# **Inpatient Treatment of Functional Neurological Disorder: A Scoping Review**

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**ABSTRACT:** Background: The aim of this scoping review is to describe the characteristics of patients requiring admission to hospital for functional neurological disorder (FND), assess interventions provided, and evaluate outcomes in the context of acute hospital presentation or elective admission with chronic symptoms (>3 months). *Methods:* A scoping review was performed. Included articles described adult patients admitted with FND to an inpatient care setting. Articles focusing on psychogenic non-epileptic attacks (PNEA) were excluded. Results: The search strategy identified 1963 citations. A total of 34 articles met inclusion criteria, with 458 patients (66%) female) described. The pooled mean age of patients in all studies was 40.6 years. Eleven studies described patients with acute presentation, and 16 studies described patients with chronic FND symptoms admitted to the hospital. Motor symptoms were most common. Interventions were most commonly physiotherapy and psychotherapy. Most studies reported partial or complete resolution of symptoms. Conclusions: This scoping review summarizes the literature on the characteristics of patients admitted to the hospital, both with acute and chronic symptoms, for inpatient treatment of FND. When comparing patients with acute to those with chronic symptoms, we found that acute presentations were older (46.9 vs. 43.7 years) and had a higher representation of men (33% vs. 30%). Those presenting with chronic symptoms were more likely to not improve or relapse. We postulate that early diagnosis and inpatient rehabilitation could have a positive impact on outcomes for patients with FND.

RÉSUMÉ : Le traitement hospitalier des troubles neurologiques fonctionnels : une étude exploratoire. Contexte : L'objectif de cette étude exploratoire est de décrire les caractéristiques de patients admis dans un établissement hospitalier pour des troubles neurologiques fonctionnels (TNF) mais aussi d'évaluer les interventions qui leur ont été fournies et l'évolution de leur état de santé soit dans le contexte de symptômes sévères ou lors d'une admission facultative pour des symptômes chroniques (> 3 mois). *Méthodes*: Nous avons ainsi effectué une étude exploratoire sur le sujet. Pour ce faire, nous nous sommes penchés sur des articles décrivant des patients adultes ayant bénéficié de soins hospitaliers en raison de TNF. À noter que les articles mettant l'accent sur des crises psychogènes non-épileptiques ont été exclus de notre analyse. Résultats : Notre stratégie de recherche nous a permis d'identifier 1963 citations. Au total, 34 articles ont répondu à nos critères d'inclusion, ce qui représente 458 patients dont 66 % étaient de sexe féminin. L'âge moyen commun de tous ces patients était de 40,6 ans. Précisons que 11 articles ont décrit spécifiquement des patients s'étant présentés à l'hôpital avec des symptômes sévères tandis que 16 autres ont plutôt décrit des patients aux prises, lors de leur admission, avec des symptômes chroniques. Si des symptômes moteurs se sont avérés les plus courants, les interventions les plus fréquentes ont été de nature de nature physiothérapeutique et psychothérapeutique. Enfin, la plupart des articles ont fait état d'une rémission partielle ou complète de leurs symptômes. Conclusions : Cette étude s'est donnée pour objectif de synthétiser des articles scientifiques ayant abordé les caractéristiques de patients présentant des symptômes de TNF sévères et chroniques admis dans un établissement hospitalier en vue d'un traitement. En comparant les patients aux prises avec des symptômes sévères à ceux dont les symptômes étaient chroniques, nous avons constaté que les premiers étaient plus âgés (46,9 contre 43,7 ans) et plus fréquemment de sexe masculin (33 % contre 30 %). Ajoutons aussi que ceux dont les symptômes étaient chroniques étaient plus susceptibles de voir leur état ne pas s'améliorer, voire empirer. Nous faisons l'hypothèse qu'un diagnostic davantage précoce et une réadaptation en milieu hospitalier pourraient avoir un impact positif sur l'évolution de l'état de santé de ces patients.

**Keywords:** Functional neurological disorder, Functional movement disorder, Psychogenic movement disorder, Inpatient rehabilitation, Treatment

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## BACKGROUND

Functional neurological disorder (FND) is a common cause for disabling neurological symptoms, including altered awareness, motor, and sensory changes. Many terms are used to describe this disorder: psychogenic, conversion, somatization, nonorganic, hysteria, shell shock, and medically unexplained. 1-4 The criteria for

diagnosis have shifted from a diagnosis of exclusion to a diagnosis emphasizing positive signs and other features.<sup>5,6</sup> Multiple diagnostic criteria have been proposed over the years for various types of functional symptoms (psychogenic non-epileptic attack [PNEA], functional movement disorders), resulting in significant heterogeneity in how the diagnosis is made.<sup>6</sup>

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Due to the lack of demonstrable structural abnormalities, a common misconception among care providers is that FND is benign. A systematic review examining the prognosis of FND found that a majority of patients continue to experience symptoms and disability years after their initial presentation, often with morbidity equal to or greater than that of other neurological conditions, although the included studies likely underestimate recovery potential, as patients did not receive treatment. Thus, there exists a considerable need to investigate treatment approaches for FND.

There is evidence for multidisciplinary treatment of FND with a symptom-based approach. 8,9 Multidisciplinary teams often include specialists from neurology, psychiatry, psychology, physical therapy, occupational therapy, speech and language pathology, and chronic pain.<sup>8,9</sup> However, what has not been clearly defined is the preferred setting for delivery of treatment, whether it be as an inpatient in acute, psychiatric, or rehabilitation wards, or provided through outpatient clinics. In the current landscape of FND treatment, patients are seen and managed in outpatient specialty clinics, when available. 10 Unfortunately, resources for the treatment of FND are frequently limited, and globally, many patients do not have access to specialists for diagnosis or treatment. <sup>10</sup> Inpatient treatment of FND, especially in the case where admission is elective, is uncommon due to a severe lack of resources. In some centers, patients may be admitted to the hospital if outpatient treatment fails to improve symptoms, if disability is high, or if the symptoms present acutely requiring further investigation and intensive treatment. 11 Of particular interest are the outcomes of patients requiring inpatient care due to severity of symptoms, as this subset of FND patients may have higher disability, risk of iatrogenic harm, and incur a greater cost to health care systems. 11

Inpatients with FND can be divided into two distinct subgroups: (1) patients who present acutely after developing symptoms and (2) patients admitted electively for intensive inpatient treatment of chronic symptoms. The aim of this scoping review was to understand the clinical characteristics of FND patients admitted and treated in the hospital setting. Additionally, this review was intended to better delineate which patient subgroups may benefit from inpatient treatment. This is necessary for the appropriate allocation of limited resources, as well as to maximize the recovery potential of patients. Given the well-known heterogeneity within treatment approaches, direct comparison between programs was not possible. Thus, the purpose of this paper was to understand and not to compare individual treatment programs. Furthermore, the ideal inpatient setting for delivery of therapy, whether it be an acute care unit, psychiatric unit, or rehabilitation unit, is unknown.

