

Perspectives on the clinical significance of functional pain syndromes in children

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Abstract: Functional pain syndromes (FPS) characterize a subset of individuals who experience pain and related symptoms and disability without clear structural or disease etiology. In the pediatric population, FPS hold high clinical importance due to significant prevalence rates and potential to persist into adulthood. Although extensive research has been executed to disambiguate FPS, the syndromes that fall within its spectrum remain conceptually complex and sometimes ill-defined. This paper provides an overview of available research on the classification and multifaceted etiology of FPS in youth and their effects on interpersonal, psychological, and familial function. Vital aspects of a successful multidisciplinary approach to treating this population are described; however, it is evident that future research requires more longitudinal studies.

Keywords: overlapping chronic pain, functional pain, primary pain disorders, pediatrics, biopsychosocial model

Introduction

Functional pain syndromes (FPS) are a diagnostic group of conditions that remain loosely defined. Pain is often a warning signal from the body indicating an abnormality in structure or function. However, there is a subset of patients who experience pain and are subject to symptoms, suffering, and disability with unclear disease etiology or biomechanical cause. Many patients who experience FPS also experience clusters of symptoms that can affect a wide array of systems, often due to a combination of varying physiological, affective, and cognitive influences.¹ As a result, physicians have the difficult task of labeling a cluster of symptoms that do not neatly fit into a disease-driven diagnostic category and implementing a treatment plan that at times relies on speculation. This is due to several reasons: 1) these diseases often present with a wide variety of symptoms and so the diagnostic criteria are vague and constantly under revision;²⁻⁶ 2) these diseases arise from a complex mix of biological, psychological, and interpersonal factors;⁷⁻¹⁰ and 3) these diseases often share similarities with each other and it can be difficult to make a definitive distinction,¹¹ and frequently co-occur, making definitive distinction difficult. As a result, these diseases can often be misdiagnosed, especially in a pediatric population.¹²

An investigation conducted by the Pain of Unknown Origin in Children Study Group one decade ago supports this premise. The study assembled a panel of 17 Dutch pediatricians who work in psychosomatic medicine and presented them with the medical reports and standardized psychiatric assessment of 134 children who had FPS for more than 3 months.¹³ All the participating pediatricians were asked to use

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the information to diagnose the children, with a consensus being defined as agreement among more than 80% of the pediatricians. Consensus for the underlying cause was only found for 47% of the cases, and consensus regarding the appropriate diagnostic approach was reached for 63% of the cases.¹³ Further work needs to be done on developing a diagnostic procedure that is reliable and encompasses most if not all FPS.¹⁴

Lack of a common language hampers clinical and scientific progress

Too often, a patient receives a diagnostic label or labels that reflect the specialist's area of expertise as opposed to the patient's overall experience. For example, a patient with excessive bowel symptoms might present to a gastroenterologist who diagnoses her with irritable bowel syndrome (IBS), whereas a patient who has widespread muscle pain and tenderness may go to a rheumatologist who diagnoses him with fibromyalgia.¹⁵ A recent study showed that these different conditions may actually reflect a common entity but have labels that reflect the specialists' area of expertise.¹⁵ The most common diagnoses that reflect this functional overlap in pediatrics include headache, functional gastrointestinal disorders (FGIDs), fibromyalgia, and chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ME).^{15,16} The involvement of organ-system specialists in evaluating and diagnosing these complex chronic pain conditions mean that very little research has been done across disciplines to determine if these conditions are truly different or do in fact reflect a common entity.

