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Functional communication disorders: a systematic review of interventions to improve outcomes in adults

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ABSTRACT

Purpose: To determine what evidence there is for the effectiveness/efficacy of interventions to improve outcomes in adults with Functional Communication Disorders (FCDs).

Materials and Methods: Seven electronic databases were searched *via* two platforms. The review was performed according to PRISMA guidelines. Population comprised adults with any specific communication diagnosis with a functional aetiology. We included studies of any type of behavioural intervention which targeted FCD, with any comparator. All outcomes related to communication were included. There were no restrictions on year or language of publication.

Results: Seven studies were included in the review. Studies used different interventions and outcome measures thus meta-analysis was not performed. Six studies described interventions for functional voice disorders and one for functional stuttering. Interventions tended to include elements of patient education, standard voice or speech therapy, and a psychological support component. Study quality was generally poor.

Conclusion: There is a lack of high-quality research to guide clinicians on evidence-based interventions for the full range of FCDs. There were some common themes within the interventions offered, but the overall poor quality of studies makes it difficult to draw conclusions on the effectiveness and efficacy of these interventions.

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KEYWORDS

Functional neurological disorder; speech and language therapy; communication disorders; functional voice disorders; functional stuttering; systematic review



> IMPLICATIONS FOR REHABILITATION


- There is a lack of high-quality intervention studies on the full range of Functional Communication Disorder (FCD) presentations.
- The strongest evidence base is for Functional Voice Disorders.
- Generalisable evidence regarding the effectiveness and efficacy of Speech and Language Therapy interventions for FCDs is limited.
- Literature recommends that intervention for FCDs should include patient education, techniques used in standard speech or voice therapy, and psychological support.

Introduction

Functional Communication Disorders (FCDs) previously referred to as psychogenic conversion reaction, are specific functional communication symptoms such as dysphonia, dysfluency, language and articulation disorders that can occur as a result of Functional Neurological Disorder (FND). In FND, there is a problem with the 'functioning' of movements, sensation or cognitive processes that is not a result of disease, damage, or structural abnormality [1]. Functional disorders can occur in isolation or alongside other neurological and psychiatric conditions [2,3]. FND is diagnosed on the basis of positive clinical features of internal inconsistency, and not by exclusion of structural damage or disease [4]. Underlying mechanisms and complexity of FND has changed significantly over the last 150 years. It is beyond the scope of this paper to discuss this in detail and the reader is referred to Stone and colleagues for further discussion [5]. Instead, our focus is specifically on treatment approaches for FCDs [4].

Speech and Language Therapists' (SLTs) expertise is long-established in the management of developmental and acquired communication disorders secondary to neurological disease and injury. Yet management of FCDs is not widely established. For example, in the UK, the Royal College of Speech and Language Therapists (RCSLT) has not published specific guidance on the management of FCDs. FND assessment, diagnosis and management is recommended in the context of an interdisciplinary team [6] with access to a range of services into which people can be referred depending on need and complexity [7]. In relation to FCDs, though different professionals are involved in their management, e.g., psychologists may lead on addressing psychogenic components, SLTs have specialist knowledge of communication and are therefore the key clinicians to support diagnosis and management in those with functional symptoms [8]. This is pertinent because FCDs are not an uncommon presentation in SLT clinics. FCDs may affect any aspect of communication, such as, voice, speech, or language; and can occur in isolation or concurrently with a combination of other FND symptoms [4]. Though

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prevalence and incidence data for overall FCDs is not known, there is evidence for specific presentations. Prevalence of voice disorders range from 3–17%, but anecdotally the prevalence of *functional* voice disorders (FVDs) is thought to be higher than this [9]. Acquired psychogenic/nonorganic speech disorders were found to present in 3% of referrals in a large 3-year study in the US [10]. Overall, FCD referrals are presenting to SLTs and the lack of evidence-based management and intervention guidance remains a challenge.

In terms of existing evidence, two reviews have addressed FCDs. Barnett and colleagues' scoping review (2019) aimed to determine what was known about speech and language symptoms in patients with FND [5]. As a result, they took a broad approach to identify papers that made mention of any speech and language symptoms in FND. They concluded that research in this field is typically centred around FVDs and functional stuttering though other symptoms including, speech and language symptoms were also reported, though less frequently [8]. The paper did not specifically report on interventions and their effectiveness, but they did include four papers (6%) that reported SLT input or involvement (including within the context of MDT interventions). A systematic review was conducted by Ruotsalainen and colleagues in 2008 on interventions for functional dysphonia as well as interventions to prevent voice disorders [11]. They found that a combination of direct and indirect therapy was effective at improving vocal performance. However, an issue in this paper, which will be addressed further in the discussion, is the use of terminology for FVDs. FVD describes a loss of voluntary control of phonation in the absence of a structural or neurological explanation [9]. Yet, the Ruotsalainen study used the term 'functional dysphonia' defined as "impaired voice [...] in the absence of organic lesions" (p. 557), but this definition included voice symptoms related to vocal misuse and strain (muscle tension dysphonia, MTD), which goes beyond the present focus on FVDs. Only one of the included studies described an intervention for FVD, which used a combination of 'classical therapy' with relaxation for "39 patients suffering from psychosomatic or conversive voice problems" (p. 563) [12].

SLTs feel that management of FCDs is part of their role, though they report lacking the skills, knowledge, and resources to provide optimal care [13]. The evidence base is reported to be lacking and variable across different FCDs: with some evidence for the treatment of FVD but relatively little on other FCDs. This has in part been addressed by the publication of international consensus recommendations for SLT [4]. Here a panel of international experts were asked to provide their recommendations for the management of a wide range of functional communication and swallowing disorders, which resulted in the most comprehensive recommendations for effective management of the full range of FCDs. It is acknowledged, however, that due to the lack of evidence-based interventions, recommendations were based largely on the lowest hierarchy of evidence: expert advice and opinion.

The aim of this review is to identify intervention studies conducted which describe interventions specifically targeting FCDs. There are no previous systematic reviews of interventions for the full range of FCDs. By combining studies on a range of FCDs (voice, speech, and language), intervention approaches can be contrasted and compared in this review, to determine whether there are similarities or differences depending on the FCD type. This can inform future intervention approaches. This review will address the research question: What is the evidence for effectiveness/efficacy of interventions to improve outcomes in adults with FCDs?

Method

The reporting of this review follows the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines

[14]. The systematic review protocol is registered on PROSPERO (17/04/2023 ref: CRD42023417186).

Eligibility criteria

This review sought intervention studies in published, peer reviewed journals, where the full text was available. There was no restriction on year of publication or language of publication. Foreign language papers were translated to English using Google Translate prior to screening. Though an older paper (2013) had highlighted some accuracy issues, especially in some languages (Chinese) in using Google Translate [15] a more recent (2019) paper has found it to be more accurate than previously reported [90% (range, 85% to 97%)] and to be a viable tool for extracting data for systematic reviews [16]. Overall the benefits of including non-English articles were deemed sufficient to justify the concerns raised in the literature.

The population included adults with any specific communication diagnosis with a functional aetiology which includes FVDs, functional stuttering (adult-onset), functional articulation symptoms, functional language symptoms; as described by Baker [4]. One specific functional communication disorder diagnosis was excluded: 'Foreign Accent Syndrome'. This was because there is research specifically focused on this condition, and it was deemed that this review would not add significantly to what is already known. Additionally, the diagnosis of 'muscle tension dysphonia' (MTD) was excluded. FVDs have two main subcategories: MTD and psychogenic voice disorders. MTD occurs due to dysregulated vocal behaviours which can be resolved through behavioural change and vocal exercises. Psychogenic voice disorders manifest due to a loss of volitional control of phonation, with observable symptom incongruity and reversibility. The latter being the focus of this review. See Baker (2016) for further discussion of the differentiation between these diagnoses [9]. Finally, only adult-onset stuttering was included, which is consistent with the diagnostic elements described in the consensus recommendations [4].

