

**Interpersonal Dynamics and Therapeutic Relationship in Patients**  
**with Functional Somatic Syndromes: A Metasynthesis of Case Studies**

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## **Abstract**

Patients with Functional Somatic Syndromes (FSS) often display troubled relationships with health care providers, psychotherapists, and significant others. Research shows that patients' history of trauma, attachment disturbances, and mentalization deficits may result in the emergence of maladaptive interpersonal patterns, which may later contribute to the onset and maintenance of FSS, "doctor-hopping," and dropout in psychotherapy. As the nature and therapeutic consequences of such maladaptive interpersonal patterns in FSS cannot be understood sufficiently by quantitative methods alone, there is a need for in-depth qualitative research. To address this issue, we conducted a metasynthesis of 23 published case studies of patients with FSS from various psychotherapeutic orientations. Results show that patients with FSS from our sample perceived others as unreliable, i.e., unavailable, overcontrolling, and overprotective. To adapt to such unreliable others, patients attempted to please and to control them. Patients also suppressed their emotional awareness and expression. Although alexithymia could also play a role, the primary reason for emotional avoidance seemed to be interpersonal in nature, i.e., patients were avoiding negative emotions in order to please and control the unreliable others. The onset and worsening of FSS were associated with both interpersonal and physical triggers. Showing signs of physical or emotional distress lead to more rejection, overcontrol, and overprotection from unreliable others, which could create a "vicious circle." Our results suggest that offering a more interpersonal perspective on emotion regulation difficulties would be beneficial for patients with FSS, counselors, psychotherapists, and other health care professionals.

*Keywords:* Functional Somatic Syndromes, Medically Unexplained Symptoms, Somatic Symptom Disorder, alexithymia, emotional avoidance.

## **Public significance statement**

Emotional avoidance and traumatic interpersonal history may play a role in the etiology and treatment of mind-body disorders. Therapists and health care providers should be sensitized for possible therapeutic impasses and adapt their strategies towards patients' interpersonal dynamics: pleasing others, controlling others, and emotional avoidance.

## **Introduction**

Patients with Functional Somatic Syndromes (FSS) present an ongoing challenge to medical professionals and psychotherapists. About 20-40% of general medical practitioners' consultations deal with various FSS (Haller, Cramer, Lauche, & Dobos, 2015), such as Irritable Bowel Syndrome, Fibromyalgia, Chronic Fatigue Syndrome, and other conditions of persistent pain, fatigue, or functional organ disturbance, for which no sufficient organic cause can be found (Henningsen, Zipfel, & Herzog, 2007; Henningsen, Zipfel, Sattel, & Creed, 2018). Despite the excessive costs for health care systems (Barsky, Orav, & Bates, 2005), neither medical nor psychotherapeutic treatments show consistent success in treating such complaints (Henningsen et al., 2018; Williams, Eccleston, & Morley, 2012).

Patients with FSS often experience frustration when dealing with health care specialists (Bertram, Kurland, Lydick, Locke, & Yawn, 2001), whereas practitioners perceive these patients as "difficult to treat" (Fischhoff & Wessely, 2003) and often feel overburdened and stuck (olde Hartman, Hassink-Franke, Lucassen, van Spaendonck, & van Weel, 2009). Both medical specialists and psychotherapists report considerable communicational and relational difficulties (Hahn, 2001; Hausteiner-Wiehle et al., 2011) and adverse transference/countertransference reactions (Luyten, Van Houdenhove, Lemma, Target, & Fonagy, 2012) in the treatment with these patients. Consequently, mutual mistrust and hostility may arise (Dixon-Woods & Critchley, 2000), as a result of which patients may

terminate treatments prematurely (Martens, Rempel, Zipfel, Enck, & Teufel, 2014) and engage in “doctor hopping” (Norton et al., 2011).

Given the high comorbidity between different FSS, as well as between the FSS and other mental disorders, it remains challenging to provide a consistent definition and guidelines for diagnosis and optimal treatment pathways for these patients (Henningsen et al., 2007). The recent attempt to introduce a broader category of Somatic Symptom Disorders in the fifth edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5; American Psychiatric Association, 2013) reflects increasing attention to shared psychological factors among FSS, hypochondria, conversion disorder, and other conditions of somatic distress (Dimsdale, & Creed, 2009). However, by broadening this nosological construct, possible etiological differences between FSS and other disorders risk becoming blurred (Henningsen et al., 2018).

Currently, it is hypothesized that FSS emerge from complex interactions between genetic factors, central sensitization, habituation, and interpersonal dynamics (Luyten et al., 2012). Several studies demonstrated genetic predispositions to FSS (Kato, Sullivan, & Pedersen, 2010). Furthermore, neurophysiological studies showed various anomalies in pain processing and modulation in patients with FSS, for instance a disruption in diffuse noxious inhibitory control (i.e., diminished ability to suppress pain response to repetitive stimuli; Edwards, Ness, Weigent, & Fillingim, 2003). Psychological factors such as prolonged interpersonal trauma, parentification (Imbierowicz, & Egle, 2003), or current stressful life events have been linked to higher risk of FSS as well (Aggarwal, McBeth, Zakrzewska, Lunt, & Macfarlane, 2006). Maladaptive illness behavior and “secondary gains” have shown to lead to habituation and psycho-biological conditioning (Witthöft & Hiller, 2010). All these factors appear to be connected in a complex, multidirectional way. This vast complexity of the bio-psycho-social interactions differentiates the FSS from stress-exacerbated organic disorders

(such as asthma or Crohn's disease) and from conversion disorder (where psychological factors appear to have a more unidirectional effect). Subsequently, recent theory-building around FSS has come up with "vicious-circle" models, where physiological, psychological, and interpersonal factors have complex but equally important contributions to the onset and sustenance of FSS complaints (Ezra, Hammerman, & Shahar, 2019).

The dominant evidence-based approach for treating FSS is derived from cognitive-behavioral theory. In this approach, biased perception of internal stimuli, catastrophizing appraisal, and habituation is assumed to cause and maintain FSS. Consequently, Cognitive Behavioral Therapy (CBT) for FSS consists of behavioral activation, modifying catastrophic cognitions, and cutting off reinforcement of attention from others (Withhöft & Hiller, 2010). However, such evidence-based treatments are moderately effective in treating FSS at best (Hauser, Bernardy, Arnold, Offenbacher, & Schiltenwolf, 2009; Luyten, et al., 2012), and, according to Cochrane Collaboration review, cannot maintain long term effects on pain, disability, mood, and catastrophizing (Williams et al., 2012). The fact that CBT is overall less effective in treating FSS than other mental disorders, could indicate that it fails to target certain mechanisms specific to FSS (Erkic et al., 2018). One important reason might be that CBT focuses overly on perception and cognitive processes, while interpersonal factors in FSS need more attention (Nickel, Ademmer, & Egle, 2010).

In response to limitations of current treatment models, modern attachment- and psychodynamic-informed models of FSS have recently arisen (Luyten et al., 2012; Meredith, Ownsworth, & Stronga, 2008; Nickel et al., 2010). These approaches are rooted in a well-established finding that patients with FSS frequently exhibit insecure attachment (Waller, Scheidt, & Hartmann, 2004), often accompanied by a history of emotional and physical abuse in childhood (Salmon, Skaife, & Rhodes, 2003), trauma (Afari et al., 2014), and parentification (Schier et al., 2011).

According to Luyten and colleagues (2012), children who cannot rely on their primary caregivers, may develop *attachment hyperactivation* or *attachment deactivation* strategies to cope with distress. Attachment hyperactivation usually results in clinging behavior toward the caregiver, whereas attachment deactivation results in pseudo-independence and hyperactivity. In adulthood, patients who rely on these strategies may face problems in communicating their needs and in resolving interpersonal conflicts. Resulting conflicts may lead to prolonged hyperarousal and set off a chain of psychoneuroimmunological events that may contribute to FSS (Lumley et al., 2011). Furthermore, maladaptive relational strategies may lead to complaining and clinging behavior towards health care providers (Luyten et al., 2012), resulting in the perception of patients with FSS as “hard to treat” (Fischhoff & Wessely, 2003).

Additionally, it is assumed that mentalization deficits and *alexithymia* play an important role in problematic interactions of FSS patients with others (Luyten et al., 2012). Several studies have shown that patients with FSS exhibit diminished emotional awareness and recognition in themselves and others, as well as reduced emotional expression (Güney, Sattel, Witthöft, & Henningsen, 2019; Lumley et al., 2011; Subic-Wrana, Beutel, Knebel, & Lane, 2010). Alexithymic patients were long considered “prototypical” for FSS (Bronstein, 2011), but recently, doubts have arisen as to whether alexithymia accounts for the majority of the FSS population or rather only represents a sub-group (Gil, Scheidt, Hoeger, & Nickel, 2008; Luyten, et al., 2012). Likewise, the debate on whether or not to include alexithymia into the definition of the FSS has been dampened by findings that alexithymia may represent a major risk factor for various types of psychopathology, and not only or specifically for FSS (Bach, Bach, Böhmer, & Nutzinger, 1994). It is also unclear whether alexithymia should be considered as a stable (or even “inborn”) trait or rather as a situational state. In the former case, it could mean that genetic, biological, or neurological factors account for the deficiency

in perception and processing of emotions. In the latter case the learning history and the interaction with the environment could result in temporary (but theoretically reversible) deficits in emotional processing (Bronstein, 2011; Mikolajczak & Luminet, 2006). Thus, alexithymia probably represents a more complex clinical phenomenon that cannot be studied with standard psychometric methodology alone (Meganck, Vanheule, & Desmet, 2008). Overall, attachment- and psychodynamic-informed models see alexithymia as an important factor contributing to interpersonal tensions in FSS regardless of its roots (Luyten et al., 2012).

