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Insights into the caregiver perspective: Involvement, well-being, and interventions

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Insights into the caregiver perspective: Involvement, well-being, and interventions

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Editorial: Insights into the caregiver perspective: involvement, well-being, and interventions

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caregiving, rewarding caregiving, demanding caregiving, physical illness, mental illness

Editorial on the Research Topic
Insights into the caregiver perspective: involvement, well-being, and interventions

The range, duration, and intensity of informal caregiving across different illnesses and disabilities have increased in the 21st century due to the increase in longevity and de-institutionalization in many countries. De-institutionalization has led to increase in the demand of and ability to provide homecare in the community by informal caregivers.

Caregiving is demanding, and hence can be stressful in terms of time, effort, emotional and social impact, as well as financial requirements, depending on the nature of the illness or disability, the relationships between the person supported and the caregiver, and the role played by available health and social care services. Yet, research evidence has demonstrated that caregiving can be also rewarding, as a different type of bonding is enabled than was the case before caregiving became a necessity.

The thirteen articles published in this issue on the theme of caregiving make for an original in-depth contribution to existing research on this sensitive and important issue.

The articles cover a range of countries (Canada, China, India, Italy, Malaysia, Singapore, Spain, UK, and US), and a wide range of physical and mental ill health issues which caregivers are responding to, leading to differentiated impact on them too.

The issues looked at include caregiving to children and adults, focusing on:

- Borderline Personality Disorder,
- Chronic illnesses,
- COVID-19,
- Down syndrome,
- First Episode of Psychosis,
- Lung cancer,
- Recovery from mental illness,
- Sclerosis,
- Stroke,
- Total knee Replacement.
- Young Homeless Refugees

Although caring for older people is in frequent demand, it is not looked at any of the articles in this Research Topic.

Likewise, a range of research methodologies have been applied in the articles. Most findings are based on cross sectional research of carers' responses to a number of questionnaires in each study, some of which have already been verified, as well as constructing new questionnaires and verifying their validity and reliability.

The foci of the questionnaires range from looking at dimensions of care burden, such as anticipatory grief, anxiety, care rewards, care benefit, depression, parental perspective on the quality of life of children with a disability, Posttraumatic stress, and the carers' quality of life.

Some articles provide a systematic review of the existing literature, indicating through narrative synthesis potential improvements in caring which would be of help to the family member in need of caring and to the caregivers too (San Juan et al.).

A few of the articles aim at evaluating training in enhancing the skills that carers have and their resilience (e.g., Sharbafshaaer et al.), and introducing new forms of the support they can offer to their ill relatives, such as motivational interviewing by carers for adolescents undergoing first episode of psychosis (Kline et al.).

Sample size varies among the different studies, from 12 (in a study of telemedicine during the COVID-19 pandemic by Sharbafshaaer et al.), to 395 (Zhou et al.) for the articles which focus on measuring responses either to questionnaires or to training. Samples range in most such studies between 200 and 300+ (13, 73, 233, 243, 254, 363), enabling a reasonable degree of representation.

Statistical expertise was demonstrated in particular in one article where a network analysis of family caregivers' needs when their family member experiences cancer is outlined (Yang W. F. Z.).

Only one article follows the qualitative autoethnographic approach in providing a narrative of three episodes of being cared for. It is also the only article to be written by a service user about her experience of being cared for by her mother, and the support she has received from a health provider in enabling her to discuss discomforting experiences of care in the context of her gradual journey toward recovery and increased independence (Fox). This article highlights the value of adhering to the Triangle of Care, consisting of the person, the parent-carer, and the health professional.

Individual interviews were applied in the study of homeless youth refugees in Canada (1).

The context of the specific illness dictates to a great extent the type and degree of care required from informal caregivers, but the different articles highlight the high level of care entailed, and the high impact caregiving has on different aspects of the lives of the carers.

Most articles did not focus on policy issues and on entitlement of carers to specific support, be it financial, shared care with other family members, payment to caregivers for their input, reduction in working hours, and enabling respite care for the caregivers. However, the authors of one of the systematic reviews (San Juan et al.) indicated the lack of sufficient studies of low income countries. This study recommends that future research will entail policy and practice aspects, as well as socio-political aspects of caregiving in the different countries. This article also compares the perspective of service users vs. the perspective of the caregivers on the issue of recovery from severe mental illness.

The articles in this Research Topic provide a unique contribution to understanding the issues caregivers face, and how they make sense of their role in a range of illnesses and social contexts.

Author contributions

SR have reviewed several of the articles and acted as the first associate editor of this Research Topic of Frontiers Psychiatry.

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Conflict of interest

The author declares that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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ORIGINAL RESEARCH
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Creating a Caregiver Benefit Finding Scale of Family Caregivers of Stroke Survivors: Development and Psychometric Evaluation

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In recent years, increased attention has been paid to the benefit finding of family caregivers due to the important role they play. Although some instruments measure benefit finding of caregivers, they do not comprehensively address it in terms specific to the family caregivers of stroke survivors, who require long-term, consistent care. This study is the first effort to develop a comprehensive Caregiver Benefit Finding Scale for the family caregivers of stroke survivors in a Chinese cultural setting. First, 50 items were extracted from a systematic literature review, and a semi-structured interview was conducted with 20 stroke family caregivers to develop the preliminary version of the scale (Version 1). Second, Delphi procedures with 20 experts were used to revise the first version and create Version 2 (37 items). Another six experts were recruited for content validation. Item content validity index (I-CVI) values ranged from 0.83 to 1.00, and the value of the scale CVI was 0.97. Third, 309 family caregivers completed the Version 2 questionnaire and the Chinese version of the Positive Aspects of Caregiving. Two weeks later, 35 family caregivers once again completed the questionnaires. An exploratory factor analysis produced four components (personal growth, health promotion, family growth, and self-sublimation) and 26 items for Version 3 (the cumulative proportion variance was 74.14%). Subsequently, 311 family caregivers completed Version 3. A confirmatory factor analysis confirmed the structure. The goodness of fit index (GFI) = 0.921, adjusted GFI = 0.901, normal fit index = 0.951, incremental fit index = 0.990, comparative FI = 0.990, and the root mean square error of approximation = 0.02 were within the acceptable range. Criterion-related validity was equal to 0.803. The model-based internal consistency index was 0.845 and the values of the Cronbach'α coefficient of the four dimensions were 0.885–0.953. The split-half reliability was 0.92, and the test-retest reliability was 0.994. These findings provide preliminary evidence of the validity and reliability of the Caregiver Benefit Finding Scale. The scale can help researchers and clinicians to achieve a more comprehensive understanding of stroke family caregivers' positive experience. This understanding is necessary for future efforts to address issues in benefit finding by targeting the underlying mechanism and intervention.

Keywords: benefit finding, family caregivers, stroke, scale development, validation

INTRODUCTION

Strokes pose a major health threat and are, on a global scale, the leading cause of mortality and disability (1). In China, stroke is the leading cause of death and the most common origin of diseases that cause disabilities (2). More than 4.5 million stroke survivors live with the resultant disabilities (3) and are consistently being cared for by family caregivers. Caregiving is detrimental to the physical and psychological health of caregivers, a fact that also affects the care recipients' quality of care and quality of life (4–7).

Caregivers, however, also experience benefits such as personal growth, better relationships with patients, and finding personal meaning during the caregiving experience (8, 9). More importantly, benefit finding may mitigate caregiver burdens, reduce the negative impact on the caregiver's quality of life, and help caregivers cope with stress caused by caregiving (10, 11). Intervention focused on benefits finding was found to reduce caregivers' depression and promote caregivers' physical health by effectively strengthening their immunity (12–14). Hence, in recent years there has been increased attention on measuring the benefit finding for caregivers in the field, to determine benefits finding, explore its effects on caregiver outcomes, and uncover how it works (9, 15).

Some instruments have been developed to measure positive outcomes related to caregiving. These include the Stress Related Growth Scale (16) and Post Traumatic Growth Inventory (PTGI) (17). The validity of these two scales, however, has been questioned (18–21). Other than these particular instruments developed for diverse caregiver populations, some measurements were developed for specific types of caregivers. Positive Aspects of Caregiving (22) was developed for caregivers of dementia patients and the Gains in Alzheimer's Care Instrument (23) and Reward of Caregiving Scale (24) were developed for palliative caregivers. The Scale for Positive Aspects of Caregiving Experience was developed for caregivers of schizophrenia patients (25). There is a requirement to validate an existing construct within a new population or assess whether adaptation and additions to the construct are required for it to translate to stroke samples.

Benefits finding is the most commonly reported type of meaning-focused coping, which is very important in the revised stress and coping model (26). A meta-analysis defined benefit finding as the positive effects that result from a traumatic event (27). A integrative review of the literature highlighted that benefit finding is an important component of positive outcomes of caregiving (9). Scales have been developed to specifically measure benefit finding, such as the Benefit Finding Scale and Benefit Finding in Multiple Sclerosis Scale. The Benefit Finding Scale was developed for patients with breast cancer (28). This scale has been used with caregivers of patients with other types of cancer (29) and as well as those with heart failure (30). The Benefit Finding in Multiple Sclerosis Scale was developed for caregivers of those with multiple sclerosis (31). These benefit finding scales may not be applicable for caregivers of stroke survivors because the nature of strokes differs significantly from these other diseases.

Moreover, the experience of family caregivers can differ significantly because of specific cultural differences (32). Chinese caregivers may value the benefits gained from receiving praise from the neighborhood, communities, and society in general (33) or from being a role model for younger generations (34). These particular factors are not measured by existing scales. The limitations of the previous scales provided the grounds to conceive the present study to develop a new comprehensive scale, the "Caregiver Benefit Finding Scale for Stroke Caregivers," and evaluate its psychometric properties.

MATERIALS AND METHODS

This study was performed in three phases. In the first phase, a literature review and semi-structured interviews were used to generate items to be included in the questionnaire. In the second phase, Delphi procedures were conducted to revise the item pool and an expert panel was used to assess the content validity. In the third phase, the psychometric evaluation of the Caregiver Benefit Finding Scale was performed. An overview of the tool development process is shown in **Figure 1**.

Participants and Design

Family caregivers of stroke survivors from five hospitals and four communities in Zhengzhou, China, were recruited to participate in the study. Three inclusion criteria were established for family caregivers. The first criterion was that the caregivers were primary family members (i.e., non-professional and unpaid) of a stroke survivor aged 18 or above, who had a formal diagnosis of cerebrovascular disease and had a functional disability (Barthel Index < 100) (35). The second criterion was that the caregiver provided care for at least 4 h each day for 4 weeks. The third, and final, criterion was that caregiver could communicate in Mandarin and was willing to participate in the study. The study was conducted from June 2015 to December 2017.

Phase One: Item Generation - Scale Version 1 (50 items)

The Caregiver Benefits Finding Scale, Version 1, was developed through a systematic literature review and semi-structured interviews conducted with family stroke caregivers.

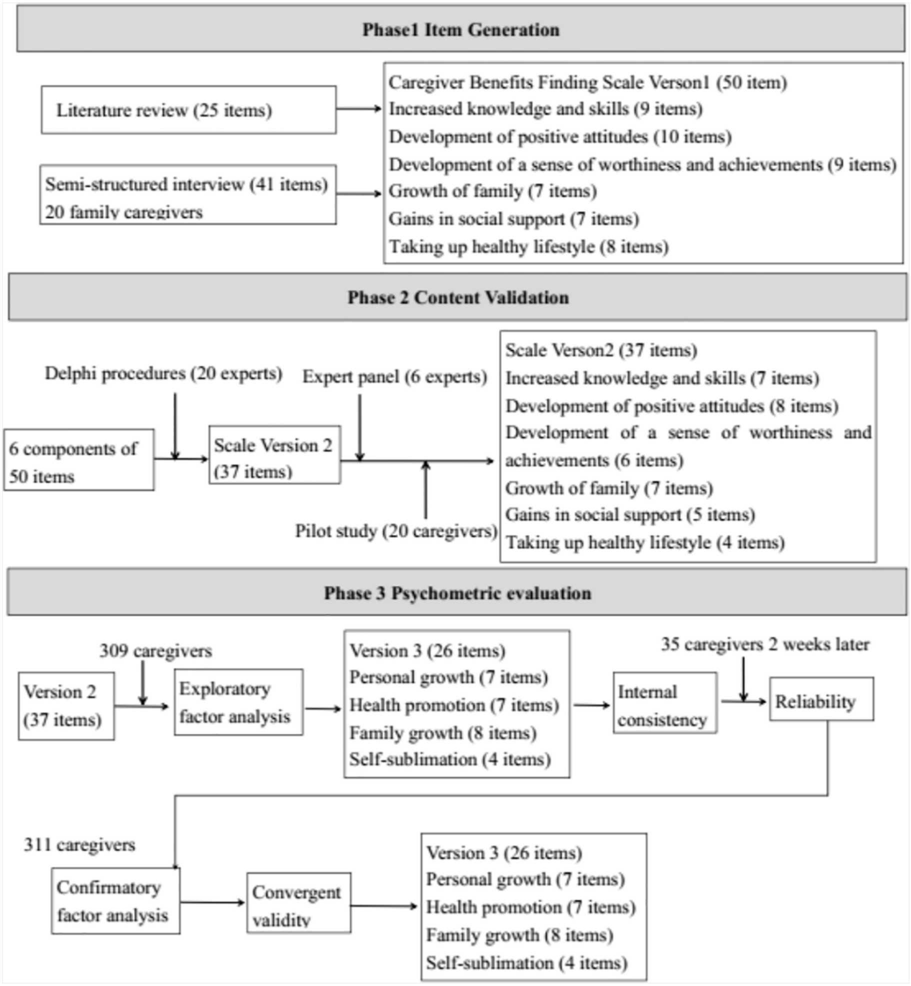

FIGURE 1 | Overview of the three-phased tool validation study.

The review was conducted using six English databases (PubMed, CINAHL, PycINFO, Embase, Web of Science, ProQuest Health, and Medical Collection) and three Chinese databases (CNKI, Wanfang, and CMB) to identify previous scales related to caregiving benefits findings. The database search criteria were articles published before December 2015 that used the terms "caregiver" (including synonyms) AND "benefit finding" (including synonyms). The inclusion criteria were articles focused on benefit finding of caregivers. The exclusion criteria were as follows: caregivers were younger than 18 years old; caregivers were professional personnel; patients were younger than 18 years old; and articles not written in English or Chinese. The related existing scales were compared (the details can be found in **Supplementary Table S1** online). All of the items in those scales were read and similar items were removed after discussion with the research team, As a result, 25 items related to benefit finding in family caregivers were obtained.

Semi-structured interviews were performed to determine the perceived benefits from the perspective of the family caregivers and how they interpreted those benefits. The interview included several questions including the following. How have you changed as a result of caring for your wife/husband/father/mother? How have these changes affected your life? What does caring mean to you? What benefits have you perceived that have come to you from being a caregiver? When you have perceived benefits that come from caring, how have your perceptions of the act of caring changed? How do others (other family members, friends, and neighbors)view your caring? What caused you to keep taking care of your wife/husband/father/mother for so long? Purposive sampling and snowball sampling were used to recruit stroke family caregivers. Sample size was determined by the saturation of interview data; the primary researcher perceived that the content of the interviews was becoming repetitive and that no new information was emerging during the interviews, and the interview transcripts were also reviewed by the research team to ensure that no new content was emerging (36). All interviews were performed in the caregivers' homes and in the presence of two researchers. The interviews were recorded and each interview lasted between 30 and 60 min with an average duration of approximately 42 min. The interviews were transcribed verbatim and then coded independently by two researchers.

Twenty stroke family caregivers were interviewed. Thirteen were females, and seven were male. There were seven wives, five husbands, five daughters, two sons, and one mother of stroke survivors. The caregivers' duration of care ranged from four months to 14 years. Eight of the caregivers provided care for eight to 12 h each day and six provided care for at least 12 h while six provided care for 4 to 8 h per day. Thematic analysis (including data familiarization, coding and developing themes and subthemes) was performed to analyze the interview transcripts (37). As a result, six themes were identified; increased knowledge and skills, development of positive attitudes, development of a sense of worthiness and achievement, growth of family ties, gains in social support, and adopting a healthy lifestyle. Within the six themes, 41 items were obtained.

As a result of the literature review and semi-structured interviews (and removing 16 duplicate items), a pool of 50 items were generated to measure caregiver benefit finding (for Version 1 of the scale). A five-point Likert scale ranging from 1 to 5 was used to assess the level of benefit finding in caregivers on each item.

Phase Two: Content Validation - Scale Version 2 (37 items)

The Content of Caregiver Benefit Finding Scale, Version 1, was validated in two rounds of Delphi survey. The 21 national experts were invited by email and 20 responded to the two rounds of Delphi procedures. Of these experts, five were nursing experts in psychology, four were researchers with expertise concerning caregivers of stroke patients, and three were researchers with expertise in scale development. Two were nursing experts working in the community, two were psychiatric nurses, two worked in clinical rehabilitation, two were neurological doctors and one was a nursing expert who worked in stroke clinical wards. In the first round, the initial version of the scale was emailed separately to the experts. A five-point, Likert-type, scale with values ranging from 1 to 5 was used to evaluate the relevance and clarity of the items. The experts could write comments and revise or add items. Discussions were held on the comments by the experts in the research group, and the revision, based on a consensus of opinion, was built. The second round was held 2 weeks later to ensure that there was agreement on the revision. The overall authority grade of the Delphi consultation was 0.895. Kendall's coefficient of concordance W of the two rounds consultation was 0.138 ($\chi^2 = 135.14$, $P < 0.001$) and 0.232 ($\chi^2 = 180.67$, $P < 0.001$). After the Delphi procedure, 37 items were included in Version 2.

Another six experts (two researchers with expertise in stroke patients, two researchers with expertise in scale development, one nursing expert working in the community, and one researcher with expertise in caregivers) were invited to assess content validity of Version 2. Item content validity index (I-CVI) and scale content validity index (S-CVI) were used to evaluate content validity, using a four-point scale ranging from 1 (not relevant) to 4 (highly relevant) (38). The I-CVI values ranged from 0.83 to 1.00, and the value of S-CVI was 0.97.

A pilot survey was conducted with 30 family caregivers. The survey took 5 to 8 min to complete and the items were clearly stated.

Phase Three: Psychometric Evaluation of the Scale (from Version 2 to Version 3)

Construct validity was assessed by an exploratory factor analysis (EFA) to identify the possible components in the scale. Moreover, criterion-related validity, internal consistency reliability, and test-retest reliability were used to evaluate the validity and reliability. To perform EFA, the sample size must be greater than 300 (39). Ten percent of the family caregivers were randomly chosen again after 2 weeks to assess test-retest reliability (39). Version 2 was refined into Version 3 (26 items) after EFA. Then, confirmatory factor analysis (CFA) was performed to verify the components of Version 3. Additionally, convergent validity was used to evaluate validity. According to Boomsma's advice, the minimal sample size for performing CFA is 300 (40).

Measures

Participants' demographic characteristics included gender, age, marital status, education, household income per month, health insurance, daily hours of caring, relationship with patients for family caregivers, and stroke patients' severity of disability (Barthel Index) (35).

Version 2 of the Caregiver Benefit Finding Scale was used for the first survey in order to perform EFA. Version 3 was used for the second survey to perform CFA.

Positive Aspects of Caregiving (Chinese version; PAC-C) (22): PAC-C was used to evaluate the scale's criterion-related validity, which is a nine-item self-report tool, including self-affirmation and outlook on life as two components. PAC-C items are scored from 1 (strongly disagree) to 5 (strongly agree) (22). PAC-C has an adequate internal consistency reliability, with the Cronbach'α of 0.90 (41).

Data Analysis

Data were analyzed using IBM SPSS, Version 21.0 software and AMOS 17.0. The demographic characteristics of patients with stroke and their caregivers were analyzed descriptively and presented as numbers and percentages.

A Pearson correlation analysis was conducted to evaluate the item total correlation, and the value of item total correlation > 0.4 with a statistical significance testing was considered to indicate a desirable discriminating power and the criteria-related validity. Extreme group (27% and 73% of the score of the Caregiver Benefits Finding Scale) comparison was performed using an independent-samples *t*-test.

EFA was performed to explore construct validity under the situation that the Kaiser-Meyer-Olkin measure of sampling adequacy ≥ 0.8 and the Bartlett's test of sphericity with

$P < 0.05$ (42). Additional criteria included a factor loading of at least 0.4, with the difference between a loading and any cross loading of at least 0.15 for an item to remain on its factor. Each factor was also required to have at least three items for that factor to be retained.

CFA with the normal theory maximum likelihood estimation was then conducted to verify the construct validity (43). The value of relative chi-square ($\chi 2$/df) < 2, the values of goodness-of-fit index (GFI), the adjusted GFI (AGFI), a comparative fit index (CFI), and Bentler and Bonett's normed-fit index (NFI) ≥ 0.9, and the value of a root mean square error of approximation (RMSEA) < 0.06 were considered to indicate an acceptable model fit (44). After conducting CFA, average variance extracted (AVE) and composite reliability were used to examine the convergent validity. The value of AVE > 0.5 and value of composite reliability > 0.7 were considered evidence of convergent validity.

Cronbach's α and a model-based internal consistency index were used to evaluate the internal consistency reliability of the scale (45). A Pearson correlation analysis was used to assess the test-retest reliability, and the Guttman Split-half coefficient was used to evaluate the split-half reliability.

Ethical Consideration

This study was approved by the Ethical Committee of Zhengzhou University. Informed consent statements were obtained from each participating hospital and community. All study participants agreed to join voluntarily and signed informed consent statements.

RESULTS

Sample Characteristics

A total of 710 (20 for the semi-structured interview) family caregivers of stroke survivors were recruited at different phases. Of these, 340 participants were recruited to obtain data for EFA and 315 answered the questionnaires. Eight invalid questionnaires were removed. A total of 307 questionnaires were analyzed. Among those caregivers, 35 participants were randomly chosen to answer the questionnaires again 2 weeks later. Another 350 participants were recruited to obtain data for CFA, and 320 caregivers answered the questionnaires. Nine invalid questionnaires were removed and a total of 311 questionnaires were analyzed. The demographic characteristics of participants for recruited for EFA and CFA are listed in **Table 1**.

Item Analysis and Extreme Group Comparison

All 37 item total correlation values ranged from 0.529 to 0.838 (all with $P < 0.001$) and show good inter-item associations. Extreme groups were divided into high- and low-score groups. All comparisons showed a significant difference (all with $P < 0.001$).

Results of EFA

The Kaiser-Meyer-Olkin measure of sampling adequacy (0.965) and Bartlett's test of sphericity ($\chi^2 = 13330.00$, $P < 0.001$)

TABLE 1 | Characteristics of the participants.

Variable	Variable category	EFA (n = 307)N (%)	CFA (n = 311)N (%)
Age (years old)	<45	110 (35.8)	151 (48.6)
	45~	140 (45.6)	100 (32.1)
	60~	57 (18.6)	60 (19.3)
Gender	Male	117 (38.1)	132 (42.4)
	Female	190 (61.9)	179 (57.6)
Marital status	Married	264 (86.0)	270 (86.8)
	Single/divorced/widowed	43 (14.0)	41 (13.2)
Education level	Primary	48 (15.6)	34 (10.9)
	Secondary	84 (27.4)	86 (27.7)
	High school	92 (30.0)	76 (24.4)
	University and above	83 (27.0)	115 (37.0)
Household income per month (Yuan)	<1000	36 (11.7)	30 (9.6)
	1000~	45 (14.7)	44 (14.1)
	2000~	99 (32.2)	67 (21.6)
	3000~	127 (41.4)	170 (54.7)
Relationship with the patients	Spouse	110 (35.8)	100 (32.2)
	Daughters/sons	149 (48.5)	151 (48.5)
	Parents	38 (12.4)	36 (11.5)
	Other relatives	10 (3.3)	24 (7.8)
Daily hours caring	4~	59 (19.2)	66 (21.2)
	8~	59 (19.2)	72 (23.2)
	12h~	189 (61.6)	173 (55.6)
Health insurance	Province level	22 (7.2)	5 (1.6)
	City level	174 (56.7)	248 (79.7)
	New Rural Cooperative	57 (18.6)	41 (13.2)
	Others	54 (17.6)	17 (5.5)
Severity of disability of care receiver	Minimum dependence	151 (49.2)	142(45.7)
	Moderate dependence	62 (20.2)	55(17.7)
	Maximum dependence	94 (30.6)	114(36.6)

provided support for an EFA. Principal axis factoring with an oblimin rotation was chosen to perform the EFA, with the scree plot (**Figure 2**) used to determine the number of factors to rotate. Examination of the scree plot to determine the point at which the line/slope begins to flatten yielded four as the best starting point. Accordingly, solutions with three, four, and five factors were examined to find the most valid solution. As a result, 11 items were removed and EFA extracted four factors that accounted for 74.15% of the total variance with 26 items. Based on factor loadings, these four factors were termed "personal growth (seven items)," "health promotion (seven items)," "family growth (eight items)," and "self-sublimation (four items)" as shown in the component matrix (**Table 2**).

Criterion-Related Validity

The Caregiver Benefit Finding Scale and PAC-C were in positively correlated ($r = 0.760$, $P < 0.01$), and all domains were also positively correlated (all $P < 0.01$).

Reliability

The model-based internal consistency index of the scale was 0.845. The Cronbach's α of each component was between 0.885 and 0.953. The values of split-half and test-retest reliability were 0.929 and 0.994 respectively.

Results of CFA

The initial model had a χ^2/df of 1.660 ($P < 0.001$), with GFI = 0.888, AGFI = 0.869, NFI = 0.932, IFI = 0.972, CFI = 0.972, and RMSEA = 0.046. The modification index indicated that the model fit could be improved. Ten correlation covariances were added, which could be explained in the content. Thus, the modified model produced a χ^2/df of 1.254 ($P = 0.002$), with GFI = 0.921, AGFI = 0.901, NFI = 0.951, IFI = 0.990, CFI = 0.990, and RMSEA = 0.029. Although the χ^2 is significant, this significance may exist because of the larger sample size. Overall, the model is a good fit and confirmed the results of EFA that suggested the Caregiver Benefit Finding Scale had four factors.

The value of AVE in the four domains ranged from 0.621 to 0.700. The value of composite reliability ranged from 0.891 to 0.942, which indicated a good convergent validity.

DISCUSSION

The main objectives of this study were to develop and analyze benefit finding psychometric properties and to develop a caregiver benefit finding scale to measure benefit finding for stroke family caregivers. The final scale comprises 26 items and possesses good validity and reliability. This finding expands the study of benefit finding by proposing a tool to directly measure benefit finding that can be used in the specific context of family caregiving of stroke survivors (see below section 4.1 for applications).

The EFA supported a four-dimensional scale structure, comprising a sense of personal growth, health promotion, family growth, and self-sublimation. Compared with the previous scales, the item "Made me be regarded as a good example by my family and friends" in the family growth domain was new added, and the health promotion and self-sublimation domains were new constructs. In Chinese culture, caregivers believe it is their duty and responsibility to care for their family members (32), and they are proud to be role models

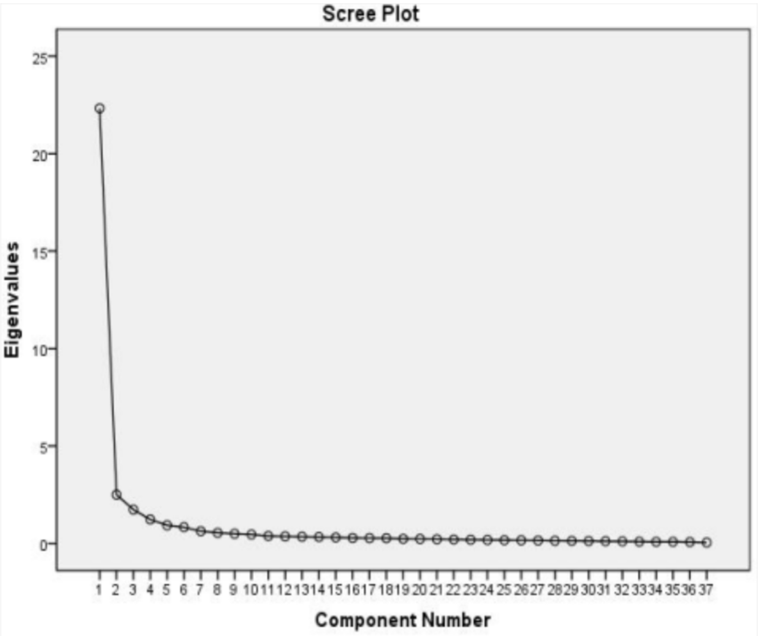

FIGURE 2 | Scree plot for the exploratory factor analysis.

TABLE 2 | Four factors extracted from factor analysis using matrix rotation (n = 307).

Taking care of patients				
	Factor 1	**Factor 2**	**Factor 3**	**Factor 4**
Personal growth				
Made me continuously improve my problem solving skills	0.911			
Made me improve my ability to care	0.861			
Made me cope with stress and difficulties better	0.818			
Made me know more about diseases	0.770			
Made me become more careful	0.749			
Made me improve my efficiency	0.688			
Made me improve my ability to guide others in healthy living	0.555			
Health promotion				
Made me pay more attention to the health of myself and other family members		0.816		
Made me focus on healthy eating		0.807		
Made me develop good habits		0.770		
Made me become aware of the significance of my health to the family and society		0.764		
Made me quit bad habits		0.716		
Made me see myself stronger and more brave		0.628		
Made me see things more positively		0.555		
Family growth				
Made our families become more united and harmonious			0.888	
Made me become more closer to my family			0.793	
Made my other family members have more time to do other things			0.672	
Made me feel more caring and support from my family			0.671	
Made me inherit and carry forward the tradition of loving each other			0.509	
Made my other family members become aware of love, giving and responsibility			0.502	
Made me spend more time with patients			0.500	
Made me be regarded as a good example by my family and friends			0.464	
Self-sublimation				
Made me feel more grateful and valued				0.781
Made me gain affirmation and praise				0.594
Made me feel more useful				0.558
Made me feel the sense of achievements				0.551

and set standards of care for the next generation (34); thus, they obviously perceive setting a good example for their families and friends as a benefit. Additionally, Chinese family caregivers place patients at the center of attention rather than themselves (32), and they are willing to do anything for their loved ones; thus, they promote healthier behaviors to take better care of stroke patients. Moreover, the Chinese government encourages the caregivers to take initiative in caring for their loved ones who need support at home (46), and thus caregivers possessed a great sense of contentment and value given recognition from their family and society. In addition, the CFA was also conducted to confirm the four-dimensional scale structure. The fit indices of CFA were satisfied. PAC-C was significantly correlated with the scale and all of its component factors, indicating that the Caregiver Benefit Finding Scale had acceptable criterion-related validity.

In this study, the "traditional" methods (Cronbach's α) and an innovative method (i.e.,model-based internal consistency) were both used to test reliability (45). Cronbach's α may not be the best method to test reliability of a multidimensional scale such as the Caregiver Benefit Finding Scale. As a result, the model-based internal consistency index for the total scale was 0.845. Cronbach's α had a range of 0.885 to 0.953 for the subscales, which indicated that the scale had acceptable internal consistency. Moreover, split-half reliability was 0.929, and test-retest reliability was 0.994, which confirmed the stability of the tool.

The survey with the Caregiver Benefit Finding Scale was conducted with 350 family caregivers being surveyed. A total of 320 caregivers responded with nine of the surveys judged as invalid, an effective rate of 97.46% (47). It should be noted that it took caregivers 9 min to complete the survey which suggests this Caregiver Benefit Finding Scale has some advantages for the caregivers who are busy caring patients and with a limited amount of time (47). In addition, caregivers thought that the items were clear and easy to understand which confirms the idea that the effort expended in answering the survey was acceptable to caregivers and could be used further.

Implications for Clinical and Research Practice

The Caregiver Benefit Finding Scale has important applications in the clinical and research areas. First, the scale provides a tool to measure the benefit finding of caregivers during acute and chronic phases of stroke recovery. Caregivers with a low level of benefit finding may be identified as a risk factor for negative outcome, requiring further assistance and resources (10). Second, the scale provides a tool to further study the mechanism of benefit finding and the experience of family caregivers, which includes both negative and positive experiences (48). Third, the scale provides a perspective on benefit finding intervention, which could focus on the personal growth, health promotion, family growth, and self-sublimation. Fourth, the Scale can be implemented as a measure in studies of interventions attempting to improve the mental health of family caregivers. Finally, the scale may help researchers to better understand how benefit finding may affect the outcomes of stroke patients (49).

Limitations

The study has limitations. First, the participants in the interviews in phase 1 were self-selected and came from a relatively affluent region of China; thus, their experiences may not reflect those of other caregivers in less affluent or rural areas in China. Moreover, we selected caregivers for the interviews in phase 1 who were dealing with a wide duration of illness (4 months to 14 years) to identify various types of benefit finding in a qualitative way. However, we did not explore the differences in benefit finding as perceived by caregivers at different stages. It would be advisable to perform a longitudinal to examine the trajectories of the levels and different types of benefit finding. Second, all caregivers participating in this study were living in Zhengzhou. The fact that they all came from one particular geographical area may have a limited application. Thus, it would be advisable to conduct the study using a larger and more diverse sample from different areas in China. This expanded study would allow more confidence in using the Caregiver Benefit Finding Scale to confirm the psychometric properties.

CONCLUSION

This study described the development of a Caregiver Benefit Finding Scale, a reliable and valid tool to evaluate important aspects of the experience of Chinese family caregivers taking care of stroke patients. The scale is shown to have adequate psychometric properties, which could be used to measure the positive experience that caregivers might have as a direct result of being family caregivers for stroke patients.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by ethical committee of Zhengzhou University. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

Y-XM was involved in design, data collection, analysis, and writing. B-LL was involved in data collection and analysis. W-HZ was involved in design and writing. S-SW was involved in data collection and writing. Z-XZ, D-BY, and DC were involved in design and writing. All authors contributed to the article and approved the submitted version.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2020.00734/full#supplementary-material

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The Outcomes of Health Education Programme on Stress Level Among the Caregivers of Post Total Knee Replacement Surgery

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Introduction: Stress level among the caregivers is often related to caregivers' lack of knowledge and skill to care for the patients. A health education program to the caregivers is one of the important elements in increasing the knowledge and skills in managing patients at home. The specific objectives of this study were to determine caregiver's stress level in managing post total knee replacement (TKR) patients pre and post of a health education program.

Materials and Methods: A clinical intervention trial design was conducted in Hospital Universiti Sains Malaysia (HUSM) with a sample size of 32 caregivers. A validated Zarit Burden Interview (ZBI) questionnaire was used to measure the stress level pre and post of the health education program on the management of patients post-TKR surgery which was adopted from Fresno Surgical Hospital in California.

Results: The findings showed that there was a significant difference between pre and post level of stress ($p \leq 0.001$).

Conclusion: This study revealed the positive outcome of the health education program. It reduced the stress level among the caregivers in caring for their relatives with post-TKR surgery.

Keywords: caregiver burden, stress level, health education programme, total knee replacement, mental health

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INTRODUCTION

With the increasing number of healthcare costs, medical advances, shorter hospital stays, and limited discharge planning, many responsibilities of long-term health care have moved from the hospital setting to the home environment (1). Once the care recipient is discharged home, family members are expected to provide much care to an adult with chronic illness and/or disability, for example; patients with post TKR (1). Caregivers of patients who underwent elective knee replacement play important roles in the early recovery process due to today's short hospital stays, and the increased age of patients (2). Caregivers often experience lower levels of psychological well-being, as well as a financial and physical burden during caregiving the patients of post-TKR surgery (3). It may therefore be a challenge for them to support the patients during the recovery period in their home environment. Also, caregiving responsibilities have expanded well-beyond assisting the patients' post-TKR with traditional household chores to now include performing medical and nursing tasks provided by the medical professionals in the hospital setting (4). According to Caron et al. (5), hospital admission is a stressful ordeal and often represents only the beginning of a long rehabilitation period of which hospital discharge is an important landmark.

It is estimated between 40 and 70% of caregivers exhibit clinically significant depressive symptoms with approximately one-quarter to one-half indicates the diagnostic criteria for major depression (6). Studies have shown that the stress of caregiving has put caregivers at a higher risk for chronic health problems such as cardiovascular problems and high blood pressure with an estimated 17–35% of caregivers perceiving their health as fair to poor (7).

Total knee replacement (TKR) is rapidly becoming one of the most common elective inpatient surgeries in the United States (8). In 2003 the number of TKRs performed in the United States was 402,100 (9). This number is expected to double by the year 2015 according to growth trends, even without factoring in the growing elderly population (9). TKR is often an effective elective surgery for patients whose quality of life has decreased because of pain and functional disability resulting from osteoarthritis (10).

Informal caregiving can facilitate a positive outcome for patients (11). Caregiver experienced more burden and less self-esteem especially among the spouses, and spouses of older adults reported that they felt less informed during the discharge process than caregiving adult children. It is known that emotional support from family caregivers can improve patients' recovery outcomes after knee replacement, for example by strengthening patients' beliefs in their ability to manage recovery and providing positive emotional responses to improve the patients' recovery (12).

Results from the European project showed that caregivers and patients who underwent knee replacement expected a wide range of knowledge and those expectations were not adequately met during the hospital stay (13). Caregivers may therefore lack empowering knowledge and may not be able to experience empowerment during patients' recovery process. Patient and caregiver education is linked to and promote the recovery process, and continuous and active involvement in healthcare results in better postoperative outcomes for the patients and their caregivers (14). Patients from the Nordic countries who underwent knee replacement surgery reported that caregiver involvement in patient education should be improved to make them more satisfied with the care they received (15). Caregivers with fulfilled knowledge expectations may feel empowered to support the patients during the early recovery period, and this may reduce their stress level in caregiving and at the same time may have a positive effect on patients' quality of recovery (QoR).

Research on caregiver's support of patients who underwent knee replacement is limited, in contrast to research on other medical issues, for example, persons with heart failure, dementia, chronic illness, or diabetes (16). To our knowledge, few studies have been made on caregiver's support of patients who underwent hip or knee replacement (17). However, the stress level among caregivers with patients post-TKR has not been studied before. As patients in the Nordic sample of the main project were least satisfied with how nurses prepared their caregivers before discharged from the hospital (15), we decided to conduct further analyses on the stress level among the caregivers with patients post-TKR; pre and post-intervention of health education program related to specific care after TKR surgery. This knowledge may help nurses to identify caregivers in need of support during the period of hospitalization.

In recent years, considerable effort has been made to describe the population of caregivers and examine the positive and negative consequences of caregiving (18). Positive outcomes of caregiving include personal growth, strengthening of the relationship between caregivers and care recipients, feelings of satisfaction, and increased self-esteem (19). Negative consequences of caregiving may be physical, financial, psychological, or social (18). Specifically, these consequences may include isolation, increased responsibilities, loss of employment, depression, a decline in physical health, financial strain, feelings of burden, and stress (20). The outcomes of stress and burden are central features of most caregiving models (18). Hence, the current study aimed to determine the difference in the stress level between pre and post health education program among caregivers of post-TKR patients.

MATERIALS AND METHODS

A clinical intervention trial design was used to conduct this study. This study used "one-group pre-test and post-test design," and "health education programme" was the independent variable which also served as an intervention in this study. This study was conducted among the caregivers of the patients who underwent TKR surgery in male and female orthopedic wards in Hospital Universiti Sains Malaysia (HUSM) Kubang Kerian, Kelantan during day one post-operation (POD1) and orthopedic clinic during the follow-up appointment.

The participants for this study were 32 caregivers of chronic knee osteoarthritis (OA) patients who were selected by using purposive sampling. Definition of caregivers are consist of patient's family members such as husband or wife, children or relatives who accompanied, consistently in delivery care, and managed the patients inward and at home, after they underwent TKR surgery and during follow-up. The sample size determination for participants in this study was by applying the formula of Krejcie and Morgan sample size calculation. Based on the calculation, the sample size of the participants supposed to be 40 participants. However, due to a few factors, the researcher obtained only 32 participants for this current study. The participants were those who accompanied, cared and managed the patients inward and at home, after they underwent TKR surgery and during TCA in the orthopedic clinic at HUSM. The participants who were selected must have the required inclusion criterion such as relatives who consistently in delivery care for the patients post-surgery, Malaysian, aged 21-year-old and above and able to understand the Malay language well while for exclusion criterion were such as having any medical problems (e.g., cancer under active treatment, diabetes, hypertension, and renal failure), frequent change of caregiver and first TCA is more than two weeks after the patients are discharged.

The intervention protocols involved in this study were the implementation of the structured health education programme (HEP) and pre and post self-administered questionnaire by Zarit Burden Interview. The health education programme used the post-TKR education package by Fresno Surgical Hospital in California. It consists of 10 elements that are; (1) pain management, (2) exercise, (3) preventing constipation, (4) preventing blood clot, (5) diet/nutrition, (6) medications, (7) surgical wound care, (8) durable medical equipment, (9) TCA, and (10) complications. The HEP was validated by two expert panels in orthopedic management. The health education programme was conducted once a day to each caregiver between 15 and 20 min on the day one of post-operation (POD1) of TKR. In conducting the HEP session, explanation to the caregivers was given in both English and Malay language. This is considering that most of the caregivers were not proficient users of English and the researcher speaks both languages fluently. Before the HEP session, the caregivers were required to answer the questionnaire (pre-test). The same questionnaire was given to the caregiver during the TCA to re-assess the outcome of the health education programme that was conducted to them before.

A validated questionnaire of Zarit Burden Interview was used as an instrument for this current study. The questionnaire consists of 22-items and each item uses a 5-point Likert scale ranging from 0 to 4; where 0 is never and 4 is always or almost always stress. The pilot study was conducted to 30 caregivers before the actual study to evaluate the proficiency of the questionnaire and intervention of the HEP. The result was 0.967 which in the range of 0.70 to 0.83 as suggested by Schrag et al. (21).

Ethical approval for this study was obtained from the Universiti Sains Malaysia Human Research Ethics Committee (JEPeM) and UKMMC Research Ethics Committee. All participants were given written consent before the session.

The data were analyzed by using SPSS version 24 for descriptive analysis such as mean, standard deviation, frequencies and percentage to outline the participants' descriptive characteristics. Categorical data such as age, gender, race, education level, marital status, employment, monthly income, and relationship of participants were analyzed using frequency and percentage while continuous data such as stress level was analyzed using mean and standard deviation. All continuous data had first been tested for its normality before proceeding to the inferential statistics.

RESULTS

The findings of the subjects' socio-demographic characteristics were illustrated in **Table 1**. Almost half of the participants fell into the 51–60-year-old age group (43.8%), while another half of them were from a different category of age with percentage, respectively. From this intervention group, there was 19 female (59.4%) caregivers and 13 males (40.6%). The data for socio-demographic characteristics in this current study were collected based on a study about caregivers' morbidity in palliative care unit: predicting by gender, age, burden and self-esteem (22).

TABLE 1 | Data of socio-demographic characteristics of participants.

Variables	Mean	Std deviation	Frequency ($n = 32$)	Percentage (%)
Age	51.19	12.09		
21–30			1	3.1
31–40			7	21.9
41–50			4	12.5
51–60			14	43.8
61–70			5	15.6
71–80			1	3.1
Gender				
Male			13	40.6
Female			19	59.4

To determine the difference in the stress level between pre and post HEP among caregivers of post TKR patients, a repeated measurement ANOVA was used. Based on the analysis of the Zarit Burden Interview (ZBI) questionnaire, there was a significant difference between the means of the pre and post-stress level of participants ($p \leq 0.001$). The total mean score for the post-stress level was 28.03 ($sd = 14.36$), which was lower than the total mean score for a pre-stress level, 40.91 ($sd = 19.48$) with mean different was −12.87 and its 95% confidence interval of different of lower was −17.15 and upper was −8.60 as shown in **Table 2**.

DISCUSSION

In managing patient care after surgery, most caregivers often experience stress and anxiety. The stress level increases when the caregivers lack the knowledge and appropriate skills and have no experience in patient care post-surgery. According to studies which adopted an online method of delivering psycho-educational interventions to participants, revealed a substantial improvement in caregivers' knowledge levels (22), stress, and social support levels (23). Additionally, lack of exposure to the structured and systematic discharge planning and HEP among the caregivers before the patients were discharged, contributed to the increase of caregivers' stress level. According to Sigurdardottir et al. (24), family psycho-educational programmes for patients and their caregivers were effective in improving physical and emotional health. There was evidence that psychosocial interventions improve coping, self-efficacy and reduce psychological distress among the caregivers of patients with OA (24).

Similar to this study, the level of stress among the caregivers decreased in which the results indicated a significant difference before and after the intervention. This indicated that the HEP conducted on the caregivers of patients who had undergone TKR surgery was significantly effective and the interventions showed additional positive effects on the caregivers. Sufficient health education and training among caregivers can greatly reduce

TABLE 2 | Means of pre and post-stress level of participants ($n = 32$).

Mean (SD)							
Variables	**Pre**	**Post**	**Mean differ**	**95% CI of the difference**		***t* (df)**	***p***
				Lower	**Upper**		
Total score of stress	40.91 (19.48)	28.03 (14.36)	−12.88	−17.15	−8.60	−6.15 (31)	≤0.001

A repeated measurement ANOVA, $p < 0.05$.

anxiety, depression and stress within their caregiving duties, ultimately prolonging their ability to provide care at home (25).

Another study by Tay Swee Cheng et al. (26) revealed that primary caregiver gained benefit in terms of reducing depression, more notably, strain at 6 months. Based on their findings, intervention caregivers' mean depression scores trended downward more than controls; mean strain scores remained stable in the intervention group but trended toward an increase in the control group. The findings were amplified among caregivers who provided more than 14 h of weekly assistance at baseline, the strain at 6 months was significantly lower in the intervention group. The observed effects of the intervention were both stronger among higher intensity caregivers and consistent across two distinct outcomes suggested that observed effects were due to the intervention (26).

Furthermore, a study by Wang et al. (27) mentioned that a greater reduction in caregiver stress scores baseline to post-intervention was attributed to their participation in the health education intervention. They might have gained new caregiving skills in coping with patient care post-surgery. Besides, they might have gained more confidence to deal with their relative's care during at home. This is consistent with earlier studies about the positive effects of psycho-education interventions on family burden (27). In this current study, it is found that HEP plays the main role and an important element in reducing stress level among the caregivers and also increases their knowledge and skills in patient care post-TKR surgery.

This study revealed that the sample size of the total population was too small ($n = 32$). This is because it was conducted with the participants of osteoarthritis patients from one hospital only which was Hospital Universiti Sains Malaysia (HUSM). Thus, this factor was the limitation of the current study.

It is essential to equip the caregivers with knowledge on TKR and its management especially when caregivers have limited formal education. This is because an increase in knowledge about patients' care post-TKR surgery might help reduce the caregivers' sense of burden and reduce their stress level. Further, information regarding the patients' care after TKR surgery, as well as potential complications facing the patient post the operation, should be explicitly explained to the caregivers. Hence it can reduce reliance on hospital services. Thus, more research is needed to explore the concept of optimal HEP for caregivers. This research could be extended to experimental study to investigate the effectiveness of HEP with the control and intervention group.

This study highlights the caregivers can be the helpful resources in the management of patients post-TKR. More consideration needs to be given to caregivers' of the patient, such as considering caregivers' health knowledge in the development of a health education programme. More information must be provided to those who live with knee osteoarthritis patients; caregivers' knowledge should be improved, especially for the management of patients post-TKR, for which they depend on the help of others. With information delivering as a major factor in perceived health-related competence, caregivers should be informed about their important role in managing patients post-TKR. Information about medications, exercises or physiotherapy, appointment, symptoms, diet and supportive measures could enhance their motivation and behavioral skills in such situations. Providing caregivers of patients post-TKR with such information could improve the quality of recovery; however, further studies are needed to confirm these findings and to explore health education programme in managing patients post-operative for other chronic bone diseases.

CONCLUSION

This study emphasized the importance and outcome of HEP for the caregivers of post TKR patients. More understanding of these specific caregiver needs and concerns by nurses could play an important role in developing caregiver centered care of patients with post-TKR surgery. Meanwhile, the facilitated application of the health education programme to the caregivers in caring for relatives who underwent TKR surgery provided a way of identifying their educational needs. It thus facilitated in examining and comparing the patterns of health needs in caregiver education. The design and findings of this study provided useful information on needs assessment and subsequent design of appropriate education programmes by nurses and healthcare professionals when providing caregiver-centered care in community settings.