To capture all relevant research studies there were minimal restrictions on the intervention, comparator, and outcome. Any behavioural intervention which targeted the FCD was included, interventions could be led by any suitably trained professional, of any type and format and carried out in any setting. The only exclusion criteria were non-behavioural interventions e.g., surgery or medication. The comparator could be over time (with a minimum of three timepoints, e.g., double baseline where no change is expected and then post-intervention), group or task. Finally, any outcome related to communication (voice, speech, or language) was included.

Information sources

Searches were carried out on 16/05/2023 via two platform: Ovid, where Embase, Ovid Emcare and Cochrane Central Register of Controlled Trials (CENTRAL) were searched; and EBSCOhost, where APA PsycINFO, CINAHL Ultimate, Communication Source, MEDLINE complete were searched. All databases were searched from conception to present day. Subject index searches were carried out on databases with this function, and title and abstract keyword searches were conducted on all databases (see Table 1 for example of search strategy – Ovid Emcare). References and citations from articles included from the first round of screening were also searched using Scopus and Web of Science.

Table 1. Ovid Emcare search strategy (Ovid database).

Concept: Functional Neurological Disorder diagnosis		
#1	Subject heading [sh.]	Conversion disorder/ Somatoform disorder/ Psychosomatic disorder/ Functional disease/ Medically unexplained symptom/
#2	Title, abstract [ti.] [ab.]	(Functional neurological disorder* OR FND OR functional neurological symptom* OR non-organic OR non organic OR conversion disorder OR psychogenic OR psychosomatic OR hysterical conversion OR hysteria OR somatic symptom disorder OR somatoform OR somati?ation OR Medically Unexplained Symptoms OR psychomotor disorder* OR ganser syndrome OR dissociative disorder*)
#3	#1 OR #2	
Concept: Communication disorder diagnosis		
#4	Subject heading [sh.]	Communication disorder/ Language disability/ Agraphia/ Speech disorder/ Aphasia/ Anomia/ Stuttering/ Fluency disorder/ Dyslexia/ Dysgraphia/ Voice disorder/ Voice disorders/
#5	Title, abstract [ti.] [ab.]	(Language disorder OR *phasia OR word finding difficult* OR speech disorder OR slurred OR dysarthria* OR articulation OR voice OR stutter* OR stammer* OR dysfluency OR dysphonia OR laryngeal OR prosody OR phonat* OR communicat*OR mutism OR speech arrest)
#6	#4 OR #5	
Concept: Intervention (behavioural)		
#7	Subject heading [sh.]	Language therapy/ Rehabilitation/ Speech and language rehabilitation/ Speech rehabilitation/ Group therapy/ intervention study/ Speech language pathologist/ (Therap* OR treat* OR strateg* OR rehabilitat* OR program* OR speech therap* OR intervent* OR train* OR language therap* OR stimulat* OR speech patholog* OR language patholog* OR manag* OR education OR coaching OR counsel?ing)
#8	Title, abstract [ti.] [ab.]	
#9	#7 OR #8	
#10	#3 AND #6 AND #9	

Search strategy

To take a focused approach on interventions for FCDs: three key terms were used for searches: 1) FND diagnosis, 2) communication disorder diagnosis, 3) intervention. Search terms were identified through reading key texts and those used in a previous scoping review by Barnett and colleagues [8]. Text frequency analysis tools (TerMine and Yale MeSH Analyzer) [17,18] were used using primary papers on the subject to ensure all key words were identified. See Appendix 1 for all search terms.

As this is an under researched area, and no prior systematic reviews are known on this topic, there were no limits applied to the search.

Selection process

Search results were exported to Rayyan [19] and duplicates removed. Screening was conducted independently by two reviewers: KM screened all articles, LR screened 30%. Initially all titles and abstracts were screened to remove any articles that clearly did not meet the eligibility criteria. Included articles then moved to the full-text stage.

A range of avenues were used to obtain full texts of included articles: articles were searched for via the University's library services; online platforms such as Google Scholar, ResearchGate, journal websites; the British Library; authors were emailed where

possible; and as a final resort, papers were requested via interlibrary loans for a small fee. Despite the lengths taken four articles could not be located. Foreign language articles were translated using Google Translate. Quality of translation varied, but both reviewers felt they were sufficient to confidently screen papers. A screening hierarchy was developed with support from senior researchers KH and NB, and reasons for exclusion were documented at the full text stage. There were few disagreements (<15%), KM and LR reviewed conflicting papers jointly until agreement was reached. Disagreements were easily resolved so input from a third reviewer was not required.

Included articles and some excluded articles felt to be relevant but failing to meet inclusion criteria were used for citation and reference searching using Scopus, with results checked on Web of Science (02/07/2023). This found a further 21 articles. These articles were then screened on Rayyan as described.

Data collection process

A data extraction form was developed. Data extraction was conducted by KM. To ensure consistency of data collection, LR independently performed data extraction on 3 out of the 7 papers (43%), which were selected using a random number generator. Percentage agreement on data items across the double-coded papers was 82%, any variations in data collected was checked and agreed with senior researchers KH and NB.

Data items

Data extracted included: Country, language of publication, declared conflicts, ethical approval, funding sources, study characteristics (study design, setting, aims), FCD diagnosis as described by the author(s), participant information (inclusion and exclusion criteria, number, age (range and mean), sex, ethnicity, and time since FCD diagnosis), intervention content, dosage (including intensity, frequency, and total hours, depending on how it was reported); comparator, targeted outcomes, timepoints of assessment, outcome measures used, and outcomes achieved.

Study risk of bias assessment

Risk of bias was rated according to study design: the Physiotherapy Evidence Database (PEDro) scale [20] was planned for randomised control trials (RCTs); the NIH study quality assessment tool [21] for before/after (pre-post) studies with no control group; and the JBI Critical Appraisal Checklist for case reports or case series [22].

Two reviewers, KM and LR, assessed each study's risk of bias independently. Findings were discussed and disagreements reviewed jointly until agreement was reached.

Effect measures

We planned to pool eligible group-level studies using similar outcomes into a meta-analysis using standardised mean differences. However, there was such heterogeneity in study designs, interventions evaluated, and outcome measures used, that we did not perform these analyses.

Synthesis methods

A narrative synthesis was carried out due to the heterogeneity of study designs and outcome measures used. The Synthesis Without Meta-analysis (SWiM) reporting guideline was followed [23].

Results

Study selection

The study selection process is illustrated in Figure 1. The initial search found 2,463 articles, with a further 21 found through reference and citation searching. 794 duplicates were removed, initial title and abstract screening removed a further 1,532 papers. 158 full texts were sourced for full text screening, although four papers could not be found. Searching for missing articles was deemed exhaustive following searches of the library services and Inter-Library loans, The British Library catalogue, and for one paper the Federal Institute for Drugs and Medical Devices in Germany was contacted but received no response. Reasons for exclusion at full text stage are detailed in Figure 1. Of the 154 papers assessed for eligibility, 147 were excluded resulting in a final set of seven articles, describing seven studies, included in the review.

Identifying whether the condition of focus was a FCD was made challenging by historical inconsistencies in terminology. This was particularly challenging in the voice disorder papers, where terminology usage has already been highlighted as an issue [9]. Non-specific terms such as 'benign dysphonia' or 'nonorganic dysphonia' were used without clear definition; or 'FVD' has been used interchangeably to describe muscle tension dysphonia (MTD) and psychogenic voice disorders (the latter being the focus of this review). Some papers did not specify the aetiology of their

participants, and as it was not possible to determine if the study was assessing the target population these papers were excluded.

Translation of non-English papers

Twenty-eight full text articles were translated to English by the lead author using Google Translate. One foreign language study was deemed to meet the inclusion criteria [24].