Although the attachment- and psychodynamic-informed models consider interpersonal patterns as pivotal for the emergence and maintenance of FSS, our understanding of these interpersonal patterns remains limited. Most state-of-the-art findings stem from quantitative cross-sectional research, which typically relies on self-report questionnaires applied to large samples. For example, Henker et al. (2019) found maladaptive interpersonal schemas “Self-Sacrifice” and “Unrelenting Standards” in patients with FSS and Saariaho, Saariaho, Karila, and Joukamaa (2012) reported similar patterns. However, such findings cannot illustrate the full interpersonal dynamics of the patients, as they only show average tendencies, devoid of patient’ real-life interpersonal and therapeutic context.

Since the nature and therapeutic consequences of maladaptive interpersonal patterns in FSS cannot be understood sufficiently by quantitative methods alone, there is a need for in-depth qualitative research. Despite increasing interest of qualitative researchers in FSS, current studies have mainly focused on coping, stigmatization, and adjustment of patients with FSS in health care systems (cf. Sowińska & Czachowski, 2018), but have only marginally considered the aspect of interpersonal dynamics. A notable exception is the interview study by Arnd-Caddigan (2006), who linked relational patterns in FSS to early experiences of abuse, and to their later recreation in troubled relationships with therapists.

Furthermore, a focus-group study by Olde Hartman, et al. (2009) has explored interpersonal problems and strategies of general medical practitioners in contact with FSS patients.

However, up to this point, similar qualitative studies in the context of psychotherapy did not exist.

Despite little qualitative research on interpersonal dynamics in FSS, extensive reports on psychotherapy with patients with FSS exist in the form of case studies. Recently gaining more attention from the field of psychotherapy research, case studies can reveal especially rich insights into the interpersonal dynamics of patients and the therapeutic relationship (Buchholz, 2019). For example, Blaustein and Tuber (1998) and Lauterbach (1996) explored interpersonal conflicts in patients with FSS, while combining clinical observations and longitudinal research designs in case studies. As opposed to static “snapshots” derived from self-report questionnaires, case studies can capture complex and dynamic interpersonal processes as they unfold over time (McLeod & Elliott, 2011). Case studies also reveal unique insights into possible alliance ruptures and transference issues, which can advance and deepen the understanding of characteristic interpersonal patterns in FSS (Safran, 1993). In this way, case studies also facilitate the theory-building regarding FSS, as they go beyond mere efficacy questions and focus on in-depth exploration of the underlying mechanisms and processes (Stiles, Hill, & Elliott, 2015). During the past decade, case study methodology has rapidly evolved, providing guidelines on data collection, reporting, and reflexivity (Buchholz, 2019; Fishman, 2017; Iwakabe, & Gazzola, 2009).

Unfortunately, case studies are scattered across different databases and until recently have been hard to locate (Meganck, Inslegers, Krivzov, & Notaerts, 2017). Yet, due to methodological and technological advances in the field, it is now possible to systematically locate and analyze sizeable samples of high-quality peer-reviewed cases (Desmet et al., 2013;



Krivzov, Hannon, & Meganck, in press). This makes a systematic investigation of interpersonal dynamics in case studies of FSS now possible.

Understanding interpersonal dynamics has shown to be pivotal for improving treatment outcomes for various conditions, such as depression (Blatt, 2004), personality disorders (Linehan, 1987), and eating disorders (Arcelus, Haslam, Farrow, & Meyer, 2013). Consequently, in-depth understanding of the interpersonal dynamics, as well as its implications for the therapeutic relationship could provide a major step forward for improving psychotherapy for patients with FSS. In the current study, we therefore target the following research questions: 1) How do case studies describe interpersonal patterns in patients with FSS, (a) in their interactions with others and (b) in the therapeutic relationship? 2) How do case studies describe the circumstances of symptom occurrences and worsening? Since FSS are reported to occur, worsen, or resolve during major interpersonal events, such as break-ups, losses, conflicts, and interpersonal dilemmas (Hatcher & House, 2003; Hills, Lees, Freshwater, & Cahill, 2018), the second research question aims at additional exploration of the real-life context of patients' interpersonal dynamics. Thereby, exploring the circumstances of symptom onset can contribute to in-depth understanding of interpersonal and other factors accompanying the symptomatic course.

While a lot of case studies on FSS may exist in databases, a systematic approach is necessary for retrieval and comparison of findings from these cases. To meet this aim, we will answer our research questions by means of a *metasynthesis* of psychotherapeutic case studies of patients with FSS from various theoretical orientations. Metasynthesis is a rather novel approach in the area of psychotherapy research (Hannes & Lockwood, 2012; Levitt, 2018; Timulak, 2009). Unlike a regular literature review, metasynthesis aims at theoretical integration of existing qualitative research, while preserving its depth and complexity (Thorne 2017). Therefore, metasynthesis “goes beyond” a mere summary of reported findings or

themes and attempts active theory-building (Benoot, Hannes, & Bilsen, 2016; Krivzov et al., in press).

Recently, metasynthesis methodology has been adapted especially for case studies (Krivzov et al., in press; Willemsen, 2015). The data from case studies have a somewhat limited applicability in an *aggregative type* of metasynthesis, since case studies are produced by different research teams in different contexts and may represent idiosyncratic rather than typical findings (Levitt, Pomerville, & Surace, 2016; Levitt, 2018). As such, “generalizability” of case material in the traditional sense is limited. At the same time, data from case studies may be especially valuable for a *theory-building type* of metasynthesis (Krivzov et al., in press). Most especially, the unique and idiosyncratic aspects of case studies allow valuable clinical inferences to be made for the development of novel theories and enriching existing ones (Flyvbjerg, 2006; Thorne, 2017). Synthesizing only “typical” findings may result in less elaborate theory-building. Thus, the current metasynthesis should result in an enhanced theoretical understanding of the interpersonal processes in FSS beyond a mere summary of themes. Although our research design does not allow one to derive generalizations or compare interpersonal dynamics in FSS to other populations, it should allow to synthesize clinically relevant inferences for practitioners and researchers dealing with FSS.

## **Method**

### *Sampling.*

The current metasynthesis is reported in accordance with the Enhancing Transparency in Reporting the Synthesis of Qualitative Research guidelines (ENTREQ; Tong, Flemming, McInnes, Oliver, & Craig, 2012), whereby the conceptual choices, epistemological position, data retrieval and analysis, as well as the background of the researchers are explicitly stated. Next to the ENTREQ, which provides more general quality guidelines for the overall research

process, we implemented the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement as an often used instrument to assure the transparency of the data collection procedure (Moher, Liberati, Tetzlaff, & Altman, 2009).

The data was retrieved from the Single Case Archive (SCA; [www.singlecasearchive.com](http://www.singlecasearchive.com)), the largest online database for case studies, which has recently been established by an international group of researchers (Desmet et al., 2013). The SCA was constructed by systematic screening of major peer-reviewed journals of various theoretical orientations from 1985 until recently and contains cases that cannot be easily retrieved in other databases with regular search terms. More than 3000 case studies are currently included into the collection, which is constantly growing and can already be considered representative for the field. The coding system and the online search tool of the SCA allow for quick retrieval of cases based on various descriptive characteristics of patients and therapists, process and outcome measures, as well as relevant keywords. The SCA is therefore suited to both practitioners and scientists in the field of psychotherapy research. The quality of case studies is ensured by the inclusion criteria for the SCA core collection, i.e., being published in a peer-reviewed ISI-ranked journal and having a minimal length of two pages (Meganck et al. 2017).

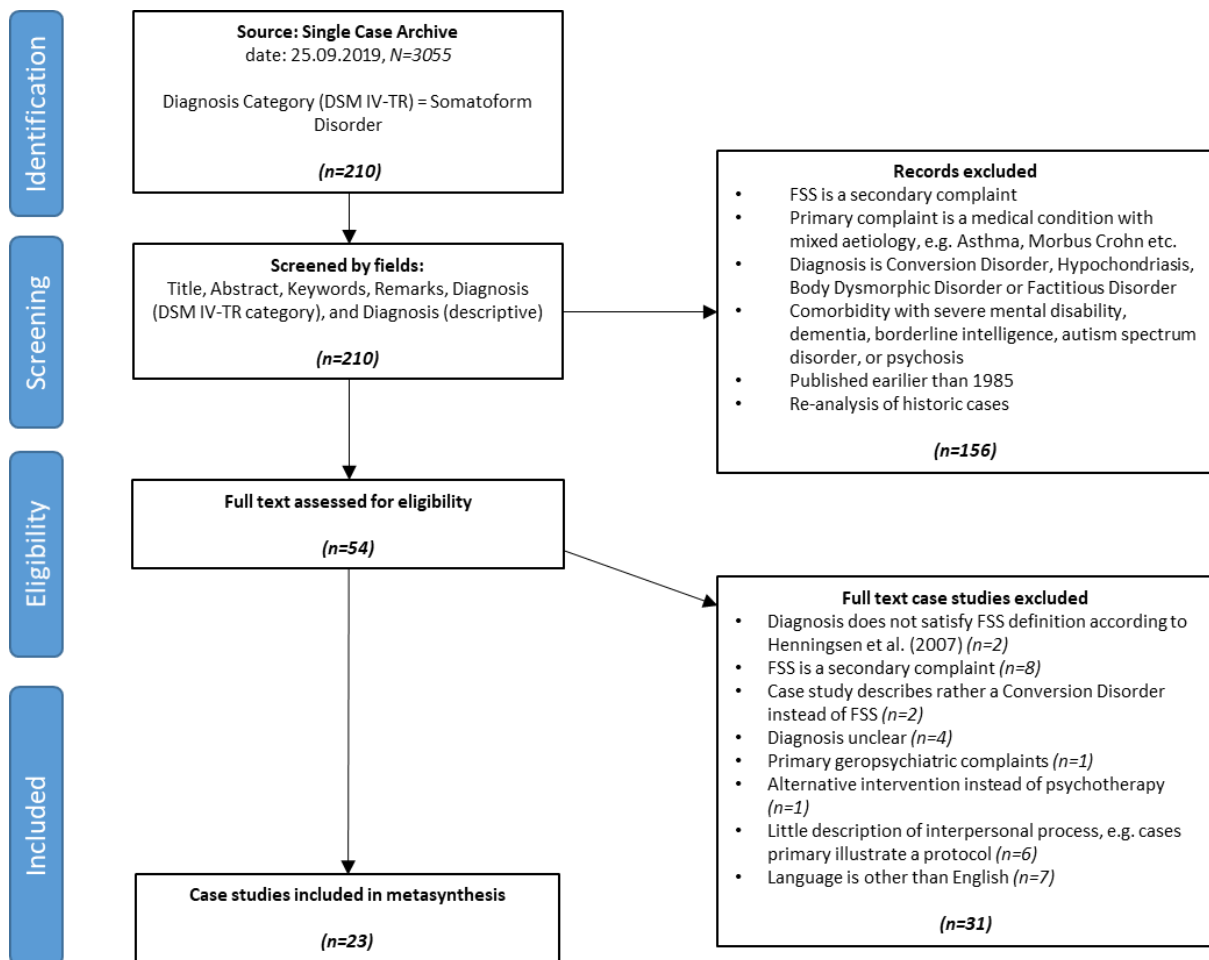


Figure 1: Overview of the selection procedure (PRISMA flow diagram, adapted from Moher et al., 2009).

