ASHA American Speech-Language-Hearing Association

AJSLP

Review Article

Behavioral Management of Respiratory/Phonatory Dysfunction for Dysarthria Associated With Neurodegenerative Disease: A Systematic Review

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ARTICLE INFO

Article History:
Received July 24, 2023
Revision received November 2, 2023
Accepted November 2, 2023

Editor-in-Chief: Katherine C. Hustad
Editor: Nancy Pearl Solomon

https://doi.org/10.1044/2023_AJSLP-23-00274

ABSTRACT

Purpose: This systematic review represents an update to previous reviews of the literature addressing behavioral management of respiratory/phonatory dysfunction in individuals with dysarthria due to neurodegenerative disease.
Method: Multiple electronic database searches and hand searches of prominent speech-language pathology journals were conducted in accordance with Preferred Reporting Items for Systematic Reviews and Meta-Analyses standards.
Results: The search yielded 1,525 articles, from which 88 met inclusion criteria and were reviewed by two blinded co-investigators. A large range of therapeutic approaches have been added to the evidence base since the last review, including expiratory muscle strength training, singing, and computer- and device-driven programs, as well as a variety of treatment modalities, including teletherapy. Evidence for treatment in several different population groups—including cerebellar ataxia, myotonic dystrophy, autosomal recessive spastic ataxia of Charlevoix–Saguenay, Huntington's disease, multiple system atrophy, and Lewy body dementia—were added to the current review. Synthesis of evidence quality provided strong evidence in support of only one behavioral intervention: Lee Silverman Voice Treatment Program (LSVT LOUD) in people with Parkinson's disease. No other treatment approach or population included in this review demonstrated more than limited evidence, reflecting that these approaches/populations require urgent further examination.
Conclusion: Suggestions about where future research efforts could be significantly strengthened and how clinicians can apply research findings to their practice are provided.
Supplemental Material: https://doi.org/10.23641/asha.24964473

This project is a first step in an effort by the Dysarthria Working Group (DWG), which is part of the Evidence-Based Clinical Research (EBCR) Committee of the Academy of Neurologic Communication Disorders and Sciences (ANCDS), to update previously published systematic reviews related to behavioral management of respiratory/phonatory dysfunction from dysarthria (Yorkston et al.,

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2003); treatment of loudness, rate, or prosody in dysarthria (Yorkston et al., 2007); and related clinical practice guidelines (Spencer et al., 2003). Yorkston et al. (2003) reported on the findings from 35 intervention studies, mostly published between 1995 and 2001. None of the studies were randomized controlled trials (RCTs), and most studies focused on examining the effectiveness of biofeedback devices (delayed auditory feedback [DAF], amplifiers, masking) and the Lee Silverman Voice Treatment Program (LSVT LOUD). Four years later, this research group reported on the findings from 51 intervention studies (Yorkston et al., 2007). Studies consisted mainly of Phase I and II trials, with eight Phase III trials. Studies focused on examining the effectiveness of biofeedback devices (DAF, pacing board, metronome), verbal cueing, and LSVT LOUD. Given the substantial expansion of the literature on treatments for respiratory and phonatory disorders, we have focused this review article on neurodegenerative disease specifically.

Since the publication of this first ANCDS study of respiratory/phonatory treatments, four systematic reviews and one Cochrane review have been published; all of these focused on Parkinson's disease (PD; Atkinson-Clement et al., 2015; Barnish et al., 2016; Herd et al., 2012; Pu et al., 2021; Yuan et al., 2020). Three of these systematic reviews concluded there is strong evidence for the use of LSVT LOUD in people with mild speech symptoms. No benefits of auditory feedback devices, or singing, could be established (Atkinson-Clement et al., 2015; Barnish et al., 2016). High-intensity respiratory treatments were found to have positive effects on pitch and volume, but the long-term benefits extended only to pitch (Atkinson-Clement et al., 2015). The Cochrane Review concluded that there is no evidence to support that one type of speech therapy is better than another (Herd et al., 2012). In general, these reviews indicated that more studies are needed, particularly ones with larger sample sizes and more rigorous designs. The purpose of this systematic review was to investigate behavioral management of respiratory and/or phonatory dysfunction in individuals who have dysarthria related to neurodegenerative disease.

Method

Search Strategy

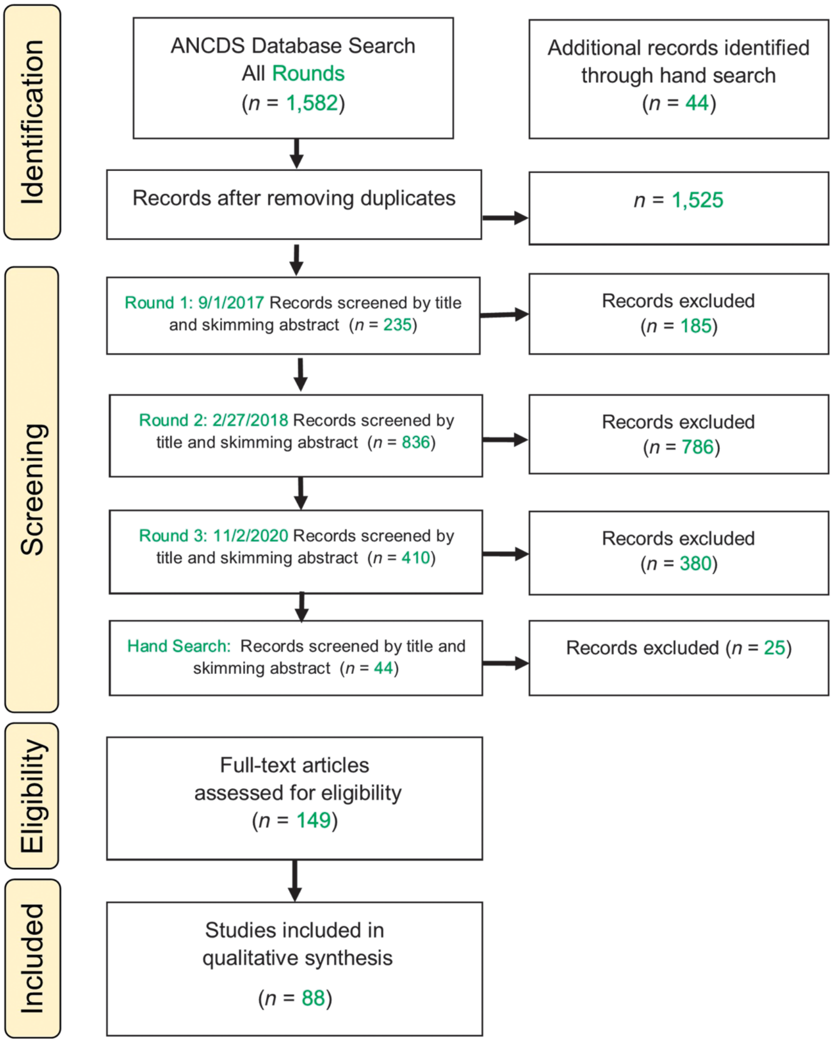
This systematic review was completed in accordance with Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) standards (Moher et al., 2015; Shamseer et al., 2015). PubMed, PsycINFO, and CINAHL electronic databases were searched to identify peer-reviewed articles published through November 2, 2020, using keywords and subject headings (see Supplemental Material S1). Inclusion in the final review was determined using predefined criteria pertaining to population and study characteristics. Specifically, therapeutic behavioral intervention studies of any design (including behavioral and device-driven interventions, compensatory strategies, and stimulability for use of speech-related cues), with a focus on dysarthria secondary to respiratory or phonatory dysfunction, in adults with an underlying neurodegenerative condition were included in the review. Articles were excluded if the dysarthria was unrelated to neurodegenerative disease (e.g., stroke, traumatic onset, spinal cord injury), they had a diagnostic focus or described respiratory/phonatory function without intervention, they described populations requiring mechanical ventilation, they examined respiratory/phonatory interventions in individuals without impairment (except for the purpose of experimental control) or individuals with disorders other than dysarthria (e.g., vocal nodules, psychogenic dysphonia, muscle tension dysphonia), they were not behavioral in nature (e.g., surgical or pharmacological interventions), or outcomes did not include speech-related function. Articles published in languages other than English, books, review articles (except systematic reviews), conference abstracts, correspondence articles, case studies, and case series were also excluded. Additionally, a hand search was completed to capture articles published prior to PubMed indexing dates for prominent speech-language pathology journals.

The overarching search strategy was to combine (Boolean AND) topic searches with (Boolean OR) focused on dysarthria and speech terms, respiratory and phonatory terms, intervention terms, and neuromuscular disease terms, respectively. Search terms were generated by members of the ANCDS DWG, which is part of the EBCR Committee. Supplemental Material S1 provides a detailed list of databases and search terms used.

Article Screening Process

Following the initial search, articles were assigned a code and entered into a spreadsheet along with citation information. Duplicates were removed. Article titles and abstracts were screened over three rounds by 12 research assistants (and verified by three writing group members) to identify articles meeting inclusion criteria or exclude articles from further analysis based on the exclusion criteria (see Figure 1). Electronic versions of the articles selected for full review were saved to an online repository available to all members of the writing group. Next, articles were assigned to six pairs of DWG reviewers for extraction of relevant data, including study design, participant characteristics, methods, outcome measurement, results, and evaluation of study quality using the

Figure 1. Preferred Reporting Items for Systematic Reviews and Meta-Analyses flowchart detailing the article screening process. ANCDS = Academy of Neurologic Communication Disorders and Sciences.



Physiotherapy Evidence Database (PEDro) scale (“PEDro Scale,” 1999).

Quality and Methodological Review

Six reviewer pairs read and evaluated the scientific quality and methodological caliber of articles identified during the screening process. Their findings were summarized in template format using a secure electronic data capture tool hosted at Teachers College, Columbia University (Research Electronic Data Capture, REDCap; Harris et al., 2009, 2019) that also included the PEDro rating scale (“PEDro Scale,” 1999). The PEDro scale is a tool developed for assessing the quality of randomized clinical trials. Each article was rated by two reviewers. Discrepancies between the reviewers regarding study inclusion or PEDro ratings were resolved by a third rater (S.E.P.). At the conclusion of the review, data were extracted from REDCap and summarized in table format by the first author (S.E.P.).

Results

Article Selection and Review

The database searches produced 1,525 articles, and additional manual searching revealed 44 articles. Following three rounds of screening and a full review of 149 articles, ultimately 88 studies met criteria for qualitative synthesis (see Figure 1 for details within the PRISMA flowchart). Results are presented by disease type and treatment modality below. For each treatment modality, details including study designs, participant characteristics, and PEDro scores are provided in the supplemental materials indicated with the headings.

Reliability

Reviewers obtained 39 items of data for each of the 88 articles (3,432 items across all of the articles). Discrepancies included missing details on inclusion or exclusion criteria (in 28 articles) in one of the reviewers and differences in judgments of PEDro ratings for at least one of the rating categories (in 10 articles). These discrepancies were all resolved by the first author (S.E.P.) following a consensus discussion with the senior authors (D.B., M.T., and J.E.H.)

