health

Generic measures of health incorporated both physical and mental health as components to establish an overall impact on the individual. Five studies measured overall health as an outcome. Two studies used the Health Utilities Index (Feeny, Furlong, Boyle & Torrance, 1999), one study used the EQ-5D (The EuroQol group, 1990), one study used Health Related Quality of Life (Moriarty, Kobau, Zack, & Zahran, 2005) and one study measured health on a four point scale – poor (1) to excellent (4). All five studies found significant findings.

Two studies reported that parents of adults with intellectual disability have poorer overall health than parents of adults without disability (Ghosh et al., 2012; Seltzer et al., 2011). However, another study found that family caregivers of adults with intellectual disability tended to rate their overall health more favourably than the general population, but no significant difference was found in overall health between older age and middle age family caregivers. (Yamaki, Hsieh & Heller, 2009). Three studies examined mediating factors within parents of adults with intellectual disability and found poorer health in parents whose spouse developed a disability, unemployed working age mothers and black mothers when compared to white mothers (Chou, Pu, Kroger & Fu, 2010; Ghosh et al., 2012; Miltiades & Pruchno, 2002).

Physical Health

Four studies measured physical health. Two studies measured physical health using the Medical Outcome Study SF-36v2 (Ware et al., 2002). Two studies measured physical health through self-reporting of the number of symptoms experienced from a list of somatic symptoms. Two studies did not report any significant findings in physical health of parents of adults with intellectual disabilities and two studies did report significant findings.

One study reported that middle-aged parents of adults with intellectual disability describe experiencing significantly more somatic symptoms than parents of adults without disability (Ha et al., 2008). However, another study found middle-aged mothers who care for an adult child with intellectual disability had significantly better physical health than the general population when compared to norms, but by old age their physical health was similar to norms (Chen, Ryan-Heller, Heller & Chen, 2001). Further studies found no significant difference in physical health when comparing parents of adults with intellectual disability to norms and to parents of adults without disability (Cairns, Brown, Tolson & Darbyshire, 2014; Ghosh et al., 2012).

All the papers examining health were judged to be of high quality. However, there were a number of limitations identified across the studies that may impact on generalisability of findings. These included: a lack of ethnic diversity within sample, use of convenience sampling, low response rate, confounding variables not described in sufficient detail and small sample size (Chen et al., 2001; Chou et al., 2010; Ghosh et al., 2012; Seltzer et al., 2011; Yamaki, Hsieh & Heller, 2009).

Summary of Health

The evidence is that parents of adults with intellectual disabilities have poorer health than parents of adults without disabilities (Ghosh et al., 2012; Seltzer et al., 2011). There were also some factors that were predictors of poor health in this caregiving group – unemployment, caring for a spouse and being black (Chou et al., 2010; Ghosh et al., 2012; Miltiades & Pruchno, 2001). When considering physical health specifically, the main findings suggest that physical health is similar to that of the general population for parents with adults with intellectual disabilities (Cairns et al., 2014; Ghosh et al., 2012). One study suggested that at midlife parents of adults with intellectual disabilities have better physical health than the general population (Chen et al., 2001).

Mediating variables

A range of factors was associated with better psychosocial outcomes for parents of adults with intellectual disabilities. These included: support, behavioural problems, employment, health, age, residence of adult child and additional caring responsibilities. (see Table 2).

Table 2. Mediating variables

Authors (Country)	Study design	Mediating variables
Ben-Zur, H., Duvdevany, I., & Lury, L. (Israel)	Cross-sectional	Mental health positively correlated: social support r=0.6; p<0.0001, hardiness r=0.72; p<0.0001 Social support and hardiness inter-correlated r=0.75, p<0.0001 Stress negatively correlated with mental health, social support and hardiness (-0.54, -0.54 and -0.47; p<0.0001)
Carr, J. (UK)	Longitudinal	Mothers age significantly associated with malaise – older mothers have higher mean malaise score (p=<.05) Those with fewer friends – higher mean malaise (p=< .05)
Chen, S. C., Ryan-Henry, S., Heller, T., & Chen, E. H (USA)	Cross-sectional	Mid-life: Physical Component Summary (PCS) mediated by: Employment F(2.30)=3.37, p=0.047*, Family income F(2.24)=3.87, p=0.035*, Arthritis F(1.31)=10.56, p=0.003** Mental Component Summary (MCS) not mediated by any characteristics examined Later-life: PCS mediated by: Employment F(2.72)=4.23, p=0.018*, Arthritis F(1.72)=12.76, p=0.001** MCS mediated by: Family income F(2.55)=3.21, p=0.048* *p<0.05**p<0.01
Chou, Y., Pu, C., Kroger, T., & Fu, L (Taiwan)	Cross-sectional	For employed mothers QOL associated with: Substitute person to care for the adult with ID (β = -0.16, p<0.05), Family income (β = 0.28, P<0.001), Social support (β = 0.23, P<0.01) Non-employed mothers QOL associated with: Absence of substitute person to care for the adult with ID (β = -0.30, P<0.01), Family income (β = 0.18, P<0.05), Social support (β = 0.17, P<0.05), ADL (β = 0.36, P<0.01)
Chou, Y. C., Lee, Y. C., Lin, L. C., Kroger, T., & Chang, A. N (Taiwan)	Cross-sectional	Differences were found between the two groups in relation to: Age 55+ Younger than 55 Overall family support*** 17.1(6.6) 19.9(6.9) Formal support* 5.9(1.8) 6.2(1.7) Informal support** 8.0(1.8) 8.5(2.3) *p<0.05 **p<0.01 ***p<0.001

Table 2. Mediating variables

Authors (Country)	Study design	Mediating variables
Einam, M., & Cuskelly, M (Australia)	Cross-sectional	Employment outside the home related to maternal mental health (r=0.46, p<0.01), increasing hours in the paid work force associated with reduced symptomatology.
Ghosh, S., Greenberg, J. S., & Seltzer, M. M (USA)	Longitudinal	Having a child with developmental disability and having a spouse develop an age related disability associated with: significantly poorer health related quality of life (β=-.07, p<.01), and a trend to report lower levels of psychological wellbeing (β-.04, p<.10) when compared to control group
Ha, J., Hong, J., Seltzer, M. M., & Greenberg, J. S (USA)	Cross-sectional	Significant age by condition interaction effects for negative affect (β=-.05, p<.05) and psychological wellbeing (β=.06, p<.05) for parents of children with developmental disability (DD). Although parents of children with DD have poorer negative affect and poorer wellbeing than the comparison group, these effects attenuate with age. Parents who were older when their child was diagnosed with a DD reported lower levels of negative affect and marginally better psychological wellbeing than those who were younger at their age of their child's diagnoses (β=-.26, p<.01; β=.15, p<.10). Those who had children with long duration of disability showed lower levels of negative affect and better psychological wellbeing than those with a shorter duration of disability, net of parental age at the onset of disability (β=-.39, p<.001; β=.37, p<.001). Having more than one child with a disability was associated with significantly higher levels of negative affect and marginally greater somatic symptoms (β=.16, p<.05; β=.16, p<.10). Being currently employed predicted significantly lower levels of negative affect and better psychological wellbeing (β=-.18, p<.05; β=.17, p<.05) Being currently married predicted significantly lower levels of negative affect and marginally better psychological wellbeing (β=-.21, p<.05; β=.17, p<.10)

Table 2. Mediating variables

Authors (Country)	Study design	Mediating variables
Hong, J., Seltzer, M., & Krauss, W (USA)	Longitudinal	Increase in network size significantly predicted purpose in life scores, only for mothers who had not obtained guardianship (β = .209, p<.05) For older middle age mothers without guardianship – increase in network size predicted increase in personal growth (β = .229, p< .05) Increase in emotional support had positive effects on personal growth for older middle age group (β = .323, p<.01) For older mothers receiving higher levels of emotional support at time 3 and increasing levels between time 3 and 6 – predicted significant increases in both purpose in life (β = .22, p<.01; β = .18, p<.05) and personal growth (β = .19, p<.05; β = .23, p<.01)
Kim, H. W., Greenberg, J. S., Seltzer, M. M., & Krauss, M. W (USA)	Longitudinal .	Mothers of adults with mental illness reported significantly more frequent use of emotion focused coping at both time points, than mothers of adults with intellectual disabilities (ID) (β = 13.58, p < 0.001) Predictors of subjective burden at time 2 (ID group): Subjective burden at time 1 (β = 0.57, p < 0.001) Co-residence (β= 0.13, p<0.01) Behavioural problems at time 1 (β = 0.13, p<0.5) Increase in behavioural problems (β = 0.20, p<0.001) Problem focused coping at time 1 (β = -0.11, p< 0.05) Increase in problem focused coping (β = -0.15, p<0.001) Emotion focused coping at time 1 (β = 0.28, p< 0.001) Increase in emotion focused coping (β = 0.31, p<0.001) Predictors of depressive symptoms at time 2 (ID group): Level of depressive symptoms at time 1 (β = 0.35, p< 0.001) Mothers age (β = 0.09, p<0.05) Increase in behavioural problems (β = 0.13, p< 0.05) Problem focused coping at time 1 (β = -0.26, p< 0.001) Increase in problem focused coping (β = 0.46, p<0.001) Emotion focused coping at time (β = 0.39, p< 0.001)

Table 2. Mediating variables

Authors (Country)	Study design	Mediating variables
		Increase in emotion focused coping (β = 0.55, p<0.001)
Magana, S., Seltzer, M. M., Krauss, M. W., Rubert, M., & Szapocznik, J (USA)	Cross-sectional	Predictors of depressive symptoms: Poor health of mother (β = .37***) Fewer years of education (β = .21*) Fewer years in US (β = .22*) Family problems (β = .28*) Predictors of burden: Poor health of mother (β = .25**) Being married (β = .24*) Ethnicity (β = .46***) Severity of behaviour problems (β = .22*) Family problems (β = .49***) *p<.05 **p<.01 ***p<.001
Magana, S., Seltzer, M. M., & Krauss, M. W (USA)	Cross-sectional	Maladaptive behaviours were related to family problems, r = .40, p < .001, and family problems were related to maternal depressive symptoms, r = .46, p < .001 Controlling for ethnicity having a child with behavioural problems was a significant predictor of maternal depression (β = .14*) When family problems variable was added (β = .45***), maladaptive behaviours were no longer a significant predictor of maternal depression, but family problems remained significant. Indicating family problem mediate the effect of the child's maladaptive behaviour on depression Significant interaction between ethnicity and maternal health status (β = .3***). For both groups mothers who were in good health had low rates of depression, but for poor health – Group 1 (Puerto Rican mothers) had extremely elevated levels of depression whereas Group 2 (non-Latina white mothers) were not substantially higher than for those in good health.

Table 2. Mediating variables

Authors (Country)	Study design	Mediating variables
Miltiades, H. B., & Pruchno, R (USA)	Cross-sectional	Being black was associated with lower self-rated health (r = 0.23, p<.01), higher relationship quality (r = 0.17, p<.05), higher levels of religious coping (r = 0.34, p< .01) Higher burden was associated with more maladaptive behaviours (r = -.38, p<.01), mothers' health (r = 0.24, p<.01), poorer relationship quality (r = 0.26, p<.01) and being white (r = 0.14, p<.10)
Miltiades, H. B., & Pruchno, R (USA)	Longitudinal	Burden at follow up was correlated significantly with burden at baseline (r=.66, p<.01), child behaviours (r= .27, p<.01), being on a waiting list at baseline (r= .13, p<.05), being on a waiting list at follow up (r= .32, p<.01), and co-residence (r= .23, p<.01) Only 2% of variance in burden at follow up explained by placement (F(7, 304), P<.01)
Minnes, P. M., & Woodford, L. M (Canada)	Cross-sectional	Stressors - Maladaptive behaviour (β = .221, p<.05) and caregiver adverse age changes (β = .274, p<.03) were positively correlated with depression. Service use for the person with a developmental disability (β = -.238, p<.03) was negatively correlated with depression
Piazza, V. E., Floyd, F. J., Mailick, M. R., & Greenberg, J. S (USA)	Cross-sectional	Predictors of depression: High levels of burden (β = .41, p<.001) Use of disengagement coping (β = .25, p<.001) Use of distraction coping (β = .22, p<.01) Burden significant interactions with following styles of coping: Secondary engagement (β =.15, p<.05) Disengagement (β =.19, p<.01) Distraction (β = .19, p<.01)
Pruchno, R. A (USA)	Cross-sectional	Caregiving satisfaction associated with affection from mother to child (β = .57, p<.01) and with affection from child to mother (β = .34, p<.01). Caregiving burden had significant negative relationships with mother's functional ability (β = .12, p< .05), affection from mother to child (β = .21, p<.01) and affection from child to mother (β = .12, p<.05)

Table 2. Mediating variables

Authors (Country)	Study design	Mediating variables
		Caregiving burden had a positive relationship with functional support from mother to child (β = .20, p<.01) but was not related to functional support from child to mother. Caregiving burden and caregiving satisfaction had a significant inverse relationship (β = .30, p<.01)
Pruchno, R. A., & Meeks, S (USA)	Cross-sectional	Correlation between positive affect and negative affect: Low stress β = -.38*, High stress β = -.52* Correlations between positive affect and depressive symptoms: Low stress β = -.19*, High stress β = -.23* Correlations between negative affect and depressive symptoms: Low stress β = .48*, High stress β = .53* *p.01
Rimmerman, A., & Muraver, M (Israel)	Cross-sectional	Among the 68 years and older age group mothers who had child living at home had significantly fewer undesired life events, as compared to out of home group of mothers (F= 6.81 (1), p 0.01) Among the 68 years and older group, those in one parent families whose adult child lived at home, had greater levels of life satisfaction than the comparable group of mothers in two parent families (F= 3.96 (1), p 0.05) Among the 68 years and older group those in two parent families whose adult child lived out of home, had higher level of wellbeing than the mothers in one parent families (F = 3.89 (1), p 0.05) Mother's social support (high) served as a moderator in relationship between mother's age and undesired life events (F = 10.17 (1), p 0.002)
Walden, S., Pistrang, N., & Joyce, T (UK)	Cross-sectional	Higher levels of emotional support associated with greater psychological wellbeing (F= 2.10, p<0.05) Higher levels of challenging behaviour are associated with higher levels of parenting stress (F = 3.10, p< 0.01), depression (F = 3.38, p< 0.01) and anxiety (F = 3.89, p<0.001)

Table 2. Mediating variables

Authors (Country)	Study design	Mediating variables
		Informal support and physical dependency related to positive affect. Higher levels of informal support are associated with higher levels of positive affect ($F= 2.07$, $p<0.05$), and the more able the offspring the higher the level of positive affect between them ($F = 2.45$, $p<0.05$)

Support

Seven studies report that informal support had a significant association with psychosocial outcomes and this was for both middle and old age groups (Ben-Zur et al., 2005; Carr, 2008; Chou et al., 2010; Chou et al., 2009; Hong et al., 2001; Rimmerman & Muraver, 2001; Walden et al., 2000). More informal support was associated with better psychosocial outcome. Only one study reported formal support as being associated with any of the psychosocial outcomes: use of services by the person with intellectual disabilities was negatively correlated with symptoms of depression (Minnes & Woodford, 2004). Furthermore, Chou et al. (2009) found that older aged parents of adults with intellectual disability received less support, both informal/formal support, than their middle age counterparts - which may explain why in their study the old age parents had poorer psychosocial outcomes.

Behavioural problems

Six studies report behavioural problems to be associated with the psychosocial outcomes of parents of adults with intellectual disabilities. High frequency and severity of behavioural problems were associated with burden, symptoms of depression and anxiety, family problems and high stress (Kim et al., 2003; Magana et al., 2002; Magana et al., 2004; Miltiades and Pruchno, 2002; Minnes & Woodford, 2004; Walden et al., 2000).

Employment

Four studies found employment to be associated with better psychosocial outcomes. Chen et al. (2001) found parents who were employed had better physical health than unemployed parents, both in mid and later life. The other three studies all found that

employment was associated with better quality of life and mental health (Chou et al., 2010; Einam & Cuskelly, 2002; Ha et al., 2008).

Health

Three studies found that poor health was a predictor of depression and burden in parents of adults with intellectual disabilities (Magana et al., 2002; Magana et al., 2004; Miltiades & Pruchno, 2002). Two studies found an association with ethnicity and health. A study by Magana et al. (2004) found that Puerto Rican mothers were more vulnerable emotionally to poor health status, with the association between poor health and depression more evident for this group that for Non-Latina white mothers. A study by Miltiades and Pruchno (2002) found that high levels of burden were associated with poor health, and being black was associated with lower self-rated health.