Case selection for the current metasynthesis was performed on September 25, 2019; the data gathering process is reported in Figure 1, according to the PRISMA statement (Moher, et al., 2009). The case selection was performed by the first author, who was being supervised by the third author. During the entire process the first author and the third author strived for a consensus in defining and applying the criteria and discussed all doubts that arose (Tong et al., 2012). We first selected the DSM-IV-TR<sup>1</sup> (American Psychiatric Association, 2000) category ‘Somatoform disorders’ in the drop-down search field ‘Diagnosis’ of the SCA, this being the closest term for FSS in the DSM-IV-TR (Dimsdale & Creed, 2009; Erza et al.,

<sup>1</sup> The SCA operates the DSM-IV-TR categories instead of more recent DSM-5 categories, as the majority of published cases uses earlier versions of the DSM (Meganck et al., 2017).

2019). This step resulted in 210 cases with Somatoform Disorder as a single diagnostic category or comorbidity. In the next step, these records were screened by the SCA fields 'Title', 'Abstract', 'Keywords', 'Remarks', 'Diagnosis: DSM Category' and 'Diagnosis: descriptive terms.' Cases were excluded if they were not clearly differentiated from Conversion Disorder, Hypochondriasis, and stress-exacerbated organic disorders, such as Crohn's disease or asthma (Ezra et al., 2019). Also, cases with FSS as a secondary complaint were excluded, as well as cases with severe comorbidities that could impose a bias on interpersonal dynamics (e.g., dementia, autism spectrum disorders, or psychosis). However, excluding all cases with comorbidities would create an artificially extreme sample and would not reflect clinical reality, since FSS are typically accompanied by mood or anxiety disorders (Henningsen et al., 2007). Therefore, we decided not to exclude cases with such prototypical comorbidities. Additionally, we excluded cases published earlier than 1985, since they did not represent the Single Case Archive core collection. As a result, 156 cases were excluded in this step.

In the next step, another 31 cases were excluded after reading full texts. Here the same criteria from the previous step were applied, if reading full texts revealed more information relevant for exclusion. Further at this stage, case studies that merely described a treatment protocol and did not describe the interpersonal dynamics of patients were excluded. Studies in languages other than English were also excluded. In the end, 23 case studies remained in the final sample. Since such number is considered eligible for a metasynthesis (Timulak, 2009), and the sample appeared balanced according to various characteristics, such as diagnoses, comorbidities, and demographic characteristics (see next paragraphs), the authors did not attempt further purposive sampling (Benoot et al., 2016).

### *Characteristics of the sample*

The final sample consisted of 23 case studies drawn from 22 articles published between 1985 and 2016 (see Supplemental Material S1 for an overview of study characteristics, and Supplemental Material S2 for the full reference list). Ten cases represented one of the three most prototypical FSS conditions according to Henningsen et al. (2007), namely, Chronic Fatigue Syndrome, Fibromyalgia, and Irritable Bowel Syndrome or their mixed presentations. Other FSS in the sample were chronic headaches, low back pain, idiopathic arm pain, rashes, abdominal and pelvic pain, for all of which no sufficient medical cause could be identified. Thus, both the prototypical phenomenon of interest as well as its variations were included into the sample, which is recommended to ensure the variability of the material in the metasynthesis (Benoot et al., 2016).

Two thirds of patients (15 cases) showed comorbidities with mood disorders, anxiety disorders, (interpersonal) trauma, or secondary medical conditions, being consistent with epidemiological findings and typical clinical presentation in FSS (Henningsen et al., 2007). The sample contained 18 female and 5 male patients, also consistent with the epidemiological finding that women are more often diagnosed with FSS (*ibid*). Fourteen patients were adults (range: 26 years to “mid-50ies”), one was young adult (20 years), five were adolescents (range: 12 to 17 years) and three were children (range: 7 to 10 years). Five patients belonged to ethnic minorities in the context of the study’s country of origin (two Asian, one African American, one Hispanic, and one Indian).

The theoretical orientations of the case studies included ten cases of psychodynamic psychotherapy (ranging from classical psychoanalysis to integrative-interpersonal approaches), nine cases of cognitive-behavioral therapy (ranging from operant conditioning to collaborative- and context-sensitive approaches), two cases of systemic therapy, and two cases of eclectic therapy. Most cases described an individual therapy, three cases a group therapy, one case a couples’ therapy, and one case a family therapy. The outcome of thirteen

case studies was considered successful by the authors, eight were considered mixed outcomes, and two were considered a failure. Standard outcome measures were additionally implemented in six cases.

As for study type, fourteen cases were peer-reviewed clinical case studies, and nine cases were systematic case studies, i.e., they were conducted in a research setting involving additional psychometric outcome measures, behavior observations, or formal qualitative research (Iwakabe & Gazzola, 2009). The therapist was the first author in 17 cases, in one case it was the supervisor, and in one case an external researcher; in four cases the authorship of the therapist is not explicitly mentioned. The focus of two case studies corresponded directly with the research question of the current metasynthesis: a psychometric assessment study (Lauterbach, 1996) and a longitudinal qualitative interview study (Blaustein & Tuber, 1998) both explored interpersonal conflicts in a patient with FSS in the course of a therapy. Two other cases focused on a treatment failure due to a complicated therapeutic relationship (Gold, 1995; Tasca, Mcquaid, & Balfour, 2016). Fifteen cases focused on the general process and technical approach in treatment of FSS and four cases had a focus other than FSS (such as trauma, bereavement, or developmental transition).

### *Data Analysis*

Our epistemological position is rooted in Critical Realism (CR; Sims-Schouten, Riley, & Willig, 2007). Consequently, we assume that the authors of the cases studies (who are often the therapists as well) can be influenced by their theoretical and personal background, as well as by the constraints of the publication culture in respective journals (Westen, 2002). At the same time, we do not consider the interpersonal aspects described in the case studies as merely reducible to a certain “narrative.” We believe that therapists’ reports of interpersonal dynamics are especially valuable due to their first-person perspective and that their observations are potentially translatable to other theoretical narratives (Dixon-Woods,

Agarwal, Jones, Young, & Sutton, 2005). In accordance with the CR, passages of case studies that appeared dominated by certain theoretical narrative (i.e., “overinterpreted”) and not translatable to other theoretical terms, were first excluded and then re-read and discussed by several team members. An example of such a general theoretical statement could be: “[...] the vicissitudes of separation and the Oedipus may become insurmountable when the capacity for psychophysiologic regulation is not age and phase-appropriate.” (Shapiro, 2003, p. 557). Such passages that contained purely theoretical speculations, not grounded in descriptive material of the case, were excluded from the data analysis. In contrast, we included theoretical passages that were grounded in the data such as: “Oedipal level conflicts were also embedded in her struggles over culture. Clearly Kai had the intelligence and ambition to pursue higher education and a career. However, such achievement represented a terrible loss of the symbiotic mother.” (Shapiro, 2003, p. 557).

To ensure transparency and reflexivity of the data analysis process, as well as diversity of interpretations, the research team was composed of members with different levels of research and clinical experience and different theoretical background. Possible expectations and preconceptions of the team members were discussed prior to the analysis and assessed during the data analysis and synthesis phases (Levitt, 2018). The first author was the primary researcher of the project and a clinical psychologist with 2.5 years of experience with treating FSS. He mostly identified with psychodynamic perspective and the German psychosomatic school (Zipfel, Herzog, Kruse, & Henningsen, 2016). Based on his clinical experience and theoretical background, his possible preconceptions included a tendency to search for conflicts underlying symptoms in FSS and to emphasize trauma as a primary contributor to FSS. The second author was a clinical psychologist with 6 month of clinical experience and identified with the “third wave” CBT and Acceptance and Commitment Therapy (ACT). Her expertise in the area of illness-related stigma and other psychosocial factors could drive her



attention towards injustice and away from more psychological level of disturbed communication in FSS. Also, it is possible that she would be more prone to see interpersonal factors as reinforcing “sick role behavior”. The third author was a clinical psychologist, supervisor, and psychotherapist with over 10 years of experience and identified with Freudian-Lacanian psychoanalytic tradition. The fourth author was a postdoctoral researcher and a clinical psychologist and completed her training from Freudian-Lacanian perspective. The third and the fourth authors could also be possibly driven towards seeing conflicts at the root of FSS. Furthermore, they could be inclined to look for the difference between anaclitic (i.e., clinging) and introjective (i.e., pseudo-independent) interpersonal styles in FSS in accordance with Blatt’s (2004) specificity hypothesis, on which they published previously.

