

Title: Siblings of children with life-limiting conditions: Psychological adjustment and sibling relationships

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Abstract

Background This study explored psychological adjustment and sibling relationships of siblings of children with life-limiting conditions (LLCs), expanding on previous research by defining LLCs using a systematic classification of these conditions.

Methods Thirty-nine siblings participated, aged 3-16 years. Parents completed measures of siblings' emotional and behavioral difficulties, quality of life, sibling relationships, and impact on families and siblings. Sibling and family adjustment and relationships were compared to population norms, where available, and to a matched comparison group of siblings of children with autistic spectrum disorder (ASD), as a comparable 'high risk' group.

Results LLC siblings presented significantly higher levels of emotional and behavioral difficulties, and lower quality of life than population norms. Their difficulties were at levels comparable to siblings of children with ASD. A wider impact on the family was confirmed. Family socio-economic position, time since diagnosis, employment and accessing hospice care were factors associated with better psychological adjustment.

Conclusions Using a systematic classification of LLCs, the study supported earlier findings of increased levels of psychological difficulties in siblings of children with a LLC. The evidence is (a) highlighting the need to provide support to these siblings and their families, and (b) that intervention approaches could be drawn from the ASD field.

Introduction

'Life-limiting conditions' (LLCs) is an umbrella term encompassing conditions for which there is no medical hope of cure, and which ultimately leads to the death of the child (Department of Health and Children 2009). Four types of LLCs have been identified (ACT/RCPCH, 1997): (1) LLCs for which there is curative treatment, but can fail (e.g., cancer, irreversible organ failures of the heart), (2) LLCs where premature death is inevitable (e.g., cystic fibrosis, muscular dystrophy), (3) progressive LLCs without curative treatment options (e.g., Batten disease), and (4) non-progressive, irreversible LLCs causing severe disability, health complications and premature death (e.g., cerebral palsy). Over 40,000 children in England are living with LLCs (Fraser *et al.* 2012).

Family members of children with LLC, including siblings, experience family life differently to families where children do not have LLCs. Living with a brother or sister with LLC is likely to impact on siblings' psychological well-being and quality of life. However, research evidence on the impact of LLCs on siblings is limited, due, in part, to difficulties defining LLCs systematically. Previous studies have tended to consider conditions under the broader grouping of chronic conditions/illnesses. There is inevitably some overlap between LLCs and chronic conditions/illnesses. According to the ACT/RCPCH definition (1997) however, LLCs form a distinct group in which the sufferer is not expected to survive into adulthood. Therefore, the overlap between chronic illness and LLC is not complete, as certain chronic conditions are not life-limiting (e.g., asthma, diabetes). Other studies focus on single clinical conditions, for example cancer.

Negative psychological adjustment and high levels of emotional and behavioural problems have been reported in siblings of children with cancer, cystic fibrosis, cardiac and kidney difficulties, compared to their peers (Barlow & Ellard 2006; Sharpe & Rossiter 2002), and to siblings of children with muscular dystrophy (Read *et al.* 2010). Sibling relationships

have been highlighted as fractious, resentful, and competitive within families of children with diabetes, Down syndrome, and orthopedic problems (Nielsen *et al.* 2010). Sibling age and relative position within the family, time since diagnosis, gender, and socio-economic status have been associated with negative well-being in these siblings (Barlow & Ellard 2006; Breslau *et al.* 1981; Houtzager *et al.* 2003; Read *et al.* 2010). Studies have also indicated positive impacts, such as increased maturity, empathy and involvement (O'Brien, Duffy & Nichol 2009; Sloper 2000).

Existing findings cannot be easily generalized to siblings of children with LLCs due to lack of consistency in the definitions used (e.g., chronic conditions). In the present study, we address this limitation by using a recently developed classification system of LLCs (Hain *et al.* 2013). Known as the directory of LLCs, it is the first systematic attempt to group LLCs among children. It was developed by outlining several hundred LLCs according to the ACT/RCPCH 1997 definition, by reviewing referrals to hospice and specialist palliative care services, as well as death certificates from 2002-2007 across Wales (Hain *et al.* 2013; Noyes *et al.* 2013). ICD-10 labels were then assigned to the conditions to create the directory. We used the directory to identify siblings of children with conditions that would be classified as life-limiting according to the ACT/RCPCH definition.

