

Remote delivery of cognitive behavioral therapy to patients with functional neurological disorders: Promise and challenges



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ABSTRACT

Functional neurological disorders (FND) are an important source of healthcare utilization and morbidity. While there are no formal guidelines for treating these disorders, cognitive behavioral therapy (CBT) is emerging as a safe and effective treatment. Currently, there is a global shortage of CBT providers, with only a small subset trained in and comfortable with treating patients with FND. We highlight four types of remote CBT delivery to patients with FND to alleviate the access obstacle: workbooks, internet-guided CBT, app-based CBT, and teletherapy. CBT workbooks and teletherapy have been studied in FND, with preliminary studies suggesting efficacy; internet-guided CBT and app-based CBT have not but have been effectively used in patients with psychiatric disorders, particularly depression, anxiety, and post-traumatic disorders. As these disorders are often comorbid and share overlapping neurobiology with FND, internet-guided CBT and app-based CBT represent promising delivery options of CBT for FND. Although remotely-delivered CBT is unlikely to replace in-person CBT and there are technical and logistical challenges to overcome prior to widespread deployment, it holds promise as an adjunct treatment when in-person CBT is inaccessible. We propose a rational approach to future allocation of remote CBT treatment options and highlight important research gaps to bridge beforehand.

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1. Introduction

Functional Neurological Disorders (FND) are a collection of disorders characterized by neurologic symptoms that are inconsistent and incongruent with clinical and neuroanatomic patterns of known neurological diseases [1]. Previously termed variously as “psychogenic disorders,” “conversion disorders,” and “hysteria,” FNDs have an incidence of 4–12 per 100,000 people and a prevalence of 50 per 100,000 people [1]. The two most common types of FND are functional (or dissociative) seizures and functional movement disorders (FMD), with a yearly incidence of 1.5–4.9 and 4–5 per 100,000 people, respectively [1]. FND disability can be similar in severity to equivalent non-functional neurological disorders (e.g., functional tremor and essential tremor, or functional seizures and epileptic seizures) but yields comparatively increased rates of total symptom burden and mental distress [2–4].

While there is no definitive treatment for FND, the general consensus is that assertive and compassionate delivery of the diagnosis and its understanding and acceptance by the patient are the key ingredients for increasing the odds of therapeutic success [5,6]. Cognitive behavioral therapy (CBT) is emerging as a preferred treatment to address the dysfunctional core beliefs and behaviors believed to underpin the generation of FND. After identification, these dysfunctional beliefs and behaviors are targeted, with a goal of providing the patient with a way to shift dysfunctional core beliefs (or cognitive distortions) into functional core beliefs. Symptom improvement hinges on the recognition that symptomatic exacerbations are connected with a particular dysfunctional core belief, with symptom improvement or cessation dependent on the unmasking of such core belief as untruthful and unhelpful, with its subsequent replacement with the corresponding truthful and helpful counterpart.

CBT addresses cognition in multiple ways including identifying and challenging unhelpful thinking styles [7], reattributing the patient’s symptoms to psychosocial issues [8], learning to accept panic without panicking [9], and targeting catastrophic symptom expectations and a low sense of control over symptoms [10].

In addition, CBT addresses behaviors in multiple ways including teaching relaxation and breathing techniques [7,9] and developing competing responses to patient’s symptoms [7,10]. Relaxation techniques emphasized include progressive muscle relaxation where patients progressively flex and relax different muscle groups, various breathing exercises, and mindfulness [7,9]. Examples of developing competing responses to patient’s symptoms include tackling the avoidance to various activities or avoiding decreased activity because of symptoms, and using principles of habit reversal [7,10].

While CBT is a promising treatment for FND, there is a shortage of CBT providers [11], and an even greater shortage of CBT providers comfortable with treating FND patients. Thus, this review assesses the promises and challenges of using remote, particularly self-guided CBT, to manage FND away from specialized treatment centers. Notably, remote treatment of FND with CBT is a target for future research as few studies have been conducted in this area. Thus, much of the evidence cited in this paper pertains to remote CBT treatment in psychiatric disorders, which are often comorbid and share overlapping neurobiology with FND. The studies that specifically address remote CBT for FND generally have small sample sizes. Future randomized controlled trials (RCT) are needed to

confirm the overall effect of remote CBT on FND symptoms, the aspects of FND most susceptible to benefiting from remote CBT, and the predictors of treatment success.