1. Parkinson's Disease (PD)

1.1. Treatment Modality: Lee Silverman Voice Treatment LOUD (LSVT LOUD)/LSVT Extended (LSVT-X)/LSVT Companion™ (Supplemental Materials S2, S2.1, and S2.2)

Forty studies testing the effects of LSVT LOUD/LSVT Extended (LSVT-X)/LSVT Companion/LSVT verbal cueing techniques met inclusion criteria. These studies tested the effects of treatment on a wide variety of perceptual and acoustic speech and voice outcomes, brain imaging outcomes, listener-reported and quality-of-life (QOL) outcomes, stroboscopic outcomes, respiratory kinematic and laryngeal aerodynamic outcomes, and cognitive outcomes. Over 800 participants with PD and controls were examined. Most included studies had prospective, quasi-experimental (37/41, 90%) designs, along with three (7%) RCTs and one (2%) single-subject–design study. There were four studies (10%) that examined the effects of LSVT LOUD in group therapy, with the remainder (90%) focusing on individual treatment. Most treatments were delivered face-to-face (36/41, 88%), with five studies (12%) examining LSVT LOUD in the context of telehealth. Patient-reported outcomes (PROs) were analyzed in 18 studies (44%). Four studies (10%) reported measures of treatment effect size.

1.1.1. Acoustic Findings

The most commonly examined treatment outcome was vocal intensity. Twenty-eight studies reported changes in vocal intensity immediately following LSVT LOUD (Cannito et al., 2012; Constantinescu et al., 2011; de Azevedo et al., 2015; El Sharkawi et al., 2002; Griffin et al., 2018; Halpern et al., 2012; Huber et al., 2003; Körner Gustafsson et al., 2019; Moya-Gale et al., 2018; Nakayama et al., 2020; Narayana et al., 2010; Ramig et al., 1996, 2018; Ramig & Dromey, 1996; Ramig, Sapir, Countryman, et al., 2001; Ramig, Sapir, Fox, & Countryman, 2001; Sale et al., 2015; Sapir et al., 2007; Sauvageau et al., 2015; Searl et al., 2011; Spielman et al., 2007, 2011; Theodoros et al., 2006, 2016; Traverse, 2016; Tripoliti et al., 2011; Wight & Miller, 2015; Wohlert, 2004); one of these was descriptive in nature (Wohlert, 2004). The combined results of these studies suggest post-treatment improvements in vocal intensity between 2.3 and 31.0 dB, depending on the elicitation task (see Supplemental Material S2.2). In some studies, improvements in intensity were maintained at 1 month (Moya-Gale et al., 2018), 2 months (Howell et al., 2009), 3–4 months (Traverse, 2016; Wohlert, 2004), 6 months (Halpern et al., 2012; Ramig et al., 1996; Ramig, Sapir, Fox, & Countryman, 2001; Spielman et al., 2007, 2011; Tripoliti et al., 2011), 7 months (Ramig et al., 2018), 12 months (Körner Gustafsson et al., 2019; Nakayama et al., 2020; Ramig et al., 1996; Wight & Miller, 2015), and 24 months (Ramig, Sapir, Countryman, et al., 2001; Wight & Miller, 2015) following treatment. The results of two studies indicated that improvements were not dependent on treatment modality, that is, in-person versus teletherapy (Griffin et al., 2018; Theodoros et al., 2016), and that, for some outcomes, teletherapy may be the superior modality, as indicated by greater improvements in vocal intensity (Griffin et al., 2018). Two studies included participants treated with deep brain stimulation of the subthalamic nucleus (DBS-STN; Spielman et al., 2011; Tripoliti et al., 2011), with conflicting results. One of these studies found no improvements in vocal intensity in the DBS-STN group, while participants treated with medication only showed significant gains (Tripoliti et al., 2011); another study reported significant gains in both participant groups (Spielman et al., 2011). In other studies, both descriptive and inferential findings suggested that improvements in vocal intensity were achieved regardless of whether the treatment was administered intensively (i.e., four times per week for 4 weeks) or at a less intense schedule (i.e., two times per week for 4–8 weeks, LSVT-X; Spielman et al., 2007; Wohlert, 2004), with the exception of picture description vocal intensity, which was found to be higher following LSVT-X compared to LSVT (Spielman et al., 2007). Another study compared outcomes between LSVT LOUD administered with clinician feedback versus the LSVT Companion computer software, reporting similar therapeutic gains across both modalities (Halpern et al., 2012). One study also compared LSVT LOUD to LSVT ARTIC—a treatment that is focused on articulation. As expected, findings revealed that increases in vocal intensity following LSVT LOUD were significantly larger than those observed following LSVT ARTIC. Only two studies reported treatment effect sizes (Sapir et al., 2007; Searl et al., 2011). One study found a large treatment effect for vowel vocal intensity improvements (Sapir et al., 2007); another study found a small effect (partial $\eta^2 = .245$) of the treatment on vocal intensity combined across various tasks (Searl et al., 2011).

There were conflicting results related to fundamental frequency (f_o). Two studies reported that mean f_o and/or f_o variability increased for reading aloud following both LSVT LOUD and non–LSVT LOUD interventions (Ramig, 1995; Ramig, Sapir, Countryman, et al., 2001).

In contrast, three studies reported no main effect of LSVT LOUD on mean f_o during reading (El Sharkawi et al., 2002; Ramig et al., 1996; Searl et al., 2011). Another study reported an increase in mean f_o but no change in the f_o contour pattern or standard deviation (Whitehill et al., 2011). There was also conflicting evidence regarding mean f_o during conversational monologue. No significant effects on mean f_o were reported in three studies (Ramig et al., 1995, 1996; Searl et al., 2011), although one study reported a significant improvement in f_o range (Manor et al., 2005). A final study reported significant increases in f_o maximum and range—but not f_o minimum—during sustained /a/ (Searl et al., 2011). Vocal pitch range—measured during pitch glides or steps—was reported to improve in two studies (Constantinescu et al., 2011; Theodoros et al., 2006), but not in another study (Theodoros et al., 2016) and not in most participants in a descriptive study (Wohlert, 2004). Another study reported improvements in the mean, but not maximum, value of high frequencies in Hertz following LSVT LOUD (Sale et al., 2015). Amplitude range was not found to improve posttreatment (Manor et al., 2005).

Additional studies examined further acoustic findings. The accuracy of lexical tone did not improve following LSVT LOUD (Whitehill et al., 2011). Another study reported significant improvements in the amplitude of the stressed word in the sentence as well as the unstressed word before the stressed one, along with increased intensity variation across the sentence, suggesting that prosodic variations were higher after treatment (de Azevedo et al., 2015). Another study reported significant changes in cepstral peak prominence (CPP) following LSVT as well as reduced variability in CPP, reduced adjusted low/high spectral ratio, and cepstral/spectral index of dysphonia (Alharbi et al., 2019), all suggesting improvements in voicing periodicity.

1.1.2. Articulatory Findings

One study found significant improvements in acoustic vowel space and increased stop consonant–vowel distinctiveness related to increased vowel intensity following LSVT LOUD (Sauvageau et al., 2015). Posttreatment improvements in the vowel articulation index were observed among participants with and without DBS-STN, suggesting less centralized corner vowels after treatment (Spielman et al., 2011). In addition, significant improvements in the F2i/F2u ratio following LSVT LOUD (Sapir et al., 2007) were reported with a moderate-to-large effect, reflecting greater vowel distinctiveness.

1.1.3. Perceptual Findings

Perceptually, both LSVT LOUD and LSVT-X improved speech (Spielman et al., 2007) compared to no-treatment controls. LSVT LOUD was also found to reduce breathiness (Baumgartner et al., 2001; Constantinescu et al., 2011; Dias et al., 2016; Theodoros et al., 2006), voice scratchiness and shakiness (Wight & Miller, 2015), and strain (Wight & Miller, 2015); improvements in phonation (Tripoliti et al., 2011), respiration (Tripoliti et al., 2011), target vowel representation (Sapir et al., 2007), and vocal quality (Dias et al., 2016) were reported. LSVT LOUD was not associated with improvements in resonance or rate (Tripoliti et al., 2011). There were mixed findings regarding hoarseness, with two studies reporting significant improvements (Baumgartner et al., 2001; Halpern et al., 2012) and two studies reporting no change (Ramig et al., 1995; Theodoros et al., 2006). There was also disagreement regarding perceived articulatory precision, with one study reporting improvements (Wight & Miller, 2015) and three studies reporting no change (Theodoros et al., 2006, 2016; Tripoliti et al., 2011). There were also mixed findings regarding prosody, with several studies reporting improved prosody (Constantinescu et al., 2011; Halpern et al., 2012; Theodoros et al., 2006; Tripoliti et al., 2011; Whitehill et al., 2011; Wight & Miller, 2015), while others reported no change (Ramig et al., 1995; Theodoros et al., 2016). Study findings regarding the effect of LSVT LOUD on perceptual ratings of roughness were also inconsistent, with two studies reporting improvements (Constantinescu et al., 2011; Dias et al., 2016) and one study reporting no changes (Theodoros et al., 2016). Perceived improvements in prosody, phonation, and respiration were only observed in a medically treated group, but not in a group treated with DBS-STN (Tripoliti et al., 2011). There were no differences in perceptual voice features between participants who received LSVT LOUD face-to-face versus via teletherapy (Constantinescu et al., 2011).

Eight studies examined listeners' perceptions of participants' loudness following LSVT LOUD (Constantinescu et al., 2011; Halpern et al., 2012; Ramig et al., 1995; Sapir et al., 2002; Searl et al., 2011; Theodoros et al., 2006, 2016; Wight & Miller, 2015). In three studies, family members rated the loudness of participants as significantly higher posttreatment (Halpern et al., 2012; Ramig et al., 1995; Wight & Miller, 2015), although, in one study, this effect was present for male speakers only (Ramig et al., 1995). Family members also continued to rate loudness as significantly improved at 12 months, but not 24 months (Wight & Miller, 2015). Speech-language pathology graduate students rated the volume of reading and monologue samples as louder following treatment for most participants (Searl et al., 2011). In other studies, speech samples taken at 12 months of follow-up were judged by both expert (Sapir et al., 2002) and lay (Halpern et al., 2012; Sapir et al., 2002) listeners to be louder (Halpern et al., 2012; Sapir et al., 2002) and of "better quality" (Sapir et al., 2002) compared to pretreatment samples as well as louder and of better quality than nontreatment/delayed treatment samples (Sapir et al., 2002). Perceptual ratings of loudness and loudness variability were shown to improve following treatment, regardless of whether the treatment was delivered in person (Constantinescu et al., 2011; Theodoros et al., 2016) or via telehealth (Constantinescu et al., 2011; Theodoros et al., 2006, 2016).

In terms of intelligibility, there were mixed results. Eight studies reported improvements in intelligibility/ease of understanding/requests for repetition and/or accuracy in listeners' transcription of participants' speech immediately following treatment (Cannito et al., 2012; Constantinescu et al., 2011; Levy et al., 2020; Moya-Gale et al., 2018; Nakayama et al., 2020; Ramig et al., 1995; Theodoros et al., 2016; Wight & Miller, 2015), regardless of whether the treatment was delivered via teletherapy or in person. One of these studies included measures of treatment effect size, which were large (Levy et al., 2020). However, in two studies, no improvements in intelligibility were found (Theodoros et al., 2006; Tripoliti et al., 2011). In some studies, gains were maintained at 1 month following treatment (Moya-Gale et al., 2018), but there was disagreement in findings at 12 months (Nakayama et al., 2020; Wight & Miller, 2015). Examination of the effects of an articulation-focused treatment (LSVT ARTIC) on intelligibility in a randomized controlled design revealed no significant improvements (Levy et al., 2020).

