

The invisible patients: posttraumatic stress disorder in parents of individuals with cystic fibrosis

Os pacientes invisíveis: transtorno de estresse pós-traumático em pais de pacientes com fibrose cística

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Abstract

Background: Besides the growing acknowledgment of the relevance of posttraumatic stress disorder (PTSD) related to medical illness, there is no study in cystic fibrosis yet. **Objective:** To assess the prevalence of PTSD and the three clusters of posttraumatic stress symptoms (PTSS) in parents of patients with cystic fibrosis. **Methods:** Parents of patients with cystic fibrosis (age range: 2 to 33 years) were drawn from the Cystic Fibrosis Association of the city of Rio de Janeiro. In this cross-sectional study, parents were asked to fulfill a questionnaire for social and demographic characteristics and were interviewed by means of the PTSD module of the Structured Clinical Interview for DSM-IV. **Results:** The sample was comprised 62 subjects (46 mothers and 16 fathers). Current prevalence for full PTSD was 6.5% and that for partial PTSD was 19.4%. Parents with and without PTSS differed significantly in two psychosocial aspects: the former reported more emotional problems ($p = 0.001$); and acknowledged more often the need for psychological or psychiatric interventions ($p = 0.002$) than the latter. However, only 6.3% of the parents with PTSS were in psychological/psychiatric treatment. **Discussion:** This preliminary study showed that the frequency of PTSD symptoms is fairly high among parents of patients with cystic fibrosis, and although these parents recognize they have emotional problems and need psychological/psychiatric treatment, their suffering remains “invisible” to the medical system, leading to underdiagnosis and undertreatment.

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Key-words: Cystic fibrosis, parents, prevalence, posttraumatic stress disorder, posttraumatic stress symptoms.

Resumo

Contexto: Apesar do crescente reconhecimento da relevância do transtorno de estresse pós-traumático (TEPT) secundário a doenças médicas, ainda não existem estudos em fibrose cística. **Objetivo:** Verificar a prevalência de TEPT e dos três grupos de sintomas de estresse pós-traumático em pais de pacientes com fibrose cística. **Métodos:** Pais de pacientes com fibrose cística (idade média: 2 a 33 anos) foram recrutados da Associação Carioca de Mucoviscidose. Neste estudo transversal, os pais preencheram um questionário sociodemográfico e foram entrevistados por meio do módulo de TEPT do Structured Clinical Interview for DSM-IV. **Resultados:** A amostra era composta de 62 indivíduos (46 mães e 16 pais). A prevalência atual de TEPT foi 6,5% e de TEPT parcial, de 19,4%. Os pais com e sem sintomas de TEPT diferiram significativamente em dois aspectos psicossociais: os primeiros relataram mais problemas emocionais ($p = 0,001$) e reconheceram mais frequentemente a necessidade de tratamento psiquiátrico ou psicológico ($p = 0,002$) que os últimos. Entretanto, somente 6,3% dos pais com sintomas de TEPT estavam em tratamento psiquiátrico/psicológico. **Conclusões:** Este estudo preliminar demonstrou que a frequência dos sintomas de TEPT é bem elevada em pais de pacientes com fibrose cística e, apesar de esses pais reconhecerem que tem problemas emocionais e precisam de tratamento psiquiátrico/psicológico, seu sofrimento permanece invisível para o sistema médico, levando ao subdiagnóstico e ao subtratamento.

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Palavras-chave: Fibrose cística, pais, prevalência, transtorno de estresse pós-traumático, sintomas de estresse pós-traumático.

Introduction

Posttraumatic stress disorder (PTSD) is a usually chronic condition that is often associated with marked functional impairment and decreased quality of life. It is characterized by the presence of three symptoms clusters (reexperiencing, avoidance and hyperarousal) that occur in response to a traumatic event¹. In a Canadian community sample, current PTSD prevalence was found to be 2.7% for women and 1.2% for men². Lifetime PTSD prevalence in the general population was estimated to be 7.8%³. Existing evidence suggests that partial or subclinical PTSD (PTSD symptoms that fall short of meeting full criteria) is not only more common than full PTSD in the general population but is also accompanied by significant disability².