Age

Three studies found associations between age and psychosocial outcomes. Two studies found that as parents of adults with intellectual disabilities age they have poorer psychosocial outcomes (Carr, 2008; Kim et al., 2003). In contrast, Ha et al. (2008) found that although parents of adults with intellectual disabilities had poorer wellbeing when compared to parents without a child with disabilities, this impact attenuates with age. So as the parent ages the impact of having a child with intellectual disabilities decreases.

Caring for others

Two studies found that caring for others as well as the adult child with intellectual disabilities was associated with poorer psychosocial outcomes. Ghosh et al. (2012) found that the additional responsibility of caring for a spouse with a disability was associated with significantly poorer quality of life than parents who care only for an adult child. Ha et

al. (2008) report that caring for more than one child with a disability was associated with significantly higher levels of negative effect when compared to parents caring for one adult child with an intellectual disability.

Discussion

The aim of this review was to identify the psychosocial outcomes for parents caring for an adult child with intellectual disability. A secondary aim was to identify mediating variables that impact on parental outcomes for this ageing caregiving group. There were a range of psychosocial outcomes measured, but for the purpose of the review only the four outcomes measured most frequently have been discussed – depression, psychological wellbeing, effect on overall health and effect on physical health.

There was a mixed pattern of evidence for depression as an outcome which made it difficult to draw firm conclusions. Overall, the evidence suggested that parents of adults with intellectual disabilities have poorer psychological wellbeing and poorer overall health when compared to parents of the same age who have adult children without disabilities. However, there were some robust studies that suggested that this was not the case. Physical health did not appear to be adversely affected for parents of an adult child with intellectual disabilities. In line with the secondary aim, the results from the review identified a number of variables which mediate psychosocial outcomes.

What are the psychosocial outcomes of parents caring for an adult child with intellectual disabilities?

There were contrasting findings both at midlife and at old age when comparing depression in parents of adults with intellectual disabilities and parents of adults without disabilities (Einam & Cuskelly, 2002; Minnes and Woodford, 2004; Seltzer et al., 2011). A lack of ethnic diversity was found across the studies measuring depression. This is important as there is known variation amongst ethnic groups with regard to a number of

demographic factors, including: health, education and income (Sorensen & Pinquart, 2005). Magana et al. (2004) found that differences in poor maternal health between the ethnic groups accounted for the difference in depression rather than ethnicity directly. The wider literature on ethnic diversity in caregiving reports differences in outcomes across ethnic groups (Dilworth-Anderson, Williams & Gibson, 2002). There are number of explanations that may account for differences in outcomes across ethnic groups, including: demographic variation, perceived stressors, internal and external resources and types of coping utilised (Aranda & Knight, 1997; Pinquart & Sorensen, 2005).

One study provided evidence that parents of adults with intellectual disabilities had fewer symptoms of depression when compared to parents of adults with mental illness. The authors concluded that this may be due to the very different challenges these two caring roles may have. The challenges associated with intellectual disability tend to be more constant and predictable than those faced by parents of children with mental health difficulties (Kim et al., 2003). Based on the findings of this review, it is currently difficult to compare findings about depression in parents of adults with intellectual disability with other caring groups due to the contrasting findings about depression in parents of adults with intellectual disability.

The evidence for psychological wellbeing as an outcome indicated that parents of adults with intellectual disabilities have poorer psychological wellbeing than parents of adults without disabilities (Ha et al., 2008; Walden, Pistrang & Joyce, 2000). There was only one study of parents at midlife and at early old age that that did not find any significant findings when comparing psychological wellbeing of parents of adults with intellectual disabilities and parents of adults without disabilities (Seltzer et al., 2011). One study supported the cumulative stress model theory that wellbeing decreases from midlife

to old age for parents of adults with intellectual disabilities (Hong et al., 2001), a possible indicator that the increase in stress associated with this caring role eventually takes its toll. This is perhaps unsurprising considering many carers will also be contending with their own age-related issues at this stage of their lives (Cuskelly, 2006).

Two studies compared psychological wellbeing in parents with an adult child with intellectual disability whose child either lived at home or outside of home. No significant differences were reported between the two groups (Rimmerman & Muraver, 2001; Seltzer et al., 2011). This is in contrast to existing research into other caregiving groups, such as caring for an elderly family member, which found residing with a care recipient had a more negative impact on carers' wellbeing, than not co-residing, even when carers reported caregiving as a positive experience (Berg-Weger, Tebb, Rubio & Berg-Weger, 2000). This may indicate the resilience of parents of adults with intellectual disability who continue to care and have their child co-residing, but do not differ in wellbeing to parents whose adult child with intellectual disability does not live at home. Or it may be highlighting that, for some parents, psychological wellbeing remains the same even when people are no longer providing the daily care. Previous studies have described how carers have an ongoing worry about their adult child's needs being met, and that they often remain actively involved in their child's care even following transition to residential care (Seltzer, Greenberg, Krauss & Hong, 1997). So, for some parents, although they are not physically meeting their adult child's needs, they remain a 'perpetual parent' in terms of their feeling of responsibility.

The findings for effects on health as an outcome indicate that parents of adults with intellectual disabilities have poorer health than parents of adults without disabilities (Ghosh et al., 2012; Seltzer et al., 2011). However, there was evidence to suggest that

physical health is similar to that of the general population for parents with adults with intellectual disabilities (Cairns et al., 2014; Ghosh et al., 2012). One study suggested that at midlife parents of adults with intellectual disabilities have better health than the general population (Chen et al., 2001). A meta-analysis that examined the differences in psychological and physical health in caregivers and non-caregivers reported that, although physical health was poorer in caregivers, the difference was relatively small in comparison to the psychological impact (Pinquart & Sorensen, 2003).

What is unique about parents of adults with intellectual disabilities is the prolonged nature of the caregiving role. It is well established in caregiving literature that caregivers have poorer psychosocial outcomes than non-caregivers (Pinquart & Sorensen, 2003). However, from the results of the review it was difficult to separate whether the findings of poor psychosocial outcomes were a result of specifically caring for an adult child with intellectual disability or a result of a prolonged caregiving role and would be seen in any long term caregiver.

Are there any mediating factors for outcomes?

The review highlighted a number of variables that were associated with psychosocial outcomes for parents of adults with intellectual disabilities, with many supporting previous research findings. Obtaining a clear understanding about what mediates the impact of the caring role informs professionals and families about what support may be required to help maintain the physical and psychological health of this group of ageing parent caregivers.

There was strong evidence that the following factors mediated outcomes: informal support, behavioural problems, employment, health and caring for others (Ben-Zur et al., 2005; Carr, 2008; Chen et al., 2001; Chou et al., 2009; Chou et al., 2010; Einam & Cuskelly, 2002; Ghosh et al., 2012; Ha et al., 2008; Hong et al., 2001; Kim et al., 2003; Magana et al., 2002; Magana et al., 2004; Miltiades and Pruchno, 2002; Minnes & Woodford, 2004; Rimmerman & Muraver, 2001; Walden et al., 2000) . There was evidence that age impacts on the psychosocial outcomes of parents of adults with intellectual disabilities. Two studies found the older parents get the poorer the psychosocial outcomes (Carr, 2008; Kim et al., 2003). However, another study found that the impact of caring for an adult child with intellectual disabilities attenuates with age (Ha et al., 2008).

The findings from the review did not indicate the type of relationship – i.e. whether or not being the mother or a father of an adult child with intellectual disability mediates the psychosocial outcome. The wider caregiving literature suggests that relationship does mediate the outcome for other caregiving groups, such as caring for elderly family members with dementia (Berg-Weger et al., 2000). It is known that mothers often take the main caring responsibilities (Essex & Hong, 2005) and, therefore, it could be hypothesized that in ageing parents the mothers of adults with intellectual disabilities are likely to have poorer psychosocial outcomes. The literature is reflective of the demographics and the majority of the research is focused on the outcomes of mothers. More research is needed on both parents to establish if relationship to the care recipient is a significant mediating factor in psychosocial outcomes for mothers and fathers of adults with intellectual disabilities.

Quality of evidence

All the studies included in the review were assessed using an established quality assessment tool and found to be of high quality standard. However, there were some limitations that need to be considered. Across a number of the studies there were factors that may restrict the generalisability of findings. This included small sample sizes (which may increase the risk of inaccurate findings) and convenience sampling. Lack of ethnic diversity may also be an issue as the samples may not be representative of the wider population.

Limitations of the literature and directions for future research

Although the main findings indicate that, overall, parents of adults with intellectual disabilities have poorer psychosocial outcomes than parents of adults without disabilities, there is currently not enough evidence measuring the same outcome to be able to draw strong conclusions. There was a lack of consistency across studies that made comparison between the studies more difficult. A large range of outcomes were measured and for each outcome a number of assessment measures were used and this, consequently, made it difficult to categorize the psychosocial outcomes.

Most of the studies were cross-sectional in design which limits directional inferences that findings were a result of the caregiving role. For future research more longitudinal design studies would allow the relationship between prolonged caregiving and psychosocial outcomes to be examined across time. Longitudinal studies would also be helpful in establishing the predictors of negative psychosocial outcome; this would help to enable the identification of vulnerable groups of parents. There was a lack of research examining the oldest age category, with only one study using parents over 85. Therefore,

further studies are needed to examine the older age parents to establish the impact of prolonged caregiving on a potentially vulnerable group of carers.

The majority of the studies were conducted in the USA. Therefore, it is difficult to know whether the main findings can be generalised to other countries where there are different health and social care systems. More research is needed in European countries to be able to compare findings. In addition, more up-to-date research is needed as less than half of the studies included in the review were published in the last 10 years, and only two of those used data obtained in the last five years.

Implications for future families/professionals

This review demonstrates that parents of adults with intellectual disabilities may be at risk of poorer psychosocial outcomes as a result of this prolonged caregiving role. What is evident is that this is a growing group of family carers as there are many adults with intellectual disabilities remaining in the family home. Therefore, services need to consider how best to support both the adult with intellectual disabilities and the family carer to ensure that they are able to continue caregiving in the best possible health for as long as the carer feels able to.

There was a noticeable absence of financial circumstances and formal support in the mediators of outcomes for these parents. The lack of formal support may indicate a lack of services for these parents or barriers to these parents accessing the services. As people enter old age, their social and emotional support network is likely to reduce, which was evident in this review, and this may make the availability of appropriate formal support even more vital. There has been a recent significant development for carers in the UK under the Care Act (Department of Health, 2014) that promotes the wellbeing of the

carer as well as the care recipient. Carers are now legally entitled to an assessment for their own needs by the local authority and may be eligible for support, including financial, social and employment support. This may have implications for clinical practice in the NHS, in terms of volumes of referrals, if carers are deemed to have mental health needs. For professionals working with adults with intellectual disabilities and their carers, it is essential to inform families about available service provision and their right to a carer's assessment under the new legislation.

References

Aranda, M. P., & Knight, B. G. (1997). The influence of ethnicity and culture on the caregiver stress and coping process: A sociocultural review and analysis. *The Gerontologist*, 37: 342–354.

Baker, B. L., McIntyre, L. L., Blacher, J., Crnic, K., Edelbrock, C., & Low, C. (2003). Pre-school children with and without developmental delay: Behavior problems and parenting stress over time. *Journal of Intellectual Disability Research, 4–5*, 217–230.

Barnett, W. S., & Boyce, G. C. (1995). Effects of children with down syndrome on parents' activities. *American Journal on Mental Retardation, 100,* 115–127.

BenZur, H., Duvdevany, I., & Lury, L. (2005). Associations of social support and hardiness with mental health among mothers of adult children with intellectual disability. *Journal of Intellectual Disability Research, 49* (1), 54-62.

Berg-Weger, M., Rubio, D. M., & Tebb, S. S. (2000). Living with and caring for older family members. Issues related to caregiver well-being *Journal of Gerontological Social Work*, 33 (2), 47-62.

Bradburn, N. M. (1969). *The structure of psychological well-being.* Chicago: Aldine.

Cairns, D., Brown, J., Tolson, D., & Darbyshire, C. (2014). Caring for a child with learning disabilities over a prolonged period of time: An exploratory survey on the experiences and health of older parent carers living in scotland. *Journal of Applied Research in Intellectual Disabilities, 27* (5), 471-480.

Carr, J. (2008). Families of 40-year olds with down syndrome. *Journal on Developmental Disabilities, 14 (2),* 35-43.

Chen, S. C., Ryan-Henry, S., Heller, T., & Chen, E. H. (2001). Health status of mothers of adults with intellectual disability. *Journal of Intellectual Disability Research, 45* (5), 439-449.

Chou, Y., Pu, C., Kroger, T., & Fu, L. (2010). Caring, employment, and quality of life: Comparison of employed and nonemployed mothers of adults with intellectual disability. *American Journal on Intellectual and Developmental Disabilities, 115* (5), 406-420.

Chou, Y. C., Lee, Y. C., Lin, L. C., Kroger, T., & Chang, A. N. (2009). Older and younger family caregivers of adults with intellectual disability: Factors associated with future plans. *Intellectual & Developmental Disabilities, 47* (4), 282-294.

Cuskelly, M. (2006). Parents of adults with an intellectual disability. *Family Matters, 74,* 20-25.

Department of Health. (2014). *The care act – the law for carers.* Available at: https://www.gov.uk/government/uploads/system/uploads/attachment_data/file/366089/Factsheet_8_-_Carers.pdf (Accessed: 28 April 2015).

Einam, M., & Cuskelly, M. (2002). Paid employment of mothers and fathers of an adult child with multiple disabilities. *Journal of Intellectual Disability Research, 46* (Pt 2), 158-167.

Ellison C., White, A., & Chapman, L. (2011) Avoiding institutional outcomes for older adults living with disability: the use of community-based aged care supports. Journal of *Intellectual and Developmental Disability 36*, 175–183.

Essex, E. L., & Hong, J. (2005). Older caregiving parents: division of household labor, marital satisfaction and caregiver burden. *Family Relation, 54,* 448-460.

Essex, E. L., Seltzer, M. M., & Krauss, M. W. (1999). Differences in coping effectiveness and well-being among aging mothers and fathers of adults with mental retardation. *American Journal of Mental Retardation, 104*, 545–563.

Derogatis, L. R. (1994). *Symptom Checklist 90–R: Administration, scoring, and procedures manual.* Minneapolis, MN: National Computer Systems.

Dilworth-Anderson, P., Williams, I. C., & Gibson, B. E. (2002). Issues of race, ethnicity, and culture in caregiving research: A 20-year review (1980-2000). *The Gerontologist*, 42: 237–272.

Feeny, D., Furlong, W., Boyle, M., & Torrance, G. W. (1999). Multi-attribute health status classification systems. Health utilities index. *PharmacoEconomics*, 7 (6), 490-502.

Floyd, F. J., & Gallagher, E. M. (1997). Parental stress, care demands, and use of support services for school-age children with disabilities and behavior problems. *Family Relations, 46*, 359–371.

Ghosh, S., Greenberg, J. S., & Seltzer, M. M. (2012). Adaptation to a spouse's disability by parents of adult children with mental illness or developmental disability. *Psychiatric Services, 63* (11), 1118-1124.

Ha, J., Hong, J., Seltzer, M. M., & Greenberg, J. S. (2008). Age and gender differences in the well-being of midlife and aging parents with children with mental health or developmental problems: Report of a national study. *Journal of Health and Social Behavior, 49* (3), 301-316.

Hassall, R., Rose, J., and McDonald, J. (2005). Parenting stress in mothers of children with an intellectual disability: the effects of parental cognitions in relation to child characteristics and family support. *Journal of Intellectual Disability Research, 49 (6),* 405–418.

Hayden, M., & Goldman, J. (1996). Families of adults with mental retardation: Stress levels and need for services. *Social Work, 41,* 657–667.

Hayden, M. F., & Heller, T. (1997). Support, problem-solving/coping ability, and personal burden of younger and older caregivers of adults with mental retardation. *Mental Retardation, 35*, 364–372.

Hong, J., Seltzer, M., & Krauss, W. (2001). Change in social support and psychological well-being: A longitudinal study of aging mothers of adults with mental retardation. *Family Relations: An Interdisciplinary Journal of Applied Family Studies, 50* (2), 154-163.

Hoyert, D. L., & Seltzer, M. (1992). Factors related to the well-being and life activities of family caregivers. *Family Relations, 41*, 74–81.

Kim, H. W., Greenberg, J. S., Seltzer, M. M., & Krauss, M. W. (2003). The role of coping in maintaining the psychological well-being of mothers of adults with intellectual disability and mental illness. *Journal of Intellectual Disability Research, 47* (Pt 4-5), 313-327.

