Family-based interventions for children and adolescents with functional somatic symptoms
a systematic review
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TITLE:

Family-based interventions for children and adolescents with functional somatic symptoms: A systematic review

SUGGESTED RUNNING HEAD:

Family interventions for functional symptoms in youngsters

ABSTRACT:

Functional somatic symptoms (FSS), defined as physical symptoms that cannot be fully explained by organic pathology, are prevalent in youngsters worldwide. When severe, they can pose a major burden on the affected individual as well as on society. FSS seem to aggregate in families, and family variables may play a key role in symptom development and perpetuation. It may therefore be important to work with the family when managing FSS.

The aim of this systematic review was to explore and describe family-based interventions used for children and adolescents with FSS, further to evaluate the overall quality of the research in this area. Sixteen studies were included. The psychological orientation for the applied treatment was based on systemic or cognitive behavioral therapy. Treatment focused on family illness beliefs and on a shift away from somatic symptom attribution towards alternative explanations that were shaped by the psychological orientation of the applied treatment.
PRACTITIONER POINTS

1. Family influences are significant for functional somatic symptoms in youngsters.

2. Family intervention may be a relevant treatment approach for youngsters with functional somatic symptoms.

3. Family-based psychotherapy for youngsters with functional somatic symptoms is mainly based on cognitive behavioral therapy or on systemic family therapy.

4. Illness beliefs of the child/adolescent as well as illness beliefs of the family as a whole could be important treatment targets in young people with functional somatic symptoms.
MANUSCRIPT

Background

Functional somatic symptoms (FSS) are prevalent in youngsters worldwide and in all medical settings. Clinical presentation of FSS varies from self-limiting to disabling symptoms (Garralda et al., 2015), and when severe, FSS may pose a major burden on the individual, health services, and society as a whole (Konijnenberg et al., 2005). Early intervention is important because FSS in childhood may continue into adolescence and increases the risk of functional and related disorders later in life (Horst et al., 2014, Campo, 2012).

Definition, terminology and classification

FSS can be defined as physical symptoms that cannot be fully explained by organic pathology or any other physical disease or psychiatric disorder (van Gils et al., 2014, Garralda et al., 2015). Recurrent abdominal pain is one of the most common and well described types of FSS in early childhood (Apley, 1975). With increasing age the clinical presentation is often more complex with various symptoms, including headaches and other pains, fatigue, and pseudoneurological symptoms (Cottrell, 2016). In daily clinical practice and in the literature, efforts are made to conceptualize FSS. In medical clinical settings, somatic syndrome diagnoses are used, such as fibromyalgia, chronic fatigue syndrome (CFS) or irritable bowel syndrome (IBS) (Wessely et al., 1999) while in psychiatric settings, FSS are classified by using psychiatric diagnoses such as somatoform or conversion disorders (Garralda et al., 2015, Schulte and Petermann, 2011b).

The differences in diagnostic classification practices have been a challenge in FSS research. However, it has recently been suggested that common pathophysiological mechanisms and similarities in patient characteristics and treatment responses could speak in favor of regarding functional somatic syndromes and somatoform disorders as a single family of disorders in adults.
(Fink and Schroder, 2010). Based on this it has been recommended that intervention research should explore these simultaneously (Schroder et al., 2015). There is also a growing body of literature describing similarities across different FSS in youngsters (Rask, 2012, Eminson, 2007), which could support adaptation of these recommendations for intervention research for younger age groups as well. Therefore, in the present paper, FSS is used as an overarching term to embrace the different diagnoses, as shown in Figure 1.

About here: Figure 1

Family influences on functional somatic symptom etiology

A biopsychosocial framework, where the relative contribution of biological, psychological, and social factors is considered, is often used to explain FSS etiology (Garralda et al., 2015). Due to the focus of this paper, only explanatory family factors are discussed in the following. However, other explanatory factors for FSS in youngsters, including individual and iatrogenic factors, may be equally relevant (Garralda et al., 2015).

Though primarily based on clinical experiences and observations rather than systematic empirical research, the literature generally agrees that family influences are important for both the development and perpetuation of FSS in youngsters (Cottrell, 2016, Schulte and Petermann, 2011a). Prevailing explanatory models for FSS often categorize possible risk factors into vulnerability factors (susceptibility to develop FSS), precipitating factors (factors triggering symptom onset), and maintaining factors (factors perpetuating the pathological process) (Garralda et al., 2015), as illustrated in figure 2. This framework will also be applied in the following to provide a brief overview of suggested family factors which have been found to be related to FSS in youngsters (Schulte and Petermann, 2011a).
Figure 2

Family-related vulnerability and precipitating factors

Various family health problems have been described to increase vulnerability to FSS in children, and mothers of children with FSS report more somatic symptoms, depression, and anxiety (Schulte and Petermann, 2011a, Campo et al., 2007).

Also adverse family climate, where psychosocial stressors and high levels of conflict are present, may form an environment where the child’s vulnerability to develop and maintain FSS is increased (Schulte and Petermann, 2011a, Garralda et al., 2015, Craig et al., 2002).

