



Health-Related Quality of Life Among Siblings of Children with Special Needs and Typically Developing Children in Japan: A Cross-Sectional Study

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博 士 論 文

Health-Related Quality of Life Among Siblings of Children with Special Needs and Typically
Developing Children in Japan: A Cross-Sectional Study

(日本における特別支援を要する子どもと定型発達児のきょうだいの

健康関連 QOL：横断的研究)

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Title

Health-Related Quality of Life Among Siblings of Children with Special Needs and Typically Developing Children in Japan: A Cross-Sectional Study

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Health-Related Quality of Life Among Siblings of Children with Special Needs and Typically Developing Children in Japan: A Cross-Sectional Study

Abstract

With the global rise in children with disabilities, their siblings may experience various impacts on their daily lives. This study investigates health-related quality of life (QOL) and related factors among siblings of students attending special-needs schools in Japan. A cross-sectional study was conducted with two groups: 71 siblings of children with disabilities and a control group of 398 siblings of typically developing children, using the KINDL^R QOL scale. Results showed a median QOL score for siblings of children with disabilities of 72.92 versus 71.88 for siblings of typically developing children ($p = 0.711$). This lack of significant difference may be attributed to existing social support systems and special-needs schools in Japan, which may reduce the burden on siblings of children with disabilities. These findings highlight the importance of maintaining and improving support systems for siblings of children with disabilities while continuously monitoring their well-being at home and in their communities.

Keywords: children with special needs, siblings, quality of life, cross-sectional design, Japan

Health-Related Quality of Life Among Siblings of Children with Special Needs and Typically Developing Children in Japan: A Cross-Sectional Study

Recent years have seen an increase in the number of children and adolescents with disabilities and diseases, driven by decreasing premature childhood deaths and population growth (GBD 2017 Child and Adolescent Health Collaborator, 2019). Globally, the population of children and adolescents suffering from epilepsy, intellectual disabilities, or sensory disabilities reached 291.2 million in 2017, accounting for 11.2% of the total population of that age group (Olusanya et al., 2020). Children with disabilities are considered a vulnerable population with an increasing risk for new or worsened health disparities across their lifespan. Therefore, addressing these disparities and promoting health equity requires not only the involvement of medical professionals but also society at large (Houtrow et al., 2023). In response, corresponding laws and specialized services have been developed worldwide to ensure the rights and safety of children with disabilities and their families (McTavish et al., 2022; Naz & Akhtar, 2020). Particular emphasis has been placed on empowering children with disabilities and their families to have equitable access and participation within the social community from an early age (Olusanya et al., 2024).

Existing laws related to disability and child welfare in Japan have been revised in recent years, and in 2021, a law providing support for children receiving medical care and their families was enacted. These legislations have facilitated the expansion of essential services for these children and their families, such as modifications to daycare support service systems, enhanced training from specialists for daycare centers and educational institutions, strengthened

consultation support and collaboration among childcare centers, schools, and related organizations; and increased assignment of nurses to schools and daycare centers (Matsui, 2022; Ohtoshi, 2016). Another important issue is the provision of appropriate education for children with diverse disabilities. Although Japan's declining birthrate has led to a decrease in the number of children in compulsory education, the number of children with disabilities requiring special education support is rising (Teruya et al., 2019). Japan offers several compulsory education pathways for children with disabilities, including special-needs schools and classrooms, regular classrooms, and *tsukyu* classrooms, which are special programs for children with disabilities in regular schools (Isogai, 2017). In particular, special-needs schools provide education to students with various disabilities, such as hearing and visual impairments, intellectual disabilities, and physical disabilities, but they also serve as resource centers that provide advice and support for children enrolled in local kindergartens, elementary schools, junior high schools, and high schools (Isogai, 2017; Ohtoshi, 2016).

However, while support for children with disabilities and their families is being expanded in Japan and globally, disability remains a low priority in the broader child and adolescent health agenda, with services reported as woefully inadequate (Cieza et al., 2021). Of particular concern are disparities in healthcare service access and quality, which depend on disability type or illness (Cheak-Zamora & Thullen, 2017). The same is seen in the field of education; research has noted the physical and social barriers children with disabilities face in participating in school activities and social interactions with peers outside the classroom (Coster et al., 2013). As a result, intense caregiving demands related to raising children with disabilities decrease family time and socialization while also exposing siblings to parental stress, anxiety, and irritability (Mulroy et al., 2008).

Siblings of children with disabilities are greatly affected in various ways both inside and outside their homes, including their behavior at school, completing school work, and participating in leisure or sports activities (Goudie et al., 2013). For example, previous research on children with intellectual disabilities and their siblings has reported that siblings of children with intellectual disabilities tire of accompanying siblings to medical appointments and face restrictions that may prevent them from playing with their friends. In addition, the needs of siblings with disabilities and the demands of managing specific situations compel siblings to have patience and self-control (Correia & Seabra-Santos, 2022; Múries-Cantán, Giné, et al., 2023). Therefore, they may not be able to concentrate on their studies or get enough sleep compared to children whose siblings do not have disabilities (Rana & Mishra, 2015). As a result, these children have feelings of depression, anxiety, guilt, and self-blame toward their siblings with disabilities and hence require adequate support (Çelik et al., 2018; Hanvey et al., 2022).

