

Mediating effects of care burden and sense of coherence between patients' disabilities and siblings' emotions in psychotic disorders

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Abstract

Aim: Care burden and sense of coherence (SOC) can facilitate an understanding of how family members interpret their caregiving experiences regarding a relative with psychosis. In informal caregiving, understanding siblings' experiences in this regard is necessary to mitigate negative emotions and strengthen positive ones. This study investigated whether care burden and SOC mediate the relationship between the disabilities of patients with psychosis and their siblings' emotions.

Methods: A nationwide online survey was used to screen and recruit adult siblings of patients with psychotic disorders. The patients' disabilities in their work lives, socializing, and family communication, the siblings' care burden and SOC, and the siblings' positive and negative emotions were assessed. Structural equation modeling (SEM) was employed to test the path model that illustrated the mediating effects of care burden and SOC on patients' disabilities and siblings' emotions.

Results: The sample included 237 siblings aged 42.3 years (mean) with a male-to-female ratio of 0.88 and 237 patients with schizophrenia (80.2%) or schizoaffective disorder (19.8%). In the SEM analyses, care burden connected each of the three disabilities with the two types of emotions through the pathways in which care burden positively predicted both negative and positive emotions. Additionally, SOC functioned as a mediator between disability in family communication and both types of emotions.

Conclusion: Siblings' negative emotions should be alleviated through interventions aimed at suppressing the care burden and enhancing SOC to address disabilities in patients' socializing and family communication. Nevertheless, care burden might contribute to their positive emotions.

KEYWORDS

caregiver burden, mediator, psychosis, sense of coherence, sibling

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INTRODUCTION

In recent decades, clinical research has focused on the period of first-episode psychosis. Approximately 54% of the affected individuals entered remission, and 32% recovered during a 6-year follow-up period.¹ However, the incurable nature of psychosis may confront patients, and consequently, their family members, with challenges of living with psychotic disorders even after the first episode. The onset of schizophrenia commonly occurs in people (males and females) in their 20s.² The parents of these individuals (hereafter, patients) usually become the key caregivers, especially when the patient's mean age at their first marriage exceeds 30 years (for both genders), as is the case in developed countries.³ As their parents age and eventually die, depending on the family's circumstances, the patients' siblings take over as caregivers. Smith et al.⁴ investigated the future expectations of 137 healthy siblings about caregiving roles. They identified siblings' sense of responsibility, personal gain, sex (female), and relationship with the patient as significant predictors of siblings' involvement in providing instrumental or emotional support. Similar to parents, siblings are negatively and positively impacted by their involvement in caring for relatives with psychosis.^{5,6} Therefore, understanding sibling caregiving for psychotic disorders is essential. It can help mitigate the negative impacts of caregiving while increasing its positive impacts.

A conceptual model of family caregiving for schizophrenia was proposed based on a qualitative synthesis that included siblings' experiences in continuous caregiving.⁷ Family members feel negative emotions (sadness, fear, anger, shame, or guilt) due to the care burden and positive emotions from a sense of coherence (SOC). SOC is a common health factor in individuals who succeed in maintaining physical and mental well-being under intense stressors and traumas. It consists of three core dimensions: the sense of comprehensibility, manageability, and meaningfulness.⁸ SOC strengthens individuals' confidence in understanding and handling difficulties and cooperative relationships with others. This sense of comprehensibility and manageability helps individuals integrate their traumatic and negative experiences with the sense of living a meaningful life.

In the above-mentioned conceptual model, care burden is presumed to be a potential mediator. A meta-analysis⁹ demonstrated a positive association between functional impairment in schizophrenia and family burden in caregiving. Tarcijonas et al.¹⁰ defined "functional impairment" as disabilities in the daily aspects of life related to sustained employment, independent living, and social relationships. Psychotic disorders cause social withdrawal, including "hikikomori," a Japanese term referring to extreme social reclusiveness.^{11,12} The aspect of disability with the strongest relation to care burden in daily life remains unclear. The care burden in schizophrenia is associated with family caregivers' states of anxiety, depression, and expressed emotions (EE).¹³ "Burden" (assessed using the Zarit Burden Interview) was identified as a potential predictor of the pathological levels of anxiety and depression experienced by informal caregivers in a local community.¹⁴ EE represents the emotional attitudes of family caregivers and involves critical comments, hostility, and over-

involvement with patients with schizophrenia. The relationship between family caregivers with high-EE status and patients with psychosis having low social functioning led to a high perceived care burden.¹⁵

In addition to care burden, SOC potentially functions as a mediator in the proposed conceptual model. However, there is a paucity of evidence on the function of SOC in caregiving for psychotic disorders. In previous studies,^{16,17} SOC worked as a direct protective factor against care burden. SOC could also enhance the sense of satisfaction that family members feel when caring for relatives with schizophrenia.¹⁸

In this study, we aimed to test the hypothesis that care burden and SOC function as mediators in the relationship between the disabilities of patients with psychosis and their siblings' emotions. In addition to the mediating effects, we examined the influence of the potential moderating effects of siblings' gender and the awareness of their key-person status in caregiving.