## **METHODS**

A scoping review was done to map the emerging research on inpatient functional treatment, reveal methodological gaps, and identify areas for future research. This review included the following five key phases: (1) identifying the research question, (2) identifying relevant studies, (3) study selection, (4) charting the data, and (5) collating, summarizing, and reporting the results. This framework was used to answer the following research questions:

- 1. What are the characteristics of adult patients who require admission to the hospital for functional neurological symptoms?
- 2. What interventions are provided for inpatients with functional neurological symptoms?
- 3. What are the outcomes for inpatients with acute and chronic presentations of functional neurological symptoms?

# Search Strategy

Relevant healthcare databases were chosen for the initial search strategy in consultation with a research librarian for the scope of this review. A search strategy was developed after a preliminary iterative search of the databases. The initial search of databases was conducted on January 22, 2019 in PubMed (1946-Present), Medline (1946-Present), CINAHL (1981-Present), and PsychINFO (1987-Present). A hand search of relevant review and expert articles was undertaken in February 2019. Given the diverse terms used to describe functional symptoms, both historically and across disciplines, a broad search strategy was employed. The terms functional movement disorder, functional symptoms, conversion disorder, conversion, psychogenic, and somatoform were combined with the following appropriate MeSH terms: inpatient, hospital, ward, acute care, neurology, rehabilitation, psychiatry, physiatry, physiotherapy, therapy, and behavioral therapy. Grey literature searches were conducted through relevant websites (e.g. neurosymptoms.org, fndhope.org), relevant conference proceedings, and Google Advanced Search. An a priori decision was made to only search the first 100 Google results given the time necessary to review each result. We also utilized Mendeley Reference Management "Suggest" function based on uploaded articles. The search was limited to French and English language articles only. No date restrictions were applied.

# Eligibility Criteria

Articles were included in the final review if they included data on adults (18+ years) who were admitted with functional neurological symptoms to an inpatient care setting. Articles that focused on the care of those with PNEA or treatment delivered exclusively in an outpatient setting were excluded.

# **Study Selection**

All citations were imported to the Mendeley Citation Management, and duplicates were removed. All potentially relevant citations were independently screened by the two authors by title and abstract. Full-text articles were screened by the two authors to ensure criteria for inclusion were met. Meetings between the two authors were held regularly to discuss differences or ambiguity in the application of inclusion criteria during selection.

# **Data Extraction**

A standardized form was created to extract data from included studies. Detailed information, including author, title, publication year, country of origin, methodology, sample size, institutional setting, admission type, characteristics of admitted patients, interventions, outcomes, follow-up, identified gaps, challenges, and limitations, were obtained upon the second reading of the

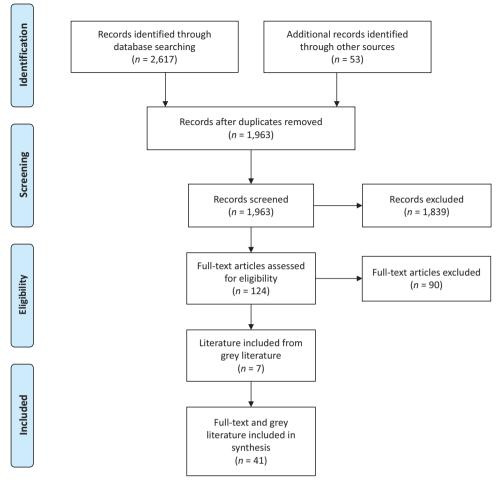


Figure 1: Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flow diagram indicated the number of citations included at each stage of review.

articles. Data extraction was performed on all eligible studies by both investigators.

## **Data Synthesis**

Given the heterogeneity of the included articles, a narrative synthesis was used to organize and summarize the data. <sup>13</sup> Papers were grouped according to the duration of symptoms prior to admission, with symptoms present for less than 3 months being considered acute presentations and longer than 3 months being chronic presentations. Publications were reported by the admission setting and admission type. The quality of included studies was not assessed, as this is not typical for a scoping review.

## RESULTS

## Search and Selection

In total, the search strategy identified 2670 citations. After duplicates were removed, 1963 citations were screened using the inclusion criteria. The title and abstract screen resulted in the retrieval of 124 articles for full-text review. A total of 34 full-text articles met criteria for the final synthesis (Figure 1). After the review of relevant grey literature, seven abstracts were selected from conference proceedings. Attempts were made to contact the

authors to obtain supportive evidence and other data relevant to the research questions.

# **General Characteristics**

Full-text studies (Tables 1–3) and available grey literature (Table 4) were organized by symptom onset, admission type, and study design. The earliest published study of inpatient treatment of functional patients was in 1970.<sup>37</sup> The majority of studies were published between 2010 and 2019 (n = 16). An evolution in terms used to describe functional symptoms has occurred over time. The term "conversion" was used most frequently (n = 18) by studies in this review. More recently, the term "functional" has gained traction in the inpatient literature. Since 2014, there were 13 studies that used the term. Other terminologies included psychogenic (n = 3), hysteric (n = 2), and nonorganic (n = 1).

Most studies were located in the United States (n = 13) and the United Kingdom (n = 6). Fifty percent of studies were case series. The majority of studies were performed on rehabilitation units (n = 16); other units included medical/surgical wards or psychiatric units. Most studies included patients who were admitted electively (n = 27) from outpatient specialty clinics or from referral from general practitioners.