Furthermore, based on findings of this current study it showed that stress level among the caregivers decreased after the intervention of HEP. This had proven that the HEP conducted on the caregivers of patients who had undergone TKR surgery was significantly effective and the intervention might have additional positive effects on the caregivers. This indicated that providing caregivers with health education and training can greatly reduce anxiety, depression and stress within their caregiving duties, ultimately prolonging their ability to provide care at home.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Universiti Sains Malaysia Human Research Ethics Committee and Research and Ethics Committee of Universiti Kebangsaan Malaysia Medical Center. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

MS: data collection, data analysis, and manuscript writing. JK: conceptual framework and manuscript editing. AS: conceptual framework and manuscript editing. All authors contributed to the article and approved the submitted version.

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Motivational Interviewing for Loved Ones in Early Psychosis: Development and Pilot Feasibility Trial of a Brief Psychoeducational Intervention for Caregivers

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Treatment delay and non-adherence in first episode psychosis is a pressing public health problem. Ambivalence regarding psychiatric intervention and labeling among young people with psychosis is a contributing factor. For these individuals, caregivers often facilitate the pathway to care and support ongoing engagement and adherence. Caregivers describe distress and burden associated with this role. This manuscript describes the development and pilot feasibility testing of a motivational interviewing-derived communication training for caregivers of individuals with untreated or under-treated early course psychosis. Individuals with lived experience were consulted in the intervention development process. The training consisted of four 60-min sessions teaching the philosophy and basic skills of motivational interviewing as well as two brief practice calls. Feasibility was assessed with regard to study enrollment, retention, and completion. Satisfaction was assessed through the Client Satisfaction Questionnaire and qualitative feedback. Thirty-one caregivers consented to this pilot feasibility trial and participated via telehealth over the course of 5 months. Intervention completion and reported satisfaction were high, with 94% of consented participants completing at least three training sessions and 84% reporting that they would "definitely" recommend the training to a friend in similar circumstances. There were no between-clinician differences in MILO session attendance ($F_{[2]} = 0.53$, $p = 0.596$) or satisfaction total scores ($F_{[2]} = 1.03$, $p = 0.371$). Brief motivational interviewing skills training appears to be a feasible and valued intervention for caregivers of individuals with poorly managed early course psychosis.

Clinical Trial Registration: ClinicalTrials.gov Identifier: NCT04010747

Keywords: schizophrenia, first episode psychosis, motivational interviewing, family, caregiver, clinical trial, feasibility studies

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INTRODUCTION

First episode psychosis (FEP) often represents a time of crisis for young people and their families. Although some psychoses are self-limiting, more often these symptoms portend a potentially chronic and disabling psychiatric disorder such as schizophrenia. Meta-analyses indicate that coordinated multidisciplinary intervention early in the course of psychosis, including family intervention, can alleviate symptoms and restore functioning more effectively than "standard" community treatment (1, 2). The Recovery After Initial Schizophrenia Episode research initiative established that coordinated specialty care for FEP could be feasibly implemented in the United States, and is more effective than treatment as usual for decreasing clinical symptoms, improving quality of life, and increasing participation in school and work (3). However, this study found that many patients entered care with long duration of untreated psychosis (DUP), adding to a consensus that treatment benefits are generally far greater for psychosis patients with shorter as opposed to longer DUP (3–5).

Treatment delay and non-adherence in FEP is a pressing public health problem. A review of privately insured adolescents and young adults in the US showed that 62% of young people in the US with FEP filled no outpatient prescriptions, and 41% received no outpatient psychotherapy, in the year following their index diagnosis (6). Among those who do encounter specialized FEP outpatient care, high attrition is a common problem, with 20-50% of individuals initially enrolled in first episode programs dropping out (7). The reasons underlying long DUP and poor engagement in care are myriad. Many individuals experiencing psychosis are reluctant to seek or adhere to mental health treatments due to lack of insight and/or concerns about the usefulness of psychiatric interventions (8, 9). Young adults may be torn between distress and dissatisfaction relating to their symptoms and functioning on the one hand, and mistrust of mental health providers, treatments, and labels on the other (10). Family members and other loved ones often endure confusion and distress as they endeavor to convince the individual with psychosis (IP) to accept and utilize psychiatric services (11–13).

Motivational interviewing (MI) is a well-established strategy for facilitating behavior change across a wide range of treatment targets, including enhanced adherence to treatment. The theme of MI is non-judgmental exploration of ambivalence regarding behavior change (14). MI is not didactic or confrontational; rather, it is a set of communication strategies designed to decrease defensiveness and rigidity. Clinician-delivered MI has been identified as effective for enhancing adherence once individuals with psychosis are involved in care (15, 16), and may also be useful for engaging those who are not yet interested in treatment (14). Several studies have found positive results in training and deploying non-professionals to use MI to influence target health behaviors such as substance use and diet (17, 18). Only one study to date has trained parents to use MI in the context of recent-onset schizophrenia; the authors reported that individuals whose caregivers learned MI used less cannabis and had less severe symptoms over the following 15 months than those whose families received routine care (19–21). MI-derived communication training for caregivers may represent a promising approach through which parents or other relatives may be able to improve relationships, decrease conflict, and influence a loved one's decision to seek care and adhere to treatment plans (20, 22–25).

The aim of the current study is to develop and test the feasibility of a brief MI-derived psychoeducational intervention for parents and other close contacts of individuals with early course psychosis who are sub-optimally engaged with treatment. The goal is not that the caregiver becomes a "therapist" to the individual with psychosis (IP), but rather that they learn and use MI-based communication strategies to decrease conflict in the relationship and play a more effective role in helping to connect the IP to relevant clinical services. The aim of this paper is to describe the development of the intervention and study procedures, determine the feasibility of the pilot protocol, and assess participants' satisfaction with the intervention.

MATERIALS AND METHODS

Intervention Development

The author group conducted stakeholder interviews to inform the development of the "motivational interviewing for loved ones" (MILO) intervention. Consultants with lived experience were interviewed about the process of seeking care for themselves or their child, and their impressions of family needs during care initiations and transitions in general. We then attended formal trainings for providers offered by certified trainers in motivational interviewing (EK) and Community Reinforcement and Family Training (HT), a related evidence-based practice that teaches skills pertaining to behavior change and reflective listening to caregivers of individuals with substance use disorders. We also consulted with a Motivation Interviewing Network of Trainers-certified trainer (Angela Cooper) about the curriculum structure of MI training for clinical providers. After these meetings, we (EK, HT, AS, KE) reviewed our notes and impressions to reach a consensus on which core MI-consistent skills to include in the training. Once the core skills were identified (see **Table 1**), we created a manual for clinicians to use in MILO sessions. Clinicians were to both teach the MILO skills and also model them consistently during sessions by being fairly non-directive, for example asking open-ended questions and using reflections to help caregivers process their own ambivalence about using MILO skills or other dilemmas.

Concerns about feasibility and cultural relevancy were of foremost consideration in designing the content and duration of this intervention. In order to maximize feasibility and minimize burden to participants, we prioritized keeping both the intervention and the assessment battery brief. The intervention was designed to be completed in four 45–60 min sessions, and the assessment battery in 25 min or less. Additionally, we strove to create a culturally conscious intervention informed by diverse needs and perspectives that would not need to be "adapted" at a later point to fit the cultures and concerns of non-white families (26). To do so, we consulted stakeholders representing a diverse range of cultural backgrounds and relevant lived experiences throughout intervention development, minimized the use of

TABLE 1 | Motivational interviewing for loved ones: session content.

Core skills	• The "spirit" of motivational interviewing • Learning not to fix or minimize others' problems • Reflections • Questions • Affirmations • Raising difficult topics • Obtaining permission before giving advice
Session structure	• Session 1: Review the individual with psychosis's current treatment status, well-being, treatment history, and relationship with the participant. If needed, offer information about relevant treatment (e.g., coordinated specialty care). Inquire about impact of illness on participant. Teach participant about the concept of motivational interviewing (MI) and the "spirit" of MI. • Session 2: Teach and practice reflections, open-ended questions, and affirmations • Session 3: Teach and practice raising difficult topics and asking permission before giving advice • Session 4: Review a conversation, plan a conversation, and/or devote more time to in-session practice

psychological jargon in the manual, and chose images for the manual that represented diverse families.

Just as recruitment for the study was beginning (February 2020), the COVID-19 pandemic struck the United States and non-essential in-person activities were suspended indefinitely at the study site (Beth Israel Deaconess Medical Center). At this point, MILO was re-designed as a telehealth intervention, and the manual was translated into a digital slide deck that would be shared over the screen with caregivers during a video-conference meeting.

MILO facilitators included the first author (EK) as well as two additional clinicians (BD, AF). The first author and senior clinician (EK) is a licensed doctoral level psychologist with training in both psychosis treatment and motivational interviewing. The other study clinicians were a post-doctoral psychology fellow (BD) and an advanced student in a clinical psychology doctoral program (AF). Clinicians trained in the intervention by reviewing the manual with the first author, observing her in three MILO sessions, and discussing cases with her weekly. Fidelity was assessed by each clinician documenting which MILO-relevant skills and themes were covered in each session and reviewing these in supervision sessions.

Procedures

Eligibility requirements for caregiver-participants were: age 18 or older, able to communicate in English, a primary caregiver and/or close contact who has ≥20 h weekly contact with an IP, and able to provide informed consent. Additionally, in order for caregivers to be eligible, the IP had to be 15–35 years old, diagnosed with a DSM-5 affective or non-affective psychotic disorder by a health professional OR have observable symptoms or behaviors (e.g., responding to internal stimuli, describing delusional ideas, or showing grossly disorganized speech or behavior) indicating psychosis, with onset of observed symptoms or first psychosis diagnosis within past 5 years. IP were either untreated or not optimally engaged in outpatient treatment (e.g., not adhering to prescribed medications, using substances in conflict with the treatment plan, or refusing to meet with providers). Due to the inevitable diagnostic uncertainty of relying only on caregiver report, study staff attempted to obtain collateral diagnostic information from another source when the diagnosis seemed very unclear. After each participant completed MILO sessions, the study clinician revisited the most likely diagnoses to confirm the presence of recent-onset psychosis.

The recruitment goal for the feasibility phase of the study was set at thirty. To recruit participants, the study's first author sent study information to clinicians and referral coordinators at FEP programs in the Boston area as well as through a national (U.S.) early psychosis-focused listserv. Clinicians and referral coordinators were encouraged to let potential participants know about the study depending on their clinical judgment and institutional policies.

This protocol was reviewed and approved by the Beth Israel Deaconess Medical Center institutional review board. Potential participants were screened for eligibility over the phone. If they were eligible, they then provided verbal informed consent via telephone. Self-report assessments were emailed to participants via a secure Redcap survey link. Participants were then asked to schedule a brief call with research staff for a pre-intervention recorded "real-play" in which research staff described a personal dilemma and asked participants to discuss it with them for 10 min. Once all pre-intervention assessments were completed, participants scheduled an initial session with a study clinician. All MILO sessions were conducted using a secure telehealth platform called Starleaf (as required by the institutional review board). After the intervention was concluded, participants recorded a second "real-play" and were emailed surveys at 0-, 8-, and 12-weeks following intervention completion. Participants were reimbursed for completing assessments ($25 per time point). No reimbursement was provided for attending MILO sessions.

Measures

The following domains were selected to measure MILO feasibility: number/pace of inquiries, percent of inquiries eligible for participation, percent of eligible trial candidates who enrolled (the goal was two participants per month), intervention completion (number of sessions attended), assessment completion, and participant satisfaction. Participants were assessed in their first MILO session by a clinician to determine whether they met criteria for an adjustment disorder as a consequence of their loved one's psychotic illness, using the adjustment disorder section of the Structured Clinical Interview for DSM-5 (27). Participant satisfaction was measured at the post-intervention assessment via a seven-item version of the Client Satisfaction Questionnaire (28) (one original item about returning for additional services was omitted). We also surveyed participants on whether they had tried using MILO skills with the IP. Participants were then prompted to respond to three open-ended questions: what they had found helpful about MILO, suggestions for improving MILO, and what barriers they encountered to MILO skills with the IP.

The following domains were selected to measure MILO effects and were administered at each of the four assessment time points: past-month treatment attendance and adherence by the IP (as reported by the caregiver); expressed emotion [measured via the Family Questionnaire; Wiedemann et al. (29)]; family conflict and cohesion [measured via the Conflict Behavior Questionnaire, Robin and Foster (30); and the Score-15, Stratton et al. (31), respectively]; self-efficacy [measures via the Parenting Self-Agency Measure, Dumka et al. (32); and the General Self-Efficacy Scale, Chen et al. (33)]; and stress [Perceived Stress Scale; Roberti et al. (34)]. To assess the extent to which participants were able to learn and demonstrate MI skills (i.e., target engagement), caregivers completed a 10-item test of their knowledge of MI concepts and an audio-recorded behavioral skill demonstration at the baseline and immediate post-intervention time points. The present study reports on the feasibility rather than the effects of MILO.

Analyses

Feasibility targets and client satisfaction were assessed using descriptive analyses only. Differences in MILO completion rates and client satisfaction scores between study clinicians were assessed via one-way ANOVA using SPSS. Qualitative responses

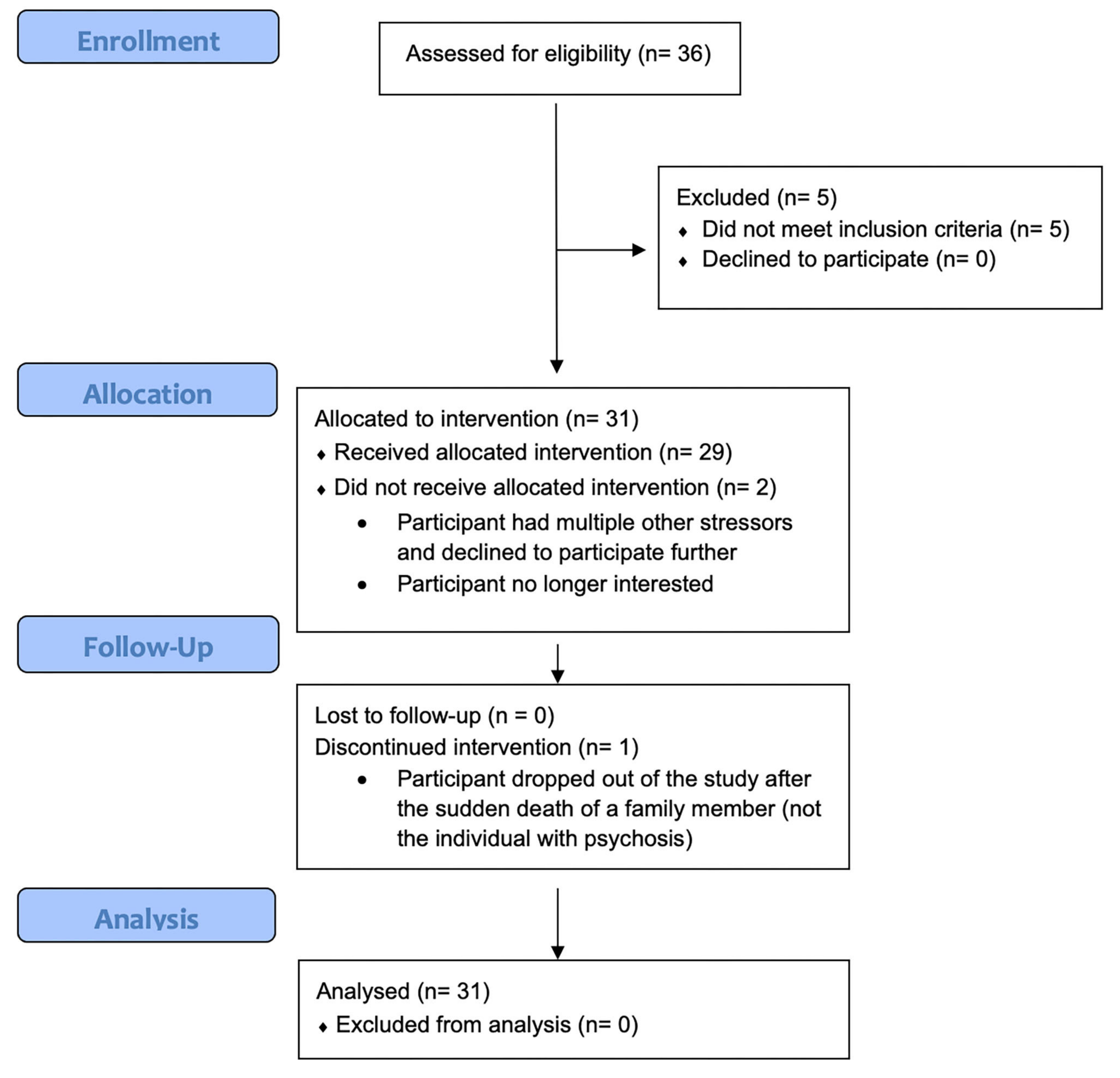

FIGURE 1 | CONSORT flow diagram.

were reviewed by the first author, who conducted an inductive thematic analysis to summarize responses (35).

RESULTS

Feasibility

See **Figure 1** for a CONSORT flow diagram reflecting this pilot feasibility trial.

A total of 43 people contacted study staff via email or phone to inquire about MILO participation between May 1, 2020 and September 11, 2020. Thirty-six were assessed by study staff and 31 were found to be eligible. Reasons for non-eligibility included not having a loved one with a psychotic disorder, duration of psychotic illness more than 5 years, IP older than 35, and being neither a primary caregiving nor spending at least 20 h weekly with the IP. Three participants who did not know their child's diagnosis were determined to meet eligibility criteria, since the participants each described a qualifying symptom that they and others had observed (delusional pre-occupation and/or disorganized speech) that had begun in the past five years, and the child had declined to participate in a psychiatric evaluation.

All eligible participants ($N = 31$) representing 25 families (some caregivers enrolled along with or subsequent to a co-parent) enrolled for the "phase 1" feasibility stage of this pilot trial. Participant characteristics are listed in **Table 2**. On average, the time from initial contact to informed consent was 3.6 days, and from consent to first MILO session was 16.7 days.

Two of the 31 participants dropped out of the study prior to attending any MILO sessions. The remaining 29 attended at least three sessions of MILO, and 27 fully completed the intervention.

All 31 participants completed baseline (pre-training) survey assessments and recorded skills demonstrations. The two participants who dropped out prior to participating in MILO sessions were not contacted for post-intervention assessments. Of the 29 who attended at least three sessions of MILO, 28 completed post-intervention surveys and recorded skill demonstrations.

Satisfaction

Scores from the CSQ (sent to participants within one week of MILO completion) are displayed in **Table 3**. Twenty-five of the 28 participants (89%) who completed post-intervention assessments reported that they had used the skills they learned in the intervention when communicating with the IP. An incidental finding was that at least eight of the 29 caregivers who participated in MILO sessions recommended the study to a family member or other social contact, suggesting high client satisfaction.

The PI (EK) was the study clinician for 19 participants, while co-authors BD and AF were the study clinicians for four and six participants respectively. There were no between-clinician differences in MILO session attendance [$F_{(2)} = 0.53, p = 0.596$] or CSQ total scores [$F_{(2)} = 1.03, p = 0.371$].

Results from the thematic analysis of participants' written qualitative responses are displayed in **Table 4**. Themes that emerged were participants' enthusiasm for the MILO principles and skills, their desire for additional memory aids and practice opportunities so that they could feel more confident using MILO skills, and their eagerness and ability to implement their newly acquired skills with their teen/young adult children.

TABLE 2 | Participant Characteristics ($N = 31$).

Participant characteristics	
Age	Range: 45-71 Mean (SD): 57.97 (7.43)
Gender	Male: 8 (26%) Female: 23 (74%)
Relationship to individual with psychosis	Parent: 31 (100%) Other: 0 (0%)
Residing with individual with psychosis	Yes: 19 (61%) No: 12 (39%)
Race	White: 25 (81%) Black: 1 (3%) Asian: 3 (10%) Other: 1 (3%) Prefer not to say: 1 (3%)
Ethnicity	Hispanic/Latino: 1 (3%) Not Hispanic/Latino: 30 (97%)
Immigration history	Born in United States: 26 (84%) Born elsewhere: 5 (16%)
Educational attainment	High school diploma or higher: 31 (100%) Bachelor's Degree or higher: 25 (81%)
Adjustment disorder diagnosis	Adjustment disorder: 14 (45%) No adjustment disorder: 14 (45%)[a] Missing: 3 (10%)
Characteristics of Individuals with Psychosis (as reported by participant)	
Age	Range: 16-30 Mean (SD): 23.13 (3.89)
Gender	Male: 26 (84%) Female: 4 (13%) Unknown: 1 (3%)
Diagnosis	Schizophrenia: 6 (19%) Schizoaffective disorder: 9 (29%) Schizophreniform disorder: 2 (6%) Bipolar disorder with psychotic features: 6 (19%) Clinical high-risk for psychosis: 1 (3%) Other unspecified psychosis: 4 (13%) Unknown: 3 (10%)
Co-occurring substance use	Yes, current: 21 (68%) Yes, past: 2 (6%) No: 6 (19%) Unknown: 2 (6%)
Duration of psychotic illness (years)[b]	Range: 0.25-4.67 Mean (SD): 2.10 (1.32)
History of psychiatric hospitalization	Yes: 23 (74%) No: 8 (26%)
Past-month psychiatric service utilization	Stayed overnight in hospital: 8 (26%) Visited emergency room: 10 (32%) Took any medication: 19 (61%) Took medication as prescribed: 10 (32%) Attended ≥1 outpatient appointment: 13 (42%)

[a] *Two participants who did not meet DSM-5 criteria for Adjustment Disorder disclosed that they had other established diagnoses of Major Depressive Disorder and Post-Traumatic Stress Disorder, respectively.*

[b] *$N = 27$; duration of illness could not be estimated for those with unknown or CHR diagnosis.*

TABLE 3 | Participant satisfaction ($N = 28$).

Item (response range for each is 0–3, with "0" representing poor satisfaction and "3" representing full satisfaction)	Mean (SD)
How would you rate the quality of service you have received?	2.89 (0.31)
Did you get the kind of service you wanted?	2.57 (0.50)
To what extent has our program met your needs?	2.50 (0.58)
If a friend were in need of similar help, would you recommend our program to them?	2.93 (0.26)
How satisfied are you with the amount of help you have received?	2.71 (0.46)
Have the services you received helped you to deal more effectively with your problems?	2.68 (0.55)
In an overall, general sense, how satisfied are you with the service you have received?	2.86 (0.36)

TABLE 4 | Qualitative response themes ($N = 28$).

Prompt	Identified theme (number of responses within this theme)
What have you found most helpful about this program?	MILO skills (22) Motivational interviewing "spirit" (7) Expertise and/or empathy of facilitator (6) Individualized advice about a specific family situation (6) Role plays (6) Convenience of telehealth (1)
What changes would improve this program in the future?	Offer more sessions and practice opportunities (11) Change wording/response options in one or more questionnaire (4) No changes (3) Provide scripts or memory aids to help with skill implementation (3) Improve telehealth platform (2) Offer training in a group format (2) Expand to diagnoses beyond FEP (2) Provide more rationale for motivational interviewing (1)
What barriers to implementing the MILO skills did you experience?	No barriers (9) Limited contact with the individual with psychosis (7) Not enough training/practice (5) IP not responsive to attempts to use skills (4) Difficulty managing own emotions during conversations (3) Not enough time for longer conversations (1)

DISCUSSION

The results of this pilot feasibility study indicate that MILO is a feasible brief intervention for parents of individuals suffering from early-course psychosis. The pace of recruitment proceeded more quickly than anticipated. Clinical staff at FEP treatment centers supported the intervention by sharing information about the study with families, and as the study progressed, additional participants were referred to the study by past participants who encouraged others in their families, support groups, or social networks to participate.

All participants were parents of IP, and many expressed gratitude for a resource that they could access even though their children were refusing to participate in treatment. Parents of untreated or under-treated individuals with early course psychosis may represent an underserved constituency. Although a number of participants expressed optimism that the MILO skills could be useful in facilitating their loved one's connection with or adherence to psychiatric care, for many participants, this outcome was secondary to their general relief at receiving guidance that would reduce overall conflict and stress in their family relationships. Illustrating this sentiment, one participant wrote that "[It was helpful to] learn specific techniques for interacting in difficult conversations and situations to produce a different outcome, to empower my child to assume more control for decisions affecting her life, [and] to feel heard and understood by someone with a relevant skill set who encouraged me and gently challenged my thinking so I could shift and think about a situation in a different way."

Retention over the course of the intervention was strong. No participants dropped out after one or two sessions. Ninety percent of consented participants completed at least three MILO sessions as well as pre- and post-intervention assessments. This is favorable relative to median drop-out rate for non-pharmacologic interventions in schizophrenia, which a 2017 meta-analysis estimated as 19% (36). Three participants requested and were granted a fifth session to obtain additional coaching on how to use MILO skills with the IP. This is notable in the context of the study design, in which participants were not reimbursed for attending study sessions.

Participant satisfaction with the MILO intervention was high. Twenty-six out of 28 participants who completed a post-intervention satisfaction survey reported that they would "definitely" recommend the service to a friend in need of similar help. Qualitative responses to satisfaction-related prompts described how participants appreciated acquiring concrete communication skills, learning the philosophy of MI, receiving individualized advice, practicing skills via role plays, working with MILO clinicians, and meeting via telehealth. Satisfaction may have been influenced not only by MILO session content but also by the fast turnaround from inquiry to enrollment to first MILO session. The MILO team prioritized responding to inquiries and making eligibility decisions quickly. This required some tolerance of uncertainty with regard to IP who did not have a well-established (e.g., via inpatient hospital record) diagnosis of a primary psychotic disorder.

The primary theme that emerged in participants' suggestions for strengthening the intervention was that they felt they needed more time to review and practice the skills. Some participants noted that the skills were difficult to remember, especially in stressful moments. In response, the author group is creating a short video series explaining and illustrating the skills, which caregivers can watch on demand to help them remember the skills, which will be publicly available when complete. Additionally, we may consider offering an optional fifth or even sixth session a few weeks or months after the four "core" sessions, so that caregivers can practice the skills again and discuss difficulties that may arise over time.

A weakness of this study is that non-Hispanic whites and individuals with college degrees are over-represented in the study sample. Overall, these groups tend to be over-represented in clinical trial samples in the U.S. (37–39); this trend was likely exacerbated by the use of telehealth as a modality. Individuals without college degrees may have encountered barriers such as lack of high-quality internet, devices compatible with video conferencing, or paid time off to seek out mental health support. In phase two of this study (pilot efficacy trial), study authors will partner with a FEP clinic that primarily serves under-represented minority groups and offer some MILO sessions in-person rather than via telehealth. Another inherent weakness of the study design is uncertainty regarding the accuracy of parent-reported diagnoses, treatment utilization, and adherence. Even co-parents who both participated in the study sometimes disagreed on their child's medication adherence. In some cases, this could be because co-parents staggered their participation by a month or more; in others, the divergence in their reporting may be due simply to their differing perceptions of the situation. In phase 2 of this study, the study team will ask participants representing multiple members of the same family to reconcile any divergent responses relevant to the IP's treatment history and utilization.

Recruitment and data collection for this study took place at the height of the COVID-19 crisis in the United States, from May through December 2020. This may have impacted the results in a few ways. First, the telehealth modality, which was not part of the original study design, was well-received by study participants and increased the pool of potential participants beyond the Boston metro area. Second, caregivers may have been especially interested in learning new strategies to address conflict during this time when many were sheltering in place with their families and experiencing unfamiliar stressors. Third, two participants disclosed that they contracted COVID during the course of their study participation but elected to remain in the study while they isolated and convalesced.

The next steps following this pilot feasibility study are to 1, implement small changes to the intervention recommended by phase one participants; 2, alter the recruitment strategy to obtain a more demographically diverse sample; and 3, move to a randomization design that will enable evaluation of the impact of MILO relative to a control condition. In phase two of this study, participants will be randomly assigned to either immediate MILO or a six-week waitlist condition, after which they will be offered MILO sessions, which will enable evaluation of intervention effects. Effects will be evaluated based on intent to treat analysis. Overall, MILO appears to be a highly feasible intervention that yielded strong retention and very high satisfaction among participating caregivers. Intentional consultation at multiple stages of intervention development and study design with a range of experts and individuals with lived experience likely contributed to the design of a feasible and well-received intervention and assessment battery.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Committee on Clinical Investigation Institutional Review Board Beth Israel Deaconess Medical Center. Written informed consent for participation was not required for this study in accordance with the national legislation and the institutional requirements.

AUTHOR CONTRIBUTIONS

EK was responsible for the study design, supervision of the study team, accurate reporting of data, and drafting this manuscript. HT, AS, MK, and KE contributed to intervention development, measure selection, and participant recruitment. BD and AF contributed to intervention design and interpretation of results. All authors have reviewed and approved this submission.

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Associations of Caregiving Knowledge and Skills With Caregiver Burden, Psychological Well-Being, and Coping Styles Among Primary Family Caregivers of People Living With Schizophrenia in China

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Background: There is a lack of clarity regarding the correlation of caregiving knowledge and skills with caregiving experiences of people living with schizophrenia (PLSs). To address this gap, this comprehensive study examines the relationships of caregiving knowledge and skills to the primary family caregiver's experiences of burden, psychological well-being (stress, anxiety, depression, caregiving rewarding feelings), and coping styles in China.

Methods: A total of 395 primary family caregivers of PLSs were enrolled in a cross-sectional study between May 2019 and September 2019. Each family caregiver was independently assessed on caregiving knowledge and skills, caregiver burden, and psychological well-being, as well as coping styles.

Results: A higher level of caregiving knowledge and skills was positively correlated with less stress ($b = -0.48$, $P < 0.001$), anxiety ($b = -0.23$, $P = 0.029$), depression ($b = -0.29$, $P = 0.013$), and more caregiving rewarding feelings ($b = 0.54$, $P < 0.001$). Also, caregivers with more knowledge and skills were more inclined to adopt positive coping strategies ($b = 0.44$, $P < 0.001$). Despite these differences, caregivers with different levels of caregiving knowledge and skills reported comparable caregiver burden ($b = 0.11$, $P = 0.705$) and the use of a passive coping style ($b = 0.10$, $P = 0.169$).

Conclusion: Caregiving knowledge and skills are a reliable predictor of psychological well-being and active coping among the primary family caregivers of PLSs. These findings inform the development of psychoeducational interventions to support family caregivers of PLSs.

Keywords: schizophrenia, caregiving knowledge and skills, caregiver burden, psychological well-being, coping styles, primary family caregivers

INTRODUCTION

With a prevalence of 1–3% worldwide (1), schizophrenia is a psychiatric disorder characterized by profound disturbance in language, perception, thinking, and frequent psychotic experiences (2). People living with schizophrenia (PLSs) are characterized by positive symptoms such as delusions and hallucinations (3). They may also present negative symptoms reflected in alteration of motivation and volition, such as social withdrawal, difficulty in maintaining interpersonal relationships, apathy, and anhedonia (4–6). PLSs may also have other accompanying symptoms, such as cognition impairment and emotional disorder (e.g., mania and depression) (7, 8). Schizophrenia can occur at different stages of life course, while the risk varies across different age groups (9). Reports have shown that the prevalence of schizophrenia changes with age in an inverted U-shape, with the highest value at around 40 years of age. Globally, PLSs aged 25–54 years are estimated to account for 70.8% of total cases. There is no sexual difference in the prevalence (10). PLSs usually have reduced life expectancy in comparison with individuals from the general population (11). This disorder afflicts both PLSs and their family caregivers (12), particularly in countries with insufficient mental health resources and community health service systems (13). In China, more than 90% of PLSs live with their family members and receive family care because of insufficient community health resources (14).

Caring for a family member with schizophrenia requires considerable time, energy, and money from the family caregivers over a prolonged period, which may result in increased caregiver burden and psychological distress (15, 16). Numerous studies have demonstrated a high level of burden among caregivers of PLSs (12, 15, 17, 18) and psychological distress, such as stress, anxiety, and depression (19). For instance, more than half of caregivers report a moderate to severe level of caregiver burden (18), whereas the prevalence of psychological distress is as high as nearly 80% (1, 20). Notably, high levels of caregiver burden and psychological distress among caregivers may also impair the mental health of PLSs (21), who tend to experience physical or verbal domestic violence from their distressed caregivers (22, 23).

Despite significant caregiver burden and distress, there are also reports of positive impacts of caregiving (24, 25). Caring for PLSs may lead to positive transformations among caregivers (26, 27) and promote positive caregiving rewarding feelings, such as enhanced self-satisfaction, self-esteem, self-confidence, and self-affirmation (28, 29). Moreover, caring for PLSs can lead to improvements in stress-coping styles of caregivers (30), which are behavioral coping strategies adopted by individuals to resolve adversities and stressors (31). Evidence has shown that active coping is also associated with better health outcomes (32).

Over the past few decades, various interventions have been developed to support family caregivers of PLSs, of which psychoeducation is the most common and widely shown to be effective (33). Psychoeducation provides PLSs and family caregivers information about strategies and resources to promote the goals of treatment and rehabilitation (33). For caregivers, the key element of psychoeducation lies in improving knowledge and skills in caregiving (34). However, inconsistent findings about the correlations between caregiving knowledge and skills and caregiver burden have been reported by Sefasi et al. (35) and Jagannathan et al. (15). Sefasi et al. found caregivers of PLSs with more knowledge tended to bear greater burden, whereas Jagannathan et al. suggested there was no relationship of caregiving knowledge with burden. To date, no quantitative studies exist that examine the associations of caregiving knowledge and skills with psychological well-being (stress, anxiety, depression, caregiving rewarding feelings) and coping styles (active coping) among the primary family caregivers of PLSs in China.

The theoretical background of this study refers to the stress process model proposed by Pearlin et al. and their assumption that stress results from an interaction of two domains: stressors and mediators (36). Each of these domains can be divided into a series of subparts that have been intensively studied and developed in recent years. According to this model, it can be inferred that stress perceived by caregivers is partially mediated by coping and individual appraisal. Elevated caregiving knowledge and skills help enhance caregivers' ability to cope with adversity and enable them to adopt active coping strategies to address current dilemmas, thus contributing to the reduction of stress (37). Individual appraisal involves subjective assessment on controls of adversity and self-worth. A high level of caregiving knowledge and skills enables caregivers to resolve difficulties in the process of caring for PLSs, thus contributing to increased feelings of personal accomplishment and self-efficacy (38, 39). Furthermore, caregiving knowledge itself is a mediator in the path between stressors and burden (36, 40). Therefore, in the conceptual framework of this study, we assumed that caregiving knowledge and skills may be associated with caregiving experiences (caregiver burden, stress, coping styles, etc.) in a direct or indirect path.

The aim of this study was to evaluate the relationships between caregiving knowledge and skills and a range of caregiving experiences among primary family caregivers of PLSs in China.

METHODS

Participants and Procedure

This cross-sectional study using one-stage cluster sampling was conducted in 12 community health centers of Changsha City, Hunan Province. The targeted population for this study was the primary family caregivers of PLSs who were registered in the "686 Program," China's largest demonstration project in mental health services (41, 42). For each person living with schizophrenia, only one primary family caregiver was enrolled in this study based on closeness with PLSs and direct involvement in caregiving. The inclusion criteria were as follows: (1) aged over 18 years; (2) caring for individuals diagnosed with schizophrenia according to the criteria of Chinese Classification of Mental Disorders Version 3 (43) or the *International Classification of Diseases, Tenth Revision* (44); (3) living with PLSs for over 2 years and taking major responsibility for caregiving; (4) able to understand, read, and communicate with investigators in Chinese. The exclusion criteria included (1) caring for PLSs who received a diagnosis other than schizophrenia; (2) unable to speak Chinese; and (3) lack of literacy, or having serious physical or mental diseases that made them unable to communicate effectively with others.

Data were collected by well-trained researchers from May 2019 to September 2019. First, all participants were informed about the purpose of the study and provided written informed consent before participating. Second, the overall content of questionnaires was introduced to each eligible participant, and they were invited to complete the questionnaires (see assessment below) by face-to-face interviews. Next, a quality-control investigator checked the answers to ensure accuracy, integrity, and consistency. A total of 414 primary family caregivers volunteered to participate in this study, of which 395 participants completed the questionnaire (response rate = 95.41%). No significant difference was detected between individuals who completed and those who did not complete the survey in terms of gender, age, marriage, education, and employment.

The institutional review board of the Xiangya School of Public Health of Central South University approved the protocol of this study. Participants who completed the questionnaire were reimbursed with RMB 20 yuan in cash ($3).

Data Collection

Sociodemographic Data

Sociodemographic data of the primary family caregivers were collected by face-to-face interviews, including sex, age, marital status, education, employment status, financial circumstances (annual income per household), length of caregiving, and the relationship between PLSs and the primary caregivers. Besides, whether there were any co-caregivers, additional dependents, and physical illnesses were also recorded.

Caregiving Knowledge and Skills

The understanding of schizophrenia and mastery of caregiving skills were evaluated using the Knowledge and Skills of Caregiving Scale (KSCS). The KSCS is a five-item questionnaire, which was originally designed according to the survey on the demands of mental health knowledge among the Chinese caregivers of PLSs (45–47). Items are scored on a 4-point Likert scale from 0 (do not understand) to 3 (totally understand) to measure the caregiver's understanding of the following aspects: the symptoms of schizophrenia, medication and its side effects, the care of PLSs, and management of symptoms. The total KSCS score ranges from 0 to 15, with a higher score demonstrating higher knowledge of schizophrenia and better skills in caregiving. The detailed contents of the KSCS are illustrated in **Supplementary Table 1**. The KSCS showed good internal consistency in the current study, with a Cronbach α of 0.87.

Caregiver Burden

Caregiver burden was evaluated using the Zarit Burden Interview (ZBI) (48). The ZBI consists of 22 items, and each item is rated on a 5-point response scale from 0 (never) to 4 (nearly always), except for the final item evaluating global burden from 0 (not at all) to 4 (extremely). The total score ranges from 0 to 88, and higher scores are indicative of higher burden perceived by caregivers. This instrument was first translated into Chinese by Lu et al. (49) in 2009 and found to be reliable and valid in evaluating caregiver burden (50, 51). The ZBI also demonstrated excellent internal consistency in this sample, with a Cronbach α of 0.92.

Stress

Perceived stress was assessed using the 10-item Perceived Stress Scale (PSS-10) (52). This instrument covers two domains: perceived helplessness (six items) and perceived self-efficacy (four items) (53, 54). Each item is scored on a 5-point Likert scale from 0 (never) to 4 (often); the total score ranges from 0 to 40, with higher scores indicating greater perceived stress. First translated into Chinese by Yang et al. (55) in 2003, the PSS-10 is reliable in evaluating perceived stress (56, 57). In this study, the PSS-10 exhibited acceptable internal consistency, with a Cronbach α of 0.79.

Anxiety

The seven-item Generalized Anxiety Disorder Scale (GAD-7) (58) was used to reflect the anxiety symptoms of primary family caregivers in the past 2 weeks. Items rated on a 4-point Likert scale from 0 (not at all) to 3 (nearly every day) are summed into a total score, which ranges from 0 to 21, and a cutoff score of 10 has been suggested to differentiate anxiety and non-anxiety (59). The GAD-7 was first translated into Chinese by He et al. (60) in 2010 and is a valid and reliable measure of anxiety among the Chinese population (61). In this study, the GAD-7 demonstrated good internal consistency with a Cronbach α of 0.95.

Depression

The depression symptoms of primary family caregivers were screened using the self-reported Patient Health Questionnaire (PHQ-9) (62). The PHQ-9 consists of nine items rated on a 4-point Likert scale from 0 (not at all) to 3 (nearly every day) to evaluate the frequency of depression symptoms in the preceding 2 weeks. The total score ranges from 0 to 27, with higher scores indicative of more depression symptoms. The PHQ-9 was first translated into Chinese by Yeung et al. (63) in 2008 and showed good validity and reliability in Chinese context (64). In the present study, the Cronbach α for the PHQ-9 was 0.93.

Caregiving Rewarding Feelings

The Caregiving Rewarding Feelings (CRF) scale was applied to evaluate positive feelings about caregiving (65). Thirty individuals with schizophrenia and their primary caregivers were qualitatively interviewed for the initial development of the CRF, pretesting, and Delphi method for further revision and then a test with a larger sample to confirm its validity and reliability. The detailed process of development and validation of the CRF has been illustrated elsewhere (65). The CRF is a 12-item questionnaire developed in a Chinese context, and each item is scored on a 4-point Likert scale (0 = "never" to 3 = "nearly always") to assess whether caring for PLSs makes the primary caregivers "become more loving and patient," "feels more worthy," "be more active and optimistic," "have stronger sense of responsibility," etc. The total score ranges from 0 to 36,

TABLE 1 | Sociodemographic characteristics of the caregivers and comparison of KSCS scores between groups.

Variables		No. (%)	KSCS score		
			Median (IQR)	*Z/H*	*P*
Gender	Male	178 (45.06)	10 (8–11)	−0.572	0.567
	Female	217 (54.94)	10 (8–12)		
Age (years)	18–44	38 (9.62)	10 (7–11)	5.151	0.076
	45–59	105 (26.58)	10 (9–12)		
	≥60	252 (63.80)	10 (8–11)		
Marital status	Not married[†]	93 (23.54)	10 (7–11)	−1.981	**0.048**
	Currently married[††]	302 (76.46)	10 (8–12)		
Education	Primary	123 (31.14)	10 (7–11)	6.141	**0.046**
	Middle	238 (60.25)	10 (9–12)		
	High	34 (8.61)	10 (8.75–10)		
Employment status	Unemployed	342 (86.58)	10 (8–11)	−0.807	0.420
	Employed	53 (13.42)	10 (9–13)		
Annual income (yuan/person)	<10,000	236 (59.75)	10 (8–11)	2.750	0.253
	10,000–19,999	112 (28.35)	10 (8–12)		
	≥20,000	47 (11.90)	10 (9–11)		
Length of caring (years)	<10	75 (18.99)	10 (9–11)	−0.567	0..570
	≥10	320 (81.01)	10 (8–11)		
Relationship to the PLSs	Parents	231 (58.48)	10 (8–11)	4.378	0.357
	Spouse	98 (24.81)	10 (8–13)		
	Children	23 (5.82)	10 (9–11)		
	Siblings	34 (8.61)	10 (7–10)		
	Others	9 (2.28)	10 (6–10)		
Co-caregiver	No	221 (55.95)	10 (8–12)	−0.991	0.322
	Yes	174 (44.05)	10 (8–11)		
Additional dependents	No	236 (59.75)	10 (8–11)	−0.109	0.913
	Yes	159 (40.25)	10 (8–12)		
Physical illness	No	156 (39.49)	10 (8–12)	−1.018	0.308
	Yes	239 (60.51)	10 (8–11)		

KSCS, Knowledge and Skill of Caregiving Scale; IQR, interquartile range; PLSs, people living with schizophrenia.

[†] *Includes single, widowed, divorced, and separated.*

[††] *Includes married and cohabited.*

Values in bold font are statistically significant (P < 0.05).

with a higher score suggesting better positive feelings. In this study, the CRF demonstrated excellent internal consistency with a Cronbach α of 0.96.

Coping Styles

The Simplified Coping Style Questionnaire (SCSQ) was used to measure a caregiver's coping style (66). This measure is a 4-point Likert questionnaire (0 = "never" to 3 = "nearly always") that comprises 20 items classified into two dimensions: active coping (12 items) and passive coping (eight items). Active coping mainly reflects one's active coping preference when encountering difficulties, such as "working out several different ways to solve the problem." Passive coping mainly reflects one's passive coping tendency when encountering difficulties, such as "relying on others to solve the problem." The total score ranges from 0 to 60, and a higher score for each dimension indicates a higher possibility that the participant would adopt the corresponding coping style. Good reliability and validity of the SCSQ in a Chinese context were reported by Xie (66), and a Cronbach α of 0.88 for the total scale, 0.90 for the subscale of active coping, 0.72 for the subscale of passive coping were detected in the current study.

Sample Size

Sample size was calculated using the correlation power and sample size calculation in G*Power. Assuming a correlation of 0.2 between caregiving knowledge and skill and stress, a Z value of 1.98 at a confidence interval of 95%, and allowable α error of 5%, we needed a sample size of 319. Considering a rejection or loss-to-follow-up rate of 10%, we expanded our final sample size to 350. Our study recruited 395 participants, which satisfies the sample size requirement with a power of 95%.

Statistical Analyses

All statistical analyses were performed using STATA software version 16.0. The categorical variables were presented as counts (percentages), whereas the continuous variables were expressed as mean (SD) or median [interquartile range (IQR)]. Normality of data was determined using frequency distributions (histogram), Kolmogorov–Smirnov test, and skewness and kurtosis statistics. The difference of KSCS scores in different sociodemographic groups was checked using Mann–Whitney *U*-test or Kruskal–Wallis *H* test. The correlations between knowledge and skills of caregiving and various types of caregiving experiences (caregiver burden, stress, anxiety, depression, caregiving rewarding feelings, coping styles) were examined by Spearman rank correlation analysis. Adjusted by potential confounders (gender, age, marital status, education, employment, annual income, etc.), several multivariate linear regressions were separately performed to examine the associations between KSCS score and seven types of caregiving experiences among primary family caregivers. Potential multicollinearity between predictor variables was checked by variance inflation factor (VIF), with VIF > 5 indicating collinearity (13). The difference was considered statistically significant at $P < 0.05$.

RESULTS

General Characteristics and Group Comparison of KSCS Score

The sociodemographic characteristics of the participants are described in **Table 1**. There were slightly more females (55%) than males (45%). Most of the caregivers were older than 60 years (64%), were married (76%), had a middle school education (60%), and were unemployed (87%), with an annual family income of <10,000 yuan (60%). The largest proportion of caregivers were parents (58%). Most caregivers had been caring for PLSs for over 10 years (81%) and without co-caregivers (56%). Sixty percent of caregivers had no additional dependents and had physical illness such as hypertension, diabetes mellitus, and cardiovascular diseases.

The median KSCS score was 10, with an IQR of 8–11. We further compared the KSCS score between groups with different sociodemographic characteristics and found no significant difference except for marital status and education. Caregivers who were married, with higher educational attainment, had better caregiving knowledge and skills ($P = 0.048$, $P = 0.046$).

Correlations of Knowledge and Skills With Caregiving Experiences

Table 2 summarizes the caregiving experiences concerning caregiver burden, psychological well-being, and coping styles, divided into negative and positive caregiving experiences. Negative caregiving experiences comprised caregiver burden (mean = 43.21), stress (mean = 18.72), anxiety (mean = 7.68), depression (mean = 8.30), and passive coping (mean = 11.80). Positive caregiving experiences consisted of caregiving rewarding feelings (mean = 26.20) and active coping (mean = 20.02).

Caregivers' knowledge and skills about caregiving were negatively correlated with stress ($r = -0.210$, $P < 0.001$), anxiety ($r = -0.104$, $P = 0.039$), and depression ($r = -0.167$, $P < 0.001$), and positively associated with caregiving rewarding feelings ($r = 0.208$, $P < 0.001$) and active coping ($r = 0.168$, $P < 0.001$). But caregiving knowledge and skills did not correlate with either caregiver burden or passive coping, despite a small trend in favor of a positive correlation.

TABLE 2 | Caregiving impacts and Spearman correlation coefficients between outcomes and KSCS scores.

Variables	Mean (SD)	Spearman correlation	
		r	*P*
Subjective burden (ZBI)	43.21 (18.26)	0.008	0.878
Stress (PSS-10)	18.72 (6.84)	−0.210	**<0.001**
Anxiety (GAD-7)	7.68 (6.45)	−0.104	**0.039**
Depression (PHQ-9)	8.30 (7.26)	−0.167	**<0.001**
CRF	26.20 (9.11)	0.208	**<0.001**
Active coping (SCSQ subscale)	20.02 (7.72)	0.168	**<0.001**
Passive coping (SCSQ subscale)	11.80 (4.54)	0.004	0.930

KSCS, Knowledge and Skills of Caregiving Scale; ZBI, Zarit Burden Interview; PSS-10, 10-item Perceived Stress Scale; GAD-7, seven-item Generalized Anxiety Disorder Scale; PHQ-9, nine-item Patient Health Questionnaire; CRF, Caregiving Rewarding Feelings; SCSQ, Simplified Coping Style Questionnaire; SD, standard deviation.
Values in bold font are statistically significant (P < 0.05).