In the initial phase of the project the first and the third authors read several case studies together, developed preliminary hypotheses and designed the study. The second author was not involved at the beginning of the project and was later invited as an external team member with a non-psychodynamic background. The data analysis was then jointly conducted by the first and the second authors. During the data analysis, the first and the second authors tried to formulate their interpretations both in CBT- and in psychodynamic terms and wrote memos as to surpass the perspective of own their own theoretical orientations. Besides having a different theoretical background, the second author was less involved in case comparisons at the initial stages of data analysis and thus could approach the cases in a more holistic manner, which contributed more specific and nuancing aspects. Being senior researchers, the third and the fourth authors supervised the coding and the synthesis.

For the data analysis, we followed a modified procedure of *thematic synthesis* (Krivzov et al., in press; Thomas & Harden, 2008), which consists of: 1) close reading of the text and line-by-line coding that yields “descriptive codes,” 2) developing “descriptive themes,” and 3) developing overarching “analytical themes.” No data analysis software was

used. The first step involved close reading of the text and identifying passages that described interpersonal events between the patient and the others, as well as between the patient and the therapist. Next to the documented interactions, the interpretations made by the authors of the case studies were selected for the analysis as well, which resulted in inclusion of substantial amount of context information from the case studies. This was followed by line-by-line coding of selected passages and developing “descriptive themes,” i.e., themes that were still close to the data but contained first abstractions and aimed at “translating” concepts between different case studies. To preserve the context of the findings, we considered the case as the primary unit of analysis (Hoon, 2013) and first constructed descriptive themes within each case. In the next step, we put descriptive themes and representative quotes from different cases in a separate table by domains from the research question 1 (domains “interpersonal dynamics,” and “therapeutic relationship”) and the research question 2 (domains “symptomatic course” and “significant events and tipping points”). After comparing the first seven cases (two CBT cases, three psychodynamic cases and two eclectic cases; see Supplemental Material S1), we re-read the case studies, refined and adjusted the descriptive themes, and constructed preliminary analytical themes. Subsequently, we alternated between close reading on case level and synthesis phases several times, each time deriving more nuanced analytical themes. The subsequent cases were mostly coded into the pre-existing categories, but each new case was first read separately to ensure that new emerging patterns received additional descriptive themes.

Contrary to the manual by Thomas and Harden (2008) at first, we did not perform data extraction from the cases but attempted to gain a holistic understanding of the interpersonal and therapeutic process in each case at the beginning. In this way we discovered an additional theme on the therapist’s intervention, “Therapist being a different (reliable) other to the

patient.” This theme could be directly linked to other themes (e.g., “Other as unreliable”) and therefore was incorporated into the final model.

Since the aim of the metasynthesis is “to go beyond” mere aggregation of themes and to strive for overarching theory-building (Benoot et al., 2016; Thorne, 2017), in the final step we developed a “vicious circle model” (see Figure 2), which integrates the themes on interpersonal dynamics and symptomatic course. The first, second, and third authors developed the model together, while relying on their reflections on how different themes were linked within the cases. This model aims at the “bigger picture,” which cannot be grasped in a single case.

## Results

We identified four themes concerning interpersonal dynamics in patients with FSS: *Others perceived as unreliable; Pleasing others; Controlling others; and Emotional avoidance*. All themes but one were present in the interactions with others, as well as in the therapeutic relationship; the theme “*Controlling others*” was predominantly present in the therapeutic relationship. Additionally, we identified one theme on interpersonal strategy used by the therapist: “*Therapist being a different (reliable) other to the patient.*” Occurrence and worsening of FSS were described by two themes: “*Triggering of FSS symptoms in the interpersonal context*” and “*Triggering of FSS symptoms in the context of physical illness or strain.*” The distribution of themes per theoretical orientation is visualized in Supplemental Material S3. The themes, sub-themes, and example quotes are summarized in Supplemental Material S4. In following, we first elaborate on each theme in detail and then introduce the “vicious circle model” as a synthesis of the themes.

### *1) Others perceived as unreliable*

Often from childhood on and well before the onset of FSS, most patients report to have been surrounded by unreliable significant others. Patients were not able to count on the others' presence and emotional support, as a result of which they felt the need to adapt to the others' unavailability. Often, patients reported harsh criticism, high expectations, and control from significant others.

For example, patients often perceived their parents as *unavailable and abandoning*, and many shared the experience of abuse, parentification, neglectful or harsh parenting [3, 6, 8, 9, 11, 12, 20, 21]<sup>2</sup>. So, one patient experienced a purely mechanistic care provided by her depressed mother [3], and another reported “being robbed of her childhood” due to having to look after siblings and “to mother her mother and ask for little caretaking in return” [8].

In addition to emotional unavailability, patients also feared physical unavailability of others due to (real or imagined) illness, death, or accident [11, 12, 15, 17, 18]. For example, for a Jewish patient who escaped Nazi Germany with his parents, the threat of losing significant others was omnipresent in his childhood. He needed to “keep low profile” and be ready to relocate frequently and unexpectedly, while his parents could neither provide emotional safety, nor openly mourn the relatives who died in the Holocaust. [11]. Even if significant others were alive and doing well, strong and uncontained separation anxiety was present in several cases, as if the other could disappear every moment [1, 17, 18].

Next to being unavailable or absent, others were also described as *controlling, demanding, and prescribing identity* to patients [1, 5, 6, 7, 9, 13, 23]. For example, Joan strived to become a “dream daughter” to her criticizing father and was afraid to disappoint his high expectations [23]. Jason, another young patient, felt abandoned and isolated from his

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<sup>2</sup> The numbers in square brackets indicate the number of the case study in which the theme has been identified from the Supplemental Material S1 and 3.

family if he did not comply with the masculine role, which included not complaining about his pain and depression [7].

Later in life, these experiences of abandonment and feeling overcontrolled by others were repeated at work and in romantic relationships [5, 8, 9, 10, 11, 12, 13, 21]. Patients reported feeling lonely, misunderstood, and neglected by significant others. They also reported incidents of traumatization and emotional and physical abuse by others [8, 11, 12, 20]. After the onset of FSS, the others often became *overprotective* of the patient [1, 13, 15, 16, 17, 18, 19], for example by taking over daily duties of the person, while still being controlling and demanding.

The perception of others as unreliable was sometimes transferred onto the therapist and the therapeutic relationship [1, 2, 3, 6, 9, 11, 20]. One patient, for example, perceived interpretations of his therapist as intrusions or “proclamations of how he should think and feel” [2]. Another patient associated the therapist with “a teacher shouting at her in the front row at school” [6]. Distrust or skepticism towards the therapist and psychotherapy approach were also present [9, 11], as well as a fear of being abandoned and rejected by the therapist [2, 6].

## 2) *Pleasing others*

In an attempt to adapt to unreliable others, patients typically tried to *please others*, for example, by caring for others’ needs more than one’s own [5], by being overly obedient [12], by being a “yes-girl” [4], or by pushing oneself too hard [5]. In several cases, patients engaged in exaggerated and compulsive caretaking activities, spending most of their time helping others while driving themselves into exhaustion. The pattern of taking responsibility for unreliable others was also connected to a history of parentification [8, 12]. Interestingly, pleasing others was mentioned even in case studies that did not explicitly focus on patients’ early interpersonal history. For example, Tamisha’s tendency towards “being a yes-girl” was

targeted in here-and-now oriented CBT treatment, although little information could be obtained from this case study on why and how Tamisha acquired it [4]. Notably, in their striving to satisfy high expectations of others, one third of patients in the sample became *high achievers*, i.e., excelling academically, professionally, in sports, receiving scholarships, etc. [3, 4, 7, 8, 12, 14, 17, 23].

Patients' attempts to please others were also experienced by therapists in the therapeutic relationship, e.g., by patients being overly cooperative, trying to become "good patients" [18], and "making themselves perfect to the therapist" [2]. Notwithstanding, therapists felt that patients in fact remained on the surface of their problems and avoided emotional exploration in sessions. Whereas ruptures in the therapeutic alliance usually provide valuable information as "windows into core themes" of the patient (Safran, 1993), sometimes ruptures could not arise at all due to overcompliance of the patients. In line with this, Gold (1995) reports on a treatment failure that was accompanied by a patient's ongoing enthusiasm and "compliance" in sessions, without her making any changes in real life and never daring to express any anger towards the therapist [5].

### *3) Controlling others in the therapeutic relationship*

Another strategy to adapt to the unreliability of others was to attempt to control them. Interestingly, the patients barely mentioned this pattern themselves during the therapy. Whereas patients typically described themselves as self-sacrificing altruists in relationships with others [20], their rather controlling attitude was detected in the therapeutic relationship by the therapist. As such, several therapists felt an inherent mistrust from the patients and a sense of being observed and controlled. For example, patients were carefully preparing and pacing the session [6], tried to determine the topics of the session [1, 6, 9], or dismissed and devalued the therapist altogether [11, 20]. Two therapists described an almost identical experience of *being controlled by non-verbal means*: The patient would signal distress with

bodily tension and freezing, and subsequently the therapist began to experience feelings of tension and immobilization in return [2, 3].

Sometimes controlling and pleasing of the therapist were present simultaneously, a dynamic which could be described as “appeasement.” Representative of such a dynamic is the behavior of Mrs. L. who constantly showed up 30 minutes before the start of the session, signaling her eagerness and respect towards the authority of the therapist, only to stubbornly defend against any attempts of emotional exploration in the subsequent hour [9].