METHODS

Data collection

Data were collected online by screening a nationwide panel of survey participants in November 2021. The inclusion criteria were family members who (1) were aged between 18 and 74 years; (2) had patients diagnosed with schizophrenia or schizoaffective disorder; (3) were patients' siblings; (4) had patients being treated through regular visits, having had their last visit to a medical facility within the past year, or having been admitted to a medical facility within the past year; and (5) had patients aged between 16 and 74 years. We excluded family members who (1) had patients with depression or bipolar disorder, (2) were patients' parents or spouses, and (3) had patients who had their last visit to a medical facility over a year ago or were on an unknown treatment regimen. Eligible participants provided demographic information regarding gender, age, occupation, and residence. Our preliminary data collection had a sample size of 240 participants, which was estimated to include more than 100 participants in each of the subgroups of siblings' gender and siblings' relation to patients. Rakuten Insight, an internet research agency, managed the nationwide panel of 2,200,000 participants (45% male; 55% female). The panel demographics were comparable to a recent census of Japan's population.¹⁹

Measures

Questions for patients' disabilities (QPD)

Three questions were used to measure the extent to which mental health problems interfered with patients' abilities in their work lives, socializing, and family communication during the past month. The questions were "To what extent has any mental problem interrupted

his/her work, schoolwork, or housework?”, “To what extent has any mental problem interrupted his/her socializing with people outside the home?”, and “To what extent has any mental problem interrupted his/her communication within the family?” Siblings of patients rated each question with a score ranging from 0 (*not at all*) to 10 (*extremely*).

Japanese version of the Zarit Burden Interview short version (J-ZBI_8)

The Zarit Burden Interview (a self-report instrument) was originally developed to assess the care burden for geriatric patients.²⁰ It was found valid for assessing the caregivers of patients with psychotic disorders.²¹ Arai et al.²² developed its eight-item short version with a Cronbach's α of 0.89. The validity of the J-ZBI_8 has been confirmed using two factors: “personal strain” and “role strain.”²³ The former includes items such as, “Do you feel uncertain about what to do about your relative?” while an example for the latter includes, “Do you feel uncomfortable about having friends over because of your relative?” Both subscales use a five-point Likert scale with scores ranging from 0 to 20 and 0 to 12, respectively.

SOC Scale 13-item version (SOC-13)

This self-report scale has 13 items forming three subscales on the sense of comprehensibility (e.g., “Has it happened in the past that you were surprised by the behavior of people whom you thought you knew well?”), manageability (e.g., “How often do you have feelings

that you're not sure you can keep under control?”), and meaningfulness (e.g., “How often do you have the feeling that there's little meaning in the things you do in your daily life?”).⁸ A seven-point Likert scale scores the three subscales from 5 to 35, 4 to 28, and 4 to 28, respectively. The reliability and validity of the original²⁴ and the Japanese version^{25,26} have been established.

Profile of Mood States 2nd edition Adult Short (POMS2-AS)

This self-administered questionnaire assesses the states of seven moods during the past week for adults aged 18 and above.²⁷ It consists of anger–hostility (AH), confusion–bewilderment (CB), depression–dejection (DD), fatigue–inertia (FI), tension–anxiety (TA), vigor–activity (VA), and friendliness (F). Each subscale includes five words as items that represent its mood state, such as “Angry” for AH and “Lively” for VA. Each mood score ranges from 0 to 20 on a five-point Likert scale. The Japanese version is reliable and valid, and it has been standardized in a representative Japanese population.²⁸ The standardization enables the conversion of the seven mood scores into standard scores by gender.

Data analyses

The means and SD of the QPD, J-ZBI_8, SOC-13, and POMS2-AS were calculated. We also calculated the Pearson's coefficient correlations between all the variables. After this, basic SEM models were created to test two hypotheses (Figure 1)—patients' disabilities