Multivariate Linear Regression

Table 3 shows the results of several multivariate linear regressions of caregiving knowledge and skills on seven types of caregiving experiences (caregiver burden, stress, anxiety, depression, caregiving rewarding feelings, active coping, passive coping). After controlling for covariates, caregiving knowledge and skills were still significantly negatively associated with stress ($b = -0.48$, $P < 0.001$), anxiety ($b = -0.23$, $P = 0.029$), and depression ($b = -0.29$, $P = 0.013$), as well as positively associated with caregiving rewarding feelings ($b = 0.54$, $P < 0.001$). Besides, primary caregivers with more caregiving knowledge and skills were more inclined to adopt positive coping strategies ($b = 0.44$, $P < 0.001$). No significant relationship was detected between caregiving knowledge and skills with either caregiver burden ($b = 0.11$, $P = 0.705$) or passive coping ($b = 0.10$, $P = 0.169$).

DISCUSSION

To our knowledge, this is the first study that investigates comprehensively the associations of caregiving knowledge and skills with caregiver burden, psychological well-being, and coping styles among primary family caregivers of PLSs in China. Although no relationship was found between caregiving knowledge and skills and caregiver burden, caregiving knowledge and skills were negatively associated with stress, anxiety, and depression and positively associated with caregiving rewarding feelings. In addition, caregivers with more caregiving knowledge and skills were more inclined to adopt positive coping strategies, but not passive coping strategies. These findings add further evidence about the positive associations of caregiving knowledge and skills with a range of health outcomes and

TABLE 3 | Multivariate linear regression analysis of KSCS scores predicting caregiving impacts.

Variables		Caregiver burden (ZBI)	Stress (PSS-10)	Anxiety (GAD-7)	Depression (PHQ-9)	Caregiving rewarding feelings (CRF)	Active coping style (SCSQ subscale)	Passive coping style (SCSQ subscale)
KSCS score		0.11 (−0.45, 0.66)	**−0.48*** (−0.70, −0.27)**	**−0.23* (−0.43, −0.02)**	**−0.29* (−0.52, −0.06)**	**0.54*** (0.25, 0.84)**	**0.44*** (0.20, 0.68)**	0.10 (−0.04, 0.25)
Gender	Male	Ref	Ref	Ref	Ref	Ref	Ref	Ref
	Female	1.67 (−1.99, 5.33)	0.55 (−0.84, 1.95)	0.25 (−1.08, 1.57)	0.76 (−0.75, 2.27)	−0.89 (−2.80, 1.03)	0.63 (−0.94, 2.20)	−0.13 (−1.10, 0.85)
Age		−0.03 (−0.23, 0.18)	0.01 (−0.07, 0.09)	0.00 (−0.08, 0.07)	−0.01 (−0.09, 0.08)	−0.09 (−0.20, 0.02)	**−0.09* (−0.18, −0.00)**	−0.02 (−0.07, 0.03)
Marital status	Not married	Ref	Ref	Ref	Ref	Ref	Ref	Ref
	Currently married	1.11 (−3.35, 5.56)	0.58 (−1.11, 2.28)	1.01 (−0.60, 2.63)	−0.52 (−2.36, 1.32)	0.77 (−1.55, 3.10)	−0.10 (−2.01, 1.81)	0.10 (−1.08, 1.29)
Education	Primary	Ref	Ref	Ref	Ref	Ref	Ref	Ref
	Middle	**5.98** (2.12, 9.85)**	0.34 (−1.13, 1.81)	0.75 (−0.65, 2.15)	0.72 (−0.87, 2.32)	**2.58* (0.55, 4.61)**	**2.78** (1.12, 4.44)**	0.56 (−0.47, 1.59)
	High	4.46 (−2.65, 11.58)	−1.04 (−3.74, 1.67)	0.98 (−1.60, 3.55)	0.99 (−1.94, 3.93)	**5.53** (1.81, 9.25)**	**5.33** (2.28, 8.38)**	0.34 (−1.55, 2.24)
Employment status	Unemployed	Ref	Ref	Ref	Ref	Ref	Ref	Ref
	Employed	−3.95 (−9.60, 1.71)	−1.26 (−3.41, 0.89)	−1.28 (−3.33, 0.76)	−1.80 (−4.13, 0.53)	−0.04 (−2.99, 2.91)	−0.55 (−2.97, 1.87)	−0.56 (−2.07, 0.94)
Annual income (yuan/person)		**−0.00* (−0.00, −0.00)**	−0.00 (−0.00, 0.00)	**−0.00** (−0.00, −0.00)**	**−0.00** (−0.00, −0.00)**	0.00 (−0.00, 0.00)	0.00 (−0.00, 0.00)	**−0.00* (−0.00, −0.00)**
Length of caring (years)		−0.01 (−0.17, 0.14)	0.03 (−0.03, 0.09)	0.01 (−0.05, 0.06)	0.00 (−0.07, 0.06)	−0.07 (−0.15, 0.01)	−0.06 (−0.13, 0.01)	0.00 (−0.05, 0.04)
Relationship to the PLSs	Parents	Ref	Ref	Ref	Ref	Ref	Ref	Ref
	Spouse	**−9.02*** (−13.94, −4.09)**	−1.81 (−3.68, 0.07)	**−2.41** (−4.20, −0.63)**	−1.15 (−3.19, 0.88)	−0.96 (−3.53, 1.61)	−0.20 (−2.31, 1.91)	−1.14 (−2.45, 0.17)
	Children	**−12.04** (−20.90, −3.19)**	−0.65 (−4.02, 2.72)	−3.00 (−6.20, 0.20)	−3.47 (−7.12, 0.18)	2.27 (−2.36, 6.89)	0.80 (−3.00, 4.60)	−2.00 (−4.35, 0.36)
	Siblings	**−6.57* (−13.08, −0.05)**	−1.59 (−4.07, 0.89)	−1.20 (−3.55, 1.16)	−0.82 (−3.50, 1.87)	0.15 (−3.30, 3.60)	0.72 (−2.07, 3.52)	−0.21 (−1.94, 1.52)
	Others	**−18.42** (−30.04, −6.81)**	−0.45 (−4.87, 3.97)	−2.03 (−6.24, 2.17)	1.82 (−2.97, 6.61)	−0.85 (−6.91, 5.22)	**−5.10* (−10.09, −0.12)**	−0.94 (−4.03, 2.15)
Co–caregiver	No	Ref	Ref	Ref	Ref	Ref	Ref	Ref
	Yes	−3.45 (−7.16, 0.26)	**−2.09** (−3.50, −0.68)**	**−2.36** (−3.70, −1.02)**	−1.15 (−2.68, 0.38)	**2.26* (0.32, 4.20)**	−0.30 (−1.29, 1.89)	**−1.04* (−2.03, −0.06)**
Additional dependents	No	Ref	Ref	Ref	Ref	Ref	Ref	Ref
	Yes	**6.66*** (3.07, 10.25)**	**1.53* (0.16, 2.89)**	**1.70* (0.40, 3.00)**	**1.98** (0.50, 3.46)**	−1.32 (−3.20, 0.56)	−0.97 (−2.51, 0.57)	0.23 (−0.73, 1.18)

(Continued)

TABLE 3 | Continued

Variables		Caregiver burden (ZBI)	Stress (PSS-10)	Anxiety (GAD-7)	Depression (PHQ-9)	Caregiving rewarding feelings (CRF)	Active coping style (SCSQ subscale)	Passive coping style (SCSQ subscale)
Physical illness	No	Ref	Ref	Ref	Ref	Ref	Ref	Ref
	Yes	**4.33* (0.43, 8.22)**	1.15 (−0.33, 2.63)	0.83 (−0.58, 2.24)	0.67 (−0.93, 2.28)	−0.96 (−3.00, 1.08)	0.52 (−1.15, 2.19)	0.37 (−0.66, 1.41)
R^2		16.41%	13.84%	12.28%	10.04%	15.81%	14.07%	4.65%
VIF	Mean (range)	1.30 (1.05–2.12)	1.30 (1.05–2.12)	1.30 (1.05–2.12)	1.30 (1.05–2.12)	1.31 (1.06–2.12)	1.30 (1.05–2.12)	1.30 (1.05–2.12)

KSCS, Knowledge and Skills of Caregiving Scale; ZBI, Zarit Burden Interview; PSS-10, 10-item Perceived Stress Scale; GAD-7, seven-item Generalized Anxiety Disorder Scale; PHQ-9, nine-item Patient Health Questionnaire; CRF, Caregiving Rewarding Feelings; SCSQ, Simplified Coping Style Questionnaire.

VIF, variance inflation factor. Collinearity existed if VIF >5, and there was little possibility of multicollinearity as all VIF <5.

Values in bold font are statistically significant ($P < 0.05$).

$^*P < 0.05$, $^{**}P < 0.01$, $^{***}P < 0.001$.

inform future psychoeducational interventions for improving caregiver outcomes.

Our study informs the relationship between caregiving knowledge and skills and caregiver burden. Some previous reports indicated that as the level of knowledge about schizophrenia increased, the perceived burden decreased (67, 68), whereas other studies showed the opposite (35). Consistent with previous research reported by Lim et al. (69) and Jagannathan et al. (15), our study showed that primary caregivers' knowledge about schizophrenia did not have any relationship to caregiver burden. Previous research suggested that avoidant coping may lead to increased caregiver burden (70), and the lack of association of caregiving knowledge and skills with caregiver burden in the current study may be due to the null association of caregiving knowledge and skills with passive coping. It is likely that coping style may mediate the relationship between caregiving knowledge and skills and caregiver burden, which warrants further study (71).

The current study also suggests that increased knowledge and skills of caregiving may relieve psychological distress, such as stress, anxiety, and depression. The stress process model argues that stress source, such as family conflicts and financial issues, is an important domain contributing to caregiver burden (36). Wan et al. (1) reported that lack of understanding of psychotic symptoms can lead to conflicts between caregivers and PLSs, which subsequently resulted in a high level of psychological distress among caregivers. Furthermore, caregivers with little knowledge may be more likely to hold false beliefs about schizophrenia, such as thinking that it is the result of bewitchment or drug abuse (72). When bewitchment is viewed as the cause of schizophrenia, caregivers usually hold pessimistic views (73) and lose hope for the recovery of PLSs, leading to psychological distress (35). Thus, increased knowledge and skills of caregiving may help caregivers change their attribution about the cause of schizophrenia so as to better understand the challenges faced by PLSs and take more active steps when crises occur (74). Improved caregiving knowledge and skills may also reduce caregivers' stigma associated with schizophrenia, which is known to be associated with depression and suicidal thoughts (75–77).

Under the framework of the stress process model, individual evaluation on self-worth is considered as a significant moderator of stress and is susceptible to subjective psychological factors. In this study, we demonstrated that improved caregiving knowledge and skills were associated with more caregiving rewarding feelings among the caregivers, an observation that may be explained, in part, by the Chinese culture. Under the influence of Confucianism, the dominant concept of collectivism is deeply rooted in the Chinese culture (13). Family members usually consider care for PLSs as their responsibility, and they may make every effort to facilitate the patient's recovery. A higher level of caregiving knowledge and skills allows them to assume more responsibility and provide better support for their loved one with schizophrenia, which may bring in a higher sense of self-achievement (38). A recent study with primary family caregivers of PLSs showed that improved knowledge and understanding of schizophrenia were effective in increasing their social interactions and promoting greater optimism for the future (74). Also, more knowledge about schizophrenia helps improve the caregiver's ability to monitor symptoms and initiate better collaboration with mental healthcare professionals, thus leading to improved self-efficacy (39).

In the stress process model, the interaction between stressors and psychological outcomes can be mediated by coping strategy (36). Similarly, a heuristic multivariate model proposed by Vitaliano et al. considers that burden results from three domains: stressor, vulnerability, and resource (78). In this model, passive coping is a risk factor for vulnerability. The current results showed that family caregivers with a higher level of caregiving knowledge and skills were more likely to adopt positive coping behaviors, which was related to better mental health (79). This finding also partially explained the phenomenon that a higher level of caregiving knowledge and skills predicted improved psychological well-being. For caregivers, increased knowledge and skills of caregiving may enhance individual confidence and competence in challenging difficulties (80), which is revealed in adopting active coping strategies. As noted above, caregiving knowledge and skills also correlated with hopefulness, which was found to be a predictor of problem-oriented coping (81). Besides, the stress–appraisal–coping paradigm is another theoretical framework for understanding the relationship between caregiving knowledge and stress (82). A better understanding of schizophrenia and caregiving may enable caregivers to reappraise the demands of caregiving, develop more effective strategies to cope with the problems related to caregiving, and better handle maladaptive behaviors of the PLSs, thus leading to reduced stress (74, 83).

Limitations

The present study has several limitations. First, there was a potential selection bias as the participants in the current study were from the "686 Program," who may have different caregiving experiences due to the availability of health services compared with those outside the program. A related limitation is that participants who were not willing to participate in the survey were also excluded. Second, as the caregivers in this study were PLSs' family members (informal caregivers), our findings cannot be extrapolated to all caregivers of PLSs. Third, the cross-sectional design of this study did not allow for investigating causal relationships among the variables, which should be further examined using longitudinal data. Fourth, a number of family caregivers in our sample had been caring for their loved one with schizophrenia for a long time, which may be different for caregivers of PLSs after a first episode. Fifth, given that the ZBI is a non-specific scale for the evaluation of caregiver burden, this instrument may not fully reflect the caregiver burden in schizophrenia.

CONCLUSIONS

The current study demonstrated that caregiving knowledge and skills were positively related to less stress, anxiety, and depression symptoms. Also, family caregivers with a higher level of caregiving knowledge and skills experienced more caregiving rewarding feelings and were more willing to adopt positive coping strategies in the face of difficulties and challenges. These findings inform future psychoeducational interventions to enhance family caregivers' knowledge and skills of caregiving to improve their psychological well-being and promote active coping.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by The Institutional review board of Xiangya School of Public Health, Central South University. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

All authors have made substantial contributions to the study conception and design, data collection and analysis, as well as to the development and editing of the manuscript. ZZ, RL, and YY contributed to the conception and design of the study. ZZ, YW, PF, TL, and YY contributed to the research conduction and data collection. ZZ and YW contributed to data analyses. PF, TL, JT, RL, and YY contributed to data interpretation. ZZ drafted the article while YW, PF, TL, JT, RL, and YY critically appraised it and revised it. All authors approved the final version of manuscript for submission and publication and agreed to be accountable for all aspects of the work.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2021.631420/full#supplementary-material

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SYSTEMATIC REVIEW
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Recovery From Severe Mental Health Problems: A Systematic Review of Service User and Informal Caregiver Perspectives

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Introduction: The recovery approach aims to have users' perspectives at the heart of service development and research; it is a holistic perspective that considers social needs, personal growth and inclusion. In the last decade recovery-oriented research and practice has increased greatly, however, a comprehensive model of recovery considering exclusively the perspectives of people with lived experience has not been devised.

Aims: This review aimed to develop a framework and contextualize service users' and informal caregivers' understanding of recovery from severe mental health problems.

Methods: We systematically searched 6 databases including key terms related to knowledge, experience and narratives AND mental health AND personal recovery. The search was supplemented with reference sourcing through gray literature, reference tracking and expert consultation. Data analysis consisted of a qualitative meta-synthesis using constant comparative methods.

Results: Sixty-two studies were analyzed. A pattern emerged regarding the recovery paradigms that the studies used to frame their findings. The resulting recovery framework included the domains Social recovery; Prosperity (Legal, political, and economic recovery); Individual Recovery; and Clinical Recovery Experience (SPICE). Service users' definitions of recovery tended to prioritize social aspects, particularly being accepted and connecting with others, while caregivers focused instead on clinical definitions of recovery such as symptom remission. Both groups emphasized individual aspects such as becoming self-sufficient and achieving personal goals, which was strongly linked with having economic means for independence.

Conclusions: The recovery model provided by this review offers a template for further research in the field and a guide for policy and practice. Predominant definitions of recovery currently reflect understandings of mental health which focus on an individual perspective, while this review found an important emphasis on socio-political aspects.

At the same time, only a small number of studies took place in low-income countries, focused on minoritized populations, or included caregivers' perspectives. These are important gaps in the literature that require further attention.

Systematic Review Registration: The review protocol was registered on PROSPERO (CRD42017076450); https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=76450.

Keywords: recovery, well-being (definitions of), severe mental disorder, service user, carer, systematic review, disability rights

KEY MESSAGES

- This systematic review generated a comprehensive model of recovery from severe mental health problems from the perspective of service users and informal carers, that can guide further research, policy, and practice.
- Four dimensions of recovery emerged: Social Recovery; Prosperity; Individual Recovery; and Clinical Recovery Experience (SPICE).
- These dimensions align with existing paradigms of recovery identified in the references of the literature: Social and political recovery models; the REFOCUS CHIME model of recovery; the United States user/survivor movement; and the clinical recovery model.
- Users' knowledge, experience, and narratives of recovery tended to prioritize social aspects, whereas caregivers focused on clinical understandings of recovery.
- Recovery is a multifaceted concept and requires a comprehensive/ecological approach. Each dimension of recovery identified in this review would benefit from specific therapeutic techniques or referral to specific professionals, including integrating civil rights or social work services into mental well-being response efforts.

INTRODUCTION

The ways in which people conceptualize mental health problems vary across cultures, and therefore there are also variations in the meaning of recovery (1, 2). Much of mental health practice, research, and policy relies on what is known as a bio-medical understanding which speaks of mental distress in terms of diagnosis, and frames recovery in terms of clinical outcomes (3, 4). From that perspective, recovery is focused on reduction of symptoms and functional impairment. The concept of *clinical* recovery derives from research led by mental health professionals: it involves diagnosis, and measures of symptoms and psychosocial functioning designed and rated by professionals (5, 6). This type of recovery underpins a large number of data collection instruments that have been used in epidemiological research.

However, critics of the clinical recovery model have highlighted limitations regarding the lack of sensitivity to variability across individuals and contexts, and not including outcomes that are meaningful to service users (7). Since the 1990's, the focus in the field of recovery has shifted to an approach derived from literature led by mental health service users/survivors. This has been referred to as *personal* recovery, it stems from and focuses on attitudes toward life, personal growth and abilities, contribution to the community, and life satisfaction (8, 9). This approach aims to have users' perspective at the heart of service development and research, and it is considered distinct from "clinical recovery" that focuses on achieving clinically-defined goals (10–13).

The personal recovery approach is an ideology that encourages a broader understanding of mental ill health experiences and how people who are feeling mentally unwell can be helped. Placing service users at the center of decision-making in mental health has initiated a major shift in traditional philosophical views of mental health, resulting in reduced discrimination and reduced association of mental health problems with deficit and chronicity (14). This definition of recovery is becoming a key concept in mental health research, policy, and service development world-wide, thus progressing toward the recognition of human and civil rights of those affected by mental health problems and their carers (15).

There has, however, been criticism about personal recovery being defined in individualistic terms (16) that neglect collectivist values that are more present in some cultural groups (17–20). A perspective that has been lacking in conceptualizations of recovery is that of informal caregivers, whose views are not typically taken into account in recovery definitions, and thus their key role in the users' recovery journey is not recognized. Acknowledging informal carers' perspectives of recovery could facilitate a deeper understanding of less common paradigms which emphasize the systemic nature of recovery and take into consideration socio-economic needs and inclusion (21, 22). Less widely cited recovery paradigms propose social and political factors to be taken into account, and add pursuing civil rights to the aims of recovery (23, 24).

In the last decade recovery-oriented research and practice has increased greatly. Recovery is now a focus world-wide and the intention to develop recovery-oriented services is typically present in official mental health service strategies (25). However, a synthesis of experts by experience's definitions of recovery has not been devised and, therefore a comprehensive model that reflects their views is not in place. The purpose of this research is to develop a comprehensive model that encompasses the full range of dimensions of recovery which are relevant to experts by experience (i.e., individual and systemic recovery), while at the same time providing context for this construct. This will be achieved by: (1) reviewing the evidence for mental health service users' and their informal caregivers' understandings of recovery from mental health problems, and (2) compiling key recovery paradigms referenced in this literature and specifying their characteristics and origins.

METHODS

This review followed the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement (26). A protocol was developed a priori and registered on PROSPERO (CRD42017076450).

Search Strategy and Study Selection

Six electronic databases (Embase, PsycINFO, Medline, ScIELO, LILACS, and CINAHL) were searched in October 2020. The search strategy included key terms related to knowledge, experience and narratives AND mental health AND personal recovery. A complete search strategy is provided in **Supplementary File 1**. Further articles were sourced by searching for publications by authors of relevant gray literature identified in the database searches. Due to most publications identified being based in Europe and North America, a convenience sample of 10 recovery experts working in seven countries across Africa, Asia, and Latin America were contacted for suggestions of further literature relevant for inclusion. Additionally, the search was supplemented by reference searching through included literature, and the five authors with most publications were contacted to enquire about potential missed studies or work in press.

Initial screening was conducted based on the titles and abstracts of the search results using the web application Rayyan (27). Full texts were sourced for articles deemed relevant for inclusion and these were then screened against the full review eligibility criteria.

To establish consistency in the study selection, 300 randomly selected records at the title and abstract screening stage, and 50 records at the full text screening stage were independently reviewed by the author and a second screener, and discrepancies were resolved via discussion.

Eligibility Criteria

Studies were included in this review if (1) their focus was recovery from severe mental health problems, (2) as understood by service users and informal caregivers, and (3) enquired through methodologies where participants' perspectives were explored in an open-ended manner; studies with fixed survey responses were excluded. There were no restrictions on publication date or language.

Recovery was understood as changes toward feeling well, reaching meaningful outcomes or experiencing a positive sense of self. The term informal caregiver refers to people who provide unpaid care or support for people with mental health problems.

Articles were excluded if mental health problems were not the participants' primary condition, or if the focus of the study was limited to a specific aspect of recovery. Studies where the primary condition was substance misuse or exposure to traumatic events were excluded due to these fields having their own extensive bodies of recovery literature which describes specific recovery paths (28).

A full list of the inclusion/exclusion criteria is provided in **Supplementary File 2**.

Data Extraction and Risk of Bias Assessment

Data collected from the studies included the recovery paradigms used to frame their findings in the introduction/background section (either in terms of a paradigm explicitly stated by study authors, or a paradigm as interpreted by the review team), and the recovery themes that studies reported in the results section/discussion. When themes were not explicitly presented, results were categorized into themes. Special attention was paid to extract themes of recovery described as an outcome, rather than when presented as helping or hindering recovery. In addition, data were collected on core study details (year, setting, population and methodological characteristics, and authors' interpretations and further discussions on the data). Missing details were requested from study authors.

Given the plurality of methodologies used in the identified studies, seven criteria for quality appraisal were adopted from different published tools (29–31)[1] with the aim of appraising transparency, description of key terms, and coherence. The full risk of bias assessment checklist is provided in **Supplementary File 3**.

Qualitative Meta-Synthesis

An interpretative synthesis using constant comparison was conducted to develop a definition of core dimensions of recovery and an understanding of how they may be related (32, 33). This method involved using reciprocal translational analysis to group the themes identified in the literature into higher order themes that best reflected their content, while keeping the theory grounded in the data and context of each study to gain a broader picture of the construct of recovery. Additionally, negative cases were kept in a log to have them present during data synthesis.

At a final stage, study characteristics were condensed into ecological sentences (i.e., "in this year, within this paradigm of recovery, in this setting, recovery meant…") to facilitate mapping the concept of recovery (34).

RESULTS

Study Selection

A flow diagram of the screening and selection process, according to PRISMA guidelines, is presented in **Figure 1**. A full list of citations and reasons for exclusion is provided in **Supplementary File 4**. The remaining 62 studies were included in this review.

Study Characteristics

From the 62 papers included in this systematic review, one was published in 1967, while the rest were conducted between 1999 and 2020. Study settings were primarily English-speaking ($n = 51$, 82%), high-income countries ($n = 58$, 94%). However, six

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[1] Critical Appraisal Skills Programme (CASP) Checklist.

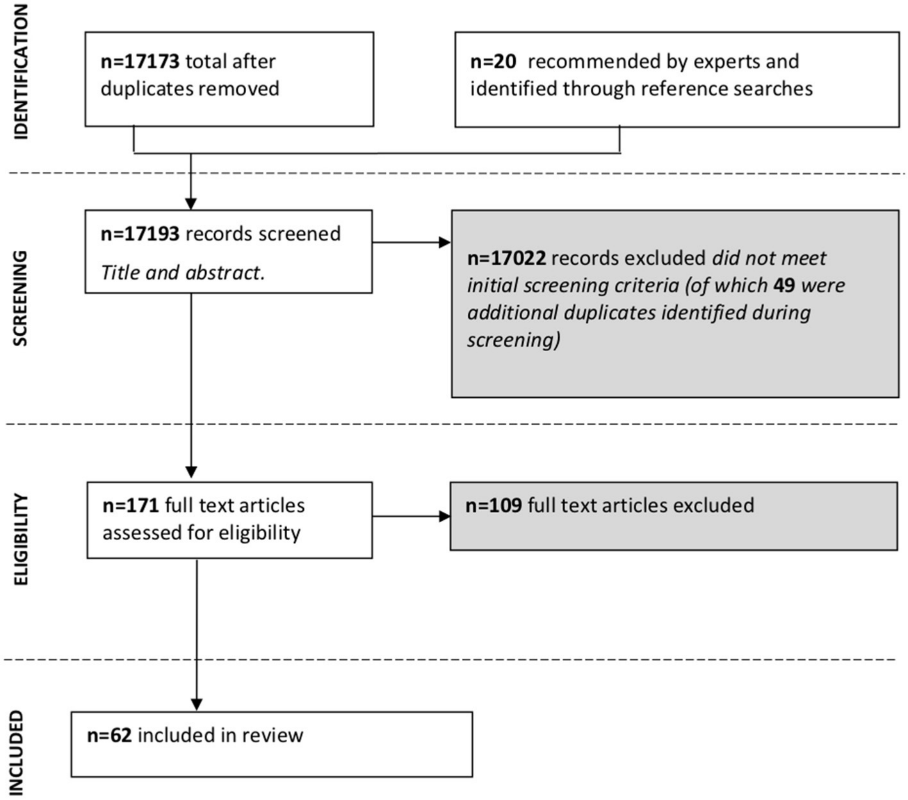

FIGURE 1 | Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flow diagram of the screening and selection process conducted in this systematic review.

(10%) of these papers focused on a low-income sample. In all included studies, recruitment was done through convenience or purposeful sampling in all studies, generally participants were reached through clinical contacts or announcements posted in recovery or service user groups.

Data were collected using in-depth interviews in 47 (76%) of the studies. Other methods included focus groups, photo-voice, ethnography field notes, and narrative interviews. Thematic analysis ($n = 27$, 44%) and grounded theory ($n = 11$, 18%) were the most commonly used analysis methods. Two studies (3%) applied a quantitative methodology, one followed a Delphi process for data collection and analysis (35), and one study used a snowball technique for data collection and Chi squared analysis (36).

Sample sizes ranged from 1 to 177 participants in qualitative studies, and 180–381 in the quantitative studies. Sixty (97%) studies included a user sample, and nine (15%) included a caregiver sample. Studies typically included both male and female participants between 18 and 65 years of age. Twenty-five (40%) studies specified participants' ethnicities; out of these, 19 were predominantly of white-European background. The remaining six studies included two in the USA and Canada which had specific interest in users of black-African descent (37, 38); one that contrasted perspectives of Euro-Canadian and Caribbean-Canadian participants (39); one focused on the perspectives of women in Swaziland (40); one about Indian service users and caregivers (36); and one focused on individuals from a Chinese community in Hong Kong (41).

Participant information concentrated around stage of recovery and diagnosis. Authors described the stage of recovery in various ways such as length of service use or feeling well enough to participate in the study. Studies included heterogeneous transdiagnostic samples, with the exception of 17 (27%) studies that focused on psychosis/schizophrenia, 3 (5%) on depression, 3 (5%) on personality disorder, 3 (5%) on bipolar disorder, and 1 (2%) focusing on voice hearing following the single complaint approach (42). Limitations were stated in relation to comorbidity with other diagnoses and relevance and usefulness of diagnostic criteria.

User employment and education were reported in 18 (29%) and 13 (21%) studies, respectively. Based on these data, users were most commonly unemployed and education levels varied from no schooling to "25 years of education."

A pattern emerged regarding the recovery paradigms that the studies referenced in their introduction and used to frame their findings. Five distinct categories/models were identified: USA consumer/survivor recovery movement (including Substance Abuse and Mental Health Services Administration—SAMHSA-model) ($n = 19$, 30%); REFOCUS-CHIME model of recovery ($n = 12$, 19%); Social recovery ($n = 8$, 13%); Political recovery ($n = 3$, 5%), and Bio-medical recovery ($n = 3$, 5%). Further exploration of the paradigms referenced highlighted that the perspectives of experts by experience were present particularly in the development of models of recovery in the USA, and to a lesser extent in the REFOCUS-CHIME model and the Political recovery paradigm. The remaining paradigms emerged mainly from reflections of mental health professionals and social/political theorists. All the identified recovery paradigms concurred in acknowledging the potential to feel better after experiencing mental health problems, however, they differed in their position regarding four aspects of recovery: (1) The extent to which they focused on internal conditions such as individual's attitudes, vs. external conditions such as policies and social circumstances; (2) the importance placed on diagnosis; (3) the literature by which they were influenced, and thus (4) the recovery goals they proposed to focus on.

A brief description of each recovery paradigm is provided in **Table 1**, and the overall main characteristics of the included studies are listed in **Supplementary File 5**.

Risk of Bias

All studies met 50% or more of the quality criteria assessed, and 31 studies (50%) fulfilled all 7 criteria. Additionally, a substantial number of studies included user participation or mindful interviewer selection ($n = 29$, 47%) to enhance rigor.

RECOVERY THEMES

This list of themes is the result of the synthesis of the empirical data extracted from the results section of the studies included in this review. **Table 2** illustrates the four core parent themes present in these data: Social Recovery; Prosperity; Individual Recovery; and Clinical Recovery Experience (SPICE). All themes were present to a greater or lesser extent in users' definitions of recovery; the cases where themes were also part of caregivers' understanding of recovery are highlighted where applicable. These themes are elaborated upon below, with selected quotes from the included studies illustrating the key characteristics of the parent themes and subthemes within these. **Figure 2** provides a visual representation of how the findings in this review are related. Theme one (Propsperity) was aligned with the social and political recovery paradigms; themes two and three (Social and Individual Recovery) overlapped with the definition of recovery of the REFOCUS-CHIME, SAMHSA, and USA consumer/survivor movement, and theme four with the bio-medical recovery paradigm. At the same time, social and political aspects of recovery were more common among user samples, while clinical recovery goals were more prevalent among carer samples.

Theme 1: Prosperity

Framing recovery as a social construct was highly present in the literature. Examples of this can be found in Basso et al. (43) "*recovery has to be understood also as a social process, where people face, along with the disease, other tests such as the need for tangible resources, jobs, availability of housing, financial independence, and efficient services*" or (38), who studied recovery from the perspective of racialized women in Canada and remarked on the lack of discussion around symptoms and treatment in participant's recovery narratives: "*their challenges were very much framed as social rather than psychiatric.*"

This recovery theme was especially common in literature linked to the user/survivor movement or advocating for collective action against human rights violations in mental health treatment.

Subthemes that fell under this theme were: "Legal and political recovery" and "Economic recovery."

Legal and Political Recovery

Empowerment was one of the central aspects underlying this theme; recovery goals were related to rebelling against socially imposed rules or practices which users considered to stand in the way of their well-being and advocating for fairer legislation. This idea was especially prominent in the literature analyzing women's understanding of recovery, where these thoughts were discussed under the terms "*breaking away from limited woman roles*" (38) and "*doing and being beyond gendered responsibilities*" (44). Fullagar and O'Brien (44) concluded "*Practitioners and advocates in women's health movements have historically recognised that personal recovery is political.*" At the same time Armour et al. (37), pointed out that black and minority ethnic (BME) groups experienced oppression both because of their mental health problems and because of their race, which would involve two different approaches when fostering empowerment.

Economic Recovery

A key recovery goal from both a user and caregiver perspective was reaching economic stability. Recovery was understood as having sufficient resources available to have an acceptable quality of life and live independently from family. Participants in Borg and Davidson's (45) study in Norway, included shopping and paying bills as part of their notion of achieving "normality" (see *normalcy* subtheme). Similarly, service users and carers in Italy considered recovery involved actions to reduce external barriers that impeded independent living, such as lack of jobs in the open market and lack of accessible living solutions which prolonged cohabitation with the family (43). The need for financial support and/or access to employment to mitigate adverse material circumstances was highlighted particularly in studies with participants from ethnic minorities or hard to engage populations (37, 40, 46).

Theme 2: Social Recovery

Two interrelated types of social recovery were identified. One was an externally derived social recovery which required approval and acceptance from the group. In this sense, recovery meant being trusted, being assigned responsibilities and being treated as an equal. Cárcamo Guzmán et al. (47) wrote about the meaning of recovery to service users in Chile, "*it is understood as the legitimacy of the user as a person, this implies the respect for their experiences, points of view and needs.*" The other type of social recovery was derived from personal initiative and consisted of: socializing and establishing meaningful relationships, being a productive member of the community, and fulfilling family roles. Participants in Hancock et al. (48) study spoke about learning to navigate complex relationships, avoiding unhelpful

TABLE 1 | Description of recovery paradigms identified in the literature.

Paradigm	Informed by	Emphasis	Key authors
1. USA consumer/survivor recovery movement	First- person accounts of members of the psychiatric survivor community in the late 80s	Self-management, reclaiming identity beyond diagnosis, self-acceptance and maintaining hope	William Anthony Patrick Corrigan Marianne Farkas Courtenay Harding
Substance Abuse and Mental Health Services Administration (SAMHSA)	Expert by experience advisory committees and mental health professionals	Self-directed, based on hope to reach full potential. Major dimensions: Health (physical and emotional); having a stable home; finding purpose; and living in community	Experts recruited by U.S Department of Health and Human Services
2. REFOCUS CHIME model of recovery	Positive psychology and wellbeing literature	Connecting with others, living a meaningful life and concentrating on individual strengths for personal growth	Work conducted by the REFOCUS team in the early 2010s
3. Social recovery	The deinstitutionalization movement, community psychiatry, and social psychology literature	Social inclusion and psycho-social rehabilitation. A key goal is users participating in research and society	Larry Davidson Steve Onken Arthur Kleinman Graham Thornicroft Ron Coleman Benedetto Saraceno
4. Political recovery	Post-modern social theory	Social inequities and breaking from forms of social control. A key goal is user collectives participating in political contexts	Franco Basaglia Kim Hopper Michel Foucault John McLeod
5. Bio-medical recovery	Traditional Western understandings about mental states	Recording users' perspectives to make clinical decisions and predict health outcomes	Nancy Andreasen

Key authors listed in this table are limited to those that were most cited in the papers included in this review. This is not an exhaustive list of authors or their publications published in these areas.

TABLE 2 | Parent themes identified in the data, the subthemes that fall within these and the number of user/carer studies which included them.

Parent theme	Subthemes	*N* (U = 53/C = 9)*	Description
Prosperity	• Legal and political recovery • Economic recovery	• 7/0 • 21/2	Linked to empowerment; covering basic economic needs and co-construction of recovery
Social recovery	–	• 41/4	Returning to a basic form of social awareness; being a part of society, functioning well within groups, treated as an equal
Individual recovery	• Normalcy • Temporal understandings and identity • Recovery and knowledge • Recovery as an individual responsibility • Appearance and hygiene • Recovery as a positive frame of mind	• 21/2 • 21/0 • 49/5 • 4/1 • 19/2	Being "normal"; completing everyday activities and/or focusing on achieving personal goals; fulfilling roles and responsibilities; gaining relevant knowledge about mental health or enrolling in formal education
Clinical recovery experience	–	17 /5	Considerations about diagnosis and treatment

This column indicates the number of User/Carer articles that included each theme. Articles with a User sample total N = 60; articles with a carer sample total N = 9.

interactions and managing the impact of their mental health problems on others.

Nxumalo Ngubane et al. (40) presented being accepted and able to contribute to their family and community as an important part of recovery for Swazi women diagnosed with schizophrenia. The socially constructed nature of recovery was emphasized repeatedly, with social discrimination and experiences of stigma being perceived as the opposite of recovery in many of the studies (36, 40, 47, 49–52). The definition and achievement of recovery was thought to be co-constructed in society and developed by engaging in honest and genuine mutuality (53). In this sense, others offering help or feedback, and users being willing to accept it, were equally important recovery goals, as pointed out by Moltu et al. (54) in Norway saying "*In our analyses, we were struck by how important others were in noticing improvement and positive change, in a way that the suffering person could embody.*"

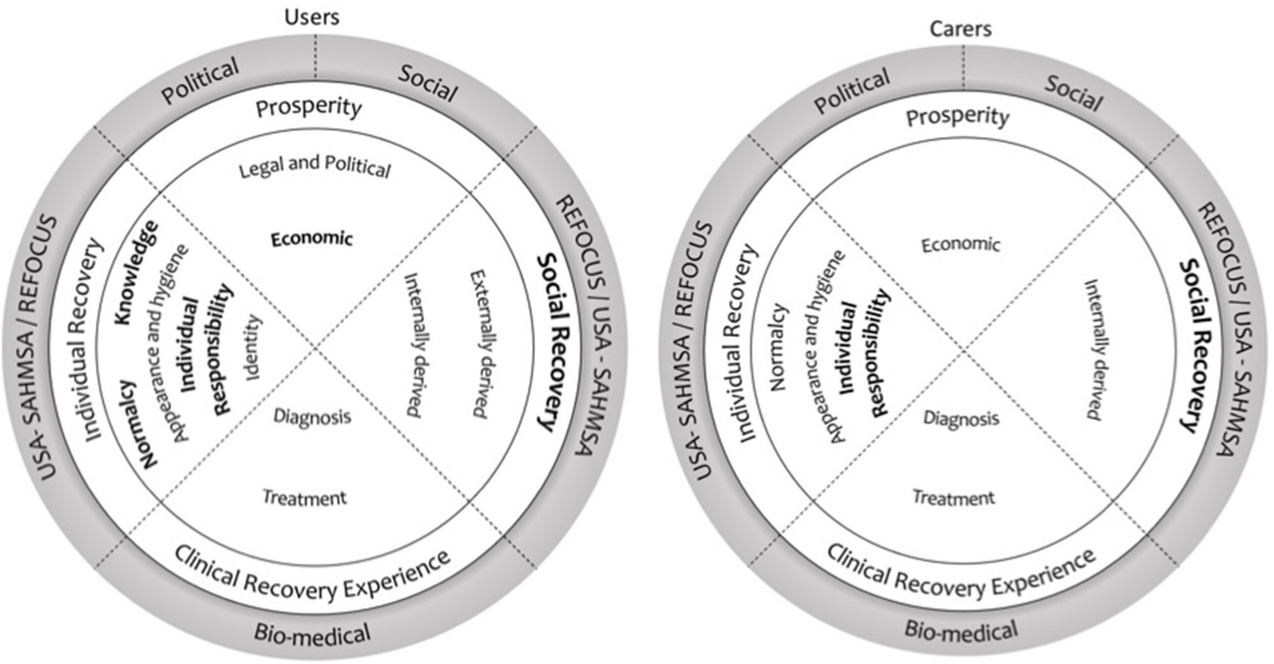

FIGURE 2 | Meta-synthesis map. Visual representation of how the recovery paradigms and themes identified in this systematic review are related and their predominance in user/carer samples. The circles on left and right represent recovery as understood by users and carers, respectively. The outer circle presents recovery paradigms, while the inner circles refer to the themes and subthemes. The most prevalent themes are highlighted in bold letters.

An important part of externally derived social recovery was being allowed to take risks, this is to be considered to have adequate judgement in everyday life and legal capacity to consent in formal contexts. As written by Pitt et al. (55) "*ultimately recovery requires active participation in life. This involves taking risks and suffering setbacks.*" Fullagar and O'Brien (44) described how an environment that allowed for free decision-making provided users with the opportunity to experience "*dignity of risk*" and realize their capabilities.

Some studies described a spiritual form of connection with a "*higher power*" or "*God*" as important for recovery (37, 40, 56). Allusion to spiritual or religious recovery concepts was present across the literature in the different populations and settings.

People with mental health problems which affect social interaction, such as people with a diagnosis of personality or bipolar disorder, were thought to face a greater challenge to achieve social recovery. This was both related to personally derived social recovery, as described by (57) "*improving relationships for this group might also be more complex than solely addressing social isolation* [discrimination], *which is commonly discussed in recovery literature*," and externally derived social recovery, Kverme et al. (53) "*The experience of becoming safer as a human among other humans constituted a core meaning of recovery.*"

Within this theme, caregivers' definitions of recovery concentrated mainly around users being attentive to others' needs and able to establish positive connections. As mentioned by (58) "*Families described changes in amount and content of interaction, noted their relative being helpful in the home, showing consideration for a parent, remembering a family member's birthday*," and by (41) "*She* [carer's daughter] *can integrate into society through such things as going to church, having a job, returning to a normal life, going out.*"

Theme 3: Individual Recovery

The third parent theme focused on individual goals, needs, and responsibilities. As expressed by (59) "*Contrary to the common belief that mental illness involves a purely degenerative condition, it appears that many people discover new potentials and new self-growth at various points throughout their recovery.*"

This theme of individual recovery encompassed six subthemes: "Normalcy"; "Temporal understandings of recovery and identity"; "Recovery and knowledge"; "Recovery as an individual responsibility," "Appearance and hygiene," and "Recovery as a positive frame of mind."

Normalcy

Related to social recovery was the idea of not feeling different from most people and achieving the goals that are considered the norm by your social group. Borg and Davidson (45) found "*being normal*" to be one of the major themes in recovery: "*What seems most crucial to "being normal" is spending time in ordinary environments with ordinary people.*" Katsakou et al. (57) identified a link between employment and feeling normal, as expressed in one of their participant's quotes: "*I still haven't* *managed to get back to work and I can't see friends, I've been cut off because I've stopped working.*"

A line of the recovery literature focused on understanding recovery through ordinary everyday activities. In this sense, the main recovery goal consisted of completing routine tasks and participating in common leisure activities. Milbourn et al. (46) noted that in order to appreciate participants' understandings of recovery, the list of everyday routines needs to be broadened to include personally meaningful activities which may be considered negative by others, such as "*recreational drugs and paying for sex.*"

McCabe et al. (60) pointed out in forensic mental health services "*everyday activities such as walking and discussing books were talked about in the language of therapies administered by services. The 'reader group' and the 'walking group' were all discussed in terms of therapeutic interventions rather than fulfilling hobbies that people adopt in everyday life*."

Temporal Understandings of Recovery and Identity

There were two contrasting views regarding the temporal focus of the recovery journey: one described recovery as the ability to focus on future goals, having hope and seeing "*the light at the end of the tunnel*" (61), while the other described it as the ability to live in the present and complete daily routines (related to the "Normalcy" subtheme). This contrasting view of recovery was also present in different identity goals, with some service users striving to develop a "new self" by learning from their experience (62, 63), and others wanting to return to the roles and occupation or everyday activities from before experiencing mental health problems (58, 64). Recovery was not a single state of being but a complex mix of the past, the here and now and, the future (60).

This distinction was discussed by (65), who found participants wishing to reflect on and integrate the disorder experience into a new identity, while others wished to leave the experience behind and focus on symptom management. Participants in both groups were described as currently not having symptoms, good quality of life and no psychological distress, for this reason the authors advocated for the latter approach to recovery to not be pathologized. Instead, they promoted a broader understanding of recovery that does not require active engagement or reflecting on the mental health problems experienced.

The idea of returning to a former identity was a prominent topic throughout the literature, however, it was particularly highlighted in the definition of recovery of older adults presented by (66) "*The single core category identified from the analysis was 'Continuing to be me.' This related to the permanent and established sense of identity which service user participants held [. . .].*"

Recovery and Knowledge

An important recovery goal was gaining new knowledge. This included knowledge about yourself (personal growth), knowledge about mental health, and knowledge gained through formal education. The latter was highlighted as particularly important in (67) study about adolescent service users.

Service users in (49) study underlined the role of understanding early lived experience as informing sense of self "*Most participants framed their understanding of their experiences within a description of their early life within their family, particularly their sense of belonging and the interpretations of their behaviour made by key family members.*" Self-discovery was also a significant part of recovery for young people in (68) study, pointing to the limited life experience before mental health problems creating an additional vulnerability.

Knowing more about mental health was approached both as part of embracing a given diagnosis [e.g., the goal "*coming to know your illness*" (69)] and discarding it [e.g., "*developing a critique of mental health services*" (55)]. These considerations about diagnosis are explored further later under the theme "Clinical recovery experience." In both cases the final aim was to develop strategies to feel better, building higher self-esteem, and self-awareness. As described by (70) "*Recovery usually occurs when people with mental disabilities discover or rediscover their strengths and the opportunities to pursue personal goals and a sense of self that allows them to grow, despite any residual symptoms and difficulties.*"

Recovery as an Individual Responsibility

Being self-sufficient and having control over one's mental health problems and their consequences were highly prevalent recovery goals. Recovery within this theme is described as an internal fight, coming to the realization that "*It needs to be me*" (48). In most of the literature, recovery was presented as a personal choice to actively cope with mental health problems. An important aspect of reaching autonomy was no longer being reliant on mental health services. As stated in (71), participants did not consider mental health services to promote self-management and this was seen as going against their recovery. Participants wished to assert their position as experts by experience and those who did not engage with services were seen as "*winners*": "*taking responsibility is at the heart of the recovery process as people are empowered to make their own choices and focus on their own outcomes.*"

This understanding of recovery is summarized by a participant in the study conducted by (72) "*It reminds me of an author who said she's never avoided challenges but put her "sails full tilt into the wind." There's a certain bravery in facing obstacles head-on. With my mental health challenges, I've learned to put my sails full tilt to the wind and move towards my goals.*"

This conceptualization of recovery as an individual responsibility was strongly linked to empowerment, which in turn was linked to having economic means for independence (43, 73, 74). A person with mental health problems reaching independence was a particularly important recovery goal for caregivers, this included financial autonomy and independent living that reduced the reliance on caregiver/family support (as mentioned in the subtheme "Economic recovery") and reaching emotional stability. An example of this are the findings from the study by (58): "*They longed for their relative to be able to take care of themselves, live independently, or have improved judgment and concentration, or to work and become functional and self-sufficient.*"

A distinctive understanding of recovery was presented by (75) who studied the views of forensic psychiatric patients (offenders with mental health problems). For the most part participants chose to rely on medication and medical guidance, rather than their own judgment and active participation: "*Their lack of control was in most cases, simply stated as an incontrovertible fact.*"

Appearance and Hygiene

Some studies described improving appearance and keeping up good hygiene as part of personal recovery; the focus of this goal was adding to a personal sense of worth, rather than complying with social rules. Davis (76) who conducted an ethnography in a women's psychiatric ward noted "*Wearing their own clothing again adds to their appearance of well-being. [...] this makes it all the more difficult for them to see themselves as*" "*sick persons.*" A participant in the study by Santos et al. (74) expressed "*[I want to] maintain... good hygiene..., fitness, exercise, nutrition....*"

Recovery as Having a Positive Frame of Mind

A representative description of this understanding of recovery can be found in Kartalova-O'Doherty and Tedstone Doherty (77) "*Personal definitions of recovery fell into two broad areas: getting rid of negative feelings, such as anxiety, depression, or panic attacks; and acquiring positive feelings and actions, such as peace of mind [...].*"

Accounts of recovery found in the literature that fall within this category include "*being positive*" (78), "*being happy and successful*" (67, 77), "*finding hope and purpose*" (48), or "*having a meaningful and satisfying life*" (69), without a deeper description about what this meant. Recovery was described as general feelings and attitudes that were considered positive or the opposite of being unwell, dissatisfied, or unsuccessful.

Another important aspect within this theme was the idea of recovery as having peace of mind (59, 61, 77). This was described as feeling at ease, enjoying leisure moments or not experiencing constant anxiety and fear.