1.1.4. Respiratory–Laryngeal Findings

Several studies investigated the effects of LSVT LOUD on respiratory and laryngeal outcomes, with mixed findings. One study reported significantly increased estimated subglottal air pressure following LSVT LOUD (Ramig & Dromey, 1996), with an average pressure increase of 2 cmH_2O and an average maximum flow declination increase of 215 L/s/s in the LSVT LOUD group. Three studies from the same research group reported no treatment effect on measures of forced vital capacity (Ramig et al., 1995, 1996; Ramig & Dromey, 1996). Another study examined the respiratory kinematic patterns (lung volume at speech initiation and termination) and percent vital capacity expended per syllable, finding no consistent effect of LSVT LOUD on respiratory kinematics or variability of the respiratory subsystem (Huber et al., 2003). Qualitative results indicated that different respiratory/laryngeal strategies for increasing volume following LSVT LOUD were used among participants (Huber et al., 2003).

A single study used the relative width of the electroglottographic waveform at 25% of its height as a proxy of vocal fold adduction, reporting a main effect of LSVT LOUD on this outcome (Ramig & Dromey, 1996). However, there was no effect of the treatment on measures of vocal fold open quotient.

While the majority of studies found improvements in maximum phonation time (MPT; Griffin et al., 2018; Ramig et al., 1995; Sauvageau et al., 2015; Searl et al., 2011; Traverse, 2016; Wohlert, 2004), two studies reported no changes (Constantinescu et al., 2011; Narayana et al., 2010). Findings suggest that improvements were only made when the treatment was administered face-to-face compared to via telehealth (Griffin et al., 2018). However, there were no (descriptive) differences between treatments administered intensively (i.e., four times per week for 4 weeks) or at a less intense schedule (i.e., two times per week for 4–8 weeks; Wohlert, 2004).

Significantly shorter utterance duration during reading after LSVT LOUD was reported, but no statistically significant change to utterance duration during monologue (Ramig et al., 1995); another study reported significant reductions in utterance length following treatment (task unspecified; de Azevedo et al., 2015). Pause duration during reading or monologue did not improve following LSVT LOUD (Ramig et al., 1995).

1.1.5. Nonspeech Findings

Three studies reported family members' descriptions of participants initiating conversation, with mixed findings. One study found improvements in participants starting and participating in conversation immediately after treatment, but this was not maintained at 12 or 24 months of follow-up (Wight & Miller, 2015). Another study found no perceived improvements in conversational initiations following LSVT LOUD (Ramig et al., 1995), while a third study found improvements in a nonmetropolitan group who received therapy via telehealth compared to a metropolitan group (Theodoros et al., 2016). A fourth study reported family members' ratings of participants' communication effectiveness via the Modified Communication Effectiveness Index (CETI-M), with posttreatment improvements noted up to 6 months following LSVT LOUD (Halpern et al., 2012). A single study reported on objective measures of pragmatics following LSVT LOUD, finding that both the mean number of turn-taking counts and mean number of speech initiation counts significantly increased (Manor et al., 2005).

Regarding PROs, the Voice Handicap Index (VHI; Jacobson et al., 1997) was the most commonly used outcome measure, although findings were not consistent across studies. Three studies reported improvements in VHI total score following treatment (Searl et al., 2011; Spielman et al., 2011; Wight & Miller, 2015), including participants with DBS-STN (Spielman et al., 2011), with a large treatment effect (Wight & Miller, 2015). Another study reported improvement in the functional subscale of the VHI, but not in the physical or emotional subscales (Moya-Gale et al., 2018). A final study reported significant improvements in self-rated participant CETI-M scores following LSVT LOUD (Ramig et al., 2018). In terms of maintenance, posttreatment effects were noted to remain at 3 months in both an LSVT LOUD group and a speech therapy control group (Sackley et al., 2018) as well as in the LSVT LOUD group at 7 months (Ramig et al., 2018) and 12 months (large treatment effect; Wight & Miller, 2015), but not 24 months (Wight & Miller, 2015). However, another study of participants treated with DBS-STN reported no (descriptive) gains maintained at 6 months (Spielman et al., 2011). By contrast, three studies reported no significant changes in VHI scores following LSVT LOUD (El Sharkawi et al., 2002; Halpern et al., 2012) or LSVT-X (Spielman et al., 2007).

For participant self-ratings, men who received LSVT LOUD reported the greatest improvements in loudness (Ramig et al., 1995). Improved loudness and fluency were reported following treatment in a clinical case (Körner Gustafsson et al., 2019), with treatment effects observed at 6, but not 12, months. There was, however, one study that did not support improvements in self-perceived loudness, as measured via the Visual Analog Perceptual Rating Scale (Manor et al., 2005). A single study reported improvements on the Speech Assessment Scale (a measure of self-perceived speech clarity; D. D. Johnson, 1975) following treatment (Manor et al., 2005), although another study by a different research group reported no improvements on either this measure (El Sharkawi et al., 2002) or a visual analog scale relating to the statement: "I speak so others understand" (El Sharkawi et al., 2002). Another study sought participant feedback following treatment that was administered over 2 months in a weekly group setting (Searl et al., 2011). Descriptive results indicated that most participants perceived improvements in their voice/speech after the treatment and noticed others commenting on positive changes to their voice/speech, less requests for repetition, and increased talking. However, some participants also noted that the treatment was tiring.

Several QOL indices were also used to examine treatment effects, with mixed findings. One study found significant, large increases in voice-related QOL (V-RQOL) scores following LSVT LOUD among people with mild PD compared to no-treatment controls and healthy controls (Saffarian et al., 2019). Another reported descriptive improvements on this measure at 3 months posttreatment in both the LSVT LOUD group and a speech therapy group (consisting of a range of exercises and cues targeting respiration, phonation, articulation, and prosody, as well as augmentative and alternative communication [AAC] strategies and therapeutic devices) compared to controls (Sackley et al., 2018). No improvements were reported on the EuroQol (EQ-5D; Sackley et al., 2018). Descriptive improvements in the Carer-Reported Quality of Life scale were reported at 3 months posttreatment in both the LSVT LOUD group and a speech therapy group compared to controls (Sackley et al., 2018).

In terms of dysarthria impact, results indicated general improvements for participants following treatment, which were maintained in the short term, such as descriptive improvements in Parkinson's Disease Questionnaire-39 (PDQ-39) communication score (Sackley et al., 2018), Living with Dysarthria Questionnaire (Sackley et al., 2018), and self-rated fatigue (Körner Gustafsson et al., 2019). Significant improvements in the Speech Assessment Scale (Manor et al., 2005), acceptance of dysarthria (Theodoros et al., 2016), and Dysarthria Impact Profile (DIP) and Questionnaire on Acquired Speech Disorders scores (Körner Gustafsson et al., 2019) were reported following treatment, with gains maintained at 3 months (Sackley et al., 2018) and 6 months (Körner Gustafsson et al., 2019), but not 12 months (Körner Gustafsson et al., 2019). In general, it did not seem to matter whether the treatment was delivered in person or via telehealth (Theodoros et al., 2016). It is worth noting that, in one study, significant improvements in dysarthria impact scores were also noted in a speech therapy control group (Sackley et al., 2018), suggesting that speech therapy in general, rather than LSVT in particular, may be the variable of influence.

Studies were generally in agreement that the effects of LSVT LOUD did not extend to nonspeech outcomes, such as depression via the Beck Depression Inventory (Beck et al., 1987; Ramig et al., 1995, 1996); Sickness Impact Profile (Gilson et al., 1975); social interaction scores (Ramig et al., 1995, 1996), although there was a temporarily reduced impact of PD on communication scores that was not maintained posttreatment (Ramig et al., 1996); Unified Parkinson's Disease Rating Scale (UPDRS) motor scores (Ramig et al., 1996); and cognition based on a battery of four cognitive tests (Ramig et al., 1996).

Two studies used brain imaging techniques to characterize treatment-induced neural changes following LSVT LOUD (Baumann et al., 2018; Narayana et al., 2010). Functional magnetic resonance imaging (Baumann et al., 2018) and positron emission tomography (Narayana et al., 2010), characterization of treatment-induced changes, were generally interpreted to indicate that there was more recruitment of cortical speech networks and recruitment of new cortical areas (including right hemisphere) during speech production after treatment.

1.2. Treatment Modality: SPEAK OUT!® and The LOUD Crowd (Supplemental Materials S3 and S3.1)

Two prospective, quasi-experimental studies investigating the effects of SPEAK OUT! and The LOUD

Crowd on outcomes including vocal intensity, voice acoustics, and V-RQOL met inclusion criteria (Behrman et al., 2020; Boutsen et al., 2018). SPEAK OUT! targets vocal function by prompting participants to speak with "intent," that is, with a deliberate focus on increasing vocal loudness and varying intonation. SPEAK OUT! is similar to LSVT LOUD in that treatment is intensive and involves hierarchical speech, voice, and cognitive exercises such as vocal warm-ups, sustained vowel production, pitch glides, and reading aloud. However, the approach differs in the sense that the number of sessions is based on patient progress. In one of the studies included in this review, participants were also encouraged to attend group therapy—The LOUD Crowd—once per week for 7 weeks for treatment maintenance (Behrman et al., 2020).

1.2.1. Acoustic Findings

Significant posttreatment changes in vocal intensity were reported for reading (average = 8- to 9-dB increase; Behrman et al., 2020; Boutsen et al., 2018), monologue (average = 7.70-dB increase; Behrman et al., 2020), and conversation (average = 7-dB increase; Boutsen et al., 2018). The greatest improvements were observed in participants with the least amount of time since disease onset (Boutsen et al., 2018). No measures of treatment effect size were provided.

Although there were no significant changes in intensity variation during reading after treatment, significant improvements during monologue (average = 1.33- to 2.29-dB variation) were reported (Behrman et al., 2020). Participants with more severe PD and/or lesser time elapsed since diagnosis showed the greatest improvements in this area (Behrman et al., 2020). Improvements in f_o variability during both reading (Behrman et al., 2020; Boutsen et al., 2018) and monologue (Behrman et al., 2020) were reported.

Behrman et al. (2020) reported significant improvements in the mean CPP following SPEAK OUT!, with the greatest improvements observed in women. This research group also measured changes in the normalized pairwise variability index (nPVI) for pitch, intensity, and duration, reporting that only nPVI for pitch improved following treatment, suggesting that prosody was only partially improved by treatment (Boutsen et al., 2018).

1.2.2. Articulatory Findings

Only one study revealed a significantly slowed rate of speech following SPEAK OUT! (Boutsen et al., 2018). The same study also measured listener perceptual ratings, finding that, following SPEAK OUT!, expert listener ratings of intensity, intonation, hoarseness, and overall dysarthria severity showed improvements.