#### 4) *Emotional avoidance*

The most common and striking characteristic of patients in our sample was *diminished emotional awareness and restricted expression of (negative) emotions*, such as anger, sadness, and frustration. Instead, patients had learned to hide their feelings and “to present a happy face to the public” [12]. Some patients actively denied experiencing negative emotions, even when these were detected and pointed out by others (e.g., during group therapy) [3, 10, 22]. However, this denial of having negative emotions does not necessarily mean that patients had no concept of them (e.g., due to severe alexithymia). In fact, emotional avoidance could only rarely be attributed to classical alexithymia in our sample [6, 20]. Conversely, in several cases the authors stressed that their patients were not typically alexithymic or were even capable of sophisticated psychological insights regarding their own and other’s mental life [2, 3, 5, 23]. Rather, patients tended to withhold emotional expression for interpersonal reasons. In six cases, patients’ emotional avoidance was explicitly described as a strategy *to please and control the unreliable others*. Patients felt that others expected them to restrict the expression of negative emotions, often under threat of social exclusion, shaming, or even abuse [12]. Jason, for example, reported that he had to be happy, “or else no one would want to be around [him]” [7]. Furthermore, patients avoided emotional expression in order not to overburden others or the therapist and were proud of “being strong and not complaining to others” [14].

Whereas emotional avoidance appears at the first glance as an inherent “trait” or “deficit” of the patients, it can also serve as a strategic response towards the expectations of the others, in line with the interpersonal strategies of pleasing and controlling.

During therapeutic sessions, therapists also observed emotional avoidance and often tried to target it in therapy, independent of their theoretical orientation. For example, one therapist addressed how a patient avoided crying in front of her (attributing tears to “allergies”) and linked it to a wish to protect others from negative emotions “to the extent that she did not even feel her own sadness nor feel tears as tears” [3].

#### *5) Triggering of FSS symptoms in the interpersonal context*

Several patients were reported to experience increasing FSS symptoms after suppressing negative emotions [3, 7, 18, 21]. For example, Blaustein and Tuber (1998) concluded in their longitudinal case study that Cindy’s “symptom eruptions were intimately and temporally connected to her inability to consciously own and represent her reactions to interpersonal conflicts” [21]. In this and in other cases, worsening of FSS was reported in the interpersonal context, especially *in the context of perceived abandonment and conflict*. Thereby, patients experienced bouts of symptoms when they felt isolated or rejected by others [7, 21], when confronted with separation anxiety [17, 18], in context of a relational break-up [9, 23], or when getting involved in arguments [1, 12, 21]. Similarly, in the therapeutic context, a transition toward a different therapist or perceived rejection by the therapist could lead to FSS [3, 6]. Furthermore, symptoms were also reported to worsen during or around the psychotherapy session [2, 3, 6, 11], which can possibly be linked to a re-experiencing of typical interpersonal patterns and avoided emotions in therapy.

#### *6) Triggering of FSS symptoms in the context of physical illness or strain*



Another major trigger for symptoms was a (non-FSS) physical event or condition, such as influenza, viral infection, or menstruation. For example, Marla's symptoms of FSS worsened when she got pneumonia [1]. Also, overexerting physical activity, such as exercising excessive sports or physical labor, was recognized by patients and therapists as a trigger for FSS [3, 6, 7, 13, 14]. Sometimes physical illness and perceived abandonment were interconnected, for example when infection coincided with a relational break-up [23]. Similarly, pleasing others could go hand in hand with physically overexerting oneself [5]. Overall, in six cases, both interpersonal and physical triggers of FSS were present simultaneously.

#### *7) Being a different (reliable) other to the patient*

As a reaction towards patients' perception of others as unreliable, some therapists adapted a relational strategy towards *being a different (reliable) other to the patient*. Therapists attempted to facilitate mentalization and tried to accept their patients' feelings without judgement and without being overly directive and hypercritical. If the intervention was successful, patients learned that their emotional reactions mattered to others and that expressing their feelings would not result in a rejection or an emotional breakdown of the other [2, 6]. It should be noted that therapists implemented numerous other interventions aimed at correcting maladaptive cognitions and behaviors, integrating of traumatic experience, learning new ways of coping, etc. (for metasyntheses of interventions in FSS see Krivzov et al., in press; Řiháček & Čevelíček, 2019) However, the strategic relational position of being a different (reliable) other seemed to provide an especially solid base for the therapeutic alliance and to facilitate change.

#### *Synthesis*

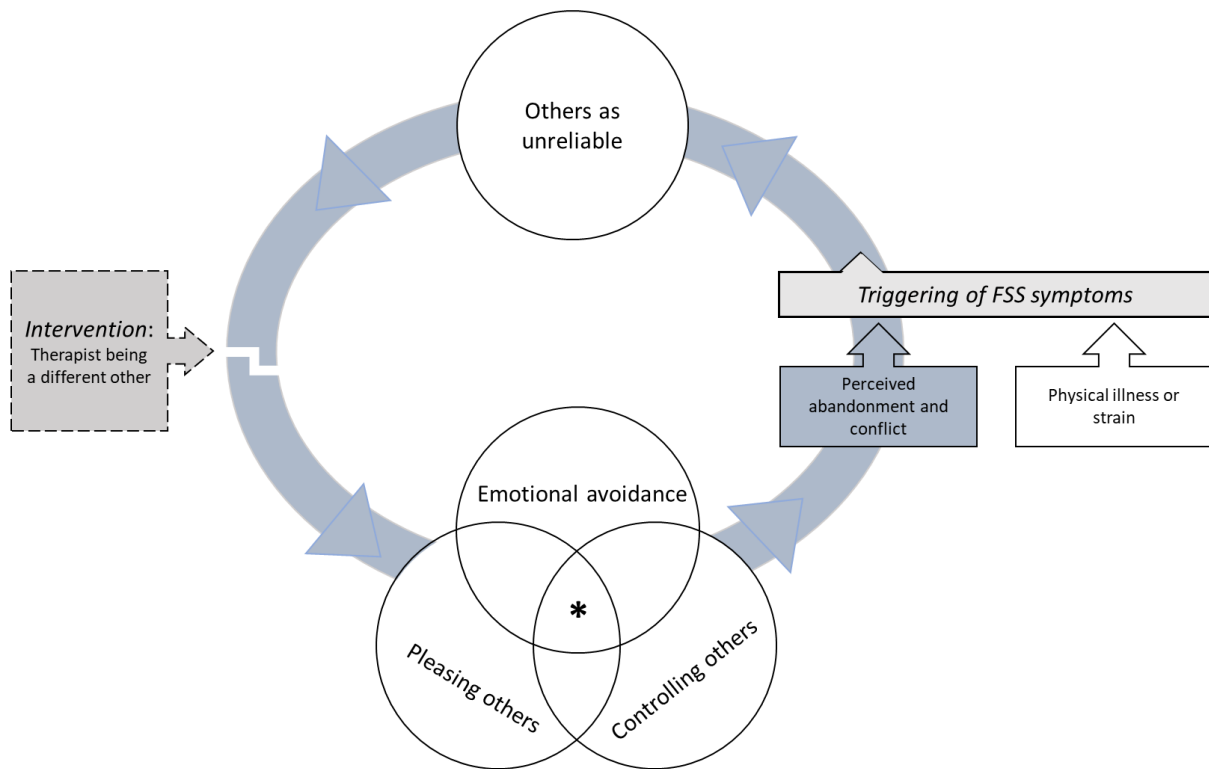


Figure 2. Synthesis of themes. The arrows indicate the suggested vicious circle resulting from the interpersonal dynamics. The overlap of three interpersonal themes (indicated by \*) marks the central pattern: *Patients' emotional avoidance in order to please and control unreliable others.*

A metasynthesis strives to go beyond mere thematic summary and aims to facilitate theory-building (Benoot et al., 2016; Thorne 2017). Also, the themes discussed above did not emerge in separation from each other but were strongly interconnected within cases. This leads to the identification of a model representing the overarching interpersonal dynamics in patients with FSS in our sample. In constructing this model, we complied with the logic of the “vicious circle” which has been described in several theories on FSS (see Introduction) and which could be recognized in several case studies. The presented “vicious circle” model does not imply that all elements were described in every case. Rather, it synthesizes information from different case studies as contributing inferences to the whole picture, which a single study cannot grasp. This understanding follows the metaphor of blind men touching the elephant, each discovering only one part of its body, whereas the reality may be more

complex and nuanced (Thorne, 2017). The resulting “vicious circle” model (Figure 2) will be discussed forthwith.

The vicious circle originates in early childhood relational experiences, in which significant others are perceived *as unreliable*. Patients report growing up in the midst of abandoning, overcontrolling, and overprotective others. In later life, these experiences can be repeated and generalized onto other contexts, such as romantic and professional relationships. In an attempt to adapt to others, who are perceived as unreliable, patients typically try *to please* them, for example by engaging in excessive caretaking, while at the same time neglecting their own needs. Next, this pleasing attitude is often accompanied by attempts to *control* unreliable others. The latter pattern is particularly salient in the therapeutic relationship, for example when patients do not disclose certain information to the therapist or attempt to dominate the session.

*Emotional avoidance* is a common characteristic of patients with FSS in the sample. They exhibit both diminished awareness of emotions and suppressed emotional expression. Other than a mere trait, emotional avoidance can also constitute *a strategy to please and control unreliable others*. In this way, the dynamics of pleasing others, controlling others, and emotional avoidance can be present in patients with FSS simultaneously (as indicated by \*, Figure 2). Despite being distinct in form, these patterns constitute a common interpersonal position in relating towards the unreliable other. Therefore, the sub-theme *emotional avoidance in order to please and control the unreliable other* builds a central theoretical link of the vicious circle model.

In some cases, patients are shown to experience FSS after suppressing negative emotions. More broadly, a triggering of FSS often occurs within the *interpersonal context*, especially in situations of *perceived abandonment and conflict*. In addition, physical stimuli such as (non-FSS) *physical illness* or *physical overexertion* may also trigger FSS. In these

cases, physical stimuli are not always related to the interpersonal dynamics and are thus placed outside of the vicious circle on the Figure 2. However, both interpersonal and physical triggers can lead to FSS onset or worsening, which subsequently contributes to the vicious circle. Subsequently, worsening of symptoms may cause others to either become more overprotective, and/or to repeat the abandoning and overcontrolling behavior towards patients, while remaining unresponsive to patients' emotional needs. In this way, the vicious circle persists and potentially leads to more rejection, more emotional avoidance, and more symptoms. The therapist can interrupt the circle by attempting to become a different (reliable) other to the patient. This can be done by fostering a secure space in which negative emotions can be expressed, explored, and acknowledged.