In DSM-III-R, a traumatic event was defined as an occurrence outside the range of normal human experience (e.g., war, natural catastrophes, and rape). In consequence, medical diseases, irrespective of their seriousness and consequences, would not qualify as traumatic events. Only with the advent of the DSM-IV was the knowledge that one's child has a life-threatening illness recognized as a qualifying traumatic event for PTSD. Since then, several studies have demonstrated that posttraumatic stress symptoms (PTSS) are frequent among parents of children with severe diseases⁴⁻¹⁰. In a recent meta-analysis¹¹, the pooled prevalence of PTSD in parents of children with chronic medical illnesses (cancer, diabetes mellitus type 1, epilepsy and asthma) or undergoing high-risk procedures (bone marrow or solid organ transplantation) was estimated in 22.8% (95% CI: 16.4%-29%).

The presence of posttraumatic stress symptoms in parents may compromise their role as caregivers and can bear serious repercussions for sick children. For example, parents with hypervigilant symptoms can overburden health services by requesting frequent doctors' visits or by making an inordinate number of phone calls to health personnel¹². Furthermore, avoidant symptoms, such as excessive anxiety about doctors' visits or medical procedures, can lead to non-adherence to medical treatment¹³.

Cystic fibrosis (CF), a progressive illness that reduces life expectancy, is the most common lethal autosomal recessive disease in Caucasians, with an incidence of 1 in 2,500 births¹⁴. It is a major cause of pulmonary and gastrointestinal morbidity in children and a leading cause of death in early adulthood¹⁵. However, the prognosis of CF has improved steadily over the last 20 years and the mean life expectancy has now reached 30-40 years¹⁶. Thus, families have to adjust and learn to cope with the stresses of taking care of children or adolescents with CF for considerably longer periods.

Traumatic events could be classified as type 1 trauma (a single traumatic event) or type 2 trauma (repeated traumatic events)¹⁷. Parents of children with CF are exposed to several types of potentially traumatic events during the course of this chronic disease. They are likely to be exposed to repeated threats by witnessing their child undergoing painful procedures, recurrent hospitalization, and colonization of the airways with antibiotic-resistant bacteria. Nowadays, the majority of patients' deaths are due to respiratory failure and one of the greatest potentially traumatic events related to CF is lung transplantation. There is growing acknowledgment of the traumatic impact of transplantation and posttransplant medical care on families of patients with CF. Besides the transplant inherent risk, the patients and their caregivers face the necessity of life-long adherence to immunosuppressive medication, the development of many side effects, and invasive procedures to prevent and manage organ rejection¹⁸.

CF is a chronic and life-threatening medical condition that, besides its dire effects on the patient him/herself, frightens and overburdens the parents, being an important source of traumatic experiences. The aim of this study was twofold: 1) to estimate the prevalence of full and partial PTSD and of its three symptomatic clusters in a sample of parents of patients with CF; and 2) to compare the demographic and psychosocial features of parents with and without PTSS, as well as the clinical status of their offspring with CF.

Method

Measures

A specifically designed questionnaire was employed to assess 1) the socio-demographic characteristics (e.g., marital status, ethnicity, family income, educational attainment, and occupation) and 2) the mental health status (e.g., history of psychological or psychiatric treatment, history of suicide ideation or of suicide attempts) of parents of the patients with CF; and 3) to obtain information regarding the medical condition of the patients themselves (e.g., age of the patient at the time of the diagnosis, number of hospitalizations during the last year, presence of pancreatic insufficiency and colonization/infection of airway by *Pseudomonas aeruginosa*, *Burkholderia cepacia* and/or *Methicillin resistant Staphylococcus aureus* – MRSA).

The PTSD module of the Structured Clinical Interview for DSM-IV SCID was used to assess current and lifetime prevalence of full and partial PTSD. The SCID is a semi-structured diagnostic interview designed for the assessment of mental disorders based on DSM criteria. Partial PTSD was defined by the presence of two out of the three PTSD symptoms clusters (reexperience, avoidance and arousal) in addition to the A, E and F criteria for PTSD (the individual's response of fear, horror, helplessness; duration of the disturbance more than 1 month; and the presence of clinically significant distress or functioning impairment, respectively). Lifetime PTSD was defined as the presence of full PTSD at any time since the diagnosis of CF.