Research into the field of FPS is further hampered by the fact that the same symptom cluster is associated with various diagnostic names, for example: somatoform disorder,¹⁶ somatization,¹⁷ functional somatic syndrome,¹⁸ bodily distress disorder,¹⁹ central sensitization syndrome, amplified pain syndrome, and primary pain disorder.²⁰ As a result, a literature search into the area of FPS is very disjointed and requires use of a wide variety of keywords. To further complicate matters, the term "somatoform disorder" was introduced by the *Diagnostic and Statistical Manual of Mental Disorders-III (DSM-III)*.²¹ However, the *DSM-III* handling of somatoform disorders focused primarily on somatic symptoms caused by psychological rather than biomedical factors. The publication of the *DSM-IV* expanded the definition of somatic symptoms to include those "not fully explained by a general medical condition", thereby removing the etiologic divide between psychological and

biomedical factors. The classification still had many flaws.²² *DSM-V* has changed the term to somatic symptom disorder, a name that had not been previously misused. In this iteration of the diagnosis, the manual's authors completely removed any distinctions between medically explained and medically unexplained somatic symptoms.¹⁹ However, the classification includes somatization in the context of other diseases (eg, cancer and diabetes), so it may not be the most optimal term for uniquely capturing FPS.¹⁹ To further complicate matters, the fact that somatic symptom disorders are included in the *DSM* at all is controversial – some doctors think that these disorders have a biological cause that has not been identified yet, while others think that the disorders have a purely psychological cause.¹⁹

What's in a name?

The label used for these diseases matters because it affects perception by patients, doctors, and the public alike. A cohort of CFS patients were asked to choose their three most preferred terms from a list of commonly used names for FPS.²³ The most endorsed term was "persistent physical symptoms", followed by "complex physical symptoms" and "medically unexplained physical symptoms", reflecting a desire to stress the physical nature of these diseases.²³ Another study surveyed 844 members of the general public on their most preferred clinical term for FPS, and the most commonly selected names were "persistent physical symptoms" (20%), "functional symptoms" (17%), and "medically unexplained symptoms" (15%).²⁴ Even the name of each individual FPS disease matters. Medical trainees presented with CFS patients and prompted with either CFS, ME, or Florence Nightingale disease attributed different causes and prognoses to the patients depending on which name they were provided in the study.²⁵ Another similar study on the labeling of CFS showed similar results.²⁶ Terminology and diagnostic criteria for the condition also affect how people are diagnosed. For example, 2,674 adolescents were evaluated with *DSM-III* somatization disorder criteria. The prevalence was too low in the 4- to 11-year-old group for a reliable conclusion, but in the 12- to 16-year-old age group, 11% of girls and 4.5% of boys fit the criteria.²⁷ A later study then used the *DSM-IV* somatization disorder criteria with a group of 14- to 24-year olds, and found that the prevalence was only 2.7%, but significantly more of the same respondents met criteria for clinically significant FPS, resulting in a prevalence rate of 12.6%.²⁷ It is clear that the terminology of these diseases, both for the individual diseases and the overarching term for FPS, still need to be clarified and reworked.²¹

FPS by the numbers

The issue of pediatric FPS is not one to be taken lightly because it affects a significant patient population. Exactly how many patients struggle with this issue is unknown because little work has been done on the epidemiology of all FPS. King et al²⁸ reported in a meta-analysis that 11%–38% of children have chronic pain. It has been estimated that a range of 0.03%–1.29% of children suffer from ME/CFS.⁶ Pediatric weekly headaches are reported in 10%–30% of children and are the reason for 1%–2% of pediatric outpatient visits.¹⁶ A cross-sectional study of Norwegian adolescents showed that the 1-year prevalence rate for tension headache was 58%, and the point prevalence for any kind of headache was 38%.²⁹ Functional or recurrent abdominal pain is reported in 7%–25% of school-aged children and is the reason for 2%–4% of pediatric visits.¹⁶ Pediatric fibromyalgia has been reported anywhere from 1.2%–6.2% of children.² Eminson²⁷ reported that studies done on FPS in various countries showed 6%–20% of pediatric patients complaining of somatic symptoms. These prevalence rates are associated with significant health-care expenditure. It has been estimated that the average annual costs associated with FPS in the US total over \$11,000 among some adolescent patients.³⁰