The aim of the present study was to describe the psychological adjustment of siblings of children with a LLC, with respect to behavioral and emotional difficulties, quality of life, and sibling relationship quality. We compare sibling data to population norms, where available, but also data from a matched group of 'high risk' siblings of children with an ASD. Siblings of children with ASD experience altered family functioning, difficult sibling relationships, and reduced parental attention (Hastings 2003; Ross & Cuskelly 2006) and present heightened emotional and behavioral difficulties (Petalas *et al.* 2009). Finally, we

explored correlates of siblings' difficulties, quality of life and relationships in the LLC group so that we can begin to understand the variability in siblings' experiences.

Method

Participants

Thirty-nine families of children with LLCs participated. Primary parental caregivers were aged between 26 and 50 years-old (mean age 38 years; $SD=6.46$), and were mostly mothers (97%). Twenty-nine (74%) had a university postgraduate or undergraduate degree. Twenty-two (56%) of the families reported having an annual family income of above £35,000 (approximately \$51,000 USD). Overall, 20 (51%) held a job: six worked full-time and 14 part-time.

There were 20 boys with LLC and 19 girls, aged between 10 months and 16 years-old (with a mean age of 6.82 years ($SD=4.10$). Clinical conditions included: Congenital Heart Defects ($n=10$), Cystic Fibrosis ($n=5$), Cerebral Palsy ($n=5$), Cancer ($n=5$), Rare Chromosomal Disorders ($n=3$: 1p36 Deletion Syndrome, $n=1$, Trisomy 10 with deletion, $n=2$), Muscular Dystrophy ($n=3$), Dravet Syndrome ($n=2$), Lissencephaly ($n=1$), Metabolic Disorder ($n=1$), Kartageners Syndrome with progressive respiratory failure ($n=1$), Pallister Killan Syndrome ($n=1$), brain malformation ($n=1$), and Rett Syndrome ($n=1$). All are included in the directory of LLCs. Diagnosis had been received between one month and 12 years before this study (mean length 5.20 years, $SD=3.18$). Hospice services were accessed by 59% of families. Mean length of contact with hospices was 3.17 years ($SD=2.10$).

Among the 39 siblings, 64% were boys. They had a mean age of 8.23 years ($SD=3.65$, range 3-16 years). Twelve siblings were younger than the child with the LLC, 26 older, and one set were twins. Eighteen (46%) siblings were the same gender as the child with the LLC.

Comparison group of high risk siblings

Siblings of children with ASD were identified from an earlier study (Petalas et al., 2012), and matched to the current group on sibling gender and age; gender and age of the child with the condition (LLC/ASD); position in relation to the child (e.g., older/younger); and same or different gender. Matching was done manually 1:1 following ordering of the ASD database by the variables of interest. Thirty-two sibling pairs were successfully matched. Seven cases from the main study sample were excluded as no reasonable match was found, due to the younger age of LLC siblings.

In the 32 matched ASD group, all primary caregivers were mothers. They were 42 years-old on average ($SD=4.10$, range 29 to 50 years). Sixteen (50%) had a university postgraduate or undergraduate degree. The annual family income was above £35,000 in 44% families, and in 66% mothers were employed outside the home. The children with ASD were 17 boys and 15 girls, aged between 4 and 15 years (mean age 9 years, $SD=2.79$). Diagnoses had been received between 9 months and 8 years of the research taking place (mean time since diagnosis 3.22 years, $SD=2.14$). ASD siblings included 21 boys and 11 girls, with average age of 9 years ($SD=3.08$, range 5 to 17 years). Eleven siblings were younger than the child with ASD, 20 older, and there was one set of twins. Sixteen (50%) siblings were the same gender as the child with ASD.