2. CBT for FND

A 2007 systematic review of 34 RCTs, which involved somatoform disorders as well as functional disorders, found that CBT had a significantly greater effect size than antidepressants and behavioral therapy [12]. Many studies have shown CBT to be effective in functional seizures [13]. In the CODES trial, a multicenter RCT in 27 sites in the UK that compared CBT+ standardized medical care vs. standardized medical care only [9], the primary outcome showed no significant difference in monthly functional seizure frequency between groups. However, the CBT arm was significantly more effective in many of the secondary outcomes, including how bothersome the seizures were to the patients, patient-reported health-related quality of life, psychosocial functioning, psychological distress, and somatic symptoms. There was also increased patient and physician reported clinical improvement in the CBT group. In addition, a 2019 prospective study of functional tremor found that 9 of 15 patients reached full tremor remission after a 12-week course of CBT [14]. Thus, CBT is a promising intervention for patients with FND.

In the Greater Cincinnati area, 111 CBT providers were contacted to determine if they were comfortable and/or willing to accept FMD patients. Of those contacted, 12 were independent psychiatrists, 7 were psychiatry offices, 24 behavioral health clinics, 5 neuropsychology clinics, 25 doctors of philosophy (PhD), 11 doctors of psychology (PsyD), 2 licensed independent social workers (LISW), 1 a licensed independent social worker (LISW), 6 licensed professional clinical counselors (LPCC), 1 a licensed professional counselor (LPC), 1 a doctor of education (EdD), and the remaining providers each had multiple licenses, including a combination of a masters of education (M.Ed), LPCC, PhD, American Board of Professional Psychology certified (ABPP), PsyD, professional limited liability company (PLLC), Masters of Business Administration (MBA), American Board of Sleep Medicine certified (ABSM), Master of Public Health (MPH), Academy of Certified Social Workers certified (ACSW), and LISW. Those who wanted to help this patient population but were unfamiliar with FMD were offered a workbook to help guide them. Of the 111 providers, only 12 were comfortable accepting and treating these patients. Of these 12, seven were behavioral health clinics, three were independent providers with a PhD, one had an LISW, and one had a PsyD.

Remote delivery of CBT has the potential to increase access to treatment by removing the barriers of location, time of transportation, and limited availability of the few CBT providers for FND. With self-guided forms of remote CBT through the use of a workbook, the internet (iCBT), or an application on a smartphone or tablet (app-based CBT), the need for a therapist can be decreased or eliminated entirely. Currently, remote delivery of CBT most commonly involves teletherapy, in which a therapist interacts with the participant through either video-calls or telephone calls in a traditional weekly manner. This latter form of remote delivery is also limited by the availability of time by CBT therapists. These four remote delivery methods of CBT are discussed below.

3. CBT workbooks for FND

CBT workbooks have been developed for both FND, “Overcoming Functional Neurological Symptoms: A Five Areas Approach” [7], and functional seizures, “Taking Control of your Seizures: Workbook” [8]. While other workbooks have been developed as well [15], there are no published studies of their efficacy.

“Overcoming Functional Neurological Symptoms: A Five Areas Approach” was studied as a self-guided therapy and compared to usual care for adult neurology outpatients who had any form of FND and did not require specialist psychiatric care [16]. This self-guided workbook was supplemented by four 30-minute guidance sessions by trained therapists throughout a 12-week course. In total, 127 participants were enrolled, and a Clinical Global Improvement Scale (CGI) survey was collected on 125 participants. After 12 sessions, participants in the self-guided workbook group had significantly greater CGI-based improvement than usual care, with an odds ratio of 2.36 ($p = 0.016$).

