



**Quality of Life and associated factors in siblings of children
with severe motor and intellectual disabilities: A cross-
sectional study**

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Abstract (176 words)

This study examined quality of life and its associated factors in siblings of children with severe motor and intellectual disabilities in Japan. The participants were 789 siblings of children with a disability and their primary caregivers. We used the Kinder Lebensqualität Fragebogen questionnaire to assess the quality of life of siblings. The mean age of the siblings in this study was 12.21 ± 3.07 years, and the mean quality of life score was 69.63 ± 12.55 points, which is higher than that of the general population of children of the same age. It was revealed that the following factors contributed to higher quality of life scores: a closer relationship with the child with a disability, younger age, the primary caregiver's lower care burden, later birth order of siblings (i.e., younger siblings), higher family empowerment, and female gender of siblings. The relationship with the child with a disability had the strongest influence on siblings' quality of life. Our study suggests the need for nursing interventions that focus on the whole family to enhance siblings' quality of life.

Keywords: Children with disabilities, Family, Empowerment, Japan, Quality of life, Siblings

1. INTRODUCTION

Families caring children with disabilities are faced with several challenges in family life. In particular, regarding the experiences of the families of children with chronic conditions, it has also been reported that the siblings are socially resilient yet overlooked members of the family who may present psychological, academic, and peer-related difficulties following the diagnosis of a brother or sister with chronic conditions (Gan, Lum, Wakefield, Nandakumar, & Fardell, 2016). Orsmond and Seltzer (2017) provide some evidence that the siblings of children with autism spectrum disorders describe both positive and negative aspects of their sibling relationship and may be at an increased risk of social and behavioral adjustment problems during childhood and adolescence. In addition, the siblings of children with a disability have reported close relations within their families due to the potential role shifts of family members in their situation (Van Riper, 2003). Moreover, siblings of these children have feeling of forbearance (Moyson& Roeyers, 2011). The forbearance leads to difficulty in expressing their feelings.

The influence of children with disabilities on their siblings is modified by several factors relating to the characteristics and dynamics of the individual family members and of the family as a whole. Living with a child with disability is thus hypothesized to have a significant impact on the quality of life (QoL) of their siblings. QoL pertains to psychological well-being, social relationships, physical functions, and activities of daily life as perceived by both adults and children (Ravens-Sieberer &

Bullinger, 1998).

The number of severe motor and intellectual disabilities (SMID) is increasing in Japan, with the current number estimated to be about 40,000. Children with SMID have been defined as “children with severe physical and intellectual disability.” This term is included in the Children with Special Healthcare Needs (CSHCN) (McPherson et al., 1998). These children require very high-level specialist medical care, such as respiratory support, nutritional support, and daily rehabilitation. The most common diagnoses in SMID are cerebral palsy and chromosomal abnormalities. About 70% of children with SMID live at home (Okada, Iai, Ishii, & Ozawa, 2015). Children with SMID require constant long-term care, as they cannot move on their own to perform most of the activities of daily living and have difficulty in engaging in verbal communication (Tokyo Metropolis, 2014). The family caregivers of children with CSHCN and SMID has impacted from their daily caregiving. There is evidence that caring for these children affects the family caregivers’ physical and mental health (Davis & Gavidia-Payne, 2009; Raina et al., 2005), friendships, marital lives, social participation (Davey et al., 2015), and employment (Okumura et al., 2009), as well as the economic situation of the whole family (Davis et al., 2010; Ushio, 2014). Support for the lives of families of children with SMID was instituted by law in 1967 through institutional services for children with SMID in Japan. Today, there are many kinds of social services, such as short-stay services, visiting nurses, helpers, and day care services. It is still, however, necessary to consider support for these families in the effective use of

such services to increase their quality of life (Nishigaki et al., 2016).

Quality of life (QoL) is a psychological concept describing the physical, social, psychological, and functional aspects of well-being, therefore, support to improve the QOL of siblings of the families raising these children is essential. Especially, Bertelli et al. (2011) described that the QoL is perceived somewhat differently by individuals and by members of their families. Recent research on sibling quality of life has, however, found a discrepancy between parents' and siblings' reports (Houtzager, 2005). Most research on sibling adjustment is based on parental report. Therefore, when planning the care for the siblings, it is important to take into consideration their own self-reported QoL.