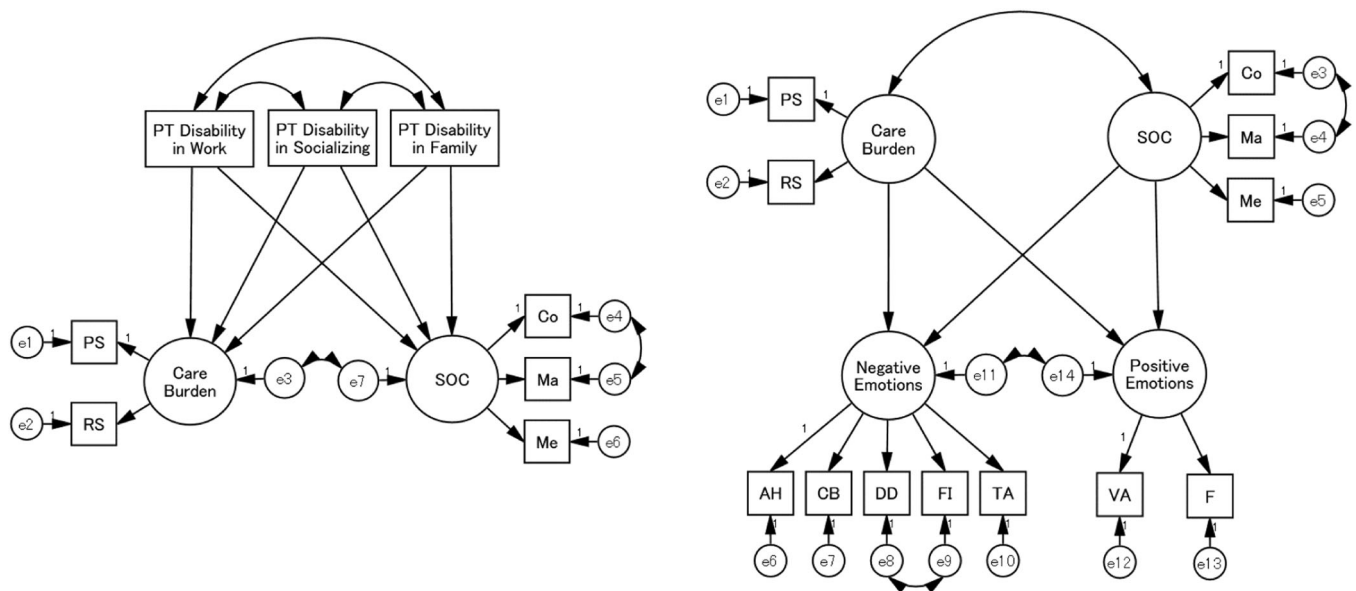


FIGURE 1 Basic path models that illustrate the first hypothesis (left) and the second hypothesis (right). AH, anger–hostility; CB, confusion–bewilderment; Co, comprehensibility; F, friendliness; 1DD, depression–dejection; FI, fatigue–inertia; Ma, manageability; Me, meaningfulness; PT, patient; PS, personal strain; RS, role strain; SOC, sense of coherence; TA, tension–anxiety; VA, vigor–activity.

TABLE 1 Sociodemographic and clinical characteristics of siblings (SIB) and patients (PT)

	SIB total (n = 237)		SIB relation to PT				Key person in caregiving			
			Brother (n = 111)		Sister (n = 126)		Yes (n = 106)		No (n = 131)	
SIB age, mean (SD), years	42.3	(11.6)	44.6	(11.6)	40.3	(11.4)	42.0	(12.7)	42.5	(10.7)
SIB male gender, n (%)	111	(46.8)	111	(100)	0		60	(56.6)	51	(38.9)
SIB educational background, n (%)										
<High school	11	(4.6)	4	(3.6)	7	(5.6)	5	(4.7)	6	(4.6)
High school	49	(20.7)	17	(15.3)	32	(25.4)	25	(23.6)	24	(18.3)
Two-year college	69	(29.1)	24	(21.6)	45	(35.7)	27	(25.5)	42	(32.1)
University	94	(39.7)	56	(50.5)	38	(30.2)	39	(36.8)	55	(42.0)
Postgraduate course	14	(5.9)	10	(9.0)	4	(3.2)	10	(9.4)	4	(3.1)
SIB occupational status, n (%)										
Regular employment	113	(47.7)	68	(61.3)	45	(35.7)	55	(51.9)	58	(44.3)
Irregular employment	61	(25.7)	18	(16.2)	43	(34.1)	28	(26.4)	33	(25.2)
Self-employment	17	(7.2)	14	(12.6)	3	(2.4)	7	(6.6)	10	(7.6)
Housewife/househusband	19	(8.0)	1	(0.9)	18	(14.3)	7	(6.6)	12	(9.2)
Student	4	(1.7)	2	(1.8)	2	(1.6)	2	(1.9)	2	(1.5)
Unemployment	18	(7.6)	7	(6.3)	11	(8.7)	5	(4.7)	13	(9.9)
Others	5	(2.1)	1	(0.9)	4	(3.2)	2	(1.9)	3	(2.3)
SIB marital status, n (%)										
Unmarried	110	(46.4)	43	(38.7)	67	(53.2)	47	(44.3)	63	(48.1)
Married	127	(53.6)	68	(61.3)	59	(46.8)	59	(55.7)	68	(51.9)
SIB living status, n (%)										
Living with PT	73	(30.8)	41	(36.9)	32	(25.4)	42	(39.6)	31	(23.7)
Not living with PT	164	(69.2)	70	(63.1)	94	(74.6)	64	(60.4)	100	(76.3)
Caregiving duration										
<6 months	75	(31.6)	22	(19.8)	53	(42.0)	20	(18.9)	55	(42.0)
6–12 months	24	(10.1)	17	(15.3)	7	(5.6)	15	(14.2)	9	(6.9)
1–3 years	30	(12.7)	19	(17.1)	11	(8.7)	16	(15.1)	14	(10.7)
3–5 years	23	(9.7)	12	(10.8)	11	(8.7)	14	(13.2)	9	(6.9)
5–10 years	29	(12.2)	12	(10.8)	17	(13.5)	17	(16.0)	12	(9.2)
≥10 years	56	(23.6)	29	(26.1)	27	(21.4)	24	(22.6)	32	(24.4)
Caregiving time/week, mean (SD), h	11.9	(23.0)	12.6	(21.7)	11.2	(24.1)	17.3	(26.6)	7.5	(18.5)

	SIB total (n = 237)		SIB relation to PT				Key person in caregiving			
			Brother (n = 111)		Sister (n = 126)		Yes (n = 106)		No (n = 131)	
PT age, mean (SD), years	42.6	(11.7)	44.6	(11.3)	40.9	(11.7)	41.9	(12.7)	43.2	(10.8)
PT male gender, n (%)	117	(49.4)	73	(65.8)	44	(34.9)	55	(51.9)	62	(47.3)
PT educational background, n (%)										
<High school	28	(11.8)	15	(13.5)	13	(10.3)	13	(12.3)	15	(11.5)
High school	91	(38.4)	36	(32.4)	55	(43.7)	40	(37.7)	51	(38.9)
Two-year college	45	(19.0)	16	(14.4)	29	(23.0)	21	(19.8)	24	(18.3)
University	67	(28.3)	42	(37.8)	25	(19.8)	28	(26.4)	39	(29.8)
Postgraduate course	6	(2.5)	2	(1.8)	4	(3.2)	4	(3.8)	2	(1.5)

TABLE 1 (Continued)

