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Original Article

Parenting stress and depressive symptoms in the family caregivers of children with genetic or rare diseases: The mediation effects of coping strategies and self-esteem

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Abstract

Objective: To elucidate how parenting stress influence depressive symptoms in the family caregivers of children with genetic or rare diseases by examining the mediation effects of coping strategies (problem-focused, emotion-focused, and dysfunctional coping) and self-esteem. **Materials and Methods:** In total, 100 family caregivers were recruited and administered a questionnaire assessing demographics and study measures. We used the PROCESS for SPSS macro with 10,000 bootstrapped samples and a 95% confidence interval to test the proposed mediation models. **Results:** Increased parenting stress was associated with more depressive symptoms, partially via dysfunctional coping, but not problem- or emotion-focused coping strategies. The serial multiple mediation pathway (parenting stress → low self-esteem → dysfunctional coping → depressive symptoms) was not significant, whereas the indirect effect of via dysfunctional coping alone had a significantly partial mediation effect. **Conclusions:** Dysfunctional coping strategies may explain the parenting stress–depressive symptom relationship. The goals of psychosocial medical care for family caregivers were suggested.

Keywords: *Coping strategies, Depressive symptoms, Genetic or rare diseases, Parenting stress, Self-esteem*

Introduction

Genetic disease is a group disorder with genetic problems caused by whole or partly abnormal change in the DNA sequence or chromosome abnormalities which typically inherit from family [1]. A genetic disease may not be a rare disease. A rare disease is defined as a disease that affects fewer than 1 in 10,000 individuals in Taiwan [2]. There are 223 rare diseases, of which up to 80% are genetic disorders. Most genetic diseases and rare diseases, such as Down syndrome, congenital adrenal hyperplasia, epidermolysis bullosa, and tuberous sclerosis, are concerned with heritability issues, complex pathology, various chronically symptoms, most early onset in childhood, and even no cure [3,4]. Family caregivers of children with genetic diseases or rare diseases face significant caring burden, especially for those children with developmental delay or defected organ functions. Past studies have noted that family caregivers have high parental stress and depressive symptoms [5,6] and lack of psychosocial supports [7]. Parenting stressors of caring a child with a chronical illness are multidimensional challenges, including communication, medical cares, emotional functioning, and role constraints [8]. How caregivers cope with these patenting stressors may not only affect their health, but also children's adjustment and well-being [9]. It is important to elucidate the relationship between parenting stressors and depressive symptoms for suggesting effective stress management for these family caregivers in medical care.

Coping is an ongoing process with constantly changing cognitive and behavioral efforts to meet with various stressful situations [10]. People usually use mixed types of coping strategies depending on the controllability of situation and induced different emotional outcomes. There are two major coping efforts served to decrease emotional distress. When the stressful situation is appraised controllable, problem-focused strategies (efforts to directly change a stressful situation, such as planning) are dominated. When the stressful situation is unable to change, the emotion-focused strategies (efforts to regulate stressor-triggered distress, such

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as acceptance) are occupied [11]. Some emotion-focused coping strategies mean to disengage or avoid the stressor, such as venting, may be useful for managing stressful responses immediately, but gradually become dysfunctional in reducing distress and may create negative mood or problems in the long term [12]. It is not clear how parenting stressors that family caregivers experienced influence the types of coping adopted which, in turn, affect the level of depressive symptoms.

Self-esteem refers to overall subjective evaluation of the self [13]. Self-esteem is one of the relative stable intrapsychic resources to affect coping process [14]. People with high self-esteem might evaluate stress as controllable and implement more efficient and less avoidant coping strategies against adverse health outcome [15]. However, chronically caring stress may decrease caregivers' sense of self-worth [16]. The self-esteem of family caregivers may also be defeated by stigmatization of their child with a rare genetic condition [17] and the subtle social discrimination caused as a problematic genetic passenger [18]. There may have an additive effect of combining low self-esteem with high using dysfunctional coping strategies on raising family caregivers' depressive symptoms.

Because few studies explain the mechanism between parenting stress and depressive symptoms in the family caregivers of children with genetic or rare diseases, the current study aims to examine the potential mediators of three coping strategies (i.e., problem-focused, emotion-focused, and dysfunctional coping) and self-esteem. We hypothesize: (1) there is a significant relationship between parenting stress, three coping strategies, self-esteem, and depressive symptoms; (2) three coping strategies mediate the parenting stress–depressive symptom relationship [conceptualized in Figure 1]; and (3) significant serial multiple mediation pathways are presented: parenting stress → low self-esteem → dysfunctional coping → depressive symptoms [conceptualized in Figure 2].

Materials and methods

Participants and procedures

This study was approved by the hospital's institutional review board (IRB103-127-B). In total, 100 family caregivers were recruited by a senior attending pediatrician at the center of genetic counseling of a medical center in East Taiwan. All participants completed a questionnaire after signing informed consent.