1.2.3. Nonspeech Findings

Behrman et al. (2020) and Boutsen et al. (2018) both examined PROs, reporting significant posttreatment improvements in V-RQOL (Behrman et al., 2020; Boutsen et al., 2018) and VHI (Boutsen et al., 2018; Jacobson et al., 1997) scores. Participants with less severe PD reported better posttreatment V-RQOL scores (Behrman et al., 2020). Finally, attendance at The LOUD Crowd sessions did not have a significant effect on mean intensity, intensity variation, f_o variation, or V-RQOL scores (Behrman et al., 2020).

1.3. Treatment Modality: Loud and Proud (Supplemental Materials S4 and S4.1)

Two prospective, quasi-experimental studies investigating the effects of Loud and Proud—a group speech maintenance program—for people with PD following LSVT LOUD met inclusion criteria. The program is designed to recalibrate participants' vocal loudness to post–LSVT LOUD levels and facilitate self-monitoring of loudness levels to promote generalization into everyday communication contexts. Edwards et al. (2018) examined acoustic and QOL-related outcomes following eight, weekly Loud and Proud sessions provided in the context of group therapy, and Quinn et al. (2019) examined Loud and Proud in the context of group therapy delivered via telehealth ("eLoud and Proud").

1.3.1. Acoustic Findings

Following 4 weeks of eLoud and Proud, participants demonstrated increased vocal intensity for sustained phonation and reading aloud, which were maintained at 3 months of follow-up (Quinn et al., 2019). Similar improvements in vocal intensity for sustained vowels, reading, and conversation were observed following 8 weeks of in-person therapy (Edwards et al., 2018). Although improvements in monologue vocal intensity were significant immediately following the treatment (Edwards et al., 2018; Quinn et al., 2019), these were not maintained at 3 months of follow-up (Quinn et al., 2019). Finally, both studies reported no significant improvements in frequency range following treatment.

1.3.2. Respiratory–Laryngeal Findings

Both Edwards et al. (2018) and Quinn et al. (2019) reported no significant changes in MPT following Loud and Proud or eLoud and Proud.

1.3.3. Perceptual Findings

Edwards et al. (2018) reported no significant improvements in perceived intelligibility following 8 weeks of Loud and Proud.

1.3.4. Nonspeech Findings

In terms of psychosocial outcomes, there was no significant impact of eLoud and Proud on DIP scores (Quinn et al., 2019), the Parkinson's Disease Questionnaire communication domain (Quinn et al., 2019), Communicative Effectiveness Index (Edwards et al., 2018), or Quality of Communication Life scale scores (Edwards et al., 2018). Participants reported a high level of satisfaction with treatment effectiveness and acceptability of the telehealth service delivery model (Quinn et al., 2019). There was, however, a slight preference for face-to-face therapy (Quinn et al., 2019).

1.4. Treatment Modality: Pitch Limiting Voice Treatment (PVLT; Supplemental Materials S5 and S5.1)

A prospective, quasi-experimental study investigating the effects of a novel treatment approach—Pitch Limiting Voice Treatment (PVLT)—met inclusion criteria (de Swart et al., 2003). Thirty-two participants took part in a single session, where they were provided with verbal cues to speak "the way you speak at home" (control condition) or "loud and low" (experimental treatment: PVLT) or to "think loud, think shout" (comparison treatment: LSVT LOUD).

1.4.1. Acoustic Findings

Results revealed an immediate and significant increase in vocal intensity for sustained vowel phonation, reciting, and reading sentences compared to spontaneous speech (i.e., the control condition). Unlike the LSVT LOUD condition, a significant increase in pitch was observed during reciting for the PVLT condition. Both conditions resulted in significantly reduced phonatory jitter.

1.4.2. Respiratory–Laryngeal Findings

No improvements in MPT were noted.

1.4.3. Nonspeech Findings

Participants reported a preference for PVLT over LSVT LOUD, because it "reduces the risks of a strained, pressed, or screaming sound, and it normalizes pitch" (de Swart et al., 2003, p. 500).

1.5. Treatment Modality: Music Therapy/ Singing (Supplemental Materials S6 and S6.1)

Eleven prospective, quasi-experimental studies investigating the effects of music therapy and/or singing on acoustic, respiratory, QOL, and functional speech/voice outcomes in people with PD met inclusion criteria. Of note, two studies also included participants with atypical PD (i.e., Lewy body dementia, progressive supranuclear palsy [PSP], multiple system atrophy [MSA]) as well as some participants who received additional speech therapy during the treatment study (Tamplin et al., 2019, 2020). In all but one study, treatment was provided in the context of group therapy.

1.5.1. Acoustic Findings

The most common outcome measure across studies was vocal intensity, although studies reported conflicting findings. Tamplin et al. (2019, 2020) reported increases in vocal loudness of 5.13 and 5.69 dB in participants who received weekly or monthly singing therapy, respectively. Of note, a greater improvement in vocal loudness was achieved earlier in participants who received weekly therapy, although participants who received monthly therapy eventually reached the same gains by 12 months. In other studies, an increase of 3.46 dB for reading aloud was noted after only 2 weeks of music therapy (Yinger & Lapointe, 2012), as well as a 5.94-dB increase after 6 weeks of therapy (Yinger & Lapointe, 2012) and a 9.91-dB increase after approximately 4 months of therapy (Haneishi, 2001). Elefant et al. (2012) reported significant improvements in singing intensity after both 10 and 20 weeks of treatment that was also associated with greater consistency of intensity and a greater proportion of voiced versus voiceless sounds. However, in three other studies, changes in vocal intensity for reading did not reach statistical significance (Elefant et al., 2012; Shih et al., 2012; Tanner et al., 2016), although in one of these studies, intensity range did significantly improve and was associated with a large treatment effect and high clinical significance (Tanner et al., 2016). These studies did not find evidence for improvements in vocal intensity in conversation/connected speech (Shih et al., 2012; Tanner et al., 2016; Yinger & Lapointe, 2012) or maximum cued volume (Shih et al., 2012).

Regarding other acoustic outcomes, several studies concluded that there were no significant changes in f_o or f_o variation (Di Benedetto et al., 2009; Elefant et al., 2012; Haneishi, 2001; Shih et al., 2012; Yinger & Lapointe, 2012), first- or second-formant frequency variation (Azekawa & Lagasse, 2018), or f_o tremor intensity index (Di Benedetto et al., 2009) following music/singing therapy. However, one study reported significant posttreatment increases in mean f_o for reading, but not conversation (Tanner et al., 2016). Although the authors noted a large treatment effect size, the clinical significance of this finding was questionable. One study noted significant improvements in singing vocal range by measuring the lowest and highest f_o; Elefant et al. (2012) noted improvements following 20, but not 10, weeks of music therapy.

Following 6 weeks of music therapy, no significant changes were observed in measures of jitter (Azekawa & Lagasse, 2018; Di Benedetto et al., 2009), shimmer (Azekawa & Lagasse, 2018; Di Benedetto et al., 2009), peak amplitude variation (Di Benedetto et al., 2009), amplitude tremor intensity index (Di Benedetto et al., 2009), or harmonics-to-noise ratio (Azekawa & Lagasse, 2018), suggesting no change in vocal fold vibratory quality. A single study noted that, following 11 weeks of group singing therapy, significant increases in the mean vowel space area were observed (Higgins & Richardson, 2019).

1.5.2. Articulatory Findings

Two studies measured articulatory outcomes following music therapy. One of these studies included neurologic music therapy techniques—specifically, vocal intonation therapy (i.e., vocal exercises targeting intonation, resonance, loudness, and breath control, among other aspects of voice control) and therapeutic singing (i.e., using singing to target speech initiation, articulation, and breath support for speech)—in a small group of participants with PD-related hypokinetic dysarthria (Azekawa & Lagasse, 2018). Both studies reported no significant changes in diadochokinetic rate following 6 weeks of music therapy (Azekawa & Lagasse, 2018) and no changes in speech or singing rate following 20 weeks of music therapy (Elefant et al., 2012). No significant changes were observed in the number of interword pauses, mean interword pause time, or pause ratio following 6 weeks of music therapy; however, total interword pause time was found to be significantly reduced compared to pretreatment measures, with a large treatment effect (Azekawa & Lagasse, 2018).

1.5.3. Perceptual Findings

There were mixed findings regarding speech intelligibility. Following 6 weeks of music therapy, one study reported no significant changes in intelligibility, as measured by the percentage of discernible words (Azekawa & Lagasse, 2018). However, another study involving 11 weeks of singing therapy reported significant improvements in sentence intelligibility (Higgins & Richardson, 2019). Using the Frenchay Dysarthria Assessment, Evans et al. (2012) estimated that “laryngeal speech” (i.e., clear phonation, appropriate volume and pitch) significantly improved following 2 years of singing therapy; however, other elements—including intelligible words/sentences/conversation—did not change. Following approximately 4 months of music therapy, Haneishi (2001) reported that participants’ self-reported speech intelligibility did not significantly improve; however, caregiver ratings of participants’ intelligibility were significantly higher.

From an auditory-perceptual perspective, one study observed improvements in perceived vocal quality for reading, but not conversation, following singing therapy (Di Benedetto et al., 2009). This same study also observed improvements in perceived fatigue during reading, but not conversation. Using perceptual measures of vocal range, another research group noted improvements following 2 years of weekly singing therapy (Evans et al., 2012). Using the Frenchay Dysarthria Assessment, Evans et al. (2012) estimated that volume significantly improved following 2 years of singing therapy.

1.5.4. Respiratory–Laryngeal Findings

Regarding MPT, findings were mixed. Following 6 weeks of music therapy, Azekawa and Lagasse (2018) reported no significant changes in MPT. This finding was supported by two other studies (Haneishi, 2001; Tanner et al., 2016). However, a single study involving a combination of voice therapy and singing reported significant improvements in MPT (Di Benedetto et al., 2009). In addition, following 6 weeks of music therapy, no significant changes were observed in measures of s/z ratio (Shih et al., 2012).

Findings regarding respiratory measures were also mixed. One study observed improvements in functional residual capacity and maximal expiratory pressures (MEPs; Di Benedetto et al., 2009) following singing therapy, but not forced vital capacity or forced expiratory volume in 1 s. Another research group reported that participants who underwent 3 months of ParkinSong treatment demonstrated significantly larger MEPs compared to control participants (Tamplin et al., 2019), although there were no differences between those who received weekly versus monthly treatment (Tamplin et al., 2019). However, after 12 months of treatment (Tamplin et al., 2020), the monthly ParkinSong group dropped to lower-than-baseline levels of MEP. Similarly, findings were mixed regarding maximum inspiratory pressures (MIPs): One study found significant improvements (Di Benedetto et al., 2009), while another found no treatment effect on measures of MIP (Tamplin et al., 2019, 2020) and sniff nasal inspiratory pressure (Tamplin et al., 2019).

