

Psychosocial Functioning in Siblings of Children With Rare Disorders Compared to Controls

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Siblings of children with chronic disorders are at increased risk of psychosocial problems. The risk may be exacerbated when the chronic disorder is rare and limited medical knowledge is available, due to more uncertainty and feelings of isolation. We examined mental health, parent-child communication, child-parent relationship quality, and social support among 100 children aged 8 to 16 years (M age 11.5 years, $SD = 2.2$; 50.0% boys, 50.0% girls). Fifty-six were siblings of children with rare disorders, and 44 were controls. The siblings of children with rare disorders (herein, siblings) were recruited from a resource centre for rare disorders and comprised siblings of children with a range of rare disorders including neuromuscular disorders and rare chromosomal disorders with intellectual disability. Controls were recruited from schools. Self-reported child mental health was significantly poorer for siblings compared to controls (effect size difference $d = 0.75$). Parent-reported child mental health was not significantly different between the groups ($d = -0.06$ to 0.16). Most child-parent relationships (anxiety/avoidance; mothers/fathers) were significantly poorer for siblings compared to controls ($d = 0.47$ to 0.91). There was no difference between groups in anxious relation with mother. Parent-child communication was significantly poorer for siblings compared to controls ($d = -0.87$ to -0.75). Social support was significantly poorer for siblings compared to controls ($d = 0.61$). We conclude that siblings of children with rare disorders display more psychosocial problems than controls. Interventions are indicated to prevent further maladjustment for siblings.

Chronic disorders impact all members of the family, including typically developing siblings (herein, siblings). Growing up with a brother or sister with a chronic disorder is a multifaceted experience involving both positive and negative aspects. Among the challenging experiences, siblings face multiple burdens, such as extra care responsibilities, worries, impaired family communication,

and reduced coping, resilience, and social support [1-3]. Consequently, siblings are at increased risk of psychological difficulties and reduced quality of life [3,4].

Several factors may contribute to the documented increased risk facing siblings. Risk factors may be directly associated with features of the disorder (eg, behavior problems, life expectancy, or degree of intrusiveness in daily

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Keywords: Rare disorders, siblings, parents, psychosocial adjustment

Author Contributions: YBH: Conception and design; Collection of Data; Data Analysis; Writing; Review and Editing. TMV (ORCID iD: 0000-0002-2591-4859): Principal Investigator; Conception and design; Collection of Data; Review and Editing. SM: Conception and design; Collection of Data; Review and Editing. KWF (ORCID iD: 0000-0001-7501-0033): Conception and design; Collection of Data; Data Analysis; Writing; Review and Editing.

life of the family [3]), or indirectly associated with risks facing other family members (eg, parenting stress or maternal depression [2]). Siblings' perceived social support and the quality of family communication have also been suggested as factors associated with sibling outcomes (eg, [5]). The presence of a chronic disorder in a child has been linked to risk of poorer family communication [6], and research has suggested that degree of topic restriction and emotional openness in the family communication is associated with child functioning [7]. Whether the degree of perceived social support and the quality of relation and communication with parents are more associated with child psychosocial functioning when a sibling has a chronic disorder than in families of typically developing children is not known. More research on this matter has been called upon [1]. Further, there is scarce knowledge about whether a chronic disorder in a sibling impact the relationship with and the quality of communication of the mother or father differently. Existing research on parents of children with chronic disorders are disproportionately mother-focused and less is known about the fathers' role in these families (eg, [8,9]) and whether the role of the mothers and fathers differ from families with typically developing children.

Research has also indicated sibling adjustment is impacted by siblings' perceived sense of control and predictability of the family situation, which in turn has been linked to siblings' access of information and understanding of their sibling's disorder [5]. Siblings are indeed found to lack knowledge and/or have misunderstandings about their brother's or sister's diagnosis [10-12]. Although there is a knowledge base documenting risk for siblings, research has not concluded about predictors for sibling psychosocial functioning. More knowledge about factors associated with risk for siblings is needed, as this can help identify subgroups of siblings that may be in particular need of support and that may benefit from intervention programs [2].