Recruitment and assessment of participants

The Cystic Fibrosis Association of the city of Rio de Janeiro, Brazil (ACAM) is a local chapter of the Brazilian National Alliance for Cystic Fibrosis, and provides assistance to patients with CF and their families. The main missions of the local chapters are to increase public awareness about CF and to improve the quality of life of those with the disease. To be eligible to participate in the study, the parents of patients with CF had to be living with them and be responsible for their care. Parents with more than one offspring with CF were asked to answer all the questions focusing on the patient they worried more about. Informed consent was obtained from all participants. The main reasons given by parents for not completing the questionnaire were lack of time or distress triggered by the questions.

A psychiatrist with clinical experience in PTSD (MC) and a medical student serving her internship in psychiatry (CR) conducted the SCID interviews. Both researchers were trained in the SCID interview by a senior researcher with extensive experience in the use of this instrument (IF). All SCID-PTSD questions were specifically directed to the worst moment experienced by the parents regarding the CF since the diagnosis was made. All SCID-PTSD items were asked, even when parents failed to endorse the criterion A2; this was done in order to provide an estimate of the prevalence of each posttraumatic symptom in the whole sample. The protocol of the study was submitted and approved by the Institutional Review Board of the Institute of Psychiatry of the Universidade Federal do Rio de Janeiro and by the executive board of the local chapter of the Brazilian National Alliance for Cystic Fibrosis.

Analysis

Parents meeting diagnostic criteria for full current and partial PTSD were combined to form the PTSS group. Then, distributions of the demographic, psychosocial and clinical condition-related variables were calculated for the groups with and without PTSS. Two-sided chi-square and Fisher exact tests were used for the categorical and two-sided Mann-Whitney test for the continuous variables. Significance was set up at a p value ≤ 0.05 . Data were analyzed using the Statistical Package for the Social Sciences for Windows (10.0 SPSS version).

Results

Participants' characteristics

The 93 parents of the 165 patients who had attended regularly the ACAM from July 2005 to August 2006 were approached by a research assistant who explained the goals and methods of the study. Two parents were excluded because they were unable to communicate effectively with the researchers (one due to a serious hearing loss and the other due to a mild mental retardation). Two parents refused to participate in the study without any further explanation. The remaining 89 parents were requested to fill out the demographic, psychosocial, and clinical questionnaire. Sixty-seven parents (75.3%) returned the questionnaire. Two parents were excluded because they failed to fill out most of the questionnaire. Of the 65 parents that answered the questionnaire, one father refused to be interviewed with the SCID. The SCID could not be administered to two other fathers because of logistical problems. The sample was thus comprised of 62 subjects (46 mothers and 16 fathers). Most of the SCID-PTSD interviews were carried out by telephone (69.4%). There were no significant differences regarding to social and demographic characteristics between respondents and non-respondents, except for an excess of fathers in the non-response group.

The parents' age ranged from 23 to 62 years (mean = 40 years, SD = 10 years). Most participants were Caucasians (64.5%), but 15 (24.2%) were of mixed Caucasian and African-Brazilian ethnicity and seven (11.3%) were African-Brazilians. Twenty-one individuals (34.4%) had not completed elementary school, twelve (19.7%) had only elementary school, 12 (19.7%) had graduated from high school,

and 16 (26.2%) had attended college. Forty-nine parents (80.3%) were married or lived a stable relationship, seven (11.5%) were single, and five (8.2%) were separated, divorced or widowed. Twenty-eight parents (45%) reported a monthly family income of less than two minimum wages (approximately US\$330).

The majority of parents (88.7%) had only one son or daughter with cystic fibrosis (CF). Seven parents (11.3%) had two offspring with CF. One mother had two daughters and one son with CF. While eight fathers (50%) reported that they were not the primary caregiver, only one mother (2.2%) did not consider herself to be the main caregiver responsible for the patient with CF.