1.5.5. Nonspeech Findings

Regarding PROs, there were mixed findings. One study reported no improvements in overall VHI scores after 12 weeks of group singing therapy (Shih et al., 2012); however, another study found significant improvements in the physical subscale of the VHI after 20 weeks of group music therapy (Elefant et al., 2012). Similarly, one research group reported that improvements in V-RQOL were observed in participants who received weekly singing therapy, while control participants experienced decreased V-RQOL, and participants who received monthly singing therapy remained relatively stable (Tamplin et al., 2019, 2020). However, another study reported no significant changes in V-RQOL (Shih et al., 2012). No improvements on the emotional or functional VHI subscales were reported by any research groups. No improvements on measures of depression (Elefant et al., 2012; Tamplin et al., 2020), perceived overall health (Tamplin et al., 2020), relationship quality (Tamplin et al., 2020), stress (Tamplin et al., 2020), or subscales of the PDQ-39 (Evans et al., 2012) were found. However, anxiety scores were found to be lower for participants who received either weekly singing therapy or weekly control therapy (an active, nonsinging condition with social elements) compared to monthly singing therapy/monthly control therapy (Tamplin et al., 2020; although anxiety scores were within normal limits for all groups). One study also noted self-reported improvements in dribbling/drooling following weekly singing therapy for 2 years (Evans et al., 2012).

Tamplin et al. (2020) were the only group to measure caregiver well-being following singing therapy. They noted significant improvements in measures of depression for caregivers who attended weekly versus monthly ParkinSong groups. However, decreases in caregiver anxiety, stress, QOL, and perceived relationship quality did not reach statistical significance.

1.6. Treatment Modality: Verbal Cueing/Articulation Therapy (Supplemental Materials S7 and S7.1)

Ten studies met inclusion criteria for investigating the effects of verbal cueing/articulation therapy/speech therapy on outcomes including vocal intensity, articulation rate, acoustics, intelligibility, respiratory kinematics, and dysarthria severity in people with PD. In total, 282 participants were analyzed, with etiologies including PD, multiple sclerosis (MS), MSA, PSP, or healthy controls. There was one retrospective RCT design, four prospective quasi-experimental designs, and five quasi-experimental secondary analyses. Most of the studies (9/10) provided in-person, individual therapy, and one study provided both individual therapy and group therapy. Two studies included PROs, and two provided measures of treatment effect size.

1.6.1. Acoustic Findings

Six studies examined the effects of verbal cueing on vocal intensity. In studies that measured the immediate effects of verbal cues, one study reported that Czech-speaking participants with PD increased vocal intensity by 1.36 dB on average during a clear speech condition compared to habitual speech (Skrabal et al., 2020). However, there was no effect of clear speech on loudness variability. Similar studies in English speakers by Tjaden et al. (2013, 2014) revealed that verbal cues for clear speech resulted in increased vocal intensity (3–5 dB) for sentences spoken by people with PD. Increased vocal intensity was also noted in the loud and slow speech conditions, though to a lesser extent. Cues to increase loudness elicited a 7- to 11.2-dB increase in vocal intensity in PD (Hsu et al., 2019; Sadagopan & Huber, 2007) and combined PD-and-MS groups (Tjaden et al., 2013, 2014) with a small-to-medium effect size (PD; Sadagopan & Huber, 2007), compared to habitual speech. Ramig et al. (2018) compared outcomes before and after a 1-month block of intensive articulation treatment (LSVT ARTIC), reporting significant improvements in participants' vocal intensity for sustained phonation, monologue, picture description, and reading aloud, but gains were not maintained at 7 months of follow-up.

Regarding other measures of acoustics, Martens et al. (2015) found in Dutch speakers that intensive speech therapy (comprising daily, hour-long training on speech rate and intonation over 3 weeks) resulted in no significant changes in f_o maximum values for statements or questions or for the final syllable in statements. However, a significant increase in the f_o maximum values for the final syllable in questions was observed. Another study found no differences in f_o range or vowel centralization among loud, slow, or habitual speech conditions in Mandarin-speaking individuals with PD (Hsu et al., 2019). This contrasts with findings from a study of English speakers by Tjaden and Wilding (2011), who found that verbal cues for loud and slow had opposite effects on f_o. Specifically, when speakers have increased vocal intensity, prosodic variation within utterances also improved, which the authors hypothesized may benefit intelligibility. Conversely, when speakers used a slower-than-normal rate, utterance-level prosodic variation declined, which Tjaden and Wilding (2011) hypothesized may be detrimental to intelligibility. Skrabal et al. (2020) found that Czech-speaking participants with PD responded to a clear speech cue by increasing f_o variability by an average of 0.36 semitones compared to habitual speech, although no significant differences in vowel space area were reported. Tjaden and Wilding (2004) reported that all three speech conditions—loud, slow, and clear—resulted in an increase in vowel space area relative to habitual speech. However, this effect was the most robust in the clear speech condition, while the slow speech condition resulted in the greatest temporal distinctiveness for some vowels (Tjaden et al., 2013).

1.6.2. Articulatory Findings

Five studies examined the immediate effects of verbal cueing on articulation rate, with mixed findings. In Czech speakers, cueing to produce clear speech had no significant effect on articulation rate (Skrabal et al., 2020). In Dutch speakers, 3 weeks of intensive speech therapy (described above) also had no effect on speech rate, articulation rate, or mean pause time, although a significant difference in pause frequency was observed during a passage-reading task (Martens et al., 2015). In English speakers, verbal cues for clear speech resulted in a reduced articulatory rate (of approximately 20%) in people with PD and MS (Tjaden et al., 2013, 2014). Verbal cues for loud speech did not alter the articulatory rate for people with PD (Sadagopan & Huber, 2007; Tjaden et al., 2014). People with PD responded to verbal cues to speak slowly;

however, their speech rate reduced to a lesser extent compared to healthy controls (Tjaden et al., 2013).

1.6.3. Perceptual Findings

In terms of auditory-perceptual measures of prosody, one study found that 3 weeks of intensive speech therapy resulted in improved intonation scores for questions, but not for statements (Martens et al., 2015). Furthermore, posttreatment sentence-reading task scores showed greater prosodic variation compared to pretreatment sentence-reading task scores, although there was no change for sentence-repetition task scores (Martens et al., 2015).

The effect of verbal cueing on speech intelligibility was measured in five studies. Intelligibility was reported to be best in the clear speech condition, followed by the loud, habitual, and slow conditions, even when speakers with a history of LSVT LOUD were excluded (Tjaden et al., 2014). One study of Mandarin speakers also concluded that intelligibility was significantly greater in the loud speech condition compared to habitual speech (Hsu et al., 2019). Three studies concluded that speaking slowly did not improve sentence intelligibility (Hsu et al., 2019; Tjaden et al., 2014; Tjaden & Wilding, 2004). Another study found significant improvements in intelligibility following 3 weeks of intensive speech therapy exercises comprising daily, hour-long training on speech rate and intonation over 3 weeks, with a large treatment effect (Martens et al., 2015).

In terms of overall dysarthria severity, Robertson and Thomson (1984) reported significant increases in all areas of a dysarthria profile score following 2 weeks of intensive speech therapy, involving exercises targeting respiration, phonation, facial musculature movements, diadochokinesis, coughing, swallowing, articulation, intelligibility, and prosody. Furthermore, the treatment group continued to show improvements on these measures 3 months after therapy had concluded. As noted by the authors, however, participants were aware they would be tested again upon follow-up and likely continued therapy exercises during this period. Measures of perceived speech severity in people with PD were found to be the best for the loud condition (~7% improvement), followed by the clear speech, habitual speech, and slow speech conditions (Tjaden et al., 2014).

1.6.4. Respiratory–Laryngeal Findings

A single study examined the effects of verbal cues to increase vocal intensity on measures of lung, rib cage, and abdominal volume initiation, termination, and excursion (Sadagopan & Huber, 2007). Both abdominal volume initiation and termination were significantly lower following verbal cues to increase volume by 10 dB, and the authors hypothesized that this would make speech more fatiguing or effortful when a patient is cued to target a specific vocal intensity. Furthermore, all excursions were significantly larger following verbal or noise cues to increase loudness.

1.6.5. Nonspeech Findings

Three studies examined PROs following treatment blocks involving verbal cueing. One study reported significant improvements in CETI-M scores immediately following LSVT ARTIC; however, these changes were not statistically significant at 7 months posttreatment (Ramig et al., 2018). Another study used qualitative assessment methods to measure increased awareness and understanding of dysarthria after therapy (Robertson & Thomson, 1984). Participants indicated an increased desire to communicate, greater confidence in speaking, self-perceived improved intelligibility, and general positivity following treatment. In another study involving Czech speakers, cueing to produce clear speech resulted in a significant reduction in self-perceived severity of dysarthria, as measured by Item 18 on the UPDRS (Skrabal et al., 2020).

1.7. Treatment Modality: Biofeedback (Supplemental Materials S8 and S8.1)

Three prospective, quasi-experimental studies investigating the effects of biofeedback on outcomes including vocal intensity, voice acoustics, and participants' experiences with wearing portable devices met inclusion criteria. In total, 38 participants were analyzed, including people with PD and healthy controls. Two studies involved wearing a biofeedback device outside of the clinic with no in-person sessions, and the final study provided biofeedback in the context of in-person clinic sessions. Two out of three studies included PROs, and no studies provided measures of treatment effect size.

1.7.1. Acoustic Findings

Wearing a portable voice accumulator (VoxLog) for 1 week significantly increased vocal intensity by 1.5 dB compared to baseline (Schalling et al., 2013). J. A. Johnson and Pring (1990) combined speech therapy exercises with biofeedback devices including the Visispeech monitor, sound-level meter, and Jedcom vocal loudness indicator and found that participants demonstrated significantly improved maximum loudness (by 16 dB), volume range (17.2 dB), and volume for speech (monologue; 11.5 dB) and reading (9.5 dB) after 4 weeks.

A single study examined voice acoustics (J. A. Johnson & Pring, 1990) and found no significant differences in f_o or modal pitch for speech following 4 weeks of combined speech therapy with biofeedback. However, pitch range significantly increased, and modal pitch for reading significantly lowered.

1.7.2. Nonspeech Findings

Two studies examined participants' experiences with wearable biofeedback devices. Participants with PD in Schalling et al.'s (2013) study generally reported positive experiences wearing the VoxLog for up to 15 days. Most found it easy to wear and reported that it served as a reminder to use a louder voice and to speak more often. Some drawbacks were that the device felt uncomfortable and/or unwieldly and drew attention. Searl and Dietsch (2015) found that most people with PD and controls reported issues with using a voice monitor (VocaLog) for 1 week. The most common complaints were issues positioning the device, cord, and microphone as well as planning outfits that would accommodate the device. Participants with PD reported significantly worse comfort with the device compared to controls.

1.8. Treatment Modality: Masking Noise/ SpeechVive™ (Supplemental Materials S9 and S9.1)

Five prospective, quasi-experimental studies met inclusion criteria for investigating the effects of masking noise/SpeechVive on auditory-perceptual, voice intensity, acoustic, laryngeal aerodynamic, and respiratory kinematic outcomes. In total, 100 participants were analyzed, including people with PD and healthy controls. Four of five studies involved measuring the immediate effects of masking noise or multitalker babble, and one study involved wearing a SpeechVive device over 8 weeks.

1.8.1. Acoustic Findings

One study measured changes in voice outcomes related to the Lombard effect while white masking noise was presented at 40, 70, and 90 dB SPL (Quedas et al., 2007). Results revealed that, for both people with PD and controls, as the intensity of masking noise increased, vocal intensity and frequency also increased in a nonlinear fashion. Similarly, as masking intensity increased, the stability of intensity and frequency within utterances improved. A second study involved presenting multitalker noise at 70 dBA to people with PD (Sadagopan & Huber, 2007). Participants' vocal intensity was found to be significantly higher in the multitalker babble noise condition, compared to verbal cues to increase loudness, with a medium-to-large treatment effect.

