

BRIEF REPORT

Resilience and difficulties of parents of children with a cleft lip and palate

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Abstract

Aim: The present study explored the differences in emotional difficulties and resilience between fathers and mothers of a child with a cleft lip and palate.**Methods:** Married couples were recruited who were accompanying their child with a cleft lip and palate (<12 years old) on regular visits to an outpatient clinic in a Japanese hospital. The participants were distributed an anonymous questionnaire that included items regarding emotional difficulties with the cleft lip and palate and a scale to measure resilience when caring for the children. In the data analysis, the paired *t*-test was used to compare the individuals within the couples.**Results:** By analyzing the data of 64 couples who provided valid responses, two items with the highest mean score for the difficulties that were faced by both the father and mother of a child with a cleft lip and palate were: “I am worried about whether the child’s teeth will be straightened” and “I am worried that the children could suffer due to their appearance.” The mothers felt significantly more worry about their child’s future and more guilt than did the fathers. In contrast, the fathers had greater resilience, in terms of problem-solving skills and recognition and acceptance than did the mothers.**Conclusion:** It is important that healthcare providers understand the difference between the fathers’ and mothers’ worries about their child with a cleft lip and palate. Specific support services should be offered to fathers with a high level of resilience, in terms of the acceptance of reality and problem-solving, which could increase their parenting ability.**Key words:** cleft lip, cleft palate, cross-sectional study, parents, resilience.

INTRODUCTION

Cleft lip and/or palate is the most common congenital craniofacial anomaly that is caused by abnormal facial development during gestation (Dixon, Marazita, Beaty, & Murray, 2011), accounting for about half of all craniofacial anomalies (Mossey & Modell, 2012). According to a global epidemiological survey (International Perinatal Database of Typical Oral Clefts Working Group, 2011),

the overall prevalence of cleft lip and/or palate is 9.92 per 10,000 births. Japan has the statistically highest prevalence (20.04 per 10,000 births), whereas countries in Europe and Africa have a lower prevalence (International Perinatal Database of Typical Oral Clefts Working Group). In particular, about half of the cases have a cleft lip and palate (CLP).

Although there are standardized treatment strategies and children rarely die due to a CLP, parents experience a significant psychological impact at the time of the child’s birth because a CLP indicates a dysplasia of the face, as well as dysfunction of the oral cavity (Fukuda, Goto, Wade, & Miyazaki, 1981).

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Additionally, children with a CLP are required to deal with tasks regarding breastfeeding, speaking, and/or dentition during development. Therefore, the parents of a child with a CLP experience difficulties, including worry, guilt, conflict, and anxiety, about continued therapy for each aspect of child development (Nelson, Kirk, Caress, & Glenny, 2012; Nitta, Fujiwara, & Ishii, 2012). This emotional distress can have long-term adverse effects on the parents' mental health, family functioning, and the adjustment of the sick child. These parents must resolve the difficulties that are faced when rearing children with a CLP. Therefore, healthcare professionals must consider how to support parents in dealing with this adversity.

In the current study, the focus is on resilience, which is a process in which persons adapt successfully when they encounter any stressors due to an adverse challenge, trauma, tragedy, threat, or serious health problem (Grotberg, 1999). The resilience of Chinese parents who have a child with a congenital disease is positively correlated with their personal strength, appreciation of life, and self-efficacy, but negatively correlated with post-traumatic stress symptoms (Li, Cao, Cao, Wang, & Cui, 2012). Moreover, lower resilience among the parents of children with cancer has been accompanied by higher distress levels, less social support, and weakened family functions (Rosenberg *et al.*, 2014). The Japanese parents of children undergoing surgery within 1 week after birth felt positive emotions when trying to cope with any of the events that resulted from the child's disease (Yamauchi, Nakayama, & Okamoto, 2016).

Thus, studies that explore the resilience that brings a satisfactory result for parents who experience traumatic stress due to having a child with a congenital disease are meaningful. However, most of the studies that have examined parents' psychological stress have focused mainly on the mother (Nelson, Glenny, Kirk, & Caress, 2012). As fathers' and mothers' parenting roles are different to each other, it is important to consider the parenting and treatment of both the father and the mother of a child with a CLP while they share and compensate for each other's strengths and weaknesses. In order to display family dynamics fully, examining the power balance between fathers and mothers can help to consider better ways of supporting family function. Therefore, the present study was designed to explore the differences in the difficulties and resilience of the fathers and mothers of children with a CLP.