The matching process resulted in groups being similar with respect to sibling age ($t(31)=1.693$, $p=.100$), gender (male $n=17$, female $n=15$), whether the sibling and child with condition were the same or different genders (same $n=16$, different $n=16$), and whether the sibling was younger or older (older $n=20$, younger $n=11$, twin $n=1$). However there was a significant difference in the age of children with the conditions ($t(31)=3.084$, $p=.004$). Children with a LLC were younger. The matching process was deemed fairly successful as it resulted in no differences between the two groups in five of the six variables used. Further, no differences were present for parental age ($t(31)=1.803$, $p=.081$), parental respondent gender

(31 mothers in LLC, 32 in ASD), and levels of out-of-home employment ($X^2(1)=5.91$, $p=.442$). Groups differed in time since diagnosis ($t(31)=4.075$, $p=.001$), parental education (more university-level parents in the LLC sample; $X^2(1)=4.27$, $p=.039$). Table 1 summarizes the demographics of the samples in the two matched groups.

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Measures

Strengths and Difficulties Questionnaire (SDQ). Emotional and behavioural problems were measured using the parent-report version of the SDQ (Goodman 1997), which includes 25-items about emotional symptoms, conduct problems, hyperactivity, peer relationship problems, and prosocial behaviours. A total behavioral difficulties score (range 0-40) is derived by the first four subscales. A further impact score (range 0-10) indicates the extent to which the difficulties distress the child and interfere with daily living. Parent-report SDQs are available for children aged 3-4 years and 4-16 years. Internal consistency among the LLC group was good. Cronbach's alpha coefficients were as follows: .86 total behavioral difficulties, .79 emotional symptoms, .68 conduct problems, .85 hyperactivity, .78 peer relationship problems, and .69 prosocial behaviour. SDQs were available in the ASD sibling comparison group. In this group, Cronbach's alpha coefficients were .80 for prosocial behaviour, and .88 for total difficulties (Petalas *et al.* 2012).

Sibling Relationship Questionnaire (SRQ). The SRQ brief version (Buhrmester & Furman 1990) is a parent-reported measure of children's relationships (child with LLC and the selected sibling). Thirty-nine items assess warmth/closeness, relative status/power, conflict, and rivalry. Cronbach's alphas among LLC siblings were .91 for warmth/closeness, .84 for relative status/power, .84 for conflict and .75 for rivalry. The SRQ was also available in the ASD group. Cronbach's alpha coefficients in this group were .91 for warmth/closeness, .63 for relative status/power, .87 for conflict and .90 for rivalry (Petalas *et al.* 2012).

Pediatric Quality of Life Questionnaire (PedsQL). The PedsQL (Upton *et al.* 2005) is a 23-item scale measuring physical well-being, emotional functioning, social functioning, and academic functioning among 2-18 year-old children. There is a parent proxy and self-report form for children aged 8+. Here, parents completed the proxy version, and the sibling completed a self-report, if over 8 years, and if /she wished. A total quality of life score (range 0-100) is available, along with domain scores for physical, and psychosocial quality of life. Cronbach's alpha coefficients were .66 and .83 for total quality of life, .84 and .64 for physical quality of life, and .59 and .79 for psychosocial quality of life, for the parent- and self-report versions respectively.

Impact on Family Scale (IoF). The IoF (Stein & Reissman 1980; Stein & Jessop 2003) was developed to measure parental perceptions of the impact of a child's medical condition on the family as a whole. It includes 19 items addressing financial impact, familial-social impact, personal strain, and mastery. A total family impact score (range 0-72) is available. Cronbach's alpha coefficients were .84 for total family impact, .78 for financial impact, .77 for familial-social impact, .80 for personal strain, and .12 for mastery. Mastery was excluded from analysis, due to its poor reliability.

Impact on Sibling Scale (IoS). The IoS (Stein & Jessop 1985) measures parental perception of the effects of a child's medical illness on the unaffected siblings. Six items measure siblings' emotional and behavioral reactions to the illness, parental concerns about the siblings' health, and their own ability to attend to the needs of all their children. It yields a total impact on siblings score (range 0-24). Internal reliability was deemed adequate at (alpha) .72.

Procedure

Ethical approval was gained by the School of Psychology, Bangor University, and the North West Wales NHS Ethics Committee. Families were recruited over 10 months through local

hospice services and UK-wide voluntary organizations for families of children with LLCs.

Research packs were posted to 143 families and 39 were returned (27% return rate).