Study characteristics

Study designs used by the seven studies included: one pilot RCT with intervention versus a control group assessed at three timepoints [25] and one a pre-post study with no control group, where timepoints were used as a comparator: a pre-intervention, post-intervention, and two follow-up data collection points [26]. There were three case series: of these, two used group comparators [27,28]; one used time comparators: a pre-intervention, post-intervention, and one follow-up data collection timepoint [29]. Two studies were single case studies which used a pre-intervention, post-intervention, and one follow up data collection timepoint [24,30]. Six of the seven studies were on FVDs [24–29] and one on functional stuttering [30]. Three studies specified where the intervention was conducted: one in an outpatient voice clinic [25], one in an outpatient voice clinic or inpatient setting [28], and the last study was in an inpatient rehabilitation unit [30]. The other studies ($n=4$) did not give precise detail on the intervention setting, but were indicative of an outpatient setting [24,26,27,29]. The studies were conducted in the United Kingdom [25], Switzerland [27], Yugoslavia [28], Japan [24], USA [26,30] and Turkey [29]. Study characteristics are detailed in Table 2.

Participant characteristics

The studies reported on 236 participants in total, 235 with FVD and one (in a single case study) with functional stuttering [30]; all participants received SLT intervention, one study had an experimental group which received Cognitive Behavioural Therapy (CBT) in addition to SLT ($n=37$) [25]. Six of the seven studies reported on participant age. Across studies where the mean age of participants was available, the weighted mean was 43.7. Where reported, range and medians are given in Table 2. Participants in included studies were predominantly female (81.8%), except for the male single case study [30]. Time since FCD diagnosis prior to intervention was not consistently reported, where it was reported the average was 104 days (range from 1 day to 4 years).

Intervention characteristics

Intervention description

All papers provided a description of the intervention provided, one included a treatment protocol [26] and another referenced where the protocol could be found [25]. However, description specifics were variable across the papers, most did not provide sufficient information about the content of the intervention, dosage, or rationale to a replicable standard. One paper reported on modifications and fidelity [25], but the others did not.

Intervention content

Interventions offered across the studies varied, but there were reoccurring treatment approaches across studies. All but one study

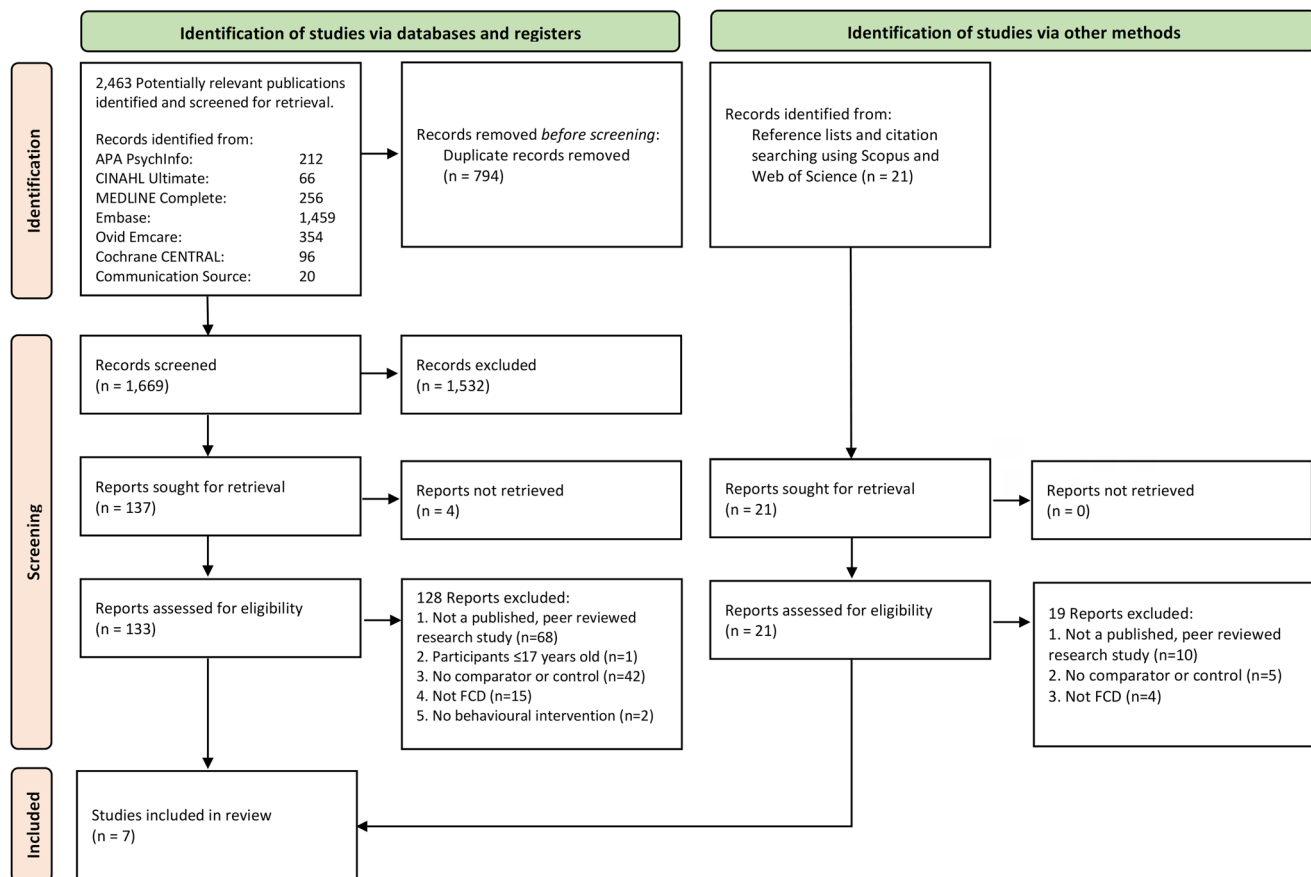


Figure 1. Identification of studies. PRISMA 2020 flow diagram adapted from page et al. [14].

[24] included education as a key part of intervention. Content of education sessions was described as: discussion and education about normal voice production, the aetiology of functional disorders, and the impact of emotion on communication. Most studies described a psychological component to intervention: such as CBT [25], psychotherapy [27], or exploring the psychogenic factors contributing to the disorder indicative of a counselling approach [26,28–30]. Most studies described using ‘standard’ voice or speech therapy, such as vocal hygiene advice [24,25], tube phonation [24,29]; ‘voice exercises’ were reported by Kolbrunner et al. (2010) but no further details were given [27], Umeda et al. (2017) specified using the pushing method and humming [24]; and Yam et al. (2016) gave the following speech therapy exercises: prolongation, melodic lines and easy onset [30]. Three studies reported using breathing exercises for FVDs [25,27,28]. A further three studies reported using relaxation as part of the intervention [27,28,30]. Two of the FVD papers reported using acupressure or ‘manual laryngeal musculoskeletal tension reduction’ to elicit a more normal voice [24,26]. A further two reported encouraging the patient to use their new skills in their normal social environment [26,29]. Dosage varied across the studies and was not consistently reported. For studies which gave clear dosage information: sessions were carried out daily to fortnightly, quantity of sessions were between one and ten, and the intervention period ranged from 50min to 3 months.

Intervention delivery

All intervention and education sessions were delivered face-to-face in individual sessions, and one study [29] involved family in education sessions. Discussion and explanation were the primary

modalities used, details about these varied between studies. Two studies prescribed homework exercises [25,29]. The intervention was reported to be carried out by a SLT [25], Speech and Language Pathologist [30], Speech Pathologist [26], Phoniatrician [29], and jointly by a Phoniatrician and Vocal Therapist in another [28]. Two studies did not specify the profession of the clinicians who delivered the intervention [24,27]. As the studies were conducted in a range of different countries, professional titles, roles, and qualifications are likely to vary and may not be directly comparable.