Theme 4: Clinical Recovery Experience

This theme includes topics traditionally related to clinical understandings of recovery such as diagnosis, medication, and symptom-related concerns. Examples when this was present in the literature were references to recovery goals such as "*chemical balance*" (63), "*adherence to treatment*" (79), or "*reducing clinical symptoms*" (47, 51, 67, 80). Brijnath (73) challenged traditional personal recovery literature writing: "*Participants' emphasis on being 'cured', achieving an endpoint in their depression and discontinuing medicines runs counter to the recovery discourse that emphasises that one can be ill and still live a meaningful, contributory life.*" In the same line, Piat et al. (12) remarked that "*The prominence of the illness perspective of recovery among consumers was unexpected. Many looked for recovery outside of themselves: in a cure, or in dreams of disappearing symptoms.*"

For service users in some studies, recovery meant being discharged. This in turn had implications for recovery milestones being prioritized by participants, as described by McCabe et al. (60) "*service users identified their relationships with staff as of greater importance than those with other service-users [...] attaining discharge was a more immediate and pressing goal and staff were seen as holding the key to discharge [...] In order to be deemed to be recovering service users were keen to demonstrate an acceptance of the bio-medical model regardless of whether this actually fitted with their view of the world.*"

TABLE 3 | Themes present in caregivers' understandings of recovery.

Theme	Carer focus
• Economic recovery	• Financial autonomy and independent living
• Social recovery	• Derived from personal initiative (socializing, fulfilling family roles…)
• Normalcy	• Participating consistently in group/family activities and fulfilling common life milestones
• Recovery as an individual responsibility	• Being self-sufficient and having control over one's mental health problems and their consequences
• Appearance and hygiene	• Self-care and hygiene habits
• Recovery as a positive frame of mind	• General positive feelings and attitudes
• Clinical recovery experience	• Adherence to treatment, accepting diagnosis, and/or being discharged

With regard to diagnosis, there were two opposing views: recovery as embracing the label and recovery as dropping the label. For the first, Ridge and Ziebland (63) used the term "coming out of the closet," since accepting the given diagnosis was understood as way to achieve authentic living without trying to pass as "normal." Assimilating the diagnosis as part of one's identity also meant giving central importance to complying with treatment and medication. Brijnath (73) found that Indian participants found meaning in life through religion, while "*For Anglo participants, meaning in life was derived from the illness experience itself. Participants talked about the importance of a diagnostic label in validating how they felt, discovering their inner strength and learning to live with depression.*"

In contrast, recovery as a rejection of the given diagnosis usually implied disengaging with services. This view was especially prevalent in literature from the user/survivor or feminist movements, and it was linked to poor practices of mental health services. Examples can be found in Adame and Knudson (81) "*Another traditional construction from the survivors' narratives was* "*recovery from the mental health system*" *[...] all four participants felt that recovering from psychiatric interventions (e.g., ECT, drugs, solitary confinement) was one of, if not the biggest, challenge in their entire healing process*" and in (40), where participants believed health professionals, traditional healers and religious leaders had used labeling as a form of coercion to support their own ideas of recovery.

At the same time, some studies found both views represented in their sample, such as (49) who studied recovery in people diagnosed with personality disorder and concluded that most found it useful and "*For a minority of participants however the diagnosis of personality disorder was seen as unhelpful - representing a direct comment on them as a person, or as a representation of their previous behaviour, not a 'mental illness' per se.*"

Clinical understandings of recovery were particularly common among carers (it was the predominant theme in five out of the nine papers that presented caregivers views) and it was normally presented as part of the guidance they received from their psychiatrist. To this respect (82) wrote "*Even though carers are the closest people that many consumers have in their life, carers had major divergence in their views on mental health recovery. Contrasting to consumers and nurses, none of the carers described regaining one's sense of self as an important aspect to mental health recovery. The carers' views on mental health recovery closely related to the traditional views of remission of symptom.*" Also, the same study reported that of importance was that this understanding of recovery led caregivers to think recovery was impossible as they understood these goals (e.g., symptom remission, retuning to pre-illness status) as unattainable: "'*I don't understand what you mean by recovery from mental illness, there isn't one... we went to the psychiatrist the other day and she said [that] the illness will never go*'."

DISCUSSION

This review aimed to define the various ways in which service users and carers conceptualized recovery and to provide context for how this construct is represented in the existing literature. Data from sixty-two studies originating mainly from high-income countries were synthesized and analyzed resulting in the SPICE model of recovery. The most prominent themes in users' definitions of recovery were Social Recovery and Individual Recovery. Within these themes, users' understanding of recovery revolved especially around connecting with others, and recovery as an individual responsibility to reach control over mental health problems. In the case of informal carers, the most common themes when defining user recovery were Recovery as an Individual Responsibility, particularly reaching autonomy/being self-sufficient, and Clinical Recovery Experience, mainly symptom remission (see **Table 3**). Marshall et al. (83) also found informal carers had pessimistic views about the potential for recovery and emphasized clinical aspects of recovery. As a possible solution they pointed to recovery training which has been found to be effective among staff (84) and could perhaps be mirrored in carer populations.

Service users' perspectives overall resonated with the more established models and definitions of recovery mentioned in the introduction (8, 9) and identified as paradigms 1 and 2 in **Table 1**. These definitions of recovery are present in the themes "Individual recovery" and "Social recovery" (derived from personal initiative) proposed in this review, which focuses on personal growth, autonomy, and individual initiatives. This is consistent with a review of user autobiographical accounts provided by (85), who concluded that recovery was "*a growing sense of agency and autonomy, as well as greater participation in normative activities, such as employment, education, and community life*," or the study conducted by (86) who wrote "*For our participants, successful living is fundamentally connected to*" "*not being dependent on mental health care.*"

However, along with providing further evidence in support of previously defined models and definitions of recovery, this review identified additional dimensions, namely social (externally derived), political and economic aspects of recovery, and factors related to social reciprocity and acceptance. These understandings of recovery were consistent with less prominent recovery paradigms (3 and 4 in **Table 2**). This is consistent with the findings of the systematic review conducted by (87) to synthesize typologies of user recovery narratives. The authors found that recovery narratives incorporated social, political and human rights aspects to a greater extent than illness narratives. Petros et al. (88) suggested an adaptation of the REFOCUS-CHIME model of recovery (paradigm 2 in **Table 2**) to underline the bi-directional nature of recovery. To this respect they wrote "*perceived reciprocity within [...] relationships is correlated with higher levels of satisfaction in support and higher levels of personal confidence, self-esteem, and perceived recovery.*" The integral role in personal recovery of family and community has been especially mentioned in literature referring to cultures that focus more on group goals than on self-responsibility (20, 89). An example of this is (90) including the domains "family involvement" and "social ties and integration" as part of their scale to measure personal recovery in Chinese culture.

Furthermore, an emphasis on availability of basic needs as exemplified in the theme "Economic recovery" was also found to be a key concern for users in the review conducted by (91) and the Australian National Survey of Psychotic Illness (92). The importance of factors related to social justice which fall under the theme "Prosperity" is widely supported by research on social determinants of health (93–95).

Price-Robertson et al. (16) and Bayetti et al. (96). There has been substantive criticism about the field of recovery being excessively focused on the individual has raised awareness on the risk of glossing over important social challenges and the stressful social conditions that can be generated by high expectations of self-control in adverse contexts (18, 97–99). Yates et al. (100) addressed this gap in recovery literature by studying in detail the social and environmental conditions in which recovery takes place, concluding recovery should be understood as an interaction of ecological processes such as the co-occurrence of personal growth and self-determination in contexts of social structures that restrict personal agency.

Thus, addressing social, political, and economic disparities and opportunities for participation in the community should also be recognized as a key dimension of recovery. This discussion is especially relevant for the development of the recovery approach in low- and middle-income countries (LMICs) that are affected to a greater extent by social inequality, violence, or other social stressors (101, 102). Despite identifying a limited amount of research from LMICs that focused on recovery, the key role of economic sufficiency, housing, and respect of basic human rights in mental health are highly present in literature relating to both LMICs and BME groups (103–105). It has been the focus of recent calls for a paradigm change in the field of global mental health (106–108), particularly in the context of the COVID-19 pandemic (109).

Another aspect of dominant definitions of recovery that is contested in our findings of the clear distinction between *clinical* and *personal* recovery. Despite an attempt in the recovery-oriented discourse to diverge from "clinical" language and make a clear-cut distinction between "clinical" and "personal" recovery, the theme "Clinical Recovery Experience" highlighted how topics traditionally considered to fall under *clinical* rather than *personal* recovery are actually important aspects of users and carers' everyday lives and notion of recovery. Clinical concepts present in users and carers' understandings of recovery, however, have a distinctive social meaning behind them. There is also a need to study the meaning of clinical language when used by lay stakeholders in order to further understand the role that it plays in their individual and social recovery. This disparity between a social and a clinical understanding of clinical language has great importance for the development of meaningful mental health evaluation tools and clinician-user communication. This would affect decisions such as that made by (110) of removing items related to symptom management and medication from a personal recovery measure.

Regarding diagnosis, the criticism about the lack of validity and practical use of diagnostic categories expressed in the background literature of the included studies contrasted greatly with the notable adherence to the diagnosis identity on the part of users and caregivers. Some authors have highlighted the social role of diagnostic labels, such as Cruwys and Gunaseelan (111) who found that people diagnosed with depression tended to identify more with their diagnosis when they faced stigma, using the identification with a group as a buffer against discrimination. Tekin (112) pointed to risks of diagnosis being a "double-edged sword" that on one hand may facilitate self-understanding and communication, while on the other hand may lead users to make sense of situations focusing only on unrealistic dichotomous outcomes. At the same time, some researchers have suggested there may be an excessive representativeness of user narratives which align with medical views due to user samples consisting for the most part of responsive persons who are in a disempowered position (78, 113, 114).

Implications

Service user and carer accounts reviewed in this study show experiences of severe mental health problems are multifaceted and require an ecological/holistic approach. In light of these results, efforts in mental health policy and service development should address users' social and legal disadvantages and economic distress. Articulating a civil rights or social work perspective on recovery from mental health problems would help to meet the recovery goals presented as most important to service users.

With respect to practice, worrying levels of stigma and discrimination in psychiatric practice were identified in users' testimonies and reflected in caregivers' notion of recovery. These are direct barriers to recovery and therefore there is a pressing need to consider the negative effects that narrow medicalized attitudes have on people's lives. At the same time, the legal or social barriers that prevent psychiatrists from promoting user freedom and participation should be addressed (16, 115, 116). Clinical and personal recovery are intrinsically related and can complement each other; optimal provision of services can be achieved by combining the strength of professional's knowledge and epidemiological research, with stakeholder's experience and feedback about their needs (117).

The particular understandings of recovery identified in this review would benefit from specific therapeutic techniques. Service users who underlined the importance of bi-directional communication for recovery may adhere better to treatments of a dialogical nature (54), while users less interested in active engagement and meaning-making, such as those searching to achieve normalcy through completing everyday routines, could find more use in mindfulness-oriented techniques (118, 119). In the same way, service users expressing concerns relating to discrimination, legal, and economic circumstances should be referred to appropriate help which focuses on facilitating access to adequate housing, employment, education, and money management, to ultimately be empowered to address their needs (120). Examples of this are initiatives such as the Bapu Trust for Research on Mind and Discourse, in India (121), and advice services set by government in the United Kingdom such as the Money Advice Service. Altogether, identifying users' personal recovery goals and mapping them onto the framework proposed in this review would in turn facilitate the development of person-centered individualized care.

There is a need for research about recovery across different cultures. Predominant definitions of recovery currently reflect Western understandings of mental health which focus on an individual perspective, without adequately addressing important socio-political aspects. Recovery-oriented research and practice should take an additional step beyond focusing on what occurs in clinical settings and empower communities for the promotion of human rights, thus shifting from questions around *why* addressing socio-political recovery to *how* we can address user's holistic well-being.

At the same time, only a small number of studies included caregivers' perspectives. Findings from these studies suggest the recovery approach has not yet permeated this group's view, and further attention to informal carers in research would be a step toward recognizing their potential to contribute to mental health care and users' well-being. Users and caregivers should be included as partners in the development of knowledge and services to ensure their personal needs and external challenges are accounted for and met.

Lastly, research into recovery identified in this review demonstrated important characteristics that helped to mitigate bias. Studies benefitted from patient and public involvement; ethnographic methodologies, which allow for study of individuals who are not usually inclined to engage in research activities otherwise; the use of measures such as autovideography to allow participants to shape their own data freely; and mixed methods that allow for the inclusion of larger samples, such as Delphi studies used for questionnaire development.

Strengths and Limitations

The findings in this review should be considered within the context of its strengths and limitations. To the authors' knowledge, this is the first systematic review to examine users and caregivers' understanding of recovery. The use of PRISMA guidelines and quality assessment of the studies added transparency and rigor to the research. However, research about recovery from the perspective of people of diverse backgrounds seemed to only start being documented in recent years. Despite applying a comprehensive search strategy, the evidence found in this review originated mainly from high-income, white-European populations due to a paucity of research in the field of recovery outside of these groups. Therefore, applicability of these findings outside of this context should be done with caution. Additionally, the proposed model of recovery could be strengthened in the future by researching gray literature or literature about concepts adjacent to recovery, such as studies which focused specifically on the notion of hope, empowerment, or social inclusion.

CONCLUSION

The SPICE model of recovery proposed in this review provide context and depth to the construct of recovery, and add further evidence to emphasize the importance of social and clinical aspects of recovery. The comprehensive recovery model provided by this review offers a template for further research in the field and a guide for policy and practice development.

Evidence-based recovery research and practice relies on accurate representations of recovery goals and experiences in order to adequately address people's needs. With sufficient attention to holistic models of recovery that represent the broad range of domains that interest users and carers, along with the promotion of their active participation, the recovery movement can continue toward fulfilling its commitment to have people with lived experience at the center of decision-making in mental health.

DATA AVAILABILITY STATEMENT

The original contributions generated for the study are included in the article/**Supplementary Material**, further inquiries can be directed to the corresponding author/s.

AUTHOR CONTRIBUTIONS

NV participated in the conception, analysis, interpretation of data, drafting, revising, and final approval. PG and MB participated in analysis, interpretation of data, revising, and final approval. MH, SE-L, and VL participated in the conception, interpretation of data, revising and final approval. AO participated in analysis and final approval. All authors contributed to the article and approved the submitted version.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2021.712026/full#supplementary-material

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Lifestyle and Symptom Management Needs: A Network Analysis of Family Caregiver Needs of Cancer Patients

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Previous research on the needs of family cancer caregivers (FCCs) have not elucidated associations between specific caregiving needs. Network analysis, a statistical approach that allows the estimation of complex relationship patterns, helps facilitate the understanding of associations between needs and provides the opportunity to identify and direct interventions at relevant and specific targets. No studies to date, have applied network analysis to FCC populations. The aim of the study is to explore the network structure of FCC needs in a cohort of caregivers in Singapore. FCCs ($N = 363$) were recruited and completed a self-report questionnaire on socio-demographic data, medical data on their loved ones, and the Needs Assessment of Family Caregivers-Cancer scale. The network was estimated using state-of-the-art regularized partial correlation model. The most central needs were having to deal with lifestyle changes and managing care-recipients cancer-related symptoms. The strongest associations were between (1) having enough insurance coverage and understanding/navigating insurance coverage, (2) managing cancer-related pain and managing cancer-related symptoms, (3) being satisfied with relationships and having intimate relationships, and (4) taking care of bills and paying off medical expenses. Lifestyle changes, living with cancer, and symptom management are central to FCCs in Singapore. These areas deserve special attention in the development of caregiver support systems. Our findings highlight the need to improve access to social and medical support to help FCCs in their transition into the caregiving role and handle cancer-related problems.

Keywords: caregiver needs, cancer, network analysis, central needs, lifestyle, symptoms management

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INTRODUCTION

Family caregivers of cancer patients play a crucial and essential role in care recipients' cancer journey, particularly as patient care moves from inpatient to ambulatory and home settings (1). Throughout this journey, family cancer caregivers (FCCs) themselves may encounter specific needs as a result of many complex factors. A caregiver's need is "unmet" if action or resources taken to attain optimal well-being do not satisfy or resolve the need (2). Previous literature found a consistent association between unmet psychosocial needs and poorer caregiver mental health across the patient's cancer journey (3). Specific unmet needs such as perceived information needs have been associated with higher odds of FCC anxiety (4). While both cancer patients' and FCCs' needs are important as they affect the patient-caregiver relationship (5), quality of life (6), and FCCs' psychological health (7), they may be more dire for FCCs than for patients (2). Therefore, identifying these needs during the cancer patient's treatment journey is crucial in supporting caregiving efforts.

Although research to date has provided much insight on FCCs needs, specific questions on complexities of needs, and their relationships and interactions remain unanswered (8). Needs are complex and increased by interactions, especially at different phases of the disease (9). Recent research methodologies are only able to inform how certain clustered needs in general, such as financial, social, medical, or other needs, influence the caregiving experience. They cannot identify how specific needs in those dimensions affect caregiving or how important specific needs are to FCCs. Identifying specific needs of FCCs can further provide an understanding of the relationship between these needs. For example, FCCs personal time may be restricted as they need to care for their loved ones (10, 11). This may lead to FCCs distancing themselves from family and friends, resulting in an escalation of needs for social and emotional support (2). As such, specific needs may interact and reinforce one another. More importantly, this crucial information may help with effective intervention designs that target these needs or cluster of needs. Therefore, investigating specific needs may answer and provide more insights into the FCC experience.

A novel method to investigate FCC needs and their association is network analysis. Network analysis is an emerging graphical methodology in psychology and has been used to analyze the relationships (edges) between variables (nodes). Complex relationship patterns can be estimated and the network structure can be analyzed to establish core features and properties between nodes (12, 13). Network analysis has been used to investigate associations between symptoms in psychopathology, e.g., symptoms of depression (14) and post-traumatic stress (15). In the context of FCC, it can be used to investigate the relationship between various FCC needs.

The classical theory test or item response theory assumes that constructs arise due to causal interactions between their elements (16). In other words, items do not necessarily arise due to a latent construct, and neither do latent constructs necessarily cause variation in item responses (17). Instead, items are causally dependent on each other to form a network or construct (18). For instance, FCC financial needs do not cause variation in the items; rather, these items covary to cause variation in financial needs. FCCs experience needs due to the accumulation of and interaction between needs in various dimensions and phenomena. For example, FCCs may need to look after their loved ones while sacrificing their own time, learn more about the disease, and manage finances. These distributed and diverse needs come together to form a complex "needs" structure which can inform us on the salient and influencing needs FCCs experience, the relationship among these needs and other needs. Based on this, interventions can be more accurately targeted and introduced to improve FCCs needs, thus modifying and improving the needs structure.

To date, no studies have applied network analysis to examine the needs of FCCs; only two studies have examined symptom experience of cancer patients (19, 20). The aim of this study was therefore (1) to explore the needs of FCCs in Singapore and their interrelatedness via network analysis, and (2) establish the strengths and "centralness" or importance of these needs to FCCs. We hypothesize high interrelatedness of needs, however no a priori hypothesis was made as to which of these were central or of importance to FCCs in Singapore. To our best knowledge, this is the first study to examine FCC needs using network analysis.

METHODS

Participants and Procedure

FCCs ($N = 517$) of cancer patients (aged 21 and over) followed up in ambulatory clinics at the National University Cancer Institute Singapore were invited to participate in this study. Participants were recruited from May 2017 to December 2017. Inclusion criteria were: (1) Singapore citizens or permanent residents between 21 and 84 years of age, and (2) able to read and understand English. Details on participant recruitment are described elsewhere (21). Convenience sampling method was used as caregivers and their care recipients were most accessible at the clinics. FCCs completed a questionnaire on socio-demography and their care recipient's cancer diagnosis and treatment, and scales to assess their mood state, quality of life, caregiving burden, and needs at home as part of a larger study (21). Four-hundred-and-five participants returned the forms. Forty-two participants were excluded from the analysis for the following reasons: they were not English speakers ($N = 6$), did not complete the NAFC-C ($N = 25$), were not family members ($N = 9$), withdrawal from the study ($N = 1$), and care recipient's diagnosis being revised to "no cancer" ($N = 1$). Hence, a total of 363 participants were included in the final analysis. The study had Ethics Board approval (NUS-IRB Reference No. 2017/000/29, Received: 25 April 2017), and written informed consent was obtained.

Measures

Sociodemographic and Medical Characteristics

Participants completed a self-report questionnaire which collected two types of variables: (1) demographic variables comprising of age, sex, ethnicity, marital status, education, employment, income per capita, and identity of care recipient; and (2) medical variables of the care recipient comprising of type of cancer, cancer stage, and type and length of treatment and whether it was completed.

Needs Assessment of Family Caregivers- Cancer (NAFC-C)

The NAFC-C is a 27-item scale that measures different cancer caregiver needs on two dimensions: the importance of the need and the satisfaction with the fulfillment of the need during the past 4 weeks (3). Both dimensions are measured on a five-point

Likert-type scale ranging from 0 (Not at all) to 4 (Extremely). Satisfaction rating was reverse coded for each item. For each item, needs score was computed by multiplying satisfaction with importance rating, yielding a range of 0 to 16, with a higher score indicating a higher index of un-fulfillment. The scale consists of four factors (1) psychosocial unmet needs, (2) unmet medical needs, (3) unmet financial needs, and (4) daily activity unmet needs. Our previous study found acceptable validity of the NAFC-C to be used in an Asian population like Singapore (21). In this sample, Cronbach's alpha was strong ($\alpha = 0.90$).

Statistical Analysis

Three steps were taken to analyze the data: (1) descriptive statistics, (2) network estimation, and (3) network stability. All analyses were conducted in R 3.5.3 loading on R Studio 1.3.842.

Network Estimation

The NAFC-C network was estimated using a Gaussian Graphical Model (GGM), in which edges (associations between needs) represent estimations of partial correlations between nodes (needs). As two nodes are connected in the resulting network, their connections have been controlled for connections to all other nodes in the network. With 27 nodes in the network, there are 351 possible pairwise connections between nodes, and these were estimated in the network model. The least absolute shrinkage and selection operator (LASSO) was applied to the network to identify relevant edges (pairwise connections) and reduce spurious connections, i.e., false-positive connections (22). In short, LASSO shrinks very small edges to zero. The tuning parameter (λ) was selected empirically by applying the Extended Bayesian Information Criterion (EBIC). A more detailed tutorial on how to perform this procedure can be found elsewhere (23).

To examine the importance of each need (node) in the network, strength centrality was computed. A central node exhibits many connections in the network; removing or altering that node will likely result in large changes in the entire network. In short, strength centrality (node strength) measures the relationship between one node and all other nodes in the network. A node with high strength centrality has many connections with other nodes relative to the rest of the network. We only reported strength centrality, as betweenness and closeness centrality were unreliable in recent network analysis (24). Hence, in line with other research we reported node strength only (15). In addition to centrality indices, the predictability of each node will also be calculated using the *mgm* package in R (25). Predictability explains the shared variance of each node with all its direct neighbors (26). It provides an absolute measure of the interconnectedness in the network and, therefore, an idea of the connections' practical relevance (26). In a way, it also quantifies how much a node can be influenced by intervening in all of its neighbors. Higher shared variance between the nodes and their neighbors indicates greater interconnectedness between these nodes.

Network Stability

In line with current best practices (27), the accuracy and stability of the network were also estimated using the *bootnet* package (27). To calculate the stability estimates of the centrality indices, each centrality index was bootstrapped 1,000 times with non-parametric samples at 95% confidence interval (27). Centrality stability coefficient ranges from 0 to 1, with values > 0.25 indicate moderate stability and values >0.50 indicate strong stability. Accuracy was investigated by plotting the bootstrapped confidence intervals to examine the variability in the edge weights (27).

Identification of Needs of Similar Construct or Processes

In network analysis, some nodes likely measure the same underlying construct, i.e., nodes are collinear. Hence, the *goldbricker* function within the *networktools* package (28) was used to identify potential pairs of nodes that correlate strongly with each other in highly similar patterns with other nodes (topological overlap). In essence, goldbricker identifies pairs of nodes that are strongly inter-correlated ($r > 0.50$) and are sharing at least 75% topological overlap, or <25% of significant divergent dependent correlations at $p < 0.05$ (29).

RESULTS

Demographics and Clinical Characteristics

The response rate of the study was 78.33% ($N = 112$ unreturned forms). **Table 1** presents the socio-demographic and clinical characteristics of the participants. About three-fifths of the participants were female ($N = 227$, 62.50%), and almost all participants had at least secondary (10 years) education ($N = 347$, 95.60%). The ethnicity distribution for this sample for Chinese ($N = 263$, 72.50%), Malays ($N = 59$, 16.30%), Indians ($N = 27$, 7.44%), and others ($N = 12$, 3.31%) is comparable to the Singapore population ethnicity distribution at 74.35, 13.49, 8.96, 3.21%, respectively (30). Furthermore, cancer distribution among this sample was similar to the latest census in Singapore (31). The most prevalent relationship with care recipients' are parents ($N = 169$, 46.60%), followed by spouses ($N = 116$, 32.00%).

Network Estimation

Figure 1 shows the estimated network of the NAFC-C, indicating the needs among FCCs. There were 149 non-zero edges out of 351 edges which indicated associations between FCC needs. Furthermore, all needs were positively correlated with each other, with higher values indicating more needs. The strongest edges (depicted by thicker blue lines in **Figure 1**) emerged between *having enough insurance coverage* (Item 7) and *understanding/navigating insurance coverage* (Item 17), *managing cancer-related pain* (Item 19) and *managing cancer-related symptoms* (Item 20), *being satisfied with relationship* (Item 22) and *having an intimate relationship* (Item 24), and *taking care of bills* (Item 3) and *paying off medical expenses* (Item 11). Of the four pairs, two were associated with financial needs, one was associated with medical needs, and the remaining was associated with psychosocial needs. *Dealing with lifestyle changes* (Item 25) and *managing cancer-related symptoms* (Item 20) had the highest strength centrality (**Figure 2**).

TABLE 1 | Participants demographics.

Socio-demographic and medical variables	***N* (%^{a})**
Sex	
Male	136 (37.50)
Female	227 (62.50)
Race	
Chinese	263 (72.50)
Malay	59 (16.30)
Indian	27 (7.44)
Others	12 (3.31)
Age group (years)	
21–30	61 (16.80)
31–40	68 (18.70)
41–50	86 (23.70)
51–60	79 (21.80)
61–70	52 (14.30)
71–80	12 (3.31)
Education	
No formal education	2 (0.55)
Primary (Some/Completed)	12 (3.31)
Secondary (Some/Completed)/*N*, O Levels / ITE	104 (28.70)
A-Levels/poly diploma	95 (26.20)
Bachelor's degree	116 (32.00)
Masters/Ph.D.	32 (8.82)
Education (≥High school)	
Yes	347 (95.60)
No	14 (3.86)
Marital status	
Single	119 (32.80)
Married	221 (60.90)
Divorced/Separated	7 (1.93)
Widowed	2 (0.55)
Employed	
Yes	238 (65.60)
No	118 (32.50)
Income (per capita)	
$2,000 & below	77 (21.20)
$2,001–$8,000	166 (45.70)
$8,001 & above	58 (16.00)
Relationship with care recipient	
Spouse	116 (32.00)
Parent	169 (46.60)
Grandparent	8 (2.20)
Son/daughter	19 (5.23)
Sibling	30 (8.26)
Others	1 (3.03)
Type of cancer	
Breast	73 (20.10)
Lung	68 (18.70)
Gastro-intestinal/Colorectal/Stomach	59 (16.30)
Hematological/Leukemia/Lymphoma/Myeloma	54 (14.90)
Gynecological	16 (4.41)
Pancreas	11 (3.03)
Multisite	12 (3.31)
NPC/Throat/Oral	13 (3.58)
Renal	8 (2.20)
Brain tumor	6 (1.65)
Cancer stage if known	
Early (stages 0–2)	63 (17.40)
Late (stages 3–4)	247 (68.10)
Is treatment completed?	
No	265 (73.00)
Yes	83 (22.90)
Type of treatment completed	
Chemotherapy	142 (39.10)
Radiotherapy	96 (26.40)
Surgery	147 (40.50)

[a] *Percentages might not sum up to 100% due to missing data, or rounding difference.*

The average predictability of nodes was 0.34, indicating that, on average, 34% of the variation in one node is explained by its direct neighboring nodes. The predictability of each need is shown in **Table 2** and **Figure 1** as a black area around the rings. *Dealing with lifestyle changes* (Item 25), *managing cancer-related symptoms* (Item 20), *understanding/navigating insurance coverage* (Item 17), *getting information about cancer diagnosis* (Item 14), *satisfaction with relationship with other family members and friends* (item 18) demonstrated the highest predictability at 0.53, 0.47, 0.45, 0.34, and 0.40, respectively.

Network Stability

The correlation stability coefficient (CS [cor = 0.7] = 0.36) for the strength centrality metric exhibited moderate stability, although it was below the recommended cut-off at 0.50 for strong stability (27). The confidence intervals around the edge weights were large, and most of the confidence intervals overlapped, indicating that their order should be interpreted with caution (see Supplementary Materials for the edge weights).

Needs Underlying the Same Construct or Process

The identification of needs with a topological overlap revealed six pairs of needs that may have a high conceptual overlap and may be better explained as multiple measurements of the same process or construct: (1) *best possible care* (Item 2) and *communicating with medical staff* (Item 6), (2) *taking care of bills* (Item 3) and *paying for his/her medical expenses* (Item 11), (3) *communicating with medical staff* (Item 6) and *getting information about cancer diagnosis* (Item 14), (4) *getting involved in medical decisions* (Item 10) and *getting information about cancer diagnosis* (Item 14), (5) *managing cancer-related pain* (Item 19) and *managing cancer-related symptoms* (Item 20), and (6) *satisfied with relationship* (Item 22) and *having an intimate relationship* (Item 24). The first

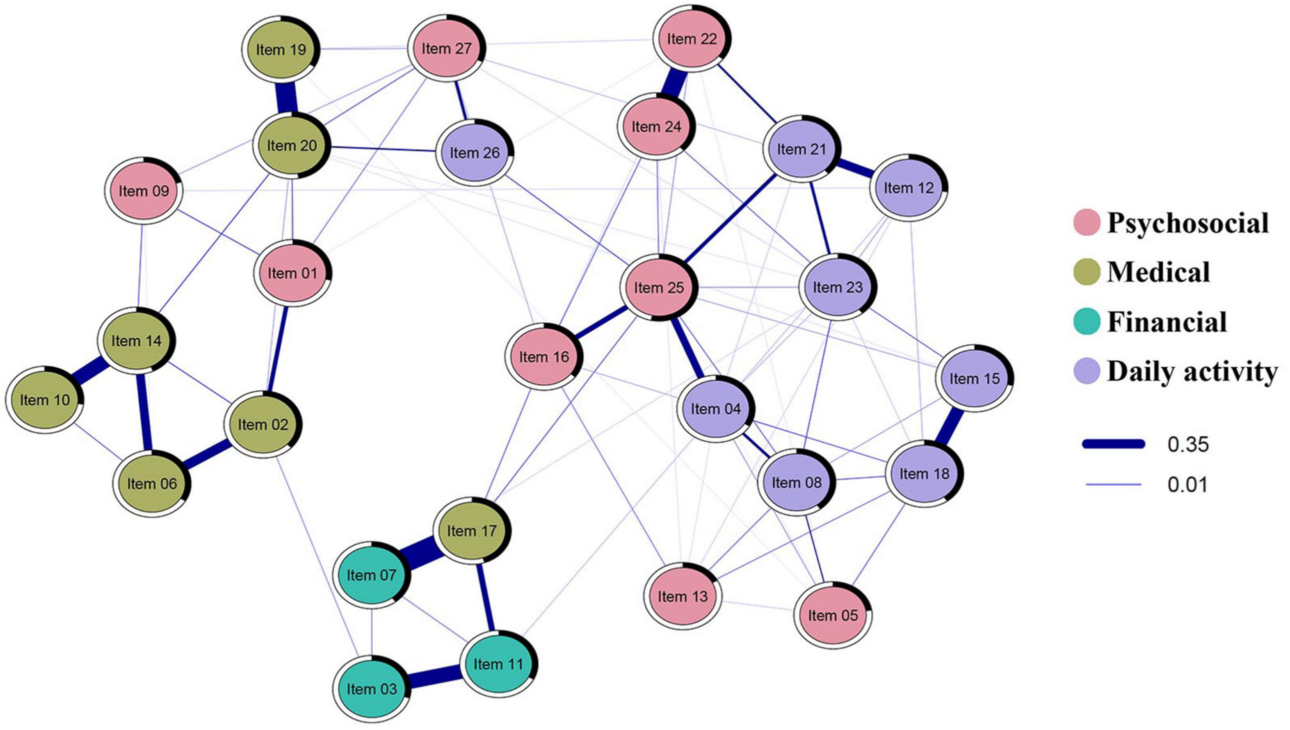

FIGURE 1 | Estimated regularized partial correlation network plot for the NAFC-C ($N = 363$). Only edges that have associations are shown. Blue lines show the positive association between needs; the thickness of the lines represents the strength of the association between needs. The thicker the line, the stronger the relationship between the two needs. The black area around the rings shows the predictability, the variance of a given node explained by all its neighbors. Nodes are colored by their factors in the NAFC; psychosocial needs are colored red, medical needs are colored olive, financial needs are colored green, daily activity needs are colored purple.

five pairs are associated with medical needs, while the last pair is associated with psychosocial needs.

DISCUSSION

This study aimed to investigate the needs of FCCs via a network approach. We used a partial correlation model applied with LASSO and EBIC to identity the NAFC-C network. Our results revealed that FCCs have critical needs across several distinct domains, i.e., financial needs regarding medical bills and coverage, social and interpersonal relationship changes, and medical-related needs.

Overall, network analysis revealed that connections between needs were positive, confirming our hypothesis. This is not surprising as FCC's needs are highly interrelated (2). However, our analysis revealed that the network has low stability (CS = 0.36), compared to the recommended threshold (CS = 0.50) for strong stability (27). One reason for this is the heterogeneity of the sample. In our study, we had recruited FCCs with care recipients in different phases of the cancer journey (including some who had completed treatment), at different cancer stages, and different cancer types. These factors are associated with distinct needs of care recipients and FCCs. For example, previous research reported that caregiving stress and lack of social support were important needs of FCCs during the early phase of the cancer journey (3). Others however, have shown that although psychological impact persists through the first 6 months of the care recipient's treatment, they reduced over the year and beyond, suggesting that caregivers adapted to their patient's condition over time (32). Similarly, our previous study found that different cancer treatment phases were associated with distinct needs and related outcomes (33). As FCCs adjust to their new roles, their needs evolve largely determined by their care recipient's situation (34). Despite this, the average predictability across all nodes was 0.36, indicating that 36% of the variance of a node that is not predicted by the intercept model is explained by its neighbors. This is an average level of predictability compared to the results of other network analyses (26).

Core Needs of Caregivers: Lifestyle Changes and Living With Cancer

Network analysis also revealed two important categories of needs: *dealing with lifestyle changes* (Item 25) and managing patients' *cancer-related symptoms* (Item 20). These two needs had the highest strength centrality among other needs, suggesting the important effect and influence of these needs on other needs.

Lifestyle changes are evident during cancer caregiving with FCCs having to adjust and transition from a family member to a caregiver role, sacrifice their personal time and work, and assist their loved ones in daily activities and routines. Our result

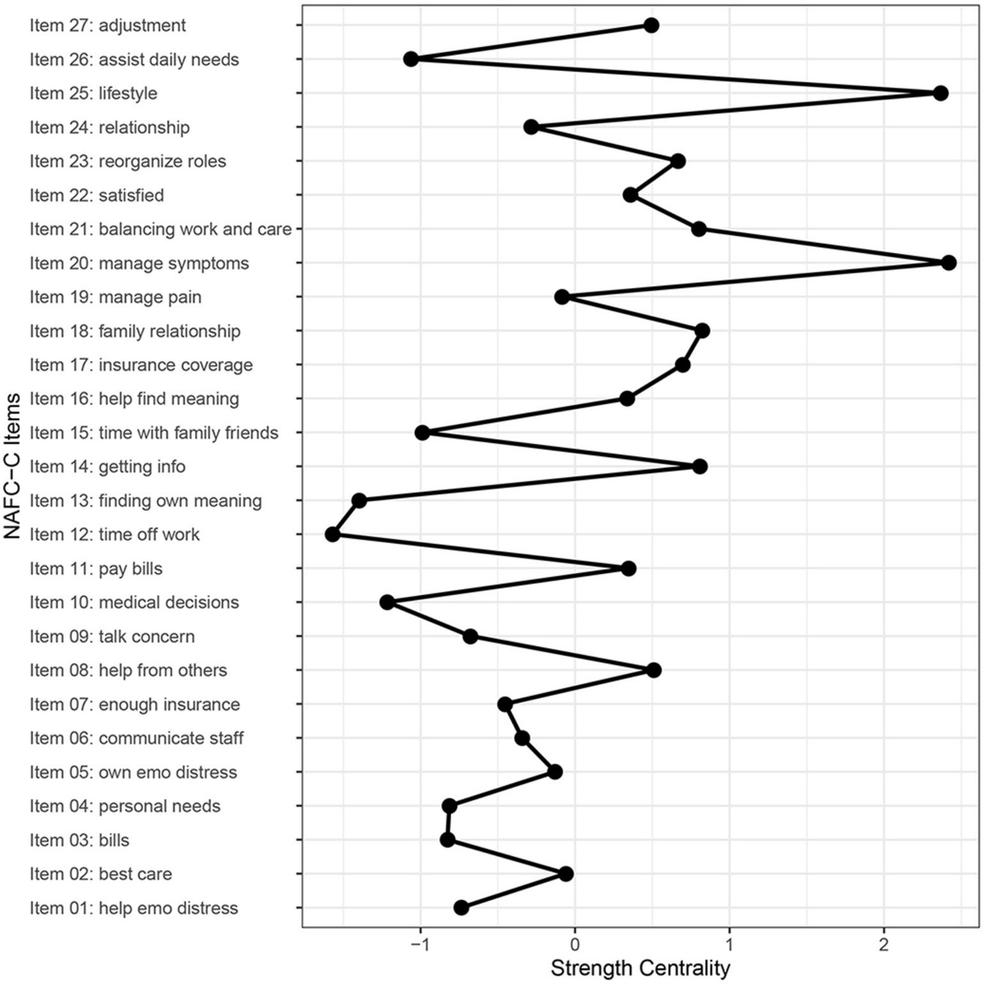

FIGURE 2 | Strength centrality for every item in the NAFC-C. Managing his/her other cancer-related symptoms (item 20) and dealing with lifestyle changes (item 25) had the highest strength while taking time off work (item 12) and finding meaning out of the experience with care recipient's cancer (item 13) had the lowest strength.

is in line with previous literature demonstrating that FCCs need to adjust and adopt a new "normal" after sacrificing their jobs, time, space, and life just to care for their sick loved ones (11). This is shown through the strong connection between dealing with lifestyle changes and *balancing work/school with caring for him/her* (Item 21). FCCs face difficulties in trying to relax and manage personal responsibilities due to the additional responsibility of caring for their ill loved ones (10). Furthermore, needs in lifestyle changes were also strongly connected to *meeting personal needs* (Item 4). Our strength centrality results suggest that *helping the loved one find meaning out of cancer* (Item 16) is an important need associated with lifestyle changes. Studies have shown that finding meaning and spirituality in the cancer illness, which can help reduce distress and enhance coping with symptoms, may not be addressed until the disease is in the advanced stages or may even be entirely unaddressed by healthcare teams (35). FCCs themselves may fail to get help in this area and it influences the quality of care they provide and their handling of personal issues (36, 37). Care provision for cancer patients and their caregivers has to be holistic and complementary to psychological, social and medical care to support various aspects of the illness and its progression.

Given that FCCs need to find a new stable job if they had previously quit their job or face added responsibilities in their current one, lifestyle changes exacerbate other problems or needs. Our data support this line of reasoning via high predictability in *lifestyle changes* (Item 25) and *satisfaction with the relationship with other family members and friends* (item 18). As predictability quantifies how much a node can be influenced by intervening in all of its neighbors, higher predictability indicates greater interconnectedness between nodes. We found that needs such as *reorganizing roles among family members* (Item 23), *assisting with daily needs* (Item 26), *meeting personal needs* (Item 4), *finding help from others* (Item 8), *and lack of time with family and friends* (Item 15) *are related to satisfaction with the relationship with other family members and friends* (item 18) *and lifestyle changes* (Item 25) (**Figure 3A**). These factors have become more prominent in recent years as cancer care shifts toward ambulatory care

TABLE 2 | NAFC network metrics.

Items	Description	Strength	Predictability
Item 1	Helping his/ her emotional distress (e.g., anger, anxiety, depression, fear, resentment, etc.)	−0.74	0.28
Item 2	Getting the best possible care for him/her	−0.06	0.37
Item 3	Taking care of bills	−0.83	0.29
Item 4	Meeting your personal needs	−0.81	0.34
Item 5	Dealing with your emotional distress (e.g., anger, anxiety, depression, fear, resentment, etc.)	−0.13	0.22
Item 6	Communicating with his/her medical staff	−0.34	0.34
Item 7	Having enough insurance coverage for him/her	−0.45	0.40
Item 8	Getting help from others in order to take time for yourself	0.51	0.40
Item 9	Talking to him/her about his/her concerns	−0.68	0.21
Item 10	Getting involved in medical decisions affecting him/her	−1.21	0.27
Item 11	Paying for his/her medical expenses	0.35	0.32
Item 12	Taking time off work	−1.57	0.27
Item 13	Finding meaning out of your experience with his/her cancer	−1.40	0.17
Item 14	Getting information about the cancer he/she was diagnosed with (e.g., prognosis, treatment, side effects, nutrition)	0.81	0.43
Item 15	Getting together with family and friends	−0.99	0.28
Item 16	Helping him/her find meaning out of cancer	0.34	0.35
Item 17	Understanding/ Navigating medical and/or insurance coverage	0.70	0.45
Item 18	Being satisfied with your relationship with other family members and friends	0.82	0.40
Item 19	Managing his/her cancer-related pain	−0.08	0.34
Item 20	Managing his/her other cancer-related symptoms (e.g., fatigue, nausea)	2.42	0.47
Item 21	Balancing work/school with caring for him/her	0.80	0.38
Item 22	Being satisfied with your relationship with him/her	0.36	0.36
Item 23	Reorganizing roles among family members	0.67	0.39
Item 24	Having an intimate relationship with him/her	−0.28	0.38
Item 25	Dealing with lifestyle changes	2.37	0.53
Item 26	Assisting with his/her daily needs (e.g., preparing meals, transportation, etc.)	−1.06	0.27
Item 27	Helping him/her adjust to life after cancer	0.50	0.32

and home settings (1). Eventually, caregivers become lonely and require social support but rarely have the ability nor time to seek help (38). The immense caregiving demands can lead to FCCs to have no time to look after their own personal and social life. This was shown in our data where FCCs needed more time and were unsatisfied with the relationships with their family members and friends, and needed help from others to assist daily needs and other various tasks. Intervening to address these needs will directly impact the personal and social life of FCCs. These needs are therefore critical nodes that policymakers, clinical administrators and service providers need to consider to improve the personal lives of FCCs.

Network analysis also revealed the significance of managing cancer-related symptoms for FCCs. *Managing cancer-related symptoms* (Item 20) had high connections with several other items associated with living with cancer, such as *helping with patient's distress* (Item 1), *getting the best possible care and information* (Items 2 & 14), *managing cancer-related pain and symptoms* (Item 19), and *assisting with life with cancer* (Items 26 & 27) (**Figure 3B**). This cluster can be seen in two dimensions: managing medical symptoms, and managing psychosocial aspects and daily living, i.e., living with cancer. Caregivers are very involved and invested in the management of medical, psychosocial, and daily needs, of their care recipients. The strong correlation between Items 19 and 20 (cancer related pain and symptoms) demonstrate significant challenges FCCs face in helping their care recipient in dealing with, and managing pain and other symptoms such as fatigue and nausea. Pain is an unpleasant distressing emotional experience with psychological effects, which affect FCCs as well (7, 39). In addition to managing cancer-related symptoms, there were also strong connections for psychosocial distress (Item 1) and assisting with patient needs (Items 26 and 27).

Providing the Best Possible Medical Care for the Patients

Our analysis also revealed high conceptual overlap between many medical needs, such as *finding the best possible care for the patient* (Item 2), *communicating with medical staff* (Item 6), *getting involved in medical decisions* (Item 10), *getting information about cancer diagnosis* (Item 14) and *managing cancer-related pain and symptoms* (Item 19 & 20). Although there were correlations among items associated with financial needs, they were not of high predictability nor were they central needs of the FCCs. This is not surprising as in Singapore, medical care is comprehensive, readily available and supported by various healthcare financing schemes for its citizens (40). The high topological and conceptual overlap within medical needs suggest that FCCs work diligently

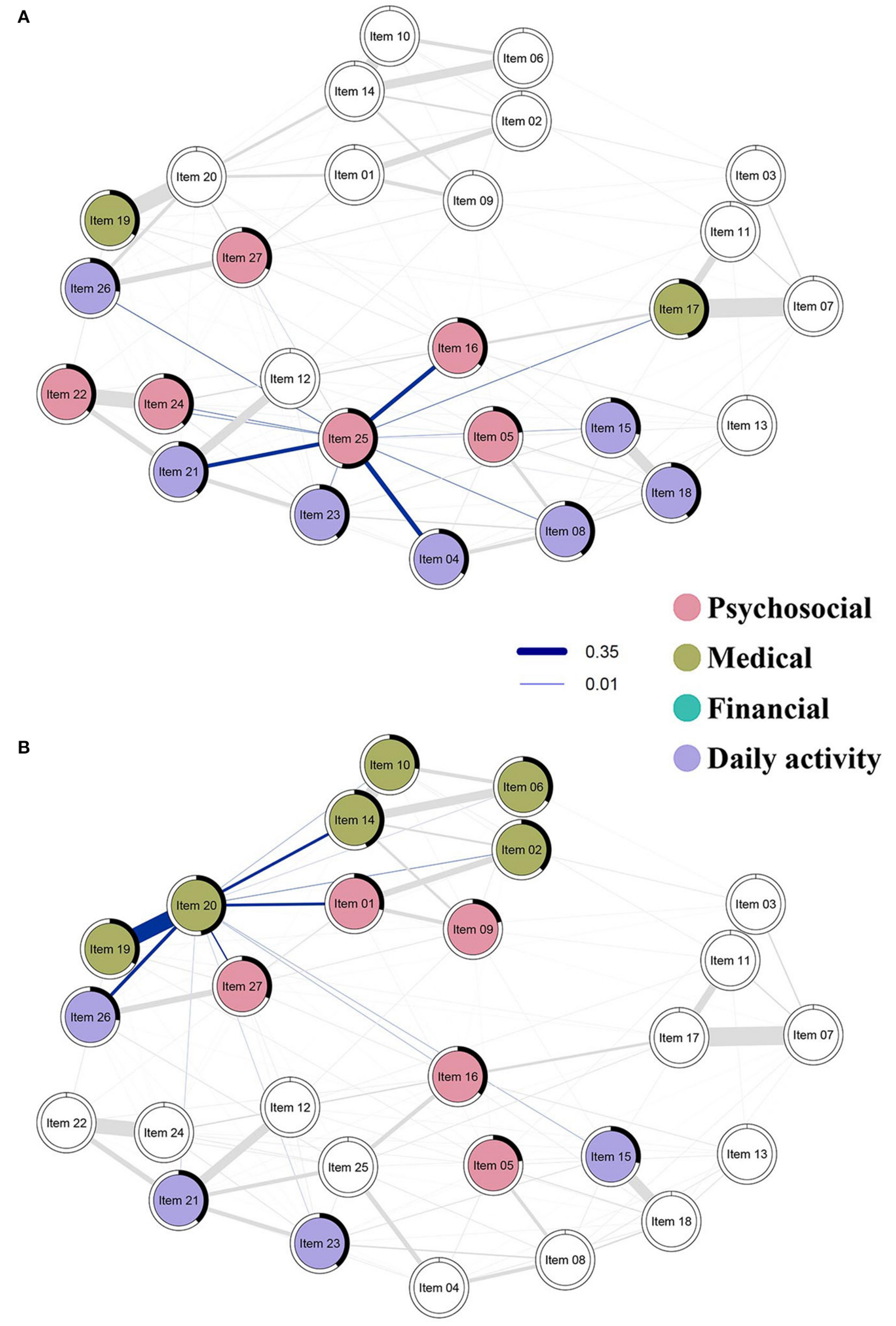

FIGURE 3 | Strength centrality and predictability of top two nodes with highest strength centrality are highlighted in this figure. Blue lines show the positive association between needs; the thickness of the lines represents the strength of the association between needs. The thicker the line, the stronger the relationship between the two needs. The black area around the rings shows the predictability, the variance of a given node explained by all its neighbors. Nodes are colored by their factors in the NAFC; psychosocial needs are colored red, medical needs are colored olive, financial needs are colored green, daily activity needs are colored purple. **(A)** Strength centrality and predictability of dealing with lifestyle changes (Item 25) and **(B)** managing other cancer-related symptoms (Item 20).

to look after their loved ones and provide the best possible care for their care recipient. This result also supports previous studies that demonstrated the involvement of family members in cancer caregiving in Asian societies (41). Duty and filial piety are drivers for FCCs to strive to provide the best possible care for their care recipients, including finding the best medical options for them. Cancer patients and their family often want more and precise communication with their doctors so that they could make the best decisions (42).