1.1. Literature review

Previous studies have reported inconsistent results on QoL of siblings of children with a disorder or illness. Some studies have reported the significant impairment of the QoL of siblings of children with a chronic illness compared to siblings of healthy children (Eladl & Atwa, 2011; Rana & Mishra, 2015; Yılmaz, Türkeli, Karaca, & Yüksel, 2017). On the other hand, other studies have reported comparatively high QoL among siblings of children with diseases such as cystic fibrosis and leukemia (Berbis et al., 2015; Havermans et al., 2011). Based on a systematic review, Limbers and Skipper (2014) found that the higher the severity of the disease in children with a chronic health condition such as cancer, type 1 diabetes, or epilepsy, the higher the risk of a decline in their siblings' QoL. It is

therefore hypothesized that the QoL of siblings of children with SMID is lower than that of siblings of healthy children.

Another study reported that illness-related problem behavior and female gender were negatively correlated with QoL of siblings of children with psychological disorders (Bowman, Alvarez-Jimenez, Wade, Howie, & McGorry, 2014). Furthermore, among siblings of children with leukemia, Berbis et al. (2015) found that an older age at diagnosis for both siblings and children with leukemia, low socioeconomic status, and cancer-related factors were risk factors for impaired QoL of siblings. Furthermore, Barrera, Atenafu, Nathan, Schulte, and Hancock (2018) developed an intervention program for siblings of children with cancer, and assessed its efficacy in addressing depression symptoms and QoL of siblings, but their results were inconclusive. In addition, no previous study has reported on QoL of siblings of children with SMID, which may be presumed to be much lower than in siblings of healthy children.

To understand the experiences and perceived needs of siblings of children with SMID and to develop support programs, we should focus on siblings' QoL via their own voice by directly asking them for their perspectives (Meadan, Stoner, & Angell, 2010). Previous studies reported differences between parent-reported and sibling-reported QoL. With respect to assessing the QoL of siblings of children with a disorder or illness, previous research revealed significant discrepancies between the siblings' self-reports and their parent-proxy reports (Baca, Vickrey, Hays, Vassar, & Berg, 2010; Houtzager,

Grootenhuis, Caron, & Last, 2005), especially on social and emotional subscales (Schulte, Wurz, Reynolds, Strother, & Dewey, 2016).

1.2. Framework

As mentioned above, the QoL of siblings is largely influenced by the functions and empowerment of the whole family and factors relating to their parents (marital status, working style, physical status, psychological status, and so forth) who raise and take care of them. Moreover, the situation of the parents also affects the QoL of siblings. In particular, since primary caregivers spend a significant amount of time with the siblings, the primary caregivers' physical and psychological situation must be assessed in order to consider the QoL of siblings. In this study, we set variables and scales in order to explore the QoL of siblings in an exploratory manner, mainly in relation to the assumed factors of families and parents.

1.3. Purpose

The purposes of this study were to clarify the self-reported QoL of siblings of children with SMID and its associated factors with a focus on family environmental factors, such as the QoL of caregivers, family empowerment, and family functioning, and to devise both appropriate and comprehensive forms of support for the entire family with vulnerabilities and practical approaches to improve siblings'

QoL.

2. METHODS

2.1. Design

This study was conducted in Japan using a self-report questionnaire-based, cross-sectional design.

2.2. Participants and settings

We recruited siblings aged 6–18 and the primary caregivers of children with SMID who attend special needs schools in Japan. The inclusion criteria included being able to read and write in Japanese.

2.3. Data collection

We phoned all 212 special needs schools that span the entire younger-to-older age ranges (6–18 years old) in Japan, of which 89 agreed to participate in this study. They sent out questionnaires to every family at their school. We contacted the primary caregivers of children with SMID and provided the consent form and questionnaires via the schoolteachers. We recruited primary caregivers, partners of primary caregivers, and siblings of children with SMID. If a child with SMID attending the above schools had siblings between the ages of 6–18 years, they were included in the study. It was clarified in the manual that if there were multiple siblings per child with SMID to be included, only the sibling

who had cohabitated the longest with the child with SMID would be selected. Siblings with disabilities and siblings living separately from the child with SMID were excluded from this study. This means that we excluded infants, toddlers, and siblings that lived away from home, and if the sibling was affected by their own disability or an even more severe SMID than the index child, they were also excluded. We included both adopted and fostered siblings if they lived with the children with SMID as a family. Each family placed their completed questionnaires in an envelope and submitted them at their child's school, which then sent the collected envelopes to the researchers' institution. Data were collected between November 2015 and March 2016.