Outcome measures

The outcome measures used and how they were reported varied across studies. There were some similarities: all studies measured perceptual voice or speech quality as an outcome. Three studies used the Grade, Roughness, Breathiness, Asthenia and Strain (GRBAS) scale [31], two papers used different versions of the Voice Handicap Index [32,33]. Two studies used ‘time to achieve normal voice’ as an outcome but did not report how this was determined [27,28]. Three of the studies reported on the psychological status of patients as an outcome measure, two used validated psychological measures [25,30], though the other used a subjective assessment of the success of psychotherapy [27]. Two studies used instrumental assessment (laryngoscopy) [24,29]. Finally, three studies used patient self-reported incidence of relapse or reoccurrence of the FCD as an outcome [26–28].

Treatment maintenance

All studies reported long-term follow up of participants, though the time periods and method of follow-up varied between studies.

Table 2. Study and participant characteristics.

Reference	Deary et al. [25]	Kolbrunner et al. [27]	Milutinović [28]	Umeda et al. [24]	Yam et al. [30]	Başer and Denizoglu, [29]	Roy et al. [26]
Title	A psychosocial intervention for the management of functional dysphonia: complex intervention development and pilot randomised trial	Psychogenic aphonia: no fixation even after a lengthy period of aphonia.	Results of vocal therapy for phononeurosis: Behaviour approach.	Voice therapy for a case of functional dysphonia with suspected psychogenic factor	Interdisciplinary rehabilitation approach for functional neurological symptom (conversion) disorder: A case study	The Efficiency of DoctorVox Voice Therapy Technique in Conversion Dysphonia / Aphonia	Manual Circumlingual Therapy for Functional Dysphonia: An Evaluation of Short- and Long-Term Treatment Outcomes
Journal	Pilot and Feasibility Studies	Swiss medical weekly	Folia phoniatrica	Japan Journal of Logopaedics and Phoniatrics	Rehabilitation Psychology	The Journal of Laryngology & Otology	Journal of Voice
Location of study	United Kingdom	Switzerland	Yugoslavia	Japan	USA	Turkey	USA
Declared conflicts	None	None	Not stated	None	Not stated	The first author: none. The second author has a conflict of interest: He is the founder, inventor, and patent owner of the DVT method and devices.	Not stated
Ethical approval	Yes	Not reported	Not reported	Not reported	Not reported	Yes	Not reported
Funding source	Medical Research Council (MRC) Research fellowship to the first author (UK)	Not reported	Not reported	Not reported	Not reported	No financial disclosure	National Centre for Voice and Speech
Language of publication	English	English	English	Japanese	English	English	English
Functional communication diagnosis	Functional voice disorder	Functional voice disorder	Functional voice disorder	Functional voice disorder	Functional stuttering	Functional voice disorder	Functional voice disorder
Study design	Pilot single-centre randomised controlled trial	Single-centre retrospective case series	Single-centre retrospective case series	Single case study	Single case study	Single-centre retrospective case series	Single-centre pre-post study with no control group
Study setting	Outpatient voice clinic	Not reported	Outpatient voice clinic, inpatient treatment for patients with slow reaction to therapy	Not reported	Inpatient rehabilitation unit	Not reported	Not reported
Participants N=	Total: 74 patients. Control group (SLT only): $n = 37$; Experimental group (CBT+SLT): $n = 37$	Total: 22 patients. By group: A. $n = 6$, B. $n = 13$, C. $n = 2$, D. $n = 1$	Total: 99 patients. By diagnosis: Aphonia psychogeneses $n = 61$, Dysphonia psychogeneses $n = 16$, mutatio falsa $n = 22$ [no figures provided]	1	1	14	25
Age = mean (SD), median (IQR)	Control group: 45.8 (13.5), Experimental group: 43.5 (16.3)	A. 46.7 (11.1), 44.5 (18.25) B. 37.5 (16.9), 38 (25) C. 49 (16.9), 49 D. 20	Description: The distribution according to age in all show that the condition appears in all age-groups, from adolescence to 59 years, with peaks in adolescence and the second half of the fourth decade. $M = 28$, $F = 71$	"30s"	45	48.93 (18.73) (range: 18–72)	40.9 (13.03)
Sex Female: F Male: M	Control group: $M = 6$ (16.2%), $F = 31$ (83.8%). Experimental group: $M = 3$ (8.1%), $F = 34$ (91.9%)	Total: $F = 19$, $M = 3$. By group: A) $F = 6$, B) $F = 10$, $M = 3$, C) $F = 2$, D) $F = 1$	$M = 28$, $F = 71$	Female	Male	$M = 2$ (14.3%), $F = 12$ (85.7%)	$F = 25$ (100%)
Ethnicity	Not reported	Not reported	Not reported	Not reported	White	Not reported	Not reported
Time since dx (and central tendency measures) in days	Not reported	Fourteen patients (64%) had previously experienced aphonic phases (on average 6 phases) lasting from a few days to up to 3 years. The time between the beginning of the current aphonia and the consultation at our department was, on average, 20 days (SD 18 days). One exceptional case was excluded from this calculation, whose aphonia had already lasted for more than one year by the day of consultation.	1 - 243 days (average 50 days)	Not stated. Reports previous aphonia in her 20s.	106 days	4-365 days (mean(SD)= 77.07 (101.22) days, median= 37.5 days)	5 - 1461 days mean 268 (371) days

Follow-up ranged from three months [24], six months [25,30], the case series reported follow up for at least two [28] and three years [27], two studies had variable follow up times of 16.5 months (± 11.4 months) [26] and 17 months (± 9.6 months) [29]. All studies repeated outcome measures for long term follow up, apart from two studies that reported on reoccurrence of symptoms but did not report how this was determined [27,28]. All studies reported significant treatment maintenance at long-term follow up, though the specificity of reporting varied across studies.

Table 3 provides further information on interventions.

Risk of bias across studies

Risk of bias assessment was carried out on all studies, assessment measures were used according to study design, scores are detailed in Appendix 2.

Risk of bias across the studies was moderate to high. All but two studies achieved <50% of the required quality criteria, with the exception of one single case study (score 6/8) [30] and one case series (score 7/10) [29]. Reoccurring issues were around incomplete reporting of participant demographics: all studies reported on participants' sex and age, though one reported age vaguely as "in her 30s." [24] Only one study included the participant's ethnicity with a detailed case description [30], another gave employment status and educational background but not ethnicity [29]. Similarly, there was a lack of specifics about the intervention provided, only two studies provided information about the treatment protocol [25,26]. Intervention terms such as 'voice therapy' were often used without explicit description of what this involved, all studies reported using 1-2 clinicians to conduct the intervention, however there was little information about what checks were made to ensure consistency and replicability of the clinician's intervention. This impacts the studies replicability and external validity.

Many of the studies failed to report specifics about inclusion criteria, recruitment, and inclusion of participants. Three studies reported inclusion criteria [25,26,29]. Yet, none of the three-case series state whether there was consecutive and complete inclusion of patients [27–29]. All studies failed to compare the participants against the wider population and comment whether participants were representative, with implications of the studies' external and ecological validity. Furthermore, a lack of clear information about the recruitment and inclusion raises concerns around whether selection bias occurred.

The RCT²⁵ did not blind participants, therapists, or assessors; though it is acknowledged that with behavioural interventions it is not practical or feasible to blind participants or therapists to the intervention they are receiving or providing. However, the researchers could have attempted to blind the assessor by using an external assessor which is deemed to be an oversight.

Finally, there is a lack of statistical comparisons in many of the studies. The single case studies, case series and RCT could have conducted between group or within group comparisons, but this was only reported in one study [29]. Though the RCT was a feasibility study which did not aim to demonstrate the effectiveness of the intervention, it seems like a lost opportunity in an area of scarce research.

Overall, there were issues with all studies with implications to replicability. All studies used perceptual ratings as part of their outcome measures, though only one described the measures taken to ensure reliability [26]. Furthermore, the outcome measures used have been designed to be used with organic communication disorders, so their use with FCDs may not be validated.

