

Positive change and sense of coherence in Japanese mothers of children with congenital appearance malformation

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Abstract

The purpose of this study was to clarify the factors related to perceived positive change and sense of coherence in mothers ($n=293$) of children with cleft lip and palate. The strongest negative correlation with sense of coherence was stigmatizing words from others; there were positive correlations with social capital and perceived positive change. Multiple regression showed that to be aware of positive changes, mothers share their feelings with mothers in the same position, are rooted in a safe community and do not feel isolated in parenting. People who have experienced adversity may have the opportunity to enhance sense of coherence by obtaining perceived positive change.

Keywords

congenital disability, malformation, mother, positive change, sense of coherence, social capital, social support, adversarial growth

Introduction

Adversity is a difficult or unlucky situation or event, such as diseases and disasters, wars, accidents, abuses, unemployment, divorce and events, that can cause an intense stress reaction. People who face adversity, struggle to reconstitute the traumatized self and the world (Park and Helgeson, 2006). The meaning of the event and the feeling of self-growth experienced after suffering through a struggle are called adversarial growth (Nishi et al., 2010). Adversarial growth, stress-related growth, post-traumatic growth, resilience, positive change and so on have attracted attention in recent years, and related studies are accumulating (Linley and Joseph, 2004).

Antonovsky (1987) first conceptualized and standardized the concept of sense of coherence (SOC) in the salutogenic theory as successful coping ability in diverse and stressful situations. SOC is highly predictive of recovery from an adversity and facilitates an individual's use of various resources when faced with hardship. SOC focuses on salutary factors (i.e. health factors) described as 'salutogenesis'. Rather than focusing on pathogenesis, or a conventional medical line of questioning regarding the causes of disease, salutogenesis aims to shed light on what creates health. SOC has three components: comprehensibility, manageability and

meaningfulness. Comprehensibility is the degree to which events are perceived to be explicable, predictable and structured. Manageability is the degree to which one feels that one can cope. Meaningfulness is how much one feels that life makes sense and how worthy challenges are of investment and engagement. People high in SOC can deal with stress successfully even when experiencing adversity, as well as being able to respond to such experiences with personal growth (Antonovsky, 1996). SOC has been studied not only in Europe and the United States but also in Japan in recent years (Eriksson and Lindstrom, 2007; Lansimies et al., 2017).

Types of adversity that families may suffer from include disease and disability. Giving birth to handicapped children greatly impacts parents' lives. Mothers of children with a congenital disability may experience sudden unavoidable difficulties such as needing urgent medical care, as well as

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economic difficulties, and social distress (Aftyka et al., 2017). For mothers, ‘ordinary life and healthy child rearing’ become unrealistic, and they embrace the feeling of losing a healthy child, which they had been hoping for every day. In that sense, this is one kind of adversity that disrupts a mother’s normal life.

One of the most common morphological abnormalities affecting appearance in newborns in Japan is cleft lip and/or cleft palate (CLCP). The incidence in Japan is about 1/500, which is higher than 1/800 in Europe and the United States (Natsume et al., 2000). The genetic predisposing cause of CLCP is not clear; therefore, the mother may ask herself unreasonable questions such as ‘Why is my child like this?’ or ‘Why me?’ (Shinohara and Nakanii, 2003).

Research on the psychosocial aspects of CLCP in Japan has centred especially on mothers’ care, including the acceptance process of mothers (Nakanii et al., 2003), prenatal notification (Nakanii et al., 2005) and the timing of prenatal diagnosis. These studies focused mainly on the pathogenesis approach. However, in recent years, more reports have focused on the positive aspects of having children with disabilities (Markoulakis et al., 2012). For mothers of children with an intellectual disability, raising children with a disability provides mothers with positive experiences such as an increased sense of purpose, priorities, spirituality, tolerance and understanding, personal growth and strength, and expanded personal/social networks (Blacher and Baker, 2007). It is meaningful to pay attention to parents’ feelings of human growth and positive changes from having children with disabilities (Rajan and John, 2016). However, there are no studies focusing on positive changes in mothers of children with CLCP. Indeed, what kinds of things and experiences are related to positive change and what maintains mothers’ high SOC even after experiencing adversity have rarely been clarified. Clarifying these will lead to suggestions for concrete support for mothers of children with CLCP or disabilities. Furthermore, considering that adversity can happen to everyone, clarifying the relationship between positive changes and SOC will provide hints to support health for all people.