**Table 1: Acute and chronic presentations** 

Setting	Study	Design	Participants	Intervention	Length of stay	Outcomes at discharge	Follow-up
Acute inpatient ward	Matthews et al. (2016) <sup>14</sup> United Kingdom	Prospective case series	n = 35 Age: 33*, 15–58 Female: 71% Onset: 1 day to 10 years (<1 month 60% >1 month 40%)	Diagnostic explanation Mean 11.2 (range 2–43) sessions of physiotherapy Eighteen patients had neuropsychology or liaison psychiatry consultation	Mean 18 days (range 2–62)	Partial: Improvement quantified using MRMI; mean score improved from 20 to 37 out of 40 points Patients with acute presentations gained more points on average (20.5 points vs. 12.2)	Not reported
	Fahn and Williams (1988) <sup>15</sup> United States	Case series	n = 21 (7 patients excluded: age of onset <18 years) Age 33.57 ± 9.7, 23–58 Female: 93% Onset: <1 month to 15 years	Diagnostic explanation Positive reinforcement 60% Psychotherapy 50% Hypnotherapy Physiotherapy 30% Placebo 70% Pharmacologic: MAOi or TCA Antidepressant 30%	Not reported	Complete: 50% Some improvement with return of symptoms: 40% No improvement: 10%	Not reported
Rehabilitation ward	Jordbru et al. (2014) <sup>16</sup> Norway	Randomized controlled trial (intervention vs.4- week waitlist)	n=60 Age: 37.6 ± 11.0, 18–62 Female: 80% Onset: 1 to 48 months	Specialty Functional Program Diagnostic explanation Adapted physical activity with an educational and cognitive behavioral framework. Reinforcement of positive behavior. Interdisciplinary team: Physiotherapy, occupational therapy, and adapted physical activity educator.	3 weeks	Partial: Improvement in gait and functional independence (change in SF-12 not statistically significant)	1 year: stable
	Shapiro and Teasell (2004) <sup>17</sup> Canada	Case series	n = 39 Age median: Acute: 39* Chronic: 42* Acute: Female 89% Chronic: Female 80% Onset: Acute: <2 months 23% Chronic: >6 months 77%	Standard behavioral physiotherapy: Patients were told that therapies were designed to help them relearn proper muscle functioning. Any reference to psychiatric terminology was avoided.  Strategic behavioral physiotherapy: If after 4 weeks of treatment there was no progress, the strategic protocol was implemented.  Patients were told failure to recover is evidence of psychiatric etiology, family meeting at discharge reinforcing psychiatric etiology if relapse	Not reported	Effectiveness of standard therapy was a function of symptom duration (8/9 acute and 1/28 chronic recovered).  Twenty-one patients transferred to strategic therapy after no improvement, 62% improved with strategic therapy.	Not reported
Psychiatric ward	Heruti et al. (2002) <sup>18</sup> Israel	Prospective consecutive case series	n = 34 Age: $30.29 \pm 11.67$ Female: $26\%$ Onset: not reported	Intensive rehabilitation program Some patients were transferred to psychiatry	Not reported	Complete: 26% Partial: 29% Unchanged: 44% (4 patients diagnosed as malingering)	Not reported
	Speed (1996) <sup>19</sup> United States	Retrospective case series	n = 10 Age: 32.7 ± 13.88, 19–69 Female: 50% Onset: mean 27.8 weeks, 0.5–112.	Diagnostic explanation Physiotherapy Occupational therapy Recreation therapy Psychology Skinner learning theory: No reinforcement of abnormal symptoms, confined to a wheelchair initially, ambulation only with treating team supervision, praise for normal ambulation only	Mean 11.8 days (range: 4–22)	Mean FIM score improved from 3.6 to 7. All patients had normal gait on discharge.	7–36 months. Normal gait 70%, partial dependence on a wheelchair 20%, lost to follow-up 10%
	Dickes (1974) <sup>20</sup> United States	Case-control series	n = 16 (2 patients excluded: PNEA) Age: 22.19 ± 4.87, 18–32 Male: 100% Onset: <24 hours to 4 months	Standard of care 50% Behavioral strategies: Earned privileges with improvement as positive reinforcement 50%. All received group psychotherapy and individualized medical therapy including antidepressants as indicated.	Not reported	Complete 69%, partial 6%, unchanged 25% Of those with full/partial improvement, 56% received intervention treatment.	Not reported

Acute and chronic presentations of FND. Study characteristics, characteristics of inpatients, interventions, and outcomes, organized by the setting of inpatient admission. FIM = Functional Independence Measure; MAOi = monoamine oxidase inhibitor; MRMI = Modified Rivermead Mobility Index; PNEA = psychogenic non-epileptic attacks; SF-12 = 12-Item Short Form Survey; TCA = tricyclic antidepressant. \*Unable to calculate the standard deviation with data provided in the study.

**Table 2: Acute presentation** 

Setting	Study	Design	Participants	Intervention	Length of stay	Outcomes at discharge	Follow-up
Acute Inpatient ward	Gargalas et al. (2017) <sup>21</sup> United Kingdom	Retrospective case series	n = 98 FAST + on presentation to ED Age: $49.1 \pm 18.8$ Female: $63.3\%$ Onset: $13.3$ hours $\pm 63$	Hospitalization to rule out stroke	Mean 1.54 days ± 1.2	94.9% discharged home from stroke unit Symptoms on discharge not reported	Not reported
	Letonoff et al. (2002) <sup>22</sup> United States	Case series	n = 3 Age: 26 ± 2.82, 24–30 Female: 100% Onset: <24–36 hours	IV Steroids 66% per spine protocol Daily physiotherapy Psychiatry 33%	4–7 days	Partial: Ambulating without assistance 100%	Interval not stated: Complete 67%, Partial 33%
	O'Neal and Dworetzky (2016) <sup>23</sup> United States	Review with case vignettes	n = 2 (one patient excluded: PNEA) Age: 25 Female Onset: <24 hours	Diagnostic explanation Physiotherapy Occupational therapy Psychiatry Antidepressant (dose not stated)	Not reported	Not reported	6 weeks ambulating with a walker
	<b>Fogel</b> (1976) <sup>24</sup> Canada	Case series	n=3 (one patient excluded: age and PNEA) Age: $35 \pm 14$ , $21-49$ Female: $100\%$ Onset: 7-30 days	Hypnosis 1. Daily for 35 days 2. Weekly for 5 weeks	~1 month	Able to walk unaided     Spasms once every few days	5 years: 1. Complete, no recurrence 2. One episode of self-limiting spasm during the dental procedure
	<b>Atan et al.</b> (2007) <sup>25</sup> Turkey	Case report	Age: 39 Female Onset: <24 hours	SSRI Physiotherapy Transcutaneous electrical nerve stimulation Faradic stimulation	1 week	Partial: Ambulating without assistance	1 month: "Completely normal"
	<b>Chou et al.</b> (2006) <sup>26</sup> Taiwan	Case report	Age: 50 Male Onset: < 24 hours	Physiotherapy Occupational therapy Functional electrical stimulation Psychology	Not reported	Partial: ADL partial independence, and MRC scale for muscle power of the affected side had improved to a grade of 4.	Not reported