METHODS

A self-administered cross-sectional questionnaire was used. The participants were married fathers and mothers who were accompanying their child with a CLP on regular visits to an outpatient clinic of a Japanese hospital. This hospital has an outpatient clinic that is mainly for cleft lip and/or palate and accepts a great number of patients from throughout Japan. The inclusion criteria were that the child had finished cheiloplasty that was conducted initially for a CLP and the child was <12 years old. The exclusion criterion was that the child had any other chronic or congenital disease (e.g. Down syndrome). The data were collected between October, 2015 and February, 2016.

The chief nurse informed the first author about the participants who were coming to the clinic who met the inclusion criteria. The chief nurse wrote down the parents' names to ensure that they did not respond to the questionnaire twice. The first author made contact with them in the waiting area, provided written information about the study, and asked them to complete the anonymous questionnaire. The first author conducted this task on the request of the last author, who headed the study. The completed questionnaires were posted in a lockbox in the waiting area or sent by mail within 1 month. If the participants did not visit as a couple, the first author was asked to give the questionnaire to one person in the couple. It was determined that an appropriate sample size was ~60, based on research that examined differences in resilience between 57 couples with normal birthweight infants (Nagatomi & Hohashi, 2015).

The questionnaire included items about the parents' and child's demographic characteristics, difficulties with a CLP, and the Scale to Measure Resilience in Child Care (SMRCC; Miyano, Fujimoto, Yamada, & Fujiwara, 2014). The difficulties with a CLP were assessed with 12 unique items that were based on the results of the authors' prior qualitative research that clarified the difficulties of parents with children with a CLP (Nitta *et al.*, 2012). The results showed that the parents encountered worries, concerns, and anxiety about parenting the child with a CLP. For validity, the co-authors reviewed and discussed these items in order to reach a consensus regarding their use. Each item was rated on a 5 point Likert-style scale that ranged from 1 ("strongly disagree") to 5 ("strongly agree"). The SMRCC is a validated self-report measure of the parent's resilience in terms of the parent adapting successfully when encountering difficult situations while caring for the child

(Miyano *et al.*). The scale consists of 36 items and three subscales: support from surrounding people (e.g. “I have a supporter whom I can count on, no matter what”), problem-solving skills (e.g. “I can cope with anything by using my experience”), and recognition and acceptance (e.g. “I think I can manage some difficulty if I face it”). Each item’s score ranges from 0 (“strongly disagree”) to 6 (“strongly agree”). A higher score indicates greater resilience. In this research, the Cronbach’s alpha of the SMRCC was 0.94.

The data analysis was conducted by using the paired *t*-test to clarify the differences in each of the items that assessed the difficulties with a CLP and the SMRCC score between the individuals in the couple. A *P*-value of <0.05 indicated statistical significance. The statistical analyses were carried out by using IBM SPSS Statistics for Windows v. 20.0 (IBM Corporation, Tokyo, Japan).

Prior to the data collection, approval was obtained from the institutional review board of the research hospital. All the participants were given an anonymous questionnaire and a participant information sheet that stated the purpose and method of the study, the privacy protection, and that participation was free with no penalty for non-participation. The parents who responded to the anonymous questionnaire were considered to

have provided their informed consent and to be the study’s participants.

RESULTS

The questionnaire was distributed to 235 couples; 64 (27.2%) provided valid responses. The mean age of the mothers was 36.8 years (standard deviation [SD] = 5.6), the mean age of the fathers was 38.5 years (SD = 5.9), and the mean age of the children was 4.4 years (SD = 3.7). The frequency distribution of the children’s ages were: infant (0–1 year): 20; toddler (2–6 years): 25; and school-aged child (7–12 years): 19. Of the parents, 42.2% of the mothers and 96.9% of the fathers were employed.