Kmet, L. M., Lee, R. C., & Cook, L. S. (2004). Standard quality assessment criteria for evaluating primary research papers from a variety of fields. *The Alberta Heritage Foundation for Medical Research, Health Technology Assessment Initiative, 13,* 1-21.

Kropf, N. P., Kelly, T. B. (1995). Stigmatized and perpetual parents: older parents caring for adult children with life-long disabilities. *Gerontological Social Work, 24 (1/2),* 3-16.

Krauss, M.W., & Seltzer, M. M. (1993) Current well-being and future plans of older caregiving mothers. Irish Journal of Psychology 14, 48–63.

Lazarus, R. S., & Folkman, S. (1984). *Stress, appraisal, and coping*. New York: Springer.

Lecavalier L., Leone, S., & Wiltz, J. (2006). The impact of behaviour problems on care-giver stress in young people with autism spectrum disorders. *Journal of Intellectual Disability Research* 50, 172–83.

Magana, S., Seltzer, M. M., Krauss, M. W., Rubert, M., & Szapocznik, J. (2002). Well-being and family role strains among cuban american and puerto rican mothers of adults with mental retardation. *Journal of Human Behavior in the Social Environment, 5* (3-4), 31-55.

Magana, S., Seltzer, M. M., & Krauss, M. W. (2004). Cultural context of caregiving: Differences in depression between puerto rican and non-latina white mothers of adults with mental retardation. *Mental Retardation, 42* (1), 1-11.

McDermott, S., Valentine, D., Anderson, D., Gallup, D., & Thompson, S. (1997). Parents of adults with mental retardation living in-home and out-of –home: caregiving burdens and gratifications. *American Journal of Orthopsychiatry, 67 (2),* 323-329.

Miltiades, H. B., & Pruchno, R. (2002). The effect of religious coping on caregiving appraisals of mothers of adults with developmental disabilities. *The Gerontologist, 42* (1), 82-91.

Miltiades, H. B., & Pruchno, R. (2001). Mothers of adults with developmental disability: Change over time. *American Journal on Mental Retardation, 106* (6), 548-561.

Minnes, P., & Woodford, L. (2004). Wellbeing in ageing parents caring for an adult with a developmental disability. *Journal on Developmental Disabilities, 11 (1),* 47-65.

Minnes, P., Woodford, L., & Passey, J. (2007). Mediators of Well-being in ageing family carers of adults with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 20* (6), 539-552.

Moriarty, D. G., Kobau, R., Zack. M. M., Zahran, H. S. (2005). Tracking Healthy Days—a window on the health of older adults. *Preventing Chronic Disease*, 2 (3), 1-8.

Murphy N. A., Christina B., Caplin D. A. & Young P. C. (2007). The health of caregivers for children with disabilities: caregiver perspectives. *Child Care Health and Development 33,* 180–187.

Parish, S. L., Seltzer, M. M., Greenberg, J. S., & Floyd, F. (2004). Economic implications of caregiving at midlife: Comparing parents with and without children who have developmental disabilities. *Mental Retardation, 42*, 413– 426.

Piazza, V. E., Floyd, F. J., Mailick, M. R., & Greenberg, J. S. (2014). Coping and psychological health of aging parents of adult children with developmental disabilities. *American Journal on Intellectual and Developmental Disabilities, 119* (2), 186-198.

Pinquart, M., & Sorensen, S. (2003). Differences between caregivers and noncaregivers in psychological health and physical health: A meta-analysis. *Psychology and Aging*, 18, 250–267.

Pinquart, M., & Sorensen, S. (2005). Ethnic differences in stressors, resources, and psychological outcomes of family caregiving: A meta-analysis. *The Gerontologist*, 45 (1), 90–106.

Pruchno, R. A. (2003). Enmeshed lives: Adult children with developmental disabilities and their aging mothers. *Psychology & Aging, 18* (4), 851-857.

Pruchno, R. A., & Meeks, S. (2004). Health-related stress, affect, and depressive symptoms experienced by caregiving mothers of adults with a developmental disability. *Psychology & Aging, 19* (3), 394-401.

Radloff, L. S. (1977). The CES-D scale a self-report depression scale for research in the general population. *Applied Psychological Measurement*, 1 (3), 385-401.

Rimmerman, A., & Muraver, M. (2001). Undesired life events, life satisfaction and well-being of ageing mothers of adult offspring with intellectual disability living at home or out-of-home. *Journal of Intellectual and Developmental Disability, 26* (3), 195-204.

Risdal, D., & Singer, G. H. S. (2004). Marital adjustment in parents of children with disabilities: A historical review and metaanalysis. *Research and Practice for Persons with Severe Disabilities, 29*, 95–103.

Rutter, M., Tizard, J., & Whitmore, K. (1970). Education, health and behaviour. London: Longmans.

Ryff, C. D. (1989). Happiness is everything, or is it? Explorations on the meaning of psychological wellbeing. *Journal of Personality and Social Psychology*, 57, 1069-1081.

Saloviita, T., Italinna, M., & Leinonen, E. (2003). Explaining the parental stress of fathers and mothers caring for a child with intellectual disability: a double ABCX model. *Journal of Intellectual Disability Research, 47*, 300–312.

Seltzer, M., Greenberg, J. S., Floyd, F. J., Pettee, Y., & Hong, J. (2001). Life course impacts of parenting a child with a disability. *American Journal on Mental Retardation, 106* (3), 265-286.

Seltzer, M. M., Greenberg, J. S., Krauss, M. W., & Hong, J. (1997). Predictors and outcomes of the end of co-resident caregiving in aging families of adults with mental retardation or mental illness. *Family Relations*, 46, 13-22.

Seltzer, M. M., Floyd, F., Song, J., Greenberg, J., & Hong, J. (2011). Midlife and aging parents of adults with intellectual and developmental disabilities: Impacts of lifelong parenting. *American Journal on Intellectual and Developmental Disabilities, 116* (6), 479-499.

Shearn, J., & Todd, S. (1997). Parental work: an account of the day-to-day activities of parents of adults with learning disabilities. *Journal of Intellectual Disability Research* 41, 285–301.

Sorensen, S., & Pinquart, M. (2005). Racial and ethnic differences in the relationship of caregiving stressors, resources, and sociodemographic variables to caregiver depression and perceived physical health. *Aging & Mental Health*, 9 (5), 482-495.

The EuroQol Group. (1990). EuroQol a new facility for the measurement of health related quality of life. *Health Policy,*16 (3),199-208.

Walden, S., Pistrang, N., & Joyce, T. (2000). Parents of adults with intellectual disabilities: Quality of life and experiences of caring. *Journal of Applied Research in Intellectual Disabilities, 13* (2), 62-76.

Ware, J. E., Kosinski, M., Bjorner, J. B., Turner Bowker, D. M., & Maruish, M. E. (2007). *User's manual for the SF-36v2 health survey.* Quality Metric Incorporated.

Weiss, M. J. (2002) Hardiness and social support as predictors of stress in mothers of typical children, children with autism, and children with mental retardation. *Autism,* 6**,** 115–30.

Yamaki, K., Hsieh, K., & Heller, T. (2009). Health profile of aging family caregivers supporting adults with intellectual and developmental disabilities at home. *Intellectual and Developmental Disabilities, 47* (6), 425.

EMPIRICAL PAPER:

Caring for a child with Down syndrome across 50 years – a qualitative study

Abstract

Objective: The aim of the study was to explore the experience and challenges that resulted from caring for a child with Down syndrome across 50 years.

Method: 15 family members, from 12 families, of an adult with Down syndrome were recruited. Participants took part in a semi-structured interview. The interview schedule was designed so that participants could share both positive and negative experiences across the lifespan of their child/sibling and to reflect on challenges and resources they accessed for support. Data was analysed using a Framework Analysis Approach (Ritchie & Spencer, 1994).

Results: Four main themes were discussed – early experiences and feelings of having child with Down syndrome, factors that enabled carers to cope with raising a child/sibling with Down syndrome, experience of caring for an adult with Down syndrome and positive experiences of caring for a child/sibling with Down syndrome. Families experienced initial feelings of grief, sadness and anger on receiving the diagnosis. Overall, findings indicated that families experienced the way in which they received the diagnosis and transition of care as particularly challenging when looking back across their child's life. The families identified a number of personal and external factors that helped them cope: family support, personal characteristics, the temperament and level of disability of the child, having more children, economic circumstances, faith, and seeing it as a mutually benefitting relationship (as the child provided companionship). Despite challenges faced across a lifetime of caring, parents felt it had been a worthwhile and rewarding experience.

Conclusion: Overall, the study highlighted the resilience of parents who care for a child with Down syndrome across their lifespan in their ability to adjust to the challenges of this

role and draw on the positives of the experience. However, further research is recommended to continue to build on understanding of the impact of caring across time on different members of the family.

Introduction

Down syndrome is a genetic condition in which an extra chromosome 21 develops. This extra chromosome can impact on development and, therefore, those that are born with this condition have a degree of intellectual disability, the extent of which varies between individuals (Sheets et al., 2011). There is also a risk of associated health conditions including low muscle tone, heart conditions, thyroid problems, hearing and visual impairment. There is, in addition, a high prevalence of early ageing including the onset of dementia and there is typically a lower life expectancy than in the general population (Carr, 1995; Wilkinson, Kerr, Cunningham & Rae, 2004). However, due to advances in the medical field, longer life expectancies than ever before are being recorded, with many individuals with the condition living into middle and old age. The average life expectancy of a person born with Down syndrome is 55 years, but some individuals may live well into their 60s (Patti, Amble & Flory, 2010; Torr, Strydom, Patti, & Jokinen, 2010).

There has been substantial social change in the last 30 years around caring for individuals with intellectual disability (Brown & Brown, 2005; Ellison, White & Chapman, 2011). In the past, UK families would have commonly sent their child to an institution to be cared for – and this would typically have been segregated from main society (Cairns, Brown, Tolson, & Darbyshire, 2014). However, in the UK today there has been a shift towards inclusion and a number of families are raising their child with intellectual disability at home or within the local community, with tailored health and social support (Ellison et al., 2011). It is likely that raising a child with an intellectual disability will present challenges and responsibilities that are unique when compared to that of parenting a typical developing child. For instance, for many families with a child with Down syndrome it is unlikely that this child will go through the transition from

dependence to independence and instead will need ongoing long term support (Minnes & Woodford, 2004). Thus, the increase in life expectancy provides new challenges for families whose child remains within the family home as parents are now likely to be still "caring" well into old age.

There has been a lot of research into the impact of caring for a child with an intellectual disability, but this has mostly focused on parents of children and young adults. There is far less evidence about the impact of prolonged caregiving on older parents of adults with intellectual disability. Existing studies that have examined psychosocial outcomes of parents caring for a child with intellectual disability have found that work and social opportunities may be restricted for parents due to the responsibilities of meeting their child's care needs (Parish, Seltzer, Greenberg & Floyd, 2004). Further to this, an association has been found between unemployment and poorer psychological wellbeing (Chou, Pu, Kroger & Fu, 2010). This may result from the stress of having less income and the feeling of isolation due to lack of social contact (Caples & Sweeney, 2010). Caring for an adult with intellectual disability has been associated with poorer health which may have future implications for older parents caring for their adult with intellectual disability (Ghosh, Greenberg & Seltzer, 2012; Seltzer, Floyd, Song, Greenberg & Hong, 2011). There is evidence to indicate that parents of an adult with intellectual disability have poorer psychological wellbeing than parents of an adult without a disability (Ha, Hong, Seltzer & Greenberg, 2008; Walden, Pistrang & Joyce, 2000).

The experience of raising a child with Down syndrome across their whole life span is not well documented. It has been suggested that parents of a child with Down syndrome experience greater wellbeing and less stress than parents of a child with other types of intellectual disability and this is termed the 'Down syndrome advantage' in recent

literature (Blacher & McIntyre, 2006; Stoneman, 2007). It raises interesting questions about what factors may mediate this difference between parents and whether this advantage is still present when the caring is prolonged over time.

There have been a number of studies that have explored developmental differences between individuals with Down syndrome when compared to a matched control group without disability. Carr's 50 year longitudinal research study into a cohort of Down syndrome individuals has been particularly unique and invaluable in this area of research as no other study has followed a cohort for this length of time across their whole lifespan (Carr, 2012). The participants in Carr's study were born between December 1963 and November 1964. All babies born with Down syndrome in this time-frame in Surrey and one area of Southeast London were invited to take part in the study. 54 babies were recruited – 25 boys and 29 girls. The study has followed the cohort over their lifespan looking at their overall development, behaviour, living and life styles, the role of services and the effects on family (Carr, 1988, 1995, 2008, 2012). In examining the effects on family Carr (2008) measured the wellbeing of mothers, findings indicated that across time mothers of children with Down syndrome report higher scores on the malaise inventory when compared to mothers of children without disabilities, however, the difference between groups was not significant.

There appears to be limited research into the longitudinal experience of caring for a child with Down syndrome and the research that does exist is mainly focused on reporting challenges and negative experiences, or the absence of negative impact. This qualitative research study intends to bridge that gap in the literature. It will be an opportunity for the parents/siblings of Carr's cohort study to document their stories of raising a child with Down syndrome across 50 years. It is hoped the data sourced from these interviews will

provide rich information to help future families who have a child with Down syndrome and professionals involved with their care. The main aim of the study was to explore the experiences and challenges that resulted from this prolonged caregiving role.

Method

Design

The study used a qualitative methodology using Framework Analysis Approach (Ritchie and Spencer, 1994). A semi structured interview schedule was designed by the research team using an abbreviated life history approach (see Appendix 4). A life history approach explores personal experiences using time points in history as an anchor for narratives. This study used an abbreviated approach so questions were framed around certain time points to give an overview across the lifespan. The aim of this was to help guide the participants to share both positive and negative experiences across the lifespan of raising their child and to reflect on challenges faced and what resources they accessed for support. To ensure the interview was accessible to participants and to examine validity, pilot interviews were conducted with a mother and sibling of an adult with Down syndrome and verbal feedback about the schedule was sought. The interview schedule was reviewed in response to the comments and a further question about fears for the future was added to the schedule, which was felt important to help capture the sibling experience.

Participants

Participants were 15 family members of an adult with Down syndrome (8 parents, 7 siblings). This represented 12 different families as the sample included a couple, two siblings from the same family and a mother and daughter. One of the siblings, who had an older brother with Down syndrome, was also a mother of a child with Down syndrome. The mean age of parents was 77.63 years (range = 67- 86, SD = 6.02). The mean age of siblings was 56 (range = 48-65, SD = 5.45). The majority of the sample were White British

(14). Education level was as follows: no qualifications (3), GCSE level/equivalent (6), A-level/equivalent (3) and Degree level or higher (3). Over half of participants were retired (8). The adults with Down syndrome were 50 years of age or soon to be 50. Two adults with Down syndrome had died, one aged 37 and one aged 49. Three of the adults with Down syndrome still remained living in the family home. Table 1 presents the demographics for each participant.

Table 1. Participant Demographics

Participant code	Age	Ethnicity	Education	Occupational group	Work Status	Relationship	Age of child/sibling with DS	Living with family
01	78	White British	GCSE/or equivalent	III NM	Retired	Father	50	X
02	58	White English/Italian	A-level/or equivalent	IV	Employed part-time	Sister	49	✓
03	65	White British	Degree level or higher	I	Retired	Sister	49	X
04	86	White British	GCSE/or equivalent	V	Retired	Mother	50	✓ •
05	78	White British	GCSE/or equivalent	II	Retired	Mother	50	X
06	74	White British	A-level/or equivalent	III M	Retired	Foster-Father	50	X
07	85	White British	No academic qualifications	V	Homemaker	Mother	50	X
08	76	White British	GCSE/or equivalent	III M	Retired	Father	Died aged 49	
09	77	White British	No academic qualifications	V	Homemaker	Mother	Died aged 49	
10	56	White British	GCSE/or equivalent	IV	Unemployed - ill health	Brother	50	✓ •
11	51	White British	GCSE/or equivalent	IV	Employed full-time	Sister	49	X
12	58	White British	Degree level or higher	I	Retired	Sister	50	X
13	67	White British	No academic qualifications	V	Retired	Mother	Died aged 37	
14	48	White British	A-level/or equivalent	III NM	Homemaker	Sister Mother	50 12	X ✓ •
15	56	White British	Degree level or higher	I	Homemaker	Sister	50	X

Procedure

Ethical approval for this study was obtained from the Science, Technology, Engineering and Mathematics Review Committee at the University of Birmingham. Confirmation can be seen in Appendix 5.