Table 2. Continued

Rehabilitation unit	Silver (1996) <sup>27</sup> United States	Case series	n=4 (one patient excluded: outpatient) Age 53.33 ± 6.48, 45–61 Female: 100% Onset: <24 hours to 10 weeks	Behavioral management techniques Double-blind communication strategies Physiotherapy Electromyographic biofeedback Family conference	Not reported	Partial ("not fully independent"): 33% Partial ("independent"): 33% Complete ("premorbid state"): 33%	Not reported
	Ness (2007) <sup>28</sup> United States	Retrospective case analysis	n=3 Age: 24 ± 7.11, 18–34 Female: 100% Onset: <24 hours to 2 months	Physiotherapy Occupational therapy SLP Recreational therapy Psychology	6–9 days	Partial: Independent with transfers, self-care, gait	3 months: No recurrence
	Roberts (1994) <sup>29</sup> United States	Case report	Age: 60 Female Onset: 8 days	Multidisciplinary rehabilitation team Behavioral management	1 month	Partial: Independent with self-care	Not reported
Psychiatric unit	Hersen et al. (1972) <sup>30</sup> United States	Case report	Age: 19 Male Onset: 4 days	Positive social reinforcement	12 days	Complete	Not reported
	Gill (1984) <sup>31</sup> India	Case report	Age: 31 Female Onset: 2 weeks	Psychology Pharmacologic: Trial of antidepressants, barbiturates, and sedatives for 7 days nitroxazepine 75 mg nightly, secobarbital 50mg PRN, chlordiazepoxide 10 mg daily and 20 mg nightly	14 days	Complete	Not reported

Acute presentations of FND. Study characteristics, characteristics of inpatients, interventions, and outcomes, organized by the setting of inpatient admission. ADL = activities of daily living; ED = emergency department; FAST = face arm speech test; IV = intravenous; MRC = Medical Research Council; PNEA = psychogenic non-epileptic attacks; PRN = as needed; SLP = speech and language pathology; SSRI = selective serotonin reuptake inhibitor.

**Table 3: Chronic presentation** 

Setting	Study	Design	Participants	Intervention	Length of stay	Outcomes at discharge	Follow-up
Acute ward	Bharadwaj et al. (2015) <sup>32</sup> Australia	Case report	Age: 61 Male Onset: 1 year	Geriatric unit Relaxation technique CBT Psychiatry Physiotherapy (unclear if this was in the hospital)	Not reported	Cognitive: Complete Motor: Partial over 3 months	1 year: complete recovery
Rehabilitation unit	Jacob et al. (2018) <sup>33</sup> United States	Retrospective chart review	n = 32 Age mean: $49.1 \pm 14.2$ Female: $75\%$ Onset mean: $7.4 \pm 10.8$ years	Specialized motor retraining program Multidisciplinary team: Neurology, psychiatry, psychology, physiotherapy, SLP, occupational therapy, and social work Motor relearning 3 hours per day physiotherapy/occupational therapy and 1-hour psychotherapy	7 days	Independent ambulatory status increased from 59.4% to 87.5% Use of a wheelchair decreased from 21.9% to 3.1%	At 6 months, 69.2% maintained symptomatic improvement from the program. Reduction in physician and eemergency room visits.
	Richardson et al. (2018) <sup>3-4</sup> New Zealand	Retrospective consecutive case series, no control	n = 12 (one patient excluded: outpatient) Age: 41.2, 19–63* Female: 50% Onset: not reported	Diagnostic explanation (hardware/software), "nocebo" model Physiotherapy: 2–4 hours per day, 5 days per week with video playback Psychology Occupational therapy	Not reported	Complete recovery: 83% FIM improved by mean 28.1 in a mean of 14.3 days	Mean 17 months, range 12–26. No recurrence 25%, self-limiting symptoms 42%, relapse 25%, no improvement 8%.
	Delargy et al. (1986) <sup>35</sup> United Kingdom	Case series	n=6 (one patient excluded: age) Age: $52.8 \pm 9.26$ , $37-65$ Female: $60\%$ Onset mean: $7.6 \pm 8.36$ years (range: $1-24$ )	Behavior therapy program Physiotherapy Demonstration of "hidden power" of limb Increased independence with decreasing nursing care	Not reported	Partial recovery (able to walk) at a mean of 5.2 ± 3.25 weeks	Mean 11.8 ± 2.56 months to No relapse
	Withrington and Parry (1985) <sup>36</sup> United Kingdom	Case series	n = 5 Age: 33 ± 5.02, 27–42 Female: 100% Onset: 1–20 years	Physiotherapy Occupational therapy Transcutaneous electrical nerve stimulation	4 weeks to 6 months	Partial return of motor power	3 months to 3 years: Complete recovery
	<b>Trieschmann et al.</b> (1970) <sup>37</sup> United States	Case series	n = 3 Age: 32.67 ± 11.61, 23–49 Female: 100% Onset: 18 months to 15 years	Physiotherapy Skinner learning theory: No reinforcement of abnormal symptoms, confined to a wheelchair initially Psychology Social work	3–5 weeks	Not reported	2–2.5 years. Complete recovery 33%, recurrence 66%
	Teasell and Shapiro (1994) <sup>38</sup> Canada	Case series	n = 3 Age: 42 ± 9.93, 28-50 Female: 100% Onset: 1.5-10 years	Nondisclosure of diagnosis Patients were told that failure to recovery is evidence of psychiatric etiology Physiotherapy Occupational therapy	15 weeks-4 months	Partial at discharge	2 months to 2 years Partial: 33% Complete 33%, Relapse 33%
	Vatine et al. (1996) <sup>39</sup> Israel	Case series	n = 2 Age: 29 ± 11, 18–40 Male: 100% Onset: 6 weeks to 6 months	Physiotherapy Occupational therapy Psychology If no improvement, then transfer to psychiatry unit	4–6 weeks	Partial     Complete	4 weeks: complete     6 months: Complete, no recurrence
	Khalil et al. (1988) <sup>40</sup> United States	Case report	Age: 42 Male Onset 3 months	Physiotherapy Vocational therapy Behavioral therapy Functional electrical stimulation	2 weeks	Partial at discharge	Recurrence at 1.5 years with repeat treatment; no further recurrence at 3 years