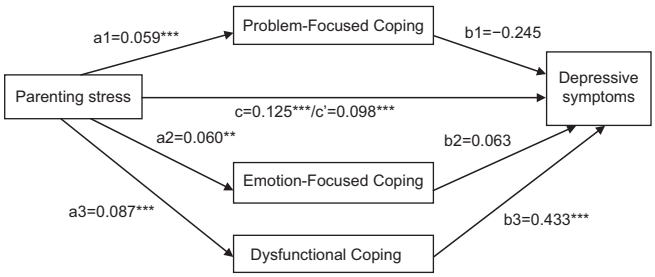

Figure 1: Parallel multiple mediator model showing the total effect (c), the direct effect (c'), and the specific indirect effects through the three coping strategies (a1*b1, a2*b2, and a3*b3). Path values represent unstandardized regression coefficients. $^{**}P < 0.01$, $^{***}P < 0.001$

One-hundred family caregivers aged between 19 and 79 years (mean = 43.4 years, standard deviation [SD] = 11.6) provided care for an average of 8.7 years (SD = 5.8). Most of the caregivers are women (58%), mothers (53%), married (84%), had senior high education (47%), employed (67%), and healthy (82%). Their 77 children with a mean age of 9.9 years (SD = 5.9) has been received a diagnosis (rare diseases with genetic problems, n = 33 [2]; genetic diseases, n = 44) averagely for 8.4 years (SD = 5.6). The diagnoses include 16 rare disease types and 16 genetic disease types.

Measures

A questionnaire was designed to assess demographic data, caring experiences, and four study measures described as follows.

Pediatric Inventory for Parents

The Pediatric Inventory for Parents (PIP) is a 42-item measure using a 5-point scale to evaluate the frequency and difficulty of caring events for children with serious illnesses in the last week in the following four domains: communication, medical cares, emotional functioning, and role constraints [8]. The PIP demonstrated acceptable internal consistency (α = 0.80–0.96) and validity. The present study used the PIP-frequency scale to evaluate the occurrence of caring events with high score indicating more parenting stress appraised cognitively.

Brief Cope

The brief COPE consists of 14 subscales to measure different coping strategies responding to stress by a 4-point scale and scored into the following three coping strategies: (1) problem-focused (active coping, instrumental support, and planning); (2) emotion-focused (acceptance, emotional support, humor, positive reframing, and religion); and (3) dysfunctional (behavioral disengagement, denial, self-distraction, self-blame, substance use, and venting) [19]. The brief COPE demonstrated adequate internal consistency (α = 0.72–0.84), concurrent criterion, and convergent validity.

Rosenberg Self-Esteem Scale

The Rosenberg Self-Esteem Scale (RSES) is a 10-item scale using a 4-point scale to measure a person's overall sense of worthiness. The RSES demonstrated adequate internal consistency (α = 0.81) and high factor loading [20]. High score indicates high self-esteem.

Center for Epidemiological Studies Depression Scale Short Form

The Center for Epidemiological Studies Depression Scale (CES-D) Short Form is a 10-item revised version assessing

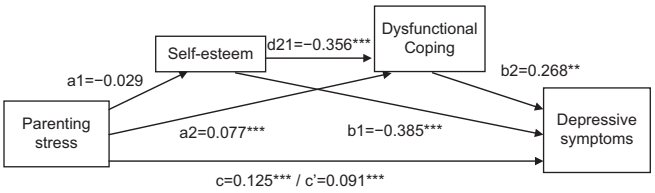

Figure 2: Serial multiple mediator model showing the total effect (c), the direct effect (c'), and the specific indirect effects through self-esteem and dysfunctional coping (a1*b1, a1*d21*b2, and a2*b2). Path values represent unstandardized regression coefficients. $^{**}P < 0.01$, $^{***}P < 0.001$

depressive symptoms in the past week [21]. The CES-D Short Form achieved satisfied internal consistency (α = 0.78–0.87) and validity. High score indicates more depressive symptoms.

Data analysis

The relationships among the study variables were analyzed through the two-tailed Pearson's product correlation coefficients.

To ensure the mediation hypothesis tests as valid as possible, we decided to control for two demographic variables (age and gender). The first reason was that younger age and female gender are related risk factors of major depressive disorder [22]. Female gender was also a risk factor for caregiver burden [23]. The second reason was that age had a significantly negative correlation with depressive symptoms in this study [Table 1]. By using independent *t*-test, we also found that there were significant gender differences in PIP-frequency (t (98) = −3.34, $P < 0.01$), problem-focused coping (t (98) = −2.09, $P < 0.05$), emotion-focused coping (t [98] = −2.22, $P < 0.05$), dysfunctional coping (t [98] = −2.96, $P < 0.01$), and CES-D (Short Form) (t [98] = −3.65, $P < 0.00$), but not in self-esteem (t [98] = 1.48, $P > 0.05$). Female caregivers had higher parenting stress, more using three coping strategies and more depressive symptoms.