One factor that may impact sibling adjustment and disorder knowledge is type of diagnosis of the child with disorder. Most sibling research is conducted with relatively prevalent diagnoses such as pediatric cancer, diabetes, Down syndrome, or autism spectrum disorder. However, the knowledge about siblings' experiences in the case of rare disorders (ie, disorders affecting less than 1 in 2000; [13]) remains very sparse. Several aspects associated with rare disorders may impact on sibling outcomes. First, less is known about the diagnosis and limited information may be available, causing more uncertainty about the condition and its implications for the future [14]. Second, parents of children with rare disorders often report feeling isolated in their experience due to the lack of knowledge about the condition in the health care system [14]. Such feeling of isolation may

also apply to siblings. Furthermore, many rare disorders are complex medical syndromes involving both physical impairments as well as intellectual disabilities, which may be difficult for siblings to grasp due to their abstract and complicated nature [15]. An additional feature of rare disorders is that 80% are of genetic origin [13] and many may imply a risk of carrier status for typically developing siblings. The concept of genetic risk may be particularly difficult for children to comprehend [16,17], and hence, also lead to limited sibling disorder knowledge. Consequently, siblings' understanding of rare disorders may be characterized by more confusion and misconceptions compared to better known disorders, with potential risk of maladjustment [12,18].

Little knowledge exists about the risk for negative sibling outcomes in rare disorders. A few studies have examined psychosocial health of siblings of children with rare disorders specifically, eg, siblings of children with 22q11.2 deletion syndrome [16], Duchenne muscular dystrophy [19,20], Mucopolysaccharidoses or Batten disease [21], spina bifida [22], or William syndrome [23]. A study by Haukeland et al. [24] exploring the emotional experiences of siblings of children with different rare disorders showed that these siblings described a range of complex and contradictory positive and negative emotions. Knowledge about the rare disorder appeared to be of high importance to the siblings and thinking about the prognosis and future health condition of their brother or sister seemed to be a particular source of emotional distress [24]. Other studies have included rare disorders in larger samples of siblings of children with more well-known diagnoses (eg, [25,26]), but few studies have focused specifically on rare disorders.

Furthermore, the studies that have examined the psychological impact on siblings of children with rare disorders, have mostly been qualitative. These studies have provided important insights, but results are not necessarily generalizable across various rare disorders. Furthermore, to our knowledge, no study has included comparisons with controls. Controls provide important contextual information for the level of risk. Another limitation in the field of sibling research is the predominant reliance on reports from parents. Due to the generally limited congruence between parent and sibling reports [27,28], the inclusion of sibling self-report is crucial when studying sibling adjustment and mental health (eg, [29,30]), also in the case of rare disorders.

In the present study, our aim was to investigate how having a brother or sister with a rare disorder impacts siblings' psychosocial functioning. We examined sibling mental health, parent-child communication, quality of the child-parent relationship, and social support perceived by the siblings across a broad range of rare disorders. To contextualize our findings, we included a broad range of

rare disorders, sibling self-report, and a control group. We investigated three research questions: 1) Is mental health, quality of parent-child communication and relation, and social support different for siblings of children with rare disorders compared to controls? Based on previous studies (eg, [3,4]), we expected these variables would be poorer for siblings. 2) Is there a difference in child relation and communication with mothers and fathers, and are such differences larger in the sibling sample compared to controls? 3) Is there larger overlap (ie, correlations) between mental health, parent-child communication, child-parent relation, and social support for siblings of children with rare disorders compared to controls? Based on limited previous findings, we explored 2) and 3) openly.

METHODS

Participants and Procedure

Data was drawn from a larger study of the intervention “SIBS” (short for SIBlingS) for siblings of children with chronic disorders and their parents (see [31,32]). Families of children with rare disorders were recruited for participation in the SIBS study through a national resource center for rare disorders. Families were invited to participate in the intervention through an information letter if the child with disorder had one or more typically developing sibling aged 8 to 16 years. Children gave their verbal assent and parents provided written consent. The study was approved by the Regional Committee for Medical and Health Research Ethics – South Eastern Norway. Both parents and siblings completed questionnaires about siblings’ mental health, and siblings completed questionnaires about parent-child communication, parent-child relationship, and social support prior to participating in the intervention. The control group was recruited from two local elementary schools through parental meetings, and parents and children from the control group completed the same measures.