Data synthesis

Meta-analysis could not be carried out due to the heterogeneity of the studies, different interventions and outcome measures used. A narrative synthesis approach was used to describe the outcomes, according to the type of FCD. Table 3 details results from individual studies. Due to the small number of studies (7) all were included in data synthesis despite quality assessment findings.

Functional voice disorders – intervention effectiveness and efficacy

There were six studies that presented evidence for interventions for FVDs. Three of these were case series, one single case study, one pre-post study with no control group, and an RCT. Though an RCT is considered the highest level of evidence, the Deary et al. (2018) paper was a feasibility study which primarily sought to describe the process of developing a CBT intervention, therefore its focus was not specifically to test the effectiveness or efficacy of SLT intervention for FVDs [25]. This accounts for some of the issues in synthesising the findings into the review. The pre-post study provided detailed information about the intervention and outcome measures used but lacked detail about participant recruitment which raises issues around the potential for selection bias. Two of the case series provided very limited information regarding the intervention offered and both papers sought 'recovery of normal voice' as the outcome, though gave no information about how this determination was gained [27,28]. The remaining case series [29] provided better reporting of intervention and outcome measurement, yet still there are potential issues with selection bias as there is no transparent reporting of the inclusion of participants. Finally, the remaining single case study [24] provided adequate information about the intervention offered but insufficient detail about the patient demographics, clinical condition, and conclusions, to determine the implications of the study's findings.

The difference in study design, aims, and outcome measurement present challenges for data synthesis, and the findings of the study need to be considered rationally due to the methodological and quality issues raised. Nevertheless, the intervention effectiveness and efficacy will now be summarised.

There is heterogeneity in the studies' interventions, but there are some similarities between approaches. All, but one [24], describe education (of normal voice production and the impact of functional disorder) as a component of intervention. All papers describe using voice therapy techniques as part of the intervention. Voice treatments can be described as either physiologic (i.e., where therapy aims to change the physiology of the voice mechanism) or symptomatic (i.e., using techniques to modify deviant vocal behaviours) [34]. Five papers describe the use of physiologic voice therapy techniques: such as Manual Circumlaryngeal Techniques [24,26], Resonant Voice Exercises [24], breathing exercises [25,27,28], and the pushing method [24]. Five papers also report using symptomatic voice therapy techniques: relaxation [27,28], Semi-Occluded Vocal Tract Exercises [24,29], and biofeedback [28]. Four papers also report using psychological approaches as part of intervention, two papers describe specific methods: CBT²⁵ and psychotherapy [27]; the other two papers describe using counselling skills within sessions to explore the psychogenic factors [26,28]. The inconsistency between the approaches taken makes synthesising outcomes very challenging, as study findings cannot be directly compared.

All studies reported improved voice post-intervention. However, this was generally determined using clinician's perceptual evaluations. Three studies [24,25,29] used the GRBAS rating scale, which

Table 3. Intervention and outcomes table.

Reference	Aim	Intervention description	Targeted outcomes and measures used	Results mean (SD), median (IQR)
Deary et al. [25]	To develop and pilot a new psychosocial intervention aimed at improving both voice and well-being by comparing patients receiving SJT to patients receiving SJT+CBT	Control group (Voice therapy only): - Vocal hygiene education - Reducing vocal abuse - Breath work - Voice resonance and flexibility exercises Experimental group (Voice therapy plus CBT): Indl. assessment, formulation, treatment Amount: Six to eight approximately 1-hour sessions delivered weekly to fortnightly.	1. Feasibility and acceptability objectives: Recruitment and retention rates (patient acceptability of intervention and trial procedures), through observation of the training process (therapist acceptability) and through monitoring the process and content of the CBT participants' treatment, in clinical supervision, case recordings, case notes and case summaries (fidelity, therapist and patient acceptability and feasibility, clinical utility). 2. Voice: GRBAS and VPO. 3. Psychological distress and anxiety: GHQ-28, HAD.	1. Feasibility and acceptability outcomes Prior to randomisation: 175 referrals – 7 DNA = 168 remaining (96%). 64 (38%) did not meet inclusion criteria: rate of eligibility: 62% (104/168). Did not consent: 30 (29%); 30/104). 74 patients (71% of those eligible) were recruited to the trial. Control group follow up attrition: 13.5%. Experimental group follow up attrition: 24.3%. CBT case formulation feasibility (32/27, 86%). 2. Voice outcomes Control group: GRBAS baseline: 1.6(0.7), post-intervention not reported. VPO scores went from 29.08 (6.36) to 17.68 (5.01). Experimental group: GRBAS 1.8(0.8), post-intervention not reported. VPO scores went from 35.28(11.42) to 16.77(6.53). 3. Psychological distress and anxiety Control group: Mean GHQ-28 scores went from 29.41 (14.67) to 14 (5.61); mean HAD depression scores went from 5.16 (4.12) to 2.12 (2.83). Experimental group: Mean GHQ-28 scores went from 30.22 (15.67) to 16.73 (11.04); mean HAD depression scores went from 7.08 (4.5) to 2.69 (2.83). Group A: Aphonia resolved before initial consultation, no longer aphonic but remained dysphonic ($n=6$) 1. Voice recovered by time of initial consultation. 2. yes $n=3$, no $n=2$, partially $n=1$. 3. 3pts relapsed with 2-7 phases of aphonia, average relapse free 2.7 years (SD 2 years). Group B: Aphonic at initial consultation, but recovered a speaking voice within a year ($n=13$) 1. mean: 11.5 weeks (9.7), median: 9, (14.15); range: 33.8. 2. yes $n=6$, no $n=2$, partially $n=4$, missing data $n=1$. 3. 1pts relapsed, average release free 3.5 years. Group C Aphonic at initial consultation, voice recovery took more than a year ($n=2$) 1. mean: 141.5 (58.7), median: 141.5, range: 83. 2. no $n=1$, partially $n=1$. 3. Release free 3.3 and 4.8 years. Group D: No recovery of voice ($n=1$) 1. Remained aphonic despite therapy. 2. unknown $n=1$. 3. N/A.