Focusing on positive change and SOC, this study aims to clarify how much and what kind of positive change mothers of children with CLCP experience and to clarify factors relevant to positive change and SOC. Based on the above, the relationship between positive change and SOC is considered, and ways of enhancing or maintaining SOC in mothers of children with cleft lip and palate and health support are suggested.

Method

Data collection

The study was conducted between July and September 2014. We targeted 514 mother members of a self-help

group (SHG) operated by parents of children with CLCP. We distributed anonymous self-administered questionnaires to members of the SHG by mail. We received 296 responses (response rate=57.9%). After excluding questionnaires from individuals that did not meet inclusion criteria (e.g. those answered by the father or other relatives), data from 293 questionnaires were analysed.

Variables

Demographic variables

Regarding the characteristics of the participants, data on each mother’s age, marriage status, educational background, employment, presence of chronic disease, standard of living (poor, somewhat poor, average, somewhat rich, rich) were collected. For children with CLCP, age, gender, disease type (cleft lip only, cleft palate only, and cleft lip and palate) and presence of complications were asked.

Mothers’ subjective experiences and receipt of support regarding rearing a child with CLCP

When creating a multi-item scale, there is a method of extracting items from interviews with target persons (John and John, 2007). Scale development was not the purpose of this research. However, to obtain suggestions for concrete support, we thought that it was important to conduct interviews and reflect on the participants’ experiences of the questionnaire. We interviewed 13 mothers, and items were extracted regarding ‘Mothers’ subjective experience in the child rearing process’ and ‘Receipt of support from medical staff’.

The content of ‘Mothers’ subjective experience in the child rearing process’ included nine items such as ‘My spouse understands my feelings well’, ‘Parents-in-law understand my feelings well’, ‘My parents (grandparents for children) understand my feelings well’, ‘I feel like I am raising and treating my child by myself’ (reverse item), ‘My spouse actively participates in treatment and care of my child with CLCP’, ‘There are unforgettably severe or stigmatic words regarding CLCP from surrounding people’ (reverse item), ‘There are people who are familiar with information about lip cleft palate near me’, ‘For me, there are people close by with whom I can share feelings about CLCP’ and ‘I think I will explain CLCP properly to my child’. Regarding ‘Receipt of support from medical staff’, we asked the following five items: ‘Medical staff listened to my feelings and story carefully’, ‘Medical staff responded with a polite attitude’, ‘Medical staff explained about CLCP to grandparents’, ‘Medical staff provided information about treatment of CLCP’ and ‘Regarding disability of the child, I was told by medical staff that “you should not be blamed”’. Each item is rated on a 5-point scale from 1 (Disagree) to 5 (Agree).

Perceived positive change

We used the perceived positive change (PPC) scale to evaluate maternal positive change and self-growth related to rearing a child with CLCP. The original PPC scale was developed for patients and families of HIV-infected haemophiliacs in Japan, and its internal consistency and construct validity were confirmed (Kumada, 2009). Kimura and Yamazaki (2014) modified this as an index to grasp positive and negative changes of parents of children with disability. According to Kimura, this assesses how parents perceive both positive and negative changes in their life, self and relationships with others following a child's diagnosis (Kimura and Yamazaki, 2014). The PPC scale includes 10 items such as 'Your psychological strength has (*weakened to become stronger*)', 'Your self-confidence to overcome life's challenges has (*decreased to increased*)', 'Have you acquired new purposes or joy in life?' (*not at all to quite a lot*), 'The desire to be useful to people and society has (*weakened to become stronger*)', 'Thoughts about everything: you have started to think as though everything is getting (*worse to better*)', 'Feelings about how every day passes by: every day (*has ceased to matter or is important*)', 'Your ties with your family have (*weakened to become stronger*)', 'Your ties with your friends have (*weakened to become stronger*)', 'Have you made new reliable friends and acquaintances because you had a child with a disability?' (*not at all to quite a lot*)', 'In your everyday life, you (*no longer pay attention to your health to have started to pay attention to your health*)'. Each item is rated on a 5-point scale from 1 to 5.