 Table 3. Continued

	Oh et al. (2005) <sup>41</sup> Korea	Male Phy Onset not reported ses wit		Psychology Physiotherapy 2 hours per day 5 days per week, 1 session on Saturday, not allowed to walk except with physiotherapy sessions, used a wheelchair at all other times	5 weeks	Partial (GARS improved from 22 to 4)	2 weeks: Ongoing improvement Lost to further follow-up
	Yam et al. (2015) <sup>42</sup> United States	Case report	Age: 45 Male Onset 3 months	Diagnostic explanation Neuropsychology SLP Canine-assisted therapy Physiotherapy Recreation therapy with group Occupational therapy Smoking cessation Pharmacologic: Antidepressant	3 months	Occasional stutter otherwise at baseline	6 months complete resolution
Psychiatric unit	McCormack et al. (2013) <sup>43</sup> United Kingdom	Retrospective comparative study	n = 33 Age: 40.8 ± 12.1, 20–59 Female: 78.8% Onset median: 48 months	CBT 84.9% Neuropsychiatry Psychology Physiotherapy Occupational therapist. Other health professionals (including SLP) consulted as needed	Median 101 days (IQR 84– 130)	Partial at discharge: Improvement in mobility, activities of daily living and mRS	Not reported
	Saifee et al. (2012) <sup>11</sup> United Kingdom	Retrospective case series	n = 26 Age: 47 ± 9.5 Female: 58% Onset: 63% > 3 years	Treatment in specialized multidisciplinary unit which included neurophysiotherapy, occupational therapy CBT 5 days a week for 4 weeks Neuropsychiatry	Median 24 days (IQR 15–32)	Improvement: 58% No change: 35% Worsened: 7%	Median 7 years (IQR 4.5–8.5), Mo (90%) reported fatigue as the main symptom at follow-up, about 60% had weakness
	Moene et al. (1998) <sup>44</sup> Netherlands	Case series	n = 8 Age: 36.5 ± 9.37, 22–48 Female: 100% Onset mean: 9 years (range: 1.5–19)	Diagnostic explanation Psychotherapy Hypnosis Physiotherapy Group therapy 2 hours of bed rest per day	Mean 2 months (range 1 week to 6 months)	Partial (one patient dropped out after 1 week)	6 months to 7 years: three patients had relapses, one did not recover from relapse
	<b>Behr</b> (1996) <sup>45</sup> Canada	Case series	n=3 Age: 36 ± 10.68, 21–45 Female: 67% Onset: 1–6 years	Physiotherapy Weekly sodium amytal interviews Video playback Psychotherapy Pharmacologic: Antidepressants	90 days-4 months	Partial	Not reported
	Radonja et al. (2010) <sup>46</sup> Croatia	Case report	Age: 52 Male Onset: 15 months	Psychotherapy Pharmacologic: maprotiline (75 mg/day), diazepam (20 mg/day), clonazepam (2 mg/day), tramadol (50 mg/day), and quetiapine (50 mg/day)	Not reported	No improvement in dystonia, some improvement in mood	Not reported

Chronic presentations of FND. Study characteristics, characteristics of inpatients, interventions, and outcomes, organized by the setting of inpatient admission. CBT = cognitive behavioral therapy; FIM = Functional Independence Measure; GARS = Gait Abnormality Rating Scale; IQR = interquartile range; SLP = speech and language pathology. \*Unable to calculate the standard deviation with data provided in the study.

**Table 4: Grey literature** 

Study	Design	Participants	Interventions	Length of stay	Outcomes at discharge	Follow-up
MacGillivray (2017) <sup>47</sup> Canada	Case	Age: 51 Female Onset: <24 hours	Expectation management Conceptual model groundwork De-medicalization Pain control Physiotherapy Occupational therapy Psychiatry	5 days	Partial: Frequency and intensity of dystonia attacks reduced	Not reported
Rutherford (2017) <sup>48</sup> United Kingdom	Case	Age: 33 Female Onset: "Chronic"	Multidisciplinary intervention diagnostic explanation Neuropsychology Neurophysiotherapy	2 weeks	Nine-Hole Peg Test decreased from 45 to 23 seconds; 10-meter walk test decreased from 10 to 7 seconds; gait quality improved with a reduction in abnormal posturing and asymmetry and the patient discarded orthosis	3 months: Return to driving and full-time work 9 months: Complete resolution of lower limb symptoms with a return to regular exercise and mild upper limb symptoms on overhead activities only
Boogers (2017) <sup>49</sup> Belgium	Case	Age: 28 Female Onset: 9 months	Physiotherapy Hydrotherapy CBT	Not reported	Partial on discharge: walking normally	Relapse
Arlien-Søborg (2017) <sup>50</sup> Denmark	Program Data	Age: 42 Gender not stated	Length of stay: Mean 8.5 weeks Neurology Physiotherapy Psychology	Not reported	Excellent effect 67%; 26% moderate improvement 26%; mild effect 2.4%; unchanged 4.7%.	3–6 months: Excellent effect 79%; moderate effect 14%
Clea (2018) <sup>51</sup> United Kingdom	Program Data	Data not available	Diagnosis documented in 66% Psychology 26%	Not reported	Not stated	Not reported
Aasnes (2017) <sup>52</sup> Sweden	Program Data	Data not available	Neurology Occupational therapy Physiotherapy Psychology Rehabilitation instructor Social work	Not reported	Partial: Manual wheelchair to walking, with or without aids 80% Patients with tremors, seizures, and other symptoms subjectively have fewer symptoms at discharge.	Not reported
Louissant (2017) <sup>53</sup> United Kingdom	Program Data	Data not available	Multidisciplinary program	4 weeks	Partial: Reduction in patient- reported symptoms 50%; improvement across the five health dimensions in the EQ5D5L 80%	Not reported

Grey literature characteristics of inpatients, interventions, and outcomes. (CBT = cognitive behavioral therapy; EQ5D5L = EuroQol-5D, an instrument for measuring quality of life).