Kolbrunner et al. [27]	To report outcomes of psychotherapeutic intervention and voice therapy on patients with psychogenic aphonia, by grouping patients into four groups (A-D) by aphonia presentation (see results column for group descriptions).	Treatment: - FVD education with videolaryngoscopic evidence - Short-term dynamic psychotherapy - Voice therapy Indl. sensory awareness, relaxation, breathing, and voice exercises. Amount: Mean therapy hours per participant: 11.8 (range 0-65). Further detail not given.	1. Recovery of voice: Time until voice recovered from initial consultation (in weeks). 2. Success of psychotherapy: Subjective assessment of success of psychotherapy: "yes"= psychosomatic context recognised and psychosocial changes initiated; "partial"= psychosomatic context recognised; "no"=psychosomatic context not recognised; or unknown. 3. Relapse: number of new aphonia phases; period of no relapse in years.	1. Time to recover normal voice Aphonia psychogenes 0-1 week $n=51$, 1-2 weeks $n=4$, >2 weeks $n=6$. Dysphonia psychogenes: 0-1 week $n=14$, 1-2 weeks $n=2$. Mutatio falsa: 0-1 week $n=22$. 2. Recurrences Aphonia psychogenes $n=3$ (3.03%); dysphonia psychogenes $n=1$ (1.01%); mutatio falsa $n=0$. Pre-therapy 1. Laryngeal findings: Laryngeal endoscopic findings showed glottic insufficiency during vocalisation and mild supraglottic hypertension. Vocal cord paralysis is not recognised. 2. MPT: 11.0 s, expiratory flow rate: 103 ml/sec. 3. G2R0B2A150, vocal range: 261 Hz speech (196 Hz – 659 Hz); VHI: 65 points (P: 30 points, F: 24 points, E: 11 points). Post-therapy 1. Laryngeal findings: Glottic insufficiency during vocalisation improved, but glottal insufficiency in the posterior part remained. Hypertension in the supraglottis remained. 2. MPT: 32.0 s, expiratory flow rate: 130 ml/sec. 3. G1R0B1A050, vocal range: 207 Hz speech (174 Hz – 783 Hz); VHI: 37 points (P: 20 points, F: 14 points, E: 3 points). 3 months after 1. Laryngeal findings: Posterior glottic closure insufficiency and hypertension in the supraglottis remained, but stroboscopy findings showed a prolonged closure period compared to immediately after completion. 2. MPT: 27.0 s, expiratory flow rate: 140 ml/sec. 3. G1R0B1A050, vocal range: 207 Hz speech (164 Hz – 698 Hz); VHI: 24 points (P: 15 points, F: 7 points, E: 2 points).
Milutinović [28]	To describe a voice therapy intervention and demonstrate its effectiveness for three types of phononeuroses (aphonia psychogenes, dysphonia psychogenes, mutatio falsa) by reporting duration of treatment and incidence of recurrences.	Voice therapy treatment: - Facilitate correct voice production - Voice production education - FVD education - Biofeedback - Breathing exercises Counselling - Explore social factors contributing to FVD Amount: Outpatients offered daily sessions, inpatients 2-3 sessions per day. Further detail not given.	1. Recovery of normal voice: duration of treatment (in weeks) required to achieve normal voice. 2. Rates of recurrences of phononeuroses: recurrence reported from long-term follow up.	1. Time to recover normal voice Aphonia psychogenes 0-1 week $n=51$, 1-2 weeks $n=4$, >2 weeks $n=6$. Dysphonia psychogenes: 0-1 week $n=14$, 1-2 weeks $n=2$. Mutatio falsa: 0-1 week $n=22$. 2. Recurrences Aphonia psychogenes $n=3$ (3.03%); dysphonia psychogenes $n=1$ (1.01%); mutatio falsa $n=0$. Pre-therapy 1. Laryngeal findings: Laryngeal endoscopic findings showed glottic insufficiency during vocalisation and mild supraglottic hypertension. Vocal cord paralysis is not recognised. 2. MPT: 11.0 s, expiratory flow rate: 103 ml/sec. 3. G2R0B2A150, vocal range: 261 Hz speech (196 Hz – 659 Hz); VHI: 65 points (P: 30 points, F: 24 points, E: 11 points). Post-therapy 1. Laryngeal findings: Glottic insufficiency during vocalisation improved, but glottal insufficiency in the posterior part remained. Hypertension in the supraglottis remained. 2. MPT: 32.0 s, expiratory flow rate: 130 ml/sec. 3. G1R0B1A050, vocal range: 207 Hz speech (174 Hz – 783 Hz); VHI: 37 points (P: 20 points, F: 14 points, E: 3 points). 3 months after 1. Laryngeal findings: Posterior glottic closure insufficiency and hypertension in the supraglottis remained, but stroboscopy findings showed a prolonged closure period compared to immediately after completion. 2. MPT: 27.0 s, expiratory flow rate: 140 ml/sec. 3. G1R0B1A050, vocal range: 207 Hz speech (164 Hz – 698 Hz); VHI: 24 points (P: 15 points, F: 7 points, E: 2 points).
Umeda et al. [24]	To demonstrate effective voice therapy in a patient with psychogenic dysphonia	Voice therapy treatment: - Vocal hygiene education - Voice exercises incl. "pushing method to induce glottal closure," humming, and tube phonation - Acupressure to promote glottis closure i.e., manually compressing the lateral lamina of the thyroid cartilage. Amount: Six sessions delivered weekly to fortnightly over 3 months.	1. Laryngeal findings: visualised larynx (scoping). 2. Aerodynamic test: MPT, expiratory flow rate. 3. Voice evaluation: GRBAS, vocal range, VHI.	1. Laryngeal findings: Glottic insufficiency during vocalisation improved, but glottal insufficiency in the posterior part remained. Hypertension in the supraglottis remained. 2. MPT: 32.0 s, expiratory flow rate: 130 ml/sec. 3. G1R0B1A050, vocal range: 207 Hz speech (174 Hz – 783 Hz); VHI: 37 points (P: 20 points, F: 14 points, E: 3 points). 3 months after 1. Laryngeal findings: Posterior glottic closure insufficiency and hypertension in the supraglottis remained, but stroboscopy findings showed a prolonged closure period compared to immediately after completion. 2. MPT: 27.0 s, expiratory flow rate: 140 ml/sec. 3. G1R0B1A050, vocal range: 207 Hz speech (164 Hz – 698 Hz); VHI: 24 points (P: 15 points, F: 7 points, E: 2 points).