Coutinho et al. (2009) examined the immediate effects of four different listening conditions—habitual, delayed feedback, amplified feedback, and masking noise—on f_o and vocal intensity. Results revealed that all conditions resulted in increased f_o, while none of the conditions significantly changed participants' vocal intensity.

There were two studies investigating the effects of the SpeechVive device—a device designed to provide monoaural, multitalker babble to elicit the Lombard effect during speech. One study measured the immediate effects of the device on acoustic outcomes, among others (Stathopoulos et al., 2014). Results revealed an immediate increase in loudness when participants spoke with the SpeechVive turned on. There were significant increases in estimated subglottal air pressure, along with expected changes to voicing (increases in peak-to-peak glottal airflow and maximum flow declination rate, decreases in open quotient). The second study measured the effects of daily wear/use of the SpeechVive device over 8 weeks, as well as following a 4-week detraining period (Richardson et al., 2014). Following 8 weeks of treatment, participants' vocal intensity significantly increased by an average of 2.9 dB. However, following 4 weeks of detraining, vocal intensity had decreased significantly by an average of 2.5 dB.

1.8.2. Articulatory Findings

Sadagopan and Huber (2007) presented multitalker noise at 70 dBA to people with PD. Results suggested that the noise condition did not affect speech rate.

1.8.3. Perceptual Findings

One study examined the immediate effects of four different listening conditions—habitual, delayed feedback, amplified feedback, and masking noise—on auditory-perceptual voice features, as rated by five expert speech-language pathologists (Coutinho et al., 2009). Delayed feedback resulted in significantly worse vocal quality, pitch, strain, rate, articulation, and loudness. Amplified feedback resulted in significant deteriorations in pitch, as well as increased strain and reduced rate for males and poorer articulation for females. In contrast, masking noise was associated with significant improvements in vocal quality, pitch, strain, rate, articulation, and loudness.

Another study measured the effects of the SpeechVive device over 8 weeks (Richardson et al., 2014). Speech intelligibility increased significantly following treatment, from 93% intelligible to 98%.

1.8.4. Respiratory–Laryngeal Findings

Coutinho et al. (2009) examined the immediate effects of four different listening conditions—habitual, delayed feedback, amplified feedback, and masking noise—on MPT. Results suggested that none of the conditions significantly changed participants' MPT. Sadagopan and Huber (2007) presented multitalker noise at 70 dBA to people with PD and found that the noise condition appeared to promote the most efficient respiratory strategies for increasing loudness, in terms of utilizing volumes in the mid-lung range. Kinematic excursions of the lungs, rib cage, and abdomen were significantly lower in the noise condition compared to verbal cues to increase loudness, with a small effect.

One study examining the immediate effects of the SpeechVive device on respiratory kinematic outcomes revealed increases in lung, rib cage, and abdominal volume initiation as well as lung, rib cage, and abdominal volume termination (Stathopoulos et al., 2014). No changes in utterance length or lung, rib cage, or abdominal volume excursion were observed. Another study measured the effects of the SpeechVive device over 8 weeks, as well as following a 4-week detraining period (Richardson et al., 2014). Following 8 weeks of treatment, participants' voice onset time and percent voicing increased, followed by a detraining effect.

1.9. Treatment Modality: Voice Therapy (Supplemental Materials S10 and S10.1)

Five prospective, quasi-experimental studies investigating the effects of voice therapy on outcomes including laryngeal aerodynamics, intelligibility, speech intensity, and acoustics met inclusion criteria. In total, 90 participants with PD were analyzed. Three of the studies delivered individual therapy face-to-face, one study provided individual therapy via telehealth, and one study provided face-to-face group therapy. Three studies included PROs, and no studies provided measures of treatment effect size.

1.9.1. Acoustic Findings

The specific voice therapy techniques included pushing, overarticulation, maximal-effort sustained phonation, maximal-effort pitch scaling, reading aloud with variable intensities, monologue, self-monitoring and caregiver monitoring, vocal function exercises, and modified LSVT LOUD exercises. In terms of immediate treatment effects, one study reported an immediate increase in the minimum and maximum f_o as well as an increase in vocal range for females, following the finger-kazoo technique (de Lira et al., 2022).

Regarding vocal intensity, increases in the intensity of /a/ (at habitual; Chan et al., 2019; de Angelis et al., 1997) and maximal (de Angelis et al., 1997) volume, number recitation (de Angelis et al., 1997), monologue (average = 6- to 12-dB increase; Chan et al., 2019; Gupta et al., 2008), and sentence reading (average = 18.8-dB increase; Gupta et al., 2008) were observed following therapy, at both intensive (three sessions per week) or less intensive (one session per week) schedules. There was some discrepancy regarding reading intensity, with one study reporting a posttreatment average increase of 25.87 dB (Gupta et al., 2008) and another study reporting a nonsignificant increase in vocal intensity (Chan et al., 2019).

1.9.2. Perceptual Findings

One study reported that fewer participants descriptively rated their voices as weak, high, low, unintelligible, monotonous, and strained–strangled following 1 month of intensive treatment (de Angelis et al., 1997). Another study revealed significantly improved speech intelligibility following intensive voice therapy delivered via a smartphone (Chan et al., 2019).

1.9.3. Respiratory–Laryngeal Findings

When voice therapy was provided over a period of time, two studies found that participants demonstrated similar, significant increases in MPT, whether the therapy was administered intensively (i.e., two to three sessions per week for 1–2 months; de Angelis et al., 1997; Gupta et al., 2008) or less intensively (i.e., once per week for 9 weeks; Gupta et al., 2008). A maintenance schedule of one session per week was sufficient to maintain treatment gains (Gupta et al., 2008). A third study also reported an immediate increase in MPT following the finger-kazoo technique in male participants (de Lira et al., 2022). However, a fourth study that involved intensive therapy delivered via a smartphone did not find any changes in MPT after 12 sessions (Chan et al., 2019). Following 1 month of intensive therapy, participants demonstrated significantly smaller s/z ratios and less glottic airflow or better laryngeal efficiency (de Angelis et al., 1997).

1.9.4. Nonspeech Findings

Significant improvements in VHI scores were observed, suggesting a positive impact of the therapy on voice-related daily life activities, following treatment delivered via a smartphone (Chan et al., 2019). Another study examined the effects of 2 weeks of intensive prosodic therapy on functional speech outcomes. Results revealed significant improvements on perceptual ratings of dysprosody, discrimination of prosodic contrasts, matching speech with facial expression, discrimination of affective and grammatical functions of prosody, discrimination of semantic functions of prosody, and producing questioning speech/tone (Scott & Caird, 1984).

1.10. Treatment Modality: Delayed Auditory Feedback/Frequency-Shifted Feedback (Supplemental Materials S11 and S11.1)

A prospective, quasi-experimental study investigating the effects of DAF/frequency-shifted feedback (FSF) on speech outcomes in people with PD met inclusion criteria (Brendel et al., 2004). This study included 16 people with PD, separated into high- and low-intelligibility groups, as well as 11 controls. Outcomes were measured within a single session.

1.10.1. Acoustic Findings

Regarding pitch, the PD low-intelligibility group showed significantly higher f_o variability in the FSF condition compared to the DAF condition. No participants showed increased mean f_o levels with FSF. In terms of intensity, people with PD and high intelligibility (PD-HI) showed a significant increase in loudness in the DAF condition compared to no altered feedback.

1.10.2. Articulatory Findings

Speech rate was significantly reduced in the DAF condition compared to no altered feedback. The PD-HI group also significantly reduced speech rate in the FSF condition compared to the no–altered-feedback condition and showed a similar pattern in the DAF condition compared to the FSF condition. No effect on the articulation/pause time ratio was observed. However, in the PD-HI group, the no–altered-feedback condition resulted in a significantly lower number—but longer duration—of pauses compared to the DAF and FSF conditions.

1.10.3. Perceptual Findings

The PD-HI group was perceived as significantly less intelligible in both the DAF and FSF conditions, compared to no altered feedback. Intelligibility ratings were also significantly lower for the high-intelligibility group in the DAF condition compared to the FSF condition. All participants with PD were rated as significantly more natural sounding in the no–altered-feedback condition, compared to the DAF condition. Naturalness ratings were also significantly higher for FSF compared to DAF in all participants and higher in the no–altered-feedback condition compared to the FSF condition (PD-HI group only).

1.11. Treatment Modality: Expiratory Muscle Strength Training (EMST; Supplemental Materials S12 and S12.1)

A single-subject study met inclusion criteria for investigating the effects of expiratory muscle strength training (EMST) on respiratory/kinematic outcomes in people with PD (Darling-White & Huber, 2017). This study involved 12 participants. Participants attended weekly in-person training sessions together with daily training at home over 4 weeks. PROs were not measured; however, measures of treatment effect size were provided.

1.11.1. Acoustic Findings

Changes to functional speech outcomes were mixed. Two participants showed positive effect sizes for vocal intensity, and two participants showed negative effect sizes.

1.11.2. Respiratory–Laryngeal Findings

Following EMST, nine participants demonstrated significant, positive changes in MEP. Three participants demonstrated significant, negative effect sizes and were excluded from further analyses due to a lack of expiratory muscle response to training. The majority of participants (7/8) showed significant effect sizes for more typical lung volume initiations. Lung volume termination and excursion were not consistently altered after EMST. Four participants also had significant changes in effect size for utterance length: three positive and one negative.

1.12. Treatment Modality: Group Dynamics/Coaching Strategies (Supplemental Materials S13 and S13.1)

A prospective, quasi-experimental study met inclusion criteria for investigating the effects of group dynamics/coaching strategies on voice and communication outcomes in people with PD (Diaféria et al., 2017). Sixteen participants were divided into either a control group who received traditional voice therapy or an experimental group who received a combination of traditional therapy and group dynamics/coaching strategies. The goal of the experimental treatment was to promote self-awareness and self-development, improve self-esteem, and share coping strategies for situations such as work. Both groups received their treatments in the context of in-person, group therapy. A mixture of PROs, auditory-perceptual ratings, and ratings of the group "climate" (i.e., engagement, conflict, avoidance within the group) were measured at two time points: following traditional therapy and following an additional 4 weeks of either traditional therapy or experimental therapy.

1.12.1. Perceptual Findings

Auditory-perceptual analyses of voice indicated that both groups improved following traditional treatment, followed by a decline in perceptions of vocal quality for the experimental group and further improvements for the control group (based on descriptive comparisons).

1.12.2. Nonspeech Findings

Results revealed significant improvements in participants' self-evaluations of their voice following 4 weeks of the experimental therapy. Interestingly, neither the experimental group nor the control group reported significant improvements on this measure following traditional therapy. However, both groups reported improvements in their self-ratings of communication and living with dysarthria. There were no significant improvements in group climate following treatment, with the exception of avoidance, for which the experimental group showed improvements.

2. Progressive Supranuclear Palsy (PSP)

2.1. Treatment Modality: Lee Silverman Voice Treatment LOUD (LSVT LOUD; Supplemental Materials S14 and S14.1)

A prospective, quasi-experimental study met inclusion criteria for examining the effects of LSVT LOUD for people with PSP (Sale et al., 2015). Sixteen participants with PSP received individual, in-person therapy, and acoustic outcomes were measured.