Theories to explain FPS

FPS can be caused by a myriad of physiological/biological and psychological influences.¹ The biology behind FPS is still poorly understood, and no work has been done on differences between children and adults. As a result, the theories that will be underlined below apply to the development of FPS in the general population. Some work has been done on understanding the mechanisms underlying specific FPS. For example, a recent study found that fructose intolerance/malabsorption can contribute to the pain symptoms of FGIDs,³¹ and another study found that more than half of a sample of 41 pediatric patients with chronic widespread pain showed signs of small-fiber polyneuropathy.³² However, a few unifying theories on the mechanisms behind all FPS have been proposed and are outlined below. It is generally considered that patient symptoms can be understood through the lens of one or more of these theories, with some relying more heavily on the biological/physiological substrates of the condition, whereas others highlight the cognitive-affective contributors.

Neuronal sensitization theory

Patients who have repeated sensations of pain can develop memory traces on a neuronal level that increase sensitivity to further sensations.³³ In these cases, a typically benign

sensation can be interpreted as pain due to the neuronal memory traces. Over time, the memory trace can rewire larger portions of the brain and increase its range until a symptom memory matrix has been sensitized, and several physical symptoms can be triggered at once with increasing intensity.³⁴ Hypersensitivity has been shown in children with FGIDs, Crohn's disease, fibromyalgia, and chronic fatigue.^{35–37}

Immune system sensitization theory

The body can trigger an immune response to danger signals and pathogen-associated molecular patterns. The resulting release of cytokines that normally tells the brain to change the focus of its priorities can lead to increased sickness behavior, metabolic alterations, and malaise.³⁸ This cytokine-induced sickness behavior can persist when the innate immune system is chronically activated, and prolonged activation of this system can restructure a patient's physiology and induce mood disorders.³⁹ These cytokines can also amplify the perception of pain due to the effect that proinflammatory mediators have on the central nervous system, thereby increasing pain, sleep disturbance, and fatigue.⁴⁰

Endocrine dysregulation theory

Dysregulation of the hypothalamus-pituitary axis' (HPA) ability to regulate the body's response to stress has been found in patients with FPS. Studies have shown that many patients with FPS have cortisol irregularities.⁴¹ A theory for the dysfunction of the HPA is that prolonged activation creates a "burnout" effect and downregulates the HPA chronically, leading to hypocortisolism.³³ Another theory is that the hypocortisolism is a protective mechanism.³³ A final theory proposes that the HPA fluctuates in activity, with hypoalgesia during acute stress and hyperalgesia during chronic stress.⁴¹ Therefore, it is clear that the axis does play some role in FPS, but it is not yet fully understood.

Signal filter theory

The body constantly sends the brain sensory information, but the signals are filtered to protect the brain from being overstimulated by information. In patients with FPS, it has been proposed that a malfunctioning filter causes the patients' brains to become overloaded with endogenous and exogenous information and increases the number of physical sensations that the patient experiences.³³ Rief and Broadbent³⁴ proposed that factors that can decrease filter activity include selective attention, infections, health anxiety, depressive mood, and lack of distraction.

Somatosensory amplification theory

The concept underlying this theory is attention. A patient who experiences a physical sensation and focuses his/her attention on this sensation develops certain behaviors and cognitions that amplify his/her perception of the sensation.³³ The amplification feeds itself back into a cycle of enhanced attention and pain. There are limitations to this theory, however. The original theory focused on the experience of hypochondriasis, not patients suffering from FPS.³⁴ Additionally, while there has been some work with children indicating that somatosensory amplification increases the risk of some pains but not others (eg, back pain but not headache in adolescent girls⁴²), most researchers agree that amplification can only explain a portion of the development of FPS.⁴³ Along similar lines is the abnormal proprioception theory, illustrated in a study by Scholz et al⁴⁴ that showed that patients with FPS had more precise but not necessarily more intense proprioception. The implications of this are still not well established, but Rief and Broadbent³⁴ interpreted it to suggest that patients with FPS perceive physical sensations more precisely, so that normally minor physical symptoms could be perceived as more amplified and threatening.