Participants were eligible for inclusion if the LLC diagnosis of their brother or sister was cited in the dictionary of life-limiting conditions (Hain *et al.* 2013). Families had to have at least one sibling, aged 3-16 years, living in the same household as the child with LLC. The sibling closest in age to the child with the LLC was selected, if there was more than one sibling in a family.

Results

Comparison of LLC siblings to normative data.

Sibling measures in this study were compared to national normative data, where available (Table 2). UK norms were available for the SDQ (Meltzer *et al.*, 2000) and PedsQL (Upton *et al.* 2005). US norms data were available for the IoF and IoS scales (Stein & Jessop 2003). Compared to peers in the general population SDQ norms, siblings of children with LLCs presented significantly higher levels of emotional symptoms, conduct problems, and hyperactivity, with medium effect sizes ($d=.77$, $d=.61$, $d=.51$ respectively). Total behavioral problems and total impact were also significantly higher, with large effect sizes ($d=.81$, $d=1.04$ respectively). Prosocial skills were significantly lower (medium effect size, $d=.76$).

Compared to population norms, siblings' overall and psychosocial quality of life were lower (Table 2), both according to parental reports (small effect sizes ($d=-.34$, $d=-.38$ for total and psychosocial, respectively), and self-reports (medium to large effect sizes: $d=-.58$ and $-.72$ for total and psychosocial self-rated quality of life, respectively). Family impact was significantly higher than available norms with respect to total impact but also financial, and familial-social impact, with moderate effect sizes ($d=.53$, $.42$, $.62$, for total, financial, and familial-social impact, respectively). Personal strain was also significantly higher than

available norms with a large effect size ($d=.83$). Interestingly, impact on siblings was rated at levels similar to available norms.

-----Insert Table 2 here-----

Comparison with ASD Siblings. SDQ and SRQ scores were compared between the two matched groups using paired samples t -tests (Table 3). Siblings of children with LLC displayed significantly higher levels of hyperactivity than siblings of children with ASD (moderate effect size, $d=-.48$), but overall there were no other differences for the SDQ. With regards to sibling relationships, siblings of children with LLC showed significantly lower relative status/power scores than ASD siblings with a large effect size ($d=1.30$), but no other SRQ scores differed between the groups.

-----Insert Table 3 -----

Correlates of Adjustment and Sibling Relationships for the LLC Siblings

We explored potential association between socio-demographic characteristics and all study outcomes among the LLC group of siblings. Only significant associations are reported. There was a higher impact on siblings reported for girls ($t(37)= -2.084, p=.044$). There were significantly higher levels of self-reported physical quality of life (PedsQL) for siblings whose families accessed hospice care ($t(16)=2.426, p=.027$). Siblings whose parent worked outside the home, either full- or part-time, had significantly higher prosocial skills ($t(37)=2.069, p=.046$). Lower family income was associated with higher sibling conflict ($r(39)=-.325, p=.043$), and more financial impact (IoF: $r(39)=-.349, p=.029$). Higher family income was associated with higher parent-rated sibling total quality of life ($r(39)=.349, p=.03$), and parent-rated sibling physical quality of life ($r(39)=.319, p=.048$). A shorter length of time since diagnosis was associated with lower SDQ total impact scores ($r(39)=-.343, p=.032$), less relative status/power in the sibling relationship ($r(39)=.455, p=.004$), and more sibling warmth ($r(39)=-.365, p=.022$). No significant associations were present for sibling age.

Discussion

Siblings of children with LLC present higher levels of emotional and behavioural problems compared to population peers, consistent with previous evidence in similar groups of siblings (Barlow & Ellard 2006; Brennan *et al.* 2013; Read *et al.* 2010; Sharpe & Rossiter 2002). Additionally, almost half of the siblings (49%), exceeded clinical cut-offs (Goodman, 1997), indicating their difficulties were at clinically significant levels. Prosocial skills were significantly lower compared to the general population. This was unexpected as quantitative and qualitative evidence suggests the complex life experiences of these children may equip them with higher levels of empathy and social skills (Alderfer *et al.* 2015; Brennan *et al.* 2013, Malcolm *et al.* 2014). It could, however, indicate the limited opportunities for social interactions in the lives of these children, as also suggested by the high levels of social strain parents reported. Findings highlighted the role of parental out-of-home employment in increasing children's prosocial skills, possibly as children take over a larger role as a carer that involves both direct caregiving but also negotiating care provision with other formal and informal carers.