2.1.1. Acoustic Findings

Following the treatment, minimum and maximum vocal intensity and MPT increased significantly for a prolonged vowel task. No significant differences were observed for measures of minimum and maximum vocal intensity for sentence reading, but there were improvements for measures of minimum and maximum vocal intensity during passage reading. During spontaneous speech, only minimum vocal intensity increased.

2.1.2. Nonspeech Findings

Responsiveness to treatment was not significantly different for the patients with PSP, as compared to those with PD. Although the response to treatment was considered objectively successful, the authors also reported some subjective, potentially negative responses to the treatment, such as high effort and potential stress caused by the treatment.

2.2. Treatment Modality: Verbal Cueing (Supplemental Materials S15 and S15.1)

A prospective, quasi-experimental study met inclusion criteria for examining the effects of verbal cueing for people with PSP (Skrabal et al., 2020). Seventeen Czech-speaking participants with PSP were cued to read a passage, as well as single words, “as clearly as possible” (i.e., clear speech). This was compared with a control condition in which participants were cued to read aloud as they would habitually. Neither PROs nor treatment effect size was measured.

2.2.1. Acoustic Findings

Measures of vocal intensity, loudness variability, and pitch variability were compared between the clear speech condition and the habitual speech condition. Results revealed no immediate effects of clear speech strategies on these outcomes.

2.2.2. Articulatory Findings

Results revealed no immediate effects of clear speech strategies on measures of vowel articulation or speech severity. However, articulation rate showed a significant increase compared to habitual speech, by 0.26 words per second.

2.3. Treatment Modality: Singing (Supplemental Materials S16 and S16.1)

Tamplin et al. (2019, 2020) examined singing therapy in a heterogenous population that included four people with PSP. Subgroup analyses by diagnosis were not completed. These studies are described in detail in the PD section.

3. Multiple System Atrophy (MSA)

3.1. Treatment Modality: Verbal Cueing (Supplemental Materials S17 and S17.1)

A prospective, quasi-experimental study met inclusion criteria for examining the effects of verbal cueing for people with MSA (Skrabal et al., 2020; see Supplemental Materials S17 and S17.1). Seventeen Czech-speaking participants with probable MSA were cued to read a passage, as well as single words, “as clearly as possible” (i.e., clear speech). This was compared with a control condition in which participants were cued to read aloud as they would habitually.

3.1.1. Acoustic Findings

Results revealed no immediate effects of clear speech strategies on vocal intensity, loudness variability, and pitch variability.

3.1.2. Articulatory Findings

Results revealed no immediate effects of clear speech strategies on measures of speech severity; however, articulation rate showed a significant increase compared to habitual speech, by 0.15 words per second.

3.1.3. Treatment Modality: Singing (Supplemental Materials S18 and S18.1)

Tamplin et al. (2019, 2020) examined singing therapy in a heterogenous population that included three people with MSA. Subgroup analyses by diagnosis were not completed. These studies are described in detail in the PD section.

4. Autosomal Recessive Spastic Ataxia of Charlevoix–Saguenay

4.1. Treatment Modality: Melbourne Ataxia Speech Treatment (MAST; Supplemental Materials S19 and S19.1)

A prospective, quasi-experimental study examining the effects of the Melbourne Ataxia Speech Treatment (MAST) in individuals with autosomal recessive spastic ataxia of Charlevoix–Saguenay (ARSACS) was included for review (Vogel et al., 2019). MAST consisted of 45 min of speech therapy per day, 5 days per week, for 4 weeks. Training was a mixture of nonsupervised, computer-based training and biofeedback as well as clinician-led training (first session and weekly phone calls). MAST targeted three elements of speech production: intelligibility, vocal control, and prosody. Exercises included repetition, reading aloud, functional questions, imitation, and self-monitoring with visual and aural biofeedback, and knowledge of results was provided via loudness and pitch displays on a laptop monitor.

4.1.1. Acoustic Findings

There were no statistically observable changes in measures of vocal control (e.g., variability of f_o) after treatment.

4.1.2. Articulatory Findings

There were no statistically observable changes in measures of timing (e.g., mean and variability of pause length) after treatment.

4.1.3. Perceptual Findings

Results revealed significant increases in ratings of monologue and passage reading intelligibility and monologue naturalness, following MAST. Four of the participants (57%) demonstrated an increase in intelligibility of greater than 10%. There were no statistically observable changes in perceptual measures of pitch breaks, prolonged intervals, equal and excess stress, imprecise consonants, or vowel distortion.

5. Cerebellar Ataxia

5.1. Treatment Modality: Lee Silverman Voice Treatment – Extended Version (LSVT-X; Supplemental Materials S20 and S20.1)

A prospective quasi-experimental study examining the effects of LSVT-X on respiratory–phonatory outcomes in cerebellar ataxia met inclusion criteria (Lowit et al., 2020). People with ataxic dysarthria secondary to Friedrich's ataxia ($n = 18$), spinocerebellar ataxia type 6 ($n = 1$), idiopathic cerebellar ataxia ($n = 1$), or spastic paraplegia 7 ($n = 1$) were enrolled in 16 LSVT-X sessions (two sessions per week over 8 weeks) and home practice. All treatment and assessment sessions were delivered remotely via Skype. Of the 19 participants, 12 received the full number of LSVT-X sessions.

5.1.1. Respiratory–Laryngeal Findings

Analysis of results indicated significant changes in MPT, although post hoc analysis provided conflicting results about the change between the first baseline and posttreatment MPT (nonsignificant) and second baseline and posttreatment MPT (significant). The authors note that the large standard deviations for MPT suggest considerable variability in participant performance. Secondary analysis indicated that, of 13 participants who scored below normal limits on MPT initially, 12 improved following treatment ($p < .05$).

5.1.2. Perceptual Findings

Voice quality measures of grade, roughness, breathiness, and asthenia all showed improvements posttreatment; strain remained highly variable. No changes in intelligibility or naturalness were observed posttreatment.

5.1.3. Nonspeech Findings

Questionnaires and patient interviews were conducted to measure the psychosocial impact of the treatment, as well as communication participation and fatigue. While none of the formal measures of psychosocial impact (e.g., Communicative Participation Item Bank [Baylor et al., 2013], VHI [Jacobson et al., 1997], visual analog scale) showed any significant improvements after treatment, posttreatment interviews revealed that most participants felt that their communication had improved following the treatment. Participants also indicated that they preferred the telehealth approach compared to in person.

6. Huntington's Disease (HD)

6.1. Treatment Modality: Speech Therapy (Supplemental Materials S21 and S21.1)

A prospective, quasi-experimental study met inclusion criteria for examining the effects of therapeutic intervention for people with Huntington's disease (HD; Giddens et al., 2010). Five participants with mild–moderate HD completed twice-daily exercises targeting labial and lingual range of motion and strength, respiratory function, and glottal adduction, over a month-long period. Participants were trained in the exercises both individually and as a group, but the exercises were completed at home. Outcomes were measured from cranial nerve examination and patient report.

6.1.1. Respiratory–Laryngeal Findings

Following 1 month of therapy, participants were asked to report any perceived benefits from the treatment. Participants most commonly reported improvements in the areas of breathing and vocal control.

6.1.2. Nonspeech Findings

Results revealed a significant change in the mean cranial nerve function score following treatment, although it is unclear how this was measured. The authors acknowledged that the introduction of dopamine-antagonist therapy for three of five participants mid-study was a potential confounding factor in the results.

7. Lewy Body Dementia

7.1. Treatment Modality: Singing (Supplemental Materials S22 and S22.1)

Tamplin et al. (2019) examined singing therapy in a heterogenous population that included five people with Lewy body dementia. Subgroup analyses by diagnosis were not completed; however, overall group findings are presented in the PD section.

8. Multiple Sclerosis (MS)

8.1. Treatment Modality: Verbal Cueing (Supplemental Materials S23 and S23.1)

The four studies meeting inclusion criteria included prospective quasi-experimental investigations of verbal cueing on speech rate, intensity, pitch, intelligibility, articulatory acoustics, and overall speech severity. A total of 123 participants were examined, with diagnoses of MS, along with those with PD or controls. All studies were from the same research group and employed a similar design where participants were provided with verbal cues to speak “loud” (Tjaden et al., 2013, 2014; Tjaden & Wilding, 2004, 2011), “clear” (Tjaden et al., 2013, 2014), or “slow” (Tjaden et al., 2013, 2014; Tjaden & Wilding, 2004, 2011). Outcomes were measured within a single session.

8.1.1. Acoustic Findings

Specific to MS, Tjaden and Wilding (2004) reported significant changes in vowel acoustic working space following cues to speak “slow” compared to the habitual condition. Vowel space area has been used to quantify articulatory distinctiveness for vowels where a larger vowel space area reflects larger articulatory differences among vowels. Each speech condition—“loud,” “slow,” and “clear”—resulted in an increase in vowel space area relative to habitual speech; however, this effect was the most robust in the clear speech condition (Tjaden et al., 2013). When quantified using measures of distance from the average habitual vowel space, results indicated that the clear speech condition maximized the spectral distinctiveness of vowels (Tjaden et al., 2013). Tjaden et al. (2013) hypothesized that this may result in maximized intelligibility for the clear speech condition.

Increasing vocal intensity (“loud” condition) resulted in differential effects on f_o during a reading passage (Tjaden & Wilding, 2011). Specifically, when speakers increased vocal intensity, prosodic variation within utterances also improved. Conversely, when speakers used a slower-than-normal rate, utterance-level prosodic variation declined. In the MS group, the “loud” condition was found to maximize formant frequency change of point vowels (/i/, /ae/, /a/, /u/), although the clear speech condition resulted in the most formant change on all vowels (Tjaden et al., 2013). Conversely, the “slow” condition was associated with a reduction in formant frequency change and the largest differences in vowel duration and temporal distinctiveness (Tjaden et al., 2013).

Subsequent studies by Tjaden et al. (2013, 2014) revealed that verbal cues for “clear” speech resulted in increased vocal intensity (of 3–5 dB) for sentences spoken by people with MS. Increased vocal intensity was also noted, though to a lesser extent, in the “loud” and “slow” conditions. The “loud” condition elicited an increase of ~7 dB in vocal intensity in the MS group, compared to habitual (Tjaden et al., 2013, 2014).

8.1.2. Articulatory Findings

Results revealed that people with MS were able to reduce their speech rate in the “slow” condition, but to a lesser extent than healthy controls (Tjaden et al., 2013). Subsequent studies by Tjaden et al. (2013, 2014) revealed that verbal cues for “clear” speech resulted in reduced articulatory rate (of approximately 20%) for sentences spoken by people with MS and, to a lesser extent, in the “loud” or “slow” conditions. In the “loud” condition, articulatory rate was also reduced for the MS and control groups (Tjaden et al., 2014).

8.1.3. Perceptual Findings

Regarding intelligibility, findings were conflicting. An earlier study by Tjaden and Wilding (2004) reported that people with MS tended to be the most intelligible in the habitual speech condition, compared to “loud” or “slow.” However, a later study reported that intelligibility was best in the “loud” condition, followed by “clear,” “habitual,” and “slow,” and intelligibility for the “slow” condition was poorer compared to those for the “loud” and “clear” conditions (Tjaden et al., 2014). Results were consistent, however, that speaking slowly did not improve sentence intelligibility (Tjaden et al., 2014; Tjaden & Wilding, 2004).