Social capital

We also used a subjective Social Capital (SC) Scale to assess existing resources. Lochner et al. (1999) pointed out that SC overlaps with the existing concepts of collective efficacy, psychological sense of community, neighbourhood cohesion and community competence. Based on these concepts, Togari (2006) developed six items in the subjective SC scale. A sample of SC items include 'Our neighbours are willing to help others who need support' (psychological sense of community) and 'The neighbourhood where I live is very safe' (collective efficacy). Each item is rated on a 5-point scale from 1 (*strongly disagree*) to 5 (*strongly agree*). A higher score indicates a higher level of SC.

SOC

We used the 5-point Japanese version of the SOC-13, which is a shortened version of the original scale (SOC-29). The SOC scale has been translated into almost 40 languages; in Japan, Togari and Yamazaki (2005) developed a 5-point Japanese version of the SOC-13 scale, and it has been

widely used in Japanese studies and comparative research. The SOC-13 includes the following three subscales: comprehensibility (five items), manageability (four items) and meaningfulness (four items). The Cronbach's alpha of this scale in this study was 0.86.

Statistical analysis

To clarify the subjective positive change (adversarial growth) and negative change experienced by mothers in raising children with CLCP, a simple tabulation of the PPC scale was performed. Following previous research, we tabulated negative changes in each item of the PPC scale, such as 'weakened' or 'weakened rather'. In addition, responses such as 'Becoming stronger' or 'Becoming stronger rather' were considered as positive changes, and 'Neither agree nor disagree' was added, responses were totalled into three groups and the distribution was observed by item. Next, to confirm the relationships between mothers' subjective experience, receipt of support from medical staff, SC, PPC, and the three components of SOC (manageability, comprehensibility, meaningfulness), correlation coefficients were calculated. Two items ('Medical staff provided information about treatment of CLCP' and 'Regarding disability of the child, I was told by medical staff that "you should not be blamed"') that did not correlate with SOC, SC and PPC were not included in the subsequent multiple regression analysis.

Next, hierarchical multiple regression analysis was performed using PPC and SOC as dependent variables. As covariates in two hierarchical multiple regression analyses, we input the age of the mother, the presence or absence of mother's chronic illness, employment, education, standard of living and the presence or absence of child complications. For PPC, covariates were entered as step 1, mother's experience and medical support as step 2 and SC was entered as step 3.

For SOC, covariates were entered as step 1, followed by mother's experience and medical support as step 2, SC as step 3 and PPC in the final model of step 4.

The statistical package SPSS 22.0 (IBM Japan, Ltd, Tokyo, Japan) was used for data analysis.

Ethical considerations

This study was conducted with the approval of Toho University School of Nursing Ethics Review Committee (approval no. 25022). When mailing, we enclosed a document explaining the purpose of the research along with the questionnaire. Additionally, this document explained that whether participants replied depended on their free choice, the SHG would not know whether participants replied, no disadvantage would occur even if they did not answer and that returning the survey form would be regarded as consent to participate.

Table 1. Demographic characteristics of the participants ($n=293$).

	<i>n</i>	%, SD
<i>Mother</i>		
Age, mean (SD), range: 28–73	49.9	8.9
Chronic disease		
Yes	61	20.8
No	232	79.2
Employment		
Regular employee	70	24.1
Non-regular staff, part-timer	129	44.3
Unemployed/homemaker	92	31.6
Education		
Junior/high school	82	28.1
Junior college, vocational school	127	43.5
University, graduate school	83	28.4
Marital status		
Currently not married	15	5.1
Married	278	94.9
Standard of living		
Poor	11	3.8
Somewhat poor	35	11.9
Average	104	35.5
Somewhat rich	113	38.6
Rich	30	10.2
SC, mean (SD), range: 5–30	22	4.0
SOC, mean (SD), range: 13–65	42.3	7.7
PPC, mean (SD), range: 5–50	38	5.7
<i>Child</i>		
Boy	162	55.7
Girl	129	44.3
Child age mean (SD)	18.7	9.4
CLCP type		
Cleft lip and palate	207	76.7
Cleft lip only	41	15.2
Cleft palate only	22	8.1
Complications yes	62	21.5

SD: standard deviation; SC: social capital; SOC: sense of coherence; PPC: perceived positive change; CLCP: cleft lip and/or cleft palate. Missing values are excluded from the table.