“Taking Control of your Seizures: Workbook” was studied as an adjunct to in-person CBT. In a 2009 prospective study, 21 participants were given 12 weekly CBT sessions by a trained therapist as well as the functional seizure workbook, with 17 participants completing the intervention [17]. Of these 17 participants, 11 had no seizures by the end of the 12 weeks. A subsequent four-arm multicenter RCT compared the workbook and 12 weekly sessions with a trained therapist, to sertraline, sertraline + CBT, and treatment as usual [18]. In total, 34 participants were included in the final analyses, which found that the CBT arm yielded a 51.5% seizure reduction ($p = 0.01$), CBT + sertraline a 59.3% seizure reduction ($p = 0.008$), and sertraline and treatment as usual both showed no significant reduction in seizures ($p = 0.08$ and $p = 0.19$, respectively).

While neither workbook has been studied as a standalone treatment, both workbooks significantly decreased FND symptoms. Future studies should assess whether workbooks can be used as a standalone therapy by subsequently decreasing the amount of therapist involvement. However, even if future studies find therapist involvement to be integral to successful treatment with workbooks, “Overcoming Functional Neurological Symptoms: A Five Areas Approach” has already shown it can increase accessibility to FND treatment by decreasing the standard weekly therapy sessions to once every three weeks over the course of 12 weeks.

4. iCBT

iCBT, or internet-delivered CBT, is usually delivered as self-paced modules that participants can access on the internet. While it has not been studied in FND, it has been evaluated in patients with mental disorders and somatic symptoms, such as depression [19], anxiety [20], fatigue [21], insomnia [22] and DSM-5 somatic symptom disorders targeting predominantly chronic fatigue and pain [23]. These disorders are responsive to in-person CBT and share with FND overlapping neurobiological brain dysfunction [24,25]. As such, the feasibility and efficacy of iCBT in psychiatric disorders informs the potential harms and benefits of iCBT as translated for FND.

4.1. Self-guided iCBT

Studies have been conducted on fully automated, self-guided iCBT protocols for psychiatric disorders. In a meta-analysis studying RCTs of self-guided iCBT for depression and anxiety, 13 trials were identified, all of which consisted of online modules that the participants completed on their own [26]. Most of these RCTs consisted of 4–11 online self-guided modules. Notably, no support on

therapeutic content was provided. Nine of the 13 trials reported significant improvement of depression and anxiety symptoms with iCBT compared to control groups, which included waitlist [27–32], treatment as usual [28,33–37], psychoeducation [38], and “attention placebo,” which consisted of weekly contact with a lay interviewer to discuss lifestyle factors that can affect depression [39]. However, across all 13 studies, there was a significantly greater rate of dropouts in the iCBT groups compared to control groups ($p = 0.01$) [26]. Together, these studies suggest that self-guided iCBT may be also effective in FND if adherence to treatment can be addressed.

4.2. Therapist-supported iCBT

A recent study examined five clinics in five separate countries that have successfully provided validated iCBT as part of routine care for psychiatric disorders [40]. The treatment procedures were similar across clinics: patients regularly read material and practice skills on their own, adherence was encouraged through automated and individualized messages, and there was a 10- to 20-minute guided therapy session per week. Thus, these are therapist-guided iCBT models rather than fully automated, self-guided iCBT models. All five clinics treated a large number of patients with therapist-guided iCBT and migrated this treatment paradigm from pilot studies to permanent services with secure funding, and published results pertaining to the efficacy of their program [40]. They all report clinically significant improvement in symptoms and high user satisfaction. While these models are not fully automated and thus do not eliminate the need for a therapist completely, they do decrease the amount of contact needed with a therapist from the traditional 60 minutes per week to 10–20 min per week. Thus, these clinics provide evidence that iCBT can help mitigate barriers to care due to a shortage of qualified therapists.