	SIB total (n = 237)		SIB relation to PT				Key person in caregiving			
			Brother (n = 111)		Sister (n = 126)		Yes (n = 106)		No (n = 131)	
PT occupational status, n (%)										
Regular employment	36	(15.2)	21	(18.9)	15	(11.9)	24	(22.6)	12	(9.2)
Irregular employment	48	(20.3)	18	(16.2)	30	(23.8)	25	(23.6)	23	(17.6)
Self-employment	14	(5.9)	9	(8.1)	5	(4.0)	8	(7.5)	6	(4.6)
Housewife/househusband	16	(6.8)	8	(7.2)	8	(6.3)	7	(6.6)	9	(6.9)
Student	3	(1.3)	1	(0.9)	2	(1.6)	1	(0.9)	2	(1.5)
Unemployment	116	(48.9)	53	(47.7)	63	(50.0)	40	(37.7)	76	(58.0)
Others	4	(1.7)	1	(0.9)	3	(2.4)	1	(.9)	3	(2.3)
PT marital status, n (%)										
Unmarried	178	(75.1)	78	(70.3)	100	(79.4)	74	(69.8)	104	(79.4)
Married	59	(24.9)	33	(29.7)	26	(20.6)	32	(30.2)	27	(20.6)
Diagnosis, n (%)										
Schizophrenia	190	(80.2)	84	(75.7)	106	(84.1)	73	(68.9)	117	(89.3)
Schizoaffective disorder	47	(19.8)	27	(24.3)	20	(15.9)	33	(31.1)	14	(10.7)
Treatment status, n (%)										
Regular visits	165	(69.6)	71	(64.0)	94	(74.6)	65	(61.3)	100	(76.3)
Irregular visits	31	(13.1)	23	(20.7)	18	(14.3)	18	(17.0)	13	(9.9)
Hospitalization	41	(17.3)	17	(15.3)	14	(11.1)	23	(21.7)	18	(13.7)
Number of hospitalizations, n (%)										
None	72	(30.4)	34	(30.6)	38	(30.2)	30	(28.3)	42	(32.1)
One time	64	(27.0)	30	(27.0)	34	(27.0)	37	(34.9)	27	(20.6)
Two or more times	101	(42.6)	47	(42.3)	54	(42.9)	39	(36.8)	62	(47.3)

can explain the care burden and SOC of siblings and the care burden and SOC can predict the negative and positive emotions of siblings. Given that the two models were fitted to the data, we built a combined SEM model that illustrated the pathways from patients' disabilities to negative and positive emotions via care burden and SOC. We then ran multigroup analyses to examine the configural invariance of the combined model in terms of being the patient's brother or sister and their regard for themselves as a key person in caregiving. Configural invariance means that different groups possess the same number of factors and pattern of loadings.²⁹ The fit of the models to the data was computed in terms of the goodness-of-fit indices of their χ^2 value (CMIN), comparative fit index (CFI), and root mean square error of approximation (RMSEA). Following the conventional criteria, a good fit would be indicated by CMIN/df < 2, CFI > 0.97, and RMSEA < 0.05, and an acceptable fit by CMIN/df < 3, CFI > 0.95, and RMSEA < 0.08.³⁰ IBM SPSS Statistics version 28 and AMOS version 28 for Windows were used to perform all analyses.

RESULTS

Participant characteristics

Table 1 shows the sociodemographic and clinical characteristics of the participants. In total, 237 siblings had a mean age of 42.3 years (SD = 11.6), and 237 patients were affected with schizophrenia (80.2%) or schizoaffective disorder (19.8%). Of the siblings, 111 were brothers and 126 were sisters. The male-to-female ratio between siblings and patients was 1.92 among brothers and 0.54 among sisters. The brothers were more highly educated and securely employed than the sisters. In caregiving, 54.1% of brothers and 36.5% of sisters considered themselves as a key person for the patients. Therefore, they were engaged in caregiving 2.3 times longer than those who did not play such a role. Nearly half of the patients with a mean age of 42.6 years (SD = 11.7) were unemployed, and three-quarters of them were unmarried. About 44.5% of 110 unmarried siblings and 18.9% of 127 married siblings lived with the patients.

TABLE 2 Correlations between patients' disabilities, siblings' care burden, SOC, and emotions for multigroup analyses