Results

Characteristics

Characteristics of participants are shown in Table 1. The average age of mothers was 49.9 ± 8.9 (standard deviation (SD)) years old, most were married (94.9%) and 20.8 per cent had a chronic illness. Regarding employment, 44.3 per cent of mothers had non-permanent employment such as part-time work, 24.1 per cent had regular employment and 31.6 per cent were unemployed. The average SOC score was 42.3 ± 7.7 , and the PPC score was 38.0 ± 5.7 . Regarding children's gender, 55.7 per cent were male, and the average child's age was 18.7 ± 9.4 years old. There were 76.7 per cent of children with both lip clefts and cleft palate, and 21.5 per cent with complications.

Regarding the PPC of mothers while raising children with CLCP (Table 2), 7 out of 10 items showed positive change in more than 50 per cent of respondents. There were no items for which the negative change exceeded the positive change. Among them, the most frequent positive change was 'psychological strength', as 82.0 per cent answered 'It was rather stronger' or 'became stronger'. In Table 3, regarding the subjective experience of the mother, the experience that one's spouse, real parents and parents-in-law understand one's feelings showed weak to moderate significant correlations with the three SOC subscales, SC and PPC (correlation coefficients $r = -0.342$ to 0.341). There was a tendency to show negative correlations with the sense of child rearing alone or stigma experience.

PPC, SOC and related variables

The results of hierarchical multiple regression analysis with PPC and SOC as dependent variables are shown in Tables 4 and 5. In step 1, covariates explained 8.4 per cent of the variance in PPC and 16.7 per cent of the variance in SOC. In step 2, 'Receipt of support from medical staff' and 'Mothers' subjective experience in the child rearing process' explained 27.3 per cent of the variance in PPC and 16.2 per cent of the variance in SOC. This was particularly the case for the relationship between 'For me, there are people close by with whom I can share feelings about CLCP' and PPC ($\beta = 0.226$, $p = 0.003$), and 'There are unforgettably severe or stigmatic words regarding CLCP from surrounding people' and SOC ($\beta = -0.223$, $p < 0.001$). In step 3, SC explained 4.5 per cent of the variance in PPC and 5.9 per cent of the variance in SOC. Finally, PPC explained 5.6 per cent of the variance in SOC. The change in all the R^2 values was statistically significant ($p < 0.001$).

As for PPC, the final model explained 35 per cent of the variance, while demographic characteristics showed no statistically significant associations with PPC. Regarding mothers' subjective experiences, 'For me, there are people close by I can share feelings about CLCP with', 'I think I will explain CLCP properly to my child', 'Medical staff explained about CLCP to grandparents' and SC were positively and significantly associated with PPC. Especially, SC was significantly associated ($\beta = 0.247$, $p = 0.001$). However, 'I feel like I am raising and treating a child by myself' ($\beta = -0.130$, $p = 0.041$) showed negative correlations with PPC.

The final model explained 44.3 per cent of the variance in SOC, and the hierarchical multiple regression results showed that SOC was associated with age ($\beta = 0.120$, $p = 0.044$), 'There are unforgettably severe or stigmatic words regarding CLCP from surrounding people' ($\beta = -0.223$, $p < 0.001$), SC ($\beta = 0.210$, $p = 0.001$) and PPC ($\beta = 0.305$, $p < 0.001$).

Table 2. Percentages of positive and negative change experienced by mothers of children with CLCP.