Patients with CF characteristics

Out of the 52 patients with CF, 30 were male. Their age varied from 2 to 33 (mean = 11.4 years, SD = 7.9 years). The majority of patients (57.7%) were Caucasians, 18 (34.6%) were of mixed Caucasian and African-Brazilian ethnicity and four (7.7%) were African-Brazilians. The time elapsed since the diagnosis of CF ranged from 2 months to 22 years (mean = 7.3 years, SD = 6.0 years). The patient's age at the time of the diagnosis varied from one week to 23 years (mean = 4.3 years, SD = 5.9 years). Twenty-eight patients (53.8%) had had at least one hospitalization during the last year, with the total lengths of the hospital stays ranging from 3 to 130 days (mean = 30 days, SD = 29.2 days).

PTSS and PTSD prevalence

Table 1 depicts current and lifetime prevalence of full and partial PTSD for the 62 parents. Four parents (6.5%) – three mothers (6.5%) and one father (6.3%) – met diagnostic criteria for current full PTSD. Moreover, 12 parents (19.4%) fulfilled diagnostic criteria for partial PTSD and 11 (17.7%) for lifetime PTSD. When parents with current full PTSD were combined with those with partial PTSD to form the group with PTSS, a “total” prevalence for PTSS of 25.8% (16/62) was obtained.

Table 1. The distribution of PTSD diagnosis in parents of patients with cystic fibrosis

	Current full PTSD (%)	Partial PTSD (%)	Current full PTSD plus partial PTSD (%)	Lifetime PTSD (%)
Parents (n = 62)	6.5 (4)	19.4 (12)	25.8 (16)	17.7 (11)
Mothers (n = 46)	6.5 (3)	19.6 (9)	26.1 (12)	19.6 (9)
Fathers (n = 16)	6.3 (1)	18.8 (3)	25 (4)	12.5 (2)

PTSD = posttraumatic stress disorder.

Table 2 displays frequencies and percentages of PTSS clusters (reexperience, avoidance and arousal) and of each specific PTSD symptom. Reexperience and arousal were the most frequent symptoms clusters among the participants: while 39 parents (62.9%) met diagnostic criteria for the reexperiencing cluster and 39 (62.9%) for the arousal cluster, only seven parents (11.3%) met diagnostic criteria for avoidance. All parents in the PTSS group fulfilled the criterion B (reexperience). With a single exception, all parents with partial PTSD (11/12) failed to fulfill the criterion C (avoidance). Hypervigilance was the most frequently reported symptom in the whole sample (71%) and in parents with PTSS (87.5%). Behavioral avoidance was the less frequently reported symptom among all participants (4.8%) and also among the parents with PTSS (12.5%).

As shown in table 3, parents with and without PTSS differed significantly in two psychosocial aspects: the former reported emotional problems ($p = 0.001$) and acknowledged they needed psychological or psychiatric treatment ($p = 0.002$) more often than the latter. Regarding the clinical condition of the patient, the time elapsed since the diagnosis of CF was significantly smaller in the group with PTSS than in the group without PTSS ($p = 0.05$).

Table 2. Prevalence of PTSD clusters and PTSD symptoms in parents of patients with cystic fibrosis

	Total sample (n = 62)	Total sample (%)	PTSD symptoms group (n=16)	PTSD symptoms group (%)	No PTSD symptoms group (n = 46)	No PTSD symptoms group (%)	χ^2	p value
Criterion B – Reexperience	39	62.9	16	100	23	5	12.72	0.001
Intrusive recollections	24	38.7	12	75	12	26	11.97	0.002
Nightmares	9	14.5	6	37.5	3	6.5	9.18	0.007 [†]
Flashbacks	10	16.1	6	37.5	4	8.7	7.28	0.014 [†]
Psychological distress	28	45.2	11	68.8	17	37	4.84	0.056
Physiological reactivity	19	30.6	11	68.8	8	2.2	14.73	0.000 [†]
Criterion C – Avoidance	7	11.3	5	31.3	2	4.3	8.58	0.010 [†]
Cognitive Avoidance	16	25.8	5	31.3	11	23.9	0.33	0.741 [†]
Behavioral avoidance	3	4.8	2	12.5	1	2.2	2.75	0.161 [†]
Psychogenic amnesia	4	6.5	2	12.5	2	4.3	1.31	0.272 [†]
Diminished of interest	12	19.4	7	43.8	5	10.9	8.22	0.008 [†]
Feeling of detachment	7	11.3	5	31.3	2	4.3	8.58	0.010 [†]
Emotional numbing	4	6.5	2	12.5	2	4.3	1.31	0.272 [†]
Sense of foreshortened future	9	14.5	5	31.3	4	8.7	4.87	0.042 [†]
Criterion D – Arousal	39	62.9	15	93.8	24	52.2	8.79	0.008
Insomnia	23	37	11	68.8	12	26	9.26	0.06
Irritability	22	35.5	11	68.8	11	23.9	10.43	0.003
Difficulty concentrating	17	27.4	10	62.5	7	15.2	13.34	0.001 [†]
Hypervigilance	44	71	14	87.5	30	65.2	2.86	0.117 [†]
Exaggerated startle response	22	35.5	10	62.5	12	26.1	6.88	0.020