	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15
1. QPD Wo	---	.820***	.730***	.239*	.216*	-.273**	-.068	-.048	.225*	.285**	.327***	.242*	.172	.044	.100
2. QPD So	.848***	---	.581***	.380***	.311***	-.268**	-.223*	-.228*	.240**	.413***	.366***	.384***	.388***	-.143	-.081
3. QPD Fa	.833***	.662***	---	.377***	.345***	-.304**	-.169	-.137	.207*	.374***	.351***	.329***	.281**	.084	.154
4. ZBI PS	.833***	.606***	.638***	---	.349***	-.197*	-.204*	-.262**	.273**	.412***	.381***	.413***	.392***	-.213*	-.136
5. ZBI RS	.662***	.645***	.322***	.404***	---	-.322***	-.103	-.116	.281**	.370***	.383***	.339***	.328***	.109	.172
6. SOC Co	.606***	.412***	.363***	.434***	.405***	-.290***	-.204*	-.369***	.353***	.449***	.396***	.490***	.414***	-.250**	-.247***
7. SOC Ma	.334***	.388***	.462***	---	.821***	-.296**	-.205*	-.263**	.518***	.594***	.556***	.633***	.575***	.037	.163
8. SOC Me	.336***	.369***	.313***	.757***	---	-.293**	-.210*	-.289**	.551***	.642***	.602***	.661***	.575***	.067	.171
9. AH	.270**	.339***	.416***	.823***	.821***	---	-.262**	-.392***	.489***	.619***	.548***	.550***	.533***	-.005	-.025
10. CB	-.139	-.108	-.097	-.179	-.218*	---	.508***	.529***	-.318***	-.423***	-.406***	-.425***	-.406***	.284**	.135
11. DD	-.318***	-.264**	-.372***	-.358***	-.367***	-.366***	---	.427***	-.189*	-.439***	-.359***	-.331***	-.392***	.182*	.136
12. FI	-.141	-.165	-.063	-.174	-.168	.433***	---	.439***	-.289**	-.277**	-.346***	-.339***	-.400***	.257**	.127
13. TA	-.150	-.157	-.169	-.347***	-.248**	.548***	.509***	---	-.299***	-.445***	-.430***	-.348***	-.359***	.210*	.054
14. VA	-.193*	-.190	-.196*	-.284**	-.346***	.435***	.357***	---	-.288**	-.309***	-.352***	-.424***	-.399***	.383***	.272**
	-.153	-.218*	-.306***	-.354***	-.339***	.509***	.502***	.502***	---	-.476***	-.485***	-.353***	-.398***	.388***	.479***
	.260**	.268**	.421***	.451***	.541***	-.058	-.203*	-.166	---	.731***	.820***	.765***	.738***	.110	.147
	.211*	.236**	.269***	.454***	.507***	-.298***	-.298***	-.406**	-.406**	.711***	.686***	.721***	.677***	.017	-.044
	.303**	.334**	.411***	.533***	.633***	-.194*	-.224*	-.320***	.784***	---	.855***	.831***	.782***	.098	.174
	.392***	.423***	.417***	.588***	.622***	-.545***	-.435***	-.456***	.676***	.827***	.858***	.788***	.858***	-.154	-.063
	.305**	.329***	.425***	.508***	.642***	-.125	-.208*	-.243*	.800***	---	.855***	.823***	.806***	.077	.113
	.367***	.383***	.365***	.525***	.529***	-.490***	-.467***	-.525***	.708***	.871***	---	.653***	.781***	-.177*	-.153
	.314**	.367***	.419***	.623***	.670***	-.205*	-.283**	-.386**	.725***	.816***	.750***	---	.796***	.089	.144
	.345***	.386***	.441***	.546***	.549***	-.456**	-.372**	-.382***	.739***	.800***	.708***	.784***	.813***	-.052	.023
	.265**	.300**	.390***	.476***	.517***	-.244*	-.336***	-.321***	.709***	.744***	.760***	.784***	---	.016	.177
	.322***	.364***	.368***	.543***	.566***	-.483***	-.405***	-.443***	.691***	.867***	.807***	.816***	---	-.140	-.064
	-.085	-.018	.056	.096	.191*	.299***	.239*	.295**	.208*	.175	.188	.165	.116	---	.801***
	-.078	-.122	-.194*	-.039	-.066	.221*	.258**	.435***	-.013	-.167	-.200*	-.071	-.168	---	.664***

TABLE 2 (Continued)

	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15
15. F	-.103	-.043	.025	.117	.077	.253**	.128	.308**	.139	.159	.120	.165	.172	.662***	---
Relation: Mean	3.4 (2.6)	3.9 (2.7)	3.8 (2.4)	8.5 (4.9)	4.6 (4.9)	20.0 (4.9)	15.8 (3.9)	16.8 (4.1)	55.8 (10.8)	56.7 (10.8)	58.3 (10.6)	53.9 (10.1)	54.4 (10.3)	50.5 (11.1)	49.7 (12.0)
(SD)	3.3 (3.2)	3.2 (3.0)	3.1 (2.9)	8.9 (6.0)	4.1 (3.4)	17.7 (5.3)	14.3 (3.7)	15.9 (4.4)	53.4 (11.2)	56.8 (12.9)	57.8 (12.1)	54.5 (11.4)	55.3 (10.5)	48.4 (10.3)	47.9 (11.1)
Key person:	3.9 (2.4)	4.0 (2.5)	4.0 (2.5)	8.6 (4.9)	4.5 (3.1)	18.6 (4.7)	14.9 (3.4)	16.6 (3.7)	55.3 (9.7)	57.4 (10.5)	58.8 (9.9)	54.1 (9.5)	54.7 (9.3)	50.8 (9.4)	50.1 (10.6)
Mean (SD)	3.0 (3.2)	3.1 (3.1)	3.0 (2.7)	8.8 (5.9)	4.2 (3.4)	18.9 (5.7)	15.1 (4.2)	16.1 (4.7)	53.8 (12.1)	56.2 (13.0)	57.4 (12.5)	54.3 (11.7)	55.0 (11.2)	48.2 (11.5)	47.7 (12.2)

Note: The correlations of the relation data (brother/sister) and the key-person data (yes/no) are above and below the diagonal, respectively.

Abbreviations: AH, anger-hostility; CB, confusion-bewilderment; Co, comprehensibility; DD, depression-dejection; Fa, family; F, friendliness; FI, fatigue-inertia; Ma, manageability; Me, meaningfulness; PS, personal strain; QPD, questions for patients' disabilities; RS, role strain; So, socializing; SOC, sense of coherence; TA, tension-anxiety; VA, vigor-activity; Wo, work; ZBI, Zarit Burden Interview.

* $p < .05$; ** $p < .01$; *** $p < .001$.

Path analysis model fit

Appendix 1 presents the correlations between all study variables in the sample. The negative emotions of the POMS2-AS were significantly positively correlated with the three domains of the QPD and subscales of the J-ZBI_8 and negatively correlated with the subscales of the SOC-13. Higher VA and F were significantly correlated with increased subscales of the SOC-13.