The sample of families accepted to the study comprised a total of 100 children aged 8 to 16 years and their parents. Fifty-six were siblings of children with rare disorders (M age = 11.3 years, SD = 1.7; 51.8% girls; 48.2% boys; herein: sibling group) drawn from the SIBS intervention study sample (see [32]), and 44 were controls (M age = 11.4 years, SD = 2.5, 38.6% girls; 61.4% boys). The age difference between the samples was not significant (p = .757). Although the percentage of boys was higher among controls, the gender frequency distribution was not significantly different between the samples (χ^2 = 1.715, p = .190).

Mean parental age was significantly lower in the sibling sample (mothers M age = 40.3, SD = 4.8; fathers M age = 42.6 years, SD = 5.3) than among controls (mothers M age = 44.6, SD = 4.9; fathers M age = 47.2 years, SD

Table 1. Overview of Diagnoses of the Children with Rare Disorders in the Sibling Sample

Diagnosis	$N = 56$
Duchenne muscular dystrophy	9
Velocardiofacial syndrome	8
Smith Magenis syndrome	5
Congenital muscular dystrophy	4
Fragile X syndrome	4
Mucopolysaccharidosis type IV (Morquios syndrome)	3
Severe progressive disorder in central nervous system	3
Spinal muscular atrophy	3
Angelman syndrome	2
Hereditary ataxias	2
Humoral immune deficiency	2
Noonan syndrome	2
Becker muscular dystrophy	1
Bethlem myopathy	1
Chromosome 5q deletion syndrome	1
Neurodegenerative disease	1
Neurofibromas type 1	1
Osteogenesis imperfecta	1
Prader-Willi syndrome	1
Rett syndrome	1
47,XXY syndrome (Klinefelter syndrome)	1

= 6.4). In terms of socio-economic status, parents in the sibling sample reported significantly lower family financial status than controls (p > .001, effect size difference d = 1.03). Note that this was a subjective scale and not reported income.

The children with rare disorders represented a range of rare diagnoses including neuromuscular disorders (eg, Becker muscular dystrophy, Duchenne muscular dystrophy, Spinal muscular atrophy) and rare chromosomal disorders with intellectual disability (eg, Angelman syndrome, Fragile X syndrome, Smith-Magenis syndrome, Velocardiofacial syndrome). See Table 1 for an overview of all the rare disorders.

Measures

To assess mental health, we used the Strengths and Difficulties Questionnaire (SDQ; [33]). Children and parents rated the child’s emotional, conduct, attention, and peer problems on a 3-point scale from 0 (not true) to 2 (certainly true) (eg, “I worry a lot”) on the 25-item total scale. Sound psychometric properties have been reported for the SDQ [34-36]. In the present study, internal consis-

tency was satisfactory for the total scale for both siblings (α child = .78, α mothers = .87, and α fathers = .76) and controls (α child = .78, α mothers = .73, and α fathers = .72).

We used the child version of the “parent communication” subscale of the Parent-Child Communication Scale (PCCSc; [37]) to measure child-perceived quality of parent-child communication. The children rated openness and problems in parent-child communication on a 5-point scale from 1 (almost never) to 5 (almost always) (eg, “Does your mother try to understand what is on your mind?”) for both their parents. Satisfactory reliability has been reported for the PCCSc ($\alpha=0.75$; [38,39]), and in the present study we found satisfactory internal consistency for the parent communication subscale in both siblings (α mothers = .77; α fathers = .82) and controls (α mothers = .79, α fathers = .71).

To measure quality of parent-child relationship we used an adapted version of the Experiences in Close Relationships-Revised (ECR-R) questionnaire [40]. ECR-R is a 9-item self-report instrument consisting of two subscales, ie, *Avoidance* and *Anxiety*, designed to assess attachment patterns in a variety of close relationships [40]. The children rated their relationship with their mother and father respectively on a 7-point scale from 1 (correct) to 7 (incorrect) (eg, “It is easy for me to trust my mother/father”) [40]. Satisfactory reliability has been reported for the ECR-R ($\alpha = .88$ to $.92$) [41]. In the present study, internal consistencies for the two dimensions of the ECR-R were satisfactory in both siblings ($\alpha = .75$ to $.82$) and controls ($\alpha = .72$ to $.81$).

Social support was measured with a child-adapted version of the Functional Social Support Questionnaire (FSSQ) [42], consisting of 7 items measuring affective and confidant support (eg, “I have a lot of people around me that care about what happens to me”) rated on a 5-point scale from 1 (as much as I need) to 5 (much less than I need). The original FSSQ has shown average item-remainder correlations of $r = 0.63$ [42], and internal consistency for the child-adapted version in the present study was satisfactory in both siblings ($\alpha = .82$) and controls ($\alpha = .83$).