Implications

Our study provides several implications for clinicians and the healthcare administration. Firstly, it must be recognized that while the needs of the FCC and their care recipient can be addressed as a dyad, the FCC and the patient have specific individual needs that need to be addressed separately. While psychosocial support for both FCCs and cancer patients benefits them in coping with daily life, support groups or individualized counseling sessions with advice and interventions based on sociocultural and personal context would provide additional benefit to FCCs. Secondly, healthcare administration must strengthen medical resources, information and awareness on cancers to help patients and FCCs to better manage symptoms and live with cancer; several measures such as psycho-education and case management services can be used to address this. Finally, the strong association with needs for more information, involvement and communication with medical staff suggest the need for streamlined communication channels and easier accessibility, engagement, and regular team meetings with healthcare staff (42). It has been suggested that a concordant model of communication supports all parties in fully participating and sharing perspectives on diagnosis and treatments (43).

Limitations and Future Research

There are several limitations in this study that need to be addressed. First, this study cannot answer if needs are temporally related; we could only inform their associations. Therefore, we did not speculate if specific needs in one dimension will lead to needs in another dimension. A longitudinal study may be better equipped to answer this important question. Second, our network stability result showed low to moderate stability (CS = 0.36). Upon examining Cronbach's alpha, our data showed very good reliability (Cronbach's $\alpha = 0.90$, 95% CI = [0.88 0.91]). The NAFC-C was originally developed with the United States population, which has very different population characteristics than the Singapore population (3). This was also shown in our previous validation study as FCCs may interpret items differently from their United States counterpart (21). Hence, other than differences in characteristics of needs, there may be other unexplored needs specific to the Asian population. Given that FCCs have different needs across the patient's cancer journey (34), and we were underpowered to split our sample into different treatment or cancer phases, we were not able to examine the differences and stability of the network structure between different cancer [treatment] phases. This may be better examined with larger sample sizes and in the subgroups. Relatedly, we did not control for possible confounding factors such as demographics and treatment or cancer phases. For example, different ethnic groups in Singapore may have distinct needs which this study could not identify. In the next step, researchers might include these variables to generate a stricter network model or examine differences in needs among ethnicities. Future research using network analysis should also include FCC-related outcomes, especially quality of life, burden, and mood symptoms, to gain insights on how specific needs bridge between other needs and outcomes. Finally, it is now recognized that some cancers can become chronic illnesses similar to other chronic medical conditions. Network analysis might be useful for a more explicit understanding of caregiver needs in the latter group and comparisons of both groups for service planning.

CONCLUSIONS

Overall, our results generate new insights into the needs of FCCs from a network perspective. This study adds relevant and crucial information regarding specific needs for research, social, and clinical support of FCCs, which could not be known through average scores. Needs pertaining to lifestyle changes, living with cancer, and symptom management seem to be central to FCCs in Singapore, and therefore deserve special healthcare administrative attention in developing a support care system for them. FCCs have been found to put effort and time into caring for their care recipients, with less time for themselves. Our findings highlight the need for improved access to and availability of, psychosocial and medical support, to help FCCs with role transitions in caregiving and dealing with cancer illness related problems.

DATA AVAILABILITY STATEMENT

The data is available from the corresponding author on reasonable request and subject to Ethics Board approval.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by National Healthcare Group Domain Specific Review Board Ethics Committee. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

The study was conceptualized by RM, YC, KG, and SK. WY did data analysis with statistical advice from YC. The manuscript was drafted by WY and RM with inputs and reviews by YC, KG, and SK. All authors contributed to the article and approved the submitted version.

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Experiences of Being Cared-For: The Perspective of an Expert-by-Experience in Mental Health

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It is difficult to understand what it feels like for people with mental ill-health to be cared-for and supported by family members; this experience is often little-explored. Narratives about caring have been increasingly written alongside first-person accounts of recovery, however, there is a dearth of literature written to gain the perspective of being cared-for because of mental distress. Thus, using autoethnography, I present three critical incidents occurring at different points in my recovery to enable exploration of experiences of being cared-for. Firstly, a critical incident at the point of acute unwellness is introduced, secondly an incident during a consultation with a health professional is highlighted, and finally a moment of transition when embarking on an independent life with my husband-to-be is described. I use autoethnography to connect "the autobiographical and personal to the cultural, social, and political". I consider how the identity of a carer is continually negotiated in a relationship with the service user in both the "private" and the "public" worlds during recovery. I reflect on how professionals can support both service users and carers in a triangle of care, by providing information and support, alongside promoting the development of independence and agency for the service user whilst in the caring relationship. Finally, I introduce a service model which promotes a family network approach to empower the service user, and highlight training programs on recovery that enable carers. I conclude by suggesting the potential of both approaches to support carers to promote the recovery of the service user.

Keywords: caregivers, carers, mental distress, autoethnography, expertise-by-experience

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INTRODUCTION

It is difficult to understand what it feels like for people with mental ill-health to be cared-for and supported by family members; this experience is often little-explored. Narratives by caregivers have increasingly been written about their experiences of providing care to people with mental distress (1–3), in which they document frustrating and challenging, as well as joyous and hopeful moments of caring; however, it is clearly noted the tasks of caring that can impact negatively on both a person's physical and mental health (4). Of interest is a burgeoning body of first-person narratives written by experts-by-experience highlighting the nature of their recovery journey and experiences of mental ill-health (5), however there is a dearth of literature written to gain their perspective of being cared-for. There is furthermore a paucity of research to understand the perspective of being cared-for using experiential and autoethnographic methods. This article thus seeks to begin to bridge this gap in understanding this phenomenon.

In this article I reflect on my experiences of being cared-for by family members both when I was in the acute phases of psychosis and later when experiencing a long recovery. Using a process of reflection and the presentation of three critical incidents (6), I draw on autoethnography (7, 8) to explore my thoughts, feelings, and memories of being cared-for at different stages in my recovery journey. These reflective memories are contextualized in the wider social and cultural context (7, 8) and their relevance is highlighted.

The Methodology: Telling Reflective Memories

Reflective practice is of key importance to both health and social care practitioners (9); it has a long tradition of being used in these professions as a method to develop both personal understanding of the lived experiences of service users and carers (10) and of innovation in practice (11). Autoethnography is a methodology that utilizes a process of reflective writing to provide an understanding of experiences in the wider social context (7, 8, 11). It has been increasingly used in health and social care research, education, and practice (11, 12). Autoethnography is thus employed in this article to explore my experiences of being cared-for, through a process of writing and reflection.

In this article I present three reflections, deliberately selected from critical points in my recovery journey because they evidence a step change in the experiences of being cared-for. These moments indicate a transition in the dyadic relationship between myself and my carers and mark a shift in my recovery journey. Alongside these three chosen moments of change, there is a concomitant reflection of the significance of this transition, thus allowing the connection of "the autobiographical and personal to the cultural, social, and political" (13). Throughout this process, autoethnographic writing (7) requires the researcher to pay careful attention to both the *epistemic* (claims to knowledge) and the *aesthetic* (practices of imaginative, creative, and artistic craft) characteristics of their texts as they seek to convey the meaning of their individual experiences and communicate their significance to the wider community of practice.

Critical incident reflections, as used in this article to capture my memories, are an approach often used in social work (6) and demonstrate how professional perspectives can provide new insights for service users, carers, practitioners themselves and their wider professional group. Although the content of the reflections is presented at a distance from my experiences, impacting on their potential accuracy, they still convey a very vivid description. Additionally, it could be argued that the accuracy of these stories may be blurred by the experiences of psychosis or could be unclear because of medication side effects; however, it is my contention that the value of first-person narratives is increasingly recognized and their role in highlighting the authentic experience of using services is highly regarded by service users, carers and many professionals (14). Moreover, as far back as the early 2000s, the importance of seeking the opinions and experiences of inpatients in mental health wards was recognized through the implementation of systematic user-focused monitoring (15). This highlighted the importance of listening to service users' opinions even when they were experiencing symptoms of mental ill-health or had blunted cognition because of the side effects of medication. In the next section, I provide three reflections on my experiences of being cared for, all of which occur at chosen points in my recovery.

The Findings: Reflective Memories

Reflection 1 took place at the beginning of first psychosis and extreme paranoia in 1990. I remember at the height of my terrors, when I was horrified at what I perceived to be happening around me, I was hearing voices in my head and had active symptoms of paranoia. At this moment I believed that I had superpowers and was going to be used by MI6 to stop terrorism in Northern Ireland. My parents had come up to Durham (in the north of England), where I was at university, knowing that something very distressing was happening to me, but not knowing what. I was sharing a room with my mother, as she stayed in a hotel in Durham whilst I was waiting for a psychiatrist's appointment. I was too terrified to stay in my room in college and became so scared that I crawled into bed with her, as I sought physical proximity to my mother to counteract the crisis I was experiencing. I now know the distress my mum experienced as she was unable to alleviate my crisis.

Reflection 2 took place about 11 years later into my recovery in 2001. I remember one mental health appointment I went to. My mother came with me. I was working in London at that time and drove a 3-h round trip each day to my place of work and then back home. My mother drove me to the local hospital because I was tired of driving. She always drove me around locally. I self-managed my mental health and had control of choices about my medication. My psychiatrist suggested that it would be a good idea to come without my mother to the hospital. From my perspective, when my mother accompanied me to the hospital, it had nothing to do with her infantilising me or taking control of my life, rather it was a way of escaping the long driving. However, after that occasion, my mother never came with me again to the hospital. My psychiatrist challenged me in a quiet and professional way to become independent which impacted strongly on my sense of self as I strove to present myself as an adult. I couldn't have survived without the care of my mother, but she fostered a sense of dependency which to some extent infantilised me and removed my agency. As I reflect now, my mother was not able to "let go" in case I experienced a crisis or period of unwellness again.

Reflection 3 took place at a time shortly before I was married in 2005. The final reflection is a turning point as I began an independent life and became responsible for myself, my home, and my own wellbeing. I had lived at home since leaving university at the age of 22. It was only when I met my husband-to-be and he expected me to be a grown adult and no longer a child, that I really grew up. I was entering an exciting and loving relationship and we had a new house together that needed a lot of work, and in the excitement of renovation I began to grow up. I was expected to do my fair share of household chores and to work full-time. I began to reduce the intimacy I had with my mother because her overwhelming care had become over-protective and intrusive. Maybe only now, as I reflect on my relationship with her at the time, do I realize the sense of fear and lack of control she felt in her caring role and the need to protect me when she felt she had failed me at the time of crisis in my first episode of psychosis. She longed to sustain a relationship of intimacy and support, although I rejected that bond. This relationship signified a status of dependency, weakness and vulnerability—all of which I rejected.

The relationship that I shared with my mother was central to my recovery journey. My mother constantly reinforced messages of hope and optimism, which are promoted in the recovery approach, to promote my wellbeing. Such concepts were under-developed at the time of my initial illness, but my mother played a pivotal role in my early recovery. My mother and I were co-dependent, and this relationship suited both her and my needs at different times in my recovery journey. Fostering a sense of dependency in my relationship with her, thus enabled her to protect me and to prevent any risk of harm, but at the same time to hinder any opportunity for growth. Additionally my mother was isolated and lacked the opportunity to learn from other caregivers. I felt uncomfortable with her going to a peer support group—I felt she was sharing my private information with other people, not understanding, as I do now, that she needed support and help to care in her own right. She thus had very little support in her own right, other than that of her husband, my father.

DISCUSSION

This section now seeks to connect "the autobiographical and personal to the cultural, social, and political" by enabling connections to be drawn between my own reflections on my relationship between my mother and me, and the wider research undertaken to consider the identity of family caregiver.

As shared in my reflections, the caring role is hard to define and understand, encompassing different tasks and different roles (16). Moreover the caregiver identity (17) is a socially constructed concept existing both in the public world of mental health services and professionals and in a private world between the carer and the cared-for. It has been socially constructed as a public identity (17) because the enactment of care in the community and the right to receive a carer's allowance and a carer's assessment, has led partners, parents, and siblings to be identified as caregivers. It has also been defined as existing in the private world between the caregiver and the cared-for as they individually negotiate their relationship of care, which is always changing and adapting. Thus, throughout the period of recovery, the caring/cared-for dyad is continually re-constructed in both the private world and the public world as the relationships change and develop, and are renegotiated between the cared for person and the caregiver. This is underlined in my reflections.

Moreover, my three reflections plot the changing relationship over time between my mother and me as we renegotiate our relationships and merge and separate our identities. Aldridge (2), a caregiver and mother of her son with undiagnosed bipolar disorder, highlight**s** the need for caregivers of people experiencing mental distress to balance both the support and care they provide against the need to respect the cared-for person's mental capacity and decision-making rights. She explores the difficulties of managing both care and control, while considering her own role of caring in a situation in which mental health services seemed unable to engage her son. It is often suggested by carers that this relationship should develop into a triad or a "triangle of care" (18) in which the service user, carer and professionals work together to support recovery of the service user. Aldridge (2) however experienced that service provision was missing, and professionals failed to provide appropriate care for her son. My reflections also revealed the importance of professionals including and supporting both me and my mother but reiterated the need to enable me to reassert my agency in treatment choices and appointments, disrupting the sense of co-dependency. Despite this, my mother emphasized a message of hope and optimism as suggested in the recovery model (19), as denoted in the CHIME model 1, and encouraged me to focus on my strengths (20). Such reinforcement can support people who experience mental distress to improve and sustain their sense of wellbeing, as my mother did in my case.

My reflections revealed the crucial role my mother played in my recovery, but also the co-dependency she fostered in our relationship. It is difficult for service providers to strike to correct position in supporting both the service users and the carer. As a population, caregivers often complain that they are under-supported and under-informed by services as they seek to care for their family members (2, 3). Although there may be conflict between the caregiver and the cared-for (21), most caregivers want to be positively involved in the lives of their family members. However, Henderson asks (18, p. 157) whether the caregiver and the cared-for experience any "shared interests and needs" or whether their needs are "incompatible and in conflict". In recognition of these differing experiences, in England and Wales, service users have the right to displace their nearest relative (a legal position that allows them to make certain decisions in the enactment of the Mental Health Act) under section 29 of the Mental Health Act (22). Such an amendment to the law, recognizes that there may be conflict between some service users and carers, and they may not always share the same goals or objectives, suggesting the inadequacy of some relatives to occupy the role of nearest relative.

However, both caregivers and service users may have differing opinions of what constitutes best practice; for my mother it was hard for her to be told that I should be attending the mental health appointment by myself. Ryan (3) shares her experiences of caring for her son, diagnosed with Autistic Spectrum Disorder, describing both the effective as well as the unhelpful services they received from health and social care professionals. She documents how caregivers can feel excluded from professional support, which reduces their involvement in the life of the cared-for and prevents them from experiencing the respect and recognition as an ally. Furthermore, studies reveal that some caregivers experienced (23) powerlessness and lack of control as caregivers alongside a sense of failure from not preventing the admission, when service users entered the hospital. Additionally, they suffered feelings of isolation from the lack of partnership working as professionals took over the management of care. Moreover, one study (24) considers the importance of family caregivers participating in the lives of people who are currently expressing suicidal behavior and are residing in inpatient accommodation. It reiterates the needs for family members to be present and emotionally available for their relative, participating in their lives in such a way as to share everyday life; as well as to participate in joint activities that nurture sources for vitality and encourage thoughts about recovery and wellbeing. My reflections support the contention that professionals should use their professional judgement to recognize the needs of service users at different times in their care, and to use their professional knowledge to enable the caregiver to provide the most effective possible support.

Taking these experiences into account, practice models, such as the Triangle of Care (18), encourage mental health service professionals to involve caregivers in supporting service users. The Triangle of Care is a model which attempts to establish therapeutic alliance between the service user, professional and caregiver. Partnership models are important for different members of the care team at different points in the recovery journey (1); however, these need to recognize when dependency is preventing the independent growth and recovery of the service user. Moreover, internationally, the Open Dialogue programme (25) implemented in Finnish Lapland uses a family-centered approach that focuses on recognizing the significance of all members of the service user's network in their assessment and treatment. This involves regular meetings of all stakeholders in the group with treatment lasting for up to 2 years. It is of increasing influence in the UK (26) and elsewhere—although, with its focus on the user's right to confidentiality and autonomy, practice in the UK would require significant change to implement fully this model.

Finally, caregivers need information and advice to enable them to care effectively, although they have historically received little training or support in their caring role (27). However, recently the implementation of caregivers' education programmes is now recommended in UK government guidance (28), and caregivers are given a right to support in accordance with the Care Act (29). Furthermore, training opportunities have become increasingly available for caregivers of people with specific mental health conditions, such as personality disorder (30, 31) or schizophrenia (32). Despite this, access to forms of peer support and training are often denied to many caregivers by the cared-for person's fears (33). This underlines the need for accessible and sensitive training to support caregivers to care effectively (34). Research I have undertaken (33, 34) has led to the development of a training programme on the recovery approach for family caregivers, co-produced by different stakeholders and co-delivered by myself, in my identity as a service user, and with a caregiver. This programme allows me to share my experiences of mental ill-health to enable carers to understand mental distress and the enablers and barriers to recovery (34). This programme reinforces the intention of this paper to enable carers and service users to reflect on and renegotiate their relationship in the caring dyad.

LIMITATIONS

This article attempts to enable carers to understand better the potential enablers and barriers to an effective relationship with their family member in the process of recovery. However, a potential limitation, is that the autoethnographic methodology itself allows the writer of the reflections to be the primary analyser of the perceptions. This leads to the potential of bias in the process. However, autoethnography emphasizes the primacy of the individual at the center of both the narrative and the analysis and acknowledges the importance of the *self* interacting with the *social context* (7, 8, 13). This thus reinforces the value of the person making sense of their individual experiences through a process of reflexivity and undertaking meaning-making through the connection of the personal to the political (13). It is this individual experience that is thus validated through the process of reflection, recognized as meaningful, and acknowledged as having an important role to play in the construction of knowledge (11).

CONCLUSION

This article has highlighted the experiences of being cared-for from the perspective of an expert-by-experience, using autoethnography (7, 8, 13). I have presented three critical incidents during my recovery journey of my experiences of being cared for, and through wider analysis of these occurrences sought to connect "the autobiographical and personal to the cultural, social, and political" (13). This reflection has led to consideration of the important role that professionals can play in supporting both the service user and the caregiver to renegotiate their relationships in their caring dyad, to foster independence but to build alliances with carers in a triangle of care (18). Such demands highlight the need for professionals to use their professional judgement to support recovery as they work with both the service user and the carer.

Furthermore, the importance of involving caregivers in service users' lives is increasingly recognized (1–3), which has led to the development of effective service partnership models (25, 26) to increase the influence of family members in the care of the person with mental distress. Training programmes which share authentic experiences of recovery, co-produced and co-delivered by service users and carers also play an important part in providing information and support to caregivers (33, 34). This article thus concludes the need for caregivers to hear more of service user's experiences of recovery and of being cared-for. Thus, this article seeks to contribute to this process and to support caregivers' effective involvement in the care of their family member.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to the corresponding author.

AUTHOR CONTRIBUTIONS

JF wrote and developed this article.

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Relationship Between Post-traumatic Stress Symptoms and Anticipatory Grief in Family Caregivers of Patients With Advanced Lung Cancer: The Mediation Role of Illness Uncertainty

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Objective: To explore the interrelationship between post-traumatic stress symptoms (PTSS), illness uncertainty (IU), and anticipatory grief (AG).

Methods: Structural equation modeling with bootstrapping estimation was conducted using data from a convenience sample of 254 family caregivers of patients with advanced lung cancer in China. Participants were recruited from a public cancer hospital in Shenyang, China. The family caregivers completed the Impact of Events Scale-Revised, Uncertainty in Illness Scale Family Caregiver Version, and Anticipatory Grief Scale.

Results: The measurement model has good reliability and validity, and the final model fit the data well. PTSS positively influenced AG (direct effect estimate = 0.391, $p = 0.002$). Moreover, IU was found to mediate the relationship between PTSS and AG (Indirect effects estimate = 0.168, $p = 0.005$). The mediating effect of IU accounted for up to 30.1% of the total effect.

Conclusion: IU mediated the relationship between PTSS and AG. Healthcare professionals should continuously assess PTSS, IU and AG levels in FCs and provide effective intervention options for mitigation.

Keywords: post-traumatic stress symptoms, anticipatory grief, illness uncertainty, lung cancer survivors, family caregivers

BACKGROUND

Lung cancer is the leading cause of cancer-related morbidity and mortality in China, and according to statistics, the number of newly diagnosed lung cancer cases in 2015 was 787,000 and the number of deaths was 630,500, with lung cancer deaths accounting for 27% of all cancer deaths, equivalent to one out of every four cancer-related deaths (1). The high mortality rate of lung cancer patients in China may be attributed to the lack of routine medical checkups leading to the fact that most Chinese are diagnosed with advanced lung cancer and miss the best time for treatment, thus about two-thirds of patients usually die within 1–2 years (2). Palliative care for patients with advanced lung cancer is often provided by family caregivers (FCs) because the place of death is more likely to be at home than in the hospital due to the traditional Chinese culture and the burden of medical expenses (3). The FCs definition includes any family member, friend, or partner who maintains a significant relationship with the patient and provides some care (4). However, the high mortality rate from advanced lung cancer leads FCs to not only take on heavy care duties but also to manage expectations and emotions associated with the fear of losing someone important to them, a phenomenon known as Anticipatory Grief (AG). AG is defined as the family response to the perceived threat to the other's life and the subsequent anticipation of loss in the context of the end-of-life caregiving relationship (4). A study by Nielsen showed that approximately one-third of FCs experienced AG, with up to 15% of those with severe symptoms (5). While AG is considered a natural progression when caring for a terminally ill relatives, its impacts are nonetheless debilitating for FCs who must learn to cope in the process (6).

Therefore, an increasing amount of attention has been paid to exploring the antecedents of AG, which is required to enhance FCs quality of life and helping them to improve negative emotion. Faced with the imminent loss, FCs react with anxiety, depression, concerns about the future, fear, sadness, feelings of helplessness, compassion fatigue or even Post-traumatic Stress Symptoms (PTSS) (7–11). PTSS is a delayed and persistent psychiatric disorder caused by various traumatic events or catastrophic psychological trauma. The clinical manifestation is a traumatic experience of repeated intrusions into dreams and avoidance of any scenario that might lead to traumatic memories and persistent hypervigilance (12). Previous studies have found that there may be a positive correlation between the level of PTSS experienced by family caregivers and the severity of the patient's illness, and that the level of PTSS experienced by family caregivers is more significant prior to the patient's death (11, 13).

In addition, exposure to PTSS may cause family caregivers to avoid any scenarios that may trigger traumatic memories, such as avoiding information related to the illness, may interfere with their ability to access health-related knowledge and increase their illness uncertainty (IU). IU is defined as "the inability to determine the meaning of illness-related events and accurately anticipate or predict health outcomes" (14). The study we conducted previously confirmed that IU may be an important factor affecting AG (15). IU, also identified as one of the core characteristics in the conceptual framework of AG for FCs, may cause FCs to develop into a permanent state of hypervigilance and traumatic distress toward illness signs, mainly after crisis episodes (4). However, there are few studies that have explored the relationship between the above three variables in depth, resulting in untargeted interventions for AG. Therefore, this study is the first to analyze the development mechanism of AG using a structural equation modeling approach, with the aim of providing a guiding direction for the construction of clinical intervention programs.

The present study was inspired by Stroebe's integrative risk factor framework (16) for the prediction of bereavement outcome. The framework includes various predictors of bereavement outcomes, which work together to describe and determine the sources of individual differences in adjustment to bereavement. In this study, AG as the outcome, PTSS as the loss-oriented stressor, and IU as appraisal process. The study proposed two hypotheses, hypothesis 1 that PTSS is positively related to caregivers AG and hypothesis 2 that IU mediates the effect of PTSS on caregivers AG. The hypothesized model is illustrated in **Supplementary Figure 1**.

METHOD

Study Design and Participants

This study is a descriptive cross-sectional study. A convenience sample was recruited from a public cancer hospital in Shenyang, China, between January and October 2021. Inclusion criteria were as follows: (1) FCs of patients with a clinicopathological histological or cytological diagnosis of lung cancer and TNM stage III or IV; (2) Age $\geq$ 18 years; (3) awareness of the patient's disease condition; (4) undertaking the primary care of the patient's daily life and being identified by the patient as their primary caregiver; (5) good reading and communication skills in Chinese; (6) and volunteer for this study. We excluded those who were unable to complete the questionnaire due to psychological or cognitive impairment and those who had <1 month of care. We estimated the sample size according to the requirements of the structural equation modeling (SEM), and the ratio of observed variables to sample size ranged from 1:10 to 1:15, with a sample size between 200 and 400 being appropriate (17). We distributed the survey questionnaires to 298 potential participants and received 254 complete and valid questionnaires out of the 274 possible questionnaires, giving an 92.7% overall response rate. The sample size met the requirements for SEM analysis.

Data Collection

The study team members were first trained before the start of the study, and a uniform guideline for participants to fill out the questionnaire was clarified to ensure the reliability of the questionnaire. The questionnaire was distributed by members of the study team and completed independently by FCs of patients with advanced lung cancer and consisted of four main sections: Sociodemographic Characteristics of FCs, Impact of Events Scale-Revised, Uncertainty in Illness Scale Family Caregiver Version and the Anticipatory Grief Scale. The assessors collected on the spot the completed questionnaires, and asked the participants to complete any missing options.

Measures

Sociodemographic Characteristics of FCs

A self-designed questionnaire was used to collect sociodemographic characteristics of FCs including gender, age, education, marital status, relationship with patients, and length of care.

Impact of Events Scale-Revised

PTSS was assessed using the Chinese version IES-R, which was developed by Weiss and Marmar (18) and modified by SuRan (19). It consists of 22 items and includes three dimensions: avoidance, intrusion, and hyperarousal. Each item has a score range of 0–4, with higher scores reflecting higher levels of PTSS.

The Cronbach's α was 0.89 and the split-half reliability was 0.93(19). Cronbach's α in the current sample was 0.77.

Uncertainty in Illness Scale Family Caregiver Version

Caregivers' IU was assessed using the Chinese version UIS-FC, which was developed by Mishel (20) and modified by Hongyan (21). It consists of 30 items and includes four dimensions: unpredictability, ambiguity, complexity, and lack of informativeness. These items were scored on five-point Likert scales. The total score ranges from 30 to 150, with higher scores indicating higher levels of IU. The content validity index (CVI) was 0.87 and Cronbach's α was 0.89 (21). Cronbach's α in the current sample was 0.81.

Anticipatory Grief Scale

The AG was assessed using the Chinese version AGS, which was developed by Theut (22) and modified by Dajun (23). It consists of 27 items and includes seven dimensions: sadness, feelings of loss, anger, irritability, guilt, anxiety, and ability to complete tasks. These items were scored on five-point Likert scales. Higher scores reflect higher levels of AG. The CVI was 0.96 and Cronbach's α was 0.90 (23). Cronbach's α in the current sample was 0.91.

Statistical Analysis

The IBM SPSS Statistics 26.0 (IBM Corp., USA) was used for data analysis. The sociodemographic characteristics of the participants were examined by computing frequencies and percentages. One-way ANOVAs and t tests were used to determine the relationship between FCs' characteristics and the three variables, and Pearson correlations were used to test for unadjusted associations between variables. Statistical significance was set at 0.05. The hypothesized model was tested using SEM with IBM SPSS AMOS version 26.0 (IBM Corp., USA). The maximum-likelihood estimation of the entire system in a hypothesized model, and enables the assessment of variables with the data (24). In our analysis, the measurement model was confirmed using confirmatory factor analysis (CFA), and then we performed SEM analysis to measure the fit and path coefficients of the structural model. The model fit indices were as follows: Chi-square (χ^2), degrees of freedom (df), value of χ^2/df, goodness-of-fit index (GFI), adjusted GFI (AGFI), comparative fit index (CFI), Tucker-Lewis index (TLI), root mean square error of approximation (RMSEA), and standardized root mean square residual (SRMR). The recommended value for GFI, AGFI, CFI, and TLI is 0.90 or higher. The RMSEA would be "close to" 0.09 or lower, SRMR would be "close to" 0.05 or lower, and χ^2/df would be "close to" 5.00 or lower, indicating a good model fit (25). Finally, we used the bootstrap test to measure the direct, indirect and total effects of the structural model (26).

Ethical Considerations

The study was approved by the Ethics Committee of Liaoning Cancer Hospital and Institute. Based on the Declaration of Helsinki, participants had the right to leave the study at any time. Written informed consent was obtained from all participants.

RESULTS

Sociodemographic Characteristics

A total of 254 FCs of patients with advanced lung cancer participated in this study. The majority were females (63.4%), age 36–59 years (58.7%), high school (53.1%), married (85.0%), parents of the patients (52.4%), and length of care <6 months (58.7%).

PTSS, IU and AG, According to Sample Characteristics

Female FCs reported significantly higher IES-R, UIS-FC, and AGS scores than male ($P < 0.05$ or 0.001). FCs aged ≤35 years reported significantly higher IES-R and UIS-FC scores than those aged 36–60 years and ≥60 years ($P < 0.05$). FCs with a bachelor's degree and above reported higher IES-R scores than those with high school education, primary school education and below ($P < 0.05$). FCs who were married reported lower IES-R, UIS-FC, and AGS scores than other ($P < 0.05$ or<0.01). Regarding relationships with patients, spouses reported lower IES-R, UIS-FC, and AGS scores than compared with parents and children ($P < 0.05$, 0.01 or 0.001). There were significant differences in the AGS scores for FCs with different lengths of care ($P < 0.05$) (**Supplementary Table 1**).

The Interrelationships Between PTSS, UI and AG

AG were significantly and positively correlated with PTSS. The Pearson's correlation coefficients ranged from 0.25 to 0.62 ($P < 0.01$). Additionally, AG were significantly correlated with UI. The Pearson's correlation coefficients ranged from 0.13 to 0.54 ($P < 0.01$ or 0.05) (**Table 1**). The PTSS were significantly and positively associated with UI. The correlation coefficients ranged from 0.20 to 0.52 ($P < 0.01$).

Reliability and Validity of the Measurement Model

To measure the internal consistency reliability, convergent validity and discriminant validity of the constructs in our hypothetical model, we performed CFA on the three constructs of PTSS, IU, and AG (**Table 2**). The results indicated that the composite reliability (C.R.) of each construct ranged from 0.765 to 0.910, exceeding the C.R. threshold value of 0.60 (27), and providing evidence of internal consistency reliability. In addition, the standardized factor loadings of the individual dimensions in the model were between 0.672 and 0.805, exceeding threshold value of 0.50 (27), and reached significant (all $P < 0.001$), giving preliminary evidence for the convergent validity of the measurement model. Meanwhile, the average variance extracted (AVE) of all constructs ranged from 0.517 to 0.592, exceeding the AVE threshold value of 0.50 (27), and thus the convergent validity was acceptable. Moreover, the estimated intercorrelations among all constructs were less than the square roots of the AVE in each construct, and this provided support for discriminant validity (28) (**Table 3**).

As with all self-reported data there is a potential for common method variance (CMV) resulting from multiple sources (29).

TABLE 1 | The interrelationships between PTSS, IU and AG (r).

AGS scores	IES-R scores				UIS-FC scores				
	Total	AVO	INT	HYP	Total	UNP	AMB	COM	LAI
Total	0.56**	0.62**	0.38**	0.38**	0.54**	0.52**	0.45**	0.31**	0.50**
SAD	0.49**	0.52**	0.33**	0.35**	0.47**	0.44**	0.41**	0.26**	0.41**
FOL	0.46**	0.49**	0.33**	0.31**	0.45**	0.43**	0.38**	0.28**	0.36**
ANG	0.46**	0.50**	0.34**	0.28**	0.45**	0.42**	0.37**	0.28**	0.43**
IRR	0.44**	0.50**	0.26**	0.32**	0.45**	0.45**	0.38**	0.27**	0.43**
GUI	0.45**	0.51**	0.31**	0.30**	0.47**	0.45**	0.41**	0.25**	0.41**
ANX	0.45**	0.51**	0.28**	0.33**	0.43**	0.42**	0.34**	0.23**	0.42**
ACT	0.40**	0.38**	0.26**	0.25**	0.31**	0.31**	0.26**	0.13*	0.35**

**P < 0.05, **P < 0.01. PTSS, Post-traumatic Stress Symptoms; IU, Illness Uncertainty; AG, Anticipatory Grief; IES-R, Impact of Events Scale-Revised; UIS-FC, Uncertainty in Illness Scale Family Caregiver Version; AGS, Anticipatory Grief Scale; AVO, Avoidance; INT, Intrusion; HYP, Hyperarousal; UNP, Unpredictability; AMB, Ambiguity; COM, Complexity; LAI, Lack of Informativeness; SAD, Sadness; FOL, Feelings of Loss; ANG, Anger; IRR, Irritability; GUI, Guilt; ANX, Anxiety; ACT, Ability To Complete Tasks.*

TABLE 2 | CFA for the measurement model.

Construct	Variable	Unstandardized factor loadings	S.E.	t-value	P	Standardized factor loadings	SMC	C.R.	AVE
PTSS	AVO	1.000				0.672	0.452	0.765	0.522
	INT	1.134	0.135	8.368	***	0.747	0.558		
	HYP	0.982	0.117	8.370	***	0.745	0.555		
IU	UNP	1.000				0.712	0.507	0.811	0.517
	AMB	2.991	0.311	9.607	***	0.723	0.523		
	COM	1.241	0.134	9.258	***	0.688	0.473		
	LAI	1.181	0.120	9.845	***	0.752	0.566		
AG	SAD	1.000				0.730	0.533	0.910	0.592
	FOL	1.311	0.115	11.425	***	0.736	0.542		
	ANG	0.956	0.076	12.495	***	0.803	0.645		
	IRR	0.937	0.075	12.526	***	0.805	0.648		
	GUI	0.899	0.077	11.705	***	0.754	0.569		
	ANX	1.039	0.085	12.276	***	0.789	0.623		
	ACT	0.785	0.066	11.849	***	0.763	0.582		

****P < 0.001. CFA, Confirmatory factor analysis; S.E, Standard errors; SMC, Squared Multiple Correlations; C.R., Composite reliability;AVE, Average variance extracted; PTSS, Post-traumatic Stress Symptoms; IU, Illness Uncertainty; AG, Anticipatory Grief; AVO, Avoidance; INT, Intrusion; HYP, Hyperarousal; UNP, Unpredictability; AMB, Ambiguity; COM, Complexity; LAI, Lack of Informativeness; SAD, Sadness; FOL, Feelings of Loss; ANG, Anger; IRR, Irritability; GUI, Guilt; ANX, Anxiety; ACT, Ability To Complete Tasks.*

Therefore, we performed statistical analyses to assess the severity of CMV. First, a Harmon one-factor test was conducted on the 14 crucial variables in our hypothetical model (30). Following Jukka, the first factor tends to explain over half of the variance indicating the presence of CMV (31). Results illustrated that 14 factors are present and the most covariance explained by one factor is 43.67 percent, showing that CMV are not a likely contaminant of our results. Second, we included in the model 1 a common method factor whose indicators included all the principal constructs' indicators. Subsequently, we interconnected the CFA constructs into fully correlated constructs in model 2. If method variance is largely responsible for the covariation among the measures, df and χ^2 values of difference between model 1 and model 2 should indicate that there was no significant (30, 32). Given the difference between two models did demonstrate the significance, we contend that the CMV is unlikely to be a serious concern for this study. ($\Delta\chi^2 = 221.065$, $\Delta df = 3$, $P < 0.001$).

TABLE 3 | Discriminant validity analysis for the measurement model.

	AVE	PTSS	IU	AG
PTSS	0.522	**0.722**		
IU	0.517	0.684	**0.719**	
AG	0.592	0.720	0.661	**0.769**

AVE, Average variance extracted; PTSS, Post-traumatic Stress Symptoms; IU, Illness Uncertainty; AG, Anticipatory Grief. The bold values indicate the square root of AVE for discriminant validity.

Test of the Structural Model

The structural modeling results showed that the hypothesized model fit the data well ($\chi^2 = 124.507$, df $= 74$, $\chi^2/df = 1.683$, GFI $= 0.937$, AGFI $= 0.910$, CFI $= 0.971$, TLI $= 0.964$, RMSEA $= 0.052$, SRMR $= 0.047$). We use a causal step strategy to investigate

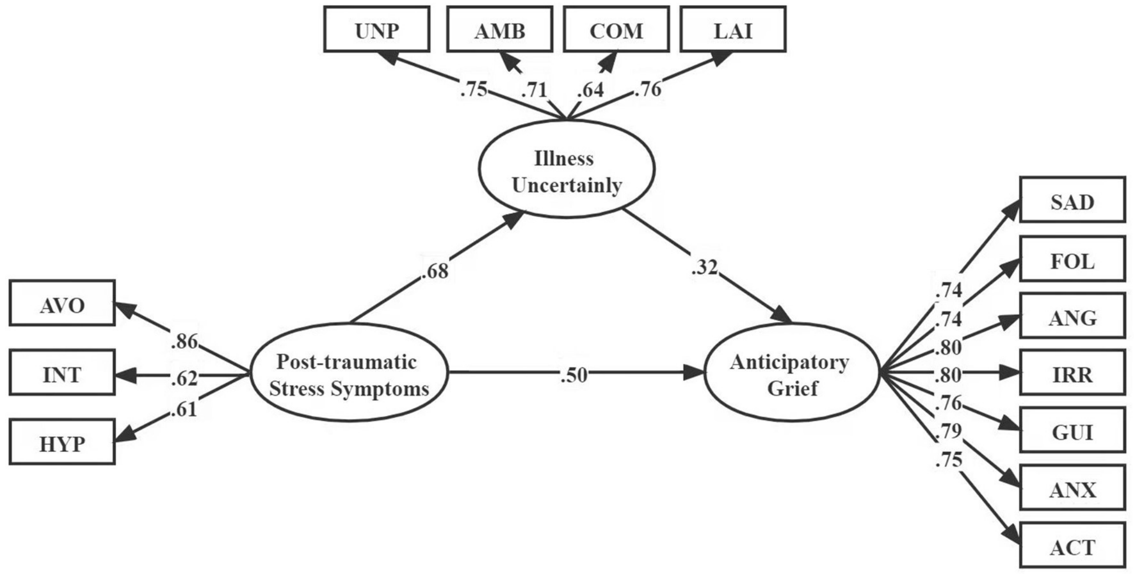

FIGURE 1 | Structural equation modeling of the hypothesized model. AVO, Avoidance; NT, Intrusion; HYP, Hyperarousal; UNP, Unpredictability; AMB, Ambiguity; COM, Complexity; LAI, Lack of Informativeness; SAD, Sadness; FOL, Feelings of Loss; ANG, Anger; IRR, Irritability; GUI, Guilt; ANX, Anxiety; ACT, Ability To Complete Tasks.

TABLE 4 | Direct, indirect, and total effects of the hypothesized model.

	Point estimate	**Product of coefficients**		**Bootstrapping**				
				Bias-Corrected 95% CI		**Percentile 95% CI**		**Two-tailed significance**
		SE	**Z**	**Lower**	**Upper**	**Lower**	**Upper**	
Direct effects								
PTSS→ AG	0.391	0.081	4.827	0.256	0.570	0.249	0.567	0.002**
Indirect effects								
PTSS→ AG	0.168	0.051	3.294	0.071	0.273	0.064	0.262	0.005**
Total effects								
PTSS→ AG	0.560	0.063	8.889	0.437	0.686	0.439	0.688	0.002**

*Estimating of 1,000 bootstrap sample, **P <0.01. PTSS, Post-traumatic Stress Symptoms; AG, Anticipatory Grief.*

the first mediation condition with respect to hypothesis 1 (33). As shown in **Table 1**, the correlation coefficients indicated that PTSS was significantly and positively associated with AG ($r = 0.56$, $P < 0.01$). In addition, the results of the direct effect of PTSS on AG (standardized direct effect $= 0.76$, $P < 0.001$) was statistically significant. Therefore, hypothesis 1 was supported.

To test hypothesis 2, we measured the second condition of mediation. The correlation coefficients indicated that PTSS was significantly and positively associated with IU ($r = 0.48$, $P < 0.01$), IU was significantly and positively associated with AG ($r = 0.54$, $P < 0.01$). In addition, the results of the direct effects of PTSS on IU (standardized direct effect $= 0.50$, $P < 0.001$), and the direct effect of IU on AG (standardized direct effect $= 0.32$, $P < 0.001$), were all statistically significant (**Figure 1**). To examine the indirect effects of the dependent variable through the mediator, we performed bias-corrected percentile bootstrapping and percentile bootstrapping at a 95% confidence interval with 1,000 bootstrap samples (**Table 4**) (34). We calculated the confidence interval of the lower and upper bounds to examine of whether the indirect effects were significant (28). The result of the bootstrap test confirmed the existence of a significant and positive mediating effect for IU between PTSS and AG (indirect effect $= 0.17$, $P < 0.01$). Hypotheses 2 was thus supported.

DISCUSSION

FCs assume the primary responsibility for the care of patients with advanced cancer in China. However, due to the irreversible nature of advanced cancer patients' disease, FCs may experience varying degrees of AG when faced with the imminent death of the patient (15). AG not only affects the FCs 's ability to assess the patient's care needs, but also leads to a decrease in the quality of palliative care. In addition, experiencing AG may also have a negative impact on the FCs 's mental state, predisposing them to negative emotions such as anger, fear, and guilt and self-blame (6). Nevertheless, there is still limited evidence of study on FCs with advanced cancer experiencing AG. Therefore, in view of the potentially serious adverse outcomes for FCs with AG, a more in-depth exploration of the mechanisms underlying the development of AG is necessary to inform the precise implementation of clinical interventions.

The present study found significant differences in IES-R, UIS-FC and AGS when PTSS, IU and AG were examined according to sociodemographic characteristics. Culturally, women are the primary source of caregiving. Meanwhile, female FCs have been found to be more likely to use emotion-focused coping strategies. Therefore, it was not amazing to find that women reported significantly greater PTSS, IU and AG than men. In this study, FCs aged <35 years reported higher levels of PTSS and IU. Similarly, Elisavet et al. found that younger age was significantly associated with higher PTSS (35). Heleen et al. reported that the age of mother caregiver of children with cancer was negatively associated with uncertainty (36). Young FCs may face the stress of work and economic and social challenges along with their caregiving responsibilities. When work-life balance is difficult it can lead to physical and psychological symptoms, which may be one reason why younger FCs may have higher levels of PTSS and IU, while older homemakers have more positive emotional responses. In terms of education, FCs with at least a bachelor's degree reported higher levels of PTSS. One possible explanation is that FCs with higher education possess more but not deeper information and knowledge about cancer treatment and nursing and can be highly alert to any subtle symptoms or even normal reactions of the patients, which may increase their PTSS. However, there was no significant difference in education level in terms of IU and AG. In addition, our results show that married FCs reported lower levels of PTSS, IU, and AG because they are more likely to have partner support. In terms of the relationship with the patient, the patient's spouse had lower levels of PTSS, IU and AG than the patient's children and parents. This is inconsistent with the results of previous study and the reason may be the influence of blood relationship (37). In China, there is a traditional belief that couples are not blood relationship, but are legal relationship. In fact, many couples are not selfless; they look more to their own interests and do not think to take on problems together when they come up. Furthermore, FCs with <6 months of care had higher AG levels. Burton et al. study showed that caregivers who spent fewer months caring for the patient before the bereavement would increase the level of grief (38). FCs in the short term do not cope well with the fact of patient's illness and are not adapted to the life of intense care. AG level can therefore exacerbate.

We found a possible causal relationship between PTSS, IU and AG and FCs in 254 patients with advanced lung cancer based on the theoretical basis and SEM analysis. The results demonstrate PTSS had a direct effect on AG. Meanwhile, IU was shown to have a mediation effect in FCs' PTSS and AG. In a full mediation model, the direct effect of PTSS would have become insignificant when the role of IU was added to the model. The model illustrates only partial mediation as the path between PTSS and AG remains significant in the full model. Overall, the results explain well the hypotheses proposed in this study.

The first aim of this study was to examine the associations between PTSS and AG in FCs of patients with advanced lung cancer. Consistent with previous studies (11), the present study showed that these associations are positive and statistically significant. Advanced lung cancer is a traumatic event for FCs, who can develop a variety of post-traumatic symptoms due to the changes in the cancer and the patient's uncomfortable response. As patients require frequent hospitalization, FCs are constantly in trauma-related scenarios while caring for the patient, and they unconsciously repeat all the information about the cancer and exhibit intrusive thinking. Some FCs will avoid cancer-related topics, not wanting to face the deterioration of the patient's cancer and displaying fear for the patient after death. FCs become hyperarousal to any subtle, even normal changes and reactions in the patient's body. These post-traumatic symptoms can lead FCs to perceive that they may lose a loved one to cancer and develop AG, or even psychological illness and suicidal tendencies. Therefore, to reduce the level of AG in FCs, this can be achieved by reducing the level of PTSS and reducing the intrusion, avoidance and hyperarousal of stressful events. It is suggested that healthcare professionals can use a variety of psychotherapeutic interventions to minimize the adverse effects of intrusive thinking and to help FCs to properly deal with the deterioration of the patient's illness and death.

The second aim was to determine to what extent IU mediates the association between PTSS and AG. The significance of the finding is that intervening in IU is an important strategy to alleviate AG. The present study showed that IU scores of FCs of patients with advanced lung cancer was positively correlated with PTSS and AG scores. FCs often have the stressful event of the patient's cancer in their minds and are reluctant to face reality and avoid any cancer-related information, which may lead to a lack of information for FCs and further interfere with their ability to acquire health-related knowledge. Due to the lack of information and ability, FCs do not predict the course of cancer, coupled with the various complex treatments, which these uncertainty factors often overwhelm FCs and thus deepen IU. IU can further increase the physiological and psychological stress of FCs, reducing their confidence in coping with and overcoming traumatic events. Moreover, FCs are at risk of losing a loved one, which makes the experience of caring for a palliative care patient unique and complex, ultimately leading to AG for FCs. Therefore, AG levels can be mitigated by reducing the IU of FCs. It is recommended that healthcare professionals should provide more care and communication to FCs, disseminate cancer-related knowledge, and satisfy FCs' information needs. It is also necessary to strengthen FCs' mental health education, assist them in establishing a correct concept of death, understand that death is an inevitable law of nature, and help them cope with the pain and grief of the impending loss of a close relative.

There are still some limitations in this study. First, the participants in this study were all from the same hospital and a convenience sampling method was used, thus limiting generalization due to potential selection bias. In addition, this study is a preliminary cross-sectional study does not enable causal inferences to be made yet. Although we used SEM to conduct a simultaneous testing of our proposed model in FCs of advanced lung cancer patients, the results still need to be treated with caution. It is desirable that future longitudinal studies will be conducted to further clarify the causal relationship between them.