The total sample data met the conventional criteria for the model fit with $CMIN/df = 1.648$, $CFI = 0.992$, and $RMSEA = 0.052$ for the first basic model (Appendix 2) and $CMIN/df = 1.901$, $CFI = 0.979$, and $RMSEA = 0.062$ for the second basic model (Appendix 3). The data were then confirmed to be fitted to the combined model with $CMIN/df = 1.627$, $CFI = 0.981$, and $RMSEA = 0.052$ (Appendix 4). Table 2 shows the correlations between all the variables grouped by the siblings' relation to the patients and their awareness of being a key person in caregiving. Multigroup analyses demonstrated the configural invariance with $CMIN/df = 1.394$, $CFI = 0.976$, and $RMSEA = 0.041$ for the former group (Figure 2), and $CMIN/df = 1.436$, $CFI = 0.973$, and $RMSEA = 0.043$ for the latter group (Figure 3). The better fit to the combined model indicated that the siblings' data were to be separated by family relation or awareness of key-person status.

MEDIATING EFFECTS OF CARE BURDEN AND SOC

Figures 2 and 3 show that care burden was mediated between the three domains of patients' disabilities and siblings' negative and positive emotions. Disability in family communication was positively associated with the care burden of brothers ($\beta = 0.34$, $p = 0.021$), sisters ($\beta = 0.33$, $p = 0.003$), and siblings without key-person awareness ($\beta = 0.39$, $p < 0.001$). Disability in socializing was positively associated with the care burden of brothers ($\beta = 0.42$, $p = 0.013$). Disability in work lives was negatively associated with the care burden of brothers ($\beta = -0.33$, $p = 0.048$). For patients' key persons, only the disability in socializing was related to care burden ($\beta = 0.41$, $p = 0.037$). Care burden connected patients' disabilities with siblings' negative and positive emotions through the pathways in which care burden positively and commonly predicted not only negative but also positive emotions. The care burden of key persons had the strongest effect on their negative emotions ($\beta = 0.70$, $p < 0.001$). Concurrently, care burden more strongly influenced the positive emotions of siblings who were aware of their key-person status ($\beta = 0.55$, $p < 0.001$) than those without this awareness ($\beta = 0.35$, $p = 0.004$). Further, care burden had a larger influence on the positive emotions of sisters ($\beta = 0.58$, $p = 0.001$) than brothers ($\beta = 0.27$, $p = 0.01$).

SOC also functioned as a mediator between the patients' disability in family communication and the siblings' negative and positive emotions (Figures 2 and 3). Disability in family communication was negatively associated with the SOC of sisters ($\beta = -0.47$, $p < 0.001$) and siblings without key-person awareness ($\beta = -0.38$, $p = 0.002$). SOC connected disability in family communication with