On the other hand, previous research has also reported positive experiences, such as the joy of looking after their siblings with disabilities in their daily lives or playing and participating in sports together (Múries-Cantán, Giné, et al., 2023). These mixed findings are also reflected in previous QOL surveys of siblings. For example, some studies have reported that siblings of children with disabilities or diseases show lower QOL compared to siblings of children without (Dinleyici et al., 2019; Koukouriki & Soulis, 2020), while others have shown that the QOL of siblings of children with disabilities is not significantly lower (Buchbinder et al., 2019; Wakimizu et al., 2020). In other words, previous research results regarding the QOL of siblings of children with disabilities or illnesses are inconsistent.

Additionally, Lamsal and Ungar (2021) show that there is a paucity of research on the QOL of siblings of children with neurodevelopmental disorders, making it difficult to draw

definitive conclusions about the impact on siblings' QOL. Thus, further research is needed to address these gaps (Lamsal & Ungar, 2021; Marquis et al., 2019; Rana & Mishra, 2015). Moreover, many factors other than having a brother or sister with a disability affect the QOL of children in general. Previous studies have found individual factors such as breakfast, sleep, and extracurricular activities are related to children's QOL (Lundqvist et al., 2019; Taylor et al., 2023; Ricci et al., 2020). Given the insufficient research on this topic, it is unclear if and how these factors affect the QOL of siblings of children with disabilities. Additionally, while several studies have examined siblings of children with disabilities in Japan, these relied on historical data for comparison with children of the same age group (Takishima, 2018; Wakimizu et al., 2020). However, support and educational systems for children with disabilities and their families vary across municipalities. Therefore, this study used children of the same age living in the same neighborhood as the control group.

This study aims to explore the QOL of siblings of children attending special-needs schools in Japan as well as related individual factors, such as lifestyle, by comparing these siblings with those of typically developing children. This study can thus add new insights to existing knowledge. Moreover, by conducting this study at a time when support systems for children with disabilities and their families are being developed, the needs of siblings can be clarified from the perspective of QOL, and recommendations can be made for future support.

Materials and Methods

Study Sample

Two groups of children were selected for this study: a group of siblings of children with

disabilities attending special-needs schools in Japan and a control group of children with siblings not receiving special-needs education. The selection criteria for the sibling group were (a) having a sibling attending special-needs school and (b) being a child between the first grade of elementary school and the third grade of junior high school. The selection criteria for the control group were (a) being a child between the first and third grades of elementary school in the same municipality and (b) having a sibling who is not receiving special-needs education. For both groups, the exclusion criteria were as follows: (a) attending a special-needs school or receiving special-needs education and (b) having no siblings (being an only child).

Setting and Recruitment

This study was conducted from October 2020 to June 2023. It targeted special-needs schools, elementary schools, junior high schools, and day care centers for children with disabilities in a district of 1,490,000 people with 11 special-needs schools. After receiving their consent to cooperate in the study, we mailed the paper questionnaire to the target institutions and asked them to distribute it in their facilities. Since it was expected that the number of siblings would be small, and there was a risk that the statistically required number of respondents could not be secured, snowball sampling was also used in conjunction with a network of parents. To facilitate parents' and siblings' responses, we used web-based survey tools (SurveyMonkey, Google Forms) with the same content as the paper questionnaire, allowing respondents to respond via smartphone or PC.

Measurements

KINDL^R QOL Measurement

This study used the KINDL^R, a health -related QOL scale for children (Ravens-Sieberer & Bullinger, 1998a, 1998b). Valid and reliable, the scale is composed of six subscales: physical well-being, emotional well-being, self-esteem, family, friends, and school, providing a comprehensive measure of children's health-related QOL. Every item was scored using a 5-point Likert scale, with total scores and subscales converted to a range of 0 to 100, with higher scores indicating better QOL. Developed in Germany in 1994, it has been translated into more than 20 languages and is used worldwide. As KINDL^R is equipped with developmentally appropriate questionnaires for use with children from elementary to junior high school, including 1 for toddlers aged 4–6 (kiddy-KINDL^R), 1 for elementary school students aged 7–13 (kid- KINDL^R), and 1 for junior high school students aged 14–17 (kiddo-KINDL^R), it is suitable for use in this study, which targeted elementary through junior high school students (Matsuzaki et al., 2007; Shibata et al., 2003). However, since the age of first-grade elementary students in Japan is 6–7 years old and the age of first-grade junior high school students is 12–13 years old, we used the kid-KINDL^R when the respondent was 6 years old in the first grade of elementary school and the kiddo-KINDL^R when the respondent was 12 years old in the first grade of junior high school. Since the use of an interview format is effective in surveying early elementary school students to elicit reliable answers, parents read the questions to the first and second-grade children in this study, and the children answered them orally (Shibata, 2012).

Investigation of QOL-Related Factors

The following items and questions were included in our questionnaire to investigate

factors related to QOL with reference to previous studies:

1. Sleep duration: The ad-hoc items “What time do you get up in the morning?” and “What time do you go to bed at night?” (Taylor et al., 2023) were used to calculate sleep duration.
2. Sleep quality: Similarly, sleep quality was investigated using the ad-hoc item “Have you slept well in the last week?” (Taylor et al., 2023). Response options were: “sleeping well” or “not sleeping well.”
3. Availability of breakfast: We asked about eating breakfast using the following ad-hoc item (Lundqvist et al., 2019): “Do you eat breakfast every morning?” The three response options were: “every day,” “sometimes,” or “not eating.”
4. Extracurricular activities: Extracurricular activities were investigated using the following ad-hoc items (Ricci et al., 2020): “Do you take part in any lessons or club activities?” “What kind of lessons or club activities do you do?” The number of lessons and club activities was calculated from the answers to the open-ended question. “How many days a week do you do lessons or club activities?” “How many hours a week do you spend in lessons and club activities?” “Is there anything that you devote all your time to?”
5. Screen time: We asked about time spent in contact with media using the following ad-hoc items: “How much do you use your cell phone or smartphone per day?” “How much do you watch TV, videos, DVDs, etc., per day?” (Kliesener et al., 2022; Motamed-Gorji et al., 2019). Here, time spent in contact with media was calculated using the total time spent from these two questions

Basic Attributes

The following questionnaire items identified basic attributes of the respondents: age, grade in school, gender, composition and number of family members living together, number of siblings, birth order within siblings, presence or absence of siblings attending special-needs schools, and their relationship to the respondent, and presence or absence of illness under treatment and its nature.

