INTRODUCTION

Congenital pseudarthrosis of the tibia (CPT) is a deformity of the tibia caused by dysplasia that is manifested as an angular deformity of the tibia, medullary cavity stenosis, or cyst. It eventually forms a non-union false joint. CPT is one of the most challenging and refractory diseases in pediatric orthopedics. Although the surgical treatment of CPT has developed rapidly in recent years, late complications remain, such as ankle valgus, unequal length of limbs, proximal tibial valgus, and re-fractures. Long-term multiple operations and complicated treatment procedures make it easy for the child to develop psychological and behavioral problems. In recent years, more attention has been paid to the psychological and behavioral problems of children caused by CPT. The behavioral problems that appear early in the process of the growth of children are relatively stable. Early detection and early intervention can often get twice the result with half the effort. Some studies have shown that the mental health status of parents or caregivers of children with CPT is generally poor, mainly reflected in obsessive-compulsive disorder, anxiety, fear, psychosis, and so on. However, there are few reports on whether the mental state of the child’s parents or caregivers has any influence on the child’s mental and behavioral problems.

This study investigated patients over 4 years of age with CPT and explored the relationship between children’s psychological behavior problems and parents’ or guardians’ psychological emotions. It is expected to provide a basis and reference for the early comprehensive intervention of psychological and behavioral problems in children with CPT.

METHODS

Research object

This study selected 90 children with CPT, who were treated in Hunan Children’s Hospital from August 2014 to August 2015, and their caregivers, as the research subjects. This study complied with the “Declaration of Helsinki of the World Medical Association” and was approved by the ethics committee of the hospital. All patients or their caregivers signed an informed consent form.
Inclusion and exclusion criteria
Inclusion criteria of this study were patients who
1. were diagnosed with CPT;
2. aged 4–18 years; and
3. voluntarily signed informed consent,

and exclusion criteria were patients with
1. other chronic diseases;
2. mental illness; and
3. incomplete case data.

Research methods
The general information about the child (gender, age, and course of illness) was collected using self-made questionnaires. The behavioral problems of the children were assessed using the Achenbach Children’s Behavior Scale, including social competence and behavioral problems. The scores of each dimension of the children’s behavior scale were used in comparison with the national norm. When the score of a certain dimension was greater than the national norm, it was considered an abnormality in that dimension. Children with abnormal scores in any dimension were defined as having behavioral problems. Parents’ emotional problems were investigated using the Self-Rating Depression Scale (SDS) and the Self-Rating Anxiety Scale (SAS). The score was compared with the national norm. The anxiety scale was divided into 50 points: 50–59 points indicated mild anxiety, 60–69 points indicated moderate anxiety, and 69 or greater points indicated severe anxiety. The demarcation of the depression scale was divided into 53 points: 53–62 points indicated mild anxiety, 63–72 points indicated moderate anxiety, and 72 or greater points indicated severe anxiety.

Questionnaire survey method
The filling results were screened by the staff, and invalid questionnaires were removed. A total of 90 questionnaires were distributed and 90 were collected, of which 90 were valid questionnaires. The efficiency was 100%.

Statistical processing
After the questionnaires were collected, all raw data were imported into SAS 9.4 for analysis. The measurement data obey normal distribution and are described by mean ± standard deviation. The measurement data are described by frequency and constituent ratio. The Student’s t-test was used for the comparison between groups that satisfied the normal distribution. The non-parametric test was used for the comparison among groups that did not satisfy the normal distribution. Logical regression analysis was used to analyze the influencing factors of behavioral problems in children with CPT. The chi-square test was used for counting data. The p-value <0.05 indicated that the difference was statistically significant.

RESULTS
General
A total of 90 children were enrolled in this study, including 58 male children and 32 female children (aged 4–16 years) with an average age of 7.10±2.58 years, an average treatment course of 5.67±2.47 years, and an average number of operations of 1.68±1.41. The main caregivers were 22–52 years old, with an average age of 33.3±5.1 years. The caregivers enrolled in this study included 33 fathers (36.7%), 55 mothers (61.1%), and 2 other relatives (2.2%). The details are shown in Table 1.