Participants were recruited from an existing 50 year longitudinal study that has examined a cohort of individuals with Down syndrome from when they were infants across their life span (Carr, 1995). All remaining parents and siblings from Carr's cohort study were contacted by letter and provided with an information sheet that outlined the aims of the study and what to expect if they consented to taking part (see Appendix 6). Participants were requested to return written consent if they were interested in being part of the study (see Appendix 7). 23 families were invited to take part and 16 consented and met criteria to be included in the study. One participant later dropped out due to the death of her child during the data collection period. Seven participants were excluded from the study for the following reasons: did not reply (2), ill health (1) and memory problem/dementia (4). In total 15 participants, from 12 families, took part in the study.

All interviews were conducted by the first author between January 2014 and December 2014. Participants were visited at their home address and interviews conducted face to face, with the exception of two participants who opted for telephone interviews. Interviews ranged in time from 50 minutes to 123 minutes. All interviews were audio recorded for which consent was sought. The data from the interviews was transcribed verbatim and any identifying information was removed or modified.

Analysis

Using a Framework Analysis Approach the data from each interview was analysed manually and coded for themes (Ritchie & Spencer, 1994). Themes were then classified and an analytic framework was developed in Excel. This systematic method of analysis organises large volumes of qualitative data and the framework allows data to be examined both across cases and theme by theme. Following consultation with a research supervisor who is experienced in qualitative research methodology, initial themes emerging from the data were discussed to enable the development of a framework to apply to the rest of the transcripts.

Results

Nine main themes emerged from the data about the longitudinal experience of caring for a child with Down syndrome – early experiences and feelings of having child with Down syndrome, factors that enabled carers to cope with raising a child/sibling with Down syndrome, factors relating to the family, experience of services, factors relating to the health of an individual with Down syndrome, experience of caring for an adult with Down syndrome, negative experiences of caring for a child/sibling with Down syndrome, positive experiences of caring for a child/sibling with Down syndrome and factors about the individual with Down syndrome (see Figure 1). The following four themes will be discussed in more detail – early experiences and feelings of having a child with Down syndrome, factors that enabled carers to cope, experience of caring for an adult with Down syndrome and positive experiences of caring for a child/sibling with Down syndrome. These four themes captured the prominent feelings within the family at the start of the journey when finding out about the diagnoses, the factors that have enabled the families to cope with the inevitable challenges of caring across 50 years, the difficulties facing families of adults with Down syndrome in making decisions about how long to continue care at home and who will take over the caring role, and, finally, the positive experiences that have resulted from caring for a family member with Down syndrome.

Figure 1. Main Themes/categories

Early experiences and feelings of having a child with Down syndrome
Pregnancy - hopes
First impressions of baby
Knowledge of Down syndrome
How/when told about diagnoses
Feelings/reactions to diagnoses
Professional information/advice given
Family/friends reactions

Experience of caring for an adult with Down syndrome
Decision/feelings - home or community placement
Transition of care/changing roles (Feelings)
Formal support - transition of care
Worries about future
Experience of being a perpetual parent

Positive experiences of caring for a child/sibling with Down syndrome
Highlights across the lifespan
Positive impact of caring for child/sibling – meaning making of the experience

Factors that enabled carers to cope with raising a child/sibling with Down syndrome
Appearance of baby
Support – informal/ formal
Personal characteristics
Family/personal values
Level of disability
Temperament of child/sibling
Having more children – a child without Down syndrome

Experience of services
Service provision
Challenges with services

Factors relating to the health of an individual with Down syndrome
Health problems
Healthcare support

Family factors
Integration/inclusion - family
Wider family involvement/support
Bond with parents

Factors related to the individual with Down syndrome
Hobbies/Interests/Enjoyment
Achievement/progress/strengths
Independence
Relationships

Negative experiences of caring for a child/sibling with Down syndrome
Negative impact on carer/family
Others' reactions (outside family/friends)
Impact of death of parent(s) on individual with Down syndrome
Traumatic events
Challenges
Failures/regrets
Loss of 'normal' child that they did not have

Early experiences and feelings of having child with Down syndrome

Diagnoses/Reactions - How and when told:

Parents were informed following the birth of their child. Most of the parents were told within the first few days. However, two families were not informed until their child was several months old. One set of family members were only informed when they asked their GP if there was something wrong with the baby. The parents had become concerned following a weigh-in clinic when their baby was 5 month old. The mother had felt health care professionals were talking about the baby, but she did not know what they were discussing and nothing was said to the family at the clinic. Another parent described that their GP had made the decision not to tell them as he was initially unsure whether the child did have Down syndrome. Then, later, after getting a paediatrician involved who confirmed the child did have Down syndrome, both health care professionals agreed to delay informing the parents as the family did not have any idea so they did not want to break the bad news. The GP decided to wait until the parents started to ask questions about the child's delayed development. However, the parents did not ask questions because, although slightly delayed compared to other children the same age, their child did meet the typical developmental milestones. So when the child turned one the GP made the decision to tell the family. After informing the family of the diagnosis, the GP proposed that the child may have to go to into an institution suggesting that the parents would not be able to cope with the child. *'Well you know dear he might have to go away' (Participant 05, parent).* This mother spoke of being grateful for the delay in discovering the diagnosis as it meant the parents had bonded with the child and, therefore, there was no doubt around their decision to keep the child.

It was a common theme across the participants that they had little or no understanding of what the term 'Down syndrome' meant (or 'mongolism' as it was called 50 years ago) upon receiving their child's diagnosis. Parents were given very little information or advice about what living with this diagnosis would mean for the child and family. Parents spoke of professionals clearly having a very negative outlook about their child's future and were given no indication of hope or positivity. Parents also spoke about their shock of being told that they did not have to take their child home, that they could be left at the hospital to be placed in an institution.

It wasn't until I was taking him home, my husband and I, you don't have to take this child home if you don't want to, Mother, because he is a Mongol (Participant 04, parent).

They didn't give you any information. Not at all, and I mean, that really followed the whole of our early period with X. There was nowhere where you could get any advice or information on what to do, how he would react, what you could do to help the reaction. There was nothing at all (Participant 08, parent).

They just said he would sit in the corner, he won't do anything, and he'll just become a cabbage (Participant 09, parent).

Reactions to receiving diagnoses:

There was similarity across the parents in the type of emotions they experienced when told the diagnosis. Parents spoke of a sense of unfairness and questioned why it had happened to them. There was shock/disbelief, a sense of loss for the 'normal baby' they

had been expecting to have, general distress and sadness. Feelings of bereavement were common in discovering the diagnosis and an overwhelming loss of expectations and hopes for their child and his/her future. This was far removed from what the parents had imagined for their child.

One of the fathers interviewed spoke about being given the diagnosis before his wife. The doctor took him to one side at the hospital and informed him of the diagnosis and was told his baby was likely to have significant intellectual disability. *'He will be slow to develop, and when he did develop, he wouldn't do anything but sit in the corner. That's all they said' (Participant 08, parent).* The father described how he felt sick and was then left to share the news with his wife, which he described as being a difficult and emotional task.

I think it was a bit of a grieving thing going on. I cried a lot and I actually remember not wanting to look at him. I was in denial really and I thought, "If I look at him, I'm going to have to accept the fact that he is" so I didn't look at him for ages (Participant 14, parent).

Oh awful, like being bereaved…Terrifying. I had no idea. I looked at this enchanting little child and just couldn't believe there was anything really, truly, wrong with him (Participant 05, parent).

I was sitting there thinking that if the Devil came and said, "Sign this and I'll make him right." I would have signed (Participant 08, parent).

However, despite the initial emotions of distress, the parents also described a strong feeling of protectiveness which was evoked at the suggestion of not keeping their baby. Parents described feelings of shock and horror that health care professionals would even suggest not keeping their baby. It seems these reactions enabled parents to see that, despite the diagnosis, this was still their baby, and there was a clear parenting instinct to protect and care for their child.

There was no chance of that happening. I immediately packed up all my things, picked up my child and left the hospital. That I just wanted to get him home. I had this absolutely overwhelming protective feeling towards X; it was more than protective. I just wanted to wrap him up not in cotton wool but just keep him safe. Keep him safe from what he'd got, I suppose, but then being ignorant I didn't even understand that (Participant 13, parent).

What? Shock, you know? I said, Well, to be quite truthful, he didn't ask to come into the world and if later on in life I can't cope, that's the time for me to make that decision (Participant 04, parent).

Sibling reactions varied and seemed to depend on when they were told about the diagnosis. Siblings that were involved or witness to family conversations about the diagnosis from the start appeared to have more acceptance than siblings who discovered the situation at a later date. Three of the siblings said that their first realisation of their brother or sister having Down syndrome came when he or she attended a different school, when others asked them direct questions or when children were cruel and called their

brother or sister names. They did not describe noticing the physical symptoms of Down syndrome prior to this, they just simply saw their sibling as their brother or sister.

I didn't have any anger at all, none whatsoever; incredibly accepting. I don't really think that changed either. Very accepting, and perhaps very innocent I don't know (Participant 12, sibling).

Only when people started asking me questions. Prior to that, I didn't think there was anything wrong. I think really, I realised once he went to school. He was six when he went to school (Participant 02, sibling).

They took me and my sister to visit her school that she was going to go to. Saw all these people that weren't right and thought, "Why are they taking my sister there?" Didn't fit in. You know, as far as I was concerned I didn't really see her as anything different. I was very angry, yes, very angry. I was angry at the fact that she was going to be subjected to what I thought were odd people, rather than being in a normal school. She wasn't going to be treated the same as me (Participant 11, sibling).

There was some variation reported in how partners, the other parent, responded to receiving the diagnosis. It was clear the majority of partners responded with love, support and acceptance, although there were initial feelings of sadness there was a sense of acceptance almost immediately. One mother described how her husband, the baby's father, responded. *He was just as upset as I was, but he was brilliant. I cannot say how brilliant he was. He was just terrific. Just took enormous care of me and of X (Participant 05, parent).* However, there was a minority of mothers who spoke of their husbands not being

so supportive. One father wanted to leave the baby at the hospital and did not support the decision to bring him home. Another mother spoke of her distress that her husband rejected the baby and said it could not possibly be his. This father subsequently left home when the child was just a few months old with the relationship ending in divorce. *My husband burst into tears and then went mad when they said he'd mongolism as they used to call it then. Then, when we got home, he accused me of – that X wasn't his son (Participant 13, parent).*

Parents spoke of their worry about sharing the diagnosis with wider family and wanting to process the information and deal with their own emotions before managing the family reactions. *We didn't say anything to anybody until we really were sure that the children, I had an elder son and two daughters, they had to accept it (Participant 04, parent).*

Family reactions were similar to that of the parents, and a range of emotions were experienced including sadness, disbelief and anger. *My poor mother was heartbroken because I'm an only, so she had no other grandchildren you see. She was heartbroken. My father said, "Oh no the man's a fool. He's making a mistake. There's nothing wrong with X." would not believe it (Participant 05, parent).*

However, all the families spoke of very quickly there being an acceptance and that they never felt their child was excluded in any way within the wider family. In fact, family became an important source of support for the parents, both in the early days of adjustment and across their child's life span. *I think certainly positive in terms of being very accepted, and being. I think perhaps there were some patronising kind of, "Oh, dear, poor thing",*

that sort of thing, from some, but I didn't really experience…but I don't think my mum and dad would have had it any other way (Participant 15, sibling).

Factors that enabled carers to cope with raising a child/sibling with Down syndrome

A range of factors was identified by the families which helped them cope with the challenges associated with caring for a child with Down syndrome. In the early days, parents spoke of distraction and keeping busy being useful. The fact that parents found their baby attractive was a factor that a number of parents said helped with acceptance. Accepting the child as part of the family and 'getting on' with family life were important parts of adjustment. Parents spoke of treating their child the same as any other child and that, looking back, family life for the most part continued as normal.

You can't sit there and wallow can you? Not forever. I do. That's when I thought, "Right come on, I've got to be a mum here." That was it, plus he was gorgeous. That did help, the fact he was so beautiful did help (Participant 14, parent).

A number of parents spoke of making the decision to have another child soon after the birth of their child with Down syndrome. Parents felt it was important to have a child without disability to provide company for their other children. *We were only going to have two children, but when we found there was a problem with X we thought, "Well, it's going to look bad for the elder boy that he's got this handicapped sister", so we decided to go for another one (Participant 01, parent).* The parents reflected that having more children also meant that they were kept busy which stopped them dwelling on any sadness:

Well you see not really because when you've got a lot of small children, and I think we might have had his brother by then as well, he would have been a baby, you're always either holding a sick bowl or opening a packet of sandwiches or saying, "No you can't." or "Stop it. Well you can't go to the loo now." that things wash over you to be honest. Things that would upset me now just washed over you. Tremendous help (Participant 05, parent).

Then of course I became pregnant…because there was only two years between them. But that was calculated. We said yes, we'll have another one, and it did, it helped (Participant 07, parent).

There was a common thread across the families' responses that the temperament and level of disability of the child made it easier to cope with the fact they had Down syndrome. Parents spoke about their child having a happy temperament, not being a burden, being easy to manage and feeling proud of their achievements. For some of the parents, once they had accepted their child had Down syndrome, there was a transition to feeling fortunate in comparison to parents of children with disabilities who were harder to manage or parents who had children who were more severely disabled.

No problems at all. As I say, we don't have any problems with him, which is why it's so easy to accept him for what he is, you see (Participant 04, parent).

I mean, you can't – when you look at what other people have to put up with, you know, we think we were lucky (Participant 09, parent).

Family support was identified as a key factor in coping across the families. Family support included both practical support with the daily care responsibilities but also emotional support – having someone to share both the positive and the negative experiences with. Parents spoke of family support playing an important role in the early days of adjustment but also making a significant contribution in terms of coping with the long term impact of the role of caring. *So I've got a lot to be thankful for over the years, for the family as well. So I've always appreciated that. But as I say, they've all been there for me, and I've always appreciated it, you know? (Participant 04, parent).*

There were contrasting views about the value of support groups. Some families really valued having others to talk to who had got shared experiences of caring for a child with intellectual disability. Parents described that being able to talk to others who had older children with intellectual disability helped as it was useful to be able to source information about what to expect/where to access support and services. It also gave rise to the realization that families can adjust and that having a child with an intellectual disability is not a complete disaster and it provided hope that they as a family would be able to cope. However, there were families that actively chose to avoid support groups and had strong views about being able to cope without such specific support.

"Well I just feel I'm the only one." She said, "No you're not. I'll put you in touch with a couple of mothers who live near you." which she did. That was the most helpful thing. Their kids weren't Down's. They had other things wrong with them but they were so

matter of fact about it all, and so calm. Not treating it as a tragedy because they were further down the line than me. That was the most helpful thing (Participant 05, parent).

People wanted to give me advice, people said, "There's this association, there's that association. There's this group, there's that group" and I didn't go to any of them. I didn't want to know, positively shunned me, it's not me. I just hate all that. No, I'm doing it on my own thanks and I don't want any support really from anyone (Participant 14, parent).

Personal characteristics were another shared factor that families felt enabled them to cope. Common characteristics described included the following: having a willingness to stand up to people - *It's any parent, most parents, really when push comes to shove. When it's your child, normal or handicapped, you get in there and fight. Absolutely (Participant 05, parent)*; determination; actively seeking out information and available sources of support; caring, a tendency to put others first - *I've always been a person who puts other people first rather than myself (Participant 06, parent)*; positive outlook - *It's tempting to assume that had he not had Down's syndrome, he would have been superb and a wonderful young man, but he might not have been. You think if he'd only been normal everything would have been fine, but it might not have been (Participant 05, parent);* an easy going temperament and a just 'get on with things' attitude.

There was a shared value amongst many of the families of respect and treating others as you would like to be treated yourself. A number of participants spoke of being grateful for how their parents had raised them, and that family values of caring for others and accepting responsibilities had helped them cope and accept their child/sibling:

I think perhaps it's in our – both, probably, both our family backgrounds that you take your own responsibilities on, and you just get on with it (Participant 08, parent).

Well, I think obviously your family, that right from day one my parents had that political – and it is a political mind-set, actually, that is a genuine care for humanity (Participant 15, sibling).

There was also a common theme among the parents, but not among the siblings, of religion/faith being an important factor in coping:

I am a practicing Christian. I am a believer. Really, frankly, I don't know how people manage who aren't. It's fundamental, yes. More so as I get older, I think that. You've got more time to think about things (Participant 05, parent).

I fervently believe in God so I fervently believe that I will see X again one day. It probably sounds utterly ridiculous but I couldn't go on if I thought I wasn't going to see him again one day (Participant 13, parent).