The proposed mediation effects were conducted using models 4 (the parallel multiple mediator model) and 6 (the serial multiple mediator model) provided by PROCESS for SPSS macro after controlling for age and gender [24]. In Figures 1 and 2, path c is the total effect of predictor on criterion through mediators. Path c' is the direct effect of predictor on criterion controlling for mediators. Path ab or adb is the indirect effect of mediators. Path estimates were calculated by Ordinary Least Squares (OLS) regression. The indirect effects were tested by bootstrapped estimation based on 10,000 resampling and 95% confidence interval (CI). When the 95% CI estimates do not include zero, the indirect effects were significant. All data were computed using IBM SPSS Statistics 20.0 (IBM Corp., Armonk, NY, USA).

Results

Correlations among study variables

Table 1 shows that parenting stress had a significantly positive correlation with depressive symptoms and three coping strategies, whereas it had a significantly negative correlation with self-esteem. The three coping strategies exhibited a significantly positive correlation with one another and also with depressive symptoms. Self-esteem had a significantly negative correlation with dysfunctional coping and depressive symptoms.

Mediation analysis

A parallel multiple mediator model [Figure 1] showed that the total (c) and direct (c') effects of parenting stress on depressive symptoms were significant (B = 0.125 and 0.098, standard error [SE] = 0.018 and 0.019, respectively; P = 0.000 for both). By using 10,000 bootstrapped samples, a point estimate of the total indirect effects of three coping strategies was significant (B = 0.027, SE = 0.013, 95% [CI] = 0.005 to 0.054). The indirect effects of the parenting stress on depressive systems via problem-focused coping (a1*b1) (B = −0.014, SE = 0.011, 95% [CI] = −0.041 to 0.004) and emotion-focused coping (a2*b2) (B = 0.004, SE = 0.010, 95% [CI] = −0.012 to 0.028) were not significant. Only dysfunctional coping (a3*b3) was a significant partial mediator (B = 0.038, SE = 0.013, 95% [CI] = 0.016 to 0.066).

According to Baron and Kenney's criterions [25] and literature deduction, the correlation results supported the significant associations between parenting stress, self-esteem, dysfunctional coping, and depressive symptoms. We further tested the hypothesis 3 by the serial multiple mediator models [Figure 2] and have showed that the total (c) and direct (c') effects of parenting stress on depressive symptoms were significant (B = 0.125 and 0.091, SE = 0.018 and 0.017, respectively; P = 0.000 for both). When 10,000 bootstrapped samples were used, a point estimate of the total indirect effect was significant (B = 0.034, SE = 0.013, 95% [CI] = 0.012 to 0.062). The indirect effects of the parenting stress on depressive systems via self-esteem (a1*b1) (B = 0.011, SE = 0.009, 95% [CI] = 0.000 to 0.036) and of via self-esteem to dysfunctional coping (a1*d21*b2) (B = 0.003, SE = 0.002, 95% [CI] = 0.000 to 0.009) were not significant. Only the indirect effects of the parenting stress on depressive systems via dysfunctional coping (a2*b2) (B = 0.021; SE = 0.011; 95% [CI] = 0.004 to 0.048) were significant, and the pairwise comparison of a1*d21*b2 minus a2*b2 (B = −0.018, SE = 0.011, 95% [CI] = −0.046 to −0.002) was significant.

Discussion

The present study explored the mediation effects of three coping strategies and self-esteem on the parenting stress–depressive symptom relationship in the family caregivers of children with genetic or rare diseases. As

Table 1: Correlations between age and study variables

	Mean±SD	1	2	3	4	5	6	7
PIP-frequency	86.5±28.6	-						
Problem-focused coping	15.2±4.0	0.45**	-					
Emotion-focused coping	23.8±5.5	0.37**	0.78**	-				
Dysfunctional coping	21.4±5.4	0.51**	0.48**	0.52**	-			
Self-esteem	29.1±4.6	−0.21*	0.50	−0.00	−0.41**	-		
CES-D (Short Form)	7.8±6.2	0.63**	0.30**	0.32**	0.60**	−0.49**	-	
Age	43.4±11.6	−0.09	−0.10	−0.11	−0.10	0.12	−0.20*	-

P values are for two-tailed tests. **P<0.01, *P<0.05. PIP: Pediatric Inventory for Parents, CES-D: Center for Epidemiological Studies-Depression, SD: Standard deviation

hypothesized, increased parenting stress was significantly associated with more depressive symptoms. This result was in consistent with past studies [5,6] and indicated that caring for children with genetic or rare diseases may impact caregivers' mental health quality. Under this situation, caregivers also exhibited higher use of three coping strategies to manage their emotional distress. The parallel mediation analysis results showed that only the dysfunctional coping was a significantly partial mediator. This result indicates that if family caregivers choose to apply dysfunctional coping strategies, such as behavioral disengagement, denial, self-distraction, self-blame, substance use, or venting, to orient away from the highly frequent and uncontrollable parenting stress, they would experience increased depressive symptoms. This result is partially in line with the similar research among frontotemporal dementia caregivers that caregivers' use of dysfunctional coping as a response to their strain increased the levels of depression [26].