Table 1. General situation of the research subjects.

|                          | n  | %  |
|--------------------------|----|----|
| **Gender**               |    |    |
| Male                     | 58 | 64.4 |
| Female                   | 32 | 35.6 |
| **Age (years)**          |    |    |
| 4–7                      | 44 | 48.9 |
| 7–16                     | 46 | 51.1 |
| **Primary caregiver**    |    |    |
| Father                   | 33 | 36.7 |
| Mother                   | 55 | 61.1 |
| Other                    | 2  | 2.2 |
| **Parental education**   |    |    |
| Junior high school and below | 38 | 42.2 |
| High school, technical school | 18 | 20.0 |
| College degree and above | 31 | 34.4 |
| **Number of operations** |    |    |
| Have not had surgery     | 11 | 12.2 |
| 1                        | 62 | 68.9 |
| ≥2                       | 17 | 18.9 |
| **Course of disease (years)** |   |    |
| <4                       | 16 | 18.2 |
| 4–8                      | 47 | 52.2 |
| >8 years                 | 25 | 28.4 |
| **Guardian anxiety symptoms** |   |    |
| No                       | 12 | 13.3 |
| Yes                      | 78 | 86.7 |
| **Guardian depressive symptoms** |   |    |
| No                       | 42 | 46.7 |
| Yes                      | 48 | 53.3 |
The current status of behavioral problems of children with congenital pseudarthrosis of the tibia

The results of this study indicate that the detection rate of behavioral problems of children diagnosed with CPT was 53.3% (48/90). The four areas of problems were thinking (24.4%), violation of discipline (16.7%), social interaction (15.6%), and aggressive (12.2%). The abnormality rate of these four dimensions was high.

Abnormal Self-Rating Anxiety Scale and Self-Rating Depression Scale scores of parents or guardians

The results of this study showed that 41 of the investigated parents had symptoms of depression and 12 had symptoms of anxiety. All had mild-to-moderate anxiety or depression.

Comparison of Self-Rating Depression Scale and Self-Rating Anxiety Scale scores between parents of the abnormal behavior group and the normal behavior group

The results of this study showed that the SDS and SAS scores of the caregivers were 52.73±11.54 and 44.87±9.03, respectively, in the children's abnormal behavior group and 46.85±11.85 and 38.78±5.97, respectively, in the children's normal behavior group. The statistical results showed that the scores for anxiety and depression were statistically higher (p<0.05) in the abnormal behavior group than in the normal group. All of the parents of children with behavioral problems had higher levels of anxiety and depression than the parents of children with no behavioral problems.

Single-factor analysis of influencing factors of behavioral problems in children

The results of this study showed that there was no statistically significant difference in the incidence of behavioral problems among different gender, ages, ethnicities, length of illness, number of operations, parental education, monthly family income, and depression scale scores. There was a statistically significant difference in the incidence of abnormal behavioral problems between the abnormal parental anxiety scale group and the normal group. The incidence of abnormal behavior in children in the abnormal score group was higher than that in the normal score group. According to clinical observations and experience, the possible influencing factors for the occurrence of abnormal behavior in children included gender, age, number of operations, duration of illness, parents’ educational level, parents’ anxiety, and depression. The above variables were taken as independent variables, and whether or not the child had behavioral problems was used as dependent variables. The logistic regression analysis method was set to be Backward, probability for stepwise (Entry 0.05, Removal 0.10). The results showed that none of the variables entered the equation, and the OR value of the two variables (the SAS and SDS scores) tended to be positive infinity, so the model was not stable. Considering that there may be multicollinearity between the two variables, the SDS score was excluded, and the SAS score was successfully introduced into the equation. The results showed that parental anxiety had a significant impact on the behavior of the children. The more anxious the parents were, the more likely children were to have behavioral problems, as presented in Tables 2 and 3.