(Continued)

Table 3. Continued.

Reference	Aim	Intervention description	Targeted outcomes and measures used	Results mean (SD), median (IQR)
Yam et al. [30]	To present an FND case study treated with a coordinated interdisciplinary approach in a rehabilitation setting. To illustrate the feasibility and importance of treating FND in an interdisciplinary setting.	Treatment: - Speech and language therapy incl. language skills, stuttering, reduced perception and discrimination, learning and memory strategies, and attention training. - Stuttering therapy incl. sound prolongation, melodic speech, easy onset. Outpatient follow-up targeting visual organisation and attention. Amount: Cognitive rehabilitation groups three times a week. Further detail not given.	1. Neuro-psychological measures: a) Performance validity measures: RDS, TOMM, WMT. b) Motor/sensory: tasks during neurobehavioral interview. c) Visual-spatial: ROCF, JLO. d) Academic: WRAT4 (Math Computation, Word Recognition, Spelling, Sentence Comprehension). e) Attention: WALS-IV (Digit Span, Letter-Number Sequencing), WMS-IV. f) Speed of processing: TMT parts A & B, DVT, WALS-IV (Symbol Search, Coding). g) Language: D-KEFS (Verbal Fluency). h) Memory: CVLT-II, BWMTR. i) Executive function: WCST-64; D-KEFS (Design Fluency, Sorting). 2. Measures of emotional functioning: BAI; WHOQOL-BREF; MPAL-4; NSI-22; PHQ-9; PCL-C; SWLS. 3. Stutter severity: Functional assessment of stutter version. 1. Severity of vocal symptoms: VHI-10 - validated Turkish version. 2. Clinician's subjective assessment of voice: GRBAS. 3. Physical examination/objective assessment of larynx: video-laryngostroboscopy.	Admission assessment 1. Markedly impaired performances across most cognitive domains, except for preserved general verbal skills and basic attention. * 2. Mild anxiety (BAI = 10), mild depression (PHQ-9 = 10), and feeling slightly dissatisfied with life (SWLS average rating of 2.8), and a moderate level of neurobehavioral difficulties (NSI-22; Total score = 38). * 3. Severe stutter. Mid treatment 1. Markedly impaired performances on most cognitive domains. * 2. Clinically significant emotional distress, including mild depression and anxiety. * 3. Mr. A. was unable to speak more than one to two words without a severe stutter. Discharge assessment 1. Showing improvements on most cognitive measure. * 2. Mr. A endorsed relatively unchanged overall dissatisfaction with life. * 3. Speech was fluent, with only occasional stutter. *Specific scores for outcome measures used not provided, unless stated.
Bayer and Denizoglu [29]	To retrospectively evaluate the efficiency of DVTT in PD/PA patients and share the mid-long term results of the method.	Treatment: - Voice and FVD education - Counseling - Voice exercises using DVTT and oral mask (maskVOX*). Amount: Five daily 25-minute sessions, followed by five further 25-minute sessions twice a week to once a week. Telephone follow up 1-month and 6-months later.	1. VHI-10 mean scores: Pre: 30.91 (29.7), Post: 8.14 (3.82), Follow-up: 3.36 (1.78). Change in pre, post and follow up statistically significant: $p=0.001$; $p<0.05$ compared to pre-treatment, decrease post treatment was significant ($p=0.024$), as was decrease at follow-up ($p=0.001$). VHI-10 pre-, post-, and follow up was not statistically significant ($p>0.05$) according to the attack status at the time of referral (whether there were single or multiple attacks), whether or not they received psychiatric treatment, and employment status. The patient group with complaints lasting longer than one month had significantly greater amount of pre-treatment - posttreatment change and posttreatment - final follow-up change in VHI-10 measurements compared to patients with complaints lasting less than one month ($p=0.041$; $p=0.044$; $p<0.05$, respectively). 2. GRBAS evaluation (Pre-treatment: 9 (0.67), post-treatment: 0.78 (0.80), final follow-up: 0.57 (0.64), there was a statistically significant change between pre-post treatment and pre-treatment - final follow-up values ($p=0.001$; $p=0.05$). 3. Physical examination 21.4% (n3) of the cases were extremely dysphonic (it was observed that one patient phonated in a breathy falsetto register, and two patients had ventricular folds closed and that transglottic airflow was almost absent). Among the patients, 78.6% (n:11) were aphonic in which the mucosal vibration was not observed in the videolaryngoscopic examination of these patients due to the large glottic gap. Eight patients of the aphonic group were whispering, while the other three were completely silent, only moving their lips.	mean scores: Pre: 30.91 (29.7), Post: 8.14 (3.82), Follow-up: 3.36 (1.78). Change in pre, post and follow up statistically significant: $p=0.001$; $p<0.05$ compared to pre-treatment, decrease post treatment was significant ($p=0.024$), as was decrease at follow-up ($p=0.001$). VHI-10 pre-, post-, and follow up was not statistically significant ($p>0.05$) according to the attack status at the time of referral (whether there were single or multiple attacks), whether or not they received psychiatric treatment, and employment status. The patient group with complaints lasting longer than one month had significantly greater amount of pre-treatment - posttreatment change and posttreatment - final follow-up change in VHI-10 measurements compared to patients with complaints lasting less than one month ($p=0.041$; $p=0.044$; $p<0.05$, respectively). 2. GRBAS evaluation (Pre-treatment: 9 (0.67), post-treatment: 0.78 (0.80), final follow-up: 0.57 (0.64), there was a statistically significant change between pre-post treatment and pre-treatment - final follow-up values ($p=0.001$; $p=0.05$). 3. Physical examination 21.4% (n3) of the cases were extremely dysphonic (it was observed that one patient phonated in a breathy falsetto register, and two patients had ventricular folds closed and that transglottic airflow was almost absent). Among the patients, 78.6% (n:11) were aphonic in which the mucosal vibration was not observed in the videolaryngoscopic examination of these patients due to the large glottic gap. Eight patients of the aphonic group were whispering, while the other three were completely silent, only moving their lips.
Roy et al. [26]	To explore the short- and long-term consequences of manual circumlaryngeal therapy as treatment for functional dysphonia.	Voice therapy treatment: - Voice and FVD education supported with instrumental findings - manual laryngeal musculoskeletal tension reduction technique to achieve voicing - Counseling incl. explore factors contributing to FVD - Telephone call to a conversation partner to stabilise voice. Amount: One session: 50-180 min duration.	1. Improvement to auditory-perceptual evaluation ratings post-treatment: auditory-perceptual evaluation: four speech pathology graduate students employed at the university judged randomised recorded sections of "The Rainbow Passage" from each assessment point. Using a seven-point-equal-appearing-interval scale (where 1 = normal voice and 7 = severe voice disorder). Several estimates of inter- and intra-judge reliability and agreement were calculated. These were calculated for (a) Connected speech ("The Rainbow Passage") and (b) sustained vowel samples /a/, /i/, /u/. 2. Relapse: comparison of post-2 or post-3 mean clinician perceptual severity rating to post-1. 3. Episodes of recurrences: Patient interview.	1. Auditory-perceptual evaluation Group means for severity data are reported because trends and significance patterns for connected speech and vowels corresponded. Auditory-perceptual evaluation of severity: 24/25 (96%) improved. 20/25 (80%) improved by 2 scale values, 16/25 (64%) were considered normal only mildly dysphonic at post-1. (a) Connected speech: Pre- to post-1 auditory-perceptual evaluation ratings reduced from a mean of 5.37 (1.50) to 1.91 (0.74) ($p=0.0001$). No significant differences were identified between post-treatment scores, therefore improvements were maintained after a single session. (b) Vowels: Pre- to post-1 auditory-perceptual evaluation ratings reduced from a mean of 5.48 (1.66) to 2.20 (1.16) ($p=0.0001$). No significant differences were identified between post-treatment scores, therefore improvements were maintained after a single session. 2. Relapse (7/25) 28% experienced partial recurrence, 1/25 (4%) complete relapse. 3. Recurrences Data was gathered from 19/25 (76%) pts. Subject attrition: 5 pts had not had sufficient time since treatment to accurately judge recurrence, 1pt gained minimal benefit from treatment and therefore was not included. 13/19 (68%) reported 1+ episodes of recurrence during the follow-up period: 10/13 (77%) within 2-months (mean = 1.4 (0.99) months; range = 1h to 3 months). The majority pts had less than three episodes of recurrences, but there was substantial variability (daily to single incidents). Duration of recurrences ranged from several hours to several weeks, but typically less than 4 days. 8/13 (62%) reported only partial recurrence with less severe symptoms than pre-treatment presentation. 3/13 (23%) reported recurrence to be as severe as pre-treatment. In 9/13 (70%) recurrence resolved spontaneously.

BAI: Beck Anxiety Inventory; BWMTR: Brief Visuospatial Memory Test-Revised; CBT: Cognitive Behavioural Therapy; CVLT-II: California Verbal Learning Test; D-KEFS: Delis-Kaplan Executive Function System; DVT: DoctorVox Voice Therapy Technique; FVD: Functional Voice Disorder; GHQ-28: General Health Questionnaire; GRBAS: Grade, Roughness, Breathiness, Asthenia, Strain scale; HAD: Hospital Anxiety and Depression Scale; Hertz: JLO: Judgement of Line Orientation; MPAL-4: The Mayo Portland Adaptability Inventory-4; MPT: Maximum phonation time; NSI: Neurobehavioral Symptom Inventory; NSI-22: Neurobehavioral Symptom Inventory-22; PAL: Personality Assessment Inventory; PCL-C: PTSD Checklist Civilian Version; PHQ-9: Patient Health Questionnaire; PHQ-9: Patient Health Questionnaire-9; RDS: Reliable Digit Span; Rey-Osterrieth Complex Figure; SWLS: Satisfaction with Life Scale; TMT: Trail Making Test; TOMM: Test of Memory Malingering; VHI: Voice Handicap Index; VHI-10: Voice Handicap Index-10; VPO: Voice Performance Questionnaire; WALS-IV: Wechsler Adult Intelligence Scale; Fourth Edition; WCST-64: Wisconsin Card Sorting Test-64; WHOQOL-BREF: The World Health Organisation Quality of Life-BREF; WMS-IV: Wechsler Memory Scale Fourth Edition; WMT: Word Memory Test; WRA14: Wide Range Achievement Test 4th Edition.

though reliable and valid has been criticised for limited sensitivity [35]. Two studies [27,28] used perceptual judgement, with incomplete information about how the findings were determined. The Roy et al. (1997) paper gave the most robust explanation of how the auditory-perceptual evaluation used in their study was evaluated for reliability [26]. Additionally, three studies [24,25,29] used patient self-reported assessments, such as the VHI-10 or VPQ, these measures have been found to be reliable and valid instruments for assessment [36]. The positive outcomes of the studies need to be considered against the issues raised in quality assessment.