Retrospective reports show that adults suffering from FSS have significantly more often been exposed to abuse or neglect by parents, and to conflicts or traumatic events within the family (Schulte and Petermann, 2011a, Brown et al., 2005, Layten et al., 2008). This is supported by a recent prospective population-based study showing that adolescents from disrupted families (divorce or death of a parent) have an increased risk of developing FSS (van Gils et al., 2014).

Further, attachment patterns may also play a role in the development of FSS, where the possible significance of insecure attachment for symptom development has been described in adults with FSS (Stuart and Noyes, 1999).

Family-related maintaining factors

Family interactions, including parent-younger, parent-parent, and parent-extended family relations, may be of primary importance in FSS youngsters. Qualitative and quantitative studies have established the relevance and significance of maladaptive illness beliefs and behaviors as maintaining factors in adult patients with FSS (Frostholm et al., 2014, Christensen et al., 2015).

Also adverse family climate, where psychosocial stressors and high levels of conflict are present, may form an environment where the child’s vulnerability to develop and maintain FSS is increased (Schulte and Petermann, 2011a, Garralda et al., 2015, Craig et al., 2002). Retrospective reports show that adults suffering from FSS have significantly more often been exposed to abuse or neglect by parents, and to conflicts or traumatic events within the family (Schulte and Petermann, 2011a, Brown et al., 2005, Layten et al., 2008). This is supported by a recent prospective population-based study showing that adolescents from disrupted families (divorce or death of a parent) have an increased risk of developing FSS (van Gils et al., 2014).

Further, attachment patterns may also play a role in the development of FSS, where the possible significance of insecure attachment for symptom development has been described in adults with FSS (Stuart and Noyes, 1999).
Salmon et al., 1999). In youngsters, the role of parental beliefs and behaviors in response to the child’s somatic complaints has been investigated (Eminson, 2007, Walker et al., 2006). High parental involvement and focus on the child’s symptoms have been shown to increase complaints (Walker et al., 2006, Rangel et al., 2005) and to affect the degree of symptom-related disability in youngsters with FSS (Logan and Scharff, 2005, Lewandowski et al., 2010, Bialas and Craig, 2007). Parental somatic attribution affects the family’s interaction with the health care system, which may have an important bearing on FSS in youngsters. In addition, physicians often tend to primarily pursue organic explanations for symptoms, which may entail iatrogenic amplification of illness worry and illness behavior in the child and family, which in turn may increase parental somatic attribution (Lievesley et al., 2014).

The literature and clinical practice report that particular family patterns may affect symptoms in youngsters with FSS. Families with psychosocial stressors and high levels of conflict form an environment in which the child’s FSS are not only precipitated, but also maintained (Schulte and Petermann, 2011a, Garralda et al., 2015). Another family pattern characterized by the apparent absence of social, family or psychological difficulties has also been suggested to be associated with FSS in childhood. In such families, perfectionistic youngsters and overprotective parents, who may be excessively involved with symptoms, have been described to perpetuate the child’s FSS, by keeping him/her in the sick role (Eminson, 2007, Beck, 2008, Garralda, 1996). This is supported by empirical findings, as summarized in a review on CFS where parental overprotection is described as a maintaining factor for CFS (Lievesley et al., 2014).

**Family-oriented interventions**

It is well established that family relations and functioning impact a child’s development in general (Rutter, 2002), and the empirical support for family-based interventions for various mental health
problems in childhood, as well as for the cost effectiveness of such interventions, is growing (Carr, 2014, Sharma and Sargent, 2015, Crane, 2007). However, the varying use of definitions of “family intervention” is a challenge to research in this field. Recently, family therapy has been described to draw on different theoretical schools simultaneously (Carr, 2014), and Diamond and Josephson suggested the following definition for family-based intervention: “family based intervention is any treatment modality involving parents as essential participants in treatment” (Diamond and Josephson, 2005, p.874). This definition covers a broad spectrum of interventions from purely educational parent management training to family psychotherapy.

Traditionally, family psychotherapy is based on systemic theory (Cottrell and Boston, 2002) where a core principle is the focus on relations, and pathology is seen as a function of family dynamics (Cottrell and Boston, 2002). The effect of systemic family therapy is well established for eating disorders, and new studies suggest that systemic therapy is also effective for depression and anxiety (Carr, 2014). During the past decades other psychotherapeutic approaches have been introduced in the area, such as Cognitive behavioral therapy (CBT), including so called third generation CBT like Acceptance and Commitment therapy (ACT). The core principle in CBT is to change dysfunctional thoughts and behaviors. Family-based CBT therefore often focus on symptom coping strategies where parents are educated to coach their children with regard to helpful symptom management.

Whereas psychological treatment as such, especially CBT, has shown promising results for FSS in youngsters (Eccleston et al., 2012, Huertas-Ceballos et al., 2008), the specific significance of employing family-based psychotherapy for pediatric FSS has yet to be explored.

**Objective**

To our knowledge, this is the first systematic review to explore family-based intervention approaches for youngsters with FSS.
The purpose of this review is to provide a descriptive overview of studies using family-based interventions for FSS in youngsters. Due to the scarcity of research, our goal was, as a first step, to describe the psychological orientation and the treatment focus for these interventions and to evaluate the quality of the existing studies. The findings could inform future empirical and clinical work in this area.