Quality of life was significantly lower compared to peers in the general population. It is interesting to note the discrepancy between parent and child-reported quality of life: when parents reported on their children, differences with population data were small (effect sizes were between .30 and .40) but when children reported about their own quality of life differences were larger (effect sizes were between .50 and .80). Recent qualitative evidence that siblings tend to protect their parents by not disclosing the full extent of impact (Malcolm *et al.* 2014) could go some way in explaining this difference between parental and self-reports. The parent-/self-difference might also be explained by parents finding it painful to admit putative negative effects on siblings. Additional high levels of parent-reported impact on

family with respect to financial difficulties, altered social relationships, and personal strain highlight the systemic effects of LLCs across family systems.

When considering just how ‘at risk’ siblings of children with LLCs are, the comparison between these siblings and another high risk group, siblings of children with ASD, revealed very similar levels of difficulties, after accounting for (by matching) factors that are associated with increased difficulties, such as children’s gender and age. This would suggest that siblings of children with LLC may be just as ‘at risk’ of negative psychological outcomes as siblings of children with ASD. O’Brien and colleagues’ (2009) review suggested siblings of children with cancer and ASD displayed similarly heightened emotional and behavioral difficulties. In the present study, the matched group design added rigour to this finding.

When we explored socio-demographic correlates of sibling psychological adjustment, to identify correlates of sibling outcomes, few gender or family position associations were found. One gender difference only was found (higher Impact on Siblings scores for females) from 18 possible comparisons, suggesting little evidence for such differences in the current sample. Less time since diagnosis was associated with significantly less total impact, less status/power relationship imbalances, and more sibling relationship warmth, but made no difference to siblings’ behaviour problems, contrary to previous findings of more behavioral difficulties immediately after diagnosis (Barlow & Ellard 2006). Differences in findings are likely due to different approaches to classification. For example, Barlow and Ellard (2006, p.16) defined chronic conditions as “*medically diagnosed ailments with a duration of 6 months or longer which shows little change or slow progression*”. Lower socio-economic status was associated with higher conflict in sibling relationships and lower quality of life, as would have been expected (Read *et al.* 2010). A potentially protective role for hospice-based

services was highlighted by findings of higher physical quality of life (self-rated) for LLC siblings in contact with such services.

To date, interventions suitable for these siblings have not been explored systematically (Lane & Mason 2014). It is suggested that psychoeducation and involving siblings in their brother or sister's treatment might reduce sibling anxiety (e.g., Gursky 2007; Kreicbergs 2010), but the efficacy of these approaches has not been demonstrated. Current findings of similar psychological profile between LLC and ASD siblings suggest we could explore whether evidence-based psychoeducational interventions for ASD siblings (Cooke & Semmens 2010; Knott 2009; Lobato & Kao 2002) might be effective for siblings of children with LLC.

Findings will not generalize to all families of children with LLC as our sample was small and self-selected. Potentially important variables, such as physical or mental health conditions of the sibling and parent were not explored. A strength of the present study is the application of a systematic framework for categorizing LLCs (Hain *et al.* 2013) that allowed us to consider a wider group of children than previous studies, all of whom presented with medical conditions that were life-limiting.

Key points

- Growing up with a brother or sister with a life-limiting condition is likely to impact on siblings' experiences.
- We used a systematic classification of life-limiting conditions to identify children and their families. Compared to peers in the population, siblings experienced higher levels of emotional and behavioural problems and lower quality of life, especially psychosocial.
- Compared to families where the child has non-life-limiting illness, the impact on the family was perceived as greater, albeit not the impact on the siblings.
- Compared to siblings of children with autism, emotional, behavior problems and sibling relationships were at similar levels.
- The findings suggest high levels of need among siblings and families with a child with life-limiting illness, yet the evidence base for appropriate interventions is limited. Siblings' psychological adjustment appears, however, similar to that of another group of high-risk siblings (autism), highlighting the potential for drawing on the autism evidence base for effective sibling intervention approaches.