PTSD: posttraumatic stress disorder.

[†] Fisher's exact test.

Table 3. Comparison of parents with and without PTSD symptoms regarding their psychosocial-demographic features

	Total sample (N = 62)	PTSD symptoms group (n = 16)	No PTSD symptoms group (n = 46)	p
Age in years, mean (SD)	40 (10)	39.9 (9.7)	40.8 (10)	0.652
	n (%)	n (%)	n (%)	p
Female	46 (74.2)	12 (75)	34 (73.9)	NS [†]
Educational level higher than elementary school	37 (60.7)	8 (50)	29 (64.4)	NS
Married or lived a stable relationship	49 (79)	14 (87.5)	35 (77.8)	NS [†]
Family income lower than 2 minimum salaries	28 (45)	9 (60)	19 (41.3)	NS
More than one offspring with CF	8 (12.9)	2 (12.5)	6 (13)	NS [†]
Primary caregiver	53 (85.5)	15 (93.8)	38 (82.6)	NS
Referred emotional problems	30 (48.4)	14 (87.5)	16 (36.4)	0.001
Psychological/psychiatric treatment	13 (21)	1 (6.3)	12 (26.7)	NS [†]
Use of psychotropic	13 (21)	3 (18.8)	10 (22.2)	NS [†]
Needs of psychological or psychiatric treatment	24 (38.7)	12 (80)	12 (30)	0.002
Ideas of suicide	10 (16.1)	4 (25)	6 (13.6)	NS [†]
Plans of suicide	4 (6.5)	2 (12.5)	2 (4.5)	NS [†]
Suicide attempts	4 (6.5)	1 (6.3)	3 (6.8)	NS [†]

PTSD: posttraumatic stress disorder.

[†] Fisher's exact test.**Table 4.** Comparison of parents with and without PTSD symptoms regarding the clinical features of patients with CF

	Patients with CF	PTSD symptoms group	No PTSD symptoms group	p value
Age in years, mean (SD)	11.4 (7.9)	9.4 (7.4)	12.2 (8.1)	0.265
Female, n (%)	22 (35.5)	7 (46.7)	15 (40.5)	0.924
Time since diagnosis of CF in years, mean (SD)	7.3 (6.0)	4.7 (4.3)	8.3 (6.4)	0.049
Age at the time of diagnosis of CF in years, mean (SD)	4.3 (5.9)	4.7 (6.4)	4.2 (5.7)	0.558
Had hospitalized in the last year, n (%)	28 (45.2)	9 (60)	19 (52.8)	0.870
Days hospitalized in the last year, mean (SD), n = 25/52	30 (29.2)	45.25 (41.8)	22.9 (18.5)	0.111
Pancreatic insufficiency, n (%)	40 (64.5)	12 (85.7)	28 (84.8)	1.000 [†]
<i>Pseudomonas aeruginosa</i> , n (%)	24 (38.7)	6 (42.9)	18 (58.1)	0.533
<i>Burkholderia cepacia</i> , n (%)	4 (6.5)	1 (8.3)	3 (10.7)	1.000 [†]
MRSA, n (%)	4 (6.5)	2 (15.4)	2 (6.5)	0.570 [†]

PTSD: posttraumatic stress disorder; CF: cystic fibrosis; MRSA: Methicillin resistant *Staphylococcus aureus*.[†] Fisher's exact test.