Method

The review was conducted according to the PRISMA guideline (Moher et al., 2010). A systematic approach to both literature searching, data extraction, analysis, and quality appraisal was used and a review protocol including the search strategy was developed a priori and published on PROSPERO [https://www.crd.york.ac.uk/PROSPERO, ID no: 42015016703].

Search strategy

The list of search terms covered different nomenclature used in the FSS literature, including terms for various functional somatic syndromes and for somatoform and related disorders in combination with different terms for family-based interventions. We searched for synonyms of identified search terms in MESH in PUBMED and EMTREE in EMBASE (Table 1).

The first author (DH) consulted a research librarian to develop a basic template adjusted to the individual databases to perform systematic searches in PUBMED, EMBASE and Psych Info. A first search was performed on January 4, 2015 followed by a supplementary search on March 4, 2016. The searches from the different databases were combined and duplicates were excluded using Endnote software. In addition, a manual search for references in selected reviews on FSS (Garralda, 1999, Ibeziako and Bujoreanu, 2011, Lewandowski et al., 2010, Eminson, 2007, Eccleston et al., 2012, Huertas-Ceballos et al., 2008) and in the reference lists of included papers was performed.
The selection process was conducted in three separate rounds: 1) All identified records in the search were appraised by their title by the first author (DH), 2) Two of the authors (DH and GD) independently reviewed the selected paper abstracts. Subsequently, the results were compared and discussed, and any disagreement regarding eligibility was resolved by consulting the last author (CUR), 3) The remaining papers were read in full, and final inclusion was determined by discussion and consensus decision between all three authors.

Table 1 to be inserted about here

Inclusion criteria

Studies had to present original data, to be limited to human studies on children and adolescents (aged 0 – 18 years), and to be published in a peer-reviewed journal in English or a Scandinavian language between January 1975 and March 2016. The included papers reported randomized controlled trials (RCTs), uncontrolled studies or larger case series (N ≥ 10) on children with FSS treated with interventions that included parents as participants. By the exclusion of smaller studies, we attempted to focus primarily on studies, which applied a somewhat, standardized treatment approach reproducible for larger groups of patients.

Data extraction and analysis

As the current body of literature was expected to be of variable quality and characterized by the use of diverse methodology, a descriptive approach to data analysis was chosen. A Data Extraction Sheet (DES) for recording the main results was developed a priori. Five studies were randomly chosen for pilot testing of the feasibility of the sheet and the relevance of its included items. The final version included information on study population (diagnosis, age, number of participants),...
treatment design and the applied family intervention (psychological foundation, number of sessions, setting, therapist competences, use of standardized treatment manual). All three authors performed and discussed the data extractions jointly. Review of data extracted under each of the predefined items was subsequently conducted. Both similarities and differences across the studies were explored. Based on the psychological orientation, the studies could be divided into two main groups: 1) studies on CBT-based family therapy and 2) studies on systemic family therapy. Accordingly, therapy characteristics and the focus of therapy were explored and descriptively compared for these two categories.

Assessment of study quality

The “Psychotherapy outcome study methodology rating form” (POMRF), revised by Ost et al in 2008 (Ost, 2008), was used to perform a standardised assessment of overall study quality. The POMRF examines 22 individual methodological elements, including sample characteristics, the psychometric properties of outcome measures, research design, therapist training, and therapeutic modality adherence. Each item is rated on a 3-point scale from 0 to 2, where 0 = Poor, 1 = Fair, and 2 = Good (range for complete score: min 0 – max 44 points). The measure has demonstrated good internal consistency (Cronbach’s alpha= 0.86) (Ost, 2008). All three authors performed the POMRF ratings independently and any disagreement was subsequently solved by consensus decision.

Results

The primary search produced a total of 1482 records after exclusion of duplicates. Another five records were identified through a manual search for references in the selected reviews. Among the
30 papers identified and read in full, 14 were included. A supplementary search in March 2016 produced another 44 papers where three additional papers could be included. For details of the study selection process, see Figure 3.

**General study characteristics**

The 17 included papers covered 16 original studies as two of them were based on the same study population (Chalder et al., 2010, Lloyd et al., 2012).

The 16 studies were all from industrialized countries and included a total of 587 youngsters (aged 5 – 18 years) and their parents. No studies described inclusion of other family members. Eleven studies focused on youngsters with functional pain syndromes, four studies on CFS, and the last study on conversion disorders. A total of 51 different outcome measures were used. Further study characteristics are displayed in Table 2.

Table 2 to be inserted about here

**Psychological orientation of applied therapy**

CBT-based family therapy was applied in 11 studies, including one study on acceptance and commitment therapy (ACT) (Wicksell et al., 2009). Systemic family therapy was used in four studies. In the study by Kozlowska et al, the treatment approach was based on a mixture of different psychological theories and could therefore not be precisely categorized (Table 3).