References

- Alderfer, M.A. Long, K.A. Lown, E.A. Marsland, A.L. Ostrowski, N.L. Hock, J.M. *et al.* (2010). Psychosocial Adjustment of Siblings of Children with Cancer: A Systematic Review. *Psycho-oncology*, **19**, 789-805.
- Alderfer, M. A. Stanley, C. Conroy, R. Long, K. A. Fairclough, D. L. Kazak A. E. *et al.* (2015) The social functioning of siblings of children with cancer: a multi-informant investigation. *Journal of Pediatric Psychology*, **40**, 309-319.
- ACT/RCPCH (1997) A guide to the development of children's palliative care services. Association for Children with Life-Threatening or Terminal Conditions and their Families and Royal College of Paediatrics and Child Health.
http://www.rcpch.ac.uk/sites/default/files/asset_library/Publications/G/Children's_Palliative_Care_Services.pdf Accessed 02/02/16.
- Barlow, J.H. & Ellard, D.R. (2006) The Psychosocial Well-Being of Children with Chronic Disease, Their Parents and Siblings: An Overview of the Research Base. *Child: Care, Health & Development*, **32**, 19-31.
- Brennan, C Hugh-Jones, S. & Aldridge, J. (2013) Paediatric life-limiting conditions: Coping and adjustment in siblings. *Journal of Health Psychology*, **18**, 813-824.
- Breslau, N. Weitzman, M. & Messenger K. (1981) Psychologic Functioning of Siblings of Disabled Children. *Pediatrics*, **67**, 344-353.
- Buhrmester, D. & Furman, W. (1990) Perceptions of Sibling Relationships during Middle Childhood and Adolescence. *Child Development*, **61**, 1387-1389.
- Cooke, J. & Semmens, C. (2010) The Development and Evaluation of a Support Group for Siblings of Children on the Autistic Spectrum. *Good Autism Practice*, **11**, 23-29.
- Department of Health & Children. (2009) *Palliative Care for Children with Life-limiting Conditions in Ireland – a National Policy*. Dublin: Stationary Office.

- Fraser, L. Miller, M. Hain, R. Norman, P. Aldridge, J. McKinney, P. et al. (2012). Rising national prevalence of life-limiting conditions in children in England. *Pediatrics*, **129**, 1-7.
- Goodman, R. (1997) The Strengths and Difficulties Questionnaire: A Research Note. *Journal of Child Psychology and Psychiatry*, **38**, 581–586.
- Gursky, B. (2007) The Effect of Educational Interventions with Siblings of Hospitalized Children. *Journal of Developmental and Behavioral Pediatrics*, **28**, 1-7.
- Hastings, R.P. (2003) Brief Report: Behavioural Adjustment of Siblings of Children with Autism. *Journal of Autism and Developmental Disorders*, **33**, 99-104.
- Houtzager, B.A. Grootenhuis, M.A. Hoeskstra-Weebers, J.E.H.M. Caron, H.N. & Last, B.F. (2003) Psychological functioning in siblings of paediatric cancer patients one to six months after diagnosis. *European Journal of Cancer*, **39**, 1423-1432.
- Knott, F. (2009) Sibshops: Supporting Siblings of Children on the Autistic Spectrum. *Good Autism Practice*, **10**, 18-26.
- Kreicbergs, U. (2010) *The Voice of the Invisible: The Experiences and Consequences of having a Brother or Sister with Cancer during Childhood*. Conference Seminar: 2010 International Society of Pediatric Oncology (SIOP): Boston. Available: http://www.cure4kids.org/ums/home/public_area/c4k_seminar/?ppts_id=2215.
- Lane, C. & Mason, J. (2014). Meeting the needs of siblings of children with life-limiting illnesses. *Nursing children and young people*, **26**, 16-20.
- Lobato, D.J. & Kao, B.T. (2002) Integrated sibling-parent intervention to improve sibling knowledge and adjustment to chronic illness and disability. *Journal of Pediatric Psychology*, **27**, 711-716.