A few factors were consistent across iCBT delivery in all five sites and identified as contributing to the success of iCBT: (1) strong clinical, IT, and organizational governance, (2) robust links with funding bodies to sustain the programs, (3) a centralized and specialized group providing this service across a population, (4) monitoring patient satisfaction and use of patient feedback to improve procedures, (5) well-developed systems to monitor the quality of care and treatment progress, and (6) self-referrals as well as referrals from health professionals to eliminate barriers of care [40]. It is likely that these same factors will be necessary for the success of therapist-supported iCBT for FND.

4.3. Self-guided iCBT vs. therapist-supported iCBT

Some trials have compared iCBT with and without support from a therapist to determine if human support increases effectiveness or adherence to iCBT. The results have been mixed. An RCT in 2011 compared iCBT + weekly telephone calls vs. iCBT only vs. weekly telephone calls only vs. treatment as usual, and found that telephone support did not provide any additional advantage in symptom improvement with iCBT [36]. This result was supported by an RCT in 2017 that compared iCBT with automated texts/emails vs iCBT with brief weekly support phone calls without clinical content vs. waitlist. They found that both intervention groups improved significantly compared to the control, with no significant difference between the group that received automated messages and the group that received human support [31]. Notably, adherence and patient satisfaction were high with and without human interaction. Particularly, the REEACT trial, a large scale RCT testing iCBT for depression, found poor adherence which prevented significant symptom reduction with iCBT despite weekly telephone support with a therapist [37].

However, in an RCT that compared iCBT only vs. iCBT with human support on request vs. iCBT with weekly scheduled human support vs. human support only vs. waitlist found that only iCBT with weekly human support group showed significant improvements in symptom reduction from baseline.

4.4. iCBT for FND

4.4.1. Safety

While iCBT has not been studied in FND and the reported studies were conducted using iCBT for psychiatric disorders, much can be gleaned from these studies about iCBT as a potential intervention for FND. In general, iCBT seems to be safe [26]. Evidence suggests that the risks outweigh the benefits for iCBT in psychiatric disorders. Since iCBT for FND would use the same treatment modality with only changes in order to adapt content to FND patients, there is no reason to believe that iCBT will be unsafe in FND.

4.4.2. Efficacy

Although results about iCBT efficacy in psychiatric disorders are mixed when viewed as a whole [27,30–32,35,36,38,39], this picture changes when examining individual interventions. For example: “Deprexis,” an iCBT intervention targeting depression, has consistently shown positive results across five trials [27–33,34]. This suggests that there are specific components within an iCBT delivery or content that affect its effectiveness. It will be important to determine what these specific components are in order to ensure the success of iCBT in FND.

4.4.3. Promises

Many of the benefits of iCBT are universal, regardless of the disorder it aims to treat: iCBT, especially self-guided iCBT, is less costly and more accessible than treatments that require intense therapist involvement. In addition, iCBT can be accessed anytime of the day or week independent from the availability and access to therapists knowledgeable in FND. Finally, as most of the treatment is conducted via the internet, the costs of widespread delivery are expected to be low beyond initial investments, likely inducing a high return on investment.

4.4.4. Challenges

There are also aspects of iCBT for psychiatric disorders that are challenging, and these will likely also be a challenge for iCBT adapted for FND. First, iCBT is more manualized and structurally rigid than in-person CBT, and thus it is harder to tailor the treatment to the needs of individual participants. In addition, there is no direct therapeutic connection in self-guided iCBT and only partial in therapist-guided iCBT, which may dampen the effectiveness of CBT [41]. Studies are needed to evaluate the effectiveness of self-guided iCBT as compared to therapist-guided iCBT in FND. In addition, it is important to determine the dose and frequency of therapist interaction needed, if any, for iCBT to be effective in FND. This type of therapist involvement can be predetermined (e.g., weekly) or participant-driven (i.e., occur only if the participant requests it). Another challenge with iCBT is that the participant must have internet access, a source of “digital divide” which can be a barrier to care in marginalized populations. Although epidemiological studies have shown that 82% of people in the United States have internet access [42], it is possible that those who do not have access to internet overlap with those without access to an in-person therapist, the very group that iCBT is targeting.