Ethical Considerations

Documents outlining the study's purpose, methods, ethical considerations, the voluntary nature of participation, participants' right to withdraw at any time without consequences, and data protection were provided along with the questionnaire. Both respondents and their parents read these documents and, if they consented, each individually checked the checkbox on the front page of the questionnaire. This research was approved by the Ethics Committee of the Graduate School of Health Sciences of Kobe University (No. 953-4).

Analysis Method

We analyzed the collected data using IBM SPSS Statistics ver. 29.0.0. If there were missing values in the QOL score, they were handled in accordance with KINDL^R regulations. Specifically, samples in which 70% or more of the items on the subscale could be answered were considered analyzable, and mean value substitution was used to process each missing value. To explain the demographic characteristics of the sample, we used descriptive statistics. Pearson's χ -square test was conducted for nominal measures, and the Mann–Whitney U test was conducted for ordinal and interval measures and proportionality measures to compare the demographics

between siblings of children with medical complexity and general siblings. Additionally, the Mann–Whitney U test was conducted for both nonparametric tests for the comparison of QOL scores of the two target groups. Furthermore, we conducted a single regression analysis using the demographic variables and QOL-related factors that showed a significant difference among the two groups as independent variables, with the overall QOL scores of the two groups as the dependent variable, to evaluate the impact of each variable on overall QOL. Multiple regression analysis was performed using the forced entry method, with variables that showed significant differences in the single regression analysis used as the independent variables and the overall QOL score for both groups as the dependent variable. Here, the standardized partial regression coefficient was tested using the t-test to calculate the p-value. Multicollinearity was measured using the variance inflation factor (VIF), and the significance level was set at $p < .05$. The Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) checklist was followed throughout the research process (Von Elm et al., 2007).

Results

Participant Attributes

The demographic characteristics of the sibling and control groups are shown in Table 1. There were 71 children in the sibling group and 398 in the control group, with a total of 469 valid responses, showing a higher percentage of girls in the sibling group. The sibling group was significantly lower in grade than the control group. The birth order within siblings was later in the sibling group compared to the control group. The proportion of siblings with illnesses was higher than that of the control group, as was the number of extracurricular activities. However,

the frequency and time spent on extracurricular activities were lower for the sibling group. Conversely, screen time was significantly higher for the sibling group.

Comparison of QOL Scores Between the Two Groups

The Mann–Whitney U test was performed to compare the two groups’ total QOL scores, as well as those on the six subscales (see Table 2). The results showed no significant differences, and the null hypothesis could not be rejected; there was no difference in QOL between the sibling and control groups.

Factors Associated with Overall QOL Scores

To investigate the impact of various variables of daily life on QOL scores, we added the presence of a sibling with disabilities (i.e., belonging to the sibling group) as one of the variables and conducted regression analysis. Initially, a single regression analysis was performed using the five variables that showed significant differences in Table 1 as independent variables and QOL scores as the dependent variable. Significant differences were found for “grade in school” and “screen time.” Therefore, we conducted a multiple regression analysis on the effect of significant variables on QOL, using the total score of the two groups ($n = 469$) as the dependent variable and the three variables of “grade in school,” “screen time,” and “whether or not there are siblings attending special-needs school” as the independent variables. The results, shown in Table 3, indicate a significant difference in “grade in school” and a decrease in QOL score as the grade increased.

Discussion

QOL Scores of the Two Groups

We began this study assuming that the QOL scores of siblings would be lower than those of the control group, but contrary to our initial hypothesis, there was no difference in QOL between the two groups. Furthermore, multiple regression analysis (as shown in Table 3) identified “grade in school” as the only significantly different factor for the overall QOL scores of the two groups, while “sibling group” had no effect. Several reasons may account for this lack of difference despite previous research suggesting otherwise. First, the siblings and families in this study may have already been receiving appropriate social support. The second reason is the separation of children with disabilities from siblings in terms of school life and extracurricular activities by attending special-needs schools. Third, the sibling group in this study had significantly lower “grade in school” and significantly later “order within siblings” compared to the control group. Each of these reasons is discussed further below.

With regard to social support, previous research on children with intellectual disabilities has found seeking support to be a factor contributing to sibling QOL; although these siblings were unable to expect support from their extended family, they were aware of the need for social support (Correia & Seabra-Santos, 2022; Moyson & Roeyers, 2011). Similarly, in research on siblings of children with autism spectrum disorder, higher levels of social support for siblings were positively associated with better family-related QOL and higher psychosocial functioning (Garrido et al., 2020; Kirchhofer et al., 2022). Given that the sibling group in this study had access to special-needs schools and daycare centers for children with disabilities, they may be connected to various specialists and have relatively easy access to social support. Furthermore, Japan has been rapidly developing support systems and laws for children with disabilities and

their families in recent years. Considering the results of previous research, this social infrastructure may also be a factor in the lack of a significant decline in QOL among the sibling group in this study.