CONCLUSION

FCs need practical and emotional support to deal with the mental trauma they endure while providing palliative care. FCs of advanced lung cancer patients may experience reduced quality of care due to AG, and interventions for FCs from the perspective of their PTSS and IU are recommended.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Liaoning Cancer Hospital and Institute. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

DS, ZM, XZ, and LZ: material preparation, data collection, and analysis were performed. DS and ZM: the first draft of the manuscript was written. JL commented on previous versions of the manuscript. All authors contributed to the study conception, design, read, and approved the final manuscript.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2022.914862/full#supplementary-material

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Psychological Support for Family Caregivers of Patients With Amyotrophic Lateral Sclerosis at the Time of the Coronavirus Disease 2019 Pandemic: A Pilot Study Using a Telemedicine Approach

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The coronavirus disease 2019 (COVID-19) pandemic confined most of the population to homes worldwide, and then, a lot of amyotrophic lateral sclerosis (ALS) centers moved to telemedicine services to continue to assist both patients with ALS and their caregivers. This pilot, randomized, controlled study aimed to explore the potential role of psychological support interventions for family caregivers of patients with ALS through resilience-oriented sessions of group therapy during the COVID-19 pandemic. In total, 12 caregivers agreed to be remotely monitored by our center since March 2020 and underwent scales for global burden (i.e., Caregiver Burden Inventory, CBI), resilience (i.e., Connor Davidson Resilience Scale, CD-RISC), and perceived stress (i.e., Perceived Stress Scale, PSS) at two-time points (i.e., at pre-treatment assessment and after 9 months or at post-treatment assessment). They were randomized into two groups: the former group underwent resilience-oriented sessions of group therapy two times a month for 3 months, while the latter one was only remotely monitored. No significant differences were found in CBI, CD-RISC, and PSS during the 9-month observation period in the treated group compared with the control group, suggesting a trend toward stability of caregiver burden together with resilience and perceived stress scores in all the subjects monitored. The lack of differences in caregivers' burden, resilience, and perceived stress scores by comparing the two groups monitored during 9 months could be due to the co-occurrence of the COVID-19 pandemic with the stressful events related to caring for patients with ALS that might have hindered the detection of significant benefits from short-lasting psychological support.

Keywords: amyotrophic lateral sclerosis, caregivers, COVID-19, telemedicine, psychological support

INTRODUCTION

Amyotrophic lateral sclerosis (ALS) is a fatal neurodegenerative and rare disease with an incidence of 1.59 per 100,000 person-years (1) that affects the motor neurons and causes progressive physical, respiratory, and swallowing impairments (2). Moreover, up to 50% of patients with ALS may develop cognitive and behavioral impairment during the disease (2) together with a high risk of severe mental disorders, which affects their function, quality of life, and mobility (3). Furthermore, the coronavirus disease 2019 (COVID-19) pandemic has affected the psychological and physical health of patients with ALS and their caregivers, leading to an increased need for assistance (4).

The patients with ALS gradually lose muscle function, thereby needing increasing care during the disease course, which mainly hurts their caregivers (5, 6). Generally, a family member (i.e., partners or sons/daughters of patients with ALS), who frequently has no previous experience in this role, may assume the role of "informal" or "family caregiver" (7). Over the past two decades, the role of family caregivers and the integration of care for patients and their families have been increasingly investigated (4, 6, 8, 9). Caregivers are crucial figures in care provision, offering emotional and physical support to the assisted patients and playing an essential role in clinical decision-making in the ALS treatment (10). On the other hand, caregivers often struggle with accepting this fatal disease, their increased responsibilities, concerns about the future, and feelings of guilt (11). Some longitudinal studies indicated that the increasing levels of motor impairment together with cognitive and behavioral deficits during the ALS progression might significantly influence caregivers' burden (12, 13). Caregiver burden represents the impact on the emotional and physical health, the social life, and the financial status of the caregivers because of adopting the caregiving role (14). As patients are more dependent on their caregivers, this, in turn, aggravates caregivers' negative emotions, such as anxiety. Considering the close relationship between the psychological well-being of the caregivers and the disease progression of the patients with ALS, it is crucial to monitor and treat the caregivers' psychological status and their care burden (15). To counteract the effects of high psychological and care burdens, caregivers of patients with ALS may increase their resilience, which represents the ability to execute active/positive coping strategies in a complex scenario of different states of mind, such as those resulting from carrying out the caregiving role (16).

During the entire disease course, caregivers of patients with ALS could need to acquire management skills for supporting patients in executing cough assistance and using home ventilators, and/or promoting nutritional interventions and enteral feeding (17, 18). Therefore, patients with ALS need to have support from informal caregivers (10). However, these new duties associated with caregiving, as well as the condition of their loved ones, may have a great impact on the caregivers' quality of life, and it reflects the importance of psychological support in the management of their condition (19). Understanding the factors associated with the caregivers can lead to more tailored support for them. The types of support could be financial, psychological, and educational relating to the condition or related to the patient's care and supports (e.g., equipment, therapists, access to services, and respite care) (20).

As the disease's relentless deterioration progresses, telemedicine is a valid instrument to provide care to patients with ALS and support their caregivers remotely (21). Additionally, telemedicine has become the way to deliver care and reduce the risk of more dysfunction in the current COVID-19 pandemic (22). Telemedicine aided in preserving patients' access to clinical care and medical expertise during the COVID-19 pandemic, allowing healthcare professionals to follow-up on patients in remote locations (23, 24). Moreover, telemedicine might be a well-suited instrument for the ongoing management of such patients, particularly during a time when social distancing is encouraged (24). In particular, psychological support through telemedicine has been implemented during the COVID-19 pandemic to reduce the intensity of burden, distress, and loneliness experienced by caregivers of patients with ALS (4, 6, 22–25). Whereas extensive research has been conducted on the psychosocial aspects of caregivers of patients with ALS (2, 4, 6, 8–13, 26, 27), only a few investigations of psychological support interventions for caregivers of patients with ALS through video consults have been conducted in an ALS population, primarily in Europe, showing differences across healthcare systems, social services, and family culture (7, 22–25). In Ireland, Burke et al. (6) performed a randomized controlled trial, comparing two intervention groups, which underwent, respectively, mindfulness-based stress reduction (MBSR), used to promote the ability to cope with the management of negative emotions, and cognitive behavioral therapy (CBT), used to treat anxiety and depression, to a control group (i.e., an untreated group from a database of 75 caregivers of patients with ALS). In Italy, during the COVID-19 pandemic, the differences in social/healthcare services and approaches were more evident also in managing remotely patients with ALS and their caregivers (22–24). In Southern Italy, Capozzo et al. (22) reported the experience of a referral ALS centre by performing telephonic calls for monitoring patients with ALS, while video consults were refused due to poor practice in digital technologies for both patients and caregivers. Differently, in Northern Italy, De Marchi et al. (23) used video-calling for monitoring remotely patients with ALS and supporting their caregivers, as the approach *via* tele-consults was received as talking face-to-face to healthcare professionals. Moreover, multidisciplinary visits were provided through an online platform [IoMT Connected Care Platform (Ticuro Reply)]. Vasta et al. (24) used a mixed approach (i.e., both video and phone calls) to perform 139 neurological or psychological tele-visits, reporting substantial satisfaction with telemedicine approaches, although the majority would have preferred in-person visits.

In the present pilot study, we aimed to explore the effect of psychological support on reducing the burden and increasing the resilience of family caregivers of patients with ALS during the COVID-19 pandemic through video consults and resilience-oriented sessions of group therapy. We expected to reveal potential differences between the two groups in terms of reduction of global burden and perceived stress and an increase in resilience in the treated group compared with the untreated one.

MATERIALS AND METHODS

In total, 12 consecutive caregivers of patients with ALS (one each) were recruited at the First Division of Neurology of “Luigi Vanvitelli” University (Naples, Italy). The inclusion criteria were as follows: age > 18 years; being a family caregiver of a person with a diagnosis of definite or (clinically or laboratory-supported) probable ALS according to the Revised El Escorial Criteria (28); spending at least 4 h per day with the patient (10); and unimpaired cognitive performances. Caregivers with communication and hearing problems, and/or inability to comply with the study commitments were excluded. The caregiver sample was matched by the age and education level of patients with ALS. In this randomized, controlled pilot study, six consenting caregivers were randomly assigned to the treatment group (TG) and six to the control group (CG). The caregivers belonging to TG underwent regular (monthly) individual video-consults and (2 times/month) resilience-oriented sessions of group therapy in March 2020 for 3 months, immediately preceded by the administration of clinical scales at baseline. The same scales were repeated 6 months after the end of psychological support (long-term assessment). The CG group, monitored by remote phone calls every 2 months (as routinely performed in all caregivers), only completed the scales at the same time points (**Figure 1**). This pilot study lasted 9 months and was conducted according to the Declaration of Helsinki; informed consent was acquired from each participant by e-mail. The project was approved by the Institutional Review Board and the Ethics Committee of the University of Campania “L. Vanvitelli” (Naples, Italy).

Description of Psychological Support

The individual tele-consults and resilience-oriented sessions of group therapy were conducted in March 2020. This treatment consisted of three individual tele-visits per participant, each lasting about 60 min, once a month for three consecutive months, combined with sessions of group therapy (2 times/month for three consecutive months), each lasting about 60 min, according to the guidelines of “counseling” formulated by the American Psychological Association (29). We adopted a model of individual, and group counseling and psychotherapy. The tele-consults/group sessions were held in a comfortable environment, by just one licensed psychologist/psychotherapist with robust expertise in ALS and cognitive disorders (DB). The primary goals were to provide non-directive support for caregivers through empathic/reflective listening and open-ended questioning. The tele-consults and group video-coaching meetings, aimed at reducing caregivers’ burden and loneliness and increasing their resilience, were focused on the physical, cognitive, and behavioral functioning and daily routines of patients; on the perceived quality of the relationship between patient and caregiver; on emotional, physical, and social burden perceived by caregivers; and on significant needs. Moreover, individual tele-consults included semi-structured qualitative interviews aimed at exploring emotions and stress perception as well as satisfaction regarding the offered telemedicine support. An interview topic guide (as shown in **Supplementary Material**) was used, with themes constructed through the clinical experience of research team members and literature reviews. The process for analyzing and interpreting the interviews included a thematic analysis reviewing the data for contents (in the form of transcripts, or detailed notes) (30). Thematic analysis was iterative and ongoing throughout the study (31). Interview transcripts were read in full by DB, CP, and MS and, then, were coded thematically by DB who developed a preliminary coding scheme with overarching themes and subthemes. In discussion with all researchers, a final coding framework was refined.

Clinical Assessment

The following scales were remotely administered (in the Italian language) to all caregivers by the same licensed psychologist (CP):

- Caregiver burden inventory (CBI) (32): a 24-item multi-dimensional questionnaire measuring caregiver burden with five subscales, namely, “time dependence,” “developmental,” “physical,” “social,” and “emotional burden.” The score for each item is evaluated using a five-point Likert scale, ranging from 0 (not at all disruptive) to 4 (very disruptive), and all scores are summed; higher scores correspond to a higher burden. Cronbach’s alpha values for each subscale range between 0.73 and 0.85, and test-retest reliability is 0.94 (33).
- Connor Davidson resilience scale (CD-RISC) (34): consists of 25 statements that respondents rated on a 5-point scale from “strongly disagree” to “strongly agree.” Answers were scored from 0 to 4 to create a total score that ranged from 0 to 100, with higher numbers denoting greater resilience. Cronbach’s alpha value is 0.89, and test-retest reliability is 0.87.
- Perceived stress scale (PSS) (35): this scale assesses perceived stressful experiences or stress responses over the previous month with a 5-point Likert scale (0 = never and 4 = very often). PSS-10 scores are obtained by reversing the responses (e.g., 0 = 4, 1 = 3, 2 = 2, 3 = 1, and 4 = 0) to the four positively stated items (items 4, 5, 7, and 8) and then summing across all scale items. The scores range from 0 to 40, with higher scores indicating greater stress. Cronbach’s alpha value is 0.74, and test-retest reliability is 0.85 (36).

Patients with ALS were assessed by a disability score (i.e., ALS Functional Rating Scale-revised, ALSFRS-R total score, where lower total reflects higher disability) (37) and a global cognitive functioning score (i.e., Edinburgh Cognitive and Behavioral ALS Screen, ECAS) (38).

Statistical Analysis

At the pre-treatment assessment, the study groups on demographics and clinical measures of the cared-for patients were compared *via* independent *t*-test or Pearson’s chi-squared test (χ^2 test) when appropriate. Moreover, the pre-treatment differences in CD-RISC, CBI, and PSS were explored *via* an independent *t*-test. In line with O’Connell et al. (39), to test the effects of psychological support on CD-RISC, CBI, and PSS,

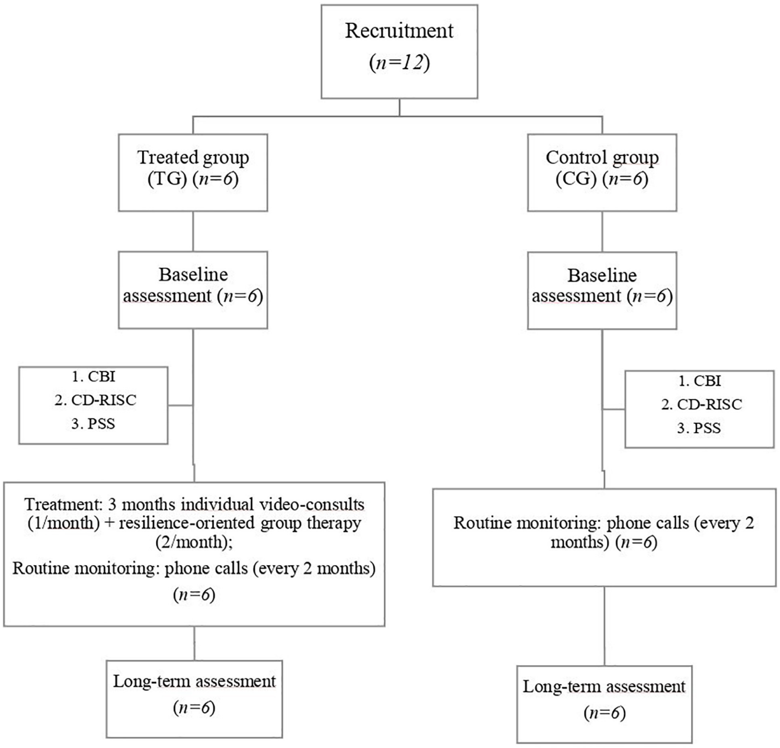

FIGURE 1 | A flowchart of the recruited and monitored caregivers.

the two study groups were compared on the post-treatment-assessment measures by analysis of covariance (ANCOVA), using the pre-treatment measures as covariates. When we carried out multiple comparisons between treated and untreated study groups, the Bonferroni correction to the alpha level was applied to avoid Type I error. All analyses were performed using the IBM Statistical Package for Social Science (SPSS) version 20.

RESULTS

At the pre-treatment assessment, the two groups of patients with ALS associated with the two studied groups did not differ in demographics or clinical measures (i.e., ALSFRS-R, *F*-statistic = 0.60, *p*-value = 0.556 and ECAS scores, *F-statistic* = –0.08, *p*-value = 0.937; **Table 1**). Moreover, no statistically significant differences were found in pre-treatment measures of the CD-RISC (*F*-statistic = –0.27, *p*-value = 0.792), CBI (*F*-statistic = –1.77, *p*-value = 0.103), and PSS (*F*-statistic = 1.61, *p*-value = 0.134; **Table 2** and **Figure 2**).

At the post-treatment assessment, CG and TG did not differ on CD-RISC (*F*-statistic = 0.25, *p*-value = 0.629), CBI (*F*-statistic = 0.29, *p*-value = 0.601), and PSS (*F*-statistic = 0.02, *p*-value = 0.888; **Table 2** and **Figure 2**). The treated caregivers represented 83% of spouses (four women; mean age 60.3 + 8.9 years), who accepted to share their most intimate, sensitive, and vulnerable parts. Through our data analysis, three overarching themes were generated: (1) activities of a caregiver of a patient with ALS; (2) changing dynamics of care and connectedness among family and friends; and (3) satisfaction regarding the health services offered by our center. Regarding themes 1–2, semi-structured qualitative interviews, performed during individual tele-visits, revealed that caregivers dealt with uncertainty, unpredictability, helplessness, and frustration and found themselves lacking even those few, but indispensable, social resources that make the difference in everyday life. Regarding theme 3, all the included caregivers declared that they were satisfied with the services of our center during the COVID-19 pandemic, although they would have preferred integration with in-person visits.

DISCUSSION

This pilot study aimed at exploring if psychological support by telemedicine services for ALS caregivers during the COVID-19 pandemic was useful to reduce caregivers' burden and their perceived stress by improving caregivers' resilience to cope with the disease condition. However, our study showed that short-lasting psychological support by telemedicine during a pandemic was not enough to help more ALS caregivers in that no significant differences were found between TG and CG regarding changes in global burden, resilience, and perceived stress scores across time. To note, caregivers' burden and perceived stress scores did not increase across 9 months in either monitored group, as well as resilience measures.

TABLE 1 | Between-group comparison at pre-treatment assessment; data are reported as mean ± standard deviation (SD) or count (percentage).

Variable	Untreated (CG)	Treated (TG)	χ^2 test[a]; *t*-test[b]	*p*-value[c]
Caregivers' sex (male)	3 (42.9%)	2 (33.3%)	0.12[a]	0.797
Caregivers' age at interview, years	53.29 ± 10.48	60.33 ± 8.98	−1.28[b]	0.224
Caregivers' years of education	12.00 ± 2.94	11.83 ± 3.18	0.09[b]	0.924
Relationship with patient:			0.25[a]	0.612
Husband/wife	5 (71.4%)	5 (83.3%)		
Son/daughter	2 (28.6%)	1 (16.7%)		
Patients' sex (male)	3 (42.9%)	3 (50.0%)	0.06[a]	0.797
Patients' age at assessment, years	57.50 ± 6.02	60.00 ± 8.94	−0.56[b]	0.583
Patients' years of education	8.60 ± 4.56	8.50 ± 2.58	0.04[b]	0.964
Age at onset, years	54.17 ± 11.75	56.67 ± 10.03	−0.39[b]	0.700
Duration of disease, months	52.50 ± 75.49	41.83 ± 23.08	0.33[b]	0.747
ALSFRS-R	24.71 ± 13.53	21.00 ± 6.81	0.60[b]	0.556[a]
ECAS-CS	92.67 ± 18.82	93.60 ± 13.50	−0.08[b]	0.937

ALSFRS-R, ALS Functional Rating Scale-Revised; CG, control group; ECAS-CS, Edinburgh Cognitive and Behavioral ALS Screen-Cognitive Score; TG, Treated Group.
[a]Marks the results of the "χ^2 test" and [b]of the "t-test". [c]Bonferroni corrected alpha level of 0.05/11 = 0.005.

In agreement with our findings, de Wit and colleagues revealed that tailored support programs for caregivers of patients with ALS and progressive muscular atrophy (PMA) (i.e., a blended intervention through face-to-face contact and e-health, based on Acceptance and Commitment Therapy or ACT) did not reduce their distress, but may be beneficial by increasing the feeling of control in caregiving situations (40). The participants positively evaluated this protocol: caregivers referred that it helped them to be more aware of their situation and to perceive more control over it, empowering caregivers to make choices according to their own needs. The online approach was also appreciated: caregivers may experience a lack of personal time, since they spend many hours providing care, especially in the advanced stages of ALS. Using the online support enabled them to enter the program at their preferred time and place. Additionally, Tang et al. (41), carrying out a face-to-face interview in 120 pairs of patients with ALS and their caregivers, revealed that higher anxiety index scores were associated with greater caregiver burden, as well as previously demonstrated also regarding the association between depression and caregiving burden in ALS (42). These findings altogether suggest that the level of disease knowledge, anxiety, and depression may be associated with caregiver burden, indicating the need for support programs to alleviate this burden.

Regarding the impact on caregivers' anxiety/depression and burden of the COVID-19 pandemic, our findings did not reveal a significant increase in perceived stress and burden in either monitored groups, despite the imposition of national quarantine and other social restrictions that have induced most caregivers to perceive more loneliness (8) and a worsening of homecare assistance (19). Although the 3-month psychological support protocol reserved for TG did not show significant benefits in caregiver burden and distress, the remote bi-monthly phone calls that we targeted to all patients and caregivers (including those from the studied TG and CG) may have reduced perceived loneliness and subsequent distress. Possible interpretations of this negative result could be related to the small sample size, the relatively short time of the psychological support, and the intrinsic difficulties of implementing effective psychological support for patients with ALS and their caregivers due to the rapid and critical progression of the disease and the severe care needs. In fact, in our sample, three patients assisted by caregivers belonging to TG died due to disease progression during the 3-month treatment, as did one patient assisted by a caregiver belonging to the CG, although the two studied groups were matched for patients' disease duration, disability (i.e., ALSFRS-R score) and cognitive performance (i.e., ECAS score) (**Table 1**). Therefore, the caregivers of patients with ALS should be informed about possible supportive interventions at an early stage of the disease and offer these interventions repeatedly (40). Moreover, offering customized care in line with the caregiver's preferences would be advised (40). As for our monitoring of resilience measures in the studied groups, the lack of changes in CD-RISC scores across time in both TG and CG may be due to the recognized co-existence and interrelation of burden, resilience, needs, and rewards in caregivers of patients with ALS (16). In this regard, Weisser et al. (16) identified a model of coping for caregivers of patients with ALS that integrates resilience (active/positive), burden (active/negative), needs (passive/negative), and reward

TABLE 2 | Between-group comparison on psychological measures at pre- and post-treatment assessment (using pre-treatment assessment as covariate).

Variable	Untreated (CG)	Treated (TG)	*t*-test[a]; ANCOVA[b]	*p*-value[c]
Connor-Davidson Resilience Scale				
Pre-treatment	62.2 ± 11.51	63.83 ± 8.54	−0.27[a]	0.792
Post-treatment	63.30 ± 8.92	58.83 ± 13.87	0.25[b]	0.629
Caregiver Burden Inventory				
Pre-treatment	31.29 ± 14.90	47.00 ± 16.95	−1.77[a]	0.103
Post-treatment	34.80 ± 16.20	51.83 ± 21.08	0.29[b]	0.601
Perceived Stress Scale				
Pre-treatment	22.86 ± 5.84	17.33 ± 6.47	1.61[a]	0.134
Post-treatment	23.40 ± 7.53	18.83 ± 7.19	0.02[b]	0.888

t-test was used for comparing the Untreated and Treated groups on pre-treatment measures; Analysis of Covariance (ANCOVA) was used for comparing the Untreated (CG) and Treated groups (TG) on post-treatment measures, using the pre-treatment ones as covariates.
[a]Marks the results of the "χ^2 test" and [b]of the t-test. [c]Bonferroni corrected alpha level of 0.05/6 = 0.008.

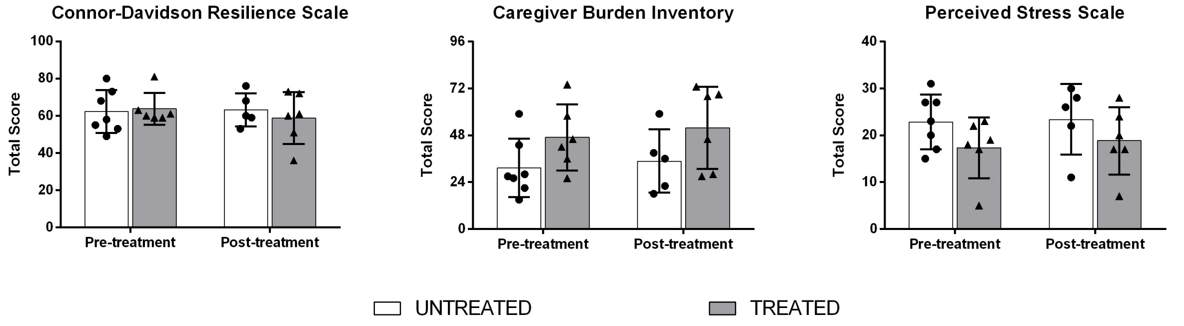

FIGURE 2 | Between-group comparison on Connor-Davidson Resilience Scale (CD-RISC), Caregiver Burden Inventory (CBI), and Perceived Stress Scale (PSS) scores at pre- and post-treatment.

(passive/positive) to develop appropriate individualized caregiver support plans for increasing resilience.

LIMITATIONS AND FUTURE DIRECTIONS

Our pilot study has several limitations: data were collected from a clinic hospital and a small sample of caregivers; thus, our findings may have limited generalizability; the time and number of interventions were too limited; the small sample of caregivers was primarily represented by women; and scales for depression and anxiety were not monitored, primarily aiming to reveal potential changes in resilience and perceived stress across time, in accordance with the tailored, psychological intervention performed. However, most limitations of our study have been frequently shown in most literature concerning psychological interventions for patients with ALS (43). A recent scoping review (43) revealed that the existing studies addressing this topic, which included three randomized-controlled trials and some observational studies, focused on a limited number of psychological outcomes, thus requiring further evaluation. Therefore, future studies are needed to examine the associations between patients' outcomes and family caregivers' psychological support needs and information-seeking behaviors. In addition, the preferences regarding these psychological support sources, such as telemedicine, as well as the accuracy of each source, should be evaluated. It will be necessary to evaluate the effects of combined approaches, such as in-person care and remote psychological support, on the well-being of caregivers of patients with ALS and to implement the number and frequency of the group-therapy sessions and the televisits.

CONCLUSION

The co-occurrence of the COVID-19 pandemic and stressful events related to caring for patients with ALS may influence the response to psychological interventions aimed at reducing caregivers' burden and perceived stress and at increasing resilience. Moreover, a combination of both remote and in-person approaches would be needed in emergencies as well as in routine conditions. In particular, the COVID-19 outbreak, which prompted the more widespread use of telemedicine services, has allowed experience that telemedicine should be intended to be complementary to in-person care in managing patients with ALS.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Institutional Review Board and the Ethics Committee of the University of Campania "L. Vanvitelli" (Naples, Italy). The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

FT, MSic, and DB: conceptualization. FT, MSic, DB, CP, MD, GD'A, FC, SE, MSil, and AR: methodology. CP, MD, GD'A, FC, SE, MSil, and AR: investigation and data collection. MSh and FT: writing—original draft preparation. MSh, FT, MSic, DB, and GT: writing—review and editing. FT, MSic, and GT: supervision. All authors have read and agreed to the published version of the manuscript.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2022.904841/full#supplementary-material

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Multicultural Quality of Life Index in Relatives of People With Borderline Personality Disorder

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Purpose: The aim of the present study was to confirm the original factor structure of the *Multicultural Quality of Life Index* (MQLI) and analyze its psychometric properties in a sample of caregivers of people with borderline personality disorder (BPD).

Methods: The MQLI was administered to 233 relatives of people with BPD. Participants completed the MQLI, the Depression, Anxiety and Stress Scale (DASS-21), and the Connor-Davidson Resilience Scale (CD-RISC).

Results: Factor analysis of the relatives indicated that the MQLI generated a one-factor solution. The MQLI showed good internal consistency, $\varpi = 0.91$ [95% CI (0.90, 0.93)] and correlated significantly and positively with the CD-RISC ($r_S = 0.576$) and negatively with the DASS-21 ($r_S = -0.583$).

Conclusion: Consistent with other studies, the MQLI demonstrated feasibility, strong internal consistency, and good convergent and discriminant validity, which means it is a psychometrically robust measure for the assessment of quality of life in relatives of people with BPD. Along with other validation studies, this measure will be a useful tool for assessing quality of life in relatives of people with mental disorders.

Keywords: quality of life, psychometric properties, relatives, borderline personality disorder, confirmatory factor analysis

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INTRODUCTION

Quality of life (QoL) is becoming one of the key concepts in the healthcare system and social policies, and it is defined as individuals' perceptions of their position in life within a cultural context and their values in relation to their life goals, expectations, concerns, and norms (1). A study by Spitzer et al. (2) indicated that the main goal of the healthcare system is to improve patients' perceptions of their health in relation to their QoL, which is quite important when calculating the cost-effectiveness of treatments (3–5).

Borderline personality disorder (BPD) is associated with severe functional impairment, high use of healthcare resources, a worrisome percentage of suicide rates, and high comorbidity with other mental disorders (6). The characteristics of this disorder, such as emotional instability, impulsivity, fear of abandonment, inappropriate anger, and chronic feelings of emptiness, among others, may explain these negative consequences (6, 7). All these aspects could impact the QoL of individuals with BPD and their relatives. In terms of the influences of this disorder on people with BPD, it should be noted that relatives play an important role in its development, and that its symptomatology affects the family climate. Some studies (8, 9) indicate that relatives of people with BPD have high rates of burden, impaired wellbeing, high levels of psychological distress, and difficulties in emotion regulation, due to their family member's illness. Stress, lack of social and emotional support, economic hardship, and negative experiences produce emotional changes in relatives' QoL (10–12). Thus, it is important to assess and improve QoL in relatives with serious mental illness (e.g., BPD) in order to foster their own health and influence their patients' health by providing them with better care (13, 14).

The Multicultural Quality of Life Index (MQLI) is a self-report originally developed by Mezzich et al. (15). It was constructed to provide a measure of QoL based on the consensus of subject matter experts from several countries, subjective assessment of satisfaction with each domain, and the importance of each domain to each person. The MQLI is a 10-item questionnaire with a Likert-type response scale ranging from 1 (Poor) to 10 (Excellent) that assesses the following areas: physical and psychological wellbeing (e.g., feeling good about oneself), self-care and independent functioning (e.g., performing daily tasks), occupational and interpersonal functioning (e.g., performing one's job; relating well to family, friends, and groups), socioemotional support (e.g., having people to confide in), community and service support (e.g., safe, resourceful neighborhood, access to resources), personal and spiritual fulfillment (e.g., feeling of personal balance; feeling of faith), and overall perception of QoL (e.g., feelings of satisfaction and happiness in one's life).

The MQLI has been validated in different languages, including Spanish, English, Chinese, Korean, and Greek. The procedure for all these validations was carried out with exploratory factor analysis, except the Greek version, which used confirmatory factor analysis. Each validation confirmed the original factorial structure while maintaining the number of items. The Spanish version (15) used two samples of Spanish speakers, one with 60 psychiatric patients and the other with 20 health professionals, obtaining a Cronbach's alpha of 0.89 (in patients) and 0.97 (in health professionals). The sample in the English version (16) consisted of 124 psychiatric patients ($\alpha = 0.91$) and 53 health professionals ($\alpha = 0.90$). For the Chinese version (17), they studied a sample containing 124 psychiatric patients ($\alpha = 0.94$) and 20 health care professionals ($\alpha = 0.95$). The Korean version (18) used two adult samples, one with 100 psychiatric patients and the other with 30 health professionals, obtaining a combined Cronbach's alpha of 0.97. Finally, 884 Community-dwelling adults participated in the Greek version (19), which showed a Cronbach's alpha of 0.90. For discriminant validity, they used the Depression, Anxiety and Stress Scale (20).

As we can see in the aforementioned studies, the MQLI has been validated in different samples and settings, but research on its psychometric characteristics in relatives of people with mental disorders is scarce. The only published validation of the MQLI in a sample of relatives was carried out by Mundal et al. (21) with a sample of 128 relatives of children with attention deficit hyperactivity disorder (ADHD). They obtained good reliability, with a Cronbach's alpha of 0.73. In addition, the correlation between the two MQLI measures and the five-item World Health Organization Wellbeing Index (22) was high ($r = 0.84$).

Moreover, a construct related to QoL is resilience. It is a dynamic process that leads to successful individual adjustment in the face of adversity (23–27). Resilience has been positively associated with QoL, and the relationship between quality of life and resilience can occur in two ways. The first suggests that having higher QoL generates more adaptive coping strategies that result in greater resilience in the person. The second indicates that having greater resilience leads to more adaptive coping and, consequently, to higher QoL (28). To date, no studies have analyzed the relationship between resilience and QoL in relatives of people with BPD. Thus, confirming the relationship between these two variables would allow us to improve current interventions designed to help relatives of people with BPD.

In sum, the MQLI has been validated in numerous studies; however, it has never been validated in relatives of people with BPD. Taking into account that relatives of people with BPD present high levels of anxiety, depression, and burden (29), it is necessary to have reliable instruments to assess their QoL. Adequately assessing the QoL of these relatives could help to detect people at risk of developing psychological problems.

Therefore, the present study has two aims: (1) to analyze the psychometric characteristics and confirm the original factor structure of the MQLI in a sample of relatives of people with BPD; and (2) to study the evidence of the scale's convergent and discriminant validity by relating it to other measures: resilience and depression, anxiety, and stress.

MATERIALS AND METHODS

Participants

The sample consisted of 233 relatives of patients with BPD who were receiving treatment at a Specialized Unit for Personality Disorders with four care facilities in the Valencian Community and one association of relatives of people with BPD in Spain. Recruitment was carried out from 2018 to 2021. The inclusion criteria were: (a) being a relative of a patient who met the criteria for BPD according to the Diagnostic and Statistical Manual of Mental Disorders (DSM-5) (30); (b) agreeing in writing to voluntarily participate in the study by signing the informed consent form. The exclusion criterion was the presence of a severe mental disorder in the relative (psychosis, schizophrenia, bipolar disorder, substance dependence, dementia, severe depression). This study was approved by the Ethics Committee of the University of Valencia with code: UV-INV_ETICA-1623849.

Regarding the gender of the participants, 67.4% ($n = 157$) were women, and 32.6% ($n = 76$) were men. Regarding the family role, 59.7% ($n = 139$) were mothers, 22.3% ($n = 52$) were fathers, 6% ($n = 14$) were son/daughters, 5.6% ($n = 13$) were partners, 3.4% ($n = 8$) were brothers or sisters, 1.3% ($n = 3$) were partners of the mother, 0.9% ($n = 2$) were uncles, and 0.4% ($n = 1$) was a grandfather. One participant did not report his/her family role. The mean age of the participants was 54.44 years ($SD = 10.09$).

Most of the relatives were married or living with a partner (56.6%; $n = 132$), 22.7% ($n = 53$) were single and 20.6% ($n = 48$) were separated, divorced, or widowed. As for the level of

TABLE 1 | Descriptive statistics and internal consistency of the scales used in the present study.

	Scale	***M* (*SD*)**	***Sk* (*SE* of *Sk*)**	***K* (*SE* of *K*)**	**ϖ (95% CI)**
Whole sample *N* = 233	MQLI	63.68 (15.82)	−0.222 (0.160)	−0.634 (0.319)	0.91 (0.90, 0.93)
	RS	2.55 (0.56)	−0.104 (0.187)	−0.240 (0.371)	0.90 (0.87, 0.92)
	DASS-21	2.54 (1.96)	0.931 (0.159)	0.369 (0.318)	0.95 (0.95, 0.96)
Men's subsample *N* = 76	MQLI	66.92 (15.58)	−0.335 (0.276)	−0.637 (0.545)	0.92 (0.98, 0.95)
	RS	2.53 (0.58)	−0.066 (0.314)	0.073 (0.618)	0.91 (0.87, 0.94)
	DASS-21	1.80 (1.66)	1.091 (0.276)	0.397 (0.545)	0.95 (0.93, 0.96)
Women's subsample *N* = 157	MQLI	62.09 (15.75)	−0.177 (0.195)	−0.596 (0.387)	0.91 (0.89, 0.93)
	RS	2.57 (0.55)	−0.122 (0.229)	−.388 (0.455)	0.89 (0.86, 0.92)
	DASS-21	2.90 (1.99)	0.867 (−194)	0.248 (0.385)	0.95 (0.94, 0.96)

MQLI, multicultural quality of life index; RS, resilience scale; DASS-21, depression, anxiety and stress scale-21; Sk, skewness; SE, standard error; K, Kurtosis.

TABLE 2 | Descriptive statistics, total-item correlations, and ϖ if item is dropped from the MQLI (whole sample).

MQLI items	***M* (*SD*)**	***Sk***	***K***	**r_s total-item**	**ϖ if item dropped**
1. Physical wellbeing**/**Bienestar físico (sentirse con energía, sin dolores ni problemas físicos)	5.83 (2.11)	−0.133	−0.632	0.648	0.91
2. Psychological/emotional wellbeing/Bienestar psicológico/emocional (sentirse bien y satisfecho consigo mismo)	5.58 (2.11)	−0.081	−0.785	0.761	0.90
3. Self-care and independent functioning/Auto-cuidado y funcionamiento independiente (cuida bien de su persona, toma sus propias decisiones)	7.22 (1.92)	−0.596	−0.018	0.698	0.91
4. Occupational functioning/Funcionamiento ocupacional (capaz de realizar trabajo remunerado, tareas escolares, y tareas domésticas)	7.73 (2.06)	−1.013	−0.651	0.620	0.91
5. Interpersonal functioning/Funcionamiento interpersonal (capaz de responder y relacionarse bien con su familia, amigos y grupos)	7.66 (1.81)	−0.613	−0.424	0.699	0.91
6. Social emotional support/Apoyo social-emocional (disponibilidad de personas en quien puede confiar y de personas que le proporcionen ayuda y apoyo emocional)	6.75 (2.18)	−0.503	−0.307	0.667	0.91
7. Community and services support/Apoyo comunitario y de servicios (buen vecindario, disponibilidad de recursos financieros y de otros servicios)	6.17 (2.27)	−0.487	−0.260	0.634	0.91
8. Personal fulfillment/Plenitud personal (sentido de equilibrio personal, de autogobierno, de solidaridad, y de disfrute sexual y estético)	5.81 (2.20)	−0.192	−0.733	0.812	0.90
9. Spiritual fulfillment/Plenitud espiritual (experimentar una elevada filosofía de vida, religiosidad y trascendencia más allá de una vida)	5.39 (2.32)	−0.150	−0.692	0.533	0.91
10. Global Perception of Quality of Life/Percepción global de Calidad de vida (sentirse satisfecho y feliz con su vida en general)	5.77 (2.13)	−0.195	−0.657	0.757	0.90

$N = 233$. Skewness Standard Error = 0.160; Kurtosis Standard Error = 0.320. MQLI $\omega = 0.91$.

education, 8.2% (n = 19) had no studies, 22.3% (n = 52) had primary education, 29.6% (n = 69) had secondary education, and 39.9% (n = 93) had higher education.

Instruments

The Multicultural Quality of Life Index

The Multicultural Quality of Life Index (MQLI) (15) has been extensively described in the introduction.

Depression, Anxiety, and Stress Scale

The Spanish version (31) of the Depression, Anxiety, and Stress Scale (20) has been used in this study.. It is a 21-item Self-report that assesses the severity of a range of problems common to depression, anxiety, and stress. It uses a Likert-type response scale ranging from 0 (it does not happen to me) to 3 (it happens to me a lot or most of the time). The Depression scale assesses dysphoria, hopelessness, and anhedonia, among others. The Anxiety scale assesses autonomic

TABLE 3 | Descriptive statistics, correlations of the MQLI, and ϖ if item is dropped from the MQLI (men's subsample).

MQLI items	*M* (*SD*)	*Sk*	*K*	r_s total-item	ϖ if item dropped
1. Physical wellbeing/Bienestar físico (sentirse con energía, sin dolores ni problemas físicos)	6.12 (1.93)	−0.149	−0.721	0.675	0.91
2. Psychological/emotional wellbeing/Bienestar psicológico/emocional (sentirse bien y satisfecho consigo mismo)	6.01 (2.06)	−0.179	−0.738	0.785	0.91
3. Self-care and independent functioning/Auto-cuidado y funcionamiento independiente (cuida bien de su persona, toma sus propias decisiones)	7.54 (1.82)	−1.105	1.481	0.658	0.92
4. Occupational functioning/Funcionamiento ocupacional (capaz de realizar trabajo remunerado, tareas escolares, y tareas domésticas)	7.92 (1.85)	−0.848	−0.253	0.569	0.92
5. Interpersonal functioning/Funcionamiento interpersonal (capaz de responder y relacionarse bien con su familia, amigos y grupos)	7.79 (1.78)	−0.684	−0.197	0.700	0.91
6. Social emotional support/Apoyo social-emocional (disponibilidad de personas en quien puede confiar y de personas que le proporcionen ayuda y apoyo emocional)	6.86 (2.22)	−0.609	−0.240	0.753	0.91
7. Community services support/Apoyo comunitario y de servicios (buen vecindario, disponibilidad de recursos financieros y de otros servicios)	6.42 (2.39)	−0.590	−0.162	0.670	0.92
8. Personal fulfillment/Plenitud personal (sentido de equilibrio personal, de autogobierno, de solidaridad, y de disfrute sexual y estético)	6.17 (2.27)	−0.395	−0.735	0.792	0.91
9. Spiritual fulfillment/Plenitud espiritual (experimentar una elevada filosofía de vida, religiosidad y trascendencia más allá de una vida)	5.87 (2.20)	−0.179	−0.441	0.601	0.92
10. Global Perception of Quality of Life/Percepción global de Calidad de vida (sentirse satisfecho y feliz con su vida en general)	6.38 (2.05)	−0.306	−0.715	0.762	0.91

N = 76. Skewness Standard Error = 0.276; Kurtosis Standard Error = 0.545. MQLI ω = 0.92.

arousal, skeletal muscle effects, situational anxiety, and subjective experience of anxious affect. The Stress scale is sensitive to levels of chronic non-specific arousal. It assesses difficulty relaxing, nervous arousal, and being easily upset/agitated, irritable/over-reactive, and impatient. Higher values indicate more severe negative emotional symptoms. Regarding the internal consistency, Cronbach's alphas were excellent, ranging from 0.94 to 0.87. In this study, Cronbach's alphas ranged from 0.94 to 0.84. In the present study, the Depression, Anxiety, and Stress Scale (DASS-21) showed good internal consistency, $\varpi = 0.95$ [95% CI (0.94, 0.96)], which did not improve if an item was dropped.

The Connor–Davidson Resilience Scale

The Connor-Davidson Resilience Scale (32) is a 25-item self-report measure that assesses a broad range of resilience characteristics, including resilience, personal competence, tolerance of negative emotions, positive acceptance of change, personal control, and spirituality. It uses a Likert-type response scale ranging from 0 (not at all) to 4 (almost always). The Connor–Davidson Resilience Scale (CD-RISC) is a psychometrically sound measure of resilience that was designed to be used as an outcome measure. In the present study, the CD-RISC showed good internal consistency, $\varpi = 0.90$ [95% CI (0.87, 0.92)], which did not improve if an item was dropped.

Procedure

Participants in the study were from two institutions located in Spain: (a) a Specialized Unit for Personality Disorders and (b) the Association for Family members of persons with BPD. The diagnostic interviews were conducted by six clinical psychologists with doctoral degrees and more than 10 years of experience in the assessment and treatment of BPD. Once the study had been explained to the family members, they were offered the opportunity to participate in the study. Interested family members signed the informed consent form, and the clinical psychologists carried out a clinical interview to verify that they met the inclusion and exclusion criteria. Then the participants filled in the assessment protocol: MQLI, DASS-21, and CD-RISC.

Statistical Analyses

First, we analyzed the descriptive statistics (means and standard deviations), measures of data distribution (skewness and kurtosis), and internal consistency (McDonald's omega, ϖ) of the scales used in the present study and the MQLI items, as well as the item-rest correlations and the change in McDonald's ϖ of the MQLI if an item was dropped.

Second, we carried out a confirmatory factor analysis (CFA) to test the fit of the unidimensional model proposed for the MQLI to date. Because Mardia's coefficient was >5 (it was 8.9123)

TABLE 4 | Descriptive statistics, correlations of the MQLI, and ϖ if item is dropped from the MQLI (women's subsample).

MQLI items	*M* (*SD*)	*Sk*	*K*	r_s total-item	ϖ if item dropped
1. Physical wellbeing/Bienestar físico (sentirse con energía, sin dolores ni problemas físicos)	5.69 (2.18)	−0.086	−0.641	0.638	0.90
2. Psychological/emotional wellbeing/Bienestar psicológico/emocional (sentirse bien y satisfecho consigo mismo)	5.37 (2.11)	−0.028	−0.784	0.737	0.90
3. Self-care and independent functioning/Auto-cuidado y funcionamiento independiente (cuida bien de su persona, toma sus propias decisiones)	7.06 (1.96)	−0.387	−0.366	0.710	0.90
4. Occupational functioning/Funcionamiento ocupacional (capaz de realizar trabajo remunerado, tareas escolares, y tareas domésticas)	7.63 (2.16)	−1.032	0.743	0.652	0.90
5. Interpersonal functioning/Funcionamiento interpersonal (capaz de responder y relacionarse bien con su familia, amigos y grupos)	7.60 (1.83)	−0.585	−0.497	0.703	0.90
6. Social emotional support/Apoyo social-emocional (disponibilidad de personas en quien puede confiar y de personas que le proporcionen ayuda y apoyo emocional)	6.70 (2.16)	−0.457	−0.295	0.630	0.90
7. Community and services support/Apoyo comunitario y de servicios (buen vecindario, disponibilidad de recursos financieros y de otros servicios)	6.05 (2.21)	−0.464	−0.255	0.608	0.90
8. Personal fulfillment/Plenitud personal (sentido de equilibrio personal, de autogobierno, de solidaridad, y de disfrute sexual y estético)	5.63 (2.16)	−0.113	−0.645	0.814	0.89
9. Spiritual fulfillment/Plenitud espiritual (experimentar una elevada filosofía de vida, religiosidad y trascendencia más allá de una vida)	5.16 (2.35)	−0.112	−0.799	0.485	0.91
10. Global Perception of Quality of Life/Percepción global de Calidad de vida (sentirse satisfecho y feliz con su vida en general)	5.46 (2.10)	−0.152	−0.620	0.745	0.90

N = 157. Skewness Standard Error = 0.195; Kurtosis Standard Error = 0.389. MQLI ω = 0.91.

and the MQLI is an ordinal scale, robust (33) and Diagonally Weighted Least Squares (DWLS) (34) methods were used (35). The fit indices used were the Comparative Fit Index (CFI; values ≥0.90 indicate acceptable fit, and values ≥0.95 indicate good fit), the Tucker-Lewis Fit Index (TLI; values ≥0.90 indicate acceptable fit, and values ≥0.95 indicate good fit), the Root Mean Square Error of Approximation index (RMSEA; values lower than 0.080 indicate acceptable fit, and values <0.50 indicate good fit), and the Standardized Root Mean Square Residual index (SRMR; values <0.080 indicate acceptable fit, and values <0.050 indicate good fit) (36).

Third, to analyze the construct validity of the MQLI, the correlations (Spearman Spitzer s *rho*, r_s) with both the CD-RISC (to test concurrent validity) and the DASS-21 (to test divergent validity) were analyzed.

All these statistical analyses were carried out with the JASP0.15 software (37).

RESULTS

The main result of this study was that the MQLI showed adequate psychometric properties, that is, a good internal consistency and both factorial and construct validity.

Descriptive Statistics and Correlations

Table 1 shows the descriptive statistics, skewness, kurtosis, and internal consistency of the scales used in the present study in the whole sample and in the subsamples of men and women.

Tables 2–4 show the descriptive statistics, skewness, and kurtosis of the MQLI items, the item-total correlations, and the MQLI's internal consistency if any item was dropped in the whole sample (**Table 2**) and in the subsamples of men (**Table 3**) and women (**Table 4**).

In the whole sample, data distribution was moderately and negatively skewed (negative skewness for the item 4 was >-1) and platykurtic. Positive kurtosis was found for Item 3 in the men's subsample and for Item 4 in the women's subsample.

In the whole sample and in the men's subsample, all the item-scale correlations were >0.50. In the women's subsample, the item-scale correlation for Item 9 was slightly below 0.50. In the present study, the MQLI showed good internal consistency, ϖ = 0.91 [95% CI (0.90, 0.93)], which did not improve if an item was dropped.

Structural Validity

The unidimensional 10-item model for the MQLI showed a good fit: $SB\chi^2_{(35)} = 35.865$, $P = 0.428$, CFI = 1.000, TLI = 1.000,

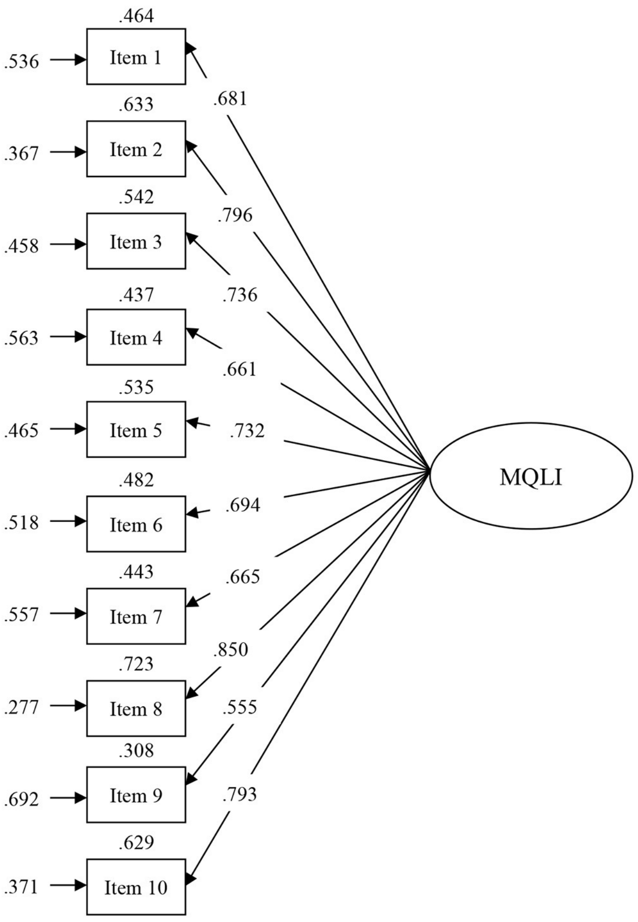

FIGURE 1 | Model obtained in the present study for the MQLI. Values at the top of each rectangle are R^2; values at the left of each rectangle are errors; parameter estimates and residual variances are standardized.