Discussion

To the best of our knowledge, this is the first study to investigate the prevalence of PTSD in parents of patients with cystic fibrosis (CF). In our sample, current prevalence for full and for partial PTSD were 6.5% and 19.4%, respectively. The figure for full PTSD was higher than that found in the Canadian general population (2.7% in women and 2.1% in men)². However, it was lower than the pooled prevalence found by our group for the only six studies^{4,5,8,9,10,19} that have assessed PTSD prevalence in parents of children with other chronic diseases or undergoing high-risk procedures (15.1%, 95% CI: 9.5%-20.6%)

(unpublished data). The prevalence of current partial PTSD (19.4%) in our study was also higher than that found in general population (3.4% for women and 0.3% for men)². The presence of partial PTSD is associated with significant functional impairment² and with suicidal ideation, even after controlling for comorbidities such as major depressive disorder²⁰. However, despite the growing acknowledgment of relevance of the diagnosis of partial PTSD, only four studies have assessed this condition in parents of children with chronic diseases or undergoing high-risk procedures^{7,9,19,21}. Two of these studies were methodologically comparable to ours since they have also employed the SCID as the main instrument to diagnose partial PTSD: our figures were comparable to those found in mothers of children with cancer (20%)¹⁹ but higher than those (7.2%) observed in mothers of children submitted to hematopoietic stem cell transplantation⁹. This inconsistency may reflect differences in the nature of the traumatic event: while hematopoietic stem cell transplantation could be considered an acute traumatic event, diseases like cancer and cystic fibrosis would be more properly classified as chronic stressors. Furthermore, Manne *et al.*⁹ have probed the mothers for PTSD only after a relatively large period of time had elapsed since the stem cell transplantation when, presumably, some of the participants may have already had fully recovered from their traumatic experience.

Terr¹⁷ proposed that type 1 trauma (single incident traumatic event) results in reexperiencing, avoiding, and increased arousal, and that type 2 trauma (chronic or prolonged exposure to trauma) results in denial, numbing, dissociation, and rage. Famularo *et al.*²² suggested that acute cases tend to exhibit sleep difficulties, physiologic hyperarousal, and reexperiencing, whereas chronic cases demonstrate more dissociation, restricted affect, sadness, and detachment. However, reexperiencing and arousal were the most frequent symptoms clusters found in the present study. Our findings dovetail with those of others studies that have assessed the prevalence of PTSS clusters in parents of children with serious medical illnesses^{5,7,9,23} and reported that the avoidance cluster was the less frequently found condition. Exceptionally, Libov *et al.*⁸ observed that the avoidance cluster was as frequent as the arousal cluster in mothers of patients with cancer.

Several factors may account for the finding that avoidance was the less frequently reported symptom cluster in our sample. This observation might, for instance, reflect a sample bias operating in two possible ways: 1) we may have failed to recruit the parents more affected by avoidance symptoms, since they may have been refusing to participate in CF-local chapter; and 2) severe avoidance symptoms may have predisposed some parents to decline to participate in our study. Either way, avoidant behavior by itself might conceivably have led to an underestimation of the prevalence of PTSD in the present study.

Alternatively, it might be hypothesized that a relatively low prevalence of the avoidance cluster reflects the fact that the parents in our study could have no effective ways of distancing themselves from the "traumatic event", since the majority of them were entirely responsible for the treatment of their children. Therefore, in conditions where avoidance is almost impossible – as is the case of parents of patients with chronic illness – reexperience and arousal may be the more prevalent symptom clusters. It follows that the diagnosis of PTSD in these cases may be less likely to be made, since all three clusters must be present to fulfill the DSM-IV criteria for PTSD.

Hypervigilance was the most frequently reported symptom in our sample and physiological reactivity was the one that achieved the highest statistical significance when parents with and without post-traumatic symptoms were compared. Both symptoms hypervigilance and physiological reactivity – may be related to physical symptoms. Physiological reactivity is in itself a physical symptom and hypervigilance is a symptom of the hyperarousal cluster which has been found to be closely associated with physical health in some trauma samples²⁴. Significant physical symptoms may predispose these parents to seek treatment in primary care settings.

