Journal of Psychosomatic Research 79 (2015) 49–54

Contents lists available at ScienceDirect

Journal of Psychosomatic Research

Fear of disease progression questionnaire for parents: Psychometric properties based on a sample of caregivers of children and adolescents with cystic fibrosis

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ARTICLE INFO

Article history:
Received 6 October 2014
Received in revised form 20 February 2015
Accepted 21 February 2015

Keywords:
Fear of progression
Cystic fibrosis
Parents

ABSTRACT

Background: Parents caring for a child with a chronic somatic condition are at risk of increased distress and impaired quality of life. Fear of disease progression (FoP) is known to be an important source of distress in patients and their partners, and may be of relevance for parents as well. Existing measures are not applicable to parents. This study describes the adaptation of the FoP questionnaire for parental caregivers and investigated its psychometric properties.
Methods: Sixteen items appropriate for parents were derived from existing measures and from interviews with clinical experts in family-oriented psychosocial care. Factor structure, internal consistency, validity and sensitivity to change were analyzed in a clinical sample of 162 caregivers ($M_{age} = 42.07$ years, $SD = 6.0$ years, 87.8% female) of a child with cystic fibrosis.
Results: The exploratory factor analysis revealed a two-factor structure, which was not supported by confirmatory analysis. Cronbach's α was examined for total score (.91) and significant positive correlations of the total score with anxiety (HADS: $r = .70$) and depression (CES-D: $r=.60$, BDI-II: $r=.59$), and a significant negative correlations with quality of life ($r = -.66$) could be demonstrated. A significant decrease in FoP was found ($d = 1.11$) in a group of highly distressed caregivers undergoing web-based cognitive behavioral intervention.
Conclusions: The FoP questionnaire is a reliable and valid instrument for parents of children with CF. Further studies in larger samples are needed to clarify dimensionality and validity among parents of children with other chronic conditions.

Background

In 1997 Lee-Jones and colleagues [1] introduced the Self-Regulatory Model by Leventhal et al. [2] and explained fear of recurrence (FoR) as an emotional reaction to the diagnosis and treatment in patients with cancer. It is assumed that the perception and interpretation of actual illness-related issues are influenced by an underlying cognitive concept or representation about the disease and its consequences. They lead to emotional reactions in various ranges [1,3], including fears of the future. Since then, further research has been conducted and the term fear of progression (FoP) has been introduced [4,5]. FoP is defined as fear 'that the illness will progress with all its biopsychosocial consequences, or that it will recur' ([6], p. 13). According to Herschbach and Dinkel [6] both concepts are basically identical with FoR clearly related to the scope of oncology. Unlike anxiety disorders involving irrational anxiety, FoP is a rational and appropriate illness-related response to a real threat, caused by the disease itself and its treatment [6,7]. Moreover, a current study indicated that FoP in patients with cancer cannot easily be covered by criteria of anxiety disorders according to the *Statistical Manual of Mental Disorders*: *Fourth Edition* [8] (DSM-IV) and therefore appears to be a distinct phenomenon [9].

The body of literature about FoP and FoR is constantly growing. FoP has already been examined in adult patients with various chronic conditions [4,10,11], their caregivers and their partners [12–18]. Its clinical relevance becomes obvious when looking at elevated levels, its persistence over time [19], its positive association with anxiety or depression [7,10,12,20] and its negative association with quality of life [6,13,14,20] in patients themselves as well as in their partners or even at its impact on health care use [21,22]. FoP may prompt self-care [3] which may result in better adherence to treatment. However, it may reach dysfunctional levels when those affected are overwhelmed by their fears and cannot follow the treatment regimen adequately, due to avoidance of medical treatment or any other triggers of their FoP [3].

As FoP has been identified as a relevant cause of distress in patients with a chronic somatic condition and their partners [5,11,16], it may be

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http://dx.doi.org/10.1016/j.jpsychores.2015.02.014
0022-3999/

of importance for parental caregivers, too. Patients across various chronic conditions experience FoP in different extents [11]. It is known that FoP is higher for chronic conditions with an unpredictable course and uncontrollable symptoms [3]. It can be concluded from the literature that FoP is present in parents of children with various chronic conditions, especially when the disease is of life-threatening and progressive character, such as cystic fibrosis (CF), pediatric multiple sclerosis (MS), or mucopolysaccharidoses (MPS). Parents are known to experience distress or mental health problems associated with their child's chronic disease [23,24], and to be at risk of impairments to their quality of life [25]. For instance, an international study recently outlined the elevated prevalence rates of the symptoms of anxiety and depression in parents of children with CF [26].