Table 3 to be inserted about here
Therapy characteristics

In all studies the therapy was performed in secondary or tertiary health care settings. The therapists’ professions were described in about half of the studies; these included physicians, nurses, psychologists, and social workers (Table 3). Only the more recent studies on family CBT provided specific information on the therapists’ training and qualifications, whereas therapist experience with family work as such was poorly described across studies on both types of psychological orientation. The amount of therapy delivered varied considerably. In contrast to the studies on family CBT, where the number of sessions typically was predefined, no precise limits for number of sessions were described in the studies on systemic family therapy.

Family sessions, here solely defined as sessions where both the child and one or both parents are present, were employed in all studies on systemic therapy, while the studies on family CBT used a varying number of family and individual sessions, with four studies using treatment programs with only separate sessions for youngsters and parents (Table 3).

Therapy focus

All treatment programs, regardless psychological orientation, reported that some of the treatment activities were devoted to working with the families’ illness beliefs, and they all adopted an explanatory model that went beyond explaining the symptoms as purely somatic. However, the studies deployed different alternative explanatory models in line with their psychological orientations (Table 3).

In the four studies on systemic family therapy, the child with FSS was described as “symptom bearer”, and FSS were explained as an expression of dysfunctional relations within the family, either between child and parent, or between parents. In the study by Turgay, symptoms were explained as a “communication channel” for psychological or relational problems (Turgay, 1990);
therapy sessions aimed to increase acceptance within the family of a psychological nature of the symptoms. The explanation by Liebman and by White (White, 1979, Liebman et al., 1976) was based on a similar symptom understanding; still, the explanations given were somewhat different. The youngster with symptoms was here described as both “symptom bearer” and “family healer”, and the symptoms as a means to shift focus away from relational problems within the family. Viner et al. used systemic theory slightly differently, focusing on the effect of a child’s symptoms on the family structure rather than trying to identify causes of the symptoms within the family (Viner et al., 2004). Taken together, the treatment approaches based on systemic therapy emphasized actively working with family relations through family sessions. The therapist had the task of uncovering and addressing familial dysfunctional relations such as parental over-involvement with the child and specifically with the child’s symptoms, parental marital problems, and/or problematic relations for the child outside the home, such as in school or with peers (White, 1979, Liebman et al., 1976). By reframing the problem and by supporting the subsystem boundaries in the family, the youngster was meant to get a sense of self-mastery, while the parents were increasing focus on their own relational problems.

In the studies on family-based CBT, dysfunctional thoughts and behaviors in the family related to the child’s FSS were considered to be important maintaining factors. Treatment approaches were focused on the child’s symptom understanding and alternation of erroneous perceptions of bodily functions, e.g. by explaining the connection between pain and stress. Further, the studies had varying focus on parental symptom understanding, and also on parental response to and coping with the child’s symptoms and symptom behaviors. The single study, using acceptance and commitment therapy, which is within the third wave of CBT, especially addressed dysfunctional avoidance behavior related to the symptoms and emphasized a shift in perspective from symptom alleviation to valued living in the presence of pain. Some of the CBT treatment approaches referred in their
explanatory model directly to the social learning theory, emphasizing that children could learn to deal with the symptoms if they were taught new behaviors through positive and negative reinforcement, and through coping skills practice (Palermo et al., 2009, Sanders et al., 1989). Parents were included in the treatment in order to facilitate the therapeutic process of the child, but also to varying degrees in order to work with their own illness perceptions and behaviors (Robins et al., 2005).

Quality assessment

The POMRF scores ranged from 2 to 32. The range was 2 to 10 for studies on systemic family therapy and 10 to 32 for studies on family CBT (Table 3). Low scores reflected various methodological problems such as no blinding of evaluators, lack of power analysis, and no handling of attrition. Scores related to degree of therapist training, therapist competences, and treatment adherences were in general low across the studies. The highest scoring item was seen for the description of symptom severity. Overall, studies of more recent origin received the best quality scores.

Discussion

Main findings

The literature suggests that psychological treatments are effective for FSS in youngsters (Eccleston et al., 2012, Huertas-Ceballos et al., 2008), but the significance of employing family-based psychotherapy instead of individual psychotherapy has yet to be explored. This review aims to describe the existing literature on family-based interventions for children and adolescents with FSS. It includes 17 papers based on 16 studies describing family-based interventions for youngsters with FSS. The majority of the studies were on youngsters diagnosed with functional somatic syndromes...
(functional pain syndromes and CFS) whereas only one study included youngsters with somatoform and related disorders as defined in the ICD and the DSM. The interventions were primarily based on CBT or systemic therapy. They focused unanimously on family illness beliefs and on a shift away from pure somatic attribution of the child’s symptoms, where the alternative explanations were shaped by the psychological orientation of the applied treatment. The studies were heterogeneous with regard to issues such as amount of therapy, outcome measures and the therapist’s profession. Treatment consisted of varying amounts of family sessions and individual sessions with the youngster and/or the parents. The overall study quality varied considerably, with the most recent studies on family-based CBT achieving the highest scores.

**Key limitations of included studies**

The main objective of the present review was to describe and characterize existing family-based interventions. Limitations of interest are therefore those that pertain to the thoroughness and transparency of the treatment descriptions offered rather than those that concern blinding, sample size, lack of power analysis and handling of attrition. These latter limitations were prominent in many of the included studies and their presence would evidently have seriously hampered any attempt to compare the outcome across the studies, and thus support the choice of a descriptive analytic approach for this review.