Data Analytic Plan

Group means were compared with independent and paired sample t-tests. Effect sizes were calculated using the formula $(SD_1 - SD_2) / SD_{pooled}$, and interpreted as small $>.20$, medium $>.50$, or large $>.80$ [43]. The correlations between variables in the two samples were compared by transforming the respective r -correlations to z -scores, and comparing them using the formula $Z_{observed} = (z_1 - z_2) / (\sqrt{[(1 / N_1 - 3) + (1 / N_2 - 3)]})$. IBM SPSS version 27 [44] was used for all analyses. Given that both samples were non-clinical and the total sample size was $n = 100$, we de-

cidated based on a skewness range from -0.65 to 2.23 and a kurtosis range from -0.25 to 6.58 that the variables were within reasonable normal distribution. There was very little missing data (range 0.7% to 6.6% across variables with a mean of 4.2%). The main reason for missing data was no data on either mothers or fathers, which mainly applied to participants from single-parent families (18.1% of the sample did not live with both parents). Thus, the level of missing data was not considered problematic.

RESULTS

First, we compared siblings of children with rare disorders to controls on mental health, parent-child communication, child-parent relation, and social support. Except for one variable, siblings had significantly poorer scores on all self-reported variables, with medium to large effect sizes. The exception was anxiety in relation to mother, which was not significantly different between the samples. There was no significant difference between the samples on mother- and father-reported child mental health. See Table 2.

Second, we examined differences between child-, father-, and mother-report in the respective samples. In the sibling sample, both mothers and fathers reported more child mental health problems than children ($p < .001$, d mothers = 0.67; d fathers = 0.80), with no difference between parents ($p = .738$, $d = 0.05$). Children reported less avoidance in relation with mothers than fathers ($p = .034$, $d = 0.30$). There was no difference between parents for anxiety in relation ($p = .603$, $d = 0.07$) or communication ($p = .102$, $d = 0.24$).

In the control sample, both mothers and fathers reported more child mental health problems than children ($p < .002$, d mothers = 0.49; d fathers = 0.64), with no difference between parents ($p = .456$, $d = 0.11$). Children reported less avoidance in relation with mothers than fathers ($p = .001$, $d = 0.50$). There was no difference between parents for anxiety in relation ($p = .268$, $d = 0.17$). Children reported better communication with mothers than fathers ($p = .009$, $d = 0.41$).

Third, we examined correlations between the variables within each sample. See Table 3. In both samples, there were several significant correlations between the main variable groups. The correlations between child-rated mental health and communication with parents were significant in the sibling sample, but not in the control sample. However, there was only one significant difference in correlation size between the samples. This was the correlation between anxiety in relation to father and social support, which was significantly higher in the sibling sample ($z = 2.327$, $p = 0.01$).

Table 2. Comparison of Psychosocial Variables Between Siblings of Children with Rare Disorders and Controls

	Siblings	Controls	Scale range	Group comparison			
	M (SD)	M (SD)		<i>t</i>	<i>p</i>	<i>d</i>	95% CI for <i>d</i>
Child-rated mental health	9.7 (5.2)	6.0 (4.6)	0-40	3.69	<.001	0.75	0.34 to 1.16
Mother-rated mental health	13.9 (5.8)	13.0 (4.5)	0-40	0.79	.433	0.16	-0.24 to 0.55
Father-rated mental health	13.6 (6.2)	13.9 (5.5)	0-40	-0.27	.786	-0.06	-0.45 to 0.34
Communication with mother	3.6 (1.0)	4.4 (0.7)	1-5	-4.25	<.000	-0.87	-1.28 to -0.47
Communication with father	3.7 (0.7)	4.1 (0.6)	1-5	-3.65	<.000	-0.75	-1.17 to -0.33
Relation to mother – anxiety	1.8 (1.4)	1.4 (1.0)	1-7 ^r	1.40	.164	0.28	-0.12 to 0.69
Relation to mother – avoidance	2.5 (1.1)	1.8 (0.7)	1-7 ^r	3.46	.001	0.70	0.21 to 1.11
Relation to father – anxiety	1.7 (1.4)	1.2 (0.7)	1-7 ^r	2.30	.024	0.47	0.07 to 0.88
Relation to father – avoidance	2.9 (1.3)	1.9 (0.8)	1-7 ^r	4.46	<.001	0.91	0.41 to 1.33
Social support	1.6 (0.7)	1.3 (0.3)	1-5 ^r	2.98	.004	0.61	0.20 to 1.01

Note. M = mean. SD = standard deviation. CI = confidence interval. ^r= lower scores indicate less avoidance/anxiety and higher levels of social support.