Contrary to our hypothesis, the study result showed that higher use of the problem- or emotional-focused coping strategies increased depressive symptom and did not play a significant role in lessening depression among the present sample. A partial explanation for this is that no matter how to use coping efforts to change or adapt to parenting stress, family remained with depressive symptoms. There might be other possible proximal mediating determinants to adjust to chronical disease such as interpersonal processes, personality attributes, or cognitive appraisals [27]. Another explanation is that using the problem- or emotional-focused coping strategies might not help to decrease depressive symptom, but benefit other health outcomes. The previous mediation studies about coping strategies and coping outcomes for caregivers were mixed. For example, the study for caregivers of people with Alzheimer's disease indicated that three coping strategies did not mediate the relationship between care burden and depression, but using more problem-focused and fewer emotion-focused coping strategies can reduce caregivers' anxiety a year later [28]. The study for frontotemporal dementia caregivers also pointed out that caregivers' use of problem-focused coping strategies increased their quality of life [27]. A meta-analysis study showed that active coping, planning, seeking instrumental social support, seeking social support for emotional reasons, positive reinterpretation and growth, acceptance, and turning to religion were significantly correlated with a high level of well-being [29].

The second focus of this study was to investigate whether self-esteem may influence on the parenting stress-coping process. As hypothesized, low self-esteem was associated with high parenting stress, dysfunctional coping strategies, and more depressive symptoms. The serial multiple mediation analysis results showed that only the dysfunctional coping was a significant partial mediator. The path of via self-esteem or via self-esteem to dysfunctional coping has no significant partial mediation effect. Contrary to our hypothesis, there is not an additive effect of combining low self-esteem with using dysfunctional coping strategies on raising family caregivers' depressive symptoms. This result suggested that coping resources (i.e., self-esteem) did not affect the stress-coping process among the present sample. Even though past studies indicated that the influence of coping resources on coping processes remained mixed, coping process is a more proximal mediator of the stress-health outcomes relationship [14]. The dysfunctional coping strategies are likely to provide a more practical and instant target of stress management for family caregivers.

These findings imply that psychosocial intervention in medical cares might screen out those family caregivers who experience high parenting stress with more depressive symptoms and simultaneously use dysfunctional coping strategies. Along with providing them caring information and family support networks [3], stress managements are provided with how to deal with the uncontrollable caring natures of diseases, replacing more efficient problem-solving techniques, and emotional-regulation skills such as mindfulness strategies [30].

This study has some limitations. First, because of the limited population of children with genetic or rare diseases, the present sample size was small (n = 100), and participants were only recruited from a genetic counseling center. Therefore, the generalization of study results should be cautioned. Second, the present study applied a cross-sectional study design recruiting 100 family caregivers of 77 children diagnosed separately with 16 rare disease types and 16 genetic disease types. The parenting stress–depressive symptom relationship might be confounded with disease course or disease severity according to different disease entities. Third, even though this study has controlled demographic variables (age and gender), there may have other distal contributors to adjustment to chronical disease, such as caregivers' socioeconomic backgrounds, culture/ethnicity, and gender-related processes [27]. Finally, the study results only demonstrated the partial mediation effect of dysfunctional coping strategies. Other factors also may contribute to the parenting stress–depressive symptom relationship.

Future studies should recruit larger sample sizes from different genetic counseling centers according to the rare incidence of each genetic disease and rare disease. Longitudinal design could be considered for exploring the changes of the parenting stress–depressive symptom relationship and coping strategies across the different stages of disease course and severity. Various coping outcomes might be included. This study found significant gender differences in some study variables and younger caregivers may have higher depressive symptoms. Future study might focus on the effect of caregivers' demographic variables on parenting stress-coping process in addition to controlling them as confounding variables. Other mediators could be considered. Dispositional optimism or finding meaning may contribute to disease-related adjustment [31].

This study indicates that increased parenting stress is associated with higher depressive symptoms in the family caregivers of children with genetic or rare diseases. Dysfunctional coping is a mechanism for the relationship.

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Conflicts of interest

There are no conflicts of interest.

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