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Research article

Agreement and disagreement in pediatric functional neurological symptom disorders: Comparing patient reported outcome measures (PROMs) and clinician assessments

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ABSTRACT

Youth with functional neurological symptom disorder (FNSD) often perceive themselves as having limited capabilities, which may not align with clinical evaluations. This study assessed the disparities between clinician evaluations and patient-reported outcome measures (PROMs) regarding pain, motor function, and learning difficulties in youth with FNSD. Sixty-two youths with FNSD participated in this study, all of whom reported experiencing pain, motor problems, and/or learning difficulties. Clinicians also assessed these domains, resulting in a two-by-two categorization matrix: (1) ***agreement***: child and clinician report "problems"; (2) ***agreement***: child and clinician report "no problems"; (3) ***disagreement:*** child reports "problems" while the clinician does not; and (4) ***disagreement:*** clinician reports "problems" while the child does not. Agreement/disagreement differences were analyzed. No significant differences in prevalence were observed between the evaluators regarding pain (clinician-85%, child-88%), motor (clinician-98%, child-95%), or learning problems (clinician-69%, child-61%). More than 80% of the children and clinicians report pain and motor disorders. Instances in which children and clinicians reported learning problems (40.3%) exceeded cases in which both reported no problems (9.6%) or only the child reported problems (20.9%). Overall, the agreement between pain and motor function assessments was high (>90%), whereas that concerning learning difficulties was moderate (49.9%). Disagreement in pain/motor assessments was minimal (<5%), whereas for learning difficulties, disagreement rates were high (>20%). In conclusion, a significant concordance exists between PROMs and clinician assessments of pain and motor problems. However, the higher frequency of disagreements regarding learning difficulties emphasizes the importance of incorporating patient and clinician evaluations in pediatric FNSD treatment.

1. Introduction

Functional Neurological Symptom Disorder (FNSD) is a condition in which patients experience neurological symptoms, such as movement and pain disorders, without any known underlying neurological conditions [1]. Manifestations of movement disorders include weakness or paralysis; abnormal movements such as tremors, dystonia, myoclonus, gait disturbances, and dysphagia; and speech impairments such as dysphonia or slurred speech [2]. Although pain is not a diagnostic requirement for FNSD, according to the Diagnostic and Statistical Manual of Mental Disorders (DSM) and other diagnostic models, a high prevalence of chronic pain as a comorbidity has been documented in patients with FNSD [3,4]. Within the pediatric population, the incidence of FNSD is estimated at 1.3 to 6.0 per 100,000 [5,6], with incidence increasing with age through adolescence years with females being diagnosed more commonly than males [7].

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Consistent functional and structural abnormalities have been observed in patients with FNSD, particularly in the motor-processing regions. Erpelding et al. [8] demonstrated that children with FNSD exhibit reduced activity in brain areas related to the motor system compared to controls. Even in the asymptomatic state, significant differences in central nervous system responses were evident after stimulating the affected and unaffected limbs, particularly in brain regions crucial for movement (for example, frontal lobes, insula, and basal ganglia). Structural abnormalities, including increased volume in the left supplementary motor area, right superior temporal gyrus, and dorsomedial prefrontal cortex, have also been observed in children with FNSD [9]. However, given the reliance on cross-sectional designs, it remains challenging to ascertain whether these structural and functional abnormalities are the cause of the functional symptoms or consequences of FNSD [10].

Regarding pain issues in individuals with FNSD, the sensory processing patterns leading to pain have yet to be thoroughly characterized using validated scales. However, it has been suggested that individuals with sensory sensitivity tend to hyperfocus on sensory experiences from the body and environment, leading to a sustained state of hyperarousal and hypervigilance, thereby intensifying pain sensations [11].

Individuals with FNSD may experience cognitive difficulties in addition to motor and pain-related issues [12]. For instance, research has indicated that approximately 40% of children and adolescents with FNSD struggle with learning difficulties, a notably higher prevalence than the reported lifetime prevalence of learning disabilities (10%) [13]. Attentional difficulties are also prevalent in this population. Rai et al. [14] suggested that attention and concentration problems, along with motor, sensory, emotional, and arousal processing, contribute to the neural basis of FNSD. Regarding attention problems, it has been suggested that FNSD involves a decrease in attention directed toward the external world and an increased focus on internal bodily processes (interoceptive awareness) [15]. This focus on internal bodily processes is of special interest as it may contribute to learning difficulties. More specifically, dysregulated interoceptive awareness has a major impact on children's educational outcomes, resulting in higher rates of disengagement, suspension, poorer learning outcomes, and difficulties in engaging in prosocial behaviors [16]. Hall et al. [17] proposed that poor verbal skills in individuals with FNSD may be linked to the development of symptoms [17]. In [18] their review of the FNSD etiology and the integrated etiological summary model, Fobian and Elliot described attentional deficits in FNSD. More specifically, the authors summarize that neurobiological research has indicated that people with FNSD have a decreased sense of agency and abnormal attentional focus on the affected area, both of which are modulated by beliefs and expectations about illness [12,19,20].

There has been a growing consensus over the past two decades that FNSD is a multifaceted phenomenon that involves biopsychosocial aspects [19]. Currently, FNSD is recognized as a complex condition influenced by a Bayesian probabilistic mechanism [20]. This mechanism suggests that somatic symptoms arise and persist because of the interplay between various factors, including psychological stimuli (such as anxiety) [21], learned experiences (such as social rejection) [22,23], heightened attention to bodily sensations (such as increased focus on pain) [24], and beliefs about illness (such as pain catastrophizing) [25,26]. Stress has also been recognized as a risk factor for FNSD development. Specifically, it has been reported that a history of adverse traumatic events may be associated with abnormalities in hypothalamus–pituitary–adrenal (HPA) axis function, potentially increasing the risk for FNSD. Moreover, reduced volumes of the hippocampus and amygdala serve as biological "trait markers" for FNSD, potentially contributing to reduced resilience to stress [27].

Adolescents diagnosed with FNSD frequently encounter challenges in participating in age-appropriate activities, including regular school attendance [28], engaging in positive social interactions with peers [29], participating in sports and extracurricular activities [30], and participating in personal and family events [31,32]. Forrest et al. [33] reported that among N = 1457 children in the fourth through sixth grades from 34 schools in three school districts, children with functional limitations or behavioral health issues were prone to reduced student engagement, disruptive behaviors, poor grades, and below-average performance on standardized achievement tests.