Autonomic nervous system dysfunction

Following a period of distress or attention, a healthy body enters a rest state that is accompanied by a decrease in heart rate associated with the parasympathetic nervous system (PNS).³³ However, in patients with FPS, there is evidence that the PNS is dysregulated and is unable to reduce the heart rate during the rest state, thus leading to chronic elevated heart rate and stress.³³ This association has been shown in children with FGIDs,⁴⁵ but not in a general pediatric patient population with FPS. Interestingly, this association may change with age, because a study found that FPS is associated with decreased PNS activity in patients aged ≤ 52 years, whereas patients with FPS aged > 52 years had increased PNS activity.⁴⁶

Illness behavior theory

The concept behind this theory is that a patient's beliefs influence his/her behavior, which can then influence the physiologic response. The fear avoidance model is a conceptualization of this theory where fear and avoidance of activities due to pain result in worsening biopsychosocial functioning. It has been validated for pediatric chronic pain,⁴⁷ but not specifically for FPS.

Sensitivity theory

Departing from the sensitization theory, this theory proposes that some patients are vulnerable to developing FPS based

on underlying genetics. There has not been much evidence for genetic predisposition, but there is evidence that genetic variants can lead to variability in inflammation, which would alter the degree to which immune system sensitization could contribute to the development of FPS.⁴⁰ Genetic variability can also change the stress response in an individual, altering how endocrine dysregulation and PNS dysfunction can affect FPS. Additionally, there is evidence that when a pregnant female is exposed to a significant stressor, the resulting stress response in her body can cross the placenta and alter the fetus's brain, endocrine, nervous, and immune function.⁴⁸ However, these mechanisms are still largely unexplored. Some other possible predisposition theories attribute FPS to preexisting personality traits, such as self-protective organization and insecure attachment, as well as environmental factors, such as abuse, parental transmission, and stress.³³ These factors will be explored later.

Biopsychosocial model

Incorporating elements of several theories described above, the biopsychosocial model proposes that FPS is a self-perpetuating cycle that involves interacting factors from physiological, cognitive, and affective domains. It also takes into account the patients' cognitions as well as any external factors that would predispose or facilitate FPS.³³ As a result, the biopsychosocial model provides a cohesive approach to FPS. Cognitive behavioral therapy (CBT), a psychological treatment approach based on this model, is validated and utilized in treating children with FPS and is generally combined with exercise and certain pharmacological approaches.⁴⁹

Despite the progress made in identifying the above mechanistic theories, the FPS pediatric population remains widely heterogeneous, making classification for research and treatment purposes extremely difficult. Similar barriers have been recognized within the pediatric headache population.⁵⁰ Connelly and Bickel⁵⁰ suggested employing the use of a bottom-up approach with large sample, population-based data in order to further identify typical mechanistic pain patterns and characteristics. Utilizing this approach for FPS could provide benefit by narrowing the focus on possible underlying mechanisms and facilitate categorization for future research studies.

The familial context in FPS

Beyond genetic and biological vulnerabilities that are present at birth, there are several internal and external psychosocial factors that have been hypothesized to exacerbate or potentially increase the likelihood of the development of FPS.

For example, insecure attachment in children has been shown to lead to somatization, especially in adult patients.^{51–53} No work has been done yet on pediatric patients with FPS and their attachment styles, but Kozłowska⁵⁴ recently proposed an intervention for children with FPS that addresses attachment.⁵⁵ Self-protection is related to attachment. Children can react to threats from close relationships by either an inhibitory, excitatory, or balanced emotional response.⁵⁶ Children tailor their reactions according to how a parent reacts to their distress. If parents fail to respond, children develop an inhibitory display of their distress. If parents are “predictably unpredictable”, the child will tend to display their distress in exaggerated ways. Parents who respond predictably and in comforting ways lead to a child who displays attachment and distress signals that are appropriate to the situation. Kozłowska and Williams⁵⁶ found that children who demonstrate inhibitory or excitatory self-protective mechanisms are at greater risk for FPS. Anxiety has also been shown to have a significant association with development of FPS; in particular, children with panic disorder and separation anxiety disorder are at higher risk of somatization.⁵⁷ Additionally, increased stress induced by a struggle to control people around us (agonistic striving) is significantly associated with increased somatization, and the association is especially apparent in people who have low pain tolerance.⁵⁸ This association is seen in both a pediatric and adult population.