5. App-based CBT

App-based CBT has not been studied to a similar extent than iCBT. A major benefit is that smartphones and tablets are more portable than a computer or laptop, making them more practical for participants to access this resource during or shortly after an exacerbation of symptoms. Otherwise, app-based CBT that uses modules that participants complete either on their own or with variable amounts of therapist guidance is presumed to have many of the same costs and benefits as iCBT, with the only difference between the two being the medium in which they are delivered.

Chatbots. In this paper, app-based CBT interventions will not refer to modular, self-paced CBT apps, but rather chatbot-enhanced CBT apps. Chatbots are systems that provide automated responses to participant responses and are more easily integrated into an application on a phone than a website on the internet. Commercially available apps that allow participants to have conversations with other people are already widely used. Since chatbots aim to mimic human interaction, they have the unique potential to use conversation to guide a participant through CBT, much like a therapist would. A few chatbots have been developed, studied in depression, and available commercially [43–45]. While there are many ways of creating chatbots, currently available chatbots all utilize various forms of decision tree models. Fig. 1 provides an example of a decision tree created in preparation for a Phase 2 app-based CBT trial in preparation.

5.1. Promises and challenges of App-based CBT

A recent review of commercially available chatbots for depression found encouraging preliminary evidence on the effectiveness of chatbots in treating depression [46]. All three chatbots identified delivered CBT by using decision trees to guide conversation. In terms of effectiveness, one chatbot had an 85% adherence rate and showed a significant reduction in depression symptoms compared to an informational e-book in an RCT [44]. Another showed that more engaged app users had a significantly greater reduction in depression symptoms compared to less engaged users [45]. The third had a 78.6% adherence rate and showed a significant decrease in perceived stress and increase in psychological well-being compared to a waitlist group [43]. Notably, these adherence rates are greater than the 50% adherence rate of internet-based interventions [47].

Comments from participants who used the apps suggest that the human-like qualities of the chatbot and the conversational delivery of CBT helped with adherence. It is then likely that adherence can be increased even more if artificially intelligent chatbots were developed that are more human-like and better mimic the flow of natural conversation. This can be done by utilizing more sophisticated machine learning techniques, such as neural networks, than those currently applied for decision-tree models. An improved naturalistic model would allow free text input by the user rather than choosing from a list of predetermined options when answering a question from the chatbot. In addition, it would allow the chatbot to learn over time and address questions and answers that it was not originally trained for, a feature that is not present in decision tree models. However, a neural network that can address the nuances and vast array of topics CBT addresses would need extensive training and require a large, representative dataset of therapy session transcripts currently unavailable and difficult to obtain due to patient confidentiality issues. If app-based CBT were to be adapted to treat FND, such a dataset would be even harder to obtain due to the scarcity of CBT sessions being used to specifically target FND in current practice.