2.4. Measurement tools

Siblings of children with SMID answered the following two questionnaires: one on the attributes of the siblings, the Kinder Lebensqualität Fragebogen (KINDL), and one on their relationship with the child with SMID. Primary caregivers answered the following questionnaires: Attributes of Primary Caregivers, Family and Children with SMID, the Japanese version of the Family Empowerment Scale (J-FES), the Eight-Item Short Form of the Japanese version of the Zarit Burden Interview (J-ZBI_8), the Family Adaptability and Cohesion Evaluation Scale Kwansei Gakuin IV-16 (FACESKGIV-16), and the Eight-Item Short Form Health Survey (SF-8). In using these scales for families of child with SMID, we verified the content validity of all of them. We then surveyed the results of the KINDL, J-

FES, J-ZBI_8, FACESKGIV-16, and SF-8 and obtained permission to use these scales.

2.4.1. QoL of siblings

The KINDL assesses health-related QoL in children and adolescents (Ravens-Sieberer & Bullinger, 1998). It has been translated into 30 languages and has been used in many international studies. Matsuzaki et al. (2007) and Shibata et al. (2003) translated and developed a Japanese version of the KINDL comprised of 24 items that yield a general QoL score and scores for the following six subscales: physical health, general health, family functioning, self-esteem, social functioning, and school functioning. Siblings with at least one chronic condition answered the disease module, consisting of six items. Each item is scored using a five-point Likert scale. Total scores and subscales are transformed into a range of 0–100, with higher scores indicating better QoL. In this study, we adopted different measurement batteries for siblings from KINDL, because it does not measure all objects with the same battery; it is recommended to use a corresponding battery according to the year range. The Cronbach's α was .83–.85, and thus internal consistency was confirmed in this study.

2.4.2. Relationship between children with SMID and their siblings

The relationship between children with SMID and their siblings was assessed with the following three items rated on a five-point Likert scale (1 = *never* to 5 = *all the time*). Item 1 asked participants

to respond to the statement “I have a good relationship with my sisters or brothers,” Item 2 was “I take care of my sisters or brothers,” and Item 3 was “I enjoy taking care of my sisters or brothers.” The items were developed by the authors based on the findings of a previous study (Wakimizu, Fujioka, Numaguchi, Nishigaki, & Sato, 2015) and validated carefully so that even young children could understand and answer them. A higher score indicated that siblings had a favorable opinion of their relationships with their siblings. The Cronbach’s α for this scale was .79 in this study.

2.4.3. Attributes of children with SMID

For the attributes of children with SMID, primary caregivers reported on the presence and type of medical care. We modified the severity assessment tool developed by Suzuki et al. (2008) to make it easier for primary caregivers to understand. Examples of questions are as follows: “What care does your child need for breathing? Check all the following applicable cares. a. Ventilator (10 points), b. Tracheostomy (8 points), c. Airway (5 points), d. Oxygen inhalation (5 points), e. Suction at least once per hour (8 points), f. Suction more than six times a day (3 points).” This tool required simplification to ensure consistency in the answers by primary caregivers without specialized medical knowledge. Children scoring more than 25 points on this scale were classified as being in the SMID-medical care dependent group (SMID-MCDG), and those scoring 10–25 points were classified as sub-SMID-MCDG. This classification is the most valuable tool to assess the need for social services in Japan.

2.4.4. Primary caregivers' burden

The J-ZBI_8 is an instrument for measuring a caregiver's perceived burden of providing family care (Arai, Tamiya, & Yano, 2003); its reliability and validity have been verified. The J-ZBI has high internal consistency (Cronbach's alpha = 0.89). Items are rated on a five-point scale ranging from 0 = *never* to 4 = *nearly always*, and the total score ranges from 0 to 32. A higher score indicates a higher caregiver burden. For primary caregivers of children with SMID, the care-related burden is an essential component of continued home care (Dambi et al., 2016). This scale is suited to assessing the care-related burden of primary caregivers of patients in various situations.

2.4.5. QoL of primary family caregivers

The SF-8, whose reliability and validity have been verified, was developed to evaluate the general aspects of health-related QoL (Tokuda et al., 2009). It consists of eight items that provide two summary scores: a physical health component summary (PCS) and a mental health component summary (MCS). A higher score indicates higher QoL.