FPS can also be induced by exogenous factors. Children who have parents with chronic pain have been shown to have significantly increased insecure attachment, internalizing and externalizing, and poor biopsychosocial functioning.^{59,60} Although this association has not been directly linked to the development of FPS, these are all precipitating factors that increase a child’s sensitivity to developing FPS. Parents with chronic pain who catastrophize about their own pain also tend to have overprotective responses to their child’s pain and increase somatization in their children with IBS.⁶¹ While there has been some discussion about early childhood abuse causing FPS, there has been no evidence to support this claim.^{62,63} There is evidence that maladaptive parenting behaviors can contribute to somatization. For example, paternal rejection and/or hostility are correlated with increased somatization in IBS patients.⁶⁴ Lastly, family life as a whole can influence the development of FPS. Family disorganization, poor family cohesion, and families less interested in political, social, intellectual, and cultural activities predict development of somatic complaints in adolescent girls in particular.⁶⁵

One last potential cause of pediatric FPS that must unfortunately be accounted for is Munchausen’s syndrome

by proxy, otherwise known as fabricated or induced illness, as well as Munchausen’s syndrome. Caretakers have been known to induce or fabricate illness, such as abdominal pain in their children.⁶⁶ Potential motivations for this behavior include extreme illness anxiety, confirmation that the child is indeed ill, material gain, maintenance of a close relationship with the child, a need for attention, and deflection away from a child’s behavioral problems.⁶⁶ Not only does this parenting behavior subject children to unnecessary hospital visits and potential additional serious harm, it has also been shown to decrease biopsychosocial functioning in these children.⁶⁶ However, Munchausen’s syndrome is also seen in some children. Patients can induce symptoms by injuring themselves, disconnecting medical equipment, switching patient charts, and interfering with test samples.⁶⁷ While this behavior is most common in women aged 20–40 years old, it has been reported in children as young as 8 years of age.⁶⁷ Jaghab et al⁶⁷ outline several cases of pediatric Munchausen’s syndrome and the implications of the behavior. Both caregiver and patient illness fabrication should be potentially considered when diagnosing symptoms that seem inexplicable. Some even posit that it is possible for Munchausen by doctor to evolve, which reflects a continued search for physiologic aberration by physicians reinforcing the perception there is an underlying treatable and undiagnosed biological explanation.

The development of FPS, therefore, seems to be attributable to a combination of several biopsychosocial factors. However, there is a serious need for further exploration into these factors, their implications, and their contribution to FPS as a whole. Just as there needs to be consensus on the language we use to talk about all FPS, we need to develop a comprehensive view of the onset and maintenance of all FPS.

The impact of FPS

Unfortunately, the complexities of FPS do not stop with unclear diagnostic terminologies and causal factors. The complex challenges that patients with FPS experience are numerous and often difficult to discern from patients with a pathophysiological explanation for their pain. Youth with chronic pain experience decreased quality of life and family disruptions,⁶⁸ disturbed sleep and inability to pursue hobbies,⁶⁹ school absenteeism and reduced socialization with friends,⁷⁰ and increased functional disability.⁷¹ When specifically examining children and adolescents with FPS, the literature is sparse, but does indicate that youth with FPS experience significant decrease across most domains of life in

comparison with healthy peers and even peers with organic disease.^{72–74} In examining the unique experience of youth with FPS, three prominent themes emerged: 1) the perception of others, 2) impact on the family, and 3) psychiatric morbidity.