#### *Nuancing and divergent findings*

Besides contributing data to the themes, some case studies delivered partially divergent evidence. For instance, in three cases, patients reported being surrounded by mostly supportive significant others, while noticing that they still felt pressure not to overburden them [4, 13, 14]. Also, in one case, a patient developed pain with no identifiable trigger, however, later bouts of symptoms were connected to physical activity [14]. Divergent evidence could mean that besides early negative interactions with others, a combination of other factors (such as physical illness plus overprotective reaction by others) may be enough to set off the vicious circle in some patients.

Therapeutic orientation of the case studies could influence the reporting of at least three themes. For instance, the themes *Controlling others in the therapeutic relationship* and *Therapist becoming a different other to the patient* were identified more often in psychodynamic case studies. This is not surprising, since psychodynamic therapists pay special attention to countertransference and respective journals would be more welcoming of the discussion of its aspects. Likewise, many psychodynamic schools embrace the idea of

“corrective emotional experience” (grosse Holtforth & Fluckiger, 2012) which is in line with the principle of becoming a reliable other to the patient.

Similarly, the sub-theme *others as overprotective* has been identified more often in cases of CBT, which can be attributed to CBT’s particular focus on maintaining factors of sick role behavior (Witthöft & Hiller, 2010). Although we did not attempt a formal discourse analysis, it was our impression that in a sub-sample of cases with younger patients, terms like “separation anxiety” were more often used and brought into relation to other’s overprotective reaction. Here, the FSS in children was viewed rather in terms of “secondary gain.” These observations could be indicative either of differences in discourses between CBT and other approaches or point to heterogeneity in patients with FSS at different ages.

## Discussion

In this metasynthesis we attempted to examine interpersonal dynamics and the therapeutic relationship in patients with FSS. In following, we will discuss how the findings of our study can be situated within recent qualitative and quantitative research and current theoretical advancements in the field. One of the central findings of the metasynthesis is the notion of a relational function of emotional avoidance in patients with FSS (Coren, 2016). In our sample, emotional avoidance appeared to be more than a mere static trait. Rather, as expressed by the theme *Emotional avoidance in order to please and control the unreliable other*, it should be interpreted as an emergent relational strategy in dealing with unreliable others. This finding replicates the results of the qualitative interview study by Lind, Delmar, and Nielsen (2014), who found that patients with FSS grew up in a “*Culture of emotional avoidance*”. In this study, patients reported being punished by social exclusion and shaming for trying to show negative emotions to significant others. For example, children were “isolated in their bedrooms if they showed feelings of anger, frustration, or sadness.” (Lind et al, p. 96). Similarly, Town, Lomax, Abbass, and Hardy (2019) identified themes “*Global*

*emotional avoidance*” and “*Anxiety about the impact of emotions*” in interviews with patients with FSS. In this study, patients reported being afraid to express emotions due to anticipated negative reactions of others, or due to others’ indifference. These findings strongly correspond with experiences of several patients from our sample, for example with Jason’s struggle for acceptance by his family when expressing sadness (see Supplemental Material S4, Category 4b).

Another finding in this metasynthesis is the perception of *Others as unreliable* by patients with FSS. It is supported by the mixed-methods study by Landa, Bossis, Boylan, and Wong (2012), who discovered that “Unmet need for closeness with others” was the typical representation of relationships in patients with FSS (Landa et al., 2012, p.413). Also, in this study, the unmet need for closeness was the strongest predictor of somatization, which is corresponds with our sub-theme *Triggering of symptoms in context of perceived abandonment and conflict*.

Our vicious circle model is also supported by the recent experimental study by Erkie et al. (2018), who found that patients with FSS recognized their own emotions poorly but performed better than controls at recognizing negative emotions in others. At the same time, patients with FSS have also been shown to be distrustful of others. These findings do not appear paradoxical, in light of the interpersonal dynamics represented in our model: Patients with FSS could be avoiding their own emotions, while closely monitoring signs of negative emotions of mistrusted others. Both “not knowing” one’s own feelings and quickly recognizing threatening emotions in others may have been an adaptive relational strategy for patients in their early social environment.

Finally, our vicious circle model adds depth and nuance to recent attachment- and psychodynamic-informed theories of FSS. The notion of simultaneous pleasing and controlling others is in line with *attachment-hyperactivation* and *attachment-deactivation*

strategies in patients with FSS described by Luyten and colleagues (2012). Wherein, patients secure the attention of the unreliable others by means of extreme clinging or, on the contrary, act extremely pseudo-independent. Since the others do not provide a secure attachment base, such seemingly “extreme” interpersonal patterns appear to be most adaptive in childhood and may become generalized in later life. Indeed, it was our overall impression that reported interpersonal patterns were strikingly harsh and “extreme” in at least 13 cases. We identified instances of extreme compulsive caretaking, overcontrolling, and overprotection both from the side of the patient and from the others. Obviously, this might be a mere result of a selection bias, i.e., more “dramatic” cases receiving more attention, which in turn gives them a higher chance of being selected for publication. However, these findings do not contradict evidence from larger and more representative quantitative studies and could thus support the notion that early attachment disturbances may play a role in the etiology of FSS. As stated by one of the authors from our sample: “The risk is that you will think a case like this to be rare—it is not.” (Shapiro, 2003, p. 548).

Notably, similarly harsh interpersonal patterns have also been reported in environments of individuals with interpersonal trauma (Van Nieuwenhove, Truijens, Meganck, Cornelis, & Desmet, 2019). Due to high comorbidity of trauma and FSS (Afari et al., 2014; van Dijke et al., 2012), traumatic origins of emotional avoidance should be taken into account as possible etiological and maintaining factors of both conditions (Dobersch et al., 2018) and more transdiagnostic research, both quantitative and qualitative, is needed.

Subsequently, our findings may also help to nuance the role of *alexithymia* in FSS. We found that patients were sometimes consciously aware of the nature of their emotional avoidance as an optimal strategy to adapt to rejective, hypercritical, or outwardly dangerous others. Such patterns are distinct from the classical perception of alexithymia as a stable trait or even inborn “deficit” (Bronstein, 2011). This observation is supported by studies indicating

that classical alexithymia is found in a rather small subgroup within the FSS patient population (Gil, Scheidt, Hoeger, & Nickel, 2008). Moreover, severe alexithymia has been associated with interpersonal patterns marked by a general *indifference* towards expectations of others, rather than a tendency to please others (Vanheule, Desmet, Rosseel, Verhaeghe, Meganck, 2007). These inconsistencies uncover a need for exploring, both in clinical practice and in research, the interpersonal patterns underlying “alexithymia” in individual cases. Our findings also suggest the need for more mixed-methods research to clarify whether alexithymia constitutes a defining feature of FSS. It is possible that several interpersonal processes underly the phenomena of alexithymia and emotional avoidance in a therapeutic setting. Subsequently, a deeper understanding of interpersonal mechanisms can give rise to more transdiagnostic options to target seemingly different clinical populations, where similar mechanisms could be operating (Kazdin, 2007).

Overall, our metasynthesis illustrates how the expectations of others could shape patterns of emotional perception and communication. Targeting emotional avoidance has shown to be an effective and cost-reducing intervention in short-term psychodynamic treatments (Abbass 2015; Abbass, Lovas, & Purdy, 2008). From the patient perspective, directly targeting emotions in therapy of FSS has also been described as a main facilitating factor (Town et al., 2019).

### *Limitations*

The current metasynthesis represents a secondary analysis of published case studies. Hence, the question arises whether published case studies could be representative of the FSS population. Relying on the Single Case Archive as a method of systematic collection across journals of various therapeutic orientations, publication cultures, and a large time frame (1985-2016) increases the potential representativity of the sample. Also, the distribution of the basic demographic variables, specific diagnoses, and comorbidities in the sample



(Supplemental Material S1) is similar to the epidemiological and clinical characteristics of the FSS patient population (Henningsen et al, 2007). Still, the current research design neither allows for claims of representativity nor for claims that our findings are specific to the FSS-population. In addition, one major shortcoming of the sample is that it does not represent elderly patients. Overall, although the sample cannot claim to be wholly representative, it can still provide necessary diversity, which has been deemed a more crucial requirement for metasynthesis (Benoot et al., 2016).

Furthermore, within the current research design, we cannot address the problem of *specificity*, namely whether the detected interpersonal patterns are unique to FSS or could be found in other disorders as well (Blatt, 2004; Cornelis et al., 2017). It is likely that several themes from our metasynthesis represent universal vulnerabilities to psychopathology, such as growing up with unavailable and detached others (Sturge-Apple, Davies, Cicchetti, & Manning, 2012). Similarly, emotional avoidance is often viewed as a common risk factor for diverse psychopathology (Kashdan, Barrios, Forsyth, & Steger, 2006). For example, interpersonal emotional avoidance has shown to be particularly characteristic of patients with anxiety disorders (Newman, & Llera, 2011). Nevertheless, our findings illustrate comprehensively how those factors interact and unfold throughout patients' lives and therapies. They combine the circumstances of symptom onset and interpersonal dynamics into the "vicious circle model" which can be applied to patients with FSS and possibly also to other clinical populations.

### **Conclusion and Suggestions for Research and Practice**

This metasynthesis investigated interpersonal dynamics and therapeutic relationship, as well as the context of the symptomatic course, as described in case studies of patients with FSS. Our results show how, starting from the early experiences with unreliable others, patients with FSS may adopt a stance of pleasing and controlling others and exhibit emotional avoidance.