2.4.6. Family functioning

The FACESKGIV-16, whose reliability and validity have been verified, is an instrument for assessing

families (Ikeno et al., 1990). The FACESKGIV-16 has high internal consistency (Cronbach's alpha = 0.71–0.99). In this instrument, family functioning is evaluated through a combination of two dimensions: adaptability and cohesion. A balanced level of cohesion and adaptability indicates a functional family. The FACESKGIV-16 measures the function and relationships of each family member (Ikeno et al., 1990). Since the siblings of children with SMID need family care to continue their daily lives, the family relationships of siblings of children with SMID are very important.

2.4.7. Family empowerment

Koren, DeChillo, and Friesen (1992) define family empowerment as “the state or ability of families to cooperate with elements outside of their living scope by controlling their life for the purpose of raising a child with severe disabilities.” It can be assessed using their Family Empowerment Scale. The reliability and validity of the Japanese version of this scale were confirmed by Wakimizu, Fujioka, Furuya, Iejima, and Yoneyama (2010).

The J-FES consists of the following three equally important domains: family, service system, and social/political. The family domain represents the ability of family caregivers to raise children with SMID by themselves. The service system domain represents awareness of social services to assist with childrearing. The social/political domain represents the capability to encourage social action and change for the sake of other children with SMID. The J-FES consists of 34 items across the

abovementioned three subscales, e.g., “I make an effort to learn new methods to help my children grow and develop,” “I can work together with administrative bodies and experts, doctors, visiting nurses, and public health nurses to decide what service my children need,” and “I contact legislators when important bills or issues regarding my children are left unresolved.” Each item is scored using a five-point Likert scale. Higher scores indicate higher family empowerment. We used only the subscale scores for our analysis, not the total score of the J-FES. The J-FES is used to address problems relating to family empowerment (Wakimizu et al., 2010).

2.5. Data analysis

SPSS version 24.0 for Windows was used for data analysis. Descriptive statistics were computed for the sample attributes and scores on the instruments. We then conducted a logistic regression analysis to reveal related factors of siblings’ QoL. Before we performed the multiple regression analysis, the Spearman’s rank correlation coefficient was calculated to select the variables relevant to the KINDL^R scores. Statistically significant variables were used to perform a stepwise multiple regression analysis to identify the predictors of QoL. In the multiple regression, the variance inflation factor was used as an indicator of multicollinearity. The level of statistical significance was set at $p < .05$.

2.6. Ethical considerations

Teachers distributed the description of the purpose, method, and ethical considerations of this study to the parents of children with a disability. Only participants who gave their consent answered the questionnaire. The questionnaires were anonymous and returned in individual envelopes. We also explained that participation was voluntary and that there would be no disadvantage arising from refusal to participate or cancellation after providing written consent. This study was approved by the Institutional Review Board of T University (Approval No. 1004).

3. RESULTS

A total of 1,659 (a participation rate of 35.2%) out of 4,707 families from 89 special needs schools in Japan answered our questionnaires. Siblings outside the age criteria and those who did not answer the entire KINDL^R were excluded; thus, only the data collected from 789 siblings of children with SMID and their primary caregivers were used for our analysis.

Table 1 shows the attributes of the sample. The mean age of the siblings was 12.21 ± 3.07 years; 419 (53.1%) of them were 6–12 years old, and 361 (45.8%) were 13–18 years old. In total, 84 siblings had at least one chronic condition. About 70% of the primary caregivers were 30–39 years old and 93.2% were married. Over 50% of the primary caregivers were unemployed. Disabilities in most children with SMID were caused by cerebral palsy, periventricular leukomalacia, or chromosomal aberration. Regarding the severity scores of children with SMID, 187 (23.7%) were classified as sub-

SMID-MCDG, and 63 (8.0%) were classified as SMID-MCDG.

Table 2 shows the scores for the KINDL^R (which reports the QoL of siblings of children with SMID), the relationship with children with SMID, the primary caregiver's burden (J-ZBI_8), family empowerment (J-FES), family function (FACESKGIV-16), and the QoL of the primary family caregivers (SF-8). The affective well-being dimension showed the highest scores, and the self-esteem dimension the lowest scores for the QoL of siblings of children with SMID (KINDL^R). Regarding the relationship with children with SMID, for the item "I have a good relationship with my siblings," 72.7% of the respondents answered "often" or "all the time." The mean total score for family empowerment (J-FES) was 101.05 ± 17.11 points.

