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ORIGINAL RESEARCH

Analysis of Influencing Factors of Family Resilience in Children with Congenital Tibial Pseudoarthrosis

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Aim: To investigate the status of the family resilience score of caregivers of children with congenital pseudarthrosis of the tibia (CPT) and analyse its influencing factors to provide a reference and basis for clinical formulation of a family resilience intervention programme.

Methods: A total of 379 families of children with CPT were included in this study, which used a combination questionnaire to investigate the families of children with CPT. The researchers sent a link to the questionnaire to all eligible subjects to conduct a survey on the characteristics of families of children with CPT. Single factor analysis was used to compare the total scores of family resilience. Multiple linear regression was used to determine meaningful variables for single factor analysis.

Results: The average family resilience score was 151.79 ± 30.79, the lowest score was 20 and the highest score was 231. The CPT usually occurred in unilateral limbs (37.63%). The payment methods of medical expenses were through a rural cooperative medical system (44.85%). The caregivers were mostly the mothers (62.80%) and caregivers were 31–40 years old (62.27%). Most caregivers were farmers (30.34%). The average monthly income of the families in the past year was 1000–5000 yuan (54.09%). There were significant differences in family resilience scores between caregivers' working conditions ($P = 0.039$) and average monthly incomes in the past year ($P = 0.008$). The working conditions of caregivers and the average monthly income of families in the past year were the main factors affecting the family resilience of caregivers of children with CPT.

Conclusion: The caregivers' working conditions and the average monthly income of families were the influencing factors in the family resilience score.

Keywords: congenital pseudarthrosis of the tibia, family resilience, family characteristics, influencing factor

Introduction

Congenital pseudarthrosis of the tibia (CPT) is a tibial deformity caused by developmental abnormalities, characterised by tibial angular deformity, medullary cavity stenosis or cysts and ultimately the formation of non-union of the pseudo-joint.[1] It is more common in the lower third of the tibia, and its pathogenesis is still unclear.[2] The data show the incidence is about 0.004‰–0.007‰ in China.[3,4] Tibial pseudarthrosis is prone to a long-term recovery after surgery and therefore is also a refractory disease. Due to the long treatment time of the disease, it has serious impacts on children's health and quality of life and requires considerable support from caregivers. It not only increases families' economic burden but also increases the psychological burden on caregivers. The physical and psychological status of caregivers directly affects the quality of care and, therefore, the rehabilitation of the children.

The heavy burden of caring for children with chronic diseases often leads to a decline in the quality of life of family members and the mental health of the whole family. This affects children's physical and mental health and has an important impact on children's psychological behaviour and health outcomes. With increasing attention to family health, Walsh proposed the concept of family resilience. Family resilience is the ability of families to help themselves become

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stronger or use potential resources when facing crises or challenges. It was shown that the family resilience level has a positive impact on the psychological resilience level of patients.[5] The key processes in three areas of family resilience are family belief systems, organisational models, and communication and problem solving. Good family functioning is conducive to promoting the recovery of patients and improving their quality of life. At present, there are few studies on the family resilience of caregivers of children with CPT in China. This study intends to investigate the status of the family resilience score of such caregivers and analyse its influencing factors to provide a reference and basis for clinical formulation of a family resilience intervention programme for caregivers of children with CPT.

Patients and Methods

Study Population

In this study, children with CPT and their caregivers treated and followed up in the Children's Hospital of Hunan Province were selected as subjects by convenience sampling. The inclusion criteria were: (1) main caregivers of children diagnosed with CPT by the hospital; (2) child caregiver age > 18 years; (3) caregivers have basic communication skills; (4) caregivers provided informed consent with the voluntary questionnaire survey. The exclusion criteria were: (1) caregivers of children with a history of mental illness or whose cognitive behaviour was abnormal; (2) failure to complete the study; (3) Neurofibromatosis-1-related CPT. A total of 379 families with CPT were included in this study, which has been approved by the Ethics Committee of Hunan Children's Hospital.

Research Scale

The research was based on the relevant literature at home and abroad and combined with the actual situation of children with CPT in China. This study used a combination questionnaire to survey the families of children with CPT to gather information about the children and caregivers. The family resilience scale, Zarit Care Burden Index (ZBI) scale, Post-Traumatic Growth Inventory (PTGI) scale, Pittsburgh Sleep Quality Index (PSQI) scale, perceived stress scale (PSS), CPT caregivers' care needs scale, follow-up needs scale, needle care methods and knowledge of nine parts were all used.