DISCUSSION

CPT has a long course of the disease, is difficult to treat, requires repeated operations, is of high cost, and patients need to be followed up until the bone is fully mature. Therefore, long-term, multiple operations and complex treatment processes make the family members of children prone to physical and mental health problems. This study found that the total detection rate of behavioral problems in 90 children with CPT was 53.3%. Among these behavioral problems, the abnormal rate in the four dimensions of thinking, violation of discipline, social interaction, and aggression was relatively high. The reasons for this could include three aspects. First, the social aspect: children with CPT had physical disorders. Abnormal bones at the prosthetic joints, fractures were easily caused by external forces and were not easy to heal, and the affected limbs needed to be protected by plaster or protective gear for a long period of time during walking until the bones matured. In daily life and at school, they were susceptible to ridicule and rejection from their peers. Studies show that neglected children were less active in all aspects, and rejected children were most likely to have aggressive and disruptive behaviors. Second, the family aspect: overprotection and spoiling were positively correlated with the behavioral problems of children. CPT is a chronic disease. More parents overprotected and took care of their children by ignoring the child's adaptability and need for social skills development. It was easy to make the children infantile, overdependent, and produce an inferiority complex. Third, the child's own aspect: to avoid fractures, children with CPT could not play sports or play games with their partners. Their activities were restricted, and this was not understood by younger children who could only express their inner distress by crying, quarreling, and tantrums.

There was a statistically significant difference in the incidence of abnormal behavior in children between the abnormal anxiety scale score group and the normal group. CPT is a rare, chronic surgical disease that is difficult to treat. The caregiver needs to take care of the child full time and pay attention to protect the child at all times. The characteristics of this disease, its treatment methods, and the family care methods
Behavioral problems in children with CPT

have a high degree of similarity so that the influences of different genders, ages, lengths of illness, number of operations, family monthly income, parents’ education, etc. are concealed.

Previous studies have shown that the psychological response of parents is an important factor affecting the behavior of children with chronic diseases. This study used the anxiety scale and the child behavior scale for the first time to verify that caregiver anxiety was a risk factor for behavioral problems in children with CPT. The reason could be that caregivers of children with CPT who were in a state of maladjustment and anxiety lacked confidence in the future of the child, ignored the child's adaptability and advantages, and demonstrated a negative attitude (such as a sad face and a sigh). The anxiety problem had the characteristics of intergenerational transmission, i.e., the parents’ anxiety traits affect the anxiety level of the children through parenting methods. In turn, this would make the children feel depressed, cause them to pay too much attention to their own image and appearance, and prevent them from forming a positive self-concept. These children were unable to actively recognize and evaluate their own condition and value, thereby generating more behavioral problems.

In this study, the detection rate of behavioral problems in children was higher. Parents of children with behavioral problems were more anxious and depressed, and children with anxious caregivers were more likely to have behavioral problems. This issue should be taken seriously by medical workers and measures should be implemented. Medical workers themselves should insist on looking at children and their families from the perspective of positive psychology, fully assess the potential advantages of their families, share more cases of good recovery with parents, encourage family members to build confidence, redefine the meaning of illness, and maintain effective communication and close cooperation to help them overcome the difficulty of the impact of illness on the family. Medical workers should also insist on treating the children with an attitude of appreciation, paying attention to their strengths, interests, abilities, and knowledge, and inspiring the confidence of the children to struggle in adversity. In addition, parents of children with CPT should overcome adverse emotions such as anxiety, maintain family unity, and avoid excessive protection. Families with children diagnosed with CPT have high medical expenditures. This may be due to rare diseases and poor treatment effects in the past. At present, they are not included in any assistance. The vast majority of mothers take care of their children full time to protect