Having a child facing a life-threatening condition adds new responsibilities, changes role expectations and may increase obligation to protect and keep constantly caring for the child [27]. Parents of a child with a progressive disease have to cope with the continuous threat that, despite available treatment, their child's disease is inevitably progressing. In patients and their partners it is hypothesized that anxiety [28] especially FoP [16] is shared interdependently or more precisely transferred. Although only one partner is affected by the disease, both perceive the same concept of fear, albeit with varying intensity [12,18,29]. This assumption is not entirely applicable to the situation of parental caregivers. The parents may perceive several aspects of FoP, like fear of pain, by proxy, as though they would be affected by the illness themselves. However, in contrast to partners of adult patients, they bear full responsibility for their child's care. After diagnosis they have to deal with a broad range of stressors [30] and several risk factors may compound their own psychosocial adaptation [31]. In addition to compassion and fear of losing their child the continuous exposure to the threat of not having done enough or not being able to protect their child from the disease and its consequences may cause FoP, which is different from FoP of patients or partners. Moreover, when shifting the perspective to parental caregivers in line with the above definition, their fears additionally encompass the consequences of disease progression for their personal life.

Despite the fact that the volume of research on FoP in patients with chronic somatic conditions is continuously growing, there has been little research on FoP in parents up to now. Nevertheless, in a review Wakefield et al. [32] discussed potential sources of increased distress in parents once their children had completed cancer treatment. In their findings, mostly based on qualitative studies, they pointed out that FoR is one major factor [32]. Overall, in this target group FoR decreased after completion of treatment but peaked especially before and during appointments for screening [33]. At the present time, the authors are not aware of any studies that have investigated FoP in parents of children with chronic somatic conditions other than cancer.

Therefore, based on the available measures we developed a FoP questionnaire to assess the caregivers' perspective and determined its psychometric properties in a sample of caregivers of children and adolescents with CF. We hypothesized that the questionnaire would cover a relatively homogenous concept of FoP, which does, however, also take the personal consequences for the parents themselves into account. We expected to find moderate to high positive associations with symptoms of anxiety and depression and negative associations with quality of life. We examined the effects of the number of children, disease severity, and gender of the parents on FoP.

Methods

Participants and procedure

The development and pre-testing of psychometric properties were conducted within a study which evaluated web-based psychotherapy for caregivers of minors with CF (WEP-CARE) [34]. Further data for the final testing of psychometric properties in a larger sample were collected within a service research study evaluating a patient-centered care model (VEMSE-CF study). The study protocols of both studies were approved by the Institutional Review Board (IRB) of the University of Ulm. All participants gave their written informed consent.

Caregivers of a child with CF (0–18 years) were eligible for this investigation. The wide age range was eligible, because we aimed to develop an instrument, which allows measuring parental FoP across the whole developmental span of childhood and adolescence.

Participants in the WEP-CARE study were recruited via flyers distributed in German speaking CF-centres and various publications of the German Cystic Fibrosis Association. Altogether, 50 caregivers were interested in participating in the study. They registered on the study website and completed baseline questionnaires on a secure online-platform. For the VEMSE study, which is ongoing, 306 patients (control and treatment condition) were recruited, of whom 164 patients were 18 years or younger. Data referring to the older child in families participating in the study were used. Cross-sectional data were available for 112 caregivers. The final combined sample consisted of 162 caregivers.

Measures

Demographic and medical information

The following socio-demographic data were collected by self-reports: age and gender of the caregivers, age and gender of the child with CF, and the total number of children with CF in the family. Current lung function of the child with CF (forced expiratory volume in one second; FEV_1%) was also assessed via self-reports of the parents in the WEP-CARE study.

Fear of disease progression

The items were derived from existing versions of the Fear of Progression Questionnaires for patients (FoP-Q, [7]) and their partners (FoP-Q-SF/P, [16]). Both measures have excellent reliability and validity [7,16,35]. The items were chosen based on interviews with clinical experts in family-oriented psychosocial care, and the item content was modified to reflect the parents' perspective. One additional item was suggested by the clinical experts and therefore included in the item pool (item 16 ‘I worry about whether I am able to impede the disease with the treatment.’). The item covers the perceived burden of responsibility for treatment outcomes, which is reported often by the parents. Overall, the original questionnaire consisted of 16 items representing different aspects of fear of disease progression. The frequency of each behavior, thought or emotion is rated on a 5-point scale from never to very often [1–5]. Pre-testing of psychometric properties in a subsample of 50 parents revealed promising results [34].

Symptoms of anxiety

The anxiety subscale of the Hospital Anxiety and Depression Scale (HADS; 36) was used to assess symptoms of anxiety. Each of the seven items in each subscale represents one symptom and its severity is rated on a 4-point scale (0–3) over the previous 7 days. Psychometric properties have been reported to be good across various studies [36] with Cronbach's α on ranging from .68 to .93 for the anxiety subscale ($M = .83$). Associations with Spielberger's State–Trait Anxiety Inventory ranged from .64 to .81 and with symptoms of depression measured with the Beck Depression Inventory from .61 to .81. Cronbach's α within our study was $\alpha = .81$.

Symptoms of depression

Different instruments for assessing symptoms of depression were used in two subsamples of our study group. In the intervention study ($n = 50$) the short form of the German version of the Centre of Epidemiologic Studies Depression Scale (CES-D) [37] was used. This 15-item screening instrument assesses frequency of depressive symptoms on a 4-point scale over the previous 7-day period. The CES-D is commonly used in research and clinical practice and features very good