The second reason relates to the separation of school environments between siblings. When siblings attend the same school, they are often assigned the role of communicating messages between teachers and parents, assisting their siblings with disabilities in moving between classrooms, the school cafeteria, and school clubs, and advising teachers regarding the care of their siblings to ensure a smooth school experience (Kolarikova, 2018). As a result, these siblings are unable to escape their responsibilities as caregivers, even at school, making it difficult for them to secure their own private time. Previous research has found that siblings of children with intellectual disabilities value having their own time, recognizing its importance for their own QOL (Correia & Seabra-Santos, 2022; Moyson & Roeyers, 2012). Given that the participants in our study attended a different school from their siblings with disabilities and participated in a greater “number of extracurricular activities,” it is reasonable to assume they were able to secure their own time and have opportunities to rest and relax. The separation of siblings’ schools unintentionally created a respite-like situation, and previous research has demonstrated that the use of respite care not only relieves parental stress but also benefits siblings (Welch et al., 2012). Thus, the ability of siblings to construct their own routines by separating school, extracurricular activities, and other social pursuits from their sibling with disabilities and their family life may have contributed to why QOL did not decline in this study.

Finally, mutual understanding and acceptance between siblings is essential for their QOL (Moyson & Roeyers, 2011). The sibling group in our study was younger than the control group and later in birth order, so it is possible that a family lifestyle centered around caring for the

sibling with disabilities had already been established and normalized while they were young. As a result, they may have been less aware that their own QOL was being negatively affected by their sibling with disabilities. Additionally, given their younger age, many of these siblings may not have been given caretaking responsibilities for their sibling with disabilities at home. Furthermore, as the siblings attended different schools from their siblings with disabilities, it is also possible that they were not required to take on such roles outside the home.

Support for Siblings and Their Families

Having discussed the reasons for the lack of difference in QOL scores between the sibling and control groups, this section discusses support strategies for siblings of children with disabilities within the community. Two types of support are highlighted: 1) support to prevent siblings' current QOL from deteriorating and 2) the provision of education and transitional support for siblings and family members.

At the time of this study, although there was no significant decline in QOL score in the sibling group compared to the control group, among the QOL subscale scores, "self-esteem" was lower in both groups. In fact, since the development of the Japanese version of KINDL^R, scores on the self-esteem subscale have been reported to be lower than those on the other subscales (Matsuzaki et al., 2007; Shibata et al., 2003). This could be because children's self-esteem is known to generally decline from childhood to adolescence (Chung et al., 2017). Children's self-esteem is greatly affected not only by their family environment but also by broader surroundings like neighborhoods and schools, and children with lower self-esteem are at risk of developing psychological and social issues (Hosogi et al., 2012; Krauss et al., 2020). One study indicated that the self-esteem of siblings of children with intellectual disabilities was not directly affected

by this condition (Saban & Arikan, 2013). However, other studies suggest that children's QOL is affected in various ways by their connection to the community. For instance, siblings can experience stigma due to the presence of a family member with a disability, resulting in reduced interaction with peers and bullying (Marquis et al., 2019; Múries-Cantán, Schippers, et al., 2023). It is therefore necessary to ensure siblings' have supportive relationships and community involvement outside the family to ensure their mental well-being.

Table 1 shows that the sibling group was able to participate in more activities than the control group, but they were not able to engage in these activities for long periods of time or more frequently. This supports previous research findings that siblings have lower levels of physical activity (Çelik et al., 2018). Furthermore, as mentioned, siblings may be required to care for children with disabilities or fulfill various roles within or outside the family, which limits their time for extracurricular activities; this may be the case in this study, as well. In addition to lower levels of physical activity, long durations of screen time are associated with a decrease in children's QOL (Kliesener et al., 2022; Motamed-Gorji et al., 2019). No decline in QOL score was observed among siblings in this study, but there is a need to continue ensuring that children have opportunities to engage with society through extracurricular activities and other means while also fostering positive peer relationships to maintain and promote QOL.

With regard to the provision of education and transitional support for siblings and family members of children with disabilities, siblings' QOL generally declines with age, and the roles required of them change as their life course changes (Hall & Rossetti, 2018). Given the increase in life expectancy of persons with disabilities, they often outlive their caregivers, at which point their siblings may take over the role. Although parents may expect siblings of children with disabilities to take over some of their caregiving responsibilities, siblings have specific plans for

how they will fulfill their guardian roles and how they will care for their disabled family members (Kruithof et al., 2021; Park et al., 2021). Consequently, siblings of children with disabilities may experience considerable anxiety and conflict in adulthood and beyond. Previous research has reported on the importance of securing the future of children with disabilities for the QOL of their siblings (Correia & Seabra-Santos, 2022). Support for siblings in performing their caregiver roles and dealing with the accompanying anxiety is thus crucial.

In particular, expanding policies and support services for families with disabled members, along with ensuring reliable local community resources, is a considerable source of strength for them (Park et al., 2021). Reducing the mental anxiety and the burden on siblings who are expected to take on the caregiver role through resources such as home visits, in-home support, and daycare support for children with disabilities is also necessary. Although the sibling group's current QOL did not decrease in this study, their circumstances may change when their siblings with disabilities eventually graduate from special-needs schools. Thus, along with providing information and support on issues that may arise after children with disabilities graduate, support should also be provided for the preparatory establishment of family structures that can respond to future issues. Furthermore, while transitional support for individual children with disabilities is an important issue, transitional support for their family members, including siblings, must also be taken into account.