Table 3 shows the correlations for the QoL of siblings of children with SMID (KINDL^R) scores, the attributes of the sample, the primary caregiver's burden (J-ZBI_8), the family empowerment score (J-FES), the family functioning score (FACESKGIV-16), and the QoL of the primary family caregiver score (SF-8). Gender ($r = .07, p < .05$), birth order ($r = .11, p < .01$), having at least one chronic health condition ($r = .09, p < .01$), relationship with the child with SMID ($r = .37, p < .001$), the family domain of the J-FES ($r = .19, p < .001$), the service system domain of the J-FES ($r = .11, p < .01$), the social/political domain of family empowerment ($r = .08, p < .05$), the physical health component summary of the QoL of the primary family caregivers ($r = .09, p < .05$), and the mental health component summary of the QoL of the primary family caregivers ($r = .14, p < .001$) were statistically

significantly and positively correlated with the KINDL^R scores. The age of siblings ($r = -.20, p < .001$) and their J-ZBI_8 scores ($r = -.13, p < .001$) were statistically significantly and negatively correlated with the QoL of siblings of children with SMID (KINDL⁸) scores.

Table 4 shows the factors associated with the KINDL^R scores based on the findings of the multiple regression analysis. The following factors contributed to higher KINDL^R scores: a more positive relationship with children with SMID ($\beta = .38, p < .001$), the younger age of siblings ($\beta = -.15, p < .001$), lower J-ZBI_8 scores ($\beta = -.10, p < .05$), later birth order (i.e., younger siblings) ($\beta = .11, p < .05$), higher FA (J-FES) scores ($\beta = .10, p < .05$), and female gender of siblings ($\beta = .09, p < .05$) (adjusted $R^2 = 0.22, F(6, 480) = 23.82, p < .001$). Multicollinearity was not observed.

4. DISCUSSION

4.1. Characteristics of the participants

The total fertility rate in Japan is 1.43 (Ministry of Health, Labour and Welfare, 2018). Against the backdrop of a declining birthrate, in this study the average number of children in a family was higher than normal (2.75). Turning to the household income of the families, the average was 7.13 million yen (Ministry of Health, Labour and Welfare, 2017) in this study. The families were thus predominantly from the lower-income group, with 72.3% having an annual income of less than 7 million yen (Approximately US 6,396 dollars). The employment rate of women raising children who are between

the ages of 7–17 in Japan is between 66.4% to 73.1% (Ministry of Health, Labour and Welfare, 2017); thus, the employment rate of the primary caregivers in this study was much lower despite many of the women having children of a similar age. It is presumed that this led to lower household incomes than in ordinary households, since in families raising children with SMID, the employment of the primary caregiver is restricted due to the care needs of the children. Finally, with respect to the severity score of children with SMID in this study, the need for medical care was considered low overall, since 68.3% of the children scored under 10 points.

4.1.1. QoL scores of siblings of children with SMID and their trends

Several previous studies have reported that QoL tends to be low in siblings of children with a disease or disorder (Bowman et al., 2014; Marciano & Scheuer, 2005; Rana & Mishra, 2015). This study was undertaken under the assumption that similar results would be obtained for siblings of children with SMID.

In a survey of healthy elementary school students in Japan, the mean KINDL^R score was 64.7 points (Shibata et al., 2003); in a study of healthy junior high school students, it was 60.9 points (Matsuzaki et al., 2007); and for elementary school students with mild developmental disabilities, it was 59.3 points (Furusho et al., 2006). However, the QoL scores of the siblings of children with SMID in this study were higher than those reported in these studies. Moreover, as in this study, a previous study reported high QoL of siblings of children with cystic fibrosis (Havermans et al., 2011). Most cystic

fibrosis cases are diagnosed through neonatal screening. Considering the commonalities between cystic fibrosis and the disabilities examined in the current study, we can infer that, as most of the children were known to have a disability since birth, their families had dealt with it over a long period of time. Unlike the onset of a sudden illness in a healthy child, family cohesion in the face of a disease and sufficient time to adapt to life with a sick child can be considered as likely reasons why the existence of a child with SMID did not result in a decline in the QoL of his or her siblings.

Moreover, since it has been reported that the presence of people with disabilities strengthens cohesion among family members and often encourages self-growth (Takano & Okamoto, 2011), it can be presumed that the presence of children with SMID does not necessarily have a negative influence on any siblings. However, the present results should be generalized carefully in view of research participation bias. In other words, it is likely that the QoL score was high because the siblings likely to respond would tend to have a positive perception of children with SMID, the relationships among family members, and their life together.