The multifaceted biopsychosocial presentation of FNSD complicates diagnosis. Consequently, a purely clinician-based ("objective") assessment strategy may not be optimal, specifically within the pediatric population. Incorporating patient-reported ("subjective") data alongside clinician evaluation holds promise for a more comprehensive diagnostic and treatment approach [34,35]. Furthermore, adolescents with FNSD may perceive themselves as having limited ability to perform basic physical activities above their clinically assessed impairment level [36]. These perceptions significantly decreased their active participation in various situations, including their ability to engage in therapeutic activities. However, despite being a common and disabling condition, there has been little research on both objective and subjective outcome measures in FNSD in general [35] and specifically among adolescents. Hence, considering adolescents' subjective perceptions of their abilities as separate but equally important, patient-reported outcome measures (PROMs) can provide valuable insights beyond evaluating their abilities in objective tasks [37].

Since FNSD in children and adolescents is characterized by multidimensional disturbances, interdisciplinary rehabilitation programs (involving physicians, physical/occupational therapists, educators, and psychotherapists) are considered promising treatment approaches for this population [38]. These programs aim to minimize symptoms (for example, movement/walking impairments) and facilitate a return to age-appropriate functioning [39].

Traditionally, a child's progress within an interdisciplinary rehabilitation program has been assessed based on the achievement of specific therapist-defined goals. However, this approach may overlook certain aspects of children's self-perceived confidence in their independent functioning. Additionally, research suggests that incorporating both therapist assessments and child/adolescent PROMs can provide a more comprehensive picture of rehabilitation needs and valuable clinical insights into this sensitive population [35]. Therefore, this study aimed to evaluate the discrepancies between therapist assessments and patient-reported outcome measures (PROMs) in children and adolescents with FNSD, specifically regarding motor function, pain, and learning difficulties. Furthermore, proxy reports (from caregivers) are often used to describe the clinical symptoms of children and adolescents. However, although caregiver proxy-reports are deemed acceptable in various conditions, such as pediatric pain assessment [40], caregiver–-child agreement is modest [41]. Therefore, it is crucial to incorporate child-specific PROMs and compare them with clinician assessments. In doing so, our study helps to address the knowledge gap regarding the use of PROMs in pediatric populations.

2. Material and methods

This observational clinical care study was conducted in an ambulatory clinical setting at a Children's Hospital in the center of the country, treating youth from the entire country.

2.1. Study participants

Between October 2017 and December 2019, children and adolescents diagnosed with FNSD were directed to the ambulatory rehabilitation clinic through referrals from their primary or secondary care physicians (for example, rheumatologists, orthopedics, and neurologists) after the exclusion of alternative underlying causes of their symptoms. This study included children and adolescents: (1) aged 6–18 years; (2) diagnosed with FNSD with abnormal movement based on the Diagnostic and Statistical Manual of Mental Disorders – 5 – Text

Revision (DSM-5-TR) and the International Classification of Diseases, 10th edition (ICD-10; F44.4);[42] and (3) presenting lower and/or upper extremity motor and/or pain problems. Children were excluded if they (1) were treated during inpatient rehabilitation or (2) had other clinical diagnoses that might explain the child's symptoms, such as other medical or psychiatric conditions.

2.2. Procedures

The study was conducted at a major public hospital (The Chaim Sheba Medical Center, The Edmond and Lily Safra Children's Hospital) in the center of the country. Referred youth with FNSD who met the inclusion criteria were admitted to an Integrative Pediatric Rehabilitative (IPR) program routinely provided to all youth with FNSD admitted to the Pediatric Rehabilitation Department. The IPR program provided comprehensive outpatient care. A multidisciplinary team comprising physicians, physiotherapists, psychologists, and specialized educators collaborated to diagnose and treat children with functional motor, pain, and learning disabilities using remedial methods. The mean duration of the IPR is 6.26 $\pm$ (3.64) months [43]. Briefly, the program consisted of psychological (for both adolescents and their parents), physical, and educational interventions provided twice a week for a duration of 45–50 min (see Box 1 for IPR treatment outline).

All the study procedures were approved by The Chaim Sheba Medical Center Medical Center Ethical Review Board (7394–20-SMC; May 2020).

2.3. Study measures

2.3.1. Demographic and clinical characteristics

Demographics (age and sex) and clinical characteristics (i.e., previous injury, additional diagnoses, and somatization symptom severity) were retrieved from medical records. Psychologists assessed the severity of somatization symptoms upon admission to the IPR using the Child's Somatization Inventory-24 (CSI-24) [44]. The questionnaire comprised a list of 24 symptoms experienced within the preceding two weeks. Participants were asked to rate the frequency of each symptom on a 5-point scale (0 = *not at all*, 4 = *a whole lot*). The mean number of symptoms (0–24) was calculated, with higher scores indicating a higher intensity of somatic complaints. Cronbach's alpha for the CSI-24 is 0.87 [44].

2.3.2. Pain assessment

2.3.2.1. PROMs pain assessment. Two categories of pain problems were created. The first category, "pain sensitivity," was established using the Budapest criteria for diagnosing complex regional pain syndrome (CRPS), which is referred to as "prolonged pain and inflammation that can occur following an injury or other medical events such as surgery, trauma, stroke, or heart attack" [45]. According to the Budapest criteria, the children were asked to report whether they experienced the following pain-related problems: allodynia (pain due to a stimulus that does not normally provoke pain) to light touch, temperature sensation, deep somatic pressure, joint movement, and/or hyperalgesia (that is, an exaggerated pain response to a stimulus that usually causes pain) [46]. Children presenting with any of the aforementioned signs were classified as having pain sensitivity (allodynia and/or hyperalgesia). The pain intensity was assessed in the second category. Pain intensity was evaluated while sitting at rest, using a numerical pain rating scale ranging from 0 (*no hurt*) to 10 (*hurts like the worst pain imaginable*) [47]. Children reporting a score greater than 4 (moderate to very severe pain) were categorized as having high pain intensity.

2.3.2.2. Clinicians' assessment of pain. Pain was evaluated using indirect assessments. Specifically, during the informal evaluation, two senior pediatric physical therapists observed the child's physical, behavioral, and emotional signs and symptoms of allodynia and/or hyperalgesia (pain sensitivity) and/or pain intensity. Such signs may

Box 1
Description of the program outline.