As previously discussed, encouraging mutual understanding between siblings and family members of children with disabilities is key for their QOL. Psychological support is important for ensuring individuals with disabilities, their siblings, and their families can maintain positive and close relationships throughout their development. Concretely, the process of accepting the disability of a sibling is considered an important area of QOL, and future support should enable

an appropriate process of acceptance as children mature (Moyson & Roeyers, 2012). Therefore, it is necessary to provide educational support to caregivers, such as parents and grandparents, from an early stage. Various institutions, including medical institutions, special support schools, public health centers, and facilities for supporting people with disabilities should cooperate and collaborate to provide comprehensive support for families.

Strengths and Limitations

This is one of the few studies examining the QOL and related individual factors of siblings of children attending special-needs schools. One of its strengths is that siblings of typically developing children living in the same community were used as the control group. Since neither of the two groups of children investigated attended special-needs schools or received special-needs education, the study's results can guide the development of school health services tailored to children and their families from diverse backgrounds who face various difficulties. The results also provide valuable data for school nurses and educators. An additional advantage of this study is that it used self-reported data from children rather than proxy responses from parents, facilitating a more accurate investigation of children's QOL.

While data collection in this study focused on the experiences of siblings of children with disabilities attending special-needs schools—a factor that could significantly impact their QOL—the sample size of the sibling group contained fewer than 100 participants. This limited sample arose from the lack of population data on siblings of children with disabilities within the survey area. Due to the limited sample size, the generalizability of the findings to a broader population may not be sufficient. In addition, the timing of data collection was during the COVID-19 pandemic, which limited the number of facilities and families that could cooperate

with this study. Finally, as this was a cross-sectional study, we were able to confirm the relationship between variables, but the causal relationships are unknown, and further investigation is required in the future.

Conclusion

The aim of this study was to clarify the actual situation of the QOL of siblings of children attending special-needs schools in Japan, as well as to explore siblings' relevant individual factors through comparison with siblings of typically developing children. The results showed that only "grade in school" was significantly different for the overall QOL scores. This lack of difference in QOL between the sibling and control groups may be because the siblings and families of children with disabilities in this study were already receiving appropriate social support and because the siblings were able to create separate school and extracurricular routines. Moreover, participants in the sibling group were generally younger than those in the control group, so a caretaking routine within the family might already have been established. While it is important to provide support to prevent siblings' QOL from deteriorating (e.g., by allowing them to participate in extracurricular activities or spending time together as a family), it is also important to support siblings in managing the various roles, responsibilities, and anxieties that may arise within the family as they grow older. Offering educational support to the family as well as social support outside the family while monitoring the family situation over time can help maintain and improve the QOL of siblings.

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Data Availability

The dataset includes highly sensitive health information, and the authors did not obtain approval from the Ethics Committee of the principal investigator's institution to publicly share the data collected. For inquiries regarding access to the data, please contact us here (bbhandm26@gmail.com).

Declaration of Conflicting Interests

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References

- Buchbinder, D., Patel, S. K., Casillas, J. N., Nugent, D. J., Neudorf, S., Sender, L. S., Torno, L., Huszti, H., Zeltzer, L. K., & Parsons, S. K. (2019). Parent proxy assessment of sibling quality of life following pediatric hematopoietic cell transplantation. *Health and Quality of Life Outcomes*, 17(1), 162. <https://doi.org/10.1186/s12955-019-1231-9>
- Çelik, E. M., Daskapan, A., & Topcu, Z. G. (2018). Physical fitness, physical activity, psychosocial status and quality of life of adolescent siblings of neurologically disabled children. *Baltic Journal of Health and Physical Activity*, 10(3), 27–37. <https://doi.org/10.29359/BJHPA.10.3.03>
- Cheak-Zamora, N. C., & Thullen, M. (2017). Disparities in quality and access to care for children with developmental disabilities and multiple health conditions. *Maternal and Child Health Journal*, 21(1), 36–44. <https://doi.org/10.1007/s10995-016-2091-0>
- Chung, J. M. H., Hutteman, R., van Aken, M. A. G., & Denissen, J. J. A. (2017). High, low, and in between: Self-esteem development from middle childhood to young adulthood. *Journal of Research in Personality*, 70, 122–133. <https://doi.org/10.1016/j.jrp.2017.07.001>
- Cieza, A., Kamenov, K., Sanchez, M. G., Chatterji, S., Balasegaram, M., Lincetto, O., Servili, C., Bermejo, R., & Ross, D. A. (2021). Disability in children and adolescents must be integrated into the global health agenda. *BMJ*, 372, 773. <https://doi.org/10.1136/bmj.n773>
- Correia, R. A., & Seabra-Santos, M. J. (2022). I would like to have a normal brother but I'm happy with the brother that I have: A pilot study about intellectual disabilities and family quality of life from the perspective of siblings. *Journal of Family Issues*, 43(12), 3148–3167. <https://doi.org/10.1177/0192513X211042845>