RMSEA = 0.010, 95% CI [0.000, 0.049], SRMR = 0.057. The CFI index was >0.95, the RMSEA index was lower than 0.050, and the SRMR was <0.080 and close to 0.050. All parameters were significant at the 0.05 level (**Figure 1**).

Construct Validity of the MQLI

The MQLI correlated significantly, $P < 0.001$, and positively with the CD-RISC ($r_s = 0.576$) and negatively with the DASS-21 scale ($r_s = -0.583$), respectively. These strong correlations were in the expected direction, according to the construct assessed with each scale, Cohen (38).

DISCUSSION

The present study had the general objective of analyzing the psychometric properties of the MQLI in Spanish relatives of people with BPD. Specifically, the first aim was to study the internal structure of the MQLI, using a one-factor confirmatory model, and its internal consistency. The second aim was to study the evidence of the scale's convergent and discriminant validity by relating it to other measures: resilience and depression, anxiety, and stress.

Regarding the first aim, our results showed that the data had a good fit to a factorial model with one factor called QoL. This result confirms the original structure found by Mezzich et al. (15), and it also confirms the good psychometric properties of the MQLI. Moreover, the data from the present study confirm the results obtained with the QLI in numerous communities (15–19).

Regarding the second objective of the present study, the MQLI showed adequate convergent validity with measures of Resilience and psychopathology, as previous studies have shown (28). QoL was highly and positively associated with Resilience. This result suggests that these two constructs are strongly related, as indicated in previous studies that found that resilience was a significant predictor of QoL in individuals with schizophrenia, bipolar disorder, and healthy controls, such that higher resilience led to higher QoL (39, 40). Moreover, our results provide evidence of the divergent validity of the MQLI. The MQLI had a high and negative association with depression and a low and negative association with anxiety and stress. These results are similar to those from other studies (19) that found that QoL was negatively associated ($P \leq 0.05$) with severe depression, anxiety, and stress. We want to highlight that this is the first study to analyze the psychometric characteristics of the MQLI in relatives of people with BPD. Our results suggest that the MQLI is an adequate measure to evaluate QoL in this specific sample.

This study has several limitations. First, the sample, although sufficient to perform a CFA of the MQLI, is not large enough to study the invariance with respect to the gender and age of the participants. Thus, future studies should check whether the structural model of the MQLI is invariant for men and women and at different ages in a larger sample of Spanish participants, which, in turn, would make it possible to analyze gender and age-related differences. Moreover, our study does not include a test-retest analysis, and so future research should replicate our results in a longitudinal study and analyze test-retest reliability. Finally, we have used resilience as a measure to analyze convergent validity. Although resilience and QoL are related, they are two different constructs, and this is a clear limitation of our study. We propose adding another instrument that evaluates QoL to analyze convergent validity in future research.

Regarding clinical practice, this is a good instrument for the assessment of QoL in relatives of people with BPD, in order to easily and efficiently identify relatives who need psychological support and treatment for the problems they have with their loved ones. Thus, it is essential that patients receive adequate and continuous treatment and strong social support (14).

In sum, the present research provides support for the good psychometric properties and reliability of the MQLI in relatives of people with BPD, and the results suggest that the MQLI is an adequate measure to assess QoL.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Ethics Committee of the University of Valencia (Valencia, Spain) (UV-INV_ETICA-1623849). The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

JM drafted the manuscript with important contributions from IF-F, AG-P, and VG. JM in collaboration with AG-P, VG, IF-F, RB, SF-B, SP, JG-A, and AG-P designed the study and participated in each of its phases. AG-P, VG, IF-F, SP, and JG-A participated in the review and revision of the manuscript and have approved the final manuscript to be published.

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Parental perspectives on the quality of life of children with Down syndrome

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Down Syndrome (DS) is the most common chromosome abnormality and the most frequent cause of developmental delay/intellectual disabilities in children. Although the investigation of the quality of life (QoL) is crucial in children with DS, relatively poor attention has been paid to this topic. The current study aimed to evaluate parent-reported QoL in a group of children with DS and identify children's individual and clinical features associated with different levels of QoL. We included in the study 73 children with DS (5–12 years) and investigated the parent-reported levels of QoL by means of the Pediatric Quality of Life Inventory. Cognitive level and the presence of behavioral difficulties were also evaluated. The overall parent-reported QoL of children with DS was high; emotional functioning was the domain with the highest level of QoL. Moreover, parents perceived low levels of QoL in children who exhibited low IQ, worse analogical reasoning, worse adaptive skills, more frequent challenging behaviors, more ritualistic/sameness behavior and more autistic symptoms. No differences emerged for family variables, namely parental education and employment, between the two groups with high and low QoL, as perceived by parents. The understanding of cognitive and behavioral factors - such as analogical reasoning, socio-communication abilities and challenging behaviors - related with different degrees of QoL in children with DS is crucial for the development of effective strategies to promote the improvement of the QoL.

KEYWORDS

trisomy 21, analogical reasoning, challenging behaviors, ritualistic behavior, autistic symptoms

Introduction

According to the World Health Organization, *quality of life* (QoL) can be defined as "individuals' perceptions of their position in life in the context of the culture and value systems in which they live in relation to goals, expectations, standards, and concerns" (1). The assessment of QoL includes the investigation of the individual's or caregiver's subjective evaluation of well-being across different domains, such as physical, emotional and social well-being. This model allows to measure subjective estimates of QoL across individuals at all developmental stages and across individuals with and without developmental disabilities. In particular, evaluation of QoL among children and adolescents is critical in detecting subsets with poor health status and in developing effective strategies to increase health of the younger population (2). The importance of a proper assessment of QoL in pediatric age becomes even clearer with reference to clinical groups. In these cases, evaluation of QoL in clinical practice can help optimizing communication between clinicians and the child and his/her caregivers as well as recognizing physical or mental health problems from the patient and caregiver perspectives and areas of potential amelioration (3).

The assessment of QoL is well-suited to conditions that have a multifaceted impact, such genetic syndromes and neurodevelopmental disorders, including intellectual disability (ID).

Children with these conditions manifest several difficulties in communication, motor and social functioning, with important consequences on the possibility to independently manage daily life activities. Consistently, children with neurodevelopmental disorders have higher health care service utilization than typically developing children (4, 5) and are more vulnerable to mental health problems (6). Given such a multidimensional impact of neurodevelopmental disorders, increasing attention has been paid to the investigation of QoL in children with neurodevelopmental disorders and genetic syndromes. In particular, children with ID display poorer QoL than their typically developing peers (7); similarly, parents of children with neurogenetic syndromes, such as Prader Willi syndrome, tend to perceive lower levels of QoL in their children (8). Research identified a number of factors associated with poorer QoL, such as older age, worse adaptive skills, the presence of challenging behaviors, and more complex needs such as autism spectrum disorder and medical comorbidities (9–19). For instance, Beadle-Brow and collaborators (12) reported life satisfaction, as perceived by proxy respondents, was related to independent functioning in childhood in a cohort of people with severe ID and/or autism; moreover, the same study provided support for a crucial role of challenging behaviors in the proxy respondents' perception of QoL. These results were confirmed and extended by a subsequent study including a group of 246 children with severe developmental disabilities that reported that higher QoL was related to younger age, higher adaptive skills, lower maladaptive behavior (7). The central role of adaptive behavior was further supported by another study on adults with intellectual and developmental disabilities; the authors found not only adaptive behavior was significant and relevant from both third-party and participants' perspective, but also problem behaviors had a modest negative impact on the QoL (17). Similar results were reported also from research on specific neurogenetic syndromes, such as Fragile X. Coffman and colleagues (19), indeed, found parent-reported QoL significantly correlated with adaptive functioning, social impairment, and aberrant behaviors; in particular, greater parent-reported QoL was associated with reduced aberrant behaviors. Findings on challenging behaviors are highly relevant with reference to QoL, given that individuals with ID are three times as likely to exhibit aggressive behaviors toward others, self-injury, or behavior destructive to property (20). To address such complex needs in this population, targeted interventions are designed to support children and their families globally.

Among children with developmental disabilities, those with Down Syndrome (DS) exhibit specific features. With a worldwide incidence rate of 1:1,000–1,100 in newborns (21), DS is the most common chromosome abnormality and the most frequent cause of developmental delay/ID in children (22). DS is caused by the presence of a supernumerary chromosome 21, resulting in a constellation of clinical features (23). This includes ID often associated with multiple health comorbidities such as cardiac defects, delayed growth, hematology and thyroid abnormalities, autoimmune diseases, and obstructive sleep apnea (23), as well as with behavioral problems (24, 25). Such a complex condition contributes to the demand for additional medical care in comparison with other forms of ID. Considering the important consequences deriving from cognitive, medical and behavioral correlates of DS on individual's functioning, it is crucial to measure the QoL in such population, as well as to identify factors that can influence it.

Despite the investigation of the QoL is crucial in children with DS, relatively poor attention has been devoted to this topic (23). However, existing research indicates that QoL of children with DS is lower than typically developing children (26–28) and, more specifically, variations in the levels of QoL across different domains emerge. For instance, children with DS exhibit low levels of physical well-being but high levels of emotional well-being (28, 29). Concerning the factors associated, sex seems to be unrelated to QoL in DS (30, 31), whereas findings on age are inconsistent. Indeed, some studies indicated a worsening of QoL with increasing age (29, 31), others reported higher QoL in young adults in comparison with adolescents (30), and others failed to detect age-related differences (32). Moreover, behavioral problems and medical comorbidity have been associated with poorer QoL in adolescents with DS (30). Finally, family resources, such as family income, have been also related to QoL in children with DS (33).

In spite of the increased awareness of the importance of evaluating QoL in children with DS, research investigating the levels of QoL and associated factors is scarce. In addition, inconsistent findings about the correlates of QoL in children with DS still persist. Finally, few evidence derives from the evaluation of objectively measured correlates of QoL in children with DS, such as IQ. Indeed, most of the previous studies mainly rely on parental reports on child's QoL and used a single instrument for behavioral evaluation excluding a detailed characterization of specific behavioral aspects - including challenging behaviors and autistic symptoms.

Specific aims of the present study were: (i) evaluating parent-reported QoL in a group of children with DS; (ii) identifying child's individual and clinical features associated with different levels of children's QoL as perceived by parents. Based on previous research on neurodevelopmental disorders, we hypothesized that lower QoL levels in the physical domain than in the emotional and social domains would be observed. We also hypothesized that the children perceived as with higher QoL would have higher cognitive abilities, better adaptive skills, less autistic symptoms, less behavior problems and less repetitive behaviors.

Materials and methods

Participants

Seventy-three children with DS (46 boys, 27 girls) ranging in age from 5 to 12.11 years of age (mean 8.97 ± 2.24 years) were included in the study. The mean IQ was 57.42 ± 7.08. Selection criteria included, besides the diagnosis of DS based on the analysis of the karyotype, the age ranging between 7 and 12 years. Exclusion criteria were as follows: age <5 and >12 years; language barrier hampering questionnaire compilation by parents. All participants underwent a child psychiatric and neuropsychological examination conducted by experienced developmental neuropsychiatrists and neuropsychologists.

Procedure

This is a cross-sectional study; data were retrospectively collected from a file review of patients with DS referred for a clinical evaluation at the Child and Adolescent Neuropsychiatry Unit of the Bambino Gesù Children's Hospital in Rome between December 2021 and April 2022. We investigated the levels of parent-reported QoL in children with DS; moreover, the differences between children with high levels of QoL and children with low QoL were analyzed. Child's characteristics included sex, age, non-verbal IQ, adaptive level, behavioral problems and autistic symptoms. The clinical evaluation of children and adolescents with DS consisted in a neuropsychiatric, neuropsychological and psychopathological/behavioral assessment performed by a team made of a child neuropsychiatrist and clinical psychologists and neuropsychologists with clinical expertise on DS. The clinical evaluation also included the administration of parent-report questionnaires, which were filled out by the parents while the children underwent neuropsychological or behavioral evaluation. All parents received precise instructions regarding filling out the questionnaires.

Due to the retrospective design, data were collected from the hospital records and clinic charts and the de-identified data were analyzed. All parents signed a written informed consent for data use for research purposes and a privacy statement that ensures that data will be kept confidential. For the current project, all subjects meeting specified criteria as described above were selected from a database. The study was conducted according to the guidelines of the Declaration of Helsinki.

Measures

Quality of life

Pediatric Quality of Life Inventor (PedsQL^{TM}) 4.0 is the standard generic core scale of QoL to measure the physical and psychosocial health of either healthy or ill children (34). The questionnaire was administered to parents. PedsQL has been previously used to assess parent-reported QoL also in children with DS (27, 28) and other forms of developmental disabilities (35). The instrument is age-specific: for this study, we used the 5–7 years and the 8–12 years versions. The PedsQL includes 23 questions with a five-point rating scale (range of 0 to 4 points based on the agreement in each statement). Questions cover the following areas: physical health and psychosocial health, including emotional, social, and cognitive/school functioning. The scores were transformed into full scores of a maximum of 100 points in each area: 100 points meant good QoL, while 0 points meant QoL with very frequent problems. The reliability and validity of PedsQL generic core scales have been well established in healthy and sick populations (36–39). There are no cutoff points to distinguish high QoL from low QoL. However, different studies have tried to distinguish between good and poor QoL. Cut offs for the current study were chosen to be based on the study by Varni and Limbers (34), who identified child self-reported total score of 69.1 and parent proxy reported total scores of 65.4 as "meaningful cut off points for impaired QoL," at one standard deviation below the average PedsQL score for healthy children. We considered the mean between these two reported cut-offs (i.e., 67.2, approximated at 67) to identify high levels of QoL.

Cognitive abilities

Cognitive development was tested by the Leiter-3 (40),which provides a nonverbal measure of intelligence and assesses the ability to reason by analogy, matching and perceptual reasoning, irrespective of language and formal schooling for individuals ages 3–70.

Adaptive functioning

To assess the presence of impairments in adaptive behaviors necessary for socialization, communication, and daily functioning, we used the Adaptive Behavior Assessment System II (ABAS-II) (41), a parent/primary caregiver questionnaire.

ABAS-II consists of eleven skill areas organized into three general domains: conceptual, practical, and social. The composite and domain scores are standard scores with a norm referenced mean of 100 and standard deviation of 15. ABAS-II was standardized and validated for Italian population, showing high internal consistency, good levels of reliability and convergent and clinical validity.

Behavioral problems and autistic symptoms

Aberrant Behavior Checklist (ABC). The ABC (42) is a caregiver rating scale used to assess the severity of core symptoms and comorbid emotional and behavioral problems for several neurodevelopmental and genetic disorders, including DS (43, 44). It consists of 58 questions and five subscales: Irritability/Agitation/Crying; Lethargy/Social Withdrawal; Stereotypic Behavior; Hyperactivity/Non-compliance; Inappropriate Speech. Several studies by the authors (42, 45) and by independent researchers (46, 47) have supported the reliability and validity of the ABC. Moreover, the instrument was used by its authors who determined a good criterion validity in a population of individuals with DS (48).

Social Communication Questionnaire (SCQ) (49). SCQ is a 40-item parent-report questionnaire investigating three major aspects of autism spectrum disorder: communication, social interaction, and repetitive behaviors. The questionnaire exists in two forms: lifetime and current. The "lifetime" form evaluates the patient's developmental history as well as current behaviors, whereas the "current" form assesses the child's behavior during the past 3-month period only. In the present study, the "lifetime" form was used. Item level validity is good (50, 51); sensitivity and specificity in school-aged samples are relatively high (0.86 and 0.78, respectively) (52); specificity and sensitivity >0.80, together with good convergent and discriminant validity, have been reported in a large sample of children with DS (53).

Repetitive Behavior Scale-Revised (RBS-R). The RBS-R (54) is a clinical rating scale including 43-items in a caregiver-completed questionnaire. Items are grouped into six subscales: (1) Stereotyped Behavior; (2) Self-Injurious Behavior; (3) Compulsive Behavior; (4) Ritualistic Behavior; (5) Sameness Behavior; and (6) Restricted Interests Behaviors. Examination of the psychometric properties of the RBS indicates that item-level inter-rater and test-retest reliability and validity are good (55). Previous research found five RBS-R factors (compiling Ritualistic and Sameness domains together) (56). Since the use of the Italian version of the instrument has not been consistently applied in preschoolers (57), we administered RBS to parents of children from 6–12 years of age.

Family variables

Parental education and parental employment were, respectively collected for fathers and mothers and considered as family resources and income indicators, possibly linked to the perceived children's QoL. Parental education was measured as the number of years of education attained. Parental employment was classified as "Not employed," "Lower supervisory, technical, (semi) routine, others," "Intermediate, small employers, own accountants," and "Managerial/professional."

Statistical analyses

Descriptive statistics were used to analyze demographic and clinical characteristics of the whole sample. To surmise the differences of children's features by QoL groups, *t* test and repeated measures analysis of variance (ANOVA) were used; Chi-squared test was used to determine the non-parametric variables. Partial eta squared ($\eta_p{}^2$) was used to measure effect size. *Post hoc* analyses were performed using Tukey HSD test. A p-value ≤ 0.05 was considered as statistically significant.

Results

Quality of life

PedsQL scores are reported in Table 1. Significant differences between domains emerged only for the emotional functioning, which was significantly higher than the other domains (all $p < 0.001$).

The participants included in the study were then split into two groups: children with high and low QoL ($\leq$66 PedsQL total score; $N = 31$; Low QoL group) and children with high QoL ($\geq$67 PedsQL total score; $N = 42$; High QoL group). The two groups did not differ for age ($p = 0.072$) nor sex distribution ($p = 0.793$). We have also investigated the presence of differences in family variables potentially influencing parental perception of children's QoL, namely parental education (in years) and family income. We did not detect differences between Low QoL and High QoL groups in both maternal and paternal education (i.e., years of schooling; $p = 0.257$ and $p = 0.372$, respectively). Similarly, differences between groups for parental occupation did not emerge for mothers nor fathers ($p = 0.252$ and $p = 0.985$, respectively). Mean scores and standard deviations of the questionnaires are reported in Table 1.

Differences in cognitive and adaptive abilities

Children in the High QoL group exhibited significant higher IQ than children in the Low QoL group (M = 59.36, SD = 6.72 and M = 54.81, SD = 6.8, respectively). ANOVA analysis with Group (Low QoL and High QoL) as between factor and the Leiter-3 subtest scores as within factor showed significant

TABLE 1 Mean score (standard deviation) of PedsQL, ABAS II, SCQ, ABC, and RBS in the total sample and in the two QoL groups.

PedsQL

	Total score	Physical health	Emotional functioning	Social functioning	School functioning	Psychosocial health
Total sample	69.01	67.6	77.53	67.57	63.78	67.44
	(16.45)	(20.41)	(16.54)	(22.19)	(19.26)	(19.41)
Low QoL group	52.35	51.26	65.14	47.93	49.71	54.56
	(7.01)	(15.56)	(13.36)	(15.01)	(15.1)	(8.86)
High QoL group	81.31	79.68	86.67	82.07	74.17	76.95
	(8.61)	(14.26)	(12.18)	(13.81)	(14.98)	(19.66)

	ABAS II				**SCQ**
	Global adaptive composite	**Conceptual composite**	**Social composite**	**Practical composite**	**Total score**
Total sample	50.86	55.68	63.67	53.32	10.22
	(13.15)	(8.96)	(14.02)	(13.91)	(6.25)
Low QoL group	46.06	53.29	59.42	48.42	13.35
	(9)	(6.73)	(12)	(10.12)	(6.55)
High QoL group	54.4	57.49	66.88	57.02	7.9
	(14.63)	(10.04)	(14.7)	(15.29)	(4.93)

ABC

	Total score	Irritability/ Agitation/crying	Lethargy/ social withdrawal	Stereotypic behavior	Hyperactivity/non-compliance	Inappropriate speech
Total sample	26.28	6.01	3.88	3.57	10.35	2.76
	(24.86)	(6.94)	(5.15)	(4.53)	(9.68)	(5.76)
Low QoL group	42.72	10.31	6.34	5.65	16.41	4.72
	(26.31)	(7.92)	(5.88)	(5.03)	(10.39)	(8.11)
High QoL group	14.05	2.82	2.05	2.02	5.84	1.31
	(14.85)	(3.78)	(3.64)	(3.43)	(6.06)	(2.19)

RBS

	Total score	Stereotyped behavior	Self-injurious behavior	Compulsive behavior	Restricted interests behaviors	Ritualistic/ sameness behavior
Total sample	20.18	3.54	1.32	3.06	2.26	9.24
	(20.02)	(4)	(2.77)	(3.86)	(2.56)	(10.11)
Low QoL group	27.03	5.10	1.86	3.76	3.21	13.38
	(21.96)	(4.31)	(2.62)	(4.13)	(2.84)	(11.69)
High QoL group	14.67	2.28	0.89	3.62	1.5	7.42
	(16.64)	(3.31)	(2.89)	(0.60)	(2.05)	(7.85)

differences among the groups, $F_{(3,204)} = 3.60$, $p = 0.014$, $\eta_p^2 = 0.05$. *Post-hoc* analyses (Tukey HSD test) revealed that the groups differed only for the Classification and Analogies subtest ($M = 2.4$, $SD = 2.1$ for the High QoL group and $M = 0.83$, $SD = 1.2$ for the Low QoL group; $p = 0.001$). Moreover, children in the High QoL group displayed higher General Adaptive Composite scores at the ABAS II than children in the Low QoL group ($M = 54.4$, $SD = 14.63$ and $M = 46.1$, $SD = 8.1$, respectively; $p = 0.006$).

Differences in behavioral problems and autistic symptoms

ANOVA analysis with Group (Low QoL and High QoL) as between factor and the ABC subscales as within factor showed significant differences among the groups, $F_{(4,264)} = 6.938$, $p < 0.001$, $\eta_p^2 = 0.09$. *Post-hoc* analyses (Tukey HSD test) revealed that children in the Low QoL group exhibited significantly higher scores at the Irritability/Agitation/Crying subscale than children in the High QoL group ($M = 10.31$, $SD = 7.92$ and $M = 2.82$, $SD = 3.78$, respectively; $p < 0.001$) and at the Hyperactivity/Non-compliance subscale ($M = 16.41$, $SD = 10.39$ and $M = 5.84$, $SD = 6.06$, respectively; $p < 0.001$).

Children in the Low QoL group exhibited significantly higher scores at SCQ than children in the High QoL group, indicating more autistic symptoms in this group ($M = 13.35$, $SD = 6.55$ and $M = 7.9$, $SD = 4.93$, respectively; $p < 0.001$).

A subsample of 65 parents completed the RBS. ANOVA analysis with Group (Low QoL and High QoL) as between factor and the RBS subscales as within factor showed significant differences among the groups, $F_{(4,252)} = 4.191$, $p = 0.003$, $\eta_p^2 = 0.006$. *Post-hoc* analyses (Tukey HSD test) revealed that children in the Low QoL group exhibited significantly higher scores at the Ritualistic and Sameness domain than children in the High QoL group ($M = 13.38$, $SD = 11.69$ and $M = 7.42$, $SD = 7.85$, respectively; $p < 0.001$).

Discussion

The aim of the present study was to investigate levels of QoL, as perceived by parents, in a sample of children with DS (ages 5–12 years) and the associated features. Results document how the overall parent-reported QoL of children with DS is high and that emotional functioning is the domain with the highest level of QoL. Moreover, consistently with our hypotheses, children with higher levels of QoL exhibit higher IQ, better adaptive skills, less challenging behaviors, less ritualistic/sameness behavior and less autistic symptoms than children with lower QoL. Moreover, differences in the family variables considered, namely parental education and occupational status, do not emerge in our sample, thus suggesting the identified child's variables result independent from family variables, at least in part.

Parents report high quality of life in children with DS

Emotional functioning is the domain with the highest levels of QoL reported by parents, whereas school functioning is the domain with the lowest scores. This result is in line with previous research reporting high levels of emotional well-being in children with DS (27, 28). Moreover, lower levels of QoL in physical health domain could be explained in light of the higher likeliness of individuals with DS to develop several medical conditions such as cardiac complications and pulmonary disorders (58).

Differently from previous studies, our results failed to document differences in age between the High QoL group and the Low QoL group. Indeed, worse QoL in adolescents with DS aged 16 to 18 years in comparison with children with DS aged 5 to 12 years was reported by Shields and collaborators (29). Lee and collaborators (31) found how the levels of emotional well-being decreases in the transition from childhood to adolescence, namely, children aged 4–5 years showed higher emotional well-being than adolescents aged 13–21 years. In the present study, the lack of significant differences on age between the High QoL and the Low QoL groups could be due to the more restricted age range of the participants included in the current study in comparison with previous research. On the other hand, findings on sex distribution are consistent with literature reporting no sex differences in QoL of individuals with DS (30, 31).

High quality of life is perceived in children with good analogical reasoning

Children with higher QoL exhibited significantly higher IQ than children belonging to the Low QoL group; intriguingly, these differences between groups could be ascribed uniquely to higher scores obtained at the Classification and Analogies subtest of the Leiter-3 by the High QoL group. This subtest includes both representational and non-representational reasoning as well as analogical reasoning problems and measures the ability to perceive and use relational similarity between two objects or geometric shapes and to *inductively* generate rules out of partial information (40). Of note, cognitive activities such as categorization, probability judgment, analogical reasoning, scientific inference, and decision-making include inductive reasoning (59). Several abilities, ranging from problem solving to social interaction, contain some forms of inductive and analogical reasoning, using what is known to make inferences about what is unknown and to find similarities (60). In particular, analogical reasoning seems to support the comprehension of the abstract similarities between seemingly different situations, especially social situations (61–64). Therefore, analogical reasoning plays a critical role in social learning, supporting children in handle social interactions (63, 65, 66). In sum, it emerges significant impairment in analogical reasoning leads to lower perceived QoL in children with DS, suggesting potential implications for cognitive intervention not only in the academic and adaptive functioning, but in the overall QoL. However, it is not clear if training analogical reasoning on non-social stimuli may directly extend to social domain. Cognitive training for individuals with DS should then include exercise on analogical reasoning applied to social sphere. Moreover, future research is required to determine whether training reasoning in relation to social and non-social aspects can improve social abilities and QoL in children with DS. It must be acknowledged that the present study identified several other factors possibly contributing to QoL in children with DS; therefore, interventions aiming to improve QoL should adopt a comprehensive approach, focusing on both cognitive and behavioral domains.

Results from cognitive evaluation were mirrored by findings on adaptive functioning, indicating that children with higher QoL had better adaptive skills than children with low QoL. Adaptive behavior includes practical domain (e.g., feeding, personal care, and staying safe), social functioning (e.g., interpersonal skills, understanding and compliance with rules, and resolution of social problems), and conceptual domain (e.g., language and communication, reading and writing, and handling figures) (46). Children with DS could exhibit reduced autonomy due difficulties with language, memory and executive functions (18). These impairments often require parents and caregivers of individuals with DS to provide support for everyday activities. Of note, children who are not autonomous with daily living skills may lose important social and educational opportunities. The association between adaptive behavior and QoL has been reported in other forms of disability, such as congenital visual disorders (67), and in neurodevelopmental disorders, such as ID not associated with DS (7, 17). The present study confirms these findings also in DS population, indicating that child's adaptive skills may play a role in the perception of QoL in parents of children with DS.

Children with more behavior problems, autistic symptoms, and ritualistic/sameness behaviors are perceived with lower quality of life

As expected, children with perceived lower QoL showed more behavior problems; in particular, significant differences between groups emerged in the Irritability/Agitation/Crying and Hyperactivity/Noncompliance scales of the ABC, with children in the Low QoL group exhibiting higher scores. This is consistent with previous research indicating the impact of behavioral problems on QoL of children with ID (2). Moreover, this finding is in line with literature reporting lower levels of QoL in individuals with attention deficit/hyperactivity disorder, that affects both physical and psychosocial domains (68). Of note, individuals with DS are at risk to exhibit hyperactivity, impulsivity, attentional problems, and non-compliance (69, 70). Given that the presence of behavioral difficulties linked to irritability and hyperactivity may influence the way parents perceive the QoL of their children, it is recommendable that interventions to improve maladaptive behaviors in children with DS should consider also QoL as outcome measure.

As concerns autistic symptoms, children perceived with low QoL exhibited more symptoms as detected by means of the SCQ. This result is consistent with literature reporting lower levels of QoL in children with autism spectrum disorder in comparison with other neurodevelopmental disorders, included DS (71, 72). In addition, previous research reported that children with ID and comorbid autism spectrum disorder exhibit worse QoL than those with only ID; specifically, they display lower physical wellbeing as well as lower scores for interpersonal relationships and social inclusion domains (73). Here, we confirm and extend this result in population with DS. Such finding should be properly took into account when setting up behavioral intervention for children with DS who exhibit autistic symptoms.

As concerns repetitive behaviors, our findings are consistent with previous studies indicating that low QoL is associated with severity of repetitive behavior in children with autism spectrum disorder (74). Of note, in our sample children perceived with low QoL only differed from the High QoL group in the ritualistic/sameness behaviors domain of the RBS-R. This domain captures both the attitude toward the performance of daily living activities in a similar matter and the resistance to change, insisting that things stay the same that can be associated to low cognitive flexibility (56). In the same vein, our findings on cognitive abilities, analogical reasoning and autistic symptoms, related to different levels children's QoL according to parental perspective, suggest that impaired cognitive flexibility could be a crucial factor playing a role for the QoL in children with DS.

The main limitation of the study is that the evaluation of QoL relied on a parent-report instrument and that we missed other sources of information, for example self- or teacher-reports. However, self-report instruments or interviews used to measure the QoL in individuals with ID, included DS, mainly target adult age (75–77). Unfortunately, the use of parent/caregiver-report instruments seems to be the only way to explore the QoL in children with moderate or severe ID. Another important limitation of the study is the lack of a comparison with other groups with different neurodevelopmental disorders. Further research is required to investigate differences and similarities in the determinants of QoL across different forms of developmental disabilities, such as autism spectrum disorder and/or other genetic syndromes. The third limitation of the current study is the cross-sectional design, which prevents drawing conclusions about the nature of the relationship between QoL and the identified variables. Therefore, future studies should aim to identify causal links between cognitive and behavioral factors and QoL levels in children with DS; moreover, longitudinal studies could allow the identification of important predictors of QoL in adolescence and adult age. Moreover, even if the reliability measures of the employed instruments are high according to literature, we did not provide the reliability measures of these instruments in the current study. Finally, given the high proneness of individuals with DS to medical comorbidities, future research on this topic should also consider the influence of physical health problems on QoL levels. It must be noticed that the study was conducted after the onset of the COVID-19 pandemic. Therefore, it cannot be established if, for some individuals, COVID-19 pandemic should have affected QoL values, at least in part. More in-depth research focused on conditions prior to the onset of the pandemic could help us better understand the impact of the pandemic on different domains of QoL in children with DS.

Despite these limitations, to the best of our knowledge this is the first study reporting a characterization of cognitive and behavioral features that play a role in the parental perspective on the QoL of their children with DS. The understanding of factors related with different degrees of QoL in children with DS is important for different reasons. For instance, some authors underlined the importance of measuring the QoL in therapeutic trials for the amelioration of cognitive function in individuals with DS (78). It cannot be excluded indeed that effective treatments, despite inducing higher potential for independence and employment, could make people with DS less satisfied with their lives (78). More importantly, an in-depth understanding of the factors affecting QoL in people with DS could be useful for the identification of strengths and weaknesses of the services provided for this population and, in turn, could help in the developing of effective strategies to promote the improvement of the QoL by addressing unmet needs. Indeed, issues revealed by a deep investigation of the QoL may lead to modifications in care and/or suggest that some interventions provide little benefit in individuals with DS. The results of the current study suggest that behavioral intervention focused on externalizing symptoms and autistic symptoms could exert a substantial effect on the QoL of children with DS. These findings can be also useful to help families anticipate possible conditions associated with DS and their treatment. Considering research on the QoL of people with DS is still limited (23), the results of the present study could provide crucial evidence for the setting up of proper interventions for children with DS and their families. Specifically, our findings suggest that, beyond medical comorbidities, healthcare professionals should encompass, in their assistance activity, interventions on cognitive functioning, autonomy and challenging behaviors to improve QoL for the children with DS.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving human participants were reviewed and approved by Bambino Gesù Children's Hospital. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

Author contributions

Conceptualization and writing—review and editing: EF, FC, and SV. Methodology, formal analysis, and writing—original draft preparation: EF and FC. Investigation and data curation: EF and PG. Supervision and project administration: SV and FC. All authors contributed to the article and approved the submitted version.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

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Relationships between care burden, resilience, and depressive symptoms among the main family caregivers of stroke patients: A cross-sectional study

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Objectives: This study aims to explore the potential mediating role of resilience between care burden and depressive symptoms in family caregivers of stroke patients.

Methods: A cross-sectional study was conducted with a convenience sample involving 245 main family caregivers of stroke patients recruited from the neurology department of a Tertiary A hospital in China. Mediation analyses were conducted using the PROCESS macro (Model 4) for SPSS, applying the Bootstrap analysis with 5,000 samples and a 95% confidence interval.

Results: The results showed that with constant hemiplegia side, Barthel Index, education level, monthly income, care time per day, and living with patients in regression equations, the resilience partially mediated the correlation of care burden and depressive symptoms with a mediation effect ratio of 26.32%.

Conclusions: Resilience plays a mediating role in the correlation between care burden and depressive symptoms.

Impact: The findings indicated a protective effect of resilience in alleviating the negative influences of care burden on depressive symptoms, suggesting that resilience-training intervention may be developed to mitigate depressive symptoms of the main family caregivers of stroke patients.

KEYWORDS

care burden, resilience, depressive symptoms, stroke, family caregivers

Introduction

Stroke is one of the leading causes of adult disability and mortality globally (1, 2), particularly in China (3). Currently, stroke survivors have to cope with severe physical, cognitive, and emotional impairments. Indeed, over two-thirds of stroke survivors require assistance in daily life (4). However, due to the limited community health service, and heavy economic burden, most patients choose to be rehabilitated at home with family caregivers providing care after discharge, and care is provided by family caregivers (5). Nevertheless, family caregivers are experiencing difficulties in employment, finance, sleeping, leisure activities (6), and social activities (7), resulting in degraded life quality, and physical and mental health (8, 9). The care burden involves physical, psychological, and social disruption related to the negative caring experience, which can be divided into objective and subjective components (10). It has been reported that 68.4% of the caregivers of stroke patients in China had a moderate burden and above burden (11), indicating that care burden is a severe issue for caregivers.

Family caregivers are facing huge financial burdens, social pressure, and mental distress (12). A previous study reported a high incidence of negative emotions in caregivers, including low satisfaction with leisure time (13), loss of happiness, loneliness, depression, and a sense of imprisonment (14). Indeed, 53.9% of the caregivers of stroke patients in China have varying degrees of depressive symptoms (11), which might be related to the care burden of family caregivers of stroke patients. Heavy burden leads to emotional exhaustion of caregivers and reduces their enthusiasm, thus affecting the quality of care provided (15). Previous studies showed that caregivers with depressive symptoms were more likely to increase the risk of patients' depressive symptoms (16) and even increased the odds of 6-month mortality of stroke survivors (17). However, some caregivers with care burdens do not experience depression, which emphasizes the essential role of protective factors, such as resilience. Resilience is defined as the ability to effectively adapt to trauma and/or adversity (18). Previous studies have shown that people with higher resilience would actively cope with adversity and rapidly adapt to changes (19, 20). Meanwhile, resilience partially mediates the correlation between negative life events and the mental health of caregivers of patients with advanced cancers (21) and diabetes (22). However, few studies have explicitly tested the mediating effects of resilience between care burden and depressive symptoms of family caregivers of stroke patients.

According to previous studies, care burden is correlated with depressive symptoms, and this correlation can be mediated by resilience. Therefore, it is assumed in this study that there is a correlation between care burden and depressive symptoms, and this relationship might be mediated by the resilience of family caregivers of stroke patients.

Materials and methods

Study design and participants

In this study, a cross-sectional study and a convenient sampling method were employed. The participants were caregivers of patients admitted to the neurology department ward of a Tertiary A hospital in Shenyang, China during 6 January—20 July 2021. An ethical counsel permit (Ref. 402/2020 on 4 January 2021) was issued by the Medical Ethical Committee of the First Affiliated Hospital of China Medical University, and informed consent was obtained from all participants under the Helsinki Accords.

Inclusion criteria

Patients: (1) the patients met the diagnostic criteria of the 4th National Cerebrovascular Disease (The 4th National Symposium on Cardiovascular Disease of the Chinese Medical Association, 1996). Classification of stroke was confirmed by brain computed tomography or magnetic resonance imaging; (2) the score of the Barthel Index $\leq$95.

Family caregivers: (1) 18 years old and above; (2) spent the longest time with the patients per day; (3) unpaid for the care provided; (4) the care time was no <3 months; (5) voluntarily participate in this study.

Exclusion criteria

Family caregivers: (1) suffering from one or more stressful life events within the past 2 weeks (e.g., divorce, widowhood, and loss of job); (2) having a severe physical illness, such as malignancy and intellectual-psychiatric issues; (3) incomplete investigation due to communication or reading and writing obstacles.

Data collection

During January 2021–July 2021, 250 questionnaires were collected from the participants and 245 of them (valid response rate = 98%) were used for data analysis. Five questionnaires were excluded due to data missing. Data collection was completed by a trained researcher using a self-reported questionnaire. The researcher explained the aims of the study to participants and informed them that the collected data will be kept confidential, and that they had the right to refuse participation. If they agree to participate, they will sign a written informed consent. Questionnaires were completed independently by the participant and collected immediately. Additionally, any participant who wished to quit anytime during the study was allowed to do so.

Measurements

Demographic characteristics

The demographic data collected from the patients include gender, age, insurance, stroke subtypes, language barriers, dysphagia, cognitive barriers and hemiplegia side. The demographic data collected from the caregivers include gender, age, education level, monthly income, employment status, relationship with the patient, total care duration, care time per day, and living with patients.

The Barthel Index (BI)

The Barthel Index (BI) was developed by Mahoney (23) in 1965 and has been widely employed to assess self-care activities including eating, bathing, grooming, dressing, using the toilet, transferring from bed to chair, walking, stair climbing, bowel control, and bladder control (23). It consists of 10 items and each item is supposed to be scored based on a 5-point Likert scale. The total score ranges from 0 (total dependence) to 100 (total independence), with 0–20 points defining total dependence, 21–60 defining severe dependence, 61–90 defining moderate dependence, 91–99 defining slight dependence, and 100 defining total independence (23, 24). The validity and reliability of this tool for use in the Chinese elderly population have been well-established (25). In this study, the Cronbach's alpha value was 0.87.

Zarit caregiver burden interview (ZBI)

The Chinese version of the ZBI scale (26), which was used to measure caregivers' perceived burden of providing informal care (27). The scale consists of 22-items assessing role strain and personal strain (28), and each item is supposed to be scored based on a 5-point Likert scale (0 = never, 1 = seldom, 2 = sometimes, 3 = often, and 4 = almost always) (29). The total score ranges from 0 to 88, with 0–20 points defining negligible or no load, 21–40 defining intermediate load, 41–60 defining large load, and 61–88 defining excessive load (30). The Chinese version of ZBI has satisfactory psychometric properties (31). In this study, the Cronbach's alpha value was 0.93.

Connor-Davidson resilience scale (CD-RISC)

The CD-RISC scale was originally developed by Connor and Davidson (32) and translated into Chinese by Yu and Zhang (33), is one of the most widely used scales to measure resilience. The scale consists of 25-items assessing tenacity, strength and optimism (33), and each item is supposed to be scored based on a 5-point Likert scale from 0 (not true at all) to 4 (true nearly all the time) (32). The total score ranges from 0 to 100 and the score is proportional to the resilience level (32). The Chinese version of CD-RISC exhibited good reliability and validity (34). In this study, the Cronbach's alpha value was 0.94.

Center for epidemiological survey depression scale (CES-D)

The Chinese version of the CES-D scale was designed to evaluate the depressive symptoms and risk of disorder in a non-psychiatric person (35). The scale consists of 20-items assessing depressed feelings, somatic complaints, positive feelings and international relationships, and each item is supposed to be scored based on a 4-point Likert scale from 0 (rarely or none of the time) to 3 (most of the time) (35). The total score ranges from 0 to 60 and a score ≥ 16 indicates an elevated level of depressive symptoms (35). Additionally, a score of 16–23 and $\geq$24 were classified as moderate and severe depressive symptomatology (36). The Chinese version of CES-D has been widely used in China with good reliability and validity (37). In this study, the Cronbach's alpha value was 0.95.

Statistical analysis

Statistical analyses were conducted utilizing SPSS version 26.0. Normal distribution tests were verified by using Kolmogorov-Smirnov and Shapiro-Wilk statistics. Continuous variables were presented as mean ± standard deviation (*SD*), whereas classification variables were presented as frequency and percentages (%). Independent sample *t*-test or single-factor variance was conducted to identify differences in depressive symptoms concerning the characteristics of caregivers and stroke survivors. Pearson's correlation analysis was employed to explore the correlation between care burden, resilience, and depressive symptoms. The mediation model was analyzed using Model 4 in the PROCESS Marco (38) version 3.3 with 5,000 iteration bootstrapping to measure the indirect effect and 95% confidence intervals (*CI*) were estimated. Parameters of indirect effects were considered statistically significant when the 95% *CI* did not include 0(39). Hemiplegia side, education level, monthly income, living with patients, care time per day, and the BI score were included as covariates since these variables exhibited significant differences in depressive symptoms and were significantly associated with depressive symptoms. A two-sided $p < 0.05$ was considered statistically significant.

Results

Sociodemographic characteristics

As shown in Table 1, stroke survivors ranged have ages from 34 to 89 years old (mean = 64.09, *SD* = 9.66), 66.53% of them were males and 90.20% of them needed help for daily activities. Of the caregivers aged 27–80 years old (mean = 59.05, *SD* = 1.00), 78.78% of them were females and 75.10% of them were the spouse of the patient. 20.00, 67.35, and 12.65% has a total score (BI) of 0–60, 61–90, and 91–99, respectively.

TABLE 1 Descriptive statistics for demographic characteristics and differences in depressive symptoms ($N = 245$).

Variable	N	%	Depressive symptoms	
			$M \pm SD$	F or t (P)
Patients				
Gender				1.398 (0.163)
Male	163	66.53	22.96 ± 10.21	
Female	82	33.47	21.10 ± 9.01	
Age (years)				1.796 (0.149)
<55	45	18.37	23.71 ± 11.45	
55–64	70	28.57	23.79 ± 9.42	
65–74	105	42.86	21.46 ± 9.71	
≥75	24	9.80	19.48 ± 7.74	
Health insurance				−0.781 (0.436)
Yes	232	94.69	22.22 ± 9.94	
No	12	4.90	24.50 ± 7.79	
Stroke subtypes				1.210 (0.300)
Ischemic stroke	204	83.33	22.40 ± 10.14	
Hemorrhagic stroke	18	7.35	24.72 ± 8.24	
Both	23	9.39	20.00 ± 8.06	
Language barriers				1.776 (0.184)
Yes	77	31.43	23.31 ± 10.22	
No	168	68.57	21.89 ± 9.67	
Dysphagia				1.960 (0.163)
Yes	23	9.39	26.30 ± 8.44	
No	222	90.61	21.92 ± 9.91	
Cognitive barriers				0.422 (0.517)
Yes	10	4.08	21.40 ± 9.24	
No	235	95.92	22.37 ± 9.89	
Hemiplegia side				2.991 (<0.050)
None	11	4.49	20.64 ± 10.24	
Left	108	44.08	21.22 ± 9.14	
Right	74	30.20	21.72 ± 10.34	
Both	52	21.22	25.88 ± 9.91	
Total scores (BI)				8.882 (<0.001)
0–60	49	20.00	27.47 ± 10.84	
61–90	165	67.35	21.05 ± 9.48	
91–99	31	12.65	21.03 ± 7.42	
Caregivers				
Gender				−0.752 (0.453)
Males	52	21.22	21.42 ± 8.82	
Females	193	78.78	22.58 ± 10.11	
Age				0.231 (0.875)
<55	83	33.88	21.96 ± 9.59	
55–64	77	31.43	22.36 ± 9.50	
65–74	71	28.98	23.00 ± 10.89	
≥75	14	5.71	21.00 ± 8.21	
Education status				8.641 (<0.001)
Primary school at most	50	20.41	27.08 ± 10.46	
Junior high school	80	32.65	23.63 ± 9.42	
High school/technical school	80	32.65	19.71 ± 8.19	
College and above	35	14.29	18.60 ± 10.42	
Monthly income (RMB, yuan)				10.811 (<0.001)
<2,500	58	23.67	27.93 ± 10.14	
2,500–3,500	93	37.96	21.72 ± 8.76	
3,500–4,500	70	28.57	20.33 ± 9.17	
>4,500	24	9.80	17.04 ± 9.61	
Working status				4.489 (<0.050)
Employed	91	37.14	22.54 ± 9.60	
Unemployed	60	24.49	25.10 ± 10.87	
Retired	93	37.96	20.32 ± 8.99	
Relationship with patient				5.843 (<0.001)
Spouse	184	75.10	22.55 ± 9.58	
Offspring	52	21.22	20.31 ± 9.48	
Parents	5	2.04	38.60 ± 10.90	
Sibling	4	1.63	18.50 ± 7.94	
Duration of care time (month)				0.677 (0.567)
3–6	89	36.33	21.34 ± 9.68	
6–12	45	18.37	22.58 ± 8.50	
12–36	52	21.22	22.31 ± 9.96	
>36	59	24.08	23.68 ± 10.96	
Care time per day (hours)				7.086 (<0.001)
<4	102	41.63	19.30 ± 7.54	
4–8	98	40.00	23.53 ± 10.16	
8–16	34	13.88	26.21 ± 11.79	
>16	11	4.49	27.82 ± 11.75	
Living with patients				2.407 (<0.050)
Yes	232	94.69	22.69 ± 9.78	
No	13	5.31	16.00 ± 9.22	

SD, standard deviation; BI, the Barthel Index.

Descriptive and correlative analysis

The average scores of care burden, resilience, and depressive symptoms of caregivers were 43.89 ± 13.40, 55.68 ± 11.01, and 22.33 ± 9.85, respectively. 72.65% of the caregivers had depressive symptoms. The results of Pearson's correlation analysis revealed that care burden was positively associated with depressive symptoms ($r = 0.58$, $p < 0.01$). Additionally, resilience was negatively associated with care burden ($r = -0.26$,

TABLE 2 Means, standard deviations, and correlations among variables.

Variable	$M \pm SD$	1	2	3
1. Care burden	43.89 ± 13.40	–		
2. Resilience	55.68 ± 11.01	−0.26**	–	
3. Depressive symptoms	22.33 ± 9.85	0.58**	−0.70**	–

**p < 0.01.

$p < 0.01$) and depressive symptoms ($r = -0.70$, $p < 0.01$), as shown in Table 2.

Mediating effect of resilience

To verify the proposed hypothesis conceptual model, Model 4 of Hayes' PROCESS macro was applied to establish three regression models (see Table 3 and Figure 1). After controlling the hemiplegia side, BI score, education level, monthly income, care time per day, and living with patients, the care burden was positively associated with depressive symptoms ($\beta = 0.51$, $p < 0.001$). After adding resilience, the positive association between care burden and depressive symptoms remained significant ($\beta = 0.38$, $p < 0.001$), while resilience was negatively correlated with care burden ($\beta = -0.21$, $p < 0.01$) and depressive symptoms ($\beta = -0.64$, $p < 0.001$).