<table>
<tr><th rowspan="2">Physical Therapy</th><th colspan="2">Psychotherapy</th><th rowspan="2">Educational Therapy</th></tr>
<tr><th>Adolescent</th><th>Parent/s</th></tr>
<tr><td>Physical therapy sessions use a combination of individual and group sessions to address the unique needs of children with FNSD. Therapists build strong rapport with each child, creating a safe and supportive environment. The program emphasizes achieving functional goals, such as improving mobility or daily living skills. Therapists utilize a fun and engaging approach, incorporating games and activities that seamlessly integrate functional exercises. Additionally, the program equips parents and caregivers with home training exercises, ensuring consistent progress and continued support beyond the therapy sessions.</td><td>Adolescents participate in individual psychotherapy sessions. Emotional challenges are discussed within a pain-related approach. The sessions focus on several themes including:
– Recognition and labeling negative emotions that may contribute to pain.
– Expressing and communicating challenges and frustrations in a proactive manner.
– Developing strong communication skills with parents and peers to foster supportive relationships.
– Exploring and resolving emotional issues related to independence and self-identity.</td><td>Parental sessions are conducted in addition to adolescents' psychotherapy sessions. Parental sessions incorporate three key components:
– Pain-related psychoeducation focused on how to manage their child's pain in a supportive way.
– Fostering a sense of autonomy, competency (mastery), and relatedness in the child.
– Developing effective communication skills to discuss pain and related challenges openly and constructively with their child.</td><td>To address learning difficulties and academic gaps resulting from school absences, a comprehensive assessment is conducted to identify the adolescent's specific needs. Following, an individualized learning program is developed in collaboration with a corrective education expert. The tailored program aims to ensure the adolescent receives targeted training to bridge the academic gaps and overcome their learning difficulties.</td></tr>
</table>

consist of, but not limited to, complaining, crying, avoidance expressions (such as “Stop, stop!”), decreased activity, dissatisfied facial expressions (for example, grimace and clenched teeth), abnormal gait, irritability, guarding of a body part, sweating, changes in mental status (e.g., confusion), muscle tension, restlessness, or exhaustion [48]. Therapists classified children presenting with such signs as high pain sensitivity (allodynia and/or hyperalgesia) and/or high pain intensity. In the current study, we indirectly assessed pain according to the unique behavioral characteristics of children during treatment [36]. Such an indirect assessment is feasible, as pain can lead to a number of physiological changes, such as sweating, pupillary dilation, grimacing, or tearing in the eyes [49].

2.3.3. Motor function assessment

2.3.3.1. PROMs motor function assessment. Motor problems were assessed using the Budapest criteria [46]. Specifically, the children were asked to report whether they experienced a decreased range of motion and/or motor dysfunction (weakness, problems in gross motor function, problems in activities of daily living, and problems in mobility). Children who reported any of these motor problems were classified as having motor problems.

2.3.3.2. Clinicians' assessment of motor function. Motor problems were assessed indirectly and directly by two experienced pediatric physical therapists during the first week of the IPR program. Indirect assessment was performed according to the recommendations of Gray et al. [50]. Indirect assessments involved observations conducted by physical therapists to determine the participants' functional capacity (such as gross motor function and mobility) while engaging in a task/game (for example, tossing a beanbag 10 times at a target on the wall). In addition, in the indirect assessment, physical therapists observed the child's ability to engage in gross motor functions (such as bed mobility, transferring from supine to sitting, and from sitting to standing) and conduct activities of daily living (getting dressed). Children who were unable to independently perform any gross motor functions or activities of daily living were categorized as having motor problems. In the direct assessment, the therapist observed the child's functioning while conducting traditional objective measures of physical function. According to a recent systematic review and recommendations on outcome measurements in FNSD [51], there are few well-validated motor-specific outcome measures. In the current study, the Six-Minute Walk test (6MWT) [52] was used, as it has already demonstrated test-rest reliability in other related populations, such as individuals with psychological or psychiatric problems, and was previously used in children and youth with FNSD [43,51]. During the test, the participants were instructed to walk for 6 min at a comfortable pace using their usual assistive devices. Those who used a wheelchair could not perform the 6MWT; therefore, their walking distance during the test was 0 m. The participants' walking ability relative to the norm was evaluated by calculating the number of standard deviations below the mean, according to the norms published by Geiger et al. [53]. Children presenting walking distances greater than or equal to 1.5 standard deviations below the norm were categorized as having walking problems. The direct assessment was limited only to walking ability within the FNSD population, especially at the beginning of the rehabilitation program, before rapport was built between the child and the treatment team, and the child is often unwilling to fully collaborate with the treatment team and engage in activities, causing them to feel uncomfortable [36]. A child was classified as having a motor problem if they presented with problems in either the direct or indirect evaluations.

2.3.4. Assessment of learning problems

2.3.4.1. PROMs assessment of learning problems. Children were directly asked whether they were experiencing learning problems in school (for example, "Do you experience learning difficulties in school, such as difficulties in writing, reading, spelling, or mathematics?). Children who responded 'yes' were classified as having learning difficulties. To better understand the nature of their learning difficulties, children who responded that they experienced learning difficulties were further asked to specify the specific domains in which they encountered difficulties (such as reading, reading comprehension, and mathematical skills).

2.3.4.2. Clinicians' assessment of learning problems. Learning problems were evaluated by testing the child's reading comprehension level and providing an index regarding the level of their performance in relation to what was expected of their peers. More specifically, the test consisted of three parts: 1) text recovery after reading (e.g., repeating explicit information details in the text, such as names, actions, time, and place); 2) answering open and closed questions at three levels of understanding; 3) repeated reconstruction of the text after answering the questions. Based on these three sections, a score ranging from 0–100 was calculated, with higher scores indicating better ability. Children with scores of < 60 were classified as having learning problems. Official text reading kits in the country's language were used for didactic diagnosis [54]. Each text had a corresponding scoring sheet. The scoring criteria included the number of recalled text details, the child's ability to maintain the narrative sequence, comprehension of context and subtext, and ability to draw relevant conclusions. Certified didactic diagnosticians administered the tests, which were conducted individually in a quiet room and lasted approximately 45 min [50].

2.4. Statistical analyses

2.4.1. Study participants' demographic and clinical characteristics

Descriptive statistics were used to present the demographic and clinical characteristics of the sample, utilizing measures such as mean, standard deviation, sample size, and percentage. Disparities in categorical demographics and clinical traits were assessed using the chi-square test.

2.4.2. Patient report outcomes vs. clinician's examination – prevalence of pain, motor, and learning difficulties

For each assessment domain (pain, motor function, and learning), the prevalence of problems was calculated separately for both patient-reported measures (PROMs) and clinician evaluations. In the pain domain, two aspects (hyperesthesia and overall pain) were evaluated, and a combined score was used to identify children with overall pain problems. Specifically, children who reported problems with either or both pain measures were categorized as experiencing pain. Chi-square tests were used to analyze discrepancies in the prevalence rates between PROMs and clinician assessments within each domain. Box-and-whisker plots were used to visually represent the distribution of continuous variables, providing insights into the children's pain, motor, and learning profiles.