Perception of others

Peers

Children and adolescents with FPS do appear to face distinct difficulties socially. Guite et al⁷⁵ revealed that healthy children reported greater liking of peers with pain conditions of an organic etiology and perceived their symptoms as more severe than peers having pain symptoms without organic disease. Additionally, adolescents with an FPS disorder, such as fibromyalgia, were perceived by healthy peers as being more sensitive/isolated and having significantly fewer friendships.⁷⁶ It is likely that youth affected by FPS syndromes recognize the negative perceptions held by their peers. It has been demonstrated that adults with functional somatic syndromes report higher levels of perceived stigma when compared with a matched medical group;⁷⁷ further research is needed to examine this same phenomenon in children.

Medical personnel

In addition to negative perceptions held by peers, children with FPS are impacted by views from medical staff. In some cases, only a small percentage of school nurses believe recurrent abdominal pain to be a serious disease and suspect that children presenting to their offices with this condition are lazy, faking, or seeking attention.⁷⁸ Similarly, when interviewed about views on adults affected with IBS, doctors often express frustration and can point out negative characteristics of these types of patients, while the patients themselves feel discredited and trivialized.⁷⁹ Very little research is available to determine if children with FPS hold similar views on the medical encounter as their adult counterparts. However, pediatric patients have recalled times when the doctors dismissed their pain when no evidence for an organic cause for the pain was found; furthermore, parents have described feelings of being judged and perceived medical professionals to be suspicious of the pain.⁸⁰ Parents of youth with FPS have also previously demonstrated lower physician satisfaction when given a functional diagnosis for their children's pain symptoms versus parents given an organic diagnosis.⁸¹

To compound the problem, the treatment providers are often struggling themselves to adequately address the needs of patients with FPS. It is not uncommon for physicians to feel “drained” by these patients due in part by not meeting

their needs and expectations as care providers.⁸² Physicians frequently struggle to explain the ambiguous and complex nature of symptoms resulting from FPS to their patients;⁸³ one can only imagine how this is compounded when addressing the patient and his/her parents. This topic warrants future investigation.

Impact on families

Given that children and adolescents live within a family system, the consequences of their pain typically extend across all family members, particularly parents. Notably, much of the available research represents the views and experiences of mothers.^{84–91} More exploration is needed in this field in order to understand the perspectives of the fathers of children and adolescents with FPS and determine if/how they differ from the mothers' perspectives.

Current research has established that mothers of children with FPS report significant effects on work and family life, including decreased productivity and increased conflicts in the household due to their children's pain.⁸⁵ Not surprisingly, Lipani and Walker⁸⁶ demonstrated that maternal worries, limitations in activities, and time constraints are related to pain severity in children with FPS. On a more critical level, the majority of parents report clinically significant scores on measures of distress, anxiety, and depression in a recent study.⁹¹

Increases in maternal distress may, in part, be explained by parental hardship in dealing with the ambiguity of FPS in their children and the search for some sort of identity or “illness label” to authenticate the symptom(s) for the child, family, and school.⁸⁵ This frustrating and exhausting process has been described as a “struggle for control and coherence” and torturous knowing that, as a parent, one cannot make the pain go away.⁹² In addition to this, mothers describe a feeling of responsibility for the pain and threat to their parental role.⁸⁴ Direct correlations have been found between perceived parental responsibility for the child's pain and poorer family functioning/parenting stress.⁸⁸

Van Tilburg et al⁹³ have touched on two central conflicts reflecting the cognitions of parents' of children with FPS: pain is real and the desire for care. Parents have struggled with this idea of “real” pain, because many times, they have seen their children experience obvious physical symptoms, but are also able to recognize the contribution of complex psychological factors. No matter the contributing factors, though, parents of children and adolescents with FPS increasingly act on their desire for care. Connelly et al⁸⁹ determined that health care utilization might be driven, at least partially, by

parental perceptions of high child vulnerability. It has also been shown that extensive medical investigations by means of laboratory tests and imaging studies are not pursued entirely for the reassurance of the parents of children with FPS; doctors need the comfort as well.⁹⁴ And since research has identified child FPS can persist into adulthood,^{95–97} it is likely that health care use will remain high for these patients whose pain continues without a clear diagnosis or etiological understanding.