The Chinese Family Resilience Assessment Scale was originally developed by Sixhey. Chaoqun et al made the scale Chinese in 2018.[6] The scale contains 4 dimensions and 44 items. Each item uses a 1–4 grade scoring method. The higher the score, the better the family resilience and the overall Cronbach coefficient. The coefficient is 0.96, and the Cronbach α coefficient of each dimension is 0.70–0.97.

The ZBI scale invented by Zarit et al in the 1980s measures the burden on caregivers. At present, the scale has been translated into many languages and is widely used in many countries. It has 22 items, including two dimensions: role burden and personal burden. Each item is scored at 0–4 points and five levels according to the weight of the burden, where 0 points indicates "never" and 4 points indicates "often". The maximum score is 88. The higher the score, the heavier the burden on caregivers. The study demonstrated that Cronbach's α coefficient showed very good internal consistency (α = 0.81). There was a correlation between all ZBI-12 items and the PSS total score (r = 0.53, P < 0.01).[7]

The PTGI scale was developed by Tedeschi, an American scholar, in 1996. The scale includes 5 dimensions and 21 items. The Likert rating method is adopted. From "no such change after trauma" to "very big change after trauma", the scores were 0–5 points, and the score range was 0–105 points. The higher the score, the higher the level of post-traumatic growth.[8] The total Cronbach α coefficient of the scale was 0.890, and the Cronbach α coefficients of each subscale were 0.848–0.893.

The PSQI was compiled by Dr Buysse, a psychiatrist at the University of Pittsburgh in the United States in 1989. The scale is suitable for evaluating sleep quality in patients with sleep disorders and mental disorders and for evaluating sleep quality generally. It consists of 19 self-assessment items and 5 other assessment items, with 18 items consisting of 7 components. Each component is scored according to 0–3 levels, and the cumulative score of each component is divided into the PSQI total score. The total score range is 0–21. The higher the score, the worse the sleep quality.[9]

The CPT care needs scale, follow-up needs scale, needle care methods and knowledge were developed according to the actual family situation of the child.

Implementation of Survey and Quality Control

After the completion of the questionnaire, the researchers sent a link to all eligible subjects through the WeChat group for parents of children with CPT to conduct a questionnaire survey. At the beginning of the questionnaire, there were unified instructions to explain the purpose, significance and completion instructions. The subjects took the voluntary questionnaire to decide whether to participate in the study. The researchers required all items to be answered to ensure the integrity of the questionnaire. To prevent bias, researchers used unified guidance to explain the completion requirements, and when there were questions, they used consistent language to explain. After the survey, the staff checked the collected questionnaires one by one, then excluded the questionnaires that did not meet the requirements. A total of 419 questionnaires were distributed, including 379 valid questionnaires, and the effective recovery rate was 90.45%.

Statistical Analysis

The SPSS™ Statistics software (version 23.0; IBM Corporation, Armonk, NY, USA) was used to conduct data analysis. Measurement data subject to normal distribution were described using mean ± standard deviation. Measurement data were described by frequency. An independent sample *t*-test or one-way analysis of variance was used to compare the total scores of family resilience of caregivers of children with CPT with different characteristics. The influencing factors of family resilience of caregivers of children with CPT were analysed by multiple linear regression. $P < 0.05$ indicated that the difference was statistically significant.

Results

Family Characteristics of Children with CPT

A total of 379 children with CPT were enrolled in this study, including 236 boys and 143 girls, with an average age of 9.23 ± 3.21 years and an age range from 1 to 23 years. The CPT mostly occurred in unilateral limbs (37.63%). The payment methods of medical expenses were through a rural cooperative medical system (44.85%). The caregivers were mostly the mothers (62.80%). Caregivers were 31–40 years old (62.27%). Most caregivers were farmers (30.34%). Most caregivers had junior high school education (37.20%). The average monthly income of the families in the past year was 1000–5000 yuan (54.09%) (Table 1). The family resilience score was 151.79 ± 30.79, the lowest score was 20 and the highest score was 231.