Table 3. Correlations Between Variables in the Sibling Sample (above Diagonal) and Controls (below Diagonal)

	1.	2.	3.	4.	5.	6.	7.	8.	9.	10.
1. Child-rated mental health	-	.37**	.66**	.36**	.34*	.37**	.47**	-.32*	-.31*	.47**
2. Mother-rated mental health	.31*	-	.59**	.40**	.03	.39**	.13	.08	.11	.20
3. Father-rated mental health	.57**	.66**	-	.30*	.12	.39**	.34*	-.08	-.11	.34*
4. Relation to mother – anxiety	.47**	.16	.07	-	.24	.77**	.27	-.16	-.27	.43**
5. Relation to mother – avoidance	.35*	.18	.32*	.30*	-	.20	.59**	-.52**	-.38**	.51**
6. Relation to father – anxiety	.47**	.11	.17	.80**	.31*	-	.27	-.11	-.30*	.58**
7. Relation to father – avoidance	.37*	.33*	.35*	.28	.75**	.40**	-	-.56**	-.73**	.41**
8. Communication with mother	-.21	.02	-.06	-.12	-.64**	-.27	-.61**	-	.75**	-.40**
9. Communication with father	-.12	-.30*	-.15	-.02	-.61**	-.14	-.78**	.66**	-	-.41**
10. Social support	.37**	.25	.27	.29	.54**	.18	.56**	-.42**	-.33*	-

Note. Correlation in the siblings of children with rare disorders sample in bold above the diagonal. Correlations in the control sample below the diagonal. *correlation is significant at the *p* < .05-level. **correlation is significant at the *p* < .001-level.

DISCUSSION

We examined psychosocial functioning in siblings of children with rare disorders compared to controls. In line with our expectation, siblings had poorer scores on several variables compared to controls. This applied both to mental health, parent-child communication, child-parent relationship quality, and social support. Effect size differences were medium to large, indicating substantial risk of poorer psychosocial functioning for siblings. These findings are in line with research on siblings of children with chronic disorders that are not rare (eg, autism spectrum disorder, diabetes, Down syndrome [3-6]), and imply that

siblings of children with rare disorders may need intervention.

We also examined differences in reports between siblings, mothers, and fathers. In both samples, parents rated more mental health problems for their children than the children self-reported. However, the effect size differences were larger in the sibling sample, indicating more inter-generational discrepancy. Although informant discrepancies are common in child mental health [45], the magnitude of the difference in the sibling sample is important to note. This is because these differences have an impact on siblings’ access to care and services. Because parents usually are initiators of services for chil-

dren, larger discrepancies between children and parents mean not all siblings who experience difficulties may access interventions. Furthermore, sibling motivation for services may be impacted by the fact that their parents believe they have larger mental health problems than what they self-report.

We found that both siblings and controls reported less avoidance with mothers than fathers. Stereotypically, mothers and fathers take on different roles and mothers tend to typically be responsible for more of the “holding on” aspect of attachment, whereas fathers are more responsible for “letting go” [46]. As such, the findings in both samples are in accord with typical gender difference findings. However, it is worth noting that whereas children in the control sample also perceived better communication with mothers, there was no difference between parents in the sibling sample. This could indicate that fathers may be particularly good communication partners for siblings in families of children with rare disorders, in which mothers typically take on a larger caretaking role for the child with disorder (eg, [47,48]). Our findings may indicate that fathers are good at compensating for such extra maternal care burdens in families of children with rare disorders and that fathers may play important roles in family and sibling interventions.