In a previous study, self-esteem scores showed a higher trend in siblings of children with SMID than in healthy elementary and junior high school students (Matsuzaki et al., 2007; Shibata et al., 2003). It is likely that because they were living with siblings with SMID, the children unconsciously compared themselves to the children with SMID, which afforded them the opportunity to realize that health is of particular value. This may explain the siblings' higher self-esteem scores.

4.2. Factors affecting the QoL of siblings of children with SMID

The results of this study indicated that the later birth order of siblings (i.e., younger siblings) is related to a higher QoL score, which supports the results of a previous study (Limbers & Skipper, 2014). Moreover, the results indicated a trend of higher QoL scores for lower ages (more so in girls than in boys), in agreement with previous studies (Limbers & Skipper, 2014; Matsuzaki et al., 2007; Shibata et al., 2003). It is likely that the parents do not talk much to younger siblings or those born later about the disease and the future of the child with SMID. Vermaes, van Susante, and van Bakel (2012) also suggested that parents may be less likely to disclose detailed information about the child's illness to younger siblings. It could be that less exposure to stressors related to the diseases and disorders of children with SMID in the family may have led to a higher QoL score. Indeed, younger siblings may have less capacity to notice stressors within the family caused by the diagnosis of a chronic health condition.

Relationships with children with SMID had the greatest influence on siblings' QoL in this study. Similar results have been reported in previous studies of patients and children with mental illness (Smith & Greenberg, 2007). Previous studies have also reported that siblings of children with chronic illnesses were relied on to assume their parents' responsibility for their siblings (Abe & Kawasumi, 2015). It is most likely that, having a good relationship with them, these children are actively involved in the care of a sibling with SMID. It is thus likely that the perception of playing an important role in

the family resulted in improved QoL scores. These findings suggest that it is important for healthcare professionals to positively recognize that their siblings' age, birth order, and relationships with children with SMIDs are relevant to the quality of life of their siblings, and to support them as needed. It is important that healthcare professionals help these parents raise not only children with disabilities but also children with disabilities, including siblings. Specifically, it is necessary to help these parents, depending on the age and stage of development of their siblings, to spend time explaining or interacting with their siblings' children with disabilities.

Moreover, it was observed that QoL was also affected by the attributes of the primary caregiver in addition to those of the siblings themselves. Although no previous study reported that the nursing care burden experienced by the primary caregiver affects siblings' QoL, it was presumed that this may be due to a lack of psychological leeway to direct attention to the siblings as the nursing care burden increases. To prevent overburdening the primary caregiver, healthcare professionals should remain vigilant and facilitate cooperation among family members. Additionally, healthcare professionals are required to perceive the siblings as well as the primary caregiver as subjects for support and provide appropriate assistance based on an assessment of the care burden and living conditions of the whole family.

This study revealed that the higher score for the family domain of the FES was related to higher QoL scores of siblings. The family domain of the FES is an indicator of the ability of a family to

respond to the problems involved in caring for children with disabilities at home (Koren, DeChillo, & Friesen, 1992). It is presumed that if the primary caregiver of a child with SMID can handle the problems involved in rearing a child with a disorder at home, they can also respond well to the problems of siblings as needed. Parents play a core role in supporting the stable mental functioning of their children (Shibata, Nemoto, Matuzaki, & Itabashi, 2013). These results therefore suggest that as the “family” is empowered, the siblings’ QoL improves, and, based on this observation, it is likely that support from healthcare professionals to improve family empowerment may also be effective in improving siblings’ QoL.

4.3. Limitations of this study

There were several limitations to this study. First, since the coefficient of determination of the model of factors affecting the QoL of siblings of children with SMID was .22, it cannot be concluded that the factors of the QoL of siblings were sufficiently elucidated. Other variables such as studies, club activities, and friendships may be assumed as important explanatory variables for sibling QOL. However, we considered the siblings as a member of a family caring for a child with severe motor and intellectual disabilities, and could not include these variables in the questionnaire. Second, because we used a cross-sectional survey design, we can only report on associations and not causality. Finally, the survey participation rate was 35.2% in this study. We therefore cannot rule out the possibility of selection bias.

Several previous studies reported that QoL is affected by the coping behaviors of siblings (Rana & Mishra, 2015; Vieira & Fernandes, 2013). We therefore deem it necessary for future studies to consider this behavior as a factor. It may be difficult to measure the QoL of siblings only in the context of caring for and living with children with SMID. As the siblings grow up, studies and club activities at school and relationships with friends become increasingly significant, especially in school-aged children. Such factors may affect the QoL of siblings of children with SMID. The relationships between individuals and family members' QoL appear to be quite complex, and such complexity needs to be clarified in future research.