The top two items with the highest mean scores regarding difficulties for both the father and the mother of a child with a CLP were: “I am worried about whether the child’s teeth will be straightened” and “I am worried that the children could suffer due to their appearance.” When comparing each difficulty item between the individuals in a couple (Table 1), the mothers’ scores for “I am worried that finding a job or employment could be affected for the child in the

Table 1 Comparison between the emotional difficulties of the fathers and mothers of a child with a cleft lip and palate (CLP)

Difficulty	N	Mother		Father		<i>t</i> value	<i>P</i> -value	95% CI	
		Mean	SD	Mean	SD			Lower	Upper
I am worried that the child’s appearance will not be beautiful	62	4.31	1.07	4.19	1.24	0.56	0.581	−0.52	0.29
I am worried about whether the child’s teeth will be straightened	62	4.48	1.01	4.48	0.91	0.00	1.000	−0.34	0.34
I am concerned about my child being able to speak well	62	4.00	1.32	3.90	1.41	0.56	0.578	−0.44	0.25
I am worried that the children could suffer due to their appearance	62	4.48	0.98	4.44	0.85	0.31	0.759	−0.36	0.26
I am concerned about my child being bullied	61	4.33	1.14	4.15	1.13	1.06	0.291	−0.52	0.16
I am worried that finding a job or employment could the child in the future	62	3.35	1.40	2.89	1.50	2.08	0.042*	−0.92	−0.02
I am worried that marrying or giving birth could be affected	61	4.43	0.86	3.84	1.40	2.91	0.005*	−1.00	−0.18
I think that the child’s disease is my fault	61	3.69	1.32	2.51	1.28	6.10	<0.001*	−1.57	−0.79
I am sorry for my child	61	3.87	1.42	3.11	1.42	3.30	0.002*	−1.21	−0.30
I would like to keep my child unclear about the CLP	61	1.30	0.55	1.18	0.50	1.36	0.180	−0.28	0.05
I do not know the appropriate time to notify my child of their CLP	62	2.39	1.40	2.19	1.34	0.97	0.338	−0.59	0.21
I do not know how to talk about the CLP with my child	62	2.56	1.45	2.24	1.38	1.48	0.144	−0.76	0.11

*Significant difference between the fathers and mothers at the 0.05 level.

This sample size was smaller than the number of participants because of missing data.

CI, confidence interval; SD, standard deviation.

future,” “I am worried that marrying or giving birth could be affected,” “I think that the child's disease is my fault,” and “I am sorry for my child” were higher than those of the fathers' scores ($P < 0.05$). The fathers' scores did not significantly exceed the mothers' scores for any item.

There was no significant difference in the total score of the SMRCC between the individuals in a couple. The scores of two of the three subscales, “problem-solving skills” and “recognition and acceptance,” were significantly higher for the fathers than for the mothers ($P < 0.05$) (Table 2).

DISCUSSION

In the present study, the mothers felt significantly more guilt, such as reporting “I think that the child's disease is my fault,” and experienced significantly more worry about their child's future, such as indicating “I am worried that finding a job or employment could be affected for the child in the future,” than did the fathers. These findings are in line with those of previous research. A cleft-specific questionnaire has revealed that parents born with a cleft lip and/or palate reported feelings of guilt significantly more often than did parents without a cleft lip and/or palate (O'Hanlon, Camic, & Shearer, 2012). Another report also suggested that the mothers of children with congenital heart disease felt more guilt than did the fathers; for example, they stated “I am sorry for my child” or “I think that it is due to myself that the child has the disease” (Shiraishi, Matsuura, & Yamagata, 2006). Furthermore, the mothers' experience of feeling guilty might influence their child's mind or behavior; for instance, children with a CLP have reported feeling that their mother experienced guilt and thus they have taken care of their mother (Matsuda, Nakanii, Nishio, & Kogo, 2016).

All the mothers of children with a CLP have felt a certain degree of guilt (Hirose, 1999; Takahashi, 2003).