Therapist training and competences were in general poorly described. In psychotherapy, it is important that the therapist is professionally qualified to deliver a particular treatment; yet, in family-based therapy, being trained to work with the family per se is no less important. Still, the studies generally failed to describe whether the therapists had prior experience with working with families. However, one could argue that systemic family therapy training implicitly includes working with a family, whereas formal training in CBT traditionally focuses on individual
psychotherapy. Also, information on the availability of complete treatment manuals describing the specific treatment elements in more detail was lacking in most of the studies, especially the studies on systemic therapy.

**Interpretation of findings**

The findings emphasize the role of family illness beliefs when targeting FSS in youngsters by using family-based therapy. This concurs with qualitative research on the role of illness beliefs in adults with FSS, which suggest that patients’ acceptance of the biopsychosocial nature of their symptoms is important for a positive outcome (Risor, 2009, Salmon et al., 1999, Salmon, 2007). Thus, change in illness beliefs has been reported to be a significant mediator for the effect of psychological treatment in adults with severe FSS (Christensen et al., 2015). Furthermore, research has shown that parental somatic attribution is associated with poor outcome in youngsters with FSS (Garralda and Rangel, 2001).

This review illustrates that the current literature on family-based interventions for youngsters with FSS lacks consensus on how to target the family. Irrespective of their psychological orientation, the included studies differ with regard to amount and combination of family sessions and individual sessions for parents and youngsters. Further, the definitions of “family intervention” and “family session” differ across the studies and are of varying clarity. These inconsistencies reflect the prevailing challenges in family intervention research in general, and in particular the lack of agreement on how the family should be involved (Carr, 2014). The existing family interventions target both child and family factors that may contribute to the onset and maintenance of symptoms in various ways. They may focus on how family strengths and resources can help remedy these problems, as in systemic therapy, or how the parents may be involved in the treatment as co-therapists or coaches for their children to change his/her maladaptive illness behaviors and
symptom coping, as in family CBT. A challenge is to acquire more knowledge on what works for whom (Diamond and Josephson, 2005, Carr, 2014).

All of the included studies were conducted in secondary and tertiary health care settings. As youngsters with FSS are prevalent in all clinical settings, future research should also be concerned with “what works in different settings”, as it has been explored in a review aiming to investigate the applicability of secondary care interventions to primary care for adult somatic symptoms (Raine et al., 2002).

**Strengths and limitations of this review**

This review was conducted in accordance with the PRISMA guidelines and thus according to current standards for systematic reviews, and a review protocol was published on PROSPERO in advance. Screening by abstract and full text and data extraction were conducted by two or three authors independently with subsequent consensus decisions, which secured good reliability and validity of the selection procedure of the papers finally included. Furthermore, a standardized assessment tool (POMRF), designed specifically to evaluate the quality of studies on psychological interventions, was applied. However, as the POMRF does not use cutoff values for overall scores, a categorization of study quality as “good” or “poor” was not possible.

A broad search of the literature on family interventions for a youngster with FSS was performed. The search included different types of study designs and broad definitions of the patient population as well as the concept of family intervention, which is in line with both the current research in the field of family therapy research (Carr, 2014) and in the field of FSS treatment research (Schroder et al., 2015). In accordance with the AMSTAR (Assessing the Medical Quality of Systematic Reviews) checklist, three electronic databases were searched. Still, a possible limitation of this review may be the exclusion of other databases such as CINAHL, which may have left out relevant
studies. Further, relevant studies may have been missed by the exclusion of paper in languages other than English or Scandinavian, and of papers with less than 10 participants. Also, only one author conducted the literature search and made the first screening of the papers, which may have reduced the reliability and validity of the first step of the selection process.

Conclusions

Systemic therapy and CBT are currently found to be the most prominent treatment approaches used in family-based interventions for youngsters with FSS. Though both types of interventions focus on family illness beliefs and on shifting focus away from a somatic attribution of the child’s symptoms, they also represent different views on how family factors may develop, precipitate and maintain FSS in youngsters. Studies based on systemic therapy employ explanations about dysfunctional family structures and patterns, and therefore target family relations, whereas studies based on CBT refer to explanations where maladaptive parental and child illness behavior and symptom coping on an individual level are significant factors to target in treatment. The research, which has the best quality, is on family-based CBT. Considering that family therapy has traditionally been based on systemic theory, it is surprising that research in this area has not developed further.

More and better research is needed, including investigation of the significance of specific family factors for the development of FSS in youngsters. Exploration of “what works for whom”, by matching specific problematic family processes with relevant therapeutic techniques, i.e., exploration of the effect of systemic versus CBT-based therapy, is highly relevant to make further progress in the area. Also the importance of various treatment conditions such as treatment dose, the combination of family and individual sessions, family sessions including more family members than parents, such as grandparents or siblings, qualification and training of the therapists, should be
explored. Finally, the general family functioning and parental distress may greatly influence outcomes and could be especially important to assess in more detail as part of delivering family-based treatment.
References


Table 1: List of selected search terms.