5. CONCLUSION

The QoL of siblings of children with SMID was observed as showing a higher trend than that reported in previous studies with participants of a similar age. It was found that the factors affecting siblings' QoL were the relationships with children with SMID, the age of the sibling, the care burden of the primary caregiver, the score on the FA subscale of the FES, and the gender of the sibling. However, it cannot be concluded that the factors of siblings' QoL were sufficiently elucidated in this study, and therefore the coping behaviors of siblings should be considered in future studies.

Healthcare professionals need to support siblings during nursing visits. Moreover, our results suggested that positively supporting siblings' perceptions of their relationships with children with

SMID, preventing the overburdening of the primary caregiver, and providing appropriate assistance related to the care of children with SMID based on assessments of the whole family's care burden and living conditions would help improve siblings' QoL.

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CONTRIBUTIONS

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Table 1. Attributes of the sample ($n = 789$)

	$n/\text{Mean} \pm SD$	%/range
Siblings		
Current age (years)	12.21 ± 3.07	6–18
6–12	419	53.11
13–18	361	45.75
No answer	9	1.14
Gender		
Male	365	46.26
Female	413	52.34
No answer	11	1.39
Number of siblings	1.75 ± 0.81	1–6
Birth order		
First	364	46.13
Second	292	37.01
Third	101	12.80
Fourth	23	2.92
Fifth	2	0.25
No answer	7	0.89
At least one chronic health condition		
Yes	84	10.65
No	705	89.35
Diagnosis (multiple answers allowed)		
Atopic dermatitis	34	4.31
Asthma	31	3.93
Allergic rhinitis	7	0.89
Heart disease	4	0.51
Kidney disease	3	0.38
Attention deficit hyperactivity disorder	2	0.25
Nettle rash hives	2	0.25
Epilepsy	2	0.25
Cleft lip and cleft palate	2	0.25
Type I diabetes mellitus	1	0.13
Pervasive developmental disorder	1	0.13
Turner syndrome	1	0.13
Hydronephrosis	1	0.13
Congenital myopathy	1	0.13
Acute Lymphocytic Leukemia	1	0.13
Spina bifida	1	0.13

1	Primary caregiver		
2	Age (years)		
3	20–29	5	0.63
4	30–39	172	21.80
5	40–49	541	68.57
6	50–59	58	7.35
7	60–	3	0.38
8	No answer	10	1.27
9	Gender		
10	Male	60	7.60
11	Female	717	90.87
12	No answer	12	1.52
13	Education		
14	Junior high school	18	2.28
15	High school	313	39.67
16	Junior college	84	10.65
17	College	205	25.98
18	University	129	16.35
19	Graduate school	8	1.01
20	Other	21	2.66
21	No answer	11	1.39
22	Marital status		
23	Married	724	91.76
24	Single	53	6.72
25	No answer	12	1.52
26	Employment		
27	Not working	404	51.20
28	Part-time	233	29.53
29	Full-time	129	16.35
30	No answer	23	2.92
31	Children with SMID		
32	Age	11.84 ± 3.35	6–18
33	Age of onset	1.08 ± 2.50	0–15
34	Severity score	11.32 ± 7.08	6–44
35	Years of home care		
36	Under 1 year	16	2.03
37	Over 1 and less than 2 years	19	2.41
38	Over 2 and less than 5 years	43	5.45
39	Over 5 and less than 10 years	227	28.77

1 Over 10 years 371 47.02

2 No answer 113 14.32

3 **Family**

4 Household income per year (million yen) †

5 Under 3 million yen 117 14.83

6 3–5 million yen 239 30.29

7 5–7 million yen 214 27.12

8 7–10 million yen 111 14.07

9 10–13 million yen 39 4.94

10 Over 13 million yen 16 2.03

11 No answer 53 6.72

12 †1 USD = 122.87 yen (at the time of the investigation); *SD* = standard deviation.