PPC items	Negative change		Neither negative nor positive		Positive change	
	<i>n</i>	(%) ^a	<i>n</i>	(%) ^a	<i>n</i>	(%) ^a
Psychological strength	11	(3.7)	42	(14.3)	241	(82.0)
Self-confidence to overcome life's challenges	8	(2.7)	63	(21.4)	223	(75.9)
New purposes or joy in life	22	(7.5)	83	(28.3)	188	(64.2)
The desire to be useful to people and society	9	(3.1)	81	(27.9)	200	(69.0)
Started to think as though everything is getting	22	(7.6)	126	(43.6)	141	(48.8)
Feelings about how every day passes by	5	(1.7)	75	(25.9)	210	(72.4)
Your ties with your family	8	(2.8)	65	(22.5)	216	(74.7)
Your ties with your friends	23	(8.0)	161	(55.9)	104	(36.1)
New reliable friends and acquaintances	32	(11.1)	118	(41.0)	138	(47.9)
Pay attention to your health	14	(4.8)	72	(24.8)	204	(70.3)

CLCP: cleft lip and/or cleft palate; PPC: perceived positive change.

^aMissing values were excluded.

Discussion

Factors related to mothers' PPC

The age range of the mothers who responded was broad (average age: 49.9 ± 8.9 (SD), age range: 28–73), and their average SOC score was slightly higher than the average score of people aged 50 years in a previous study (Togari and Yamazaki, 2005). Considering that the average age of participants was about 50 years old and that of the child was about 19 years old, it could be inferred that the treatment of CLCP had finished to a certain extent for respondents to this survey, their understanding of diseases and symptoms had progressed, and they were in good condition such as in terms of mental health.

For PPC, Kimura and Yamazaki (2014) showed that for parents of children with intellectual disability, the highest percentage of positive change experienced was for 'psychological strength'. Similarly, in our study, 82.0 per cent of respondents to the 'psychological strength' item answered, 'It was rather stronger' or 'became stronger'. There were four items for which positive change exceeded 70 per cent (apart from 'Your ties with your friends', for which 55.9 per cent answered 'Neither agree nor disagree', while 36.1 per cent indicated positive change). This result resembles that in a survey of families of HIV-infected people with phytotoxicity (Kumada, 2009) and in studies of parents of children with developmental disabilities (Kimura and Yamazaki, 2016). However, this does not mean that mothers can naturally feel positive change if they are raising disabled children. As Kimura and Yamazaki (2014) pointed out, we should focus on the fact that those who indicated negative change are currently suffering and consider how to support them.

As a factor related to PPC, there was an item 'Medical staff explained CLCP to grandparents'. The genetic predisposing cause of CLCP is not clear; however, words from parents-in law such as 'This child is not of our bloodline'

may hurt mothers of children with a disability (Nakanii et al., 2006). There are a certain number of people in Japan who emphasize the lineage of the family, and there are still elderly people who cannot accept congenital diseases appearing in their families (Nakanii et al., 2006). If the medical staff tell the grandparents that 'no-one is to blame for CLCP', the mother would feel greatly comforted, and this should be strongly encouraged. These are desirable sources of support.

Hierarchical multiple regression analysis showed no association of attributes/characteristics with positive change. To perceive positive change, it is important to receive emotional support from people such as the mother's parents and people in the same situation who can understand their feelings. However, 'I feel like I am raising and treating my child by myself' suggested that loneliness could hinder the perception of positive change. Participation in activities such as attending SHGs may help the mother.

There was also a positive association between PPC and the item 'I think I will explain CLCP properly to my child'. This study is cross-sectional, and causality cannot be specified; however, in a previous study (Omiya et al., 2012), children with CLCP were able to talk about their condition by finding out what it means to be born with CLCP. Likewise, future studies could explore whether mothers might be able to explain the condition properly to their children with CLCP when they perceive meaningfulness. Further studies using longitudinal research are needed to clarify possible causal relations between PPC and providing an explanation to children with CLCP.

Factors related to SOC

Hierarchical multiple regression analysis showed a significant association between age of the mother and SOC. This supports Antonovsky's theory that SOC becomes slightly higher with age. Additionally, stigma-like experiences, that

Table 3. Correlations among variables.