A small number of parents spoke of the area they lived in and their economic circumstances being factors that have helped with coping, as it meant they had not had the additional stress of living in an environment that was not suitable for a raising a family. *You see we've been fortunate. We've always lived in reasonably nice places. We haven't been in a top floor flat, surrounded by yobs being sick in the loo. We've been very fortunate (Participant 05, parent).*

One parent spoke of her son providing companionship since her husband had passed away and that this made coping with the caring role as a lone ageing parent much easier to deal with as she felt they both benefitted from living together. *So as I say, with my husband being gone, well it'll be 31 years in July, I've been able to spend quite a lot of time with him, and we do, you see (Participant 04, parent).*

In taking over the caring role from parents, one sibling spoke of the importance of making time for self-care, in the absence of any other support, to cope with the burden of taking on this responsibility. The sibling describes a negative experience of a battle to have their own life whilst also managing the responsibility of caring for their sibling, this is in contrast to the parent perspective. *I try to have an active life. I try to see friends regularly, keep in touch with people, go out in the evenings and have a dance you know, and erm it's just trying to do all these things, just to keep a balance. I know if I don't it is so easy just to slump, and erm you've got, you know I'm sort of fighting to have a life really (Participant 03, sibling).*

Experience of caring for an adult with Down syndrome

Transition of care - Decision-making process:

One challenge that families of a child with Down syndrome face is how long to continue to provide care within the family home. Some families made the decision that it would be in their child's best interest once they reached adulthood to move from the family home into a community placement prior to the family being forced to make the decision due to ill-health or the death of a parent. It was thought a sudden move due to the

death of a parent would be more traumatic as the individual with Down syndrome would be dealing with the death of a parent and a new environment at the same time. It took a long time for some of the families to find what they felt was an appropriate placement. The families that made this decision for their child described them moving out of the family home when they were in their 20s or 30s. This planned move was viewed as a positive transition by a number of families as it gave their child more independence and for some this was what their child was wanted/was requesting. However, this was a difficult decision. Parents spoke of feeling turmoil during this decision-making process and not knowing whether or not they were making the best decision.

It's the worst decision that I ever had to make. Thinking what would she do? Who would handle her? And all of that was going through my mind. How would she take to it? That's the way I've always regarded it, once they really are senior, that if they're at home it must be terrible for them if the parents both go, mustn't it?(Participant 07, parent).

Transition of care - Reactions and consequences:

The families that moved their child into care experienced a range of emotions. There were feelings of guilt and concern about whether this was the right decision for their child.

That was the worst part I ever experienced with X, in her whole existence. It really was the most awful experience. Yes, it was difficult. Oh, the days when I thought, "I'm going to go up there. I'm going to bring her back." Because I just couldn't see her, and couldn't

visualise what she was doing, and I just had to hope and pray that where she was, was working for her (Participant 07, parent).

I mean I felt a bit guilty when I first got her into the residential home, but then when I saw the way she had taken to it I wasn't bothered. She is enjoying it and she has got a life of her own now (Participant 01, parent).

There was a sense of relief that an appropriate placement had been found, particularly as families spent a lot of time locating somewhere they felt confident would meet their child's needs. There was also a feeling of happiness when it was clear their child had settled and was enjoying his or her new life.

I was glad because we'd always thought of her as moving on from us. We'd always envisaged that. In fact, we had tried, on several occasions, to find somewhere that she could go to…nowhere really worked because what X needs (Participant 6, parent).

I mean, they supported X in moving out because that's what she really wanted to do. They could see that she did (Participant 12, sibling).

However, there was also a sense of sadness for a change in the relationship they had with their child. One parent discussed how her child lived in a residential home for adults with learning disability which met her child's need well. However, there was a sense of sadness that the impact of old age meant she could no longer have her child home for regular visits as she was no longer able to meet her care needs. This meant frequency

and length of contact was significantly reduced following the move into care and the parent felt unable to maintain the quality of the relationship without the regular contact.

I would have her home perhaps twice a month, collect her on the Friday and take her back on the Sunday evening. But since I've had the back problem and arthritis, X needs help with all her sanitary things, and washing and bathing. It's not-I can't do. So that's how we are the moment, whereas I would have had her home more regularly. So that's our relationship at the moment. Not at close as I would have liked (Participant 07, parent).

When considering a transition of care, some parents had strong views that they did not want their other children to take on the caring role. Although they had worries about finding an appropriate community placement that would meet their child's needs, this was preferable to their other child being burdened by the responsibility. Parents felt looking after their child with Down syndrome had come with restrictions. Their own lives had revolved completely around their child and therefore it had significantly impacted on their freedom, work and social life. They did not want their other children to be restricted in the same way. They had made the decision as parents to raise their child with Down syndrome, but did not want there to be any expectation that their other children then had to take over this role.

I didn't want X and the girls to become – feel that they'd got to be there for X, you know what I mean? On a continuous basis; they've got their own lives to lead and we've always felt they should be (Participant 06, parent).

"Don't you dare take him on yourself. You'll have to find somewhere else for him." I said, "If he's still here and we haven't found anything..." Because the rest of her life would be like ours. Whereas at the moment, she can do what she wants. But there was always that sort of worry about it (Participant 09, parent).

There were other families who felt it was important to keep the child within the family. Parents spoke of their child being able to learn more in the home environment, and there was mention of worry about challenging behaviour potentially worsening if their child was living with others who had a disability. Some parent's spoke of feeling like their child was their responsibility and, therefore, it was only right that they should remain in the family home. For these families there was a shared understanding within the family that when the parents were no longer able to care due to ill health or death, that their other children would take on the caring role.

Like all these hospitals are okay, good, the parents have put them in these places and things like that. But they all copy one another and do exactly what that one does, and all them, they do exactly what everybody does. I do think because X has always lived at home, and he benefited from that (Participant 04, parent).

It's a very difficult thing to do (caring for a child with Down syndrome), but we both felt that he was our family, he was our responsibility. It's (care at home) not something you do lightly (Participant 08, parent).

They (other children) do say that they would look after him rather than him go in to a home. I wouldn't want him to go into one of the centres (Participant 04, parent)

Transition of care - Sibling experience:

Three of the siblings that have taken on the caring role spoke about the transition of care being challenging and described the expectation that they should take on this role - the responsibility of not wanting to let their parents or sibling down. There was no formal support offered to siblings and their families during this transition and it was described as a time of emotional/financial strain for the families. Siblings taking on the caring role spoke about having to cope with feelings of bereavement for the loss of the sibling relationship. The dynamics in the relationship changed and switched to a parental/child type relationship. Difficulty in providing a parenting role was described and it took a period of adjustment to be able to offer the type of support and guidance that their sibling needed from a parent figure. One sibling described the personal conflict of wanting to continue to provide the same advice she would have done as a sibling, but knowing that her sister needed something different. There were also feelings of burden, of having the responsibility of caring for their sibling at a time when they had young children themselves.

When I think back, I think I was suffering more than I knew I was, you know? Yes, because I had a little boy of four, and then I had the baby, and then taking on X. I just thought, "Oh, if they took him away, what would happen to him?" Because he wouldn't have been able to have stood that. He has always been used to having his family around him.

Nobody, nobody but nobody wanted to help me (Participant 02, sibling).

Yes, because I lost my sister as soon as Mum died and, well not as soon as, but over the years I've become more of a parent rather than a sister. It's not something I'd want. She

seems like she needs to have me by her as a parent rather than a sister. I can't not provide that, because that's what she's coming to me for. I feel like as a parent she's missing out on something. Then I'm missing out on my sister (Participant 11, sibling).

Perpetual parent:

A shared worry across the parents and siblings was the feeling of uncertainty about what the future held for their family member with Down syndrome. Families did not feel secure that changes would not be made to funding that may impact on living arrangements and there was concern about not being around to ensure that their family member was receiving the care they needed. This feeling of concern and parental responsibility appeared to continue even when the family were no longer providing the day to day care.

Well, whenever we first started, when we first started, it was a question of, "Oh, well, he's going before us anyway." But then as we got older, and he got older, you then start to worry about-you start to worry about him outliving us, you know, and what happens to him then (Participant 08, parent).

I suppose worrying about what will happen to her when I'm gone that would be my biggest fear. If anything happened to me (Participant 10, sibling).

It will be that, it will be what will happen to him when – I know he's okay in his home, but it will be that. It will be what will happen to him when Mum goes, because we will have to really take over. That's I suppose the biggest worry (Participant 14, sibling).

Positive experiences of caring for a child/sibling with Down syndrome

Despite the difficulties and restrictions the families faced across 50 years of caring for a family member with Down syndrome, the majority of the families spoke about the experience as life-changing in a positive way. Parents spoke about looking back on their lives and having no regrets about their decision to raise their child with Down syndrome. Families reported that it had been worthwhile and rewarding. Some parents felt the experience of raising a child with Down syndrome had brought meaning to their lives.

Personally I think he was sent to the whole family for a reason. Yes. That's right, yes. You know, and we've enjoyed every bit of him (Participant 04, parent).

I wouldn't say that I wish it had never happened, because we've gained a lot from her. We really have. I mean I've never thought, Oh, I wish I'd not had her (Participant 07, parent).

There was a common thread across family responses that this experience had enriched their lives. Families felt that they had gained from the experience and it had helped them develop and reflect on what was important in life. Families explained how caring for a family member with Down syndrome had developed their compassion for others, broadened their outlook and made them more in touch with their emotions. The experience also made parents and siblings want to help other families with a child with disabilities.

I think having had X as a part of our lives, it's given us a facet to our character that if you don't have someone like him in your life, you won't have. It's enabled us to have a fuller character...I think actually X is a blessing in that sense, because he has made us better people. I get very emotional when I speak about him (Participant 14, sibling).

There was also a clear theme around the pleasure the families experienced in sharing their lives with a family member with Down syndrome. There were many happy memories that the family were able to draw on, that were recounted with a smile. Families spoke of the enthusiasm and enjoyment that their child/sibling got out of simple everyday activities, and how this was very humbling to witness. Families spoke of infectious laughter and sunny personalities that really brought joy to all those that came into contact with their child/sibling.

Well, she's got a very sunny personality and she loves – enthusiastic, she loves to do things and loves to be there. Really enjoys everything that she can and she does. She really shows her enthusiasm and enjoyment of things. I enjoy that. That's a wonderful thing that she can enjoy stuff and she does. Everybody within hearing distance also enjoys it (Participant 06, parent).

The lasting feeling of caring for a child with Down syndrome was not one of regret or sadness, but of warmth and unending love. There was a shared sense that the experience had been life-changing. The families clearly had many happy memories that they cherished about their journey alongside their child with Down syndrome.

I never wanted to be away from him once, never. I never ever – I can truly sit here in front of God and say I never thought, "Why me?" Never. It was like X and I were meant to be. Yes, he taught me things. I've read lots of things and I don't think X was here to learn as much as he was here to teach. I know my life would've taken a much different course had I not have had X. How to be a proper human being and just to care. Really care (Participant 13, parent).

Discussion

The purpose of this study was to explore the experience and challenges of caring for a child with Down syndrome across their life span. This prolonged caregiving role was described by the majority of families as rewarding and worthwhile and, on the whole, families confirmed that their lasting feelings were very positive. However, inevitably, there were many challenges. The following challenges were commonly described across the families: health care professionals having a negative outlook about Down syndrome; lack of information and support from professionals; difficulty reaching a decision about how long to keep their child at home; deciding whether to transfer to a community placement or for another family member to take over the care. The families identified a number of personal and external factors that helped them cope: family support, personal characteristics, the temperament and level of disability of the child, having more children, economic circumstances, faith, and seeing it as a mutually benefitting relationship (as the child provided companionship).

Early experiences and feelings of having child with Down syndrome

Being told that your child has Down syndrome was clearly an emotional time for all the families that were interviewed. It was striking how easily and vividly the parents were able to recall the memory. Many of the parents became tearful recounting the story of exactly what was said and said it brought back a number of painful feelings. However, this supports existing literature that indicates that the first words about the diagnosis are remembered by parents with incredible accuracy (Skotko, 2005). There was a sense that this would always be difficult news to hear as a parent but it was made more distressing by

the manner in which the professionals shared the news (Helm, Miranda, & Chedd, 1998). Comparing this experience 50 years ago to how families are told the news now did not reveal a lot of change. At both time points, parents described professionals having a negative tone with lack of indication of any hope for their child and there was a lack of information about what the family was to expect and where it could access support (Skotko, 2005).

A significant difference between receiving the diagnosis 50 years ago and comparing this to today relates to the timing of when parents are told the news. There is now the option to have an amniocentesis test to discover any abnormalities during pregnancy (Skotko, Kishnani & Capone, 2009). Thus, parents today have a choice, firstly about whether they want to undergo the test, and then, if they are told they are expecting a baby with Down syndrome or any other condition/health problem, they have the choice about whether they wish to terminate the pregnancy. The literature indicates that, whether parents find out before birth or after birth, initial emotions do not appear to vary that much. Receiving the diagnosis is invariably traumatic to parents and evokes a response of grief (Nelson Goff et al., 2013; Pianta, Marvin, Britner & Borowitz, 1996).

In considering the information gathered from the parents in this study about reactions to receiving the diagnosis, the feeling of distress was quickly consumed by other emotions. Every parent interviewed felt very protective once their child was delivered and they were horrified at the suggestion of not keeping their baby when offered that option. This is important information to be shared with professionals and families so that there is an awareness that the early distress parents experience does diminish, that grief has different stages, and that most people will naturally pass through and adjust emotionally (Sheets et al., 2011).

Although finding out at such a late stage would happen less frequently today, from this study we discovered that the parents who found out the latest, sometimes several months after birth, felt grateful for this delay in discovering the diagnosis as it meant they had had the chance to bond with their baby. Parents should be allowed to make a decision that is right for them, including the timing of receiving critical information (i.e. whether they want to know during pregnancy or at birth). To be able to make an informed decision about this, parents need to be informed of the purpose of antenatal screening tests, the risks involved and possible implications of the results (Hall et al., 2007). It is equally a decision that should be respected whether parents decide to keep their baby or not. Skotko (2005) found that parents who found out pre-natally were happier at birth than parents who discovered their child had Down syndrome at birth. The parents who had found out pre-natally had experienced a period of adjustment and had also already made a conscious decision to keep the child. It is important that parents are able to make an informed decision about what is right for them as a family, for which they require both an understanding of the challenges involved and also the positive experience of caring for a child with Down syndrome.

In terms of helping future families, this study supports other literature examining receiving a diagnosis of Down syndrome. Families want more balance in the information that professionals provide and in the tone that it is delivered. Therefore, instead of reporting all the possible negative impacts such as deficits and health impacts, parents would also like professionals to talk about the potential for their child's future and to offer reassurance by explaining that families can and do adjust to having a child with Down syndrome (Skotko et al., 2009). It is hard to prepare parents fully for what life with a child with Down syndrome will be like as there is much variation in the level of intellectual

disability and co-morbid health difficulties of children with the condition (Sheets et al., 2011). However, parents of children with Down syndrome said that it would be useful if professionals gave accounts of a wide spectrum of experiences and that, by keeping this in mind, professionals would ensure they provided a more balanced perspective (Hippman, Inglis & Austin, 2012). This study alone provides hope to future families as it clearly highlights that, despite challenges, there is also a positive impact when caring for a child with Down syndrome.

Several studies have looked into the breaking of bad news and these have highlighted that health care professionals feel that the training that they have to prepare for this part of their role is inadequate. There is a need for more training and practice to develop and maintain the skills needed for this role, such as being able to support informed decision- making for families. Many professionals who deliver diagnoses to families have limited or no personal experience of individuals with Down syndrome, so there is a risk that information delivered can sometimes be like reading a list of symptoms (Hippman et al., 2012). A number of studies have made recommendations that professionals who deliver news to families should establish links with local support groups and the local Down syndrome community as this will support professionals in remaining up-to-date about what life is like for individuals with Down syndrome and their families (Hippman et al., 2012; Madeo, Biesecker, Brasington, Erby & Peters, 2011; Skotko et al., 2009).

A campaign by the Down Syndrome Association (DSA) called 'Tell it right, start it right' tried to raise awareness about the importance of how professionals inform families that their child has Down syndrome (Down Syndrome Association, 2009). The way in which families are told may influence the way they adjust to the diagnosis and form a relationship with their child (Sheets et al., 2011). It could also impact on parents'

perceptions of professionals and, in turn, their willingness to seek support when needed if they have an initially negative perception of professionals. In 2010 the DSA started to offer training days accredited by the Royal College of Midwives to health care professionals working in maternity services with the aim of ensuring that those delivering the diagnoses and those working with families in the early days have up-to-date information about what it is like to live with Down syndrome. The DSA has reported positive feedback from the health care professionals about the usefulness of the training. However more research is needed on whether this is impacting positively on the family experience.