"For Peer Review"

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Table 2. Current status of KINDL, relationship with children with SMID, J-ZBI_8, J-FES, FACESKGIV-16, and SF-8

	No. of items	Mean \pm SD	Score Range	Range
Total scores of KINDL ^R	24	69.63 \pm 12.55	0–100	20.83–96.88
Physical health	4	76.52 \pm 18.39	0–100	0–100
Affective well-being	4	80.96 \pm 16.76	0–100	0–100
Self-esteem	4	51.12 \pm 24.80	0–100	0–100
Family	4	75.30 \pm 17.55	0–100	6.25–100
Friends	4	78.06 \pm 17.73	0–100	0–100

School	4	57.02 ± 15.33	0–100	6.25–100
Disease module†	6	76.19 ± 16.56	0–100	0–100
Relationship with children with SMID	3	10.78 ± 6.18	3–15	3–15
J-ZBI_8	8	8.33 ± 6.24	0–32	0–32
FA (J-FES)	12	37.44 ± 6.89	12–60	14–57
SS (J-FES)	12	39.42 ± 6.93	12–60	16–58
SP (J-FES)	10	24.20 ± 5.53	10–50	10–47
Adaptability (FACESKGIV-16)	8	−0.73 ± 2.30	−8.0–8.0	−8.0–7.5
Cohesion (FACESKGIV-16)	8	3.50 ± 2.93	−8.0–8.0	−7.0–8.0
PCS (SF-8)	8	46.70 ± 7.53	0–100	18.70–66.20
MCS (SF-8)	8	46.00 ± 8.26	0–100	17.66–61.78

†Siblings with at least one chronic condition (84 children) answered the disease module; *SD* = standard deviation; KINDL measures QoL of siblings of children with SMID; J-ZBI_8 measures the primary caregiver's perceived burden of providing family care; J-FES measures the empowerment of the family; FA = family subscale; SS = service system subscale; SP = social/political subscale; FACESKGIV-16 measures family functioning; SF-8 measures health-related QoL in the primary caregiver; PCS = physical health component summary score; MCS = mental health component summary score.

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Table 3. Correlations for KINDL scores, attributes of the sample, J-ZBI_8, J-FES, FACESKGIV-16, and SF-8.

	ρ
Siblings	
Gender (1: Boy, 2: Girl)	0.07*
Age	−0.20***
Number of siblings	−0.01
Birth order	0.11**
At least one chronic health condition (1: Yes, 2: No)	0.09**
Relationship with children with SMID	0.37***
Primary caregiver	
Gender (1: Male, 2: Female)	0.00
Age	−0.02
Employment (1: Not working, 2: Part-time, 3: Full-time)	−0.04
Marital status (1: Married, 2: Single)	0.03
J-ZBI_8	−0.13***
FA (J-FES)	0.19***
SS (J-FES)	0.11**
SP (J-FES)	0.08*
Adaptability (FACESKG4-16)	0.01
Cohesion (FACESKG4-16)	0.08
PCS (SF-8)	0.09*
MCS (SF-8)	0.14***
Children with SMID	
Age	0.02
Age of onset	0.00
Severity score	0.06
Years of home care	0.07
Family	
Household income per year	0.03

*: $p < 0.05$, **: $p < 0.01$, ***: $p < 0.001$; ρ = the Spearman’s rank correlation coefficient with total scores of KINDL; KINDL measures QoL of siblings of children with SMID; J-ZBI_8 measures the primary caregiver's perceived burden of providing family care; J-FES measures family empowerment; FA = family subscale; SS = service system subscale; SP = social/political subscale; FACESKGIV-16 measures family function; SF-8 measures health-related QoL in primary caregiver; PCS = physical health component summary score; MCS = mental health component summary score.

Table 4. Associated factors of KINDL scores with siblings' QoL

	<i>b</i>	<i>SE</i>	β	<i>t</i>
Relationship with children with SMID	0.18	0.02	0.38	9.15***
Age of siblings	-0.64	0.18	-0.15	-3.44***
J-ZBI_8	-0.21	0.09	-0.10	-2.44*
Birth order of siblings	1.72	0.70	0.11	2.46*
FA (J-FES)	0.20	0.08	0.10	2.45*
Gender of siblings (1: Male, 2: Female)	2.35	1.05	0.09	2.24*
Adjusted R^2	0.22			
$F(6, 480)$	23.82***			

*: $p < 0.05$, ***: $p < 0.001$; A stepwise multiple regression analysis was conducted; KINDL measures QoL of siblings of children with SMID; J-ZBI_8 measures the primary caregiver's perceived burden of providing family care; J-FES measures family empowerment; FA = family subscale; Adjusted R^2 = adjusted coefficient of determination; b = partial regression coefficient; SE = standard error; β = standard partial regression coefficient.