2.4.3. Patient reported outcomes vs. clinician's examination – agreement and disagreement prevalence

A two-by-two categorization matrix was used to assess agreement and disagreement between patient-reported outcomes and clinician reports. As a result, four different categories are reported: two for agreement (child and clinician reporting "no problems" or "problems"), and two for disagreement (child reporting "problems" while therapist reporting "no problems" and vice versa). The prevalence of each of the four categories was calculated separately for each domain (motor, pain, and learning difficulties). Differences in prevalence between categories were evaluated using the chi-square test.

In addition, factors associated with the agreement were evaluated. For each participant, the total number of difficulties identified was calculated for both PROMs and clinician assessments. A discrepancy score was computed by subtracting the number of problems reported by the PROMs from the number observed by the clinician. Finally, Pearson's correlation coefficients were calculated to explore the potential associations between this discrepancy score and age, FNSD duration, and CSI scores.

3. Results

3.1. Study participants' demographic and clinical characteristics

Sixty-two youths diagnosed with FNSD, with an average age of 13.7 ± 2.9 (75% females), participated in this study. Most study participants (N = 35.0, 57.3%) sustained a previous medical event prior to FNSD diagnosis and admission to the rehabilitation program, and approximately 20% of participants (N = 12) were diagnosed with attention-deficit/hyperactivity disorder (Table 1).

3.2. Prevalence of pain, motor, and learning difficulties

No significant differences were found between the evaluations conducted by different informants (children vs. clinicians) in terms of the prevalence of pain difficulties (clinicians: 85.4%, children: 88.7%; Fig. 1a), motor function difficulties (clinicians: 98.3%, children: 95.1%; Fig. 1b), or learning problems (clinicians: 69.3%, children: 61.2%; Fig. 1c; p range: 0.35 0.58). However, from examining Figs. 2–4, it appears that children's pain level, walking ability, and learning ability varied considerably in all domains, with scores ranging from the lowest to the highest possible. Nonetheless, the groups' mean scores suggested considerable difficulty in all domains assessed (mean pain score: 6.7$\pm$2.3; mean walking standard deviations below the mean: 7.4$\pm$3.5; mean reading comprehension: 70.1$\pm$17.7).

The prevalence of children diagnosed with ADHD in our sample was relatively high (Table 1). To assess whether ADHD might confound the evaluation of learning difficulties, we compared the prevalence of ADHD

Table 1
Study participants' demographic and clinical characteristics (N = 62).

Characteristic			Mean (SD) OR N (%)
Child's socio-demographic characteristics	**Age: mean (SD)**		13.7 (2.9)
	Sex	Females: n (%)	46.0 (75.4)
		Males: n (%)	15.0 (24.5)
		Chi-square (p-value)	**31.23 (<0.01)**
Parental sociodemographic characteristics	**Marital status**	Married: n (%)	46.0 (74.1)
		Not married: n (%)	16.0 (25.8)
		Chi-square (p-value)	**29.5 (<0.001)**
	Mother's employment status	Full time: n (%)	25.0 (40.3)
		Part time: n (%)	19.0 (30.6)
		Unemployed: n (%)	18.0 (29.0)
		Chi-square (p-value)	1.6 (0.1)
Clinical characteristics	**CSI (number of symptoms): mean (SD)**		14.2 (5.1)
	FNSD duration (months): mean (SD)		4.3 (3.20)
	Previous injury	**No, number (%)**	25.0 (41.7)
		Yes, number (%)	35.0 (57.3)
		Chi-square (p-value)	**2.97 (<0.01)**
		Accident-related injury, number (%)	3.0 (4.8)
		Falling related injury, number (%)	10.0 (16.1)
		Viral or other infectious disease, number (n)	6.0 (9.6)
		Orthopedic injury, number (n)	15.0 (24.1)
		Syncope, number (n)	1.0 (1.6)
	Additional diagnoses	**Attention-deficit/ hyperactivity disorder, n (%)**	12.0 (19.6)
		Anxiety/ depression, n (%)	7.0 (11.4)
		Chi-square (p-value)	1.54 (0.21)