### **Patient Characteristics**

Thirty-four studies addressed baseline patient characteristics (Tables 1–3). A total of 458 patients with FND were described, 66% of whom are female and 34% are male. The age range of all patients was 18–70 years, with a pooled mean of 40.6 years.

All 34 included studies described the type of functional symptoms leading to admission for inpatient treatment. There did not appear to be differences in the presenting symptoms of patients with acute onset compared to those with chronic symptoms. The most common functional symptoms were motor symptoms, including weakness, dystonia, tremor, and gait disorder, present in 350 of 458 patients. Additional functional symptoms in order of frequency were: PNEA (n = 27), sensory (n = 17), urinary (n = 12), cognitive (n = 6), visual (n = 4), speech (n = 2), and hearing loss (n = 2). The authors specifically described 80 patients presenting with multiple functional symptoms, with many studies not providing details beyond the most prominent symptom. The most commonly discussed comorbid symptoms include pain (53 patients) and fatigue (6 patients).

Only nine studies specifically described psychiatric comorbidity, beyond a diagnosis of conversion disorder or somatization disorder. <sup>20,23,27,32,33,39,42–44</sup> Comorbid psychiatric diseases included depression (30 patients), PTSD (30 patients), anxiety (23 patients), ADHD (1 patient), and specific phobia (1 patient). In many larger trials, patients with comorbid psychiatric disease were excluded.

Thirty-one studies (Tables 1-3) discussed the duration of functional symptoms that patients reported prior to being admitted for treatment, with a range of less than 24 hours to 15 years reported. Seven larger studies included a total of 206 patients with both acute and chronic symptoms, with a range of less than 1 day to 15 years (Table 1). In a prospective case series of patients receiving inpatient treatment, Matthews et al. (2016) divided 35 patients into 21 patients presenting with acute symptoms (1 day to 1 month), and 14 patients admitted with chronic symptoms (2 months to 10 years). 14 Jordbru et al. (2014) enrolled 60 patients with a mean duration of symptoms of 9.5 months in a randomized, controlled trial of a 3-week inpatient rehabilitation program. 16 Shapiro and Teasell (2004) described 9 patients with symptoms for less than 2 months and 30 patients with symptoms greater than 6 months in a case series. 17 Fahn and Williams (1988) simply stated that 21 patients in a case series had a duration of symptoms between less than 1 month and 15 years. 15

# **Patient Characteristics: Acute Presentations**

Eleven studies described a total of 115 patients with acute presentations of FND admitted to the hospital, 67% of whom were female (Table 2). The pooled mean age was 46.9 years. Among studies describing patients with acute presentations, 10 studies provided details on the duration of symptoms for individual patients, with a mean duration of  $5.2 \pm 7.8$  days and a median of 1 day. <sup>22–31</sup> Gargalas et al. (2017) reported a mean of 13.3 hours for 98 patients admitted to a hyperacute stroke ward with functional stroke symptoms. <sup>21</sup>

All 115 patients in all 11 studies experienced motor symptoms. Additional symptoms included: sensory (n = 6), cognitive (n = 3), visual (n = 3), speech (n = 2), and urinary (n = 1). In addition to neurological symptoms, patients with acute symptom onset also experienced pain (n = 5) and fatigue

(n=1). Only two studies specifically described patients having psychiatric comorbidities, in both cases generalized anxiety disorder. (n=1).

### **Patient Characteristics: Chronic Presentation**

Sixteen studies described a total of 136 patients with chronic FND symptoms admitted to the hospital (Table 3). Of those patients with chronic FND symptoms, 70% were female, and the pooled mean age for all patients was 43.7 years. Eleven studies reported the duration of symptoms of specific patients prior to admission, with a mean duration of 5.8 ± 6.4 years and a median of 3 years. 32,35-40,42,44-46 Larger elective admission studies presented the duration of symptoms using alternative intervals. Thirty-two patients with a mean duration of symptoms of 7.4 years were described in a retrospective chart review by Jacob et al. (2018). The work of the 26 patients responding to a survey after inpatient admission had a duration of symptoms greater than 3 years. McCormack et al. (2014) reported a median duration of 48 months in 33 patients.

All patients in the 16 studies experienced motor symptoms. Additional symptoms included: PNEA (n=6), sensory (n=4), cognitive (n=3), speech (n=2), urinary (n=1), and dysphagia (n=1). Additionally, some studies described patients experiencing pain (n=6) and fatigue (n=1). Seven studies specifically described patients with psychiatric comorbidities, which included: major depressive disorder (n=5), post-traumatic stress disorder (n=3), generalized anxiety disorder (n=2), attention-deficit/hyperactivity disorder (n=1), and a specific phobia (n=1).  $^{32,33,42-46}$ 

# **Interventions**

Thirty-three studies broadly discussed the types of interventions given to patients while admitted to the hospital (Tables 1–3). The most common types of intervention included physiotherapy (n=29) and psychotherapeutic strategies (n=27). Other common interventions included occupational therapy (n=18), psychiatry (n=15), antidepressant medications (n=6), speech and language pathology (n=3), transcutaneous electrical nerve stimulation and electromyography biofeedback (n=5), recreation therapy (n=2), hypnosis (n=2), and confinement to a wheelchair when not in therapy (n=2). Two studies described behavioral therapy strategies in which patients were told that failure to recover was proof that their symptoms were psychological in origin, thus providing motivation for recovery. The solution and statistically discussed diagnostic explanation as a therapeutic strategy. The solution of the strategy of the solution of

Multidisciplinary approaches were utilized in 26 studies, with various combinations of specialties, including psychology, psychiatry, neurology, physiotherapy, occupational therapy, speech and language pathology, and recreation therapy often described, although not always specifically labeled as a multidisciplinary team.