In addition to hearing the news as a parent, the experience of sharing the diagnosis with the rest of the family was described. Siblings in this study spoke of either being involved in early conversations or not realizing until they had started to ask questions such as 'why are they going to a different school'. The parents in this study had very little knowledge themselves about what the diagnoses meant, which may be why siblings were told very little. Giallo and Gavidia (2006) found that good communication within the family leads to better adjustment for siblings. A study by Hames (1994) examined when parents chose to tell siblings about their brother or sister having Down syndrome. This study found that across the parents there was an agreement that their other children needed to know and that they wanted to be the ones to inform them before they found out through others. However, the main issue seemed to be about timing and the level of detail to provide. Very young children were unable to comprehend that this was a lifelong condition and they thought their sibling would get better. It may be useful for families to be provided with information about sharing the diagnosis with siblings and about what level of detail is appropriate for different ages.

Factors that enabled carers to cope with raising a child/sibling with Down syndrome

In this study the families reported a number of personal and external factors that made coping with the caring role across 50 years more manageable. The most consistent factor that came up was family support – including acceptance of the child within the family, emotional support and practical support with the caring role. Having a close relationship with immediate and extended family members meant that there was someone to talk to about the day to day challenges and that, in times of crisis, there was someone to share the problem-solving with. However, consistent with the existing literature, there was an absence of formal structures of support as a factor to help families cope. As parents age, some find it increasingly difficult to cope with the stresses and strains of caregiving, and this is particularly the case if their main source of support for coping is the family as the family support network is likely to decrease in size as they too reach old age. (Taggart, Truesdale-Kennedy, Ryan & Mcconkey, 2012).

The other common factor associated with coping in the current study was the level of disability and the individual temperament of the child. Some parents described feeling lucky when compared to families who had children with a more severe level of disability and did not know whether they would have been able to cope if their child was more challenging (Hodges & Dibb, 2010; Wills, 1981). Existing literature reports an association between behaviour problems and parental wellbeing (Blacher, Neece, & Paczkowski, 2005). Parents in this study described finding their child easy to manage, and a pleasure due to a happy temperament and enjoyment of life. Existing literature on wellbeing in parents of children with intellectual disability, has reported the 'Down syndrome advantage', indicating parents of Down syndrome children often report better wellbeing

than parents of children with other types of intellectual disability (Stoneman, 2007). These results contribute to the existing literature in that it is likely the difference in wellbeing is associated with level of disability and temperament (Baine, McDonald, Wilgosh & Mellon, 1993).

By having an understanding of factors that contribute to effective coping strategies, professionals can increase their awareness of potentially vulnerable groups of family carers. However, it is important for professionals to take a person-centred approach and to consider all contributing factors. Absence of a common coping factor does not automatically mean a parent is going to struggle. However, from the results of this study and from the findings in existing literature, the following groups are more at risk of not being able to cope: single parents who lack the support of a partner; families who are isolated either geographically or emotionally from extended family; families who have economic constraints; families who have a child with severe disabilities and who presents with frequent challenging behaviour; parents who have a negative outlook (Minnes, Woodford & Passey, 2007; Taanila, Syrjälä, Kokkonen & Järvelin, 2001).

Experience of caring for an adult with Down syndrome

Transition of care was highlighted as one of the main concerns for families in caring for an adult with Down syndrome. There were two key questions that families deliberated on – how long to keep their child at home, and who was going to take over the caring role. It evoked a range of emotions for parents including fear, anxiety, guilt and sadness. It was clear that parents were concerned about their child's needs being met and their feeling of responsibility about this. However, there was also an element of sadness

and fear about separation from the child who their life had revolved around for so many years. None of the parents felt they had received any professional support about this decision making process (Cairns & Brown, 2012; Taggart et al., 2012).

When people experience stress and high emotion it can impact on decision-making as stress-related changes in the brain can impact on the ability to consider and weigh up alternatives and the stress response can be heightened when there is uncertainty or risk around the decision (Starcke & Brand, 2012). Therefore, having someone professional and who is objective with whom to talk through the options available to their child would probably be very beneficial for many families – to help them consider the short and long term implications and to help them make an informed decision. It may be helpful for families to start conversations like this early so that whatever they decide can be a planned decision, rather than acting in an emergency situation (Grant, Ramcharan & Flynn, 2007). Having open conversations within the family early on will hopefully enable siblings to share their feelings about taking on the caring role if that is what the family would like. Open communication in families is seen as a supportive factor and is often seen in families who are functioning well (Taanila et al., 2001) If families do decide to find a community placement, to make the transition as easy as possible it would be helpful for professionals involved to support the development of a plan to maintain family relationships.

Positive experiences of caring for a child/sibling with Down syndrome

After up to 50 years of caring and factoring in all the challenges, the most enduring emotion that families were left experiencing was unending love for their child/sibling with Down syndrome. Parents spoke of having no regrets and that the experience had been life-

changing for them in a positive way. There were many examples of resilience in the stories the families shared of a lifetime of caring. They faced numerous difficulties along their journey, but their ability to draw on inner resources and continue with a positive outlook was quite remarkable.

Past research examining the experience of caring for a child with Down syndrome has mainly focused on measuring the negative impact or absence of negative impact on families, compared to parents of children with other types of intellectual disability or parents of children without disabilities (Hodapp, Ricci, Ly & Fidler, 2003; Stoneman, 2007). However, there has been a shift in recent years in research examining parents caring for a child with Down syndrome, and other types of intellectual disability, towards exploring the adjustment process including aspects such as acceptance and satisfaction of the caring role (Abery, 2006; Jokinen & Brown, 2005; Kandell & Merrick, 2007; Van Riper, 2007), and the data from this study is describing not only acceptance and satisfaction, but a pronounced level of positive emotion.

Limitations of the study/directions for future research:

This was part of a longitudinal study and relied on parents/siblings recalling information from up to 50 years ago. Although family members with known memory problems were excluded from the study, it has to be noted that there may have still been issues with the accuracy of recall of information. Therefore, this may have compromised the reliability of the findings.

Although the original sample of families who signed up to take part in Carr's cohort study (1995) were considered to be a fairly representative sample, the current

sample may not be as representative over time as would be expected with a longitudinal study as attrition has resulted due to death and ill health of parents. Therefore, the reliability of the findings may be compromised as those that are no longer part of the study may have reported different experiences and this limits the generalizability of the current findings.

The current sample of parents was made up of 6 mothers, which means that the findings may not be generalizable to fathers of children with Down syndrome. Although it is well documented that mothers typically take the primary caregiving role, there is a lack of literature that examines the experience and challenges faced by fathers of children with Down syndrome. Future research that focusses on fathers would be useful to further understanding and how best to support the whole family unit.

There is a need for further longitudinal studies that examine the experience and challenges of parents of children with Down syndrome across time. This would help to identify the key experiences and challenges of prolonged caregiving, and determine whether the impact of this role fluctuates over time. This information would help professionals to identify vulnerable time points across the life span and to tailor support from services to meet the changing needs of this group of carers. Today, parents have the choice about whether they wish to do prenatal tests to determine whether their child has Down syndrome. Future studies could examine whether there is any difference in the experience and challenges across time of parents who make a conscious choice to keep their child with Down syndrome following prenatal tests and those parents who find out at birth.

This study described transition of care and some siblings in the study had taken over the caring role from their parents. Further research is needed to examine the long-term impact of taking on the care of a sibling with Down syndrome. A comparison study could be carried out to examine what impact this extra responsibility has on the wellbeing of siblings, comparing siblings who do take on the caring role of a sibling with Down syndrome with siblings who do not take over the care due to the sibling with Down syndrome going into a care home.

Implications for future families/professionals

The findings from this study provide insight into the experience, challenges and emotional journey across 50 years of being a parent of a child with Down syndrome. New parents have described that they would find it helpful to hear more personal accounts of the experience of being a parent of a child with Down syndrome. Knowing what to expect and what others have found helpful may help new parents to increase their feeling of control over a situation that has developed out of their control. It is hoped the findings from this study will help professionals to understand the importance of sharing more personal experiences with families. It may be helpful for professionals to consider how they can communicate this information objectively to parents without it being biased by the tone of delivery of professionals or the way the initial information is interpreted by the parents. Perhaps a leaflet or website with videos could be developed which documents quotes from parents about their experiences.

With regard to professionals, the findings have highlighted a number of areas that may help professionals develop further the support they provide to families. When delivering

the initial diagnosis, families want professionals to handle this sensitively so that they feel contained and supported. Professionals need to aim to provide a balance of information tailored to meet each individual family's needs. Appropriate and up to date training and supervision is needed for health care professionals who have the role of delivering the diagnosis to families. The two major time points at which families would have liked more support were when they received the initial diagnosis and, later in life, in relation to transition of care.

Conclusion

This study provides significant contribution to the research into the longitudinal experience of caring for a child with Down syndrome. It highlights that despite challenges families adjust to this caring role, and look back on a lifetime of caring and describe the experience as being worthwhile and rewarding. However, further research is recommended to continue to build on understanding of the impact of caring across time on different members of the family.

References

Abery, B. H. (2006). Family adjustment and adaptation with children with down syndrome. *Focus on Exceptional Children*, 38 (6) 1-18.

Baine, D., McDonald, L., Wilgosh, L., & Mellon, S. (1993). Stress experienced by families of older adolescents or young adults with severe disability. *Australia and New Zealand Journal of Developmental Disabilities, 18***,** 177-188.

Blacher, J., & McIntyre, L. L. (2006). Syndrome specificity and behavioural disorders in young adults with intellectual disability: Cultural differences in family impact. Journal of Intellectual Disability Research, 50, 184–198.

Blacher, J., Neece, C. L., & Paczkowski, E. (2005). Families and intellectual disability. Current Opinion Psychiatry, 18, 507-513.

Brown, R. I., & Brown, I. (2005). The application of quality of life. *Intellectual Disabilities Research*, 49, 718-727.

Cairns, D., & Brown, J. (2012). The need for future alternatives: an investigation of the experiences and future of older parents caring for offspring with learning disabilities over a prolonged period of time. *British Journal of Learning Disabilities,* 41, 73-82.

Cairns, D., Brown, J., Tolson, D., & Darbyshire, C. (2014). Caring for a child with learning disabilities over a prolonged period of time: An exploratory survey on the experiences and health of older parent carers living in scotland. *Journal of Applied Research in Intellectual Disabilities, 27* (5), 471-480.

Caples, M., & Sweeney, J. (2010). Quality of life: a survey of parents of children/adults with an intellectual disability who are availing of respite care. *British Journal of Learning Disabilities*, 39, 64-72.

Carr, J. (1995). *Down's syndrome. Children growing up.* Cambridge: Cambridge University Press.

Carr, J. (1988). Six weeks to twenty one: a longitudinal study of a children with down's syndrome. *Journal of Child Psychology and Psychiatry*, 29 (4) 407 – 431.

Carr, J. (2008). Families of 40-year olds with down syndrome. *Journal on Developmental Disabilities, 14 (2),* 35-43.

Carr, J. (2012). Six weeks to 45years: a longitudinal study of a population with down syndrome. Journal *of applied research in intellectual disabilities*. 25 (5), 414 – 422.

Chou, Y., Pu, C., Kroger, T., & Fu, L. (2010). Caring, employment, and quality of life: Comparison of employed and nonemployed mothers of adults with intellectual disability. *American Journal on Intellectual and Developmental Disabilities, 115* (5), 406-420.

Cuskelly, M. (2006). Parents of adults with an intellectual disability. *Family Matters, 74,* 20-25.

Down Syndrome Association (2009). *Tell it right, start it right.* Available at: http://www.Downs-syndrome.org.uk/policy-and-campaigns/tell-it-right-start-it-right/ (Accessed 04 May 2015)

Ellison, C., White, A., & Chapman, L. (2011). Avoiding institutional outcomes for older adults living with disability: The use of community-based aged care supports. *Journal of Intellectual and Developmental Disability, 36* (3), 175-183.

Giallo, R., & Gavidia-Payne, S. (2006). Child, parent and family factors as predictors of adjustment for siblings of children with a disability. *Journal of Intellectual Disability Research, 50,* 937-948.

Ghosh, S., Greenberg, J. S., & Seltzer, M. M. (2012). Adaptation to a spouse's disability by parents of adult children with mental illness or developmental disability. *Psychiatric Services, 63* (11), 1118-1124.

Grant, G., Ramcharan, P., & Flynn, M. (2007). Resilience in families with children and adult members with intellectual disabilities: Tracing elements of a psycho-social model. *Journal of Applied Research in Intellectual Disabilities, 20* (6), 563-575.

Greeff, A. P., Vansteenwegen, A., & Gillard, J. (2012). Resilience in families living with a child with a physical disability. *Rehabilitation Nursing: The Official Journal of the Association of Rehabilitation Nurses, 37* (3), 97-104.

Ha, J., Hong, J., Seltzer, M. M., & Greenberg, J. S. (2008). Age and gender differences in the well-being of midlife and aging parents with children with mental health or developmental problems: Report of a national study. *Journal of Health and Social Behavior, 49* (3), 301-316.

Hall, S., Chitty, L., Dormandy, E., Hollywood, A., Wildschut, H. I. J., Fortuny, A., Masturzo, B., Santavy, J., Kabra, M., M. R., & Marteua, T. M. (2007). Undergoing prenatal screening for down's syndrome: presentation of choice and information in Europe and Asia. *European Journal of Human Genetics,* 15, 563-569.

Hames, A. (1994). Telling children that they have a brother or sister with down's syndrome. *British Journal of Learning Disabilities, 22* (2), 62-65.

Helm, D. T., Miranda, S., & Chedd, N. A. (1998). Prenatal diagnosis of down syndrome: mothers' reflections on supports needed from diagnosis to birth. *Mental Retardation*, 36, 55–61.

Hodapp R. M., Ricci L. A., Ly R. M. & Fidler D. J. (2003) The effects of the child with down syndrome on maternal stress. *British Journal of Developmental Psychology*, 21, 137–51.

Hippman, C., Iglis, A., Austin, J. (2012). What is a "balanced" description? Insight from parents of individuals with down syndrome. *Journal of Genetic Counselling*, 21 (1), 35-44.

Hodges, L., & Dibb, B. (2010). Social comparison within self-help groups. Views of parents of children with duchenne muscular dystrophy. *Journal of Health Psychology*, 15 (4), 483-492.

Jokinen, N. S., & Brown, R. I. (2005). Family quality of life from the perspective of older parents. *Journal of Intellectual Disability Research*, 49, 789–93.

Kandel, I., & Merrick, J. (2003). The birth of a child with disability. Coping by parents and siblings. *The Scientific World Journal, 3*, 741.

Kandel, I., & Merrick, J. (2007). The child with a disability: Parental acceptance, management and coping. *The Scientific World Journal, 7*, 17-99.

Korenromp, M. J., Page-Christiaens, G., van, d. B., Mulder, E. J. H., & Visser, G. H. A. (2007). Maternal decision to terminate pregnancy in case of down syndrome. *American Journal of Obstetrics and Gynaecology, 196* (2), 149 e1-11.

Madeo, A. C., Biesecker, B. B., Brasington, C., Erby, L. H., Peters, K. F. (2011). The relationship between the genetic counselling profession and the disability community: A commentary. *American Journal of Medical Genetics*. 155, 1777–1785.

Minnes, P. M., & Woodford, L. M. (2004). Well-being in ageing parents caring for an adult with a developmental disability. *Journal on Developmental Disabilities*, 11, 47-66.

Minnes, P., Woodford, L., & Passey, J. (2007). Mediators of well-being in ageing family carers of adults with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities*, 20, 539-552.

Nelson Goff, B. S., Springer, N., Foote, L. C., Frantz, C., Peak, M., Tracy, C., Veh, T., Bentley, G. E., & Cross, K. A. Receiving the initial down syndrome diagnosis: a comparison of prenatal and postnatal parent group experiences. *Intellectual and Developmental Disabilities*, 51 (6), 446-57.

Parish, S. L., Seltzer, M. M., Greenberg, J. S., & Floyd, F. (2004). Economic implications of caregiving at midlife: Comparing parents with and without children who have developmental disabilities. *Mental Retardation, 42*, 413– 426.