Therefore, the healthcare provider needs to support these mothers in order to enable them to put a positive spin on their parenting. There are some parenting programs for the parents of a child with a disease who have negative feelings, such as parent–child interaction therapy (Thomas & Zimmer-Gembeck, 2012) and the Stepping Stones Triple P Positive Parenting Program (De Graaf, Speetjens, Smit, De Wolff, & Tavecchio, 2008). The Stepping Stones Triple P program is a participatory program for parents to learn specific skills, such as ways to relate to the child, ways to cope, and ways of thinking, which is used worldwide. The Stepping Stones Triple P program is effective for the parents of children with a disability regarding the parents' depression, parental relationship, and child behavior observations (Tellegen & Sanders, 2013). Another study reported that the Stepping Stones Triple P program has short- and long-term effects on the well-being of the child, parents, and family (Sanders, Kirby, Tellegen, & Day, 2014). Thus, these parenting programs could contribute to mothers finding ways out of their psychological crisis.

In contrast to the mothers in a psychological crisis, the fathers had greater resilience in terms of problem-solving skills and recognition and acceptance. The mean resilience score of the mothers was lower and that of the fathers was higher in this study than in prior research, wherein the mean resilience score of the mothers of a child without disease was 111 (Miyano *et al.*, 2014). A recent study reported that the fathers of a preschool-aged child with a normal birthweight (Nagatomi & Hohashi, 2015) and those with a child having ongoing treatment for cancer (Shi *et al.*, 2017) had higher resilience than the mothers with such children. The fathers with a high level of resilience, in terms of the acceptance of reality and problem-solving, should be offered specific support, such as being provided with knowledge about the child's disease or enabling them to acquire care skills, which would increase their parenting ability.

Table 2 Comparison between the fathers' and mothers' scores in the three subscales of the scale to measure resilience in child care

Variable	N	Mother		Father		<i>t</i> -value	<i>P</i> -value	95% CI	
		Mean	SD	Mean	SD			Lower	Upper
Total	51	110.86	19.48	116.82	15.03	−1.97	0.054	−0.10	12.03
Support from surrounding persons	55	41.13	9.42	40.67	7.98	0.32	0.746	−3.26	2.35
Problem-solving skills	53	44.94	8.41	48.21	6.50	−2.41	0.020*	0.54	5.99
Recognition and acceptance	53	23.89	5.81	26.89	4.89	−3.19	0.002*	1.11	4.89

* Significant difference between the fathers and mothers at the 0.05 level. This sample size was smaller than the number of participants because of missing data.

CI, confidence interval; SD, standard deviation.

A limitation of the present study was that other factors that could affect parental perception of the parenting of a child with a CLP were not considered, such as support from others. For instance, one study found that the parents of a child with a CLP experienced less of a negative impact on the family, lower psychological distress, and better adjustment by having more support from friends and family members (Baker, Owens, Stern, & Willmot, 2009). In addition, it has been demonstrated that the parents of a child with a CLP require a doctor who is competent and reliable and who has effective communication skills in order for them to think positively (Nelson & Kirk, 2013). Therefore, family, friends, and doctors are significant persons for these parents and a patients' association or the establishment of a good relationship with healthcare providers are needed.

The present study focused on the differences between fathers and mothers. To grasp the psychological state of parents with children with a CLP, the factors that were related to the difficulties with a CLP, as well as the resilience of the parents, needed to be explored. Furthermore, the items of difficulties with a CLP were used in this study, which have not yet been validated as a measure. Thus, a scale of difficulties with a CLP needs to be validated that grasps the psychology of the parents with children with a CLP more deeply.

CONCLUSION

The present study demonstrated that the mothers felt more guilt and worry about their child's future than did the fathers. In contrast, the fathers had a greater resilience in terms of problem-solving skills and recognition and acceptance than did the mothers. It is important that healthcare providers understand the difference between the fathers' and mothers' difficulties with their child with a CLP and provide support based on their needs while optimally using the fathers' and mothers' strengths.

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DISCLOSURE

The authors declare no conflict of interest.

AUTHOR CONTRIBUTIONS

C. F. generated and set up this study, reviewed the manuscript, and supervised the whole study process; S. U. contributed to the acquisition of the data, the analysis, the interpretation of the data, and writing the manuscript; Y. K. and M. I. set up the research environment and introduced the participants to S. U. for the acquisition of the data; K. N., Y. F., M. K., and E. M. reviewed the manuscript critically for important intellectual content. All the authors read and approved the final manuscript.

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