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<th>Intervention</th>
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The complete search strategy is available on Prospero: [https://www.crd.york.ac.uk/PROSPERO](https://www.crd.york.ac.uk/PROSPERO)  ID no: 42015016703
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</tr>
<tr>
<td>Viner 2004</td>
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<td>CFS</td>
<td>CDC</td>
<td>9 - 17</td>
<td>78</td>
<td>56/32/ 24/17</td>
<td>ST</td>
<td>SMC</td>
<td>6/12/ 18/24</td>
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</tr>
<tr>
<td>Robins 2005</td>
<td>RCT</td>
<td>RAP</td>
<td>Apley</td>
<td>6 - 16</td>
<td>46</td>
<td>40/36</td>
<td>CBT</td>
<td>SMC</td>
<td>3/6 - 12</td>
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<td>Duarte 2006</td>
<td>RCT</td>
<td>RAP</td>
<td>Apley</td>
<td>5 - 14</td>
<td>15</td>
<td>CBT</td>
<td>SMC</td>
<td>Paediatric quality of life</td>
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</tr>
<tr>
<td>Author/Year</td>
<td>Study design</td>
<td>Study population</td>
<td>Diagnosis</td>
<td>Case - Criteria</td>
<td>Age (Years)</td>
<td>Sample size</td>
<td>Active treatment</td>
<td>Control</td>
<td>Outcome Measures</td>
<td>Follow up time (Month)</td>
<td></td>
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</tr>
<tr>
<td>Hicks 2007</td>
<td>RCT</td>
<td>Pain</td>
<td>&gt;3 episodes of head or abdominal pain; &gt;3 month interference with activities</td>
<td>9-16</td>
<td>25</td>
<td>21/18</td>
<td>CBT internet based</td>
<td>WL</td>
<td>Symptoms</td>
<td>-</td>
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<tr>
<td>Kozlowska 2008</td>
<td>Uncontrolled study</td>
<td>Pain</td>
<td>-</td>
<td>-</td>
<td>28</td>
<td></td>
<td>Psychotherapy</td>
<td>None</td>
<td>Symptoms</td>
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<tr>
<td>Palermo 2009</td>
<td>RCT</td>
<td>Pain</td>
<td>Chronic idiopathic pain &gt; 3 month; Pain interferes at least once a week with at least one area of daily functioning Duration &gt; 3 month</td>
<td>11-17</td>
<td>45</td>
<td>45</td>
<td>CBT, internet based</td>
<td>WL</td>
<td>Symptoms, Treatment acceptability</td>
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<td></td>
<td></td>
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<tr>
<td>Wicksell 2009</td>
<td>RCT</td>
<td>Pain</td>
<td>Duration &gt; 3 month</td>
<td>11-18</td>
<td>32</td>
<td>30</td>
<td>ACT</td>
<td>MDT &amp; Amitriptyline</td>
<td>Symptoms</td>
<td></td>
<td></td>
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<tr>
<td>Chalder 2010</td>
<td>RCT</td>
<td>CFS</td>
<td>CDC</td>
<td>11-18</td>
<td>32</td>
<td>31</td>
<td>CBT</td>
<td>SMC</td>
<td>Symptoms Parent illness behaviour, Comorbidity Treatment acceptability</td>
<td>3/6/12</td>
<td></td>
<td></td>
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<td>Lloyd 2012</td>
<td>RCT</td>
<td>CFS</td>
<td>CDC</td>
<td>12-18</td>
<td>68</td>
<td>67/64</td>
<td>CBT, internet based</td>
<td>SMC</td>
<td>Symptoms, Child self coping, Comorbidity</td>
<td>6/12</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Author/Year</td>
<td>Study design</td>
<td>Study population</td>
<td>Diagnosis</td>
<td>Case - Criteria</td>
<td>Age (Years)</td>
<td>Sample size</td>
<td>Active treatment</td>
<td>Control</td>
<td>Outcome Measures</td>
<td>Follow up time (Month)</td>
<td></td>
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<tr>
<td>Nieto 2015</td>
<td>Uncontrolled pilot study</td>
<td>Pain</td>
<td>Apley</td>
<td>9-14</td>
<td>15</td>
<td>CBT, internet based</td>
<td>None</td>
<td>Symptoms</td>
<td>Treatment acceptability, Comorbidity</td>
<td>-</td>
<td></td>
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</tr>
</tbody>
</table>

Apley: 1) Symptoms are recurrent, 2) more than 3 episodes in more than three month; the pain is severe enough to interfere with the child’s daily living, leisure activities, social relationships and/or school attendance.

UK (Oxford): 1) A syndrome characterized by fatigue as the primary symptom. 2) A syndrome of definite onset 3) The fatigue is severe, disabling, and affects physical and mental functioning. 4) The symptom of fatigue should have been present for a minimum of 6 months during which it was present for more than 50% of the time. 5) Other symptoms may be present, particularly myalgia, mood and sleep disturbance. 6) Certain patients should be excluded from the definition (patients with established medical conditions known to produce chronic fatigue, patients with a current diagnosis of schizophrenia, manic-depressive illness, substance abuse, eating disorder or proven organic brain disease.)