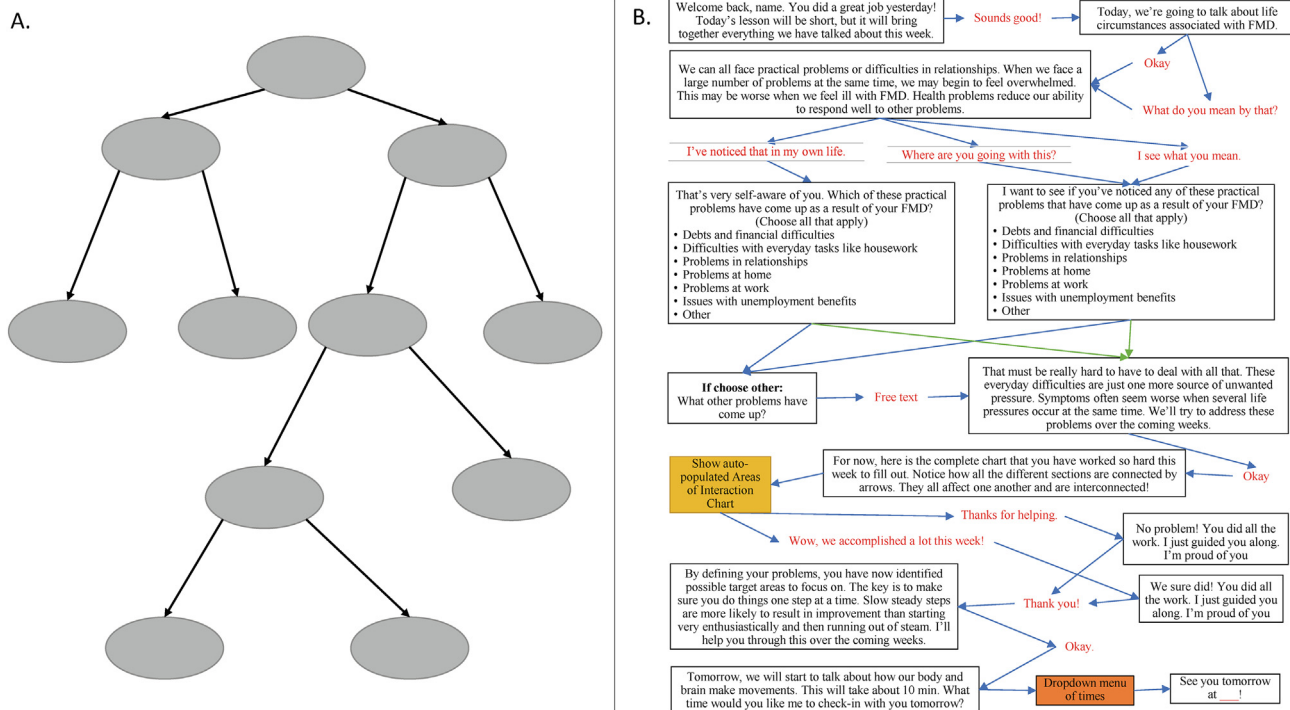


Fig. 1. Decision tree approach to chatbots. Panel A shows a schematic of a decision tree. The conversation starts at the topmost node, with subsequent branches and nodes representing potential paths for a “naturalistic” conversation. Panel B gives an example of how a decision tree can be utilized to guide a conversation with a chatbot.

Regardless of whether decision trees or neural networks are used, app-based CBT may represent an improvement over iCBT by improving adherence to treatment.

6. CBT teletherapy for FND

Teletherapy, defined here as therapy conducted one-to-one with a live therapist either through a phone or videocall, is emerging as an increasingly utilized resource in healthcare given the COVID-19 pandemic. To date, only one study has analyzed the remote delivery of CBT for FND via teletherapy. In 2020, LaFrance et al. conducted a single arm prospective cohort study on 32 patients with functional seizures. The intervention was 12 sessions of weekly manual-based psychotherapy given through video telehealth. Seizure reduction after treatment was 46% ($p = 0.0001$) per month over the course of treatment [48].

6.1. Promises and challenges of teletherapy

While this is the only study of CBT teletherapy for FND, numerous studies have been conducted on CBT teletherapy for psychiatric disorders. Meta-analyses have found therapy delivered via telephone [49] or videocall [50] to be clinically effective when compared to controls, as well as having no significant difference in effectiveness when compared to in-person CBT [51,52]. Together, this suggests that teletherapy, whether over the phone or through videocall, may be equal in effectiveness to in person CBT. This is likely because the interaction between a therapist and a patient is maintained and the content tailored during teletherapy to an extent that a workbook, iCBT, or app-based CBT cannot. The effectiveness of teletherapy for FND, if confirmed, would decrease barriers to care, including geographic location, transportation, and time-constraints, but not the shortage of CBT providers on which it depends.

7. Delivery of CBT treatment for FND

Studies are needed to compare different forms of remote therapy to each other (e.g., self-guided CBT vs. teletherapy) as well as comparing remote therapy to in-person therapy. Remote delivery of CBT for FND will probably not emerge as a replacement to in-person CBT. It is likely that in-person CBT and teletherapy will be shown to be superior to self-guided CBT, but with a shortage of CBT providers for FND still making self-guided CBT an important tool for clinicians treating FND in situations of poor access to therapists. If the current model of offering in-person CBT treatment for FND on a first-come first-serve basis remains the standard, then self-guided CBT --whether in the form of a workbook, iCBT, or app-based CBT-- can be offered to any patient while they are on the waitlist for a trained therapist. Ultimately, however, clinicians would ideally use self-guided forms of CBT to treat most patients while preferentially allocating in-person CBT to those with the greatest severity. An approach to choose the optimal treatment for an individual is suggested below.