Variables	Sense of coherence				SC		PPC	
	Meaningfulness		Comprehensibility		Manageability			
	<i>r</i>		<i>r</i>		<i>r</i>		<i>r</i>	
Receipt of support from medical staff								
1. Medical staff listened to my feelings and story carefully	0.081		-0.048		-0.047		0.157*	0.282***
2. Medical staff responded with a polite attitude	0.106		-0.062		0.031		0.143*	0.242***
3. Medical staff explained about CLCP to grandparents	0.053		-0.006		-0.015		0.031	0.202**
4. Medical staff provided information about treatment of CLCP	0.011		-0.079		-0.018		0.082	0.074
5. Regarding disability of the child, I was told by medical staff that 'you should not be blamed'	0.011		-0.064		0.078		0.078	0.092
Mothers' subjective experiences								
1. My spouse understands my feelings well	0.224**		0.153*		0.198**		0.237***	0.229***
2. Parents-in-law understand my feelings well	0.256***		0.220**		0.341***		0.314***	0.224**
3. My parents (grandparents for children) understand my feelings well	0.238***		0.145*		0.228**		0.300***	0.317***
4. I feel like I am raising and treating my child by myself	-0.156*		-0.249***		-0.267***		-0.169*	-0.238***
5. My spouse actively participates in treatment and care of my child with CLCP	0.152*		0.090		0.128		0.168*	0.128
6. There are unforgettably severe or stigmatic words regarding CLCP from surrounding people	-0.152*		-0.262***		-0.342***		-0.151*	-0.059
7. There are people who are familiar with information about cleft lip/cleft palate near me	0.129		-0.009		-0.023		-0.017	0.197**
8. For me, there are people close by with whom I can share feelings about CLCP	0.282***		0.132*		0.076		0.147*	0.340***
9. I think I will explain CLCP properly to my child	0.190**		0.161**		0.103		0.112	0.248***

SC: social capital; PPC: perceived positive change; CLCP: cleft lip and/or cleft palate.

* $p < 0.05$; ** $p < 0.01$; *** $p < 0.001$.

Table 4. PPC and related factors ($n = 269$).

Variables	Upon entry		Final model		ΔR^2	
	Coefficient β	p value	Coefficient β	Standard error	95% CI	Wald χ^2 p value
Demographics of mothers						
Mother's age	0.039	0.561	0.100	0.043	-0.014 to 0.154	1.651 0.100
Chronic illness (0 = no, 1 = yes)	0.020	0.762	-0.034	0.777	-2.011 to 1.050	-0.619 0.537
Employment (0 = unemployed, 1 = employed)	0.084	0.189	0.071	0.685	-0.465 to 2.236	1.292 0.198
Education (1–3)	-0.015	0.818	-0.022	0.456	-1.075 to 0.722	-0.388 0.699
Standard of living (1 = poor to 5 = rich)	0.228	0.001	0.099	0.364	-0.126 to 1.308	1.625 0.106
Child's complications (0 = no, 1 = yes)	-0.126	0.054	-0.065	0.767	-2.405 to 0.620	-1.163 0.246
Receipt of support from medical staff						
Medical staff listened to my feelings and story carefully	0.284	0.000	0.105	0.310	-0.168 to 1.055	1.430 0.154
Medical staff responded with a polite attitude	0.224	0.000	0.029	0.312	-0.488 to 0.742	0.407 0.684
Medical staff explained about CLCP to grandparents	0.210	0.001	0.133	0.242	0.102 to 1.054	2.394 0.018
Mothers' subjective experience in the child rearing process						
My spouse understands my feelings well	0.225	0.001	0.145	0.484	-0.102 to 1.806	1.760 0.080
Parents-in-law understand my feelings well	0.190	0.003	-0.066	0.324	-0.942 to 0.335	-0.937 0.350
My parents (grandparents for children) understand my feelings well	0.281	0.000	0.144	0.405	0.071 to 1.666	2.146 0.033
I feel like I am raising and treating my child by myself	-0.218	0.001	-0.130	0.242	-0.976 to -0.021	-2.057 0.041
My spouse actively participates in treatment and care of my child with CLCP	0.119	0.067	-0.116	0.353	-1.226 to 0.164	-1.507 0.133
There are unforgettably severe or stigmatic words regarding CLCP from surrounding people	-0.047	0.467	0.083	0.214	-0.126 to 0.719	1.384 0.168
There are people who are familiar with information about cleft lip/cleft palate near me	0.209	0.001	0.004	0.276	-0.528 to 0.558	0.053 0.958
For me, there are people close by with whom I can share feelings about CLCP	0.342	0.000	0.226	0.282	0.297 to 1.409	3.025 0.003
I think I will explain CLCP properly to my child	0.239	0.000	0.164	0.337	0.316 to 1.644	2.908 0.004
Social capital	0.247	0.000	0.247	0.089	0.182 to 0.532	4.027 0.000
Total adjusted R^2					0.350	0.045