- Malcolm, C. Gibson, F. Adams, S. Anderson, G. & Forbat, L. (2014) A relational understanding of sibling experiences of children with rare-life-limiting conditions: Findings from a qualitative study. *Journal of Child Health Care*, **18**, 230-240.
- Meltzer, H. Gatward, R. Goodman, R. & Ford, F. (2000) *Mental Health of Children and Adolescents in Great Britain*. London: The Stationery Office.
- Nielsen, K.M. Mandleco, B. Roper, S.O.R. Cox, A. Dyches, T. & Marshall, E.S. (2010) Parental perceptions of sibling relationships in families rearing a child with a chronic condition. *Journal of Pediatric Nursing*, **27**, 34-43.
- Noyes, J. Edwards, R. T. Hastings, R. P. Hain, R. Totsika, V. Bennett, V. Hobson, L. Davies, G. R. Humphreys, C. Devins, M. Spencer, L. H. & Lewis, M. (2013) Evidence-based planning and costing palliative care services for children: Novel multi-method epidemiological and economic exemplar. *BMC Palliative Care*, 12-18.
- O'Brien, I. Duffy, A. & Nicholl, H. (2009) Impact of childhood chronic illnesses on siblings: A literature review. *British Journal of Nursing*, **18**, 1358-1365.
- Petalas, M.A. Hastings, R.P. Nash, S. Hall., L.M. Joannidi, H. & Dowey, A. (2012). Psychological Adjustment and Sibling Relationships in Siblings of Children with Autism Spectrum Disorders: Environmental Stressors and the Broad Autism Phenotype. *Research in Autism Spectrum Disorders*, **6**, 546-555.
- Petalas, M.A. Hastings, R.P. Nash, S. Lloyd, T. & Dowey, A. (2009) Emotional and Behavioural Adjustment in Siblings of Children with Intellectual Disability with and without Autism. *Autism*, **13**, 471-483.
- Read, J. Kinali, M. Muntoni, F. & Garralda, M.E. (2010) Psychosocial adjustment in siblings of young people with Duchenne Muscular Dystrophy. *European Journal of Pediatric Neurology*, **14**, 340-348.

- Ross, P. & Cuskelly, M. (2006) Adjustment, sibling problems, and coping strategies of brothers and sisters of children with autistic spectrum disorder. *Journal of Intellectual and Developmental Disabilities*, **31**, 77-86.
- Sharpe, D. & Rossiter, L. (2002) Siblings of children with a chronic illness: A meta-analysis. *Journal of Pediatric Psychology*, **27**, 699-710.
- Sloper, P. (2000) Predictors of distress in parents of children with cancer: A prospective study. *Journal of Pediatric Psychology*, **25**, 79-91.
- Stein, R.E.K., & Jessop, D.J. (1985). *Tables Documenting the Psychometric Properties of a Measure of the Impact of a Chronic Illness on a Family*. NY: Albert Einstein College of Medicine, Department of Pediatrics.
- Stein, R.E.K. & Jessop, D.J. (2003) The Impact on Family Scale revisited: Further psychometric data. *Developmental and Behavioural Pediatrics*, **24**, 9-16.
- Stein, R.E.K. & Reissman, C.K. (1980) The development of an Impact on Family Scale: preliminary findings. *Medical Care*, **18**, 465-472.
- Upton, P. Eiser, C. Cheung, I. Hutchings, H.A. Jenney, M. Maddocks, A. *et al.* (2005) Measurement properties of the UK-English version of the Pediatric Quality of Life Inventory™ (PedsQL™) Generic Core Scales. *Health and Quality of Life*, **3**, 22-29.

Table 1. Demographic profile of the LLC (N=32) and ASD (N=32) matched sibling groups

Demographic variable	LLC group	ASD group
Sibling gender (<i>n</i>)		
Male	21	21
Female	11	11
Sibling age (in years) (<i>mean, SD</i>)	8.88 (3.28)	9.19 (3.08)
Child with condition gender (<i>n</i>)		
Male	17	17
Female	15	15
Child condition age (in years) (<i>mean, SD</i>)	7.78 (3.78)	8.90 (2.79)
Sibling and child sex differences (<i>n</i>)		
Same sex	16	16
Different sex	16	16
Sibling relative position within family		
Older	20	20
Younger	11	11
Twin	1	1
Parent gender (<i>n</i>)		
Male	1	0
Female	31	32
Relationship to child (<i>n</i>)		
Mother	30	32
Father	1	0
Foster parent	1	0