7.1. A stepwise approach

If the degree of therapist involvement is correlated with the effectiveness of an intervention, then a stepwise approach to FND treatment may be a practical approach (Fig. 2). In this model, participants would start with the intervention that is most accessible to them: fully automated, self-guided CBT. This can be in the form of a workbook, iCBT, or app-based CBT. If the participant does not improve with self-guided CBT, treatment can then be supplemented with varying levels of therapist involvement via phone calls or videocalls. The amount of remote therapist involvement can be increased as needed, up to the standard one-hour weekly sessions used in traditional CBT. If an hour a week of teletherapy is still ineffective for an individual, then in-person CBT may be deployed. Such a method would increase accessibility of treatment

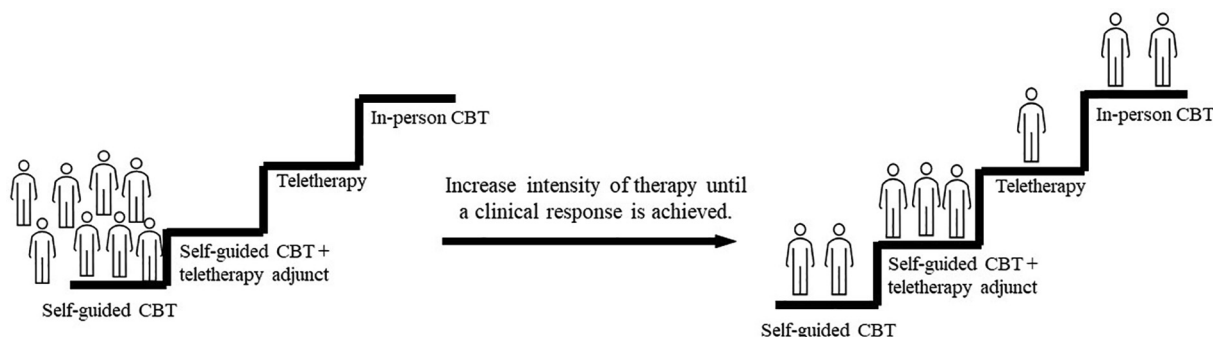


Fig. 2. Stepwise approach to providing CBT for FND. Participants start with the most accessible modality, self-guided CBT (workbook, iCBT, or app-based CBT). Further increases in complexity will be dependent on response to treatment and availability to FND-trained CBT therapist. CBT = cognitive behavioral therapy; FND = functional neurological disorder.