Family Resilience Score Comparison

The 379 families of children with CPT were classified according to the relationship between the caregivers and the children, the number of children in the family, the age range of the caregivers, the occupation of the caregivers, the average monthly income of the families in the past year, and the payment method of medical expenses. The family resilience scores were compared. The results of single factor analysis showed there was no significant difference in the scores of family resilience among different payment methods of medical expenses, the relationship between different caregivers and children, the number of children in the families or the age range of the caregivers. There were significant differences in family resilience scores between different caregivers' working conditions and different average monthly incomes in the past year. The family resilience score of caregivers who were unemployed or retired was lower than that of caregivers who were working. With an increase in family average monthly income, family resilience scores decreased (Table 2).

Multiple Linear Regression Analysis

Multiple linear regression analysis was conducted with the family resilience score as the dependent variable and two variables with statistical significance in single factor analysis (caregivers' working conditions and average monthly household income in the past year) as independent variables. When analysing caregivers' working conditions, taking unemployment or retirement as a reference, other occupations were classified as working or farming. The results of multivariate regression analysis showed that the fitting degree of the model was R = 0.327,

Table 1 Family Characteristics of Caregivers of Children with CPT

Characteristics		N	Ratio
Sex	Male	236	62.27%
	Female	143	37.73%
Location of CPT	Unilateral Limb	370	97.63%
	Bilateral Limbs	9	2.37%
Medical payment	Private Expense	140	36.94%
	Rural cooperative medical system	170	44.85%
	Urban medical insurance	69	18.21%
	Commercial insurance	0	0.00%
Patient caregiver	Father	123	32.45%
	Mother	238	62.80%
	The others	18	4.75%
Number of children	1	145	38.26%
	2	197	51.98%
	3 or more	37	9.76%
Age of caregivers (years)	≤20	1	0.26%
	21~30	62	16.36%
	31~40	236	62.27%
	41~50	69	18.21%
	51~60	9	2.37%
	61~70	2	0.53%
Education background of caregivers	Primary school	15	3.96%
	Junior high	141	37.20%
	High or secondary school	80	21.11%
	College for professional training	71	18.73%
	Bachelor	66	17.41%
	Master and above	6	1.58%
Religious belief	Having faith	23	6.07%
	Unbelief	356	93.93%
Caregiver occupation	Enterprise and institution	70	18.47%
	Individual operator	43	11.35%
	Transportation personnel	2	0.53%
	Medical personnel	11	2.90%
	Business service personnel	9	2.37%
	Educators	19	5.01%
	Retirees	3	0.79%
	Farmers	115	30.34%
	Other	53	13.98%
	Standby	54	14.25%
Average monthly household income in recent year (RMB)	≤999	29	7.65%
	1000~3000	102	26.91%
	3001~5000	103	27.18%
	5001~7000	53	13.98%
	7001~9000	27	7.12%
	9001~10,000	24	6.33%
	>10,000	41	10.82%

and the working conditions of caregivers and the average monthly income of families in the past year were the main factors affecting the family resilience of caregivers of children with CPT, which explained 24.31% of the total variation (Table 3).

Table 2 Comparison of Family Resilience Scores Among Caregivers of Children with CPT with Different Characteristics

	N	The Family Resilience Score (Mean ± Standard Deviation)	t/F	P
Medical payment				
Private expense	140	152.81±31.86		
Rural cooperative medical system	170	150.68±31.72		
Urban medical insurance	69	153.57±30.79		
Patient caregiver			2.381	0.094
Father	123	156.76±28.53		
Mother	238	149.38±31.63		
The others	18	153.83±32.05		
Number of children			0.013	0.987
1	145	152.10±31.07		
2	197	151.79±30.44		
3 or more	37	152.62±32.36		
Age of caregivers (years)			1.549	0.174
≤30	63	151.85±34.80		
31~40	236	153.32±28.53		
41~50	69	144.99±32.74		
≥51	11	164.22±32.53		
Caregiver Occupation			2.846	0.039
On-the-job	322	152.55±31.48		
Unemployment or retirement	57	146.81±26.59		
Average monthly earnings			2.932	0.008
≤999	29	157.34±32.70		
1000~3000	102	156.11±26.76		
3001~5000	103	154.93±28.19		
5001~7000	53	152.96±34.33		
7001~9000	27	144.11±33.82		
9001~10,000	24	149.96±23.01		
≥10,000	41	135.68±37.31		