Table 2. Multifactor analysis of influencing factors of CPT children’s abnormal behavior.

|                      | Behavior problem detection rate (%) | \( \chi^2 \) | p-value |
|----------------------|-------------------------------------|------------|--------|
| Gender               |                                     |            |        |
| Male                 | 50                                  | 0.527      | 0.508  |
| Female               | 59.4                                |            |        |
| Age (years)          |                                     |            |        |
| 4-7                  | 50                                  | 1.875      | 0.392  |
| 7-12                 | 60                                  |            |        |
| 12-16                | 33.3                                |            |        |
| Nationality          |                                     |            |        |
| Han nationality      | 52.4                                | 0.862      | 0.719  |
| Minority             | 62.5                                |            |        |
| Whether to go to school |                                 |            |        |
| Yes                  | 55.6                                | 0.417      | 0.339  |
| No                   | 48.1                                |            |        |
| Sick time (years)    |                                     |            |        |
| 0–4 years            | 50                                  | 0.144      | 0.930  |
| 4–8 years            | 54.2                                |            |        |
| 8–12 years           | 56                                  |            |        |
| Number of operations |                                     |            |        |
| 0                    | 54.5                                | 2.650      | 0.266  |
| 1                    | 48.4                                |            |        |
| ≥2                   | 70.6                                |            |        |
| Parents’ education   |                                     |            |        |
| Elementary to Junior High |                                 | 3.15      | 0.207  |
| High school and equivalent |                             |    |        |
| University and above | 52.9                                |            |        |
| Family monthly income|                                     |            |        |
| 1000–3000            | 51.6                                | 0.325      | 0.850  |
| 3000–8000            | 52.3                                |            |        |
| >8000                | 60                                  |            |        |
| Parental anxiety score|                                   |            |        |
| Normal               | 47.4                                | 8.510      | 0.014  |
| Abnormal             | 92.0                                |            |        |
| Parent depression scale score |                       |            |        |
| Normal               | 44.0                                | 3.451      | 0.178  |
| Abnormal             | 62.0                                |            |        |

Table 3. Logistic regression results.

|                      | Partial regression coefficient | Wald \( \chi^2 \) value | Degree of freedom | p-value | OR | 95%CI |
|----------------------|-------------------------------|--------------------------|------------------|---------|----|------|
| Constant             | 20.204                        | 3.842                    | 1                | 0.048   | -  | -    |
| SAS analysis         | 2.104                         | 3.770                    | 1                | 0.052   | 8.200 | 0.98-68.59 |
their children from accidental fractures, so their family income is single. Society should provide more financial support for children with CPT and their families. At the same time, more active attention and publicity should be given to children with CPT. This should include the encouragement of interaction and contact with other children who are also diagnosed with CPT, the presence of a fair and just social environment for children with CPT, and the promotion of mental health for children with CPT.

This is the first study to explore the influence of the mental state of parents or caregivers on the behavior of children with CPT, which is novel and innovative. Moreover, this research has shortcomings and limitations. This study is not a randomized controlled experiment, so there is a certain risk of bias. This study is a single-center clinical study, and subsequent multicenter clinical studies are needed for further discussion. Finally, the sample size included in this study is relatively small; hence, it is necessary to increase the sample size in future research.

CONCLUSION

Children with CPT have psychological and behavioral problems, and their parents’ anxiety and depression levels are significantly higher than those of parents with children who do not have CPT. In addition, the anxiety and depressive state of mind of parents or caregivers had a significant impact on the behavior of children with CPT.

AUTHORS’ CONTRIBUTIONS

JHX: Conceptualization, Writing – original draft, Writing – review & editing. HBM: Data curation, Writing – original draft, Writing – review & editing. KL: Data curation, Writing – original draft, Writing – review & editing. GHZ: Formal Analysis, Writing – original draft, Writing – review & editing. YQO: Formal Analysis, Writing – original draft, Writing – review & editing.

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