Treatment elements and disciplines delivered did not differ significantly based on the setting of admission or whether patients were admitted with chronic or acute symptoms. Physiotherapy, occupational therapy, and multidisciplinary teams were more commonly utilized for patients admitted to neurology and rehabilitation, whereas pharmacologic treatments for mood were more commonly used in inpatient psychiatry settings.

Twenty-two studies reported on the length of stay, with a total of 303 patients described. The pooled mean duration of inpatient admission was 24.4 days, with a range of 1.54–180 days. When comparing patients with acute symptoms to those with chronic symptoms, patients admitted with acute symptoms had much shorter lengths of stay (pooled mean 2.9 days) compared to those admitted with chronic symptoms (pooled mean 27.8 days). Longer durations of stay are described in psychiatric admissions (89.8 days), followed by rehabilitation admissions (20.3 days) and inpatient neurology (6.3 days).

## **Outcomes**

A large number of case studies and case series commented on the reversal of symptoms or attainment of independence as qualitative measures of outcomes. Only 10 studies captured health domains, such as degree of disability, cognitive, emotional, and quality of life with specific tools.  $^{11,14,16,19,32-34,41-43}$  Degree of disability was most commonly captured with the Functional Independence Measure (FIM)  $(n=3)^{14,32,34}$  and Modified Rankin Scale (mRS) (n=2). Impact on quality of life was assessed using the health-related quality of life scale in two studies.  $^{34,41}$ 

Many studies reported partial or complete resolution of symptoms after hospitalization. Thirty-nine patients (8%) were reported to have no change or worsening of symptoms after hospitalization. <sup>11,14,17,18,42,44,45</sup> Of those who did not improve, the authors cited premorbid psychiatric diagnosis, <sup>11,18,44,45</sup> long duration of symptoms, <sup>11,42</sup> or attrition <sup>14</sup> as likely factors for poor outcome. Among patients admitted with chronic symptoms, 8.7% had no change or worsened during hospitalization. All of these patients were admitted to psychiatric units. <sup>11,46</sup> There were no reported cases of patients worsening or remaining at their pretreatment level of disability in the acute onset group; however, this must be interpreted cautiously given the implicit reporting bias of published case reports and case series.

The follow-up period after inpatient stay was reported between 2 weeks and as long as 8.5 years, although most studies followed patients within 1 year of treatment. Follow-up symptoms were described in a total of 78 chronic patients (follow-up pooled mean 12.7 months), 14 acute patients (follow-up mean 18.0 months, median 8 months), and 60 unspecified patients (follow-up pooled mean 13.2 months). 11,16,19,22–25,28,32–42,44 A total of 16 chronic patients (18% of patients with reported follow-up) had a recurrence of symptoms in the follow-up period ranging from 1 month to 2.5 years. 34,37,38,40,44 Jacob et al. (2018) reported eight patients that either had no change in symptoms or worsening symptoms at 6 months, but did not distinguish how many of these patients were relapses or nonresponders.<sup>33</sup> None of the 14 acute patients in which follow-up was reported had relapses.<sup>22–25,28</sup> None of the patients with duration of symptoms not clearly specified had reported relapses. 16,19 Notably, reported relapses emphasized motor symptoms.

An alternate diagnosis was rarely reported in the inpatient literature. Heruti et al. (2002) found four patients who did not improve met criteria for malingering. <sup>18</sup> Thirty-two of 68 (47%) patients in Gargalas et al. (2017) had comorbid psychiatric

diagnoses (depression and stress-related conditions) upon follow-up of general practitioner records, in addition to functional symptoms.<sup>21</sup>

### DISCUSSION

We used a narrative synthesis framework to interpret the data given the heterogeneity of the included literature. Consistent with previous reviews, patients with functional symptoms who required admission to hospital were predominantly female. We found that the majority (76%) of patients admitted to the hospital had functional motor symptoms. When comparing acute to chronic onset of symptoms, we found that acute presentations were older (46.9 vs. 43.7 years) and had a higher representation of men (33% vs. 30%).

In our review of the literature, we found that no study of either patients with acute or chronic FND symptoms explicitly gave reason or provided criteria for admission to the hospital. Most studies described patients being admitted electively with functional neurological symptoms after the diagnosis is made in an outpatient specialty clinic, particularly those describing patients with chronic symptoms. Elective admissions were more likely to have symptoms for greater than 1 month, but the exact criteria for admission were not explicitly described beyond the presence of functional symptomatology and the patient being agreeable to admission.

Recently, the Diagnostic and Statistical Manual of Mental Disorders (DSM-V) has made significant revisions to the diagnostic criteria for FND, such as including positive clinical findings on physical examination, removal of identification of underlying stressors, and eliminating the need to rule out malingering or organic disease. Importantly, the addition of positive clinical findings on examination requires adoption and expert performance of a validated physical examination and consistent reporting of findings. Many studies included in this review did not specify criteria the patient met or which clinical findings best supported the diagnosis of FND. Given the prevalence of functional symptoms in inpatient practice and the need for larger scale randomized trials, validated diagnostic tools and a severity scale would be of significant benefit to advance research on effective treatment.

The evolution of terminology describing functional symptoms in the literature reflects the changes in the DSM-V.<sup>2</sup> Historically, functional symptoms have been viewed as psychopathology, either as hysteria or conversion of psychological complaints to somatic symptoms.<sup>2</sup> In this review of patients admitted with functional symptoms, we found that only 26% of studies described psychiatric comorbidity, with only 19% of included patients having diagnosed psychiatric comorbidity. The assumption that neurologic symptoms are a result of psychiatric illness is echoed throughout the literature; however, exploration of comorbid psychiatric conditions was not adequately captured in the included studies to comment on the need for psychiatric intervention for admitted patients.<sup>2,44,45</sup> Results of psychotherapeutic intervention in this review are mixed, but very few studies included tools that would capture the benefit of psychiatric intervention. 16,32,33,42 Admission to a psychiatric unit had a higher use of pharmacologic treatments and, generally, had a longer length of stay, with similar mobility outcomes to other settings. There may be a subset of patients with functional neurological symptoms who

will benefit more from psychological intervention than others, but this question requires further investigation.