Parent age (in years) (<i>mean, SD</i>)	39.50 (6.22)	42.01 (4.10)
Parent education level (<i>n</i>)		
High school qualifications	8	13
University/college education	24	16
No formal education	0	3
Parent employment status (<i>n</i>)		
Currently employed	18	21
Currently not employed	14	11
Total annual income (<i>n</i>)		
Less than £35,000	14	18
More than £35,000	18	14
Length of time since diagnosis (in years) (<i>mean, SD</i>)	5.78 (3.05)	3.22 (2.14)

Table 2. Comparison of overall LLC sample (n=39) to available normative data

Domain	Normative Mean (SD)	LLC Siblings Mean (SD)	Effect size (d)	t-test comparison
SDQ¹				
Emotional symptoms	1.9 (2.0)	3.44 (2.9)	.77	3.371, p=.002*
Conduct problems	1.6 (1.7)	2.64 (2.1)	.61	3.065, p=.004*
Hyperactivity	3.5 (2.6)	4.82 (3.2)	.51	2.616, p=.013*
Peer problems	1.5 (1.7)	2.18 (2.2)	.40	1.909, p=.064
Prosocial	8.6 (1.6)	7.38 (2.2)	- .76	-3.535, p=.001*
Total difficulties	8.4 (5.8)	13.08 (7.4)	.81	3.955, p<.001*
Total impact	0.4 (1.1)	1.54 (2.3)	1.04	3.071, p=.004*
Parent-rated PedsQL²				
Total score	81.12 (13.9)	76.45 (13.8)	- .34	-2.117, p=.041*
Physical QoL	84.99 (16.1)	83.6 (17.8)	- .09	-.487, p=.629
Psychosocial QoL	79.00 (14.7)	73.46 (17.0)	- .38	-2.035, p=.049*
Child Self-reported PedsQL²				
Total score	82.25 (13.1)	74.60 (12.7)	- .58	-2.557, p=.020*
Physical QoL	86.08 (14.1)	82.58 (12.1)	- .25	-1.229, p=.236
Psychosocial QoL	80.50 (14.1)	70.41 (15.2)	- .72	-2.823, p=.012*
Impact on Family³				
Total impact	46.20 (--)	52.03 (10.1)	.53	3.338, p=.002*
Financial support	7.60 (--)	8.62 (2.5)	.42	2.593, p=.013*
Personal strain	24.40 (--)	29.36 (6.0)	.83	5.152, p<.001*
Familial-social strain	19.90 (--)	22.72 (4.6)	.62	3.870, p<.001*

Impact on Sibling³				
Total sibling impact	13.10 (--)	13.41 (3.6)	.09	.545, p=.589

Available norms: ¹Meltzer et al. (2000), ²Upton et al. (2005), ³Stein & Jessop (2003); Standard deviations on normative data for IoS and IoF not available.

*significant at the .05 level,

QoL = quality of life

Table 3 Comparing behaviour problems (SDQ) and sibling relationships (SRQ) between matched LLC and ASD siblings

Domain	LLC Siblings Mean (SD)	ASD siblings Mean (SD)	Effect size (<i>d</i>)	t-test comparison
SDQ				
Emotional symptoms	3.69 (2.8)	3.47 (3.0)	-.07	.290, p=.774
Conduct problems	2.75 (2.2)	2.09 (1.8)	-.30	1.175, p=.249
Hyperactivity	4.78 (3.1)	3.28 (2.7)	-.48	2.795, p=.025*
Peer problems	2.31 (2.3)	2.19 (2.5)	-.05	.211, p=.834
Prosocial	7.41 (2.3)	8.03 (2.1)	.27	-1.195, p=.241
Total difficulties	13.53 (7.8)	11.03 (7.5)	.32	1.308, p=.201
Total impact	7.41 (2.3)	8.03 (2.1)	.27	-1.195, p=.241
SRQ				
Warmth/ closeness	2.91 (.77)	2.80 (.77)	.13	.515, p=.610
Relative status/ power	-.87 (.97)	.39 (1.6)	1.30	-3.832, p=.001*
Conflict	2.53 (.90)	2.95 (.98)	.47	-1.844, p=.075
Rivalry	.66 (.78)	.68 (.58)	.03	-.117, p=.907