PPC: perceived positive change; CI: confidence interval; CLCP: cleft lip and/or cleft palate.

Education as 1 = junior high school and high school, 2 = junior college/vocational school, 3 = College graduates and above.

Standard of living was a continuous variable as 1 = 'poor' to 5 = 'rich'.

Table 5. SOC and related factors ($n = 269$).

Variables	Upon entry		Final model			χ^2	p value	ΔR^2
	Coefficient β	p value	Coefficient β	Standard error	95% CI			
Demographics of mothers								
Mother's age	0.206	0.002	0.120	0.058	0.003 to 0.230	2.022	0.044	0.167
Chronic illness (0 = no, 1 = yes)	-0.108	0.085	-0.073	1.052	-3.488 to 0.659	-1.345	0.180	
Employment (0 = unemployed, 1 = employed)	0.055	0.371	0.003	0.928	-1.782 to 1.876	0.051	0.960	
Education (1-3)	-0.035	0.576	-0.028	0.613	-1.516 to 0.902	-0.500	0.617	
Standard of living (1 = poor to 5 = rich)	0.249	0.000	0.079	0.496	-0.323 to 1.633	1.320	0.188	
Child's complications (0 = no, 1 = yes)	-0.166	0.008	-0.091	1.035	-3.775 to 0.307	-1.674	0.096	
Receipt of support from medical staff								
Medical staff listened to my feelings and story carefully	-0.011	0.859	-0.109	0.310	-1.467 to 0.190	-1.520	0.130	0.162
Medical staff responded with a polite attitude	-0.004	0.952	-0.108	0.312	-1.476 to 0.184	-1.534	0.127	
Medical staff explained about CLCP to grandparents	0.052	0.398	0.016	0.242	-0.555 to 0.746	0.289	0.773	
Mothers' subjective experience in the child rearing process								
My spouse understands my feelings well	0.183	0.004	0.016	0.657	-1.164 to 1.427	0.200	0.842	0.560
Parents-in-law understand my feelings well	0.250	0.000	0.077	0.437	-0.369 to 1.355	1.127	0.261	
My parents (grandparents for children) understand my feelings well	0.178	0.004	-0.025	0.551	-1.292 to 0.882	-0.372	0.710	
I feel like I am raising and treating my child by myself	-0.217	0.000	-0.061	0.331	-0.977 to 0.326	-0.985	0.326	
My spouse actively participates in treatment and care of my child with CLCP	0.113	0.069	-0.033	0.478	-1.155 to 0.731	-0.443	0.658	
There are unforgettably severe or stigmatic words regarding CLCP from surrounding people	-0.268	0.000	-0.223	0.290	-1.672 to -0.530	-3.800	0.000	
There are people who are familiar with information about cleft lip/cleft palate near me	0.058	0.347	-0.068	0.371	-1.090 to 0.374	-0.963	0.336	
For me, there are people close by with whom I can share feelings about CLCP	0.190	0.002	0.114	0.391	-0.170 to 1.371	1.536	0.126	
I think I will explain CLCP properly to my child	0.139	0.026	0.017	0.463	-0.773 to 1.051	0.301	0.764	
Social capital	0.284	0.000	0.210	0.124	0.177 to 0.666	3.395	0.001	
PPC	0.363	0.000	0.305	0.092	0.242 to 0.604	4.600	0.000	
Total adjusted R ²					0.443			