Notes: SD, standard deviation

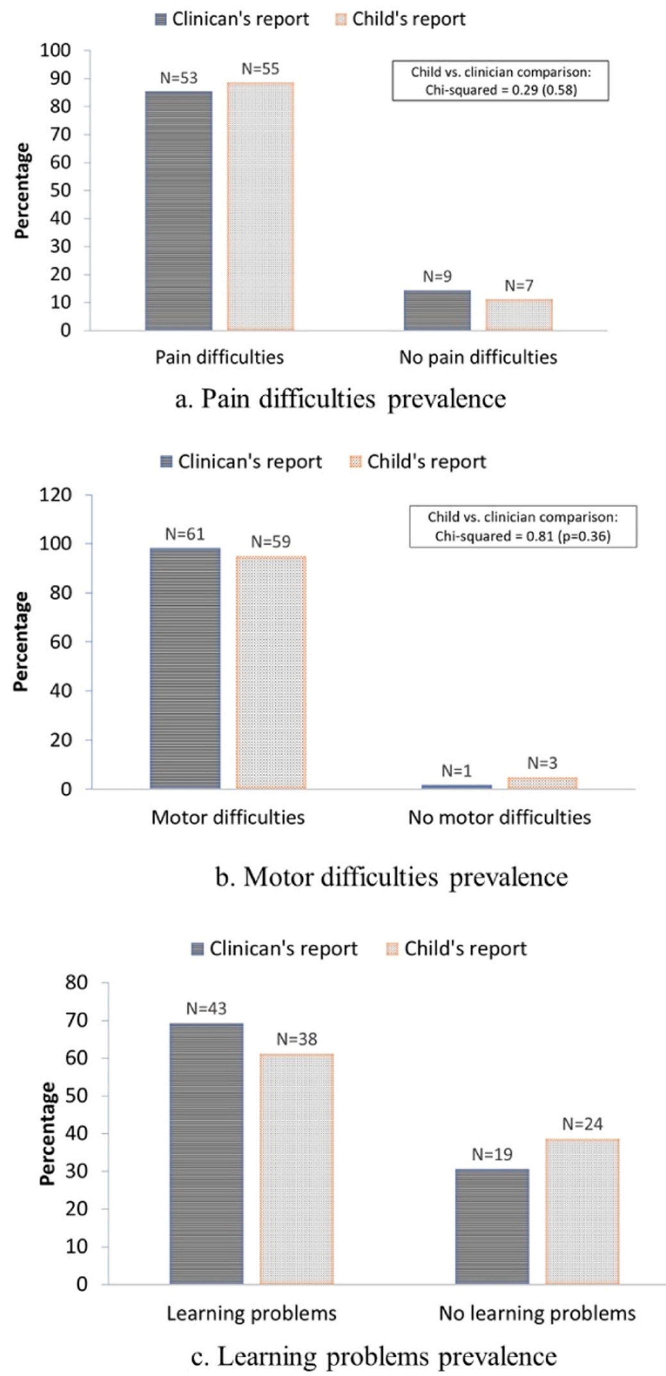

Fig. 1. Differences in difficulties prevalence based on child's and clinician's reports, a. Differences in pain (pain sensitivity and or high pain intensity) difficulties prevalence based on child's and clinician's reports; b. Differences in motor difficulties prevalence based on child's and clinician's reports; c. Differences in learning problems prevalence based on child's and clinician's reports, *Notes:* The figure depicts the prevelance of children presenting sensory (Figure a), motor (Figure b) and learning difficulties (Figure c) according to both child's and clinican's report.Between evaluators prevelance was evaluated using Chi-squared tests.

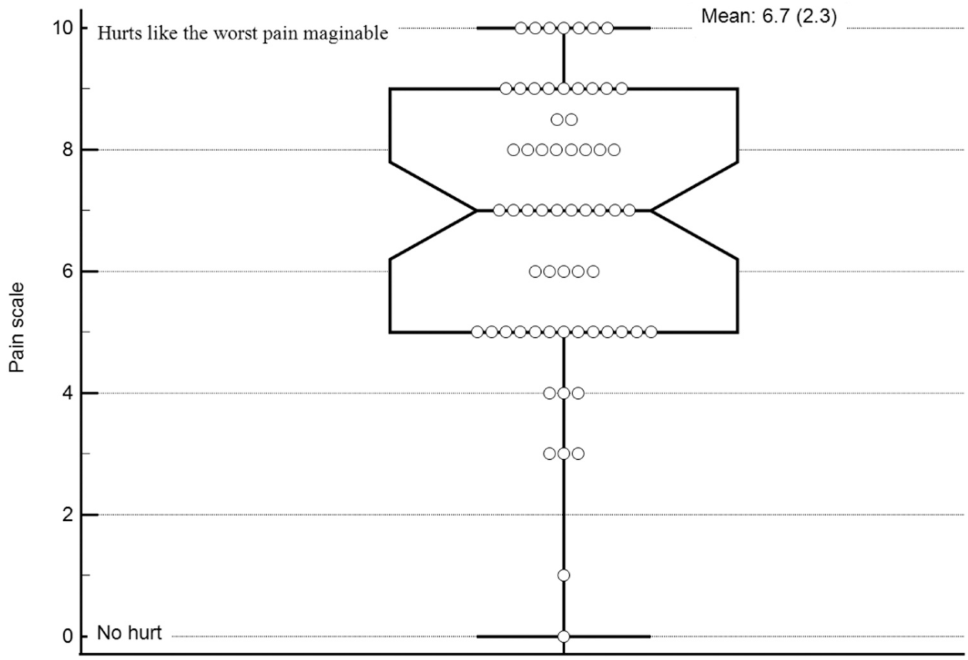

Fig. 2. Patient reported outcome measures – pain intensity. *Notes:* The central box represents the values from the lower to upper quartile (25–75 percentiles); the vertical line extends from the minimum to the maximum value, excluding outside values which are displayed as separate points. An outside value is defined as a value that is smaller than the lower quartile minus 1.5 times the interquartile range, or larger than the upper quartile plus 1.5 times the interquartile range; the middle line represents the median.

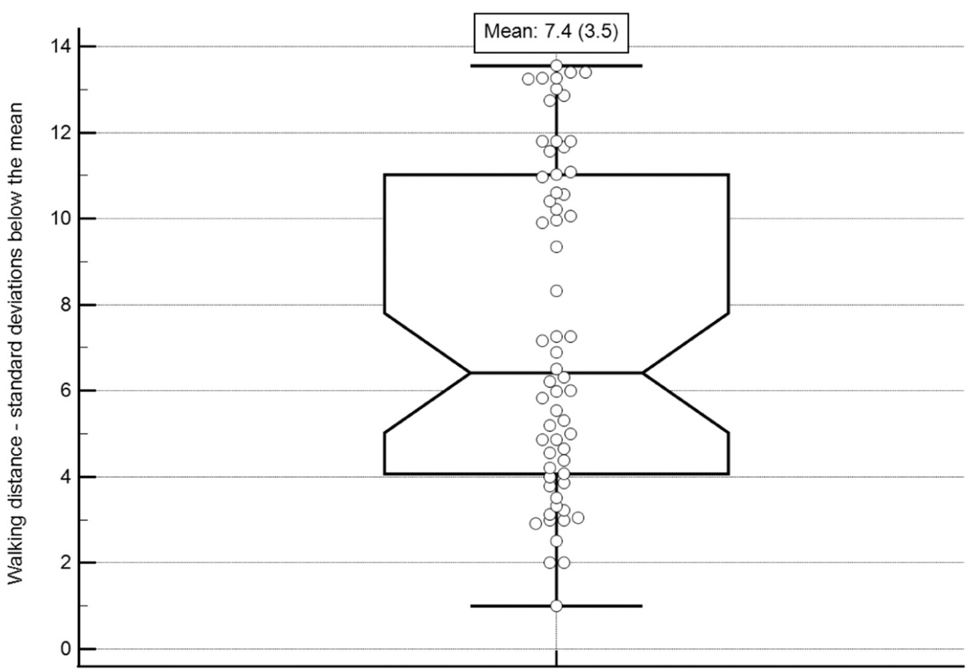

Fig. 3. Clinician's report – motor ability, *Notes:* The central box represents the values from the lower to upper quartile (25–75 percentiles); the vertical line extends from the minimum to the maximum value, excluding outside values which are displayed as separate points. An outside value is defined as a value that is smaller than the lower quartile minus 1.5 times the interquartile range, or larger than the upper quartile plus 1.5 times the interquartile range; the middle line represents the median.

in youth with and without learning problems as identified by both PROMs and clinician evaluations. The results indicated no significant difference in ADHD prevalence between these groups (PROMs: 16.6% and 20.4% among those with and without learning problems, respectively, chi-square =1.13, p = 0.28; clinician's evaluation: 16.0% and 21.6% among those presenting and not presenting learning problems, respectively, chi-square =0.86, p = 0.35).