Psychiatric morbidity: another layer of complexity

Despite no clear directional evidence regarding the relationship between pediatric FPS and psychiatric symptoms, psychiatric morbidity is quite common in this population. It has been determined that nearly 35% of a pediatric FPS sample had a clinically relevant psychiatric disorder, the most frequent being anxiety, affective, and disruptive disorders.⁹⁸ The prevalence and type of psychiatric disorder (ie, internalizing, externalizing) seem to vary with type of pain and patient sex. Egger et al⁹⁹ found that female children and adolescents who reported primarily stomach pain were more likely to also have a co-occurring anxiety disorder, while having musculoskeletal pains was more associated with depression. On the other hand, male children and adolescents were more likely to exhibit disruptive disorders, such as attention-deficit/hyperactivity disorder and oppositional defiant disorder, in association with stomach pain. This discrepancy between the sexes reflects general prevalence rates with higher rates of internalizing disorders among females and higher rates of externalizing disorders among males. Overall, however, functional abdominal pain patients seem to present with alarmingly high rates of psychological disorders.¹⁰⁰ Future research should determine why this population is particularly vulnerable to psychiatric difficulties. Additionally, psychiatric conditions co-occurring with FPS in the pediatric population is a relevant issue in other cultures. For example, Imran et al¹⁰¹ recently determined that Pakistani children with FPS also present with more anxiety and depressive symptoms than children with diagnosed medical conditions and healthy controls.

Treatment options for FPS

Taking into account the high prevalence rates and unique, challenging impacts FPS has on youth and their families, it is critical to identify treatment options for this population. Due to the uncertain nature of FPS and the varying phenotypes,

uniform recommended treatment has not been established. Currently, more well-designed longitudinal studies are needed to determine the exact prognoses of FPS treatment options. With this being said, addressing FPS, and chronic pain in general, with a multidisciplinary approach proves to be one of the most popular intervention strategies and has demonstrated much success in both inpatient and outpatient settings.^{102,103} The multidisciplinary approach acknowledges the totality of the systems that interact during an individual's experience of pain, including biological, psychological, social, and environmental factors.¹⁰⁴ It should also be rehabilitative in nature, which reframes the goal of the treatment process from “curing” to “coping” with the pain problem and puts responsibility for treatment success in the hands of the patients.¹⁰⁵ Instead of focusing on removing the pain, the ultimate goal in treatments is to increase functioning.¹⁰⁶ Upon review of limited research on multidisciplinary treatment programs, it is evident that most involve a combination of three types of therapy: 1) psychological, 2) physical, and 3) pharmacological.

Psychological therapy

CBT is being increasingly studied within the FPS population. In reference to FPS, CBT provides the opportunity for clinicians to target the negative symptom-related beliefs and behaviors that perpetuate dysfunction in many patients across a wide array of FPS.¹⁰⁷ During individual CBT sessions, a pediatric psychologist typically helps a patient identify characteristics, such as pain catastrophizing, fear of pain, and psychosocial stressors, that are potentially contributing to the pain as well as provides techniques to actively cope with and manage the painful episodes.^{47,108} Some CBT programs for FPS patients also involve acceptance, interpersonal skills training, and relapse prevention modules in group settings.¹⁰² Still others require a family therapy component, which has produced successful results in comparison to standard care.¹⁰⁹ Family involvement allows for the identification of parents' own catastrophizing, fearful, and overprotective behaviors that have proven to be maladaptive and influence the child in pain.^{110–112} The parent learns to serve as a model and reinforcer of responding to pain in adaptive healthy ways.¹¹³ In addition to CBT, evidence supporting hypnotherapy is emerging. This approach teaches children to alter the perception of uncomfortable physiological sensations in their body.¹¹⁴ A recent systematic review of randomized clinical trials of gut-directed hypnotherapy identified greater improvements in pain that were sustained at follow-up in comparison to standard care.¹¹⁵