Functional stuttering – intervention effectiveness and efficacy

Only one single case study reported a patient with functional stuttering which will be summarised, but as only one study can be reported on, a synthesis of the evidence is not possible. Yam et al. (2016) described a multidisciplinary approach to managing a patient presenting with a wide range of functional symptoms as part of his FND diagnosis [30]. Their aim was to demonstrate a good outcome when FND is treated in a multi-disciplinary inpatient setting, therefore the study did not focus on the FCD. The Speech and Language Pathology (SLP) sessions were reported to cover a broad range of communication difficulties, not specific to the functional stuttering: “sessions focused on reduced language skills, stuttering, reduced perception and discrimination, learning and memory strategies, and attention training” (p. 106). The paper reports using speech techniques commonly used for all types of stuttering behaviours (not just functional stuttering): prolongation, using melody, and easy onset. However, the paper reports that these approaches were ineffective and discontinued. Further input merged with neuropsychology and focused on cognitive rehabilitation and encouraging speaking in group settings. Though a quality assessment failing is that the paper did not provide sufficient information about dosage of the SLP sessions offered.

The paper’s lack of detail and focus on the functional stuttering makes it difficult to determine the effectiveness and efficacy of the intervention offered. A wide range of cognitive assessments were used as outcome measures, but no standardised speech outcome measures. The patient’s improvement in speech was determined by a perceptual evaluation of his speech only, though it was not reported who made the evaluation. On follow up post-discharge his speech was reported to be “fluent, with only occasional stutter” (p. 107) [30], yet it cannot be determined what caused this improvement.

Robustness of synthesis

The robustness of the synthesis is hindered by the small number of studies, the majority of which focus on FVDs only, the variety of study designs, methodological flaws, and quality limitations. Nevertheless, PRISMA guidance was adhered to, to ensure that the review was reported according to an established framework. Additionally, the broad search strategy was designed to ensure all available evidence was found.

Discussion

This systematic review followed PRISMA guidance to determine the current evidence base of effective and efficacious interventions for FCDs. Six health databases were searched. Seven studies were deemed to meet the inclusion criteria: one RCT, one pre-post study, three case series, and two single case studies. All but one study focused on FVDs, with the remaining study presenting a

functional stuttering case study. It is therefore not possible to identify similarities in approach across the full range of FCDs. Interventions offered were different between studies, but there were some common methods used. All studies reported improved outcomes post-intervention, but this needs to be interpreted cautiously considering there was moderate to high risk of bias in 5/7 studies. Most did not sufficiently verify findings statistically or report full information on participant recruitment. The studies had small participant numbers and generally did not provide enough information for replication. Participants were disproportionately female (81.8%) which aligns with findings in larger studies on FND and FVDs specifically [9,37].

Interventions for Functional Communication Disorders

Similar themes in the intervention approach across studies included: (1) educating patients about voice (in FVD papers) and FND or the psychogenic aspect to their FCD; (2) generally typical voice or speech therapy was offered i.e., similar to what is offered to patients with organic communication disorders; and (3) a counselling or psychological intervention was offered or recommended alongside SLT.

These three themes align with the international expert consensus recommendations [4]. These recommendations provide the most comprehensive guidance for the management of FCDs, and thus the best benchmark to compare the study findings to [4]. Firstly, the importance of educating and explaining the functional disorder is highlighted in the consensus recommendations (p. 4) and this was frequently included in the intervention in the review studies. Secondly, most studies incorporated a psychological treatment and/or explored the participant’s psychogenic factors. The consensus recommendations highlight the clinician’s need for counselling skills to show empathy and respect for the patient’s distress, but it goes further:

We emphasise that the therapist can helpfully and appropriately engage in supportive discussion about the role of anxiety, or about the impact that symptoms have had on relationships and everyday life, without special training in counselling or psychotherapy. These discussions might, for example, help the patient to plan for situations where symptoms may recur, and allow them to explore how best to manage future relapses. (p. 8)

Furthermore, the consensus recommendations specifically suggest incorporating principles of CBT into SLT intervention, which supports the aims of Deary et al. [25]. They reported that CBT has been found to be helpful for other FND symptoms, so it is suggested that FCD should similarly benefit [38,39].

As well as the general recommendations for the management of FCDs, the consensus recommendations provide specific advice depending on the type of FCD, these will be presented according to the included studies.

Functional voice disorders

Recommendations for the treatment of FVDs echo the approaches taken in the review studies. Firstly, reducing excessive musculoskeletal tension using palpation, massage or repositioning is advised and was used in two of the studies [24,26]. Secondly, traditional evidence-based voice therapy exercises are reported to be useful, these include: Semi-Occluded Vocal Tract Exercises, which was reported in two papers [24,29] and Resonant Voice Exercises which was reported by Umeda and colleagues [24]. Additionally, the recommendations advise generalisation of normal phonation beyond the clinical setting where possible, as also

mentioned in two studies [26,29]. Finally vocal hygiene is recommended as a helpful aspect of intervention, this was also used in two studies [24,25].

Functional stuttering

The Yam et al. (2016) study [30] described trialling specific speech techniques for stutter: easy onset and prolongating speech sounds; both these techniques are mentioned in the recommendations. The recommendations also advise that alongside symptomatic treatment clinicians should include “collaboration with mental health professionals” (p. 9); this was reported in the Yam study where SLT worked alongside a neuropsychologist.

Limitations and directions for future research

A significant limitation of the review is the small number of studies identified and the preponderance of studies on FVDs, with very limited evidence for other FCDs. This hinders drawing any conclusions on whether the evidence supports universal approaches to FCDs. The review highlights the lack of evidence-based intervention research into all forms of FCDs. It is acknowledged that there are challenges around designing rigorous studies for a heterogenous population, who may present with FCDs in combination with other functional symptoms. Nevertheless, this is a patient group underserved by research and clinical expertise, the result of which could be unnecessary costs to healthcare and diminished quality of life for those suffering with FCDs.

Furthermore, the overall quality of the research studies was poor, with small sample sizes, methodological failings, and the older studies often failing to report key information. Most studies were single case studies or case series, which are considered lower levels of evidence. This restricts the ability to draw reliable conclusions from the included studies. Researchers should use quality assessment tools and reporting guidance in the planning and design of research. This will help to ensure that publications address the key requirements which demonstrate the study's validity.

A final limitation and ongoing challenge when reviewing the evidence for FCDs relates to inconsistencies in terminology. “Functional” is a term which has been used in communication disorder research to describe both psychogenic/conversion disorder presentations and much more generally to describe the use of communication in everyday activities. Terminology inconsistencies were found to be particularly challenging when reviewing the FVD literature, where terms such as ‘non organic’, ‘benign’, and ‘MTD’ have been used interchangeably to describe functional and non-functional voice disorders. Some papers were excluded because they did not explicitly explain what was meant by the term used, and therefore it was not possible to determine if the inclusion and exclusion criteria had been met. This has been acknowledged by Payten et al. (2022) as an ongoing issue, which they aimed to address in their review on frameworks, terminology, and definitions used for the classification of voice disorders [40].

Future research should address the paucity in FCD intervention studies that use rigorous methodology with control groups and adhere to reporting guidance to ensure validity and reliability. This review has found research on FCD intervention to be lacking specificity of intervention and lack of consistent use of valid and reliable outcome measures. Future research should report in more detail intervention characteristics, e.g., using the TIDieR framework [41], so that studies can be replicated and we can begin to understand what are the most effective components of intervention. Greater consistency in the use of outcome measures will allow study findings to be synthesised in meta-analysis and thus greater

confidence in the findings. Lastly, the inclusion of qualitative data may support understanding of patient experience and help us understand better how therapies work.

Conclusion

This systematic review sought to identify and appraise FCD intervention studies. Seven studies were included, six on FVDs and one on functional stuttering. Interventions offered differed between studies, though there were some similar themes. Most interventions included education on communication mechanisms, FCDs, and how one's emotional state impact communication; a psychological component; and SLT exercises, commonly used for organic disorders. However, the quality of studies was generally poor, with unclear reporting of inclusion and recruitment practices and insufficient statistical analysis to support the study's findings. This systematic review has highlighted the lack of high-quality research evidence to guide the interventions clinicians should offer to people with FCDs. This in turn embeds SLTs feeling unsure and underprepared to manage these disorders [13]. Directions for future research to address these shortcomings have been raised.

Disclosure statement

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