Finally, we found that the overlap between mental health, quality of parent-child relations and parent communication, and social support was comparable for siblings of children with rare disorders and controls. There was one exception to this pattern, which was a much larger correlation between social support and anxiety in relation to fathers in the sibling sample compared to controls. This finding indicates that for siblings, the less anxiety they reported in their relation to the father, the more socially supported they felt. This may, again, reflect a particular role for fathers as a source of support for siblings in families of children with rare disorders, as indicated by the finding of no difference in communication quality between mothers and fathers in the sibling sample. Children and youth in general are found to disclose more to mothers than fathers about their experiences [49,50], reflecting that mothers may play a role as the most frequent provider of support for children [51]. However, mothers in families of children with disabilities typically take the major share in the caretaking of the child with a disorder (eg, [52]). Thus, siblings might perceive the mother as less available and hence, become relatively more reliant on the father for support than in families of typically developing children. Consequently, the quality of the child-parent relation to the father may be of particular importance for siblings’ sense of perceived support. More research is needed to understand the role of the father and the father’s provision of support for the psychosocial functioning of siblings.

The strengths of the current study include a relatively large sample of siblings of children with rare disorders, and the inclusion of a control group. However, there are also limitations. Various rare disorders are represented in the sample, and variations based on type (eg, primarily intellectual versus primarily somatic disorders) and degree of rarity may exist that we did not examine. Further, we did not include a control group with non-rare disorders. A major limitation is that parent-perceived financial situation was poorer in the sibling group. High socio-economic status is a common representativeness problem with control groups, and this aspect may have exacerbated differences between the samples. We cannot know the magnitude of the difference if we had asked parents to report actual income rather than their subjective experience of family economy. However, the relatively high socio-economic status of controls raises questions about the representativeness of the control sample and thus the generalizability of findings.

The main implication of the current study is that siblings of children with rare disorders may need interventions. Our results suggest that such interventions should target their mental health, communication and relation with parents, and their social support, which are all interconnected and poorer than among controls. To date, no evidence-based interventions exist for siblings across disorders [2]. Identifying such interventions represent an important step for the field towards enhancing psychosocial functioning of children who are next-of-kin.

Acknowledgments: We would like to thank all the participating families in this study.

REFERENCES

1. Kelada L, Wakefield CE, Drew D, Ooi CY, Palmer EE, Bye A et al. Siblings of young people with chronic illness: Caring responsibilities and psychosocial functioning. *J Child Health Care*. 2021 (OnlineFirst)
2. Tudor ME, Lerner MD. Intervention and support for siblings of youth with developmental disabilities: a systematic review. *Clin Child Fam Psychol Rev*. 2015 Mar;18(1):1–23.
3. Vermaes IP, van Susante AM, van Bakel HJ. Psychological functioning of siblings in families of children with chronic health conditions: a meta-analysis. *J Pediatr Psychol*. 2012 Mar;37(2):166–84.
4. McKenzie Smith M, Pinto Pereira S, Chan L, Rose C, Shafraan R. Impact of wellbeing interventions for siblings of children and young people with a chronic physical or mental health condition: A systematic review and meta-analysis. *Clin Child Fam Psychol Rev*. 2018 Jun;21(2):246–65.
5. Inledon E, Williams L, Hazell T, Heard TR, Flowers A, Hiscock H. A review of factors associated with mental health in siblings of children with chronic illness. *J Child Health Care*. 2015 Jun;19(2):182–94.
6. Murphy LK, Murray CB, Compas BE; Guest Editors: Cynthia A. Gerhardt, Cynthia A. Berg, Deborah J. Wiebe and