CDC (Fukuda): 1) Clinically evaluated, unexplained persistent or relapsing chronic fatigue that is of new or definite onset (i.e., not lifelong), is not the result of ongoing exertion, is not substantially alleviated by rest, and results in substantial reduction in previous levels of occupational, educational, social, or personal activities. 2) The concurrent occurrence of four or more of the following symptoms (substantial impairment in short-term memory or concentration, sore throat, tender lymph nodes, muscle pain, multi-joint pain without swelling or redness, headaches of a new type, pattern, or severity, un refreshing sleep, and postexertional malaise lasting more than 24 hours) These symptoms must have persisted or recurred during 6 or more consecutive months of illness and must not have predated the fatigue. Modification for children: 3-month duration.

Table 3: Type, characteristics and therapy focus of existing family interventions.

<table>
<thead>
<tr>
<th>Author</th>
<th>Year</th>
<th>Country</th>
<th>Psychological orientation/ Assumptions about the family &amp; symptoms</th>
<th>No. Of sessions</th>
<th>Therapist Profession</th>
<th>Focus of therapy</th>
<th>Treatment Manual Available</th>
<th>Setting</th>
<th>POMRF quality score (0-44)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Liebman</td>
<td>1976</td>
<td>USA</td>
<td>ST/ Symptoms shift focus away from relational problems in the family Child – symptom bearer</td>
<td>20-50</td>
<td>Not described</td>
<td>Dysfunctional family relations/ Symptom validation and management, illness beliefs</td>
<td>No</td>
<td>CAMHS</td>
<td>2</td>
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<tr>
<td>White</td>
<td>1979</td>
<td>Australia</td>
<td>ST/ Symptoms shift focus away from relational problems in the family Child – symptom bearer</td>
<td>Not described</td>
<td></td>
<td>Dysfunctional family relations/ Symptom validation and management, illness beliefs</td>
<td>No</td>
<td>CAMHS</td>
<td>2</td>
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<tr>
<td>Sanders</td>
<td>1989</td>
<td>Australia</td>
<td>CBT/ Parental reinforcement behaviours of pain. Symptoms explained by social learning theory</td>
<td>8</td>
<td>Clinical Psychologists</td>
<td>Illness behaviour (parental) Illness beliefs Child symptom coping skills</td>
<td>No</td>
<td>CAMHS</td>
<td>10</td>
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<tr>
<td>Turgay</td>
<td>1990</td>
<td>Turkey &amp; Canada</td>
<td>ST/ Symptoms have a psychological nature, shift focus away from relational problems in the family</td>
<td>27 16</td>
<td>Not described</td>
<td>Dysfunctional family relations/ Symptom validation and management, illness beliefs</td>
<td>No</td>
<td>CAMHS</td>
<td>2</td>
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<tr>
<td>Sanders</td>
<td>1994</td>
<td>Australia</td>
<td>CBT/ Parental reinforcement behaviours of pain, Symptoms explained by social learning theory</td>
<td>6</td>
<td>Psychologist</td>
<td>Illness behaviour (parental) Illness beliefs Child symptom coping skills</td>
<td>Yes</td>
<td>CAMHS</td>
<td>18</td>
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<tr>
<td>Author</td>
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<td>Country</td>
<td>Psychological orientation/ Assumptions about the family &amp; symptoms</td>
<td>No. Of sessions</td>
<td>Therapist</td>
<td>Focus of therapy</td>
<td>Treatment Manual Available</td>
<td>Setting</td>
<td>POMRF quality score (0-44)</td>
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</tr>
<tr>
<td>Chalder</td>
<td>2001</td>
<td>England</td>
<td>CBT/ Not described</td>
<td>Max. 15</td>
<td>Not described</td>
<td>Symptom validation and management, illness beliefs Child coping skills</td>
<td>Yes</td>
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<tr>
<td>Viner</td>
<td>2004</td>
<td>England</td>
<td>ST/ Not described</td>
<td>3 - 15</td>
<td>Nurse &amp; social worker</td>
<td>Dysfunctional family relations/patterns Illness beliefs</td>
<td>No</td>
<td>Liaison</td>
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<tr>
<td>Robins</td>
<td>2005</td>
<td>USA</td>
<td>CBT/ Parental reinforcement behaviours to pain</td>
<td>3 2</td>
<td>Psychologist</td>
<td>Illness behaviour (parental) Symptom validation and management, Illness beliefs Child symptom coping skills</td>
<td>Yes</td>
<td>Paediatric</td>
<td>21</td>
</tr>
<tr>
<td>Duarte</td>
<td>2006</td>
<td>Brazil</td>
<td>CBT/ Parental reinforcement behaviours to pain</td>
<td>4</td>
<td>Paediatrician</td>
<td>Illness behaviour (parental) Symptom validation and management, Illness beliefs Child symptom coping skills</td>
<td>No</td>
<td>Paediatric</td>
<td>15</td>
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<tr>
<td>Hicks</td>
<td>2007</td>
<td>USA</td>
<td>CBT/ Parental reinforcement behaviours to pain</td>
<td>7 2</td>
<td>No therapist</td>
<td>Illness behaviour (parental) Symptom validation and management, Illness beliefs Child symptom coping skills</td>
<td>Yes</td>
<td>CAMHS</td>
<td>17</td>
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<tr>
<td>Author</td>
<td>Year</td>
<td>Country</td>
<td>Psychological orientation/ Assumptions about the family &amp; symptoms</td>
<td>No. Of sessions</td>
<td>Therapist</td>
<td>Focus of therapy</td>
<td>Treatment Manual Available</td>
<td>Setting</td>
<td>POMRF quality score (0-44)</td>
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</tr>
<tr>
<td>Kozlowska</td>
<td>2008</td>
<td>Australia</td>
<td>Mixed/ Family factors may be precipitating and/ or perpetuating the child’s symptoms</td>
<td>8 8</td>
<td>Different Professions</td>
<td>Symptom validation and management, illness beliefs</td>
<td>No</td>
<td>Liaison</td>
<td>5</td>
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<tr>
<td>Palermo</td>
<td>2009</td>
<td>USA</td>
<td>CBT/ Parental reinforcement behaviours to pain</td>
<td>8 8</td>
<td>Online coaches, profession not described</td>
<td>Illness behaviour (parental)</td>
<td>Yes</td>
<td>Research unit</td>
<td>30</td>
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<tr>
<td>Wicksell</td>
<td>2009</td>
<td>Sweden</td>
<td>CBT/ The avoidance of pain and distress is considered a core problem leading to reduce quality of life and function in daily life.</td>
<td>1 10 1 - 2</td>
<td>Physician/ Psychologist</td>
<td>Pain acceptance, Symptom validation and management, Illness beliefs Address avoidance behavior Focus on valued living.</td>
<td>No</td>
<td>Paediatric</td>
<td>23</td>
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<tr>
<td>Chalder</td>
<td>2010 Lloyd</td>
<td>2012 England</td>
<td>CBT/ Psychological distress in the mother corresponds to symptoms in the child</td>
<td>13</td>
<td>Psychotherapist, profession not described</td>
<td>Symptom validation and management, Illness beliefs, Child symptom coping skills Activity scheduling</td>
<td>CAMHS</td>
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<tr>
<td>Author</td>
<td>Year</td>
<td>Country</td>
<td>Psychological orientation/ Assumptions about the family &amp; symptoms</td>
<td>No. Of sessions</td>
<td>Therapist</td>
<td>Focus of therapy</td>
<td>Treatment Manual Available</td>
<td>Setting</td>
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<tr>
<td>Nijhof</td>
<td>2012</td>
<td>Netherland</td>
<td>CBT/ Central neurobiological disturbance with triggering, sustaining and perpetuating factors both on a biological and psychosocial level</td>
<td>21 21</td>
<td>Psychotherapist, profession not described</td>
<td>Psycho education Parents: coach/ encourage their children Cognitions concerning the sustaining and perpetuating factors</td>
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<td>Research unit</td>
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<td>Spain</td>
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<td>7 7</td>
<td>No therapist</td>
<td>Illness behaviour (parental) Symptom validation and management, illness beliefs Child coping skills</td>
<td>Yes</td>
<td>Research unit</td>
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</table>