Discussion

In this study, the results showed that the working conditions of caregivers and the average monthly income of families in the past year were the main factors affecting the family resilience of caregivers of children with CPT. The long treatment time for CPT seriously affects the health and quality of life of the children and needs follow-up management after discharge. Studies have shown that follow-up after discharge can guarantee that the basic needs of patients are met.[10] Follow-up can help patients to establish a support system and can provide necessary psychological or medical support to promote the rehabilitation of patients.[11] In addition, follow-up services can effectively improve patients' compliance

Table 3 Multiple Linear Regression Results of Family with CPT

	Nonstandard Coefficient	Standard Error	Standard Coefficient	t	P	95% CI
Constant	174.26	12.07		14.435	0.000	150.52~198.001
Caregivers'working conditions						
Working	2.86	2.13	0.18	2.181	0.007	1.270~5.005
Farming	1.76	1.12	0.22	1.179	0.031	1.299~2.975
Average monthly household income in the past year	−4.297	1.108	−0.25	−3.879	0.000	−6.475~-2.119

behaviour, control risk factors and play a role in secondary prevention. It was found that children with CPT had the risk of refracture before bone maturation and should be followed up until bone maturation.[12]

The caregivers in this study participated in the whole process of the patients' surgery. They understood and accepted the surgery and postoperative nursing needs. Nursing knowledge is very helpful to caregivers. Previous investigations found that needle care methods and knowledge were needed by caregivers of children with CPT. Therefore, using targeted solutions for caregivers of children with CPT difficulties, meeting the needs of the caregivers in the care process and establishing a long-term effective follow-up mechanism increased the compliance of the families and helped children recover.

This study showed that the family resilience score of 379 caregivers of children with CPT was 151.79 ± 30.79, the lowest score was 20 and the highest score was 231. The resilience score from the survey was at a normal level. The reasons may be as follows: (1) The treatment of children with CPT is a long-term process.[13,14] It requires many operations and the postoperative wearing of an external fixator for a long time. The illness reoccurs easily, infection is common and caregivers have concerns about the prognosis of children with psychological problems; (2) Care for children with CPT requires substantial human and material resources from the family, the costs of surgery and treatment are high, postoperative recovery is uncertain and the need to respond to possible complications and refractures may hinder the formation of family resilience; (3) Related studies have found that with long-term surgery, an unfamiliar environment, long-term postoperative breaking and pain and many other factors, children who have CPT surgery have serious medical fears and strong physiological and psychological stress reactions, which seriously affects their safety, the surgical efficacy and postoperative rehabilitation.[15]

Studies suggest that male caregivers' resilience scores were significantly lower than those of female caregivers.[16] In this study, the relationship between the caregivers and the children was not a factor affecting family resilience score. This may be related to the fact that, with the progress of society, women take more responsibilities and are more likely to choose positive coping styles. The results of this study show that the age of the caregivers was not an important factor affecting family resilience. No matter what the relationship between the caregivers and the children is, we should strengthen the communication with the caregivers, manage any mental health issues in a timely manner, give more support to the caregivers, and actively guide them in facing up to the children's condition, so as to avoid affecting the physical recovery and mental health of the children.

There were significant differences in family resilience scores between different caregivers' working conditions and different average monthly incomes in the past year. Children with CPT require costly treatment, which has a heavy economic burden. Therefore, the better the family economic situation, the greater the ability to take risks; and the stronger the psychological endurance of the family members, the stronger the family resilience. The treatment time of children with CPT is long, and their caregivers need to spend considerable time and energy, so the caregivers' working conditions were also an influencing factor of the family resilience score.

There are some limitations in this study. For example, whether the family resilience level of the caregivers of children with CPT is related to the occurrence of needle tract infection in children still needs further clinical research. Other caregivers of children with CPT include grandparents, uncles and aunts. The level of family resilience may be related to the physical condition, age and length of care of children, a notion that also needs further study. The patient's self-centred nature may make the survey results subjective. The patient's exaggerated cognition of their own ability may lead to high tolerance to the disease, thus showing the illusion of good recovery, which will bias the results.

Data Sharing Statement

The datesets used or analyzed during the current study are available from the corresponding author on reasonable request.

Ethics Approval and Consent to Participate

This study was conducted in accordance with the Declaration of Helsinki and approved by the ethics committee of Hunan Children's Hospital. We obtained signed informed consent from the participants in this study.

Author Contributions

All authors contributed to data analysis, drafting or revising the article, have agreed on the journal to which the article was submitted, gave final approval of the version to be published, and agree to be accountable for all aspects of the work.

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Disclosure

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