Patti, P., Amble, K., & Flory M. (2010). Placement, relocation and end of life issues in aging adults with and without down's syndrome: a retrospective study. *Journal of Intellectual Disability Research*, 54 (6), 538-546.

Pianta, R. C., Marvin, R. S., Britner, P. A., & Borowitz, K. C. (1996). Mothers' resolution of their children's diagnosis: Organized patterns of caregiving representations. *Infant Mental Health Journal, 17* (3), 239-256.

Ritchie, J., & Spencer, L. (1994). Qualitative data analysis for applied policy research. In: Bryman., A., & Burgess, R. editors. *Analysing qualitative data*. London: Routledge, 173–194.

Sanders, J., & Morgan, S. (1997). Family stress and adjustment as perceived by parents of children with autism or down syndrome: Implications for intervention. *Child & Family Behavior Therapy, 19* (4), 15-32.

Seltzer, M. M., Floyd, F., Song, J., Greenberg, J., & Hong, J. (2011). Midlife and aging parents of adults with intellectual and developmental disabilities: Impacts of lifelong parenting. *American Journal on Intellectual and Developmental Disabilities, 116* (6), 479-499.

Sheets, K. B., Crissman, B. G., Feist, C. D., Sell, S. L., Johnson, L.R., Donahue, K. C., Masser-Frye, D., Brookshire, G. S., Carre, A. M., Lagrave, D., & Brasington, C. K. (2011). Practice guidelines for communicating a prenatal or postnatal diagnosis of down syndrome: Recommendations of the National Society of Genetic Counselors. *Journal of Genetic Counselling*, 20 (5), 432-41.

Skotko, B. (2005). Mothers of children with down syndrome reflect on their postnatal support. *Pediatrics*, 115 (1), 64-77.

Skotko, B. G., Kishnani, P. S., & Capone, G. T. (2009). Prenatal diagnosis of down syndrome: How best to deliver the news. *American Journal of Medical Genetic,* 149 (11), 2361-2367.

Skotko, B. G., Levine, S. P., & Goldstein, R. (2011). Having a brother or sister with down syndrome: Perspectives from siblings. *American Journal of Medical Genetics, 155* (10), 2348-2359.

Skotko, B., & Bedia, R. C. (2005). Postnatal support for mothers of children with down syndrome. *Mental Retardation: A Journal of Practices, Policy and Perspectives, 43* (3), 196-212.

Starcke, K., & Branda, M. (2012). Decision making under stress: A selective review. *Neuroscience and Behavioural Reviews*, 36 (4), 1228–1248.

Stoneman, Z. (2007). Examining the down syndrome advantage: Mothers and fathers of young children with disabilities. *Journal of Intellectual Disability Research, 51* (12), 1006-1017.

Taanila, A., Syrjälä, L., Kokkonen, J., & Järvelin, M. (2002). Coping of parents with physically and/or intellectually disabled children. *Child: Care, Health and Development, 28* (1), 73-86.

Taggart, L., Truesdale-Kennedy, M., Ryan, A., & Mcconkey, R. (2012). Examining the support needs of ageing family carers in developing future plans for a relative with an intellectual disability. Journal of Intellectual Disabilities, 16 (3), 217-234.

Torr, J., Strydom, A., Patti, P., & Jokinen, N. (2010). Aging in down's syndrome: morbidity and mortality. *Journal of Policy and Practice in Intellectual Disabilities*, 7 (1), 70-81.

Van Riper, M. (2007). Families of children with down syndrome: responding to "a change in plans" with resilience. *Journal of Pediatric Nursing, 22 (2), 116-128.*

Wilkinson, H., Kerr, D., Cunningham, C., & Rae, C. (2008). *Home for good? Preparing to support people with learning difficulties in residential settings when they develop dementia.* Brighton: Joseph Rowntree Foundation – Pavillion Publishing Ltd.

Wills, T. A. (1981). Downward comparison principles in social psychology. *Psychological Bulletin*, 90, 245–271.

Public Domain Briefing Document
Resilience in parents of adults with intellectual disability

The research was conducted by Leah Wood as part of the Doctorate in Clinical Psychology at the University of Birmingham. The research was supervised by Dr Chris Oliver and Dr Beth Grunfeld. There are two chapters to this research project: a literature review and a research study.

First a literature review was carried out to examine existing findings on the impact of caring for an adult child with intellectual disability. 23 studies were identified as meeting the criteria. Across the studies there were 11 different outcomes measured: depression, overall health, mental health, physical health, stress, burden, quality of life, psychological wellbeing, negative affect, family problems and life satisfaction. For the purpose of the review only the outcome measures examined most were discussed in more detail. These were as follows: depression, psychological wellbeing, overall health and physical health.

Overall, the findings from the review indicated poorer psychological wellbeing and overall health of parents of an adult child with an intellectual disability when compared to parents of an adult child without disability. There were a number of factors identified that mediated the impact of caring for an adult child with intellectual disability. These include: support, behavioural problems, employment, health, age, residence of adult child and additional caring responsibility. Due to a range of mediating variables, it was difficult to establish clear predictors of poor outcomes in parents of adults with intellectual disability. Therefore, further research is needed which examines prolonged caregiving and outcomes across time. Having a clear understanding of predictors of outcome would

inform professionals and families about what support may be required to help maintain the physical and psychological health of this group of ageing parent caregivers.

The second chapter is a research study that explored the experiences and challenges of caring for a child with Down syndrome across 50 years. Participants were parents/siblings of Janet Carr's initial Down syndrome cohort study. Fifteen family members from twelve different families were interviewed (8 parents, 7 siblings). Four themes about the experience of caring for a child with Down syndrome are discussed in more detail: early experiences and feelings, factors that enabled carers to cope, experience of caring for an adult with Down syndrome and positive experiences.

The study found that, on the whole, parents experienced medical professionals to be insensitive and unhelpful in relation to how they handled breaking the news that their child had Down syndrome. However, despite initial feelings of distress, there were no doubts for these parents about keeping their babies. A range of coping factors enabled these families to adjust and cope with the challenges associated with caring for a child with Down syndrome. Coping factors reported included the following: family support, personal characteristics, the temperament and level of disability of the chid, having more children, economic circumstances, faith, and seeing it as a mutually benefitting relationship (as the child provided companionship).

The families varied in relation to their views on transition of care once the individual with Down syndrome had reached adulthood. Some families had strong views about keeping the adult with Down syndrome within the family and would pass on the caring role to their other children when they could no longer provide the care. Other families, in contrast, held the view that they did not want to burden their other children and

that finding an appropriate community placement was preferable. These families tended to make the transition of care earlier to give their child an opportunity to build his/her own life rather than this being enforced due to ill health or death of the parent(s). In spite of the challenges that these families faced across the 50 years, there was a consensus across the families that, overall, they felt that it had been a worthwhile and positive experience to care for a child with Down syndrome.

It is hoped the findings of this study will help professionals to consider and tailor services for working with families who have a child with Down syndrome. More support is needed at the start of the journey and at critical time points across the life span, particularly when planning for transition of care. Further research has been recommended on the long- term experiences of fathers and siblings caring for a child with Down syndrome.

APPENDICES

Appendix 1: Quality Assessment Checklist (Kmet et al., 2004)

1	Question / objective sufficiently described?				
2	Study design evident and appropriate?				
3	Method of subject/comparison group selection or source of information/input variables described and appropriate?				
4	Subject (and comparison group, if applicable) characteristics sufficiently described?				
5	If interventional and random allocation was possible, was it described?				
6	If interventional and blinding of investigators was possible, was it reported?				
7	If interventional and blinding of subjects was possible, was it reported?				
8	Outcome and (if applicable) exposure measure(s) well defined and robust to measurement / misclassification bias? means of assessment reported?				
9	Sample size appropriate?				
10	Analytic methods described/justified and appropriate?				
11	Some estimate of variance is reported for the main results?				
12	Controlled for confounding?				
13	Results reported in sufficient detail?				
14	Conclusions supported by the results?				

Appendix 2: Quality Assessment Results

Study	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Q10	Q11	Q12	Q13	Q14	Total Score	Rating
Ben-Zur, H., Duvdevany, I., & Lury, L. (Israel)	2	2	1	2	N/A	N/A	N/A	2	1	2	2	2	2	1	19/22 86%	High
Cairns, D., Brown, J., Tolson, D., & Darbyshire, C. (Scotland – UK)	2	2	1	2	N/A	N/A	N/A	2	1	1	2	1	2	2	18/22 82%	High
Carr, J. (UK)	2	2	1	2	N/A	N/A	N/A	2	1	2	2	2	2	2	20/22 91%	High
Chen, S. C., Ryan-Henry, S., Heller, T., & Chen, E. H (USA)	2	2	1	2	N/A	N/A	N/A	2	1	2	2	2	2	2	20/22 91%	High
Chou, Y., Pu, C., Kroger, T., & Fu, L (Taiwan)	2	2	1	2	N/A	N/A	N/A	2	2	2	2	2	2	2	21/22 95%	High
Chou, Y. C., Lee, Y. C., Lin, L. C., Kroger, T., & Chang, A. N (Taiwan)	2	2	2	2	N/A	N/A	N/A	2	2	2	2	1	2	2	21/22 95%	High
Einam, M., & Cuskelly, M (Australia)	2	2	1	2	N/A	N/A	N/A	1	1	2	2	2	2	2	19/22 86%	High
Ghosh, S., Greenberg, J. S., & Seltzer, M. M (USA)	2	2	2	2	N/A	N/A	N/A	2	2	2	2	1	2	2	21/22 95%	High
Ha, J., Hong, J., Seltzer, M. M., & Greenberg, J. S (USA)	2	2	2	2	N/A	N/A	N/A	2	2	2	2	2	2	2	22/22 100%	High
Hong, J., Seltzer, M.,	2	2	1	2	N/A	N/A	N/A	2	2	2	2	2	2	2	21/22 95%	High

Study	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Q10	Q11	Q12	Q13	Q14	Total Score	Rating
& Krauss, W (USA)																
Kim, H. W., Greenberg, J. S., Seltzer, M. M., & Krauss, M. W (USA)	2	2	1	2	N/A	N/A	N/A	2	1	2	2	2	2	2	20/22 91%	High
Magana, S., Seltzer, M. M., Krauss, M. W., Rubert, M., & Szapocznik, J (USA)	2	2	1	2	N/A	N/A	N/A	2	1	1	2	2	2	2	19/22 86%	High
Magana, S., Seltzer, M. M., & Krauss, M. W (USA)	2	2	1	2	N/A	N/A	N/A	2	1	2	2	1	2	2	19/22 86%	High
Miltiades, H. B., & Pruchno, R (USA)	2	2	1	2	N/A	N/A	N/A	2	1	2	2	2	2	2	20/22 91%	High
Miltiades, H. B., & Pruchno, R (USA)	2	2	1	2	N/A	N/A	N/A	2	2	2	0	2	2	2	19/22 86%	High
Minnes, P. M., & Woodford, L. M (Canada)	2	2	1	2	N/A	N/A	N/A	2	1	2	2	1	2	2	19/22 86%	High
Piazza, V. E., Floyd, F. J., Mailick, M. R., & Greenberg, J. S (USA)	2	2	2	2	N/A	N/A	N/A	2	2	2	2	2	2	2	22/22 100%	High
Pruchno, R. A (USA)	2	2	1	2	N/A	N/A	N/A	2	2	2	2	1	2	2	20/22 91%	High
Pruchno, R. A., & Meeks, S (USA)	2	2	1	2	N/A	N/A	N/A	2	2	2	2	2	2	2	21/22 95%	High

Study	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Q10	Q11	Q12	Q13	Q14	Total Score	Rating
Rimmerman, A., & Muraver, M (Israel)	2	2	1	2	N/A	N/A	N/A	2	2	2	1	1	2	2	19/22 86%	High
Seltzer, M. M., Floyd, F., Song, J., Greenberg, J., & Hong, J (USA)	2	2	2	2	N/A	N/A	N/A	2	2	2	2	2	2	2	22/22 100%	High
Walden, S., Pistrang, N., & Joyce, T (UK)	2	2	1	2	N/A	N/A	N/A	2	1	2	2	2	2	2	20/22 91%	High
Yamaki, K., Hsieh, K., & Heller, T (USA)	2	2	1	2	N/A	N/A	N/A	2	1	2	2	2	2	2	20/22 91%	High

Appendix 3: Outcome Measures Examined

	Outcome Measure Examined										
	Depression	**Mental Health**	**Stress**	**Physical Health**	**Burden**	**Quality of Life**	**Health**	**Psychological Wellbeing**	**Negative affect**	**Family problems**	**Life satisfaction**
Ben-Zur, H., Duvdevany, I., & Lury, L. (Israel)		✓NS	✓S								
Cairns, D., Brown, J., Tolson, D., & Darbyshire, C. (Scotland – UK)		✓*		✓*							
Carr, J. (UK)	✓NS										
Chen, S. C., Ryan-Henry, S., Heller, T., & Chen, E. H (USA)		✓NS		✓S							
Chou, Y., Pu, C., Kroger, T., & Fu, L (Taiwan)						✓S	✓S				
Chou, Y. C., Lee, Y. C., Lin, L. C., Kroger, T., & Chang, A. N (Taiwan)						✓S					
Einam, M., & Cuskelly, M (Australia)	✓S										

	Outcome Measure Examined										
	Depression	**Mental Health**	**Stress**	**Physical Health**	**Burden**	**Quality of Life**	**Health**	**Psychological Wellbeing**	**Negative affect**	**Family problems**	**Life satisfaction**
Ghosh, S., Greenberg, J. S., & Seltzer, M. M (USA)	✓S			✓NS			✓S	✓S			
Ha, J., Hong, J., Seltzer, M. M., & Greenberg, J. S (USA)				✓S				✓S	✓S		
Hong, J., Seltzer, M., & Krauss, W (USA)								✓*			
Kim, H. W., Greenberg, J. S., Seltzer, M. M., & Krauss, M. W (USA)	✓*				✓*						
Magana, S., Seltzer, M. M., Krauss, M. W., Rubert, M., & Szapocznik, J (USA)	✓NS				✓S					✓S	
Magana, S., Seltzer, M. M., & Krauss, M. W (USA)	✓S									✓S	

	Outcome Measure Examined										
	Depression	**Mental Health**	**Stress**	**Physical Health**	**Burden**	**Quality of Life**	**Health**	**Psychological Wellbeing**	**Negative affect**	**Family problems**	**Life satisfaction**
Miltiades, H. B., & Pruchno, R (USA)					✓NS		✓s				
Minnes, P. M., & Woodford, L. M (Canada)	✓NS		✓NS								
Pruchno, R. A., & Meeks, S (USA)	✓S										
Rimmerman, A., & Muraver, M (Israel)								✓NS			✓NS
Seltzer, M. M., Floyd, F., Song, J., Greenberg, J., & Hong, J (USA)	✓S						✓S	✓NS			
Walden, S., Pistrang, N., & Joyce, T (UK)	✓S		✓NS					✓S			
Yamaki, K., Hsieh, K., & Heller, T (USA)							✓S				
TOTAL studies(0ut of 20):	**10** 3	**3** 2	**3** 2	**4** 2	**3** 1	**2** 0	**5** 0	**6** 2	**1** 0	**2** 0	**1** 1

	Outcome Measure Examined										
	Depression	**Mental Health**	**Stress**	**Physical Health**	**Burden**	**Quality of Life**	**Health**	**Psychological Wellbeing**	**Negative affect**	**Family problems**	**Life satisfaction**
Non-Significant (NS):	6	0	1	1	1	2	5	3	1	2	0
Significant (S):											
Differences reported - but not analysed for significance(*):	1	1	0	1	1	0	0	1	0	0	0

Appendix 4: Interview Schedule

DS50 – Interview Schedule

Introduction to interview process

Thank you for taking part in this interview. We are conducting these interviews as we are interested in finding out more about the life-long experience of being a family member of, or caring for, a person with Down's Syndrome. This is an interview about the story of _____'s life. I am interested in hearing about your part in that story. The story is selective; it does not include everything that has ever happened to you both. Instead, I will ask you to focus on a few key things in your life – a few key events, people, and thoughts. Some of the questions will focus on your experiences and some will focus on your thoughts about things that _____ experienced. There are no right or wrong answers to my questions. Instead, I would like you simply to tell me about some of the most important things that have happened in your life with _____. I will guide you through the interview so that we finish it all in about an hour and half or less. I would like to record the interview as I will not be able to write Down all that is said. I hope that you will enjoy the interview. Do you have any questions before we start?