Note:
Abbreviations: ST: Systemic family therapy, CBT: Cognitive behavioural therapy; Family: sessions with both parents and child present; Child: individual therapy with the child; Parents: individual sessions with the parents
POMRF (Psychotherapy outcome study methodology rating form) range for complete score: min 0 – max 44 points.
Classification of functional somatic symptoms (FSS).

In this paper, FSS is used as an overarching descriptive term, which covers the varying diagnostic categories for functional symptoms, including functional somatic syndromes and somatoform disorders.

<table>
<thead>
<tr>
<th>Setting</th>
<th>No health care/primary care</th>
<th>CAMHS</th>
<th>Diagnosis</th>
<th>Somatoform and related disorders</th>
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<td>CAMHS</td>
<td>Diagnosis</td>
<td>Functional somatic syndromes</td>
<td>Somatoform and related disorders</td>
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<td>Moderate</td>
<td>Mild</td>
<td>Functional somatic syndromes</td>
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<tr>
<td>Setting</td>
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<td>Pediatric setting</td>
<td>Diagnosis</td>
<td>Functional somatic syndromes</td>
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</tbody>
</table>
Illustration of how various family factors at different levels may be involved in the development and perpetuation of functional somatic symptoms (FSS) in youngsters.
17 papers were included covering 16 original studies as two of them were based on the same study population (Chalder et al., 2010, Lloyd et al., 2012).

Full text articles excluded with references: (1,2) (Berger et al., 1977, Sargent, 1983); (3-8) (Humphreys and Gevirtz, 2000, Gooch et al., 1997, Richtsmeier, 1985, Leslie, 1988, Sherry et al., 1991); (9) (Shorter et al., 1992); (10,11) (Sieberg, 2010, D’Souza et al., 2011); (12) (Kollbrunner and Seifert, 2013); (13-17) (Lipsitz et al., 2011, Sieberg et al., 2011, Maisami and Freeman, 1987, Woodbury, 1993, Scharff, 1995) 