SOC: sense of coherence; CLCP: cleft lip and/or cleft palate; PPC: perceived positive change.
 Education as 1 = junior high school and high school, 2 = junior college/vocational school, 3 = College graduates and above.
 Standard of living was a continuous variable as 1 = 'poor' to 5 = 'rich'.

is, unforgettable and severe words from surrounding people, had a negative relationship with SOC. A heartless phrase to the mother may impair her SOC to such an extent that it becomes difficult to recover (Omiya et al., 2016). CLCP is an unexplained congenital condition, so, even if the mother is told of the stigma about the child with CLCP, only helplessness remains. Omiya mentioned the relationship between SOC decline in HIV-infected persons and the stigma experience, stating that experiencing stigma in a place where they expect to be emotionally supported, such as a hospital, evokes helplessness and despair. Means to support mothers who experienced stigma are strongly needed.

Relationships between SOC and PPC

Hierarchical multiple regression analysis showed a strong positive correlation between SOC and PPC ($\beta=0.305$, $p<0.001$). Taku (2014) studied the relationship between resilience, which is a similar concept to SOC, and perceived adversarial growth. She concluded that those with low resilience can increase resilience by awareness of adversarial growth. Positive change is a feeling of self-growth experienced after adversity, so it is difficult to adapt the concept of PPC to those who have not experienced adversity. In that sense, positive change is conceptually limited. However, positive change is not high or low by nature. Although Taku's findings suggested that self-growth can increase resilience, research has not yet explored the related possibility that SOC can be increased by supporting PPC. Antonovsky said of SOC that 'it stabilizes by about 30 years of age, and there is almost no drastic change after that' (Antonovsky, 1979). However, the results of this study suggest the hypothesis that increases in PPC could lead to increases in SOC, and this hypothesis should be tested.

Effect of SC on SOC and PPC

Our study showed that the relationships between SC and PPC, and between SC and SOC were significant ($\beta=0.247$, $p<0.001$, $\beta=0.210$, $p=0.001$). In a population survey of Norway, Maass et al. (2014) explored the relationship between neighbourhood SC, SOC and health, and found that neighbourhood SC was significantly related to SOC (the strongest health outcome coefficient). Mato and Tsukasaki (2017) reported that SOC was significantly related to cognitive SC, structural SC and self-efficacy, as well as mental health in university students in Japan. For women who raise children, it is extremely important to interact with the local environment and local people. Especially for parents raising disabled children, safe and unprejudiced areas are important (Kimura and Yamazaki, 2016). In this research, the results of previous research were also supported. A society where children with disabilities can easily live is a

society that is easy for everyone to live in. In the future, we need to examine what kind of relationship to the region is important.

There are several limitations in this research. The target group was members of a SHG with branches all over Japan. However, in Japan, fewer than 2000 people are born with CLCP per year. It is undeniable that the representation is poor. Furthermore, the SOC of the participants was slightly higher than the population average (Togari and Yamazaki, 2005); the average age of the participants was 49.9 years, and the age range was from 28 to 73 years old. Because treatment progresses according to the child's age, results may differ between mothers of infants and older mothers. Moreover, as participants were members of a SHG, there is a high probability that respondents had already received support and were mentally stable. People with poor mental health may not respond to a survey, so careful attention is needed to generalize interpretation of the results. Finally, since this is a cross-sectional study, we cannot conclude anything about the causal relationships between the variables. In the future, it is necessary to conduct longitudinal research to identify causality.

Conclusion

The questionnaire survey for mothers of children with CLCP showed that many mothers experienced positive change through child rearing. There were strong relationships between participants' positive change scores and the experience of sharing emotions with mothers in the same situation, support from medical staff and living in safe areas. SOC was positively correlated with PPC and SC. Experiences of being stigmatized about CLCP were associated with low maternal SOC scores. To improve mothers' SOC, it may be useful to help mothers perceive positive change through child rearing, although the possibility of a causal relation between SOC and positive change will need to be confirmed by further studies using longitudinal methods.

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