Then, bootstrapping was executed to determine the statistical significance of the mediating effect of resilience. We adopted the method of random sampling to extract 5000 Bootstrap samples from the original data ($N = 245$). The results demonstrated that the total effect of care burden on depressive symptoms was 0.38 [95% *CI* (0.29–0.46)], with the direct effect and the indirect effect being 0.28 [95% *CI* (0.22–0.35)] and 0.10 [95% *CI* (0.04–0.16)], respectively. The 95% *CI* did not contain 0, indicating that resilience played a mediating role in the correlation of care burden and depressive symptoms, with a mediating contribution rate of 26.32% (0.10/0.38), as shown in Table 4.

Discussion

The study aims to clarify the correlation between care burden and depressive symptoms by using the Kumpfer's resilience model. First, the mean score of depressive symptoms was 22.33 ± 9.85, which was higher than that reported in previous studies using the same tool for caregivers of patients with other diseases, including dementia (40) and cancer (41). The difference can be attributed to stroke-related disabilities and long disease duration (42), which poses a heavy burden on caregivers of stroke patients.

As the period of data collection in this study coincided with the COVID-19 pandemic, the incidence of depressive symptoms (72.65%) among caregivers was different from those in previous studies (40–71%) (43, 44). The uncertainty induced by the epidemic would increase the psychological pressure [e.g., infection risk (45), unemployment, financial insecurity (46)] on everyone, including caregivers enrolled in this study. Specifically, the decrease of social interaction could have a negative impact on mental health, since caregivers' life and normal rest may get irregular due to restrictions on outdoor activities (47). COVID-19-related financial distress and work impairment were also associated with higher symptom levels of depression (48). A recent study (49) showed that 78.5% of caregivers of children with kidney diseases reported depressive symptoms during the pandemic, which was 32.8–48.3% higher than those in previous studies (50, 51). Meanwhile, the incidence of subjective depressive symptoms increased from 5.9 to 60% among caregivers of patients with dementia (52). On the other hand, patient caring is more challenging due to the restriction of hospitalizations and the complicated admissions process during the pandemic (53). Another recent study reported a 40% drop in stroke admissions (54), and it complained that strict measures due to the pandemic can lead to increased anxiety and distress (55).

There were statistically significant differences in the hemiplegia side, the BI and education, monthly income, working status, correlation with patients, care time per day, and living with patients among the depressive symptoms of the caregivers. Similar to previous studies (11, 16), depressive symptoms are associated with the severity of functional disability of the patients as they are more likely to rely on caregivers for support and care (56). Meanwhile, caregivers with a higher education level tend to experience fewer depressive symptoms, which may be attributed to better ways to insight into illness and seek help (57). The study has shown that unemployment and low income are risk factors for depressive symptoms as limited economic resources, substantial uncertainty and income volatility expose them to physical and mental stress (58). Additionally, the length of care time was proportional to depression. This may be attributed to the fact that a long care time would let to more disruptions in daily life, causing increased stress levels (59). In some studies, parental caregivers exhibited more depressive symptoms compared with spousal caregivers as they are more vulnerable due to physical limitations (60) and prone to have negative emotions related to the future they had envisioned for the child [e.g., care for the child after their death (61)]. Nevertheless, some studies stated that spouses exhibited more depressive symptoms (62, 63), which may be attributed to the fact that spousal caregivers tend to be overwhelmed by conflicting demands such as work, children, and household chores (5). The result indicates that healthcare workers should focus on spousal and parental caregivers

TABLE 3 Mediating effect of resilience between care burden and depressive symptoms.

Controls	Model 1 (resilience)			Model 2 (depressive symptoms)			Model 3 (depressive symptoms)		
	β	*SE*	*t*	*β*	*SE*	*t*	*β*	*SE*	*t*
Hemiplegia side	0.04	0.70	0.65	0.06	0.60	1.13	0.08	0.44	2.12*
The Barthel Index	−0.17	1.38	−2.37**	0.06	1.17	0.96	−0.04	0.88	−0.84
Education level	0.03	0.87	0.40	−0.03	0.74	−0.47	−0.01	0.55	−0.27
Monthly income	0.48	0.90	6.31***	−0.21	0.76	−2.89**	0.10	0.61	1.68
Care time per day	−0.17	0.92	−2.38	0.09	0.78	1.32	−0.02	0.59	−0.37
Living with patients	−0.10	2.72	−1.88	−0.04	2.30	−0.75	−0.11	1.72	−2.69**
Independence variable									
Care burden	−0.21	0.05	−3.37**	0.51	0.04	8.82***	0.38	0.03	8.63***
Mediator									
Resilience							−0.64	0.04	−13.93***
R^2	0.33			0.40			0.67		
F	16.91***			22.67***			60.25***		

$^{*}p < 0.05$, $^{**}p < 0.01$, $^{***}p < 0.001$. Bootstrap sample size = 5,000. β, standardized coefficients; SE, Standard Error; t, t-test value; F, F-test value; R^2, explanatory power.

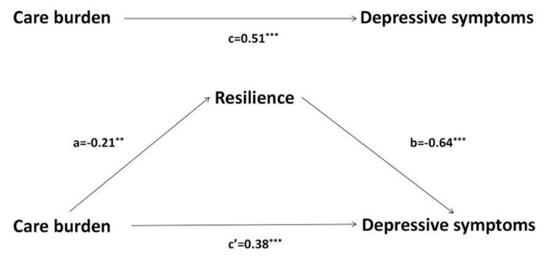

FIGURE 1
The mediating effects of resilience between care burden and depressive symptoms. **$p < 0.01$, ***$p < 0.001$. Standardized regression coefficient for the mediation model. **(a)** is the effect of care burden on resilience, **(b)** is the effect of resilience on depressive symptoms, **(c)** is the total effect of care burden on depressive symptoms, **(c')** is the direct effect of care burden on depressive symptoms (15).

TABLE 4 Bootstrap analysis of mediation effect significance test ($N = 245$).

Effect	*Effect*	*SE*	*95% CI*	
			LLCL	*UICL*
Total effect	0.38	0.04	0.29	0.46
Direct effect	0.28	0.03	0.22	0.35
Indirect effect	0.10	0.03	0.04	0.16

Bootstrap sample size = 5,000. SE, Standard Error; CI, Confidence Interval; LLCL, Lower Limit Confidence Interval; ULCL, Upper Limit Confidence Interval.

with low income, low education, unemployment, living with patients and long-term care in future work, and develop appropriate interventions to reduce depressive symptoms and improve the life quality of both stroke patient and his/her family caregivers.

Notably, the results of the present study showed that there is a statistically significant and inverse relationship between care burden and depressive symptoms in family caregivers. Our findings support previous research (64, 65) suggesting that caregivers who score high care burden have also high depression. Despite these similar findings in the literature, we don't see a ready explanation for this association. In future research we need to further examine the dynamic mechanisms between care burden and depressive symptoms of caregivers. Also, our results showed that resilience could partly mediate the relationship between care burden and depressive symptoms. This is consistent with previous studies that examined resilience as a possible mediator (66), including in the context of COVID-19 research (67). One possible explanation may be that people with a lower level of resilience tend to negatively confront adversity in unhealthy ways, such as mood disturbances, persisting fatigue, and sleep changes (68). Specifically, the psychobiological mechanisms underlying resilience has shown that resilience had a relation to neurochemical, neuropeptide, and hormonal when the response to stressful things (69, 70), people with higher resilience tend to reduce psychobiological allostatic load, and balance neural systems, which could maintain normal psychological function and thus can confront stress actively (71). Besides, individuals with higher resilience are better at coping with stressful events, they tend to make active attempts to adjust the relationship between the environment and individuals, make full use of various resources, and achieve a good state of adaptation (72, 73). Therefore, resilience seemed to be one of the possible mechanisms to resist mental disorders who exposed to care burden, which confirmed Kumpfer's resilience model.

Although family caregivers are often critical to maintain the patients' health, there has been little emphasis on how clinicians should relate to family caregivers (74). Caregivers become "the invisible patient" and often feel tense and upset (74). Hence there is a need to undertake necessary precautions to protect their health. Among patients, a handful of resilience-based interventions have shown promising results for outcomes such as resilience, stress, and anxiety (75, 76). However, there are few published recommendations for conducting intervention trials with stroke caregivers. Some recent studies suggests that a strength-oriented psychoeducational program can reduce depressive symptoms and improved life changes for caregivers (77), as well as the assessment of the risk factors of depressive symptoms (78). The current study suggests that we should assess the situation of care burden and depressive symptoms of caregivers, screen for its main influencing factors, and take effective programs such as social and financial support (79), increased post-traumatic, better patient-caregiver relationships, growth improvement in the competence and self-esteem of caregivers (80). In addition, resilience plays an important role for caregivers' mental health also means it is possible to alleviate the depressive symptoms of caregivers by promoting the level of resilience. Specifically, social support is one of the important sources for the development of resilience, which may ultimately help lessen depressive symptoms (81). Self-compassion and mindfulness training are also related to higher resilience (82). Moreover, some research has shown that love for family, feeling responsible toward the family (83), ability to analyze the current situation, and capability to establish relationships (84) are some of the motivations for resilience. In addition to the aforementioned approach, Overall, the sources of strength can provide intervention targets for promoting resilience and care burden, and thus alleviate the depressive symptoms. Insufficient evidence is available to show that psychoeducational interventions should be implemented in the families of stroke survivors.

There are some limitations in our study to be considered. Firstly, this study is a cross-sectional study, and it is difficult to determine the causal connections between the variables. Therefore, future studies can use longitudinal research to explore the causal relationship between variables. Secondly, we used a self-rating questionnaire for screening for depressive symptoms instead of a clinical diagnosis from psychiatrists. Irrespective, the instrument is a validated depressive symptoms screening tool. Thirdly, our study focused only on the association between care burden, resilience, and depressive symptoms. Further investigation needs to be taken into consideration to explore other social psychology and emotional predictors for the level of depressive symptoms in caregivers of stroke survivors, such as society, family environment factors, and so on. Finally, the COVID-19 level of psychological distress in the current sample has not been assessed, the results must be interpreted with caution. However, in large samples, the current study adds valuable information to incipient efforts to understand care burden and its consequences for family caregivers of patients with stroke, it can help to provide first insights into the research field and help to define directions for the future.

Conclusions

The correlation of care burden, resilience and depressive symptoms in the main family caregivers of stroke patients was explored. The results showed a severe mental health burden on the main family caregivers, especially spousal and parental caregivers, of stroke patients. The self-care ability of patients and conditions of caregivers (e.g., education, income, employment, relationship with the patient, care time per day, and living with patients) were had a direct correlation with depressive symptoms. Care burden was positively correlated with depressive symptoms, while the mediating effect of resilience helps to alleviate depressive symptoms of caregivers with high care burden. This study facilitates understanding and prompt assessment of mental health of the main family caregivers of stroke patients, and the development of resilience-promoting measures in the health care system.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

Ethical counsel permit (Approval number: 402/2020) was approved by the Medical Ethical Committee of the First Affiliated Hospital of China Medical University, and informed consent was received from each participant. All procedures performed in the study involving human participants were in accordance with the ethical standards of the hospital, the National Research Committee, and the 1964 Helsinki Declaration (as revised in Brazil 2013).

Author contributions

LF and JZ conceptualized and designed the study. LF collected the data and prepared Figure 1 and Tables 1–4. LF and MD analyzed the data. JZ reviewed the analyses. LF, WF, and MD drafted the initial version of the manuscript. All authors contributed to revising, editing, and finalizing the manuscript. All authors have read and agreed to the published version of the manuscript.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

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Interventions and strategies to improve social support for caregivers of children with chronic diseases: An umbrella review

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Background: Social support is an important approach to improve the psychosocial health status and promote positive coping for caregivers of children with chronic diseases. Such an approach can reduce parenting stress, help resolve parenting difficulties through the use of various social support relationships.

Methods: We performed an umbrella review methodology using the method of examination, analysis and synthesis of systematic reviews. A PRISMA flow diagram was used to show the search process. The Joanna Briggs Institute was used to appraise the quality of papers and a narrative synthesis was undertaken. Relevant English and Chinese systematic reviews were searched in Embase, PubMed, Web of science, OVID, CNKI, CBM, Wan Fang and Cochrane Library databases, until November 2021, June 2022.

Results: Out of 1,905 records, we included fourteen systematic reviews for a synthesis. Evidence to promote social support for caregivers of children with chronic diseases was identified from four key aspects: (i) Intervention content; (ii) Intervention forms; (iii) Intervention time; and (iv) Sources of support.

Conclusions: The findings of this review suggest that a combination of differing interventions, especially for early family, including the content of parenting training or education, attitude building and resource provision, which can implement online is recommend. More interventional studies and quantitative evidence syntheses are still needed.

Impact: Adequate social support is essential to promote the psychological wellbeing of caregivers of children with chronic diseases. In the early stage of children's diseases, integrating different content and forms of interventions for caregivers' families and actively helping caregivers to identify available support resources can improve social support. The findings from this review can be used to guide caregivers of children with chronic diseases and provide evidence for healthcare professionals and social workers to carry out relevant interventions.

KEYWORDS

social support, caregivers, children, chronic diseases, umbrella review

Introduction

Adequate social support is a protective factor for the mental health of caregivers of children with chronic diseases. Since caregivers are the closest contact of children with chronic diseases, and the mental health of caregivers directly affects the quality of life and psychological condition of children with chronic diseases, the objective of this paper was to determine which interventions and strategies could promote the level of social support among caregivers of children with chronic diseases.

Background

Chronic disease in children is defined as that: the affected population is between 0 and 18 years old; the diagnosis is based on valid and reliable professional criteria; the disease is currently incurable or very resistant to treatment; and the disease is active and has been present for at least 3 months and is expected to persist and/or relapse intermittently (1). Pediatric chronic diseases include diabetes, congenital deformities, asthma, cancer, kidney disease, pervasive developmental disorders, etc. Over the past decades, the prevalence of chronic conditions in children has increased (2, 3). Chronic diseases afflict more than 25% of American children (4). In China, about 10–20% of children suffer from chronic diseases (5). Children with chronic diseases are less happy and less fulfilled compared to healthy peers (6). In addition to this, chronic diseases in children can cause some delay in developmental milestones such as physical, social, and emotional growth (7). These delays not only affect the physical health and psychosocial condition of children but even bring disadvantages to family members and society (7).

Although caring for children with chronic diseases has also brought positive impacts in the form of a sense of achievement and benefit (8–10), it is particularly worth noting that due to the long course of the disease and the special stage of children's growth and development, the challenges faced by caregivers of children with chronic diseases remain central to the parenting process. As key players in parenting, caregivers of children with chronic diseases participate in the whole process of the disease. Caregivers need to be involved in different areas of childcare, including assisting with biomedical, physical, rehabilitation, psychological, and family health issues, and managing the social, financial, and emotional challenges that accompany chronic disease (11). During this process, their behaviors and mentality directly affect the mental health of children (12). Therefore, the mental health of caregivers of children with chronic diseases cannot be ignored. The impact and consequences of caring for families with chronical illness children is a global public health issue with implications for the psychological and relational health of caregivers. Research has shown that caring for children with chronic diseases translates the physical, psychological, socioeconomic, and behavioral impacts of caregivers into vulnerability, reducing the quality of life, life satisfaction, and wellbeing (13, 14). Caring for children with chronic diseases may also affect parents' work, family relationships, and friendships, and lead to personal stress (15).

Social support is usually defined as the social resources that persons perceive to be available or that are actually provided to them by non-professionals in the context of both formal support groups and informal helping relationships (16). It is mainly studied as perceived usability, satisfaction with usability competent support (17, 18) or seeking social support as a positive, problem-focused coping style (19). Social support is commonly associated with wellbeing and psychological growth. Taking adequate social support for caregivers of children with chronic diseases would be beneficial to relieve the pressure of parenting, reduce anxiety, depression and other negative emotions, and improve the quality of life and social function among caregivers of children with chronic diseases (20, 21).

Social support is a protective factor for caregivers of children with chronic diseases. Caregivers of children with chronic diseases have lower levels of social support and higher levels of loneliness than caregivers of healthy children (22, 23). Parents of children with autism in developing countries experience greater stress than parents in developed countries, partly due to the lack of social support systems (24). Researchers emphasize that the establishment of effective social support is beneficial to reduce the risk of mental health problems for parents of children (25). Therefore, how to improve the social support level of caregivers of children with chronic diseases deserves attention.

Studies have shown that the level of social support is related to perceived stress, caregiver mentality, educational attainment, employment status and the utilization of support (26, 27). Some systematic reviews have shown that connected health technologies are beneficial for providing psychosocial support for family caregivers affected by Pediatric cancer (28), and Early Family Intervention Program can increase perceived spousal emotional support for parents of children with appearance-affecting health condition (29). Parenting intervention on psychosocial adjustment can also improve social support for parents of children with type 1 diabetes mellitus (30). However, evidence is fragmented. There are few comprehensive syntheses of existing evidence and the umbrella review to improve social support for caregivers of children with chronic diseases has not been performed. Thus, this review aims to assess, analyze, and synthesize existing evidence for improving social support for caregivers of children with chronic diseases.

Methods

We presented an umbrella review following the PRISMA guidelines (31) and steps in conducting an umbrella review by Aromataris et al. (32). This type of review is more specific and addresses a focused range of outcomes (33). We chose this type of review which inform decision-making and evidence-based practice in health care to summarize existing evidence but do not re-synthesize existing synthesized data. This review followed a written protocol, accessible in Supplementary material 1. Findings were reported using a narrative synthesis.

Search strategy

A systematic search was carried out in the following databases: Embase, PubMed, Web of Science, OVID, CNKI, CBM, Wan Fang and Cochrane Library. We used specific terms embedded in each database to maximize sensitivity. Key terms searched were ("parents" OR "caregivers") AND "social support" AND ("Meta-Analysis" OR "systematic review" OR "Meta"). The full list of search terms and search strategy in per database were provided in Supplementary material 2. Hand searches were conducted by screening reference lists of included articles. Papers published in English and Chinese related to the research topic until November 2021 were set as the search limits. An updated search performed in June 2022.

Eligibility criteria

We used the PICo framework to define the study eligibility criteria. Population: caregivers (fathers and/or mothers) of children with chronic diseases under the age of 18; Intervention/Phenomena of interest: interventions or strategies aimed at social support; Context: In families of children with chronic diseases under the age of 18; Study design: systematic review, meta-analysis; Timeframe: until November 2021, June 2022; Language: English and/or Chinese. We excluded protocols, narrative reviews, scoping reviews or studies without full text.

Assessment of methodological quality

Review papers included in the final analysis were critically evaluated by two authors independently using the Joanna Briggs Institute Critical Appraisal Checklist for Systematic Reviews and Research Synthesis (32). The Joanna Briggs Institute assessment tool consists of 11 questions (see Table 2). Each item is appraised as Yes, No, Unclear or Not applicable. Each "Yes" response gains one point, and all the other answers get zero point. Based on the sum of points, the quality of papers was divided into three groups: low quality (0–4), moderate quality (5–8) and high quality (9–11) (45). According to types and the quantity of original research, under the guidance of the JBI Evidence Pre-Grading System (46), evidence was also graded (see Supplementary material 4).

Data extraction and synthesis

JY and LL took charge of data extraction on basis of the predefined criteria (e.g., authors, year, country, purpose, etc.). Then YG and WW checked the extracted content of the above one by one and improved the extracted information. LY was asked in case of disagreement until consensus was reached on all extracts. The synthesis was implemented as the preconcerted plan. Firstly, the first author sought for free codes in the articles involved line by line. Secondly, primary subthemes were raised by integrating these free codes. Thirdly, secondary themes were developed by comparing and analyzing the primary subthemes. Last, all the co-authors discussed and reached the consensus for the following results.

Results

Search outcomes

In total, 1,905 articles were identified. With the help of the reference management program EndNote X7.7, we identified and eliminated 383 duplicates. Two authors independently reviewed the titles and abstracts, those unrelated to our topic and who did not meet the inclusion criteria ($n = 1{,}474$) were excluded, 48 articles were included in the next stage. After the authors reviewed the full-text papers for the suitability, thirty-four non-compliant papers were excluded. Therefore, fourteen review papers were included in the final synthesis. A PRISMA flow diagram was presented in Figure 1. A list of excluded studies with reasons for exclusion can be found in Supplementary material 3.

Characteristics of reviewed articles

Table 1 gave an overview of the included systematic reviews. Four papers were from the United Kingdom, four from the United States, two from Australia, three from China and one from Ireland. The average number of authors was 4.36 ± 2.41. The average number of databases used was 6.36 ± 3.27. The amount of articles included was 20.57 ± 11.99. The systematic reviews included children with intellectual disability, cancer, type 1 diabetes, autism, cerebral palsy, disability, congenital heart disease, and appearance-affecting health conditions. They were published between the years of 2014 and 2022.

Critical assessment and risk of bias in included review papers

Details of the critical assessment and risk of bias in review papers were shown in Table 2. Three review papers were

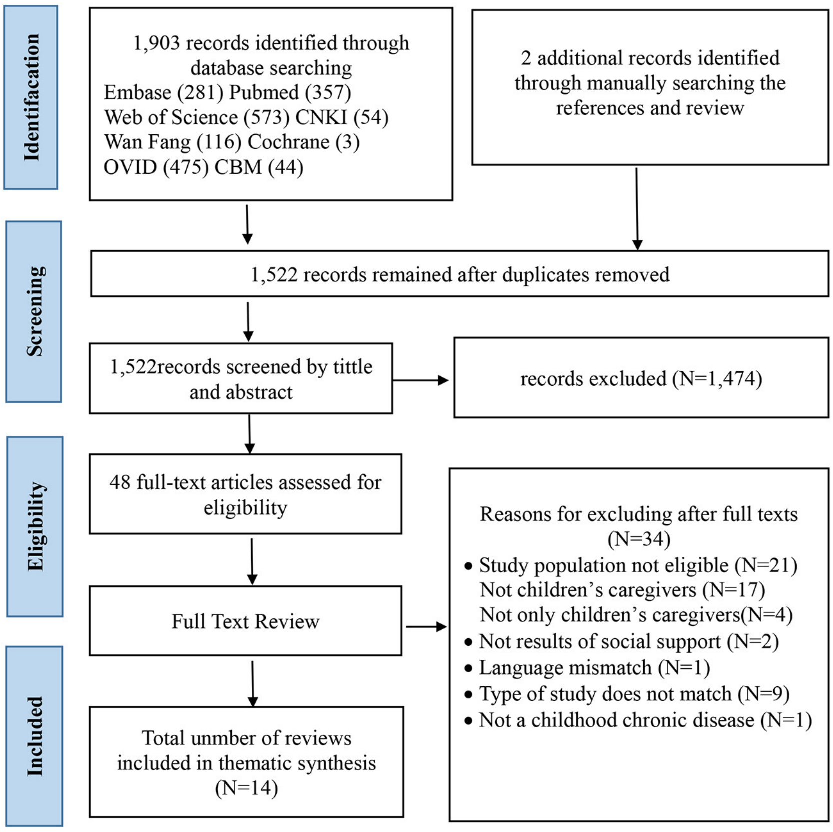

FIGURE 1
PRISMA flow diagram of included studies.

evaluated as medium quality, and eleven of them were graded as high quality.

Results of synthesis

Through repeated reading, analysis and interpretation of 14 reviews, we have raised 26 results, and summarized four themes. Finally, evidence to promote social support for caregivers of children with chronic diseases was identified from four key aspects: (i) Intervention content; (ii) Intervention forms; (iii) Intervention time; and (iv) Sources of support. The evidence graded according to the JBI Evidence Pre-Grading System (46) was shown in Supplementary material 4.

Intervention content

Within the intervention content, we identified three subthemes: (i) Psychoeducation; (ii) Training or education; and (iii) Attitudes and resources.

Psychoeducation

Two meta-analyses explored the effect of psychoeducational interventions on social support (43, 44). According to available evidence, the psychoeducational interventions had no significant effect on social support for caregivers of children with chronic diseases. Specifically, Tang et al. (43) pointed out that the outcome of psychoeducational intervention was not superior to the usual standard of care for social support of caregivers of children with cancer. Standard care mainly referred to routine medical and psychosocial care. Bourke-Taylor et al. (44) also showed no effect of their intervention using psychoeducation approaches on perceived social support for mothers with disabled children.

Training or education

One meta-analysis found that Parenting intervention was shown to be beneficial for parents of children with type 1 diabetes mellitus, Specifically, parenting interventions which include supportive parenting training or education programs could help parents of children with type 1 diabetes mellitus ask for positive social support (30).

TABLE 1 Characteristics of included reviews.

Reference and country	Study design	Including papers (N)	PIO/PICo/PICoS	Search strategy	Theme	Risk of bias	Score	Quality rating
Wilson et al. (34); United Kingdom	Systematic review	$n = 7$	P: Parents with ID I: Interventions to promote social relationships and parenting skills O: Quantitative outcome measures to judge the effectiveness of the intervention	DBs: OVID, psycINFO, EMBASE, ERIC, Medline, MIDRIS, CINAHL, ASSIA IC: In English	To review the effectiveness of interventions to strengthen social relationships and parenting skills	CQA	8/11	MQ
Gise and Cohen (35); United States	Systematic review	n=37	P: Parents of children with cancer I: Phenomenon of social support Co: Family of children with cancer	DBs: PsycINFO, CINHAL, MEDLINE IC: In English between January 2010 and May 2021	To review social support in parents of children with cancer	CQA	10/11	HQ
Costa et al. (29); United Kingdom	Systematic review	$n = 15$	P: Parents and/or guardians of children (<18 years) with AAHC I: A psychosocial intervention C: Compare the intervention group to a control group O: Psychosocial outcomes	DBs: MEDLINE, PsychARTICLES, PsychINFO,CINAHL Plus, BND, CL IC: In English	To review the effectiveness of interventions to improve psychosocial outcomes	CQA	11/11	HQ
Kimbell et al. (36); United Kingdom	Systematic review	$n = 14$	P: Parents caring for a child aged ≤8 years with T1DM I: Views and/or experiences of parents caring for a young child with T1DM Co: Family of children with TIDM S: A primary research using qualitative methods or mixed-methods studies reporting qualitative data separately	DBs: Medline, EMBASE, CINAHL, PsycINFO, WoS IC: In English	To synthesize the qualitative evidence on parents experiences of caring for a child with T1DM to identify: the challenges they encounter; their views about support received; ways in which support could be improved; and directions for future research	CQA	10/11	HQ
Boehm and Carter (37); United States	Systematic review	$n = 52$	P: Parents of individuals with ID or ASD I: Relationship and/or informal relationship Co: Family of individuals with ID or ASD	DBs: ERIC, PsycINFO, SA, SSA IC: In English before July 2014	To review informal relationships of parents and their association with a range of parent and family outcomes	NQA	8/11	MQ

(Continued)

TABLE 1 (Continued)

Reference and country	Study design	Including papers (N)	PIO/PICo/PICoS	Search strategy	Theme	Risk of bias	Score	Quality rating
Nuske et al. (38); United States	Systematic review	$n = 27$	P: Students (<18 years old) with autism spectrum disorder and their parents and teachers O: Strategies for successful student school transition	DBs: PsycINFO, ERIC IC: In English	To review the difficulties that school transitions pose for students with ASD and their parents and teachers, and the strategies used to support during school transition	CQA	10/11	HQ
Rea et al. (39); United States	Systematic review	$n = 21$	P: Parents and/or siblings of children with AAHC I: Therapeutic recreation camp O: Assessment of parent, sibling, or family outcomes	DBs: PubMed, PsycInfo, SportDISCUS, HSN/AE IC: In English between January 2000 and May 2018	To review therapeutic recreation camps impact the parents and siblings of children facing a variety of chronic health conditions.	CQA	9/11	HQ
Zhao et al. (30); China	Meta-analysis	$n = 17$	P: Parents of children or adolescents under 18 years old with T1DM I: Supportive parenting training or education programs O: Psychosocial, family-related and/or sociological outcomes	DBs: PubMed, MEDLINE, EMBASE, CINAHL, CL, WoS IC: In English from January 1978 to October 2018	To synthesize evidence about parenting interventions in parents or caregivers of children and adolescents with T1DM, and to evaluate the effect of interventions	CQA	10/11	HQ
Boshoff et al. (40); Australia	Meta-analysis	$n = 24$	P: Parents, mother, father, carer or caregiver of children diagnosed with ASD I: Experience of advocacy Co: Family of children with ASD S: Qualitative research only	DBs: OVID Medline, OVID Nursing, AACM, EMBASE, PsycINFO, ASP, CINAHL, ERIC, HSN/AE, PBSC, Scopus, WoS, CL, IFC IC: In English	To synthesize the experiences of parents advocating for their child with ASD	CQA	10/11	HQ
Delemere and Maguire (28); Ireland	Systematic review	$n = 16$	P: Family/caregivers affected by pediatric cancer I: Connected health technologied interventions O: All outcomes	DBs: PsychInfo, EMBASE, PubMed, WoS IC: In English within the past 10 years	To summarize the efficacy of Connected Health technologies for families/informal caregivers affected by pediatric cancer	CQA	10/11	HQ

(Continued)

TABLE 1 (Continued)

Reference and country	Study design	Including papers (N)	PIO/PICo/PICoS	Search strategy	Theme	Risk of bias	Score	Quality rating
Lumsden et al. (41); United Kingdom	Meta-synthesis	$n = 22$	P: Parents of Children with CHD I: Coping (manage an emotional, physical, psychological burden) Co: Family of children with CHD S: An empirical study collecting qualitative data	DBs: MEDLINE, CINAHL, PsycINFO, PubMed, ProQuest, WoS IC: No restriction was placed on language or year	To understand parental coping with their child's CHD	CQA	8/11	MQ
Zhang et al. (42); China	Meta-synthesis	$n = 8$	P: Direct caregivers of children with CP (<18 years old) I: The real experience and inner needs of caregivers of children with CP Co: Family of children with CP S: Qualitative research	DBs: PubMed, CL, Embase, WoS, SD, CNKI, CBM, VIP, Wanfang IC: In Chinese or English	To systematically review the caregiving experience of family members	CQA	9/11	HQ
Tang et al. (43); China	Meta-analysis	$n = 11$	P: Caregivers of children (<18 years old) diagnosed with any type of cancer I: PEIs O: Psychosocial and coping outcomes S: RCTs	DBs: Embase, MEDLINE, PsycINFO, CINAHL, Scopus, AMED, JBIEPBD, EBM, BNI, NAHD, ERIC IC: In English	To review the best available evidence to understand the effects of the PEIs on caregivers of children with cancer.	CQA	10/11	HQ
Bourke-Taylor et al. (44); Australia	Meta-analysis	$n = 17$	P: Mothers of children with a disability I: Interventions to improve the mental health O: Wellbeing outcomes S: Quantitative	DBs: OVID Medline, PsychINFO, Embase, Emcare, CCRoCT, CINAHL Plus, Proquest IC: In English	To investigate the effectiveness of interventions that aim to improve the mental health of mothers of children with disabilities.	CQA	10/11	HQ

DBs, databases; IC, inclusion criteria; PICO, population, intervention, comparison/control, outcome; PICoS, population, phenomenon of interest, context, study type; ID, intellectual disability; CQA, Clear quality appraisal; ASD, autism spectrum disorder; WoS, Web of Science; SA, Sociological Abstracts; SSA, Social Services Abstracts; HSN/AE, Health Source Nursing/Academic Edition; AACM, Allied And Complementary Medicine; ASP, Academic Search Premier; PBSC, Psychology and Behavioral Sciences Collection; IHC, Informit Health Collection; SD, Science Direct; JBIEBPD, Joanna Briggs Institute EBP Database; BNI, British Nursing Index; NAHD, Nursing & Allied Health Database; CCRoCT, Cochrane central register of controlled trials; NQA, Non-equality appraisal; T1DM, type 1 diabetes mellitus; RCT, Randomized controlled trial; AAHC, appearance-affecting health conditions; BND, British Nursing Database; CL, Cochrane Library; CHD, Congenital Heart Disease; CP, cerebral palsy; PEI, psychoeducational interventions.

TABLE 2 The critical assessment of included review papers.

Including review paper ($n = 14$)	**JBI critical appraisal checklist systematic review**										
	1	**2**	**3**	**4**	**5**	**6**	**7**	**8**	**9**	**10**	**11**
Wilson et al. (34)	Y	Y	Y	Y	Y	N	N	Y	N	Y	Y
Gise and Cohen (35)	Y	Y	Y	Y	Y	Y	Y	Y	U	Y	Y
Costa et al. (29)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Kimbell et al. (36)	Y	Y	Y	Y	Y	Y	Y	Y	N	Y	Y
Boehm and Carter (37)	Y	Y	Y	Y	N	Y	N	Y	N	Y	Y
Nuske et al. (38)	Y	Y	Y	Y	Y	Y	Y	Y	N	Y	Y
Rea et al. (39)	Y	Y	Y	Y	N	N	Y	Y	Y	Y	Y
Zhao et al. (30)	Y	Y	Y	Y	Y	Y	Y	Y	N	Y	Y
Boshoff et al. (40)	Y	Y	Y	Y	Y	Y	Y	Y	N	Y	Y
Delemere and Maguire (28)	Y	Y	Y	Y	Y	Y	Y	Y	N	Y	Y
Lumsden et al. (41)	Y	Y	Y	Y	Y	Y	N	Y	N	Y	N
Zhang et al. (42)	Y	Y	Y	Y	Y	Y	Y	Y	N	Y	N
Tang et al. (43)	Y	Y	Y	Y	Y	N	Y	Y	Y	Y	Y
Bourke-Taylor et al. (44)	Y	Y	Y	Y	Y	Y	Y	Y	N	Y	Y

Y, Yes; N, No; U, Unclear; NA, Not applicable. 1. Is the review question clearly and explicitly state?; 2. Were the inclusion criteria appropriate for the review question?; 3. Was the search strategy appropriate?; 4. Were the sources and resources used to search for studies adequate?; 5. Were the criteria for appraising studies appropriate?; 6. Was critical appraisal conducted by two or more reviewers independently?; 7. Were there methods to minimize errors in data extraction?; 8. Were the methods used to combine studies appropriate?; 9. Was the likelihood of publication bias assessed?; 10. Were recommendations for policy and/or practice supported by the reported data?; 11. Were the specific directives for new research appropriate?

Attitudes and resources

Two systematic reviews summarized the social support condition of caregivers of children with chronic diseases (35, 37). Parents of children with cancer reported that they needed more social support but sought less social support in the process of caring for their children (35). Additionally, there was evidence that parents' perceptions of support availability might be more important than the level of support actually received. In other words, whether parents felt they had a supportive relationship might be more important than the actual amount of support they received (37).

Intervention forms

Within the intervention forms, we identified three subthemes: (i) Supportive groups; (ii) online; and (iii) Community organizations/teams.

Supportive groups

Two systematic reviews and one meta-analysis synthesized the impact of the group-based interventions on social support for caregivers of children with chronic diseases (34, 39, 44). No consensus had been reached. Bourke-Taylor et al. (44) indicated that group support therapy which relied on peer engagement and group interactions with discussions around support, coping and information sharing had not been shown to increase the perceived level of social support due to insufficient research. Wilson et al. (34) reported the effect of group-based interventions aimed at strengthening social relationships was inconclusive. Only one review of parent-involved therapeutic concentration camps found that parents report camp was a place for providing social support for families of children with chronic health conditions (39).

Online

A systematic review synthesized the role of Connected Health technologies in supporting families affected by pediatric cancer (28). Internet-based health technologies could influence the psychosocial needs of caregivers, provided them with psychosocial support, and reduced the adverse effects of social isolation (28). Another systematic review also identified that support could be provided through networks (38).

Community organizations/teams

A systematic review synthesized coping strategies for caregivers of children with autism spectrum disorder during the new school transitions and concluded that community-based organizations and supportive teams could provide support (38).

Intervention time

A systematic review synthesized early family intervention programs in support of parents with cleft lip and palate (29). Early intervention programs aiming to support parents in adapting to having a child with a disability were implemented when the child was 6, 12, and 18 months, which demonstrated moderate evidence for the effectiveness of increasing perceived spousal emotional support (29).

Sources of support

Within the sources of support, we identified four subthemes: (i) Family members; (ii) Informal people outside the home; (iii) Professionals; and (iv) Faith/spirituality.

Family members

Three Meta-analyses and one systematic review affirmed the role of family members in providing social support (35, 40–42). Family and significant others were the most prevalent sources of support (35). Caregivers expected support from family and society (42). Parents of children with autism spectrum disorder described a strong network of support to enable advocacy, such as partners and extended family (40). For many parents whose children with congenital heart disease, close families, particularly their children's grandparents, became an invaluable source of support to help parents cope with hard times (41). What's more, parents could get emotional and practical support from someone close to them, especially couples (41).

Informal people outside the home

Two Meta- analyses and two systematic reviews integrated informal staff support for caregivers of children with chronic diseases (36, 37, 40, 41). A lot of support, including information support, could be found in informal relationships and emotional support from informal relationships outside the family system was a particularly important resource for parents (37). These informal sources of support mainly include peers, friends and others in the social or school system. While most parents visited some forms of support, they still admitted that others didn't really understand what they were going through unless they had gone through a similar experience themselves, so, parents emphasized that connecting with peers who had the same situation constituted an important source of emotional and practical support (36). Many problems could be solved, and their heads kept clear with the help of peers (41). Friends could be as validators, sounding boards and observers with constructive support (40). Social and school system support is available in rural communities (40).

Professionals

A meta-synthesis affirmed the role of professionals among parents of children with congenital heart disease (41). Parents reported that the honesty, reassurance and information which professionals provided helped them to understand their children's condition more, and in turn cope better with what they faced as a family (41).

Faith/spirituality

Spiritual support comes mainly from people of faith. Parents turned to faith, religion and often prayer to call upon a "higher power" for support, and felt comforted when procedures were successful, attributing this to divine intervention (41).

Discussion

Based on the review and thematic synthesis of the included review papers ($n = 14$), we have identified four key aspects of improving social support for caregivers of children with chronic diseases: intervention content, intervention forms, intervention time, and the sources of support.

Intervention content

Summarizing current evidence, we divided the intervention content of social support for caregivers of children with chronic diseases into three main aspects: psychoeducation, training or education, and attitudes and resources. Despite this, the intervention content of improving social support is still relatively limited. The Psychoeducational intervention is a non-pharmacological approach that involves information giving and receiving, concerns about emotions, psychological needs and family relationships (47, 48). Bourke-Taylor et al. (44) and Tang et al. (43) pointed out that psychoeducational interventions cannot play a role in improving the social support of caregivers of children with cancer or disability. In a study of caregivers of patients with lung cancer, psychological education interventions also showed no effect on the level of social support for caregivers (49). In other research, psychoeducational interventions can increase caregivers' knowledge of diseases, strengthen their stress coping skills, improve psychological outcomes and make better quality of life (49, 50). As you can see, the effectiveness of psychoeducational interventions have been demonstrated in many of the above areas. However, the role of psychoeducational interventions on social support remains to be further explored, and extensive original research is still needed. Training or education could help increase the level of social support (30). Specifically, it focuses on helping parents of children with chronic diseases feel positively supported and providing them with practical parenting guidance, available information and resources. It is worth noting that, caregivers represent they rarely actively seek social support, although they acknowledge that having available social connections is more important than actual support (35, 37). To a certain extent, this means that caregivers of children with chronic diseases have not established a positive attitude to deal with the current difficulties, while only limited to passively accepting existing assistance. This is even more dangerous in areas with inadequate medical resources, because there are not enough personnel to identify families in need. If the family does not actively seek help and does not actively use the existing social resources, the family will encounter more difficulties. They will also feel lonelier and more isolated. Therefore, it is necessary to add how to actively seek social support in the content of future interventions to guide caregivers to use intentional relationships and actively seek available resources to get social support.

Intervention forms

Intervention forms of social support for caregivers of children with chronic diseases were integrated into three types: group, online and community organizations/teams. Support groups refer to approaches that relied on peer engagement and group interactions with discussions around supports, coping and information sharing (34, 39, 44). Although existing evidence is not yet consistent that group intervention has a positive impact on social support for caregivers of children with chronic disease (34, 39, 44). However, previous research showed support groups can expand and strengthen social support networks for disabled elderly caregivers (51). The form of group intervention can greatly unite caregivers of children with chronic diseases and promote their participation and interaction. Many studies have confirmed that group support therapy can help caregivers to solve psychological burdens and adapt to the disease experience, and at the same time, caregivers have improved social functions, physical pain, general health, emotional health, role limitations caused by emotional problems (52, 53). In summary, we have reason to think that the group intervention method is still worthy of promotion in clinical practice. The online form refers to ways that rely on the Internet, through smartphone apps, online web pages, telemedicine and online groups (28). The biggest advantage of this form is that it is not affected by distance, allowing a wider range of families to participate. The online intervention focuses on providing information on disease management resulting in a significant reduction in parental anxiety and uncertainty and an increase in social functioning and knowledge (21, 54). Compared with the group intervention form, the online form is more convenient and faster. Caregivers of children with chronic diseases can obtain more comprehensive information resources according to their needs, but the real-time interaction and practice are weaker than the group form. It cannot be ignored that most interventions for caregivers of children with chronic diseases usually take 3 to 6 months (55). With such a long-term intervention, it is a challenge for many caregivers to ensure that they can participate on time. In this situation, considering the large daily burden of caregivers of children with chronic diseases, the energy and time spent on taking care of their children, and the actual effect that they want to achieve, it will be a good choice in the future to integrate these two forms. For example, in the first stage, online system learning and guidance are carried out with the help of connected health technologies. Meanwhile, problems and difficulties encountered by caregivers of children with chronic diseases are collected. The second stage is to carry out offline practice in groups, organize the learning and application of various parenting skills in the form of entertainment activities, and gather caregivers in similar situations. They can exchange experiences and provide each other with the required resources. Parenting issues or psychological confusions faced by caregivers can be guided face-to-face. This hybrid approach may be a choice for more caregivers of children with chronic diseases in the future, and it is also a good form for social workers and healthcare professionals to carry out interventions.

Intervention time

Early family intervention improves perceived spousal emotional support for caregivers of children with chronic diseases. The "Early" is the stage when caregivers of children with chronic diseases frequently experience various problems, such as psychological maladaptation, frequent anxiety and depression. Interventions for caregivers at an early stage also have positive implications for promoting adaptation, reducing threats, increasing confidence, and reducing emotional distress, anxiety, and depression (29). Early help is more like timely rain. Therefore, we advocate early intervention for caregivers of children with chronic diseases to promote the perception of spouse support, and to help them establish an orderly parenting state, a stable and positive attitude, and positive coping skills.

Sources of support

The results of our umbrella review also suggest that caregivers of children with chronic diseases can receive support from family members, friends, peers, community, self-belief, and professionals (35–38, 40–42), which is approximately consistent with the composition of the perceived social support scale. In general, informal people are an important source of social support for caregivers of children with chronic illness, especially family members and peers. The family is the living unit of the caregivers of children with chronic diseases. A close family relationship is a favorable way to relieve negative emotions and provide confidence and support. When caregivers of children with chronic diseases are faced with stressful events, support from family members can greatly relieve the pressure of parenting and economics (56–58). As special family members, partners have more contact with caregivers, and their positive emotional feedback and support are favorable factors for caregivers to cope positively (59, 60). Therefore, we suggest that partners of caregivers of children with chronic diseases should actively participate in parenting, maintain a stable mood, encourage and communicate with each other, and establish a close relationship of dependence. In addition to family members, available informal relationships outside of the home system are also a good source of support. Peers are one important part of it. It's worth mentioning that while most parents caring for children with chronic diseases access some sources of support, they still admit that others don't really understand what they're going through unless they've gone through a similar experience themselves (41). This suggests that peer support is more meaningful for caregivers. These people with similar experiences of diseases or psychosomatic conditions help each other in social and emotional aspects, which can give each other more encouragement and confidence to overcome difficulties together (61, 62). Support provided by these informal groups tends to be emotional, and it is more about improving the mentality of caregivers, and providing spiritual dependence and confidence. We recommend providing psychological counseling for family members, especially couples. When patients receive treatment, medical institutions can provide them with communication platforms among peers.

Limitations

Some limitations of this umbrella review need to be considered. Firstly, this review did not utilize all available databases (e.g., CINAHL, MEDLINE). Therefore, some evidence may have been ignored. Secondly, only articles published in English and Chinese were included, which might have led to selection bias as articles published in different languages were not considered. Thirdly, although we followed published guidelines for systematic reviews (31), we did not register our search protocol prior to the start of the review. Fourthly, quality assessments were performed using the Joanna Briggs Institute, a useful tool, although its reliability could be improved by additional assessment of the methodological quality of included studies (63). Finally, there is still a lack of research on psychoeducational interventions and group interventions for caregivers of children with chronic diseases, and their role in promoting the level of social support for caregivers of children with chronic diseases is not clear enough. At the same time, due to the strong heterogeneity of the included studies, there is not enough quantitative data for quantitative integration.

Implications for practice

The findings of the umbrella review showed that four main aspects can be taken to optimize social support for caregivers of children with chronic diseases in clinical practice. Healthcare professionals and social workers should enhance their expertise to provide available resources and health guidance for caregivers. It is also extremely important to help caregivers of children with chronic diseases identify available support relationships, especially family members and peers. This means that it is meaningful to establish a good family relationship and create a harmonious family atmosphere, and it is necessary to implement psychological counseling between the couple to establish a good supportive relationship. We encourage medical institutions and interveners to intervene early in families of children with chronic diseases, integrate diverse and effective intervention content, and use different forms at different time periods to help the caregivers of children with chronic diseases for the greatest benefit.

Conclusion

Social support plays an important role in improving mental health of caregivers of children with chronic diseases and in promoting active parenting. The findings of this umbrella review suggest that a combination of effective and diverse intervention content and forms to improve social support for caregivers of children with chronic diseases is recommended. In general, it is pivotal to follow the four aspects on how to improve social support for caregivers of children with chronic diseases, which include the content, forms, timing and the sources of social support. Specifically, the use of a combination of differing interventions, especially for early family, including content of parenting training or education, attitude building and resource provision, which can implement online, are most effective aiming at improving social support for caregivers of children with chronic diseases. Nonetheless, evidence for increasing the level of social support is still limited, and original interventional research and quantitative evidence integration for caregivers of children with chronic diseases is still needed.

Data availability statement

The original contributions presented in the study are included in the article/Supplementary material, further inquiries can be directed to the corresponding author.

Author contributions

JY: conceptualization, formal analysis, methodology, investigation, writing—original draft, and writing—review and editing. LL: conceptualization, methodology, and writing—original draft. YG: supervision, resources, and writing—review and editing. WW: methodology, resources, investigation, and writing—review and editing. LY: conceptualization, methodology, visualization, investigation, formal analysis, funding acquisition, project administration, and writing—review and editing. All authors contributed to the article and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2022.973012/full#supplementary-material

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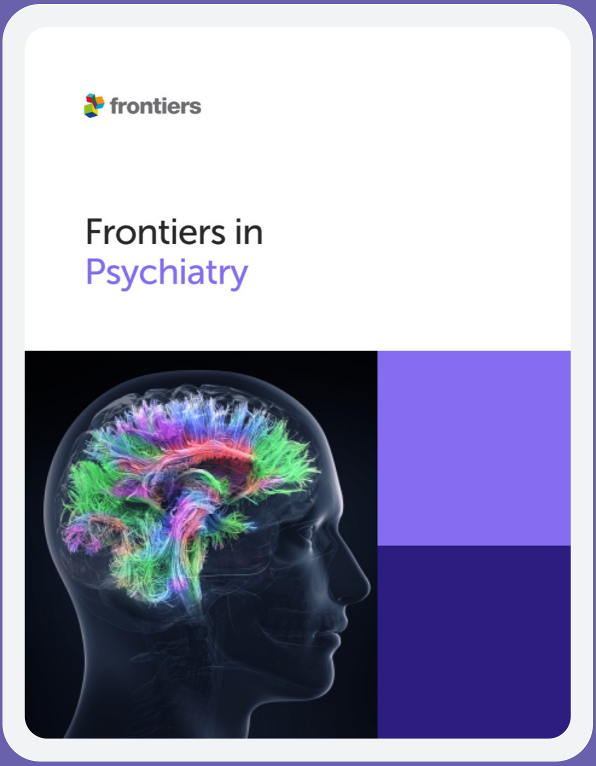

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