- Coster, W., Law, M., Bedell, G., Liljenquist, K., Kao, Y. C., Khetani, M., & Teplicky, R. (2013). School participation, supports and barriers of students with and without disabilities. *Child: Care, Health and Development*, 39(4), 535–543.
<https://doi.org/10.1111/cch.12046>
- Dinleyici, M., Çarman K. B., Özdemir, C., Harmancı, K., Eren, M., Kirel, B., Şimşek, E., Yazar, C., Duyan Çamurdan, A., & Şahin Dağlı, F. (2019). Quality-of-life evaluation of healthy siblings of children with chronic illness. *Balkan Medical Journal*, 37(1), 34–42.
<https://doi.org/10.4274/balkanmedj.galenos.2019.2019.7.142>
- Garrido, D., Carballo, G., & Garcia-Retamero, R. (2020). Siblings of children with autism spectrum disorders: Social support and family quality of life. *Quality of Life Research*, 29(5), 1193–1202. <https://doi.org/10.1007/s11136-020-02429-1>
- GBD 2017 Child and Adolescent Health Collaborators. (2019). Diseases, injuries, and risk factors in child and adolescent health, 1990 to 2017: Findings from the global burden of diseases, injuries, and risk factors 2017 study. *JAMA Pediatrics*, 173(6), e190337.
<https://doi.org/10.1001/jamapediatrics.2019.0337>
- Goudie, A., Havercamp, S., Jamieson, B., & Sahr, T. (2013). Assessing functional impairment in siblings living with children with disability. *Pediatrics*, 132(2), e476–e483.
<https://doi.org/10.1542/peds.2013-0644>
- Hall, S. A., & Rossetti, Z. (2018). The roles of adult siblings in the lives of people with severe intellectual and developmental disabilities. *Journal of Applied Research in Intellectual Disabilities*, 31(3), 423–434. <https://doi.org/10.1111/jar.12421>
- Hanvey, I., Malovic, A., & Ntontis, E. (2022). Glass children: The lived experiences of siblings of people with a disability or chronic illness. *Journal of Community & Applied Social*

- Psychology*, 32(5), 936–948. <https://doi.org/10.1002/casp.2602>
- Hosogi, M., Okada, A., Fujii, C., Noguchi, K., & Watanabe, K. (2012). Importance and usefulness of evaluating self-esteem in children. *BioPsychoSocial Medicine*, 6, 9. <https://doi.org/10.1186/1751-0759-6-9>
- Houtrow, A., Sabatini, C., & Jarvis, J. (2023). Health equity for children with disabilities. *Journal of the Pediatric Orthopaedic Society of North America*, 5(1), 634. <https://doi.org/10.55275/JPOSNA-2023-634>
- Isogai, K. (2017). Recent developments in Japan’s special needs education: Promoting an inclusive education system. *NISE Bulletin*, 16, 28–32.
- Kirchhofer, S. M., Orm, S., Haukeland, Y. B., Fredriksen, T., Wakefield, C. E., & Fjermestad, K. W. (2022). A systematic review of social support for siblings of children with neurodevelopmental disorders. *Research in Developmental Disabilities*, 126, 104234. <https://doi.org/10.1016/j.ridd.2022.104234>
- Kliesener, T., Meigen, C., Kiess, W., & Poulain, T. (2022). Associations between problematic smartphone use and behavioural difficulties, quality of life, and school performance among children and adolescents. *BMC Psychiatry*, 22, 195. <https://doi.org/10.1186/s12888-022-03815-4>
- Kolarikova, M. (2018). Sibling of disabled child at the inclusive school. *Journal of Community Medicine & Health Education*, 8(2), 609. <https://doi.org/10.4172/2161-0711.1000609>
- Koukouriki E., & Soulis SG. (2020). Self-reported health-related quality of life (HRQOL) and anxiety among Greek school-age siblings of individuals with autism spectrum disorders (ASD) in relation to parental mental health and social support. *Journal of Autism and Developmental Disorders*, 50(8), 2913–2930. <https://doi.org/10.1007/s10803-020-04395->

- Krauss, S., Orth, U., & Robins, R. W. (2020). Family environment and self-esteem development: A longitudinal study from age 10 to 16. *Journal of Personality and Social Psychology*, 119(2), 457–478. <https://doi.org/10.1037/pspp0000263>
- Kruihof, K., IJzerman, L., Nieuwenhuijse, A., Huisman, S., Schippers, A., Willems, D., & Olsman, E. (2021). Siblings' and parents' perspectives on the future care for their family member with profound intellectual and multiple disabilities: A qualitative study. *Journal of Intellectual and Developmental Disability*, 46(4), 351–361. <https://doi.org/10.3109/13668250.2021.1892261>
- Lamsal, R., & Ungar, W. J. (2021). Impact of growing up with a sibling with a neurodevelopmental disorder on the quality of life of an unaffected sibling: A scoping review. *Disability and Rehabilitation*, 43(4), 586–594. <https://doi.org/10.1080/09638288.2019.1615563>
- Lundqvist, M., Vogel, N. E., & Levin, L. Å. (2019). Effects of eating breakfast on children and adolescents: A systematic review of potentially relevant outcomes in economic evaluations. *Food & Nutrition Research*, 63. <https://doi.org/10.29219/fnr.v63.1618>
- Marquis, S., Hayes, M. V., & McGrail, K. (2019). Factors that may affect the health of siblings of children who have an intellectual/developmental disability. *Journal of Policy and Practice in Intellectual Disabilities*, 16(4), 273–286. <https://doi.org/10.1111/jppi.12309>
- Matsui, Y. (2022). Examination of the results of the "children receiving medical care and their family life survey report" by the ministry of health, labor and welfare and 1 after subsequent legal support. *Niigata Iryo Fukushi Gakkaishi*, 21(3), 92–99. https://doi.org/10.34540/niigatajohewewa.21.3_92