This can go hand in hand with FSS onset and worsening, which is typically reported in the interpersonal context. Additionally, physical stimuli are reported to trigger FSS, leading to an increased symptomatic burden. Others can react to patients' symptomatic burden in an overprotective or rejecting manner, continuing the vicious circle. The therapist can interrupt the vicious circle by becoming a different, more reliable other to the patient.

Methodologically, our metasynthesis is among the first attempts to fulfill the promise of the emerging field of systematic case comparison and to approach complex phenomena using cumulated data of published case studies (Fishman, 2017; McLeod & Elliott, 2011; Willemsen, 2015). In accordance with Flyvbjerg (2006), we could show that comparative analysis of case studies is capable of falsifying previous misconceptions. For instance, our results *do not* replicate a longstanding prototype of severely alexithymic patient with FSS from the literature (Bronstein, 2011).

Our findings suggest that the role of interpersonal dynamics in the etiology and treatment of FSS may need more attention in clinical practice guidelines. Although the German S3-guideline for health care providers has recently included a more elaborate view regarding the importance of integrating an interpersonal stance in the treatment of FSS patients (Roenneberg, et al., 2018), our study indicates that a more detailed guideline may be desirable in the future. For example, exploring possible traumatic interpersonal histories could provide useful inferences for maintaining good therapeutic alliance and avoiding possible pitfalls and impasses (Arnd-Caddigan, 2006). Also, frequent changes of health care providers and therapists (as they often occur in the managed care settings for FSS) appear less productive, as they risk repeating early negative experiences with unreliable or absent others and may result in a worsening of symptoms.

For counselors, who are often “first responders” for patients who develop FSS, it could be important to manage adverse interpersonal dynamics and be aware of early signs of a

problematic therapeutic relationship, such as overcompliance and emotional avoidance in sessions. Thus, the counselor's strength lies in supportive non-directive stance and in adopting holistic approach that surpasses reductionistic models of FSS (Hills et al., 2018). Such a stance could provide a good basis for becoming a "different other" to the patient (i.e., being emotionally present and vigilant, attentive to emotional expression and enduring in its absence) irrespective of therapeutic orientation. Behaviorally oriented practitioners could also profit from our findings when considering the patterns of pleasing others, controlling others, and emotional avoidance as reflecting patient's learning histories and targeting the resulting behavioral contingencies.

Future research should expand the application of metasynthesis methodology towards case studies from a broader range of clinical populations. For instance, populations with chronic (medically explainable) diseases, conversion disorders, anxiety disorders, or personality disorders would contribute a valuable contrast to current findings on interpersonal dynamics in FSS. Despite the limited generalizability of case study evidence in the traditional sense, such comparative metasyntheses would allow for additional valuable clinical inferences and theory-building. Samples of more homogenous case studies (e.g., stemming from mixed-methods randomized-controlled trials) could provide further insight into the interpersonal dynamics of different clinical populations and allow for testing of more specific hypotheses on interpersonal dynamics., e.g., from psychodynamic (Cornelis et al., 2017) or schema therapy domains (Henker et al., 2019).

Besides focusing on clinical populations, metasyntheses of case studies can provide rich transdiagnostic descriptions of possible underlying processes, such as emotional avoidance, (counter)transference, and various "vicious circles", where complex interactions between symptoms, social environment, and the therapist are present. This might support the

general movement away from a symptom-specific perspective and towards mechanism-specific approaches in counseling and psychotherapy research (Kazdin, 2007).

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# Supplemental Material S1

## *Characteristics of Case Studies and Patients Included in the Metasynthesis*

Nr	Case Study	Theoretical orientation	Study Type	Pseudonym	Age	Sex	FSS complaints (author's formulation)	Comorbidities (author's formulation)	Outcome
		(detailed)							
1	Allen, L. B., Tsao, J. C., Seidman, L. C., Ehrenreich-May, J., & Zeltzer, L. K. (2012)	CBT, Unified Protocol for the Treatment of Emotions in Youth with Pain (UP-YP)	Systematic case study	Marla	17	female	Whole body pain, fibromyalgia, functional abdominal pain with nausea and constipation, Irritable Bowel Syndrome.	Major Depressive Disorder, anxiety	mixed
2	Griffies, W. S. (2010)	Classical Psychoanalysis, Ego-Psychology	Clinical case study	Mr. W.	33	male	Fibromyalgia	No comorbidities reported	success
3	Shapiro, B. (2003)	Psychoanalytic psychotherapy, classical psychoanalysis	Clinical case study	Kai	13	female	Total body pain, Chronic Fatigue, Fibromyalgia.	No comorbidities reported	success

4	Taylor, R. R., Jason, L. A. (2002)	Empowerment-Oriented, Peer Counseling Group, Envelope Theory	Systematic case study	Tamisha	32	female	Chronic Fatigue Syndrome	No comorbidities currently, Past Major Depressive Episode in Full Remission.	mixed
5	Gold, J. R. (1995)	Eclectic: psychodynamically-informed with elements of CBT	Clinical case study	S.	mid-50s	female	Severe headaches, gastrointestinal distress, depression.	Depression, relational problems.	failure
6	Holland, P. (1997)	analytical psychotherapy, Winnicott, Jung	Clinical case study	Ruth	midlife	female	Chronic Fatigue Syndrome.	No comorbidities reported	success
7	Coren, S. (2016)	Eclectic: primary CBT, with psychodynamic-relational and existential elements	Clinical case study	Jason	late adolescent	male	Severe headache pain with photophobia and nausea.	Anxiety, depression	mixed
8	Lerner, P. M. (1990)	Object Relations	Clinical case study	Case example 2	38	female	Vertigo, headaches, nausea, intense	Depressive episode upon being diagnosed	success



							pain throughout her body, and continuous feelings of tiredness and depletion.	as "psychosomatic"	
9	Leung, P. W. L., & Sung-Chan, P. P. L. (2002)	Strategic reframing and culturally sensitive perspective	Clinical case study	Mrs. L.	32	female	Low back pain.	Depression, adjustment to life events	success
10	Krause, I. B. (1995)	Cross-cultural family therapy, culturally sensitive perspective	Clinical case study	R. (eldest son of Mahmuda family)	12	male	Severe headaches, missed the whole school year.	No comorbidities, relational problems in the family.	mixed
11	Blum, H. P. (2007)	Classical psychoanalysis	Clinical case study	no pseudonym	47	male	Frequent and intense somatization, diarrhea, gastric pain, spastic colitis, loss of	Cumulative trauma, disruptive behavior, narcissistic traits	success

							appetite, tension headaches. backaches.		
12	Hamberger, L. K., & Hindman, M. (2005)	CBT, solution- oriented, collaborative care approach	Clinical case study	Sue	53	female	Gastroesophageal reflux disease, irritable bowel, chronic pain syndrome, chronic fatigue syndrome, eczema, mixed headache (tension and migraine type) chronic sinusitis, weight loss.	Hypertension, kidney stones, knee injury and degeneration, chronic sinusitis, PTSD, insomnia, depression, Dysthymia, Complicated Bereavement	mixed
13	Vranceanu, A. M., Ring, D., Kulich, R., Zhao, M.,	CBT in orthopedic setting	Systematic case study	Jean	mid-40s	female	Idiopathic arm pain.	depression symptoms, anxiety symptoms	success

	Cowan, J., & Safren, S. (2008)								
14	Vranceanu, A. M., Ring, D., Kulich, R., Zhao, M., Cowan, J., & Safren, S. (2008)	CBT in orthopedic setting	Systematic case study	Laura	early 40s	female	Idiopathic hand and arm pain, headache, index finger twitching, fatigue, lightheadedness, dizziness, and heaviness in both arms.	no comorbidities reported	success
15	Chazan, S. E. (1997)	Psychoanalytic and developmental perspective	Clinical case study	Mimi	7	female	Recurrent headaches and stomachaches without physical cause.	difficulty sleeping at night, multiple fears, traumatic bereavement	success
16	Aubuchon, P., Haber J. D., &	Behavioral	Systematic case study	L.	26	female	Severe migraine headaches.	no comorbidities	success

	Adams, H. E. (1985)								
17	Szykula, S. A. (1987)	Child-focused strategic and behavioral therapy (behavioral and systemic)	Clinical case study	Ann	10	female	Extreme headaches and stomachaches, other somatic symptoms.	school anxiety, separation anxiety	success
18	Alvarez, A. (2009)	Psychoanalytic psychotherapy	Clinical case study	Luisa	15	female	Various psychosomatic problems, asthma, frequent acute abdominal pain without physical cause.	depression, severe separation anxiety, cystitis	mixed
19	Chu, B. C., Merson, R. A., Zandberg, L. J., & Areizaga, M. (2012)	Individual Behavioral Activation Therapy (IBAT)	Systematic case study	Alex	10	male	Multiple somatic symptoms, and chronic pain, motor weakness, back pains, poor ambulation.	depression, school refusal, Generalized Anxiety Disorder, Social Anxiety Disorder	mixed

20	Tasca, G. A., Mcquaid, N., & Balfour, L. (2016)	Psychodynamic-interpersonal group therapy	Systematic case study	Betty	52	female	Chronic back pain (leading to disability)	depression, history of sexual abuse	failure
21	Blaustein, J. P., & Tuber, S. B. (1998)	Psychodynamic-integrative	Systematic case study	Cindy	late 20s	female	Changeable, often vague set of symptoms, including rashes, dizziness, urinary tract infections, muscle aches, stomach problems, weight gain, fatigue.	anxiety, depression	mixed
22	Lauterbach, W. (1996)	Theoretical orientation of the study: Cognitive; of	Systematic case study	Tanya	42	female	Tension headaches (psychosomatic)	no comorbidities reported	success

		the therapy: Not mentioned							
23	Pucheu, S. (1998)	Psychoanalytic psychotherapy	Clinical case study	Joan	20	female	Vulvar burning syndrome (vulvodynia)	depression, candidiasis (fungal infection)	success

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*Note.* The ordinal number of case studies represents the chronological order of analysis. The categorization of study type and outcome was performed according to the IBISC manual of the Single Case Archive (Iwakabe & Gazzola, 2009; Meganck et al., 2017). Success is defined as a relief of most symptoms (in terms of what can be realistically expected for particular mental disorders); mixed outcome is defined as partial resolving of some symptoms, while other symptoms prevail; failure is defined as absence of an expected symptom relief.