The literature from the 1970s until now demonstrates an evolution from a paternalistic model where the diagnosis was not discussed and privileges were earned to a shared clinical diagnosis and treatment, with an increasing emphasis on selfmanaged care. Previous deceptive approaches reinforce the misconception that patients are "faking" and that "saving face" allows acceptance.<sup>54</sup> Another paternalistic approach described had patients earn privileges back slowly through participation in therapies. The concern with this technique is that this reinforces an external locus of control and downplays self-management strategies, further perpetuating reliance on the healthcare provider and system. 54 In reality, approaches that reinforce the passive and "faking" behaviors are a significant barrier to patients having access to appropriate treatment. Approaches to the management of functional symptoms need to ensure that trust and accountability underlie the care provider and patient relationship.

The studies included in this review suggest that inpatient intervention can have a positive impact on outcomes for those with functional symptoms. Inpatient populations add an additional challenge to understand the impact of each intervention, as multiple therapies are occurring simultaneously.<sup>54</sup> A multidisciplinary approach is often cited in the literature, but who is involved is dependent on available resources. The rationale for the selection of rehabilitation specialists was often not discussed.

The appropriate length of inpatient treatment remains unclear, but it can be inferred from this review that it is appropriate to admit patients with the goal of functional independence, but not total symptom resolution. When comparing symptom onset, patients presenting acutely had a mean length of stay of 2.9 days when compared to a mean of 27.8 days for those with symptoms occurring for greater than 3 months. This difference in length of stay likely reflects the differing goals of admission, with patients admitted acutely being discharged after the diagnosis is made rather than after deliberate aspects of treatment. Additionally, those with chronic symptoms had a mean onset of  $5.8 \pm 6.4$  years compared to acute presentations of  $5.2 \pm 7.8$  days. This has significant implications for healthcare utilization and cost. Future studies exploring a "staged approach" to functional symptoms, such as acute admission for motor symptoms followed by outpatient programs to support other comorbidities, such as depression, cognitive changes, pain, and fatigue, may be helpful in ensuring timely, appropriate diagnosis and treatment while reducing system burden.

The prognosis for patients with functional symptoms is often cited as poor in the literature, although previous studies likely underestimate recovery potential, as most patients do not receive treatment.<sup>55</sup> Our review of inpatient treatment of functional symptoms is promising in terms of motor outcomes. Physiotherapy was consistently used as an inpatient intervention with positive results. The majority of patients return to independent function, but remain symptomatic after an inpatient stay, with the caveat that this review captures many patients described only in case reports and small case series. Patients admitted to psychiatric units were more likely to not improve or worsen compared to those admitted to acute care or rehabilitation wards, perhaps reflecting more complex psychiatric comorbidities and manifesting more severe disease; however, given the quality of the studies available, no recommendation can be made for an optimal inpatient setting to treat FND. Consistent use of validated tools

is needed to better delineate outcomes based on settings, rehabilitation methods, and other interventions.

In terms of outcomes after discharge from hospital, most reported motor symptom recurrence occurred within a year after hospitalization with reported relapses occurring between 1 month and 2.5 years. For patients that did relapse, most recovered to functional independence with rehabilitation. All reported instances of symptom recurrence were described in patients with chronic symptoms (18% of chronic patients with reported follow-up); however, many acute presentation studies only followed patients for 1–3 months after discharge. 34,37,38,40,44

The strengths of this review included the diverse terms for FND used and lack of constraints on date range, allowing for an extensive review of available literature. Furthermore, by not limiting publication date to a specific range, this review was able to capture a historical perspective and the shifting views of FND. Limitations of this review include restriction of language to English and French due to available translation resources, and the majority of the studies are case reports and case series, thus limiting the interpretation of the available data. In keeping with the intent of a scoping review, we did not formally address the quality of the included literature. The majority of patients in the included studies had motor symptoms, making conclusions difficult to apply to FND patients with non-motor symptoms. We believe that there may be a bias among clinicians to admit patients with motor symptoms, as these symptoms are often considered to be more amenable to standard neurorehabilitation programs. The duration of symptoms is likely often underestimated by patients and clinicians, with clinical experience revealing many patients often have functional symptoms, frequently non-neurological, occurring even in childhood. There is significant heterogeneity among inpatient therapy programs provided limiting any comparison between programs. Finally, it is a limitation to group together patients admitted electively with chronic symptoms and those admitted emergently with acute symptoms, as these patients are at very different points on their illness trajectory and, in some cases, may represent different FND populations. Attempts were made to separate these patients into distinct groups as part of this scoping review, but this was not always possible with the available data.

This scoping review summarizes the available literature on the inpatient treatment of both chronic and emergently admitted patients with FND in a variety of settings. With the current evidence, it remains unclear which patients benefit most from inpatient treatment of FND. This question may be a complicated one, with specific patient characteristics, such as primary motor symptoms and acute presentations, possibly leading to better outcomes in inpatient settings. This review identified that patients presenting with acute symptoms (<3 months) had better outcomes at discharge, without any reported relapses during the follow-up period. In contrast, patients presenting with longer duration of symptoms preceding admission had worse outcomes. Although not found in this review, possibly due to reporting bias, clinical experience indicates that patients with chronic symptoms may even decompensate in hospital. We postulate, based on the available inpatient data, that early diagnosis and inpatient rehabilitation in the acute phase could have a positive impact on outcomes for patients with FND.

There are several important areas within the field identified by this review that need to be addressed to ensure treatment is informed by high-quality data. Critically, there must be agreedupon diagnostic criteria with standardized reporting, so that comparisons can be made between programs. In most countries, inpatient treatment is limited due to high cost and resource allocation. An important area of future research could be early, goal-directed therapy with the incorporation of standardized outcome tools, investigating specific patient characteristics preferentially responding to inpatient treatment that may guide the appropriate triaging of limited resources.

### CONFLICT OF INTEREST

Dr. Gilmour has nothing to disclose. Ms. Jenkins has nothing to disclose.

### STATEMENT OF AUTHORSHIP

GG: Conceptual idea for the article, literature search, data analysis, manuscript writing, and critical revisions. JJ: Conceptual idea for the article, literature search, data analysis, manuscript writing, and critical revisions. All the authors read and approved the final manuscript.

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