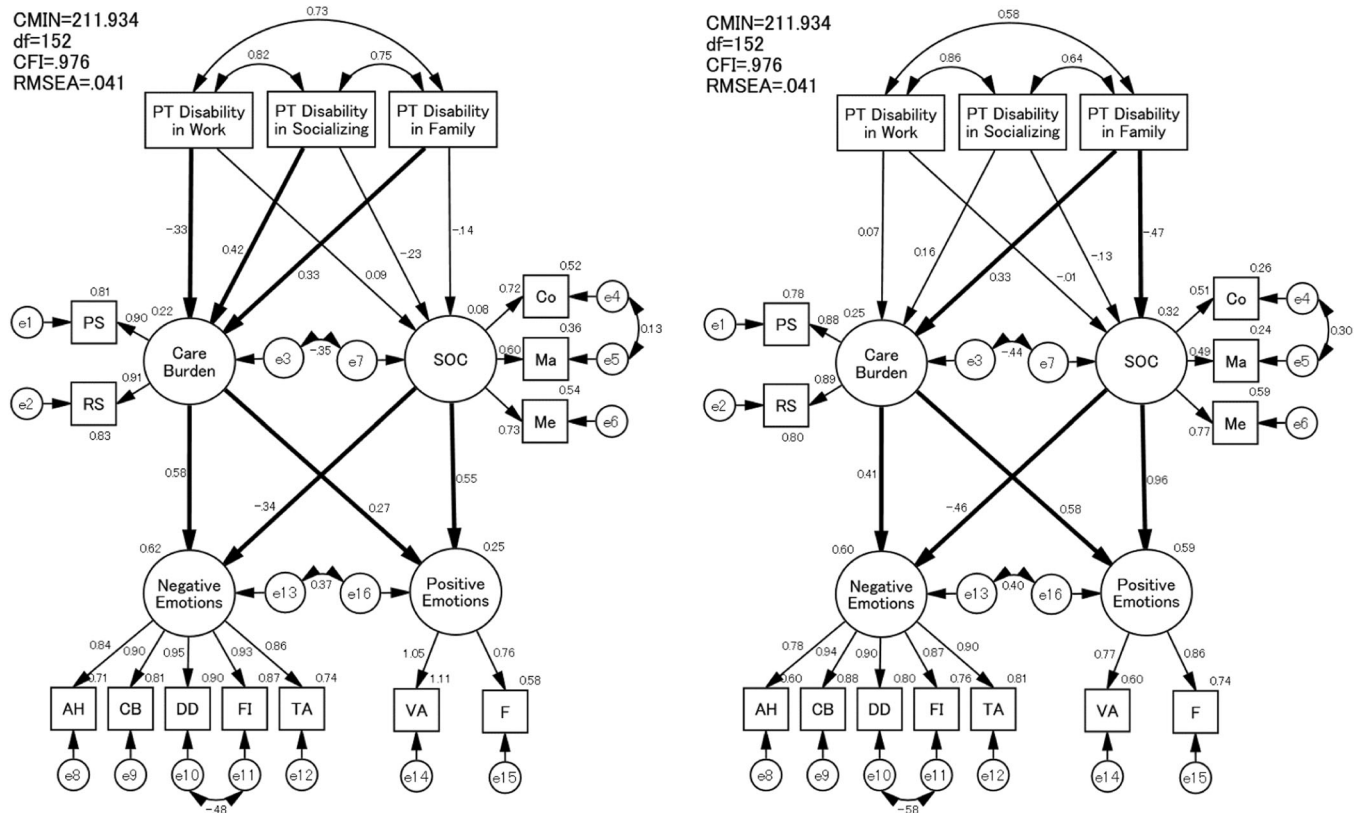


FIGURE 2 Pathways from patients' (PTs) disabilities to negative and positive emotions via the care burden and sense of coherence (SOC) of brothers (left) and sisters (right). Note: The values in bold indicate statistically significant paths ($p < 0.05$) with a standard estimate. AH, anger-hostility; CB, confusion-bewilderment; Co, comprehensibility; DD, depression-dejection; F, friendliness; FI, fatigue-inertia; Ma, manageability; Me, meaningfulness; PS, personal strain; PT, patient; RS, role strain; TA, tension-anxiety; VA, vigor-activity.

negative and positive emotions through the following pathways: SOC positively affected the positive emotions of sisters ($\beta = 0.96$, $p < 0.001$) and siblings without key-person awareness ($\beta = 0.70$, $p < 0.001$). It negatively affected the negative emotions of sisters ($\beta = -0.46$, $p = 0.001$) and siblings without key-person awareness ($\beta = -0.48$, $p < 0.001$).

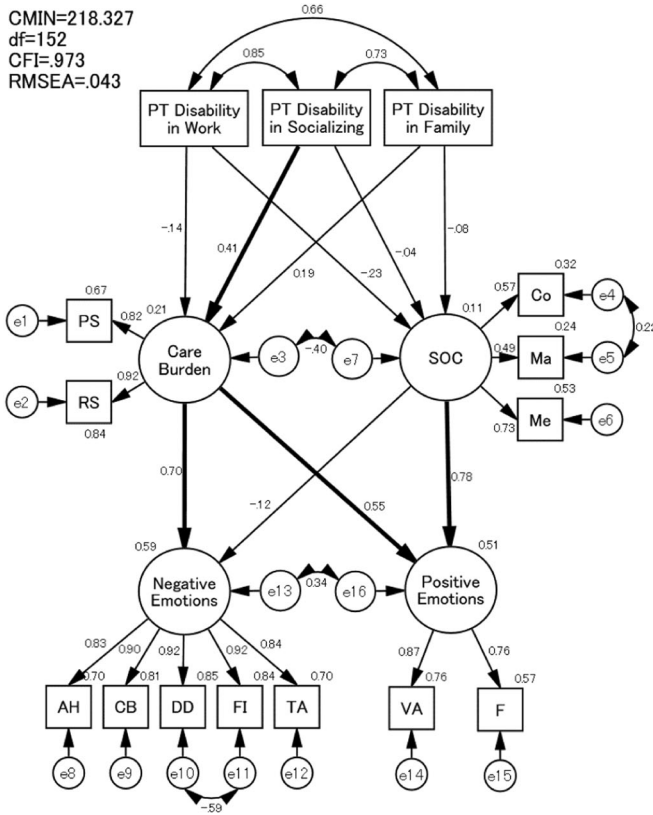
DISCUSSION

The SEM analyses revealed the functions of care burden and SOC as mediators between the disabilities of patients with psychosis and their siblings' emotions. The more severe the patients' disabilities in their socializing and family communication, the higher the care burden and the greater the positive and negative emotions of the patients' siblings. Disability in family communication was positively associated with the care burden of brothers and sisters. However, this significant association disappeared when siblings assumed a key role in caregiving, and the key persons felt exclusively burdened by the patients' disability in their socializing. Disability in family communication also undermined the function of SOC to enhance positive emotions and suppress negative emotions in patients' sisters and siblings without the key-person role.

As expected, the associations between patients' disabilities and the siblings' care burden predicted the siblings' negative emotions. To our knowledge, this is the first study to quantitatively demonstrate the mediating effects of the care burden between patients' disabilities in their socializing and family communication and their siblings' positive emotions. Previous qualitative studies are consistent with this finding, suggesting the presence of positive emotions in informal caregiving.⁷ Siblings recognized that mutual support bound the family more tightly when dealing with difficulties together.³¹ By gaining knowledge and skills, siblings could improve their self-confidence in caregiving.³² Siblings felt that they had developed because they could apply their caregiving abilities to other life difficulties.³¹ These positive aspects of the care burden facilitated siblings' expression of affection for their ill siblings³³ and motivated them to help others with mentally ill relatives.³¹

SOC showed mediating effects for patients' sisters and siblings without key-person awareness. Of the three disabilities, only family communication illustrated significant relationships with SOC. Suresky et al.³⁴ reported the negative association between family disruption and SOC in female family members of adults with mental disorders, including schizophrenia (45%) and bipolar disorders (45%). Disrupted family life could lead to a lower level of SOC experienced by female caregivers. However, healthy siblings who become primary caregivers