- Matsuzaki, K., Nemoto, Y., Shibata, R., Morita, K., Sato, H., Furusho, J., Watanabe, S., Okuruyama, M., Kubagawa, T., Maekawa, K. (2007). A study of the Kiddo-KINDL^R (questionnaire for measuring health-related quality of life in children, revised version for 13 to 16-year-olds) in Japan. *The Journal of the Japan Pediatric Society*, 111(11), 1404–1410.
- McTavish, J. R., McKee, C., Tanaka, M., & MacMillan, H. L. (2022). Child welfare reform: A scoping review. *International Journal of Environmental Research and Public Health*, 19(21), 14071. <https://doi.org/10.3390/ijerph192114071>
- Motamed-Gorji, N., Qorbani, M., Nikkho, F., Asadi, M., Motlagh, M.E., Safari, O., Arefirad, T., Asayesh, H., Mohammadi, R., Mansourian, M., & Kelishadi, R. (2019). Association of screen time and physical activity with health-related quality of life in Iranian children and adolescents. *Health and Quality of Life Outcomes*, 17(1), 2. <https://doi.org/10.1186/s12955-018-1071-z>
- Moyson, T., & Roeyers, H. (2011). The quality of life of siblings of children with autism spectrum disorder. *Exceptional Children*, 78(1), 41–55. <https://doi.org/10.1177/001440291107800103>
- Moyson, T., & Roeyers, H. (2012). ‘The overall quality of my life as a sibling is all right, but of course, it could always be better’. Quality of life of siblings of children with intellectual disability: The siblings' perspectives. *Journal of Intellectual Disability Research*, 56(1), 87–101. <https://doi.org/10.1111/j.1365-2788.2011.01393.x>
- Mulroy, S., Robertson, L., Aiberti, K., Leonard, H., & Bower, C. (2008). The impact of having a sibling with an intellectual disability: Parental perspectives in two disorders. *Journal of Intellectual Disability Research*, 52(3), 216–229. <https://doi.org/10.1111/j.1365->

- Múries-Cantán, O., Giné, C., Brown, R. I., Aguiar, N. B., & Schippers, A. P. (2023). Siblings of children with intellectual and developmental disabilities: Quality of life perceptions from Catalonia. *Journal of Policy and Practice in Intellectual Disabilities*, 20(2), 192–204. <https://doi.org/10.1111/jppi.12451>
- Múries-Cantán, O., Schippers, A., Giné, C., & Blom-Yoo, H. (2023). Siblings of people with intellectual and developmental disabilities: A systematic review on their quality of life perceptions in the context of a family. *International Journal of Developmental Disabilities*, 69(6), 797–810. <https://doi.org/10.1080/20473869.2022.2036919>
- Naz, A., & Akhtar, S. M. J. (2020). Special policies for special children: A policy review. *International Journal of Latest Research in Humanities and Social Science*, 3(10), 119–123.
- Ohtoshi, T. (2016). Current status and tasks of support systems for children with developmental disorders - Focusing on recent policy progress and clinical practice. *Journal of Allied Health Sciences*, 7(1), 11–16. <https://doi.org/10.15563/jalliedhealthsci.7.11>
- Olusanya, B. O., Wright, S. M., Nair, M. K. C., Boo, N. Y., Halpern, R., Kuper, H., Abubakar, A. A., Almasri, N. A., Arabloo, J., Arora, N. K., Backhaus, S., Berman, B. D., Breinbauer, C., Carr, G., de Vries, P. J., Del Castillo-Hegyi, C., Eftekhari, A., Gladstone, Hoekstra, R. A. ... Global Research on Developmental Disabilities Collaborators. (2020). Global burden of childhood epilepsy, intellectual disability, and sensory impairments. *Pediatrics*, 146(1) e20192623. <https://doi.org/10.1542/peds.2019-2623>
- Olusanya, B. O., Wright, S. M., Smythe, T., Khetani, M. A., Moreno-Angarita, M., Gulati, S., Brinkman, S. A., Almasri, N. A., Figueiredo, M., Giudici, L. B., Olorunmoteni, O.,

- Lynch, P., Berman, B., Williams, A. N., Olusanya, J. O., Wertlieb, D., Davis, A. C., Hadders-Algra, M., & Gladstone, M. J. (2024). Early childhood development strategy for the world's children with disabilities. *Frontiers in Public Health*, 19(12), 1390107. <https://doi.org/10.3389/fpubh.2024.1390107>
- Park, S., Ryu, W., & Yang, H. (2021). A study on the life experiences of adolescents who grew up with younger siblings with developmental disabilities: Focusing on phenomenological analysis methods. *Brain Sciences*, 11(6), 798. <https://doi.org/10.3390/brainsci11060798>
- Rana, P., & Mishra, D. (2015). Quality of life of unaffected siblings of children with chronic neurological disorders. *The Indian Journal of Pediatrics*, 82(6), 545–548. <https://doi.org/10.1007/s12098-014-1672-4>
- Ravens-Sieberer, U., & Bullinger, M. (1998a). Assessing health-related quality of life in chronically ill children with the German KINDL: First psychometric and content analytical results. *Quality of Life Research*, 7(5), 399–407. <https://doi.org/10.1023/a:1008853819715>
- Ravens-Sieberer, U. & Bullinger, M. (1998b). News from the KINDL-Questionnaire – A new version for adolescents. *Quality of Life Research*, 7, 653.
- Ricci, J. M., Clevenger, K. A., Sellers, S., Davenport, S., & Pfeiffer, K. A. (2020). Associations between extracurricular activity participation and health-related variables in underrepresented children. *Sports Medicine and Health Science*, 2(2), 102–108. <https://doi.org/10.1016/j.smhs.2020.06.001>
- Saban, F., & Arikan, D. (2013). The self-esteem and anxiety of children with and without mentally retarded siblings. *Journal of Research in Medical Science*, 18(11), 961–969.
- Shibata, R. (2012). Health-related quality of life in children and adolescents - Japanese versions