## Supplemental Material S2

### *Case Studies Included into the Metasynthesis*

- Allen, L. B., Tsao, J. C., Seidman, L. C., Ehrenreich-May, J., & Zeltzer, L. K. (2012). A unified, transdiagnostic treatment for adolescents with chronic pain and comorbid anxiety and depression. *Cognitive and Behavioral Practice, 19*(1), 56-67.
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- Aubuchon, P., Haber, J. D., & Adams, H. E. (1985). Can migraine headaches be modified by operant pain techniques? *Journal of Behavior Therapy and Experimental Psychiatry, 16*(3), 261-263.
- Blaustein, J. P., & Tuber, S. B. (1998). Knowing the unspeakable: somatization as an expression of disruptions in affective-relational functioning. *Bulletin of the Menninger Clinic, 62*(3), 351
- Blum, H. P. (2007). Holocaust trauma reconstructed: Individual, familial, and social trauma. *Psychoanalytic Psychology, 24*(1), 63.
- Chazan, S. E. (1997). Ending child psychotherapy: Continuing the cycle of life. *Psychoanalytic Psychology, 14*(2), 221.
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- Gold, J. R. (1995). Case report of a failure in psychotherapy. *Journal of Psychotherapy Integration, 5*(2), 113.
- Griffies, W. S. (2010). Believing in the patient's capacity to know his mind: A psychoanalytic case study of fibromyalgia. *Psychoanalytic Inquiry, 30*(5), 390-404.

- Hamberger, L. K., & Hindman, M. (2005). Collaborative care of a patient with multiple, chronic physical and emotional health problems. *Clinical Case Studies*, 4(2), 139-159.
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- Shapiro, B. (2003). Building bridges between body and mind: The analysis of an adolescent with paralyzing chronic pain. *The International Journal of Psychoanalysis*, 84(3), 547-561.
- Szykula, S. A. (1987). Child-focused strategic and behavioral therapy processes. *Psychotherapy: Theory, Research, Practice, Training*, 24(2), 202.
- Tasca, G. A., Mcquaid, N., & Balfour, L. (2016). Complex contexts and relationships affect clinical decisions in group therapy. *Psychotherapy*, 53(3), 314.



Taylor, R. R., & Jason, L. A. (2002). Group intervention involving a client with chronic fatigue syndrome: A clinical case study. *Clinical Case Studies*, 1(3), 183-210.

Vranceanu, A. M., Ring, D., Kulich, R., Zhao, M., Cowan, J., & Safren, S. (2008).

Idiopathic hand and arm pain: delivering cognitive behavioral therapy as part of a multidisciplinary team in a surgical practice. *Cognitive and Behavioral Practice*, 15(3), 244-254.

# Supplemental Material S3

## *Distribution of Themes Across Theoretical Orientations and Number of Case Studies Contributing to Themes*

Nr	Theoretical orientation	<i>Others perceived as unreliable (N=22)</i>	<i>Pleasing others (N=14)</i>	<i>Controlling others in the therapeutic relationship (N=7)</i>	<i>Emotional avoidance (N=19)</i>	<i>Triggering of FSS symptoms in the interpersonal context (N=15)</i>	<i>Triggering of FSS in the context of physical illness or strain (N=11)</i>	<i>Therapist being a different (reliable) other to the patient (N=7)</i>
1	CBT	+		+	+	+	+	
2	Psychodynamic	+	+	+	+	+		+
3	Psychodynamic	+	+	+	+	+	+	+
4	CBT	+	+				+	
5	Eclectic	+	+		+			
6	Psychodynamic	+		+	+	+	+	+
7	Eclectic	+	+		+	+	+	+
8	Psychodynamic	+	+		+	+		
9	Systemic	+	+	+	+	+	+	
10	Systemic	+			+	+		+
11	Psychodynamic	+		+	+	+		
12	CBT	+	+		+	+		
13	CBT	+				+	+	
14	CBT	+	+		+		+	
15	Psychodynamic	+			+			+
16	CBT	+			+		+	
17	CBT	+	+			+		
18	Psychodynamic	+	+		+	+		
19	CBT	+			+		+	
20	Psychodynamic	+	+	+				+
21	Psychodynamic	+			+	+		
22	CBT		+		+			
23	Psychodynamic	+	+		+	+	+	

*Note:* The ordinal number of case studies represents the chronological order of analysis from Appendix 1.

## Supplemental Material S4

### *Themes, Sub-Themes, Number of Case Studies Contributing to the Themes, and Representative Quotes*

#### **Category 1: Others perceived as unreliable (N=22)**

##### **(a) Others abandoning and unavailable**

*Early in treatment we began to explore what it felt like for Jason to experience intense feelings of separation, isolation, and loneliness from his family and peers. [...] Jason also experienced what he described as considerable frustration and a lack of empathy from his father and siblings, whom he felt lacked the desire to better understand his emotions and pain. [7]*

##### **(b) Others controlling, demanding, and prescribing identity**

*[Husband was perceived by patient] as a greedy, stingy, extraordinarily critical, and uncaring spouse and parent who expected total devotion, self-sacrifice, and self-denial from his wife and two (now adult) sons. [Patient] reported that she had no time or ability to have her own friends or to attend to her own interests and needs, as her husband expected her to be at his beck and call 24 hours a day. Despite his considerable wealth, he chastised her for every cent that she spent, yet he refused to allow her to get a job. [5]*

##### **(c) Others overprotective**

*Her parents also state that they were “probably very over-protective” [...] “L. avoided housework, meal preparation, working a job, socializing and sexual activity when pain occurred. Domestic duties were completed by her parents and husband. Family conversations often centered on the client’s pain. [16]*

#### **Category 2: Pleasing others (N=14)**

##### **(a) Pleasing others**

*[...] involuntary caretaking activities [...] had become central to almost all of her relationships [...]. Her family and friends had come to take for granted her seemingly endless ability to give [...] [5]*

**(b) Pleasing the therapist**

*His attention was on making himself perfect for me [therapist], not on discovering his authentic mind/body self. [...] [Patient:] I feel like I'm faking it. Like I'm attempting to playact the emotion in accordance with what I think you [therapist] think I should feel. But it's not connected to my gut. It's all being conjured up in my head.[2]*

**(c) Self as high achiever**

*Tamisha has a history of academic achievement, athleticism, and volunteerism beginning in grammar school and continuing through college. She received a scholarship to study business administration at a major university and obtained her most recent job as the general manager of sales soon after graduating from college. She describes herself as a "goal-oriented" person [...] In general, Tamisha consistently planned more activities than she was able to accomplish and undertook more activities than she had energy for. [4]*

**Category 3: Controlling others in the therapeutic relationship (N=7)**

**(a) Controlling the session setting**

*Session after session, as Ruth focused on her symptoms and her meticulous physical regime, conserving her energy by 'scrupulous preparedness' and careful 'budgeting', our relating felt carefully paced, even predictable.[...] Ruth never did use the couch; I felt she needed me to be very visible, though this rarely involved eye contact [6]*

**(b) Controlling the therapist by non-verbal means**

*[...] whenever I moved, Mr. W. perceived me as rejecting or abandoning him. [...] I was feeling constrained by him. He needed me to remain perfectly still to the extent that I felt straitjacketed in my chair, and the sense of being bound angered me.[2]*

#### **Category 4: Emotional avoidance (N=19)**

##### **(a) Restricted awareness and expression of emotions**

*Marla struggled with the concept of being aware of her emotions, as she would often react to the experience of any negative mood (i.e., anger, frustration, hopelessness) by withdrawing and disengaging from the conversation [1]*

##### **(b) Emotional avoidance in order to please and control the unreliable other**

*Ever since my first surgery, I have been depressed, but I have to be happy. I can't be sad or else no one will want to be around me. [...] Jason's father communicated harsh expectations that Jason should "be a man" and keep feelings of vulnerability, dependency, and sadness to himself. [...] Jason's restricted expression of affect shielded his mother from additional worry and protected him from his father's rage, which he experienced as rejecting and humiliating. [7].*

#### **Category 5: Triggering of FSS symptoms in the interpersonal context (N=15)**

##### **(a) In context of perceived abandonment and conflict**

*In fact, it was often precisely at the times that Cindy felt abandoned—or smothered—by important people in her life that she would report more intense somatic distress. [21]*

##### **(b) In context of therapy sessions**

*He often had tension headaches and backaches, which were more common around his analytic hours.[11]*

#### **Category 6: Triggering of FSS symptoms in the context of physical illness or strain (N=11)**

**(a) In context of physical illness**

*Near the end of treatment, Marla was diagnosed with pneumonia and her pain levels, as well as depression, increased dramatically [1]*

**(b) In context of physical strain**

*She stated that pain started while on her retail job, when she was trying to put a 30-pound box on a shelf. She noted that she immediately experienced pain in the forearm. Pain increased in intensity during the following few days, and she went to the emergency room where X-rays showed no evidence of a fracture or dislocation. [13]*

**Category 7: Therapist being a different (reliable) other to the patient (N=7)**

*Slowly, my consistent response to his thoughts and feelings as credible and meaningful started to facilitate mentalization. [...] He indicated that he was taken aback by these kinds of interactions; he expected me to make proclamations about these matters, like his father, who had constantly told him that the law was the only profession for him, or that he was an idiot if he didn't marry a particular kind of woman or live in a particular part of the country. [2]*

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*Note:* The numbers in square brackets indicate the number of the case study in which the theme has been identified from the Appendix 1 and 3.