Mothers of patients with CF experience greater stress and poorer adjustment than mothers of healthy children or the general population²⁵. A recent meta-analysis¹¹ found that mothers of children with chronic diseases had a significantly higher PTSD prevalence than fathers. Mothers are usually more affected than fathers, presumably

because they are the primary caregiver and get more personally involved in treatment. In our study, mothers and fathers of patients with CF were found to have comparable current full PTSD prevalence: 6.5% and 6.3%, respectively. The difference between our results and those of the meta-analysis should be taken with a grain of salt, since it may reflect the low statistical power of the present study.

A noteworthy finding of our study is the largely unmet need for treatment of parents with PTSS. Despite most parents with PTSS having reported that they thought they had emotional problems (87.5%) and that they felt they needed psychological or psychiatric treatment (80%), only one of them was receiving mental health assistance at the time of the study. Although these parents acknowledged their problems, they were underdiagnosed and undertreated. On the other hand, it might be hypothesized that these parents have not sought treatment for themselves because either they thought their symptoms were a normal reaction to the situation they were going through or because their personal problems were considered to have a lower priority than their children's disease.

The presence of PTSS in parents may have significant clinical implications for patients with CF. The regimen of therapy necessary to preserve health in patients with CF is time-consuming, usually takes place at home, and may require the involvement of the whole family, especially in early childhood. The CF self-care regimen typically consists of chest percussion therapy, pancreatic enzyme supplementation, antibiotic courses, and multivitamins. Parents may feel very little or no self-efficacy regarding their ability to cope with the complexity and the vagaries of the treatment of a serious chronic medical illness like CF. Parents with reexperiencing or arousal symptoms – the two most frequent symptoms clusters in our sample – may have their capacity to understand medical guidelines or to transmit essential information to the health care team significantly impaired (5). Parents with hypervigilance – the most frequent symptom reported in our sample – can overburden health services by requesting frequent doctors' visits or by making an inordinate number of phone calls to medical services¹². Further, the presence of avoidance symptoms in parents may lead to non-adherence to medical treatment in their children¹³.

Regarding the clinical characteristics of the patients with CF in the present study, the only measures related to CF severity collected in this study (e.g; infections with *Pseudomonas aeruginosa*, *Burkholderia cepacia* and/or *Methicillin resistant Staphylococcus aureus* – MRSA; and pancreatic insufficiency) were not associated with the presence of PTSS in the parents. Although this negative finding could be ascribed to the low statistical power of the present study, it is consistent with observations made by several previous studies. In a recent systematic review of posttraumatic stress in childhood cancer survivors and their parents, Bruce²⁶ also found that objective medical variables concerning treatment and/or illness severity did not correlate with PTSD in parents. The scientific literature on pediatric medical traumatic stress suggests that the objective characteristics of the medical illness (e.g., severity) and of the related treatments (e.g., intensity) are not strongly related to the subsequent development of symptoms of PTSD²⁷. These findings are in consonance with the well established notion in "traumatology" that the subjective perception of risk is more important than the "real danger".

Limitations

Some methodological concerns of the present study need to be addressed. The sample is small and there was no control group. Moreover, subjects were recruited from a CF-local chapter, and therefore are a selected group of parents that might not be representative of the larger population of parents of CF patients. Nevertheless, this data is worth to be reported as this is a new area in PTSD research. Another limitation was that almost 25% of the contacted parents did not participate; these parents may have more PTSD symptoms, especially avoidance, and this may have also biased our sample. Some important indicators of illness severity such as quality of life and pulmonary function status were not included.

Conclusions

This preliminary study was the first to report PTSD prevalence among parents of patients with CF. Like parents of patients with others serious medical diseases, those of patients with CF often suffer from PTSS. Our findings highlight the importance of identifying parents with PTSS – "our invisible patients", as Manne²⁸ once called them. To improve PTSD detection, health care teams should ask parents of patients with CF what they are their feelings about their emotional health and about the need of psychological/psychiatric treatment. Parents reporting emotional problems or need of treatment should be referred to specialized treatment. Early recognition of PTSS would avert the avenue for chronicity. Future studies should investigate if the presence of PTSS affects parent's role as caregivers and whether the treatment of these symptoms would improve the management of patients with CF, increasing their offspring's adherence to CF treatment.

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Conflict of interest statement: IF has a daughter with cystic fibrosis.

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