GMIN=218.327
df=152
CFI=.973
RMSEA=.043



GMIN=218.327
df=152
CFI=.973
RMSEA=.043

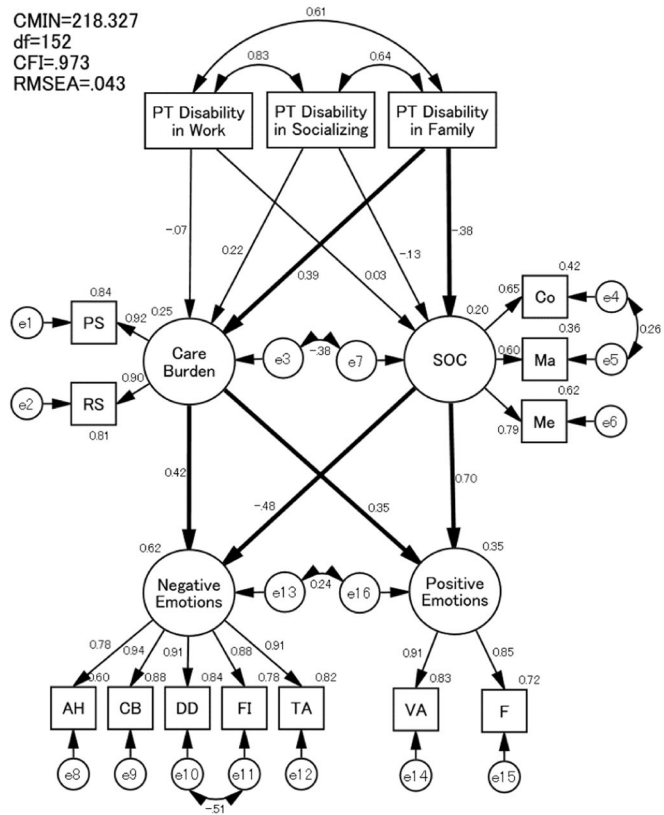


FIGURE 3 Pathways from patients' (PTs) disabilities to negative and positive emotions via the care burden and sense of coherence (SOC) of siblings with (left) and without (right) key-person awareness. Note: The values in bold indicate statistically significant paths ($p < 0.05$) with a standard estimate. Co, comprehensibility; Ma, manageability; Me, meaningfulness; AH, anger-hostility; CB, confusion-bewilderment; DD, depression-dejection; F, friendliness; FI, fatigue-inertia; PS, personal strain; PT, patient; RS, role strain; TA, tension-anxiety; VA, vigor-activity.

tend to have a positive relationship with their ill siblings,⁴ which was presumed to attenuate the negative association between disability in family communication and SOC. Additionally, SOC plays a protective role against the depression and anxiety of caregivers.³⁵ Our study suggests that the SOC does not function with the sister or the sibling without key-person awareness when patients have disability in family communication.

Our study has some limitations. First, the online data reported by the participants were not validated with the clinical data recorded by healthcare facilities. Despite the impossibility of the validation, we consider that a nationwide online survey could represent the general population of individuals with mentally ill siblings more than a survey conducted at a limited number of healthcare facilities. Second, Japan's healthcare system might make our findings less generalizable to other countries. We found a negative association between patients' disability in their work lives and their male siblings' care burden. The adverse relationship was possibly due to the Japanese welfare system, wherein severe disability in work lives makes it possible for such patients to receive more satisfactory welfare services. Third, this study is cross-sectional, and therefore we cannot confirm any causalities from the findings. For example, siblings' care burden could result in the disruption of patients' family lives.³⁴

CONCLUSION

Our study provides suggestions on clinically supporting siblings whose roles tend to move toward being a key person in caregiving as their parents age and die. The negative emotions that sibling caregivers feel should be alleviated by healthcare services for patients with disabilities in their socializing and family communication by suppressing care burden and enhancing SOC. Moreover, contribution to taking care of an ill sibling can be a potential source of positive emotions for sibling caregivers. Further longitudinal research is needed to confirm the mediating effects of care burden and SOC on the relationship between the disabilities of patients with psychosis and their siblings' emotions.

AUTHOR CONTRIBUTIONS

Nao Shiraishi conceived the survey design, analyzed the data, and drafted the manuscript. Hiroko Yatsu and Haruka Ogawa designed the survey. Tatsuo Akechi supervised the study and revised the draft.

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CONFLICT OF INTEREST

Nao Shiraishi has received lecturing fees from Mochida Pharma. Hiroko Yatsu and Haruka Ogawa declare no competing interests. Tatsuo Akechi has received lecturing fees from Chugai, Daiichi-Sankyo, Dainippon-Sumitomo, Eisai, Janssen, Kyowa, Lilly, MSD, Meiji-Seika Pharma, Mochida, Nipro, Nihon-Zoki, Otsuka, Pfizer, Takeda, and Viatrix. Tatsuo Akechi has also received royalties from Igaku-shoin and research funds from Daiichi-Sankyo, Eisai, FUJIFILM RI Pharma, MSD, Otsuka, and Shionogi. Tatsuo Akechi is the inventor of the pending patents (2019-017498 & 2020-135195).

DATA AVAILABILITY STATEMENT

The datasets analyzed in this study are available from the corresponding author upon reasonable request.

ETHICS APPROVAL STATEMENT

The present study was approved by the Institutional Review Board of Nagoya City University Graduate School of Medical Sciences and Nagoya City University Hospital (no. 60-19-0123).

PATIENT CONSENT STATEMENT

Web-based informed consent was obtained from all survey participants.

CLINICAL TRIAL REGISTRATION

This study was registered at UMIN Individual Case Data Repository (registration ID: UMIN000045247; date of registration: 1 September 2021).

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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