- Grayson N, Holmbeck. Topical review: integrating findings on direct observation of family communication in studies comparing pediatric chronic illness and typically developing samples. *J Pediatr Psychol*. 2017 Jan;42(1):85–94.
7. Huizinga GA, Visser A, van der Graaf WT, Hoekstra HJ, Hoekstra-Weebers JE. The quality of communication between parents and adolescent children in the case of parental cancer. *Ann Oncol*. 2005 Dec;16(12):1956–61.
 8. Bogossian A, King G, Lach LM, Currie M, Nicholas D, McNeill T, et al. (Unpacking) father involvement in the context of childhood neurodisability research: a scoping review. *Disabil Rehabil*. 2019 Jan;41(1):110–24.
 9. Dunn K, Kinnear D, Jahoda A, McConnachie A. Mental health and well-being of fathers of children with intellectual disabilities: systematic review and meta-analysis. *BJPsych Open*. 2019 Nov;5(6):e96.
 10. Lobato DJ, Kao BT. Brief report: family-based group intervention for young siblings of children with chronic illness and developmental disability. *J Pediatr Psychol*. 2005 Dec;30(8):678–82.
 11. Rana P, Mishra D. Quality of life of unaffected siblings of children with chronic neurological disorders. *Indian J Pediatr*. 2015 Jun;82(6):545–8.
 12. Vatne TM, Helmen IØ, Bahr D, Kanavin Ø, Nyhus L. “She came out of mum’s tummy the wrong way”. (Mis) conceptions among siblings of children with rare disorders. *J Genet Couns*. 2015 Apr;24(2):247–58.
 13. EURORDIS. Rare Diseases Europe [Internet]. Cited 2021 May 31. Available from: <https://www.eurordis.org/content/what-rare-disease>
 14. Grut L, Kvam MH. Facing ignorance: people with rare disorders and their experiences with public health and welfare services. *Scand J Disabil Res*. 2012;15(1):20–32.
 15. Glasberg BA. The development of siblings’ understanding of autism spectrum disorders. *J Autism Dev Disord*. 2000 Apr;30(2):143–56.
 16. Okashah R, Schoch K, Hooper SR, Shashi V, Callanan N. Parental communication and experiences and knowledge of adolescent siblings of children with 22q11.2 deletion syndrome. *J Genet Couns*. 2015 Oct;24(5):752–9.
 17. Metcalfe A, Plumridge G, Coad J, Shanks A, Gill P. Parents’ and children’s communication about genetic risk: a qualitative study, learning from families’ experiences. *Eur J Hum Genet*. 2011 Jun;19(6):640–6.
 18. Metcalfe A, Coad J, Plumridge GM, Gill P, Farndon P. Family communication between children and their parents about inherited genetic conditions: a meta-synthesis of the research. *Eur J Hum Genet*. 2008 Oct;16(10):1193–200.
 19. Read J, Kinali M, Muntoni F, Garralda ME. Psychosocial adjustment in siblings of young people with Duchenne muscular dystrophy. *Eur J Paediatr Neurol*. 2010 Jul;14(4):340–8.
 20. Read J, Kinali M, Muntoni F, Weaver T, Garralda ME. Siblings of young people with Duchenne muscular dystrophy—a qualitative study of impact and coping. *Eur J Paediatr Neurol*. 2011 Jan;15(1):21–8.
 21. Malcolm C, Gibson F, Adams S, Anderson G, Forbat L. A relational understanding of sibling experiences of children with rare life-limiting conditions: findings from a qualitative study. *J Child Health Care*. 2014 Sep;18(3):230–40.
 22. Holmbeck GN, Devine KA. Psychosocial and family functioning in spina bifida. *Dev Disabil Res Rev*. 2010;16(1):40–6.
 23. Cebula K, Gillooly A, Coulthard LK, Riby DM, Hastings RP. Siblings of children with Williams syndrome: correlates of psychosocial adjustment and sibling relationship quality. *Res Dev Disabil*. 2019 Nov;94:103496.
 24. Haukeland YB, Fjermestad KW, Mossige S, Vatne TM. Emotional experiences among siblings of children with rare disorders. *J Pediatr Psychol*. 2015 Aug;40(7):712–20.
 25. Fullerton JM, Totsika V, Hain R, Hastings RP. Siblings of children with life-limiting conditions: psychological adjustment and sibling relationships. *Child Care Health Dev*. 2017 May;43(3):393–400.
 26. O’Neill LP, Murray LE. Anxiety and depression symptomatology in adult siblings of individuals with different developmental disability diagnoses. *Res Dev Disabil*. 2016 Apr-May;51-52:116–25.
 27. Guite J, Lobato D, Kao B, Plante W. Discordance between sibling and parent reports of the impact of chronic illness and disability on siblings. *Child Health Care*. 2004;33(1):77–92.
 28. Sharpe D, Rossiter L. Siblings of children with a chronic illness: a meta-analysis. *J Pediatr Psychol*. 2002 Dec;27(8):699–710.
 29. Eiser C, Morse R. A review of measures of quality of life for children with chronic illness. *Arch Dis Child*. 2001 Mar;84(3):205–11.
 30. Houtzager BA, Grootenhuis MA, Last BF. Supportive groups for siblings of pediatric oncology patients: impact on anxiety. *Psychooncology*. 2001 Jul-Aug;10(4):315–24.
 31. Vatne TM, Haukeland YB, Mossige S, Fjermestad KW. The development of a joint parent-child intervention for siblings of children with chronic disorders. *Fokus på Familien*. 2012;1:20–35.
 32. Haukeland YB, Czajkowski NO, Fjermestad KW, Silverman WK, Vatne TM. Evaluation of “SIBS”: an intervention for siblings and parents of children with chronic disorders. *J Child Fam Stud*. 2020;29(8):2201–17.
 33. Goodman R, Ford T, Richards H, Gatward R, Meltzer H. The Development and Well-Being Assessment: description and initial validation of an integrated assessment of child and adolescent psychopathology. *J Child Psychol Psychiatry*. 2000 Jul;41(5):645–55.
 34. Goodman R. Psychometric properties of the strengths and difficulties questionnaire. *J Am Acad Child Adolesc Psychiatry*. 2001 Nov;40(11):1337–45.
 35. Goodman R, Scott S. Comparing the Strengths and Difficulties Questionnaire and the Child Behavior Checklist: is small beautiful? *J Abnorm Child Psychol*. 1999 Feb;27(1):17–24.
 36. Vostanis P. Strengths and Difficulties Questionnaire: research and clinical applications. *Curr Opin Psychiatry*. 2006 Jul;19(4):367–72.
 37. Conduct Problems Prevention Research Group. 1994 Conduct Problems Prevention Research Group (CPPRG). Cited 2021 May 31. Available from: <http://fasttrackproject.org/techrept/p/pcp/index.php>
 38. Pek JC. Parent-Child Communication, Child Report (Fast Track Project Technical Report). 2006. Available from:

- <http://www.fastrackproject.org>
39. McCarty CM, Doyle SR. Parent-child communication (child) (Fast Track Project Technical Report). 2001. Available from: <http://www.fastrackproject.org>
 40. Fraley RC, Waller NG, Brennan KA. An item response theory analysis of self-report measures of adult attachment. *J Pers Soc Psychol*. 2000 Feb;78(2):350–65.
 41. Fraley RC, Heffernan ME, Vicary AM, Brumbaugh CC. The Experiences in Close Relationships-Relationship Structures questionnaire: a method for assessing attachment orientations across relationships. *Psychol Assess*. 2011 Sep;23(3):615–25.
 42. Broadhead WE, Gehlbach SH, de Gruy FV, Kaplan BH. The Duke-UNC Functional Social Support Questionnaire. Measurement of social support in family medicine patients. *Med Care*. 1988 Jul;26(7):709–23.
 43. Cohen J. *Statistical power analysis for the behavioral sciences*. 2nd ed. Hillsdale (NJ): Lawrence Erlbaum Associates; 1988.
 44. IBM Corp. Released 2020. IBM SPSS Statistics for Windows, Version 27.0. Armonk, NY: IBM Corp.
 45. De Los Reyes A, Kazdin AE. Informant discrepancies in the assessment of childhood psychopathology: a critical review, theoretical framework, and recommendations for further study. *Psychol Bull*. 2005 Jul;131(4):483–509.
 46. Bögels S, Phares V. Fathers' role in the etiology, prevention and treatment of child anxiety: a review and new model. *Clin Psychol Rev*. 2008 Apr;28(4):539–58.
 47. Verma A, Srivastava P, Kumar P. Stress among parents having children with mental retardation: a gender perspective. *J Disabil Management Rehabil*. 2017;2(2):68–72.
 48. Pandey D, Dubey P. Mediating effect of social support on stress among parents of children with intellectual disability. *Indian J Public Health Res Dev*. 2019;10(2):153–69.
 49. Yau JP, Tasopoulos-Chan M, Smetana JG. Disclosure to parents about everyday activities among american adolescents from mexican, chinese, and European backgrounds. *Child Dev*. 2009 Sep-Oct;80(5):1481–98.
 50. Soenens B, Vansteenkiste M, Luyckx K, Goossens L. Parenting and adolescent problem behavior: an integrated model with adolescent self-disclosure and perceived parental knowledge as intervening variables. *Dev Psychol*. 2006 Mar;42(2):305–18.
 51. Furman W, Buhrmester D. Age and sex differences in perceptions of networks of personal relationships. *Child Dev*. 1992 Feb;63(1):103–15.
 52. Rowbotham M, Carroll A, Cuskelly M. Mothers' and fathers' roles in caring for an adult child with an intellectual disability. *Int J Disabil Dev Educ*. 2011;58(3):223–40.