3.3. Patient report outcomes vs. clinician's examination – agreement and disagreement prevalence

The most common pattern of pain and motor problems was

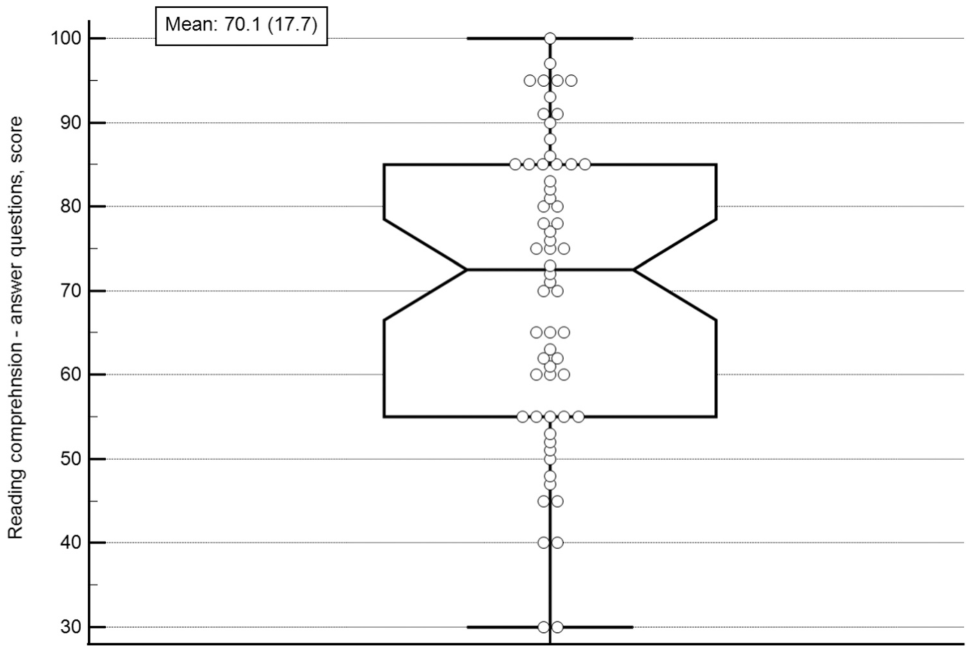

Fig. 4. Patient report outcome measure – reading comprehension, *Notes:* The central box represents the values from the lower to upper quartile (25–75 percentiles); the vertical line extends from the minimum to the maximum value, excluding outside values which are displayed as separate points. An outside value is defined as a value that is smaller than the lower quartile minus 1.5 times the interquartile range, or larger than the upper quartile plus 1.5 times the interquartile range; the middle line represents the median.

agreement regarding the presence of difficulties between children and clinicians (83.8% for pain sensitivity, 96.7% for pain intensity, and 95.2% for motor problems). Within the learning difficulties domain, the agreement between children and clinicians reporting difficulties (40.3%) was higher than that in the other agreement categories. However, this agreement rate was considerably lower than that reported for the pain and motor domains. For additional information, please refer to Table 2.

Further analysis revealed high levels of agreement between child reports and clinician evaluations. In the pain domain, the total agreement rate (including both agreement on having problems and agreement on not having problems) was 96.7%, reflecting almost perfect agreement. Similarly, the motor domain exhibits a high agreement rate (96.8%). However, the agreement rate for learning difficulties was considerably lower (49.9%), indicating moderate agreement in this domain. Similarly, the average disagreement rates for pain and motor problems were very low (3.2% in both domains). The disagreement level in learning difficulties was higher (49.9%, moderate disagreement) and significantly higher than that observed in pain and motor function ($p < 0.05$). Across all three evaluation domains, there was no significant difference in the prevalence of the two disagreement categories, that is, cases where children reported difficulties while clinicians did not, and vice versa (p range: 0.1–0.4). For additional information, please refer to Table 3.

Finally, no significant associations were observed between the difference in the number of problems reported by clinicians and those reported using PROMs and other demographic characteristics such as the child's age ($r = -0.07$, $p = 0.56$), duration of FNSD ($r = -0.11$, $p = 0.51$), or level of FNSD symptoms (CSI) ($r = -0.01$, $p = 0.51$).

4. Discussion

Functional neurological symptom disorders in children and adolescents have received growing attention in recent years because of their significant impact on the quality of life of children and their families [55]. These disorders manifest as a diverse range of neurological

Table 2
Patient report outcomes vs. clinician's examination (N = 62).

Variables		Clinician's evaluation	Patient Report		Chi-square (p value)
			No difficulties: n (%)	Difficulties: n (%)	
Pain problems	**Pain sensitivity (allodynia and/or hyperalgesia)**	**No difficulties: n (%)**	6.0 (9.6)[d]	3.0 (4.8)[d]	**84.9 (<0.001)**
		Difficulties: n (%)	1.0 (1.6)[d]	52.0 (83.8)[a-c]	
	High pain intensity (pain ≥ 4)	**No difficulties: n (%)**	2.0 (3.2)[d]	0.0 (0.0)[d]	**115.1 (<0.001)**
		Difficulties: n (%)	0.0 (0.0)[d]	60.0 (96.7)[a-c]	
	Average agreement				-
Motor problems		**No difficulties: n (%)**	1.0 (1.6)[d]	0 (0.0)[d]	**111.7 (<0.001)**
		Difficulties: n (%)	2.0 (3.2)[d]	59 (95.2)[a-c]	
Learning problems		**No difficulties: n (%)**	6.0 (9.6)[b,d]	13.0 (20.9)[a]	**7.6 (0.005)**
		Difficulties: n (%)	18.0 (29.0)[d]	25.0 (40.3)[a,c]	

Notes:
[a] statistically significantly different from clinician and patient reporting "no-difficulties" ($p < 0.05$, 2-tailed);
[b] statistically significantly different from clinician reporting "no-difficulties" and patient reporting "difficulties" ($p < 0.05$, 2-tailed);
[c] statistically significantly different from clinician reporting "difficulties" and patient reporting "no difficulties" ($p < 0.05$, 2-tailed);
[d] statistically significantly different from clinician and patient reporting "difficulties" ($p < 0.05$, 2-tailed).

Table 3
Clinician and child percentage agreement and disagreement summary table (N = 62).