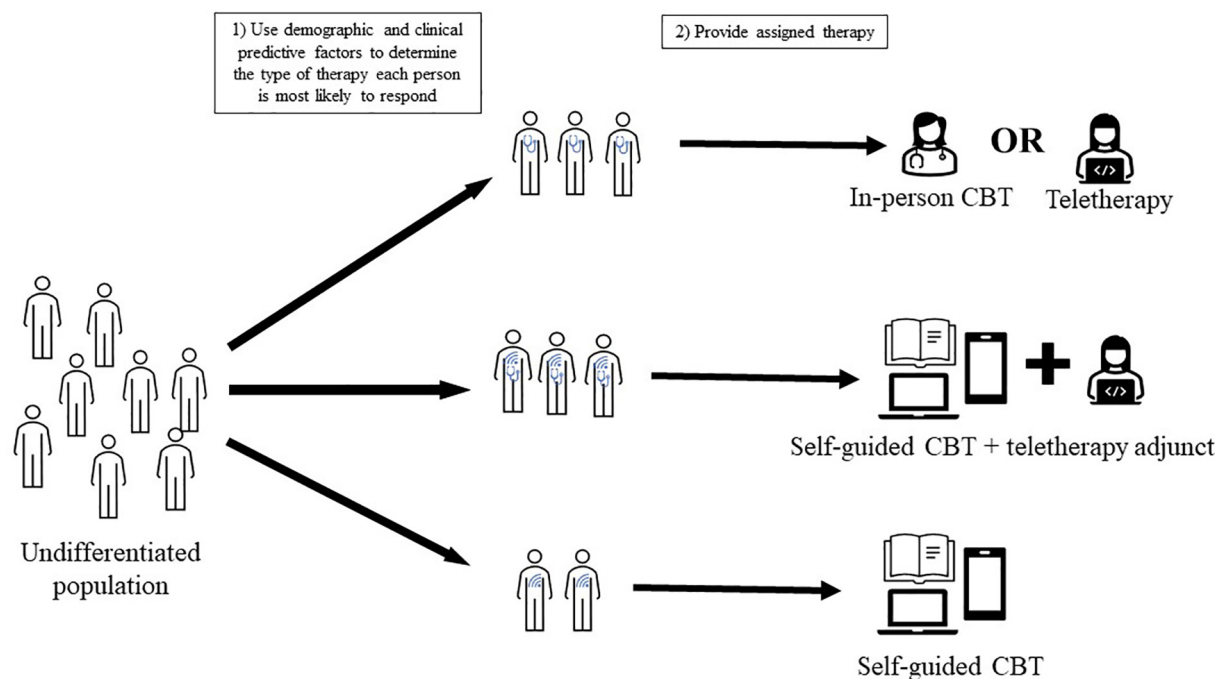


Fig. 3. Demographics- and clinical-based approach to providing CBT for FND. Based on individualized features predictive of response, patients are allocated to the most effective arm with the least need for FND-trained CBT therapist. CBT = cognitive behavioral therapy; FND = functional neurological disorder.

for all patients while restricting the scarce resource of FND-trained CBT therapists to those in whom other treatments are ineffective. Future studies will need to determine whether such a model would lead to a longer delay in treatment for those needing in-person CBT compared to the current waitlist model. If a stepwise model leads to a longer delay in treatment, worse outcomes are also possible since prognosis of FND is often correlated with symptom duration.

7.2. A demographics- and/or clinical-based approach to treatment

Another way to ensure that FND-trained CBT therapists are used for those in whom other treatments are ineffective is to identify which demographic and/or clinical characteristics make self-guided CBT beneficial for some people but not others (Fig. 3). This would allow clinicians to place participants in the treatment intervention predicted to be most effective for them, restricting CBT-trained therapists' time only to those for whom remote delivery would be futile. Future studies will be needed to determine which factors make different interventions beneficial or inadequate for different people.

8. Conclusions

There are four forms of remote delivery of CBT for FND, namely workbooks, iCBT, app-based CBT, and teletherapy, each with its own unique promises and challenges. Few studies have been conducted on each of these forms of CBT for FND. However, remote delivery of CBT has been studied extensively in psychiatric disorders. Given that psychiatric disorders and FND are often comorbid, share overlapping neurobiological features, and tend to respond to CBT, lessons learned from psychiatric studies can be reasonably applied to FND. Benefits of self-guided CBT (i.e., workbooks, iCBT, and app-based CBT) include the lower resource intensiveness, the lack of a need for a human therapist, and their promise to fill a shortage gap of FND-trained CBT therapists as most, if not all, of the therapeutic content is self-guided and adapted to FND patients. Workbooks have been shown to be effective in FND when compared to controls, but noninferiority trials are needed to determine whether they are as effective as in person or teletherapy. iCBT and app-based CBT appear to be effective for psychiatric disorders, but further studies should determine their effectiveness in FND. One

trial has shown teletherapy to be effective in functional seizures but replication studies in this and other FND subgroups would be desirable. Lastly, it will be important to understand which aspects of the established remote delivery of CBT for psychiatric disorders must be modified to suit the phenotypic variability of FND patients.

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Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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