Notes: The interviewer should feel free to ask questions of clarification and elaboration throughout the interview

Pregnancy* [for parents or older siblings who might have a memory of this]

I would like you to think about when you (or your partner/mother) were pregnant with _____. I would be grateful if you could tell me about the pregnancy including how you felt when you found out about the pregnancy, what your hopes/aspirations were and any concerns or issues that might have arisen.

Notes:

Birth

Following the birth of _____ can you talk me through how and when you first became aware of _____'s condition and how you received the diagnosis? What experience or knowledge did you have about Down's Syndrome prior to the pregnancy? I would also appreciate it if you could think about how you felt in response to the diagnosis, how you coped in those early days (including any support/advice you

received – from medical staff, friends and family) and what your concerns/fears were for the future.

Notes:

Positive childhood memory

I would like you to think of an early memory from _____'s childhood or teen-aged years – that stands out as especially positive in some way. This would be a very positive, happy memory from these early years. Please describe this good memory in detail. What happened, where and when, who was involved, and what were you thinking and feeling? Also, please say a word or two about why you think this particular moment was so good and what the experience may say about you or your life your life with _____?

N**otes:**

Negative childhood memory I would like you to think of an early memory from _____'s childhood or teen-aged years – that stands out as especially negative in some way. This would be a very negative, unhappy memory from these early years. Please describe this memory in detail. What happened, where and when, who was involved, and what were you thinking and feeling? Also, please say a word or two about why you think this particular moment was so bad and what the experience may say about you or your life your life with _____?

Notes:

Impact on the family and siblings

Can you describe the rest of your family to me? Can you tell me about _____'s brothers and sisters, their parents and extended family? [The focus will depend on who you have interviewed] What role did each of these people take in caring for _____?

What impact do you think that _____ had on each of these people, both in terms of positive and negative outcomes?

Notes:

Vivid adult memory

Moving ahead to _____'s adult years, please identify one experience or event that you have not already described that stands out as especially vivid or meaningful. This would be an especially memorable, vivid, or important scene, positive or negative. Please describe this scene in detail, tell what happened, when and where, who was involved, and what you were thinking and feeling. Also, what does this memory say about you or your life with _____?

Notes:

Recent memory

Let us think specifically now about the last five years. Please identify one experience or event that you have not already described that stands out as especially meaningful. This would be an especially memorable or important experience, positive or negative. Please describe this in detail, tell what happened, when and where, who was involved, and what you were thinking and feeling. Also, what does this memory say about you or your life with _____?

Notes:

Health

If you look back over _____'s life, what would you consider to be the major health challenge or crisis that they have faced? Please describe this health problem or challenge and how it developed. If relevant please also describe the experience you had with the healthcare system regarding this problem or crisis. In addition, please also talk about how

you coped with the problem and what impact you think this problem had both on _____ and yourself.

Notes:

Challenges

Raising any child is associated with various challenges, struggles, and problems. Looking back over your life, please identify and describe what you now consider to be the greatest single challenge you have faced in caring for _____. What is or was the challenge or problem? How did the challenge or problem develop? How did you address or deal with this challenge or problem? What support did you receive?

Notes:

Future

I would like you to think about the future now, are there any specific concerns/fears you have for _____? What is the fear, can it be addressed and have you felt able to share this concern/fear with anyone?

Notes:

Coping

What factors do you think have ensured that you have been able to cope with the challenges associated with caring for a person with Down's syndrome? Please consider your own experiences and upbringing, your personal characteristics, family factors and more general factors and how these may have influenced how you coped.

Notes:

Failure, regret

Everybody experiences failure and regrets in life, even for the happiest and fun-filled lives. Looking back over your entire life, please identify and describe the greatest failure or regret you have experienced in relation to _____. Please describe the failure or regret and the way in which the failure or regret came to be. How have you coped with this failure or regret? What effect has this failure or regret had on you and your life story?

Notes:

Personal beliefs

Now, I would like to ask a few questions about your core beliefs and values and about the meaning in your life. Please give some thought to each of these questions.

Religious/ethical values – Please consider the religious or spiritual aspects of your life. Please describe your religious or spiritual beliefs and values, if indeed these are important to you. As part of this please describe your overall ethical or moral approach to life.

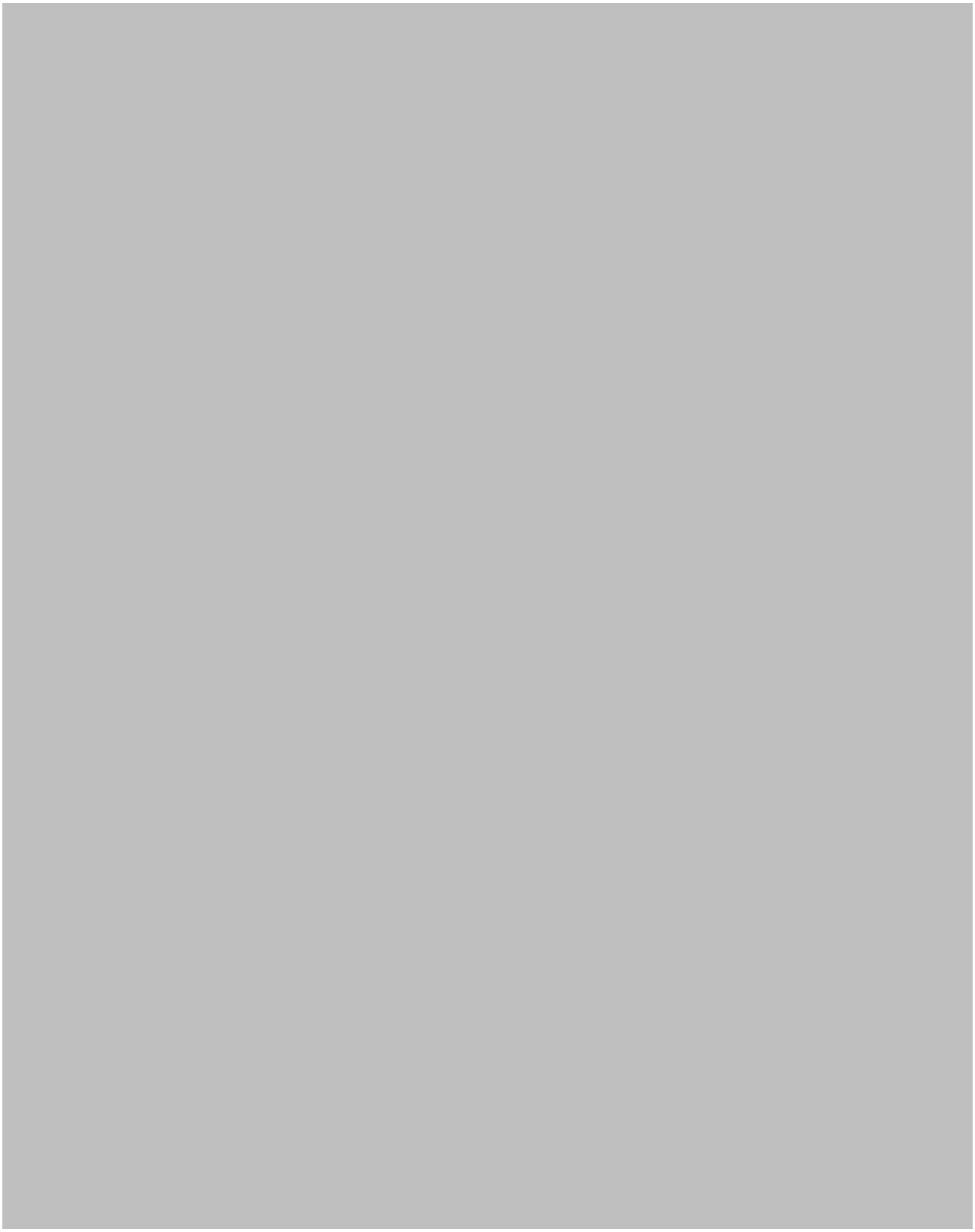
Main values – Please think about what you would consider to the most important value in human living? Please explain.

Notes:

Overall impression

I would be grateful if you would finish by thinking of a high point of _____'s life. By this I mean what would you consider to be the happy, joyous, exciting or wonderful aspect of your life with _____?

Appendix 5: Ethical Approval Confirmation



Appendix 6: Participant Information Sheet

UNIVERSITY OF BIRMINGHAM

The longitudinal experience of raising a child with Down syndrome

Information Sheet

Please read this information carefully before deciding whether you wish to take part in the study. If you have any further questions please contact [redacted] If you have any medical/ other problems which make it difficult for you to read this information, please contact Leah Wood for a verbal explanation of the research.

When you are happy that you have all of the information you need to be able to decide whether or not you would like to take part in the study, please complete the enclosed consent form and return to us in the prepaid envelope provided.

Background

We would like to invite you to take part in a research study being conducted at the Centre for Neurodevelopmental Disorders, University of Birmingham. This research work, being led by Professor Chris Oliver, looks at the life-long experience of being a family member of, or caring for, a person with Down syndrome. We hope that this information will enable us to help future families and professionals who are involved in the care of an individual with Down's syndrome.

Aims of the study

This study aims to further our understanding of being a family member of a person with Down syndrome disorder. It will be an opportunity for you to share your story of raising a child with Down syndrome. Through the use of interview you will be guided to share both positive and negative experiences you have faced, as well as considering the support you have received and resources that have helped you cope with challenges. Eventually we hope that the information gathered will help to improve the quality of life of individuals with Down syndrome and their families.

What will happen if you/ the person you care for decide(s) to participate?

Where will the research take place?
The research will take place at your home.

Who will be involved in collecting the data?
Members of the research team at the Cerebra Centre for Neurodevelopmental disorders including Leah Wood.

How long will participation in the study take?

Taking part in the study will take up 90 minutes at your home.

We will be collecting information from participants between December 2013 - August 2014. After this we will spend some time understanding the information we have collected. This means that the study will be finished in August 2015.

What will you be required to do in the study?

You will be asked to take part in an interview that will be conducted by researchers in person at your home. The interview will take approximately 90 minutes and may be recorded (with your permission) in order to assist accurate data collection. The tapes will be filed anonymously and will only be available to researchers working on the project.

Will interviews be recorded?

During the interview that we will conduct with you about the life-long experience of raising a child with Down syndrome, the interviewer's questions and your responses will be audio recorded.

The University of Birmingham will hold the copyright for the audio recordings so that the confidentiality of these recordings will be protected. But, the University of Birmingham will not be able to edit or use the recordings for teaching purposes unless you give us your written permission to do this.

We may contact you again in the future to ask your permission to use some of the recordings for teaching purposes. At that time you will be able to decide whether or not you are happy for the recordings to be used for these purposes. Agreeing to participate in this study **does not** mean that you will have to give your permission for the use of these recordings in the future.

Are there any risks that individuals taking part in the study might face?

There will not be any risks associated with participation in this study.

What are the potential benefits for participants from taking part?
This study will help us to find out more about the lives of people caring for a child with Down's syndrome. The results might help us to improve things for people with Down's syndrome and their families in the future.

Where will data be stored?
The data collected will be kept in locked or password protected storage at the University of Birmingham. All information gathered will be stored separately from any information that would allow someone to identify who you or your child are (this is known as personal identifying information, e.g. your full names, your address, your contact details). Your personal identifying information will be stored in a locked space at the University of Birmingham and only members of our research team will have access to it. We will only be able to trace the information we have collected about you and your child back to you using a special reference number which we will store in a password protected database held at the University of Birmingham. Only members of our research team will have access to that database. Personal identifying information will be treated as strictly confidential and handled in accordance with the provisions of the Data Protection Act 1998.

The audio recordings are considered to contain personal identifying information. We will therefore store the recordings of you separately to the other information we have collected about you. These

recordings will not be labelled with your names or any other personal identifying information but will be labelled with your special reference number. Recordings will be stored in a locked cabinet at the University of Birmingham and only members of our research team will have access to it.

If you decide to participate, what will happen after that participation?

The results may be presented at a conference or published in an academic journal and may be used to inform future research regarding the experience of raising a child with Down's syndrome. Please note that no identifiable information will be released in any write-up of the results. If you choose to participate in the study and would like to receive a copy of the results or final paper you may state this at the start of the interview and a copy of the results will be sent to you via the contact information you provide.

What will happen to the data afterwards?
The information that you provide will be locked in a filing cabinet at the University of Birmingham or held on a password protected database. All personal details will be kept separately from the information collected. Participants will be identified by a unique number so that it will only be possible to connect results to individuals via this number. This will ensure that results are kept anonymous. Any recording we have made of you will be destroyed 5 years after the end of the study unless you have given us your written consent for the recordings to be used for teaching and/or dissemination.

At the end of the study, your personal details will be ***destroyed unless you tell us otherwise.*** This means that we would no longer be able to trace the results back to you. ***The section below on 'The Regular Participant Database Information'*** gives information about a database that we use to store the personal details of some participants. Please read this section in order to decide if you would like to join that database.

What happens if I decide that I no longer want my details on the Regular Participant Database?
All you would need to do is contact [redacted] or at the School of Psychology, University of Birmingham, Edgbaston, Birmingham, B15 2TT. Your details would be removed from the database immediately.

For participants who are not known to us already-Regular Participant Database Information:

What is the regular participant database?
We have a database that we keep in the Cerebra Centre where we store the names and contact details of some previous participants. If you would like us to, we can add your details to this database. We would use this information for two things:

1) We will contact you with information about future research work to find out whether or not you would like to participate.
2) It is often important to find out how things change over time. By keeping your details we would be able to trace the results of the previous study that you have done with us back to you. This means that if you take part in other studies with us we would be able to look at how things have changed over time.

Who would have access to my details?
Only approved members of our research team would have access to your details. We would not share your details with anyone outside the research team.

When would I be contacted?
You would only be contacted by an approved member of the research team when we are starting another study or phase of a study that we think you might like to participate in or when we need to clarify some information that you have provided us with from participation in a research study.

What happens if I decide that I want my details to be added to the database but then I change my mind?
All you would need to do is contact [redacted] or at the School of Psychology, University of Birmingham, Edgbaston, Birmingham, B15 2TT. Your details would be removed from the database immediately.

Confidentiality

The confidentiality of participants will be ensured. However, in line with the University's Protection Procedures, the experimenter's have a duty to disclose if they have any concerns about the welfare of vulnerable adults. At this point, confidentiality may be broken to ensure safety of the individual and those around them.

If published, information on the participant will be presented without reference to their name or any other identifying information. In the unlikely event of any evidence of abuse being identified, this information will be disclosed by the research workers.

Consent
After having read all of the information and having received appropriate responses to any questions that you may have about the study you will be asked to give your consent to participate in the study if you decide that you do wish to participate. We need to receive consent from potential participants in order for them to participate.

Withdrawal
Even after consent has been granted, you can request to be withdrawn from the study and for your research data to be destroyed. You will have up to 12 months after participation to indicate your withdrawal from the study, without giving a reason. After 12 months of participating in the study, your personal details will no longer be linked to the information collected as part of this study. This means that we would no longer be able to trace the results of your assessments back to you and withdraw you from the study.

What if there is a problem?

If you have a concern about any aspect of this study, you should ask to speak to the researchers who will do their best to answer your questions. Please contact [redacted] in the first instance. If you remain unhappy and wish to complain formally, you can contact: [redacted]

Review
The study has been approved by the Science, Technology, Engineering and Mathematics Ethical Review Committee.

Further information

If you would like any more information about the study please contact [redacted]

Appendix 7: Participant Consent Form

UNIVERSITY OF BIRMINGHAM

The longitudinal experience of raising a child with Down syndrome

Consent Form for Participants

Study Director: Professor Chris Oliver

Please initial box…

1. I confirm that I have read and understood the information sheet for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily. ☐

2. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my or that of my child's/person I care for's medical care or legal rights being affected. ☐

3. I understand that all information collected during the study will be confidential. Only members of the research team at the Cerebra Centre for Neurodevelopmental disorders will know who has participated in the study. All information collected during the study will be stored in locked cabinets that only members of the research team will have access to. No names will be published in any reports. Information will be treated as strictly confidential and handled in accordance with the provisions of the Data Protection Act 1998. ☐

4. I understand that as part of the above study, voice recordings of interviews may be made and stored for further review. ☐

5. I understand that the University of Birmingham will hold the copyright of any voice recordings collected during the study but that this does not entitle the University of Birmingham to edit, copy or use the videos for teaching purposes without my written permission. ☐

6. I am happy to be contacted in the future by the University of Birmingham regarding the use of audio recordings for teaching purposes. ☐

7. I agree to take part in the above study. ☐

Print Name: _____ Telephone number: _____

Address: _____ Email: _____

Signature: _____ Date: _____

Appendix 8: Interview Transcript