Items		Child-clinician – percentage agreement				
		0.0-20.0% Slight agreement	20.1-40.0% Fair agreement	40.1-60.0% Moderate agreement	60.1-80.0% Substantial agreement	80.1-100.0% Almost perfect agreement
Pain problems	**Pain sensitivity (allodynia and/or hyperalgesia)**					$93.4^{,e}$
	High pain intensity (≥ 4)					100^{d}
	Average agreement					*96.7^{d}*
Motor problems						96.8^{d}
Learning difficulties				$49.9^{a\text{-}d}$		

Child – clinician – percentage disagreement

0.0-20.0% Very low percentage	20.1-40.0% Low percentage	40.1-60.0% Medium percentage	60.1-80.0% High percentage	80.1-100.0% Very high percentage

Items		Child – difficulties Clinician – No difficulties	Child – No difficulties Clinician – difficulties	Total disagreement	Chi-square (p value)
Pain problems	**Pain sensitivity (allodynia and/or hyperalgesia)**	4.8^{e}	1.6^{e}	6.4	1.0 (0.3)
	High pain intensity (≥ 4)	0.0^{e}	0.0^{e}	0.0	-
	Average disagreement	*2.4^{e}*	*0.8^{e}*	3.2	0.5 (0.4)
Motor problems		0.0^{e}	3.2^{e}	3.2	2.0 (0.1)
Learning problems		$20.9^{a\text{-}d}$	$29.0^{a\text{-}d}$	49.9	1.3 (0.2)

Notes: All values in the table represent percentage of agreement or disagreement between patient report outcome measures and clinicians' evaluation; a, statistically significantly different from sensory ($p < 0.05$, 2-tailed); b, statistically significantly different from pain ($p < 0.05$, 2-tailed); c, statistically significantly different from sensory problems average disagreement ($p < 0.05$, 2-tailed); d, statistically significantly different from motor problems ($p < 0.05$, 2-tailed); e, statistically significantly different from learning difficulties ($p < 0.05$, 2-tailed).

symptoms, including movement and pain disturbances [1]. The burden of FNSD extends beyond physical health, as it can also affect scholastic performance [43,56]. Studies have increasingly recognized the importance of patient self-perception in understanding their health conditions and its influence on treatment outcomes [34,37]. However, current rehabilitation assessments in FNSD often prioritize clinicians' perspectives on the child's own reported experience.

This study investigated discrepancies between therapist assessments and child-reported outcomes (PROMs) regarding motor, pain, and learning difficulties in children with FNSD. To the best of our knowledge, this is the first study to compare clinician evaluations and PROMs of pain, motor function, and learning difficulties in this pediatric population. Below, we discuss the implications of the prevalence of difficulties within each domain and explore the consequences of agreement and disagreement between therapists and children within this unique population.

4.1. Prevalence of pain, motor, and learning difficulties

Based on both clinician assessments and child PROMs, the majority of study participants exhibited pain (85% of the sample) and motor difficulties (90% of the sample). In comparison to pain and motor difficulties, the prevalence of learning problems was lower but considerable, exceeding 60%. A high prevalence of motor problems was anticipated in both evaluations, given that FNSD is recognized as a disabling condition associated with motor symptoms [42]. Similarly, an increased prevalence of pain was expected, reflecting the significance of somatosensory processing difficulties in patients with FNSD [11].

Literature on FNSD generally emphasizes the motor and pain components, with less attention paid to learning difficulties. In the current study, although the prevalence of learning difficulties was lower than that of motor and pain difficulties, it remained considerably high in both clinician evaluations and children's PROMs (>60%). This prevalence surpasses that observed in the general population (5–15%) [57]. Studies have suggested that subjective reports of cognitive difficulties, including forgetfulness, distractibility, and retrieval problems, are frequent in patients with FNSD [58]. These deficits may be attributed to various factors, such as pain, fatigue, and excessive interoceptive monitoring, common in FNSD, which can negatively impact working memory, attention, and overall cognitive processing speed [59]. Further contributions to these subjective cognitive experiences may be perfectionism and the tendency to overinterpret cognitive impairments, both of which have been reported as clinical characteristics of FNSD [60]. Similarly, school problems have been reported to be a common source of stress in children with FNSD. Common school-related difficulties include cognitive difficulties, learning problems, coping difficulties, high parental scholastic achievement expectations that are not in line with children's abilities, and school absenteeism [61,62]. Diagnosing and treating learning difficulties in the pediatric population with FNSD is of particular importance, as school participation and success are key determinants of children's health, development, and well-being [63]. Moreover, increased stress resulting from learning problems, along with difficulties in expressing these problems, may further contribute to pain difficulties and potentially exacerbate motor problems in patients with FNSD [64,65]. However, motor and pain problems may contribute to learning difficulties. Forrest et al. [33] identified that children who screen positive for special healthcare needs because of functional limitations are at risk of reduced school engagement, poor grades, and below-average performance on standardized achievement tests. Therefore, addressing the vicious cycle of motor, pain, and learning difficulties requires a multidisciplinary rehabilitation framework[39]that encompasses medical, psychiatric, and allied health professions (such as physical therapists) and teachers [66,67] for the provision of comprehensive treatments.

Differentiating learning difficulties associated with FNSD from relatively related conditions of Functional Cognitive Disorders (FCD) [68] is another critical aspect for accurate diagnosis and treatment planning. Although both conditions may involve subjective reports of cognitive challenges, FNSD encompasses a broader spectrum of neurological symptoms, including learning difficulties. Conversely, FCD presents a specific focus on cognitive problems such as forgetfulness and memory impairments [59,69]. However, given the considerable heterogeneity in the literature and methodological shortcomings, further research is warranted before dissociating the nature of cognitive difficulties in FNSD from those in FCD [58].

4.2. Agreement and disagreement prevalence – pain and motor difficulties

In the domains of pain and motor difficulties, the study's findings revealed nearly perfect agreement (>90%) between the clinicians' assessments and the child's PROMs. Consequently, the average disagreement rate remained exceptionally low at only 3%. One plausible explanation for the high percentage of concordance in pain reports between clinicians and children is the significant role of pain in the clinical presentation of FNSD. This pain often serves as a primary source of motivation for referral for rehabilitative treatment. This intense pain sensation may be related to somatosensory amplification (SA), a condition characterized by a heightened focus on and misinterpretation of both physiological and psychological sensations as abnormal or threatening. Somatosensory amplification has three elements: (1) hypervigilance (heightened attention to unpleasant bodily sensations), (2) tendency to focus on weak and infrequent sensations, and (3) tendency to overestimate visceral and bodily sensations as aberrant rather than perceiving them as normal [70,71]. Somatosensory amplification has frequently been described in adults and children diagnosed with FNSD [70–72]. Specifically, intense pain experienced by children influences their motor abilities and reduces their willingness to engage in motor activities during therapy [36]. Considering the above-mentioned findings, it appears that pain and motor difficulties are readily apparent and communicated by children and youth with FNSD to healthcare professionals. However, while the value of PROMs in adult populations is well established, research on the pediatric population with FNSD is limited, leaving our findings without a clear reference point for comparison [73]. Therefore, the findings of this study suggest that in routine clinical practice, child-reported outcome measures (PROMs) can be partially relied upon to evaluate pain and motor difficulties in youth with FNSD. This is significant, as PROMs not only capture the impact of a medical condition and/or treatment on the patient itself [74] but also enhance communication between healthcare providers and children [75–77]. Furthermore, the observed resonance between child PROMs and clinician assessments of pain and motor problems strengthens the reliability of self-reported measures in children with FNSD.