- of Kid-KINDL^R and Kiddo-KINDL^R questionnaires. *Health Science for Children*, 13(1), 39–46.
- Shibata, R., Nemoto, Y., Matsuzaki, K., Tanaka, D., Kawaguchi, T. & Kanda, A. (2003). A study of the Kid-KINDL questionnaire for measuring quality of life in elementary school children in Japan. *The Journal of the Japan Pediatric Society*, 107(11), 1514–1520.
- Takishima, M. (2018). Supporting school-age siblings of children with disabilities -The association between the result of QOL measurement that utilizes KINDL^R and the psychosocial influence on siblings of children with disabilities. *Mejiro Journal of Social and Natural Science*, 14, 81–92.
- Taylor, R. W., Haszard, J. J., Jackson, R., Morrison, S., Beebe, D. W., Meredith-Jones, K. A., Elder, D. E., & Galland, B. C. (2023). Effect of sleep changes on health-related quality of life in healthy children: A secondary analysis of the DREAM crossover trial. *JAMA Network Open*, 6(3), e233005. <https://doi.org/10.1001/jamanetworkopen.2023.3005>
- Teruya, H., Shimojo, M., & Kwon, H. (2019). Global trends in developmental disorders education and Japan's current status and new initiatives. *Asian Journal of Human Services*, 16, 101–111. <https://doi.org/10.14391/ajhs.16.101>
- Von Elm, E., Altman, D. G., Egger, M., Pocock, S. J., Gøtzsche, P. C., & Vandenbroucke, J. P. (2007). The strengthening the reporting of observational studies in epidemiology (STROBE) statement: Guidelines for reporting observational studies. *The Lancet*, 370(9596), 1453–1457. [https://doi.org/10.1016/s0140-6736\(07\)61602-x](https://doi.org/10.1016/s0140-6736(07)61602-x)
- Wakimizu, R., Fujioka, H., Nishigaki, K., & Matsuzawa, A. (2020). Quality of life and associated factors in siblings of children with severe motor and intellectual disabilities: A cross-sectional study. *Nursing & Health Sciences*, 22(4), 977–987.

<https://doi.org/10.1111/nhs.12755>

Welch, V., Hatton, C., Emerson, E., Robertson, J., Collins, M., Langer, S., & Wells, E. (2012).

Do short break and respite services for families with a disabled child in England make a difference to siblings? A qualitative analysis of sibling and parent responses. *Children and Youth Services Review*, 34(2), 451–459.

<https://doi.org/10.1016/j.childyouth.2011.12.002>

Table 1*Characteristics of Participants*

	Siblings of children with disabilities	Siblings of typically developing children	<i>p</i>
Total participants	71	398	-
Gender (boy/girl)	26/45	196/202	.054
Grade in school (median [IQR])	6 [4–8]	7 [5–8]	.015*
Single caregiver/non-single caregiver	6/65	36/362	1.000
Number of siblings (median [IQR])	1 [1–2]	1 [1–2]	.917
Order within siblings (median [IQR])	3 [3–3]	3 [2–3]	< .001***
Chronic disease (present/absent)	19/52	67/331	.065
Asthma	2	6	
Atopic dermatitis	3	23	
Kidney disease	0	1	.136
Heart disease	0	1	
Other	14	36	
Sleep quality (well/not well)	59/12	340/58	.590
Sleep time (hours: median [IQR])	8.50 [7.83–9.00]	8.00 [7.50–9.00]	.197
Breakfast (always/sometimes/none)	63/6/2	357/34/7	.836
Time spent in extracurricular activities (min/week: median [IQR])	180.00 [60.00–360.00]	330.00 [120.00–600.00]	.028*
Number of extracurricular activities (median [IQR])	2 [1–3]	1 [1–2]	.025*
Something to be passionate about (Exist/not exist)	59/12	306/92	.280
Screen time (min/day: median [IQR])	180.00 [120.00–285.00]	180.00 [120.00–240.00]	.040*

Note. “Grade in School” was calculated by converting first grade elementary school students to 1 and third grade junior high school students to 9. “Order within Siblings” was calculated by converting only child to 1, eldest sibling to 2, and second sibling from the top to 3 (and so on). IQR: inter-quartile range. * $p < .05$, ** $p < .01$ and *** $p < .001$.

Table 2*Quality of Life Scores*

	Siblings of children with disabilities	Siblings of typically developing children	<i>p</i>
	(median [IQR])	(median [IQR])	
Total QOL	72.92 [61.5–81.3]	71.88 [63.5–82.3]	.711
Physical well-being	81.25 [62.5–87.5]	81.25 [68.8–93.8]	.572
Emotional well-being	81.25 [68.8–93.8]	81.25 [68.8–93.8]	.623
Self-esteem	56.25 [37.5–68.8]	56.25 [43.8–68.8]	.946
Family	75.00 [62.5–87.5]	75.00 [68.8–87.5]	.792
Friends	75.00 [62.5–93.8]	81.25 [68.8–93.8]	.200
School	62.50 [50.0–75.0]	62.50 [50.0–75.0]	.922

Note. IQR: inter-quartile range; QOL: quality of life

Table 3*Multiple Regression Analysis of Factors Associated with Quality of Life Scores*

Factors	SE	Beta	<i>t</i>	<i>p</i>	95% CI		Collinearity	
					Lower	Upper	Tolerance	VIF
Siblings of children with/without disabilities	1.662	0.046	1.027	.305	-1.560	4.973	0.978	1.022
Grade in school	0.259	-0.240	-5.257	<.001	-1.874	-0.854	0.964	1.038
Screen time	0.005	-0.067	-1.465	.144	-0.018	-0.003	0.962	1.040

Note. CI: confidence interval; SE: standard error; VIF: variance inflation factor