Physical therapy

As stated previously, the primary aim of pain management for youth with FPS is to restore function and participation in daily activities. Physical rehabilitation is a common and often essential treatment component.¹¹⁶ More specifically, graded exercise allows for physical activity that gradually and slowly increases over time, typically after the child has established a hierarchical list of activities where each activity must be completed before pursuing the next based on successful execution rather than pain reduction.¹¹⁷ Due to the unique, complex nature of FPS patients, it is likely that graded exercise will not be beneficial for the entire population. Alternatively, yoga may serve to decrease physical and emotional pain-related symptoms.^{118,119}

Pharmacological therapy

When considering pharmacological therapy with the goal of decreasing peripheral disturbances, research is variable and generally weak, as most evidence points to focusing on function versus symptom alleviation with FPS.¹²⁰ There is still considerable use of anticonvulsants for pediatric pain management. Additionally, antidepressants have gained traction with joint effects on pain modulation and associated mood states.^{121–123} Evidence within the pediatric population is lacking; however, few studies demonstrate that antidepressant medication taken for FPS shows promising response rates in youth through improvements in quality of life as well as decreases in pain and psychiatric symptoms.^{124–127} With this being said, current research has reported greater success in decreasing pain intensity and duration by utilizing pharmacological treatment in combination with psychological intervention as opposed to standard pharmacological treatment alone. Such multifaceted treatment regimens also have the potential to provide secondary positive impacts on child psychological and social variables.¹²⁸ Still, more longitudinal studies are necessary to determine the effectiveness of antidepressant medications in the pediatric FPS population and uncover possible harmful side effects.

Where to go from here for FPS?

We have just scratched the surface in properly addressing FPS in childhood. It is apparent, though, that to better address FPS in childhood, we must shift our clinical and research approach.

In the clinic

1. Empathize: Patients with FPS are seeking, and deserve, validation. We must clarify that a lack of definitive

answers does not mean that we have given up on them or do not believe them. In fact, it should reflect that we are even more invested in finding the most appropriate treatment that is going to uniquely address their child's situation.

2. Communicate: In communicating with the patient and family about chronic pain, a coherent explanation should be given to increase patient knowledge of pain-related biology and shift conceptions of pain.¹²⁹ Additionally, communication between providers who have cared for the patient is key to offering a uniform treatment approach.
3. Normalize: Psychiatric comorbidity among youth with FPS is common. It should neither be trivialized nor become the sole focus of the child's pain treatment intervention, even in extreme cases of Munchausen. A multidisciplinary approach that includes medical, physical, and psychology teams working together is essential for treatment success.
4. Biobehavioral interventions as a first-line treatment: Biobehavioral approaches to the treatment of pain have proven efficacy, without side effects, and bringing these treatments online early reduces the stigma associated with the belief "Well, the doctors didn't know what else to do so they sent us to a psychologist".¹³⁰ Importantly, many of these strategies are now readily accessible through the internet or through apps.¹³¹

In both the clinic and in the domain of research

5. Pick a name: Decide on proper, nonjudgmental nomenclature that destigmatizes these conditions, such as the proposed term, primary pain disorders.²⁰
6. All use the same name: Ensure that this nomenclature is implemented across specialists to ensure clinicians treating these patients and researchers studying these patients are speaking a common language.
7. Put some of these theories to the test: It is possible that patients with the same symptom presentation may actually have different drivers that serve to maintain their condition, thus perhaps it is worthwhile to group patients not by their phenotype (eg, abdominal pain) but rather the underlying mechanism (eg, neuronal sensitization) to formulate treatment recommendations and new research hypotheses. The use of "big data" can also aid this effort.

In closing, the momentum is building to garner better answers and treatment for children with FPS, and there are many reasons to believe it will only get better. With this

being said, FPS should become a high-priority topic for future research.

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