4.3. Agreement and disagreement prevalence – learning difficulties

While the agreement between clinician assessments and child-reported outcomes (PROMs) on learning difficulties was moderate (49.9%), teacher evaluations identified learning difficulties in 29.0% of the children who reported no such difficulties. However, it is important to note that the teachers' evaluations in the current study focused on reading comprehension, whereas children in the PROMS group reported general learning difficulties. Nonetheless, evaluation of this specific learning skill is important because reading comprehension is perhaps one of the most essential academic skills [78]. Reading comprehension difficulties are associated with other learning problems. For example, in a recent meta-analysis aimed at illuminating the nature of the association between mathematics skills and reading comprehension, reading comprehension was found to have a significantly strong effect on students' mathematics skills [79].

To the best of our knowledge, no previous study has compared learning difficulties evaluated using PROMs with formal evaluations by clinicians in children and adolescents with FNSD. Vassilopoulos et al. [56] examined learning difficulty using children's PROMs and compared it with parents' reports. According to the authors, no discrepancies were found between the children's PROMs and parents' reports. However, this study did not include the learning assessments of educational professionals [56]. Thus, the current results emphasize the importance of including clinician-rated objective assessments of scholastic abilities in children and adolescents with FNSD. Such an evaluation has the potential to identify children with learning difficulties who may have gone unrecognized by their families before entering the rehabilitation program because of their FNSD diagnosis. Consequently, these children may not have received the appropriate learning tools and strategies to better manage their academic difficulties. Furthermore, clinicians should be particularly attuned to a child's expressions during therapy, which may suggest a diminished sense of control, potentially linked to prior experiences with academic challenges. Examples may include statements like "It's too hard," "I can't do it," and "Everyone thinks I am a failure," often linked with feelings of helplessness [61].

Furthermore, it has been documented that youth with FNSD do not attend school regularly [39,56], possibly due to unknown learning problems. The high rate of children and adolescents with FNSD who may be unaware of, or dismissive of, their learning difficulties is concerning. This lack of awareness can prevent them from receiving appropriate interventions, possibly contributing to their school absenteeism. The resulting gap between a child's actual learning abilities and their self-reported abilities can negatively impact their academic performance and social well-being [80]. This emphasizes the importance of close collaboration between the rehabilitation program's educational team and school personnel to optimize the child's successful return to their academic environment.

4.4. Future directions

The current study focused on identifying the discrepancies between PROMs and clinician assessments. Future research should investigate the broader impact of implementing PROMs in routine pediatric care. This could involve exploring how PROMs influence overall patient management, treatment outcomes, and healthcare costs. In addition, the high level of agreement observed between the PROMs and clinician evaluations, particularly in the pain and motor domains, presented a missed opportunity. Future studies should examine the factors contributing to the agreement or disagreement between assessments. This could include parental perceptions and attitudes regarding their children's learning difficulties as well as potential cultural or language barriers affecting communication.

5. Limitations

The interpretation of our findings should be considered in light of some limitations. First, the use of different assessment scales by clinicians and children is a key limitation. The choice of assessment scale can influence the results, affecting the precision, bias, reliability, sensitivity, interpretability, and validity of the assessment outcomes. Therefore, future research should explore the development and implementation of standardized PROMs and clinician evaluation methods to facilitate a more comprehensive understanding of the utility of PROMs in this population. Second, this study focused on the clinical evaluation of reading comprehension. Focusing only on the evaluation of one learning difficulty domain together with the relatively high prevalence of disagreement regarding learning difficulties suggests a need for further investigation. Future research could delve deeper into specific learning areas such as reading, reading comprehension, and mathematics. This granular analysis provides a more nuanced understanding of discrepancies in this domain.

6. Conclusion

Children with FNSD experience high levels of pain, motor skills, and learning difficulties. This study emphasizes the significant concordance between children's self-reports and clinicians' assessments of pain and motor problems, along with lower levels of agreement regarding learning difficulties. The significantly higher rates of disagreement between child reports and clinician evaluations in the latter domain, compared to pain and motor function, underscore the need for a multifaceted approach to pediatric FNSD. Thus, our findings indicate that integrating clinician assessments with child-reported outcomes (PROMs) is essential; however, further research is warranted to explore the factors contributing to the discrepancies in learning difficulty assessments among this population. Another novelty of the current study lies in its utilization of clinically relevant PROMs within a pediatric clinical setting without imposing additional demands on children. Such implementation may alleviate the burden on children with FNSD, particularly regarding pain and motor function, which are known to be sensitive to evaluation. Moreover, the high levels of agreement between child reports and clinician evaluations, particularly in the domains of pain and motor function, suggest that relying more on child PROMs for these domains could reduce the need for lengthy objective assessments, such as the 6-minute walk test (6MWT) or proxy reports. This streamlining could benefit both clinicians and children. Clinicians would have more time to focus on other aspects of the evaluation and build rapport with the child, while children would experience a less burdensome evaluation process.

CRediT authorship contribution statement

Maya Gerner: Writing – review & editing, Writing – original draft, Methodology, Investigation, Data curation. **Etzyona Eisenstein:** Writing – review & editing, Methodology, Investigation, Data curation. **Einat Neeman-Verblun:** Writing – review & editing, Methodology, Investigation, Data curation. **Tamar Ravid Vulkan:** Writing – review & editing, Methodology, Investigation, Data curation. **Tamar Silberg:** Writing – review & editing, Supervision, Methodology, Investigation, Conceptualization. **Jana Landa:** Writing – review & editing, Validation, Supervision, Resources, Methodology, Investigation, Conceptualization. **Sharon Barak:** Writing – review & editing, Writing – original draft, Methodology, Investigation, Formal analysis, Conceptualization.

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Declaration of Competing Interest

None

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