8.2. Treatment Modality: Expiratory Muscle Strength Training (EMST; Supplemental Materials S24 and S24.1)

A prospective, quasi-experimental study examining the effects of EMST on respiratory–phonatory outcomes in MS met inclusion criteria (Chiara et al., 2007). Seventeen participants underwent 8 weeks of intensive EMST, consisting of weekly in-clinic training sessions and four training sessions per week at home, followed by 4 weeks of detraining (no treatment). Training consisted of four sets of six repetitions, once per day. Training intensity was increased every week as a function of MEP.

8.2.1. Respiratory–Laryngeal Findings

Therapeutic outcome was measured via MEP, MPT, and speaking rate (words per minute). Results revealed significant improvement in MEP for participants with MS, maintained above baseline levels after detraining. Changes to speech production were not reported for people with MS.

8.2.2. Nonspeech Findings

QOL was measured via questionnaires. After training, participants with MS reported a significantly lesser impact of dysarthria on their QOL. Subgroup analyses revealed that, during the detraining period, QOL ratings returned toward baseline values for participants with mild disability. In contrast, for those with moderate disability, QOL ratings remained improved and were no different than QOL ratings of healthy controls.

8.3. Treatment Modality: Lee Silverman Voice Treatment (LSVT LOUD; Supplemental Materials S25 and S25.1)

A single study examining the effects of LSVT LOUD on respiratory–phonatory outcomes in MS met inclusion criteria (Baldanzi et al., 2020). This prospective study employed a single-subject experimental design in a consecutive sample of eight people with MS. Participants attended 16 LSVT LOUD sessions. They were also asked to complete 5–10 minutes of home practice on treatment days and up to 30 minutes of home practice on nontreatment days.

8.3.1. Acoustic Findings

Acoustic outcomes included measures of vocal intensity and MPT during the treatment and at a follow-up period 6–12 months later. Results revealed significant improvement for seven out of eight participants in vocal intensity for prolonged /a/, improvement in five out of eight participants in vocal intensity for functional sentences, and increased MPT in four out of eight participants, with gains maintained for the majority of participants during the follow-up period.

8.3.2. Perceptual Findings

Auditory-perceptual measures of voice were made using the Grade, Roughness, Breathiness, Asthenia and Strain (GRBAS) scale. Following treatment, improvements were made in grade, breathiness, and asthenia scores.

8.3.3. Nonspeech Findings

All participants reported improvements on the VHI (Jacobson et al., 1997) following LSVT LOUD, which were maintained during the follow-up period, except for one participant who returned to their pretreatment score.

9. Myotonic Muscular Dystrophy (MMD)

9.1. Treatment Modality: Speech Warming-Up Exercises (Supplemental Materials S26 and S26.1)

A prospective, quasi-experimental study examining the effects of speech warm-up exercises in people with adult-onset myotonic muscular dystrophy (MMD) met inclusion criteria (de Swart et al., 2007). Participants repeated a series of reading and repetition tasks aloud for at least 10 minutes at different rates of speech.

9.1.1. Acoustic Findings

Outcomes were compared within subjects from the start of the warm-up to the end and to healthy controls. No significant within- or between-group differences in mean speech intensity were observed.

9.1.2. Articulatory Findings

Participants with MMD produced significantly more syllables in monosyllabic sequences after the warm-up, but controls did not. The maximum repetition rate of [ka], [ta], and [pataka] significantly increased by the end of the warm-up, such that pre–warm-up differences in rate between participants with MMD and controls were eliminated. Only repetition of [pa] remained significantly slower for participants with MMD compared to controls. Variability in the rate of repetition of monosyllabic sequences was reduced after the warm-up for participants with MMD, but not controls, who showed no changes in variability. No within- or between-group differences in speech rate for reading, or reciting the months of the year, were observed in the habitual speaking rate condition. However, when cued to read aloud or recite as fast as possible, only control participants increased their speech rate from pre– to post–warm-up.

Summary of Evidence Quality

Statistical comparison of studies was not possible due to the heterogeneity in outcome measurement, intervention parameters, participant characteristics, lack of point estimates (i.e., means, medians), and/or estimates of variability (i.e., standard deviations, confidence intervals). Instead, a qualitative, evidence synthesis strategy was applied using criteria outlined in van Tulder et al. (1999) and Carnaby-Mann and Crary (2007). These studies ranked the quality of evidence for a given intervention as strong, moderate, limited, indicative, or insufficient based on the total methodological score from the PEDro scale as well as the study design. The evidence was considered to be strong if the findings were statistically significant in at least two higher quality RCTs (PEDro score $\geq$ 4). The evidence was considered to be moderate if the findings were statistically significant in at least 1 high quality RCT and at least 1 lower quality RCT (PEDro score < 3) or a higher quality controlled clinical trial (CCT). The evidence was considered to be limited if the findings were statistically significant in at least one higher quality RCT (PEDro score $\leq$ 4), or at least two high quality CCTs. Study findings were determined to be indicative if findings were statistically significant findings in a single high quality CCT or low quality RCT (PEDro score < 3), or two studies of a single group with sufficient quality. Evidence was determined to be insufficient when eligible studies did not meet any of the aforementioned criteria, and/or when there were conflicting results or no eligible studies.

Methodological Quality Analysis

Out of 12 behavioral approaches that were identified for people with PD, methodological quality ratings ranged from 1 to 9 PEDro scale points. The evidence synthesis (see Table 1) provided strong evidence for LSVT LOUD only (mean PEDro score = 4.76, *SD* = 1.93). There was limited evidence for traditional speech therapy (mean PEDro score = 4.40, *SD* = 1.26, range: 3–7). All other approaches had only indicative findings or insufficient evidence to support them.

For people with MS, there was only limited evidence to support verbal cueing (mean PEDro score = 4.25, *SD* = 0.50, range: 4–5). Both EMST and LSVT LOUD had insufficient evidence to support their use in this population. In PSP, the only approach with indicative findings was singing; both LSVT LOUD and verbal cueing had insufficient evidence. Similarly, in MSA, singing showed indicative findings, but the evidence for verbal cueing was insufficient. There was insufficient evidence to support any behavioral intervention in cerebellar ataxia, ARSACS, myotonic dystrophy, Huntington's disease, Lewy body dementia, or spastic paraplegia.

Discussion

In this review, we set out to update a previous systematic review by Yorkston et al. (2003) describing behavioral interventions for respiratory/phonatory dysfunction in adults with neurodegenerative disease. In the 20 years since Yorkston et al.'s (2003) systematic review, an additional 53 published works on this topic have been added. Historically, the evidence for respiratory/phonatory rehabilitation could be categorized as biofeedback, device-driven, LSVT LOUD, or "miscellaneous" (mainly group therapy), with no RCTs reported. Today, a large range of approaches have been added to the evidence base, such as EMST, singing, and computer-driven programs, as well as a variety of treatment modalities, including teletherapy. As one example, evidence for computer- and device-driven therapies (e.g., DAF, masking noise, amplification, EMST, MAST, SpeechVive) has increased since 2003, shifting from mainly case studies/series and one single-subject report to 11 well-designed quasi-experimental studies and one single-subject–design study. In addition, evidence for treatment in several different population groups—including cerebellar ataxia, myotonic dystrophy, ARSACS, Huntington's disease, MSA, Lewy body dementia, and spastic paraplegia—were added to the current review.

Our goal in completing this review was to examine the quality of evidence for behavioral treatments for respiratory/phonatory dysfunction, as opposed to assessing the effectiveness of such treatments. Regarding evidence quality, there was strong evidence in support of only one behavioral intervention: LSVT LOUD in people with PD (see Table 1). It is noteworthy that no other treatment approach or population included in this review demonstrated more than limited evidence. It is important to recognize that a treatment with strong evidence does not necessarily reflect the most effective or most appropriate treatment option; rather, it reflects the approach that has undergone the most scientific examination. Evidence for a treatment in a particular group does not imply that it will always be effective for individuals in that group or that it will be effective for people with dysarthria associated with a different diagnosis than studied. Likewise, treatments with insufficient or limited evidence are not necessarily ineffective; rather, further scientific enquiry is necessary before robust conclusions about treatment effectiveness can be drawn. We hope that, by highlighting the absence of strong evidence for respiratory/phonatory interventions within neurodegenerative populations, future research efforts to examine their efficacy will be stimulated.

Where does this leave clinicians? Decisions about treatment selection, including evidence- versus theory-based practice, are outside of the scope of this review; however, readers are encouraged to utilize the principles of EBP (Dollaghan, 2007; Tonelli et al., 2012)—including clinical expertise, pathophysiologic reasoning, consideration of environment and system constraints, and an informed patient's preferences—to guide their efforts in

Table 1. Summary of evidence quality for behavioral interventions for respiratory/phonatory dysfunction in neurodegenerative disease.

Population	Intervention	Evidence quality
Parkinson's disease	LSVT LOUD/LSVT-X/LSVT Companion	Strong evidence
	SPEAK OUT! and The LOUD Crowd	Indicative findings
	Loud and Proud	Insufficient evidence
	Pitch Limiting Voice Treatment	Insufficient evidence
	Music therapy/singing	Indicative findings
	Verbal cueing/articulation therapy/speech therapy	Limited evidence
	Biofeedback	Indicative findings
	Masking noise/SpeechVive	Indicative findings
	Voice therapy	Indicative findings
	Delayed auditory feedback/frequency-shifted feedback	Insufficient evidence
	Expiratory muscle strength training	Insufficient evidence
	Group dynamics/coaching strategies	Indicative findings
Progressive supranuclear palsy	LSVT LOUD	Insufficient evidence
	Verbal cueing	Insufficient evidence
	Singing	Indicative findings
Multiple system atrophy	Verbal cueing	Insufficient evidence
	Singing	Indicative findings
ARSACS	Melbourne Ataxia Speech Treatment	Insufficient evidence
Cerebellar ataxia	LSVT-X	Insufficient evidence
Huntington's disease	Speech therapy	Insufficient evidence
Lewy body dementia	Singing	Insufficient evidence
Multiple sclerosis	Verbal cueing	Limited evidence
	Expiratory muscle strength training	Insufficient evidence
	LSVT LOUD	Insufficient evidence
Myotonic dystrophy	Speech warming-up exercises	Insufficient evidence

Note. LSVT LOUD = Lee Silverman Voice Treatment LOUD; LSVT-X = Lee Silverman Voice Treatment–Extended Version; ARSACS = autosomal recessive spastic ataxia of Charlevoix–Saguenay.

the absence of strong published evidence. Readers are directed to Tonelli et al. (2012) for a framework to aid the understanding of the role of research in clinical practice, as well as practical tools for clinical decision making.

This review has highlighted several areas where research in this field of dysarthria management could be significantly strengthened. A lack of robust, high-quality studies informing treatment of respiratory/phonological deficits in neurodegenerative dysarthria is evident. Of the 88 studies included in the current review, only five were RCTs (four of these were related to LSVT LOUD, and one was related to verbal cueing). The benefit of RCTs lies in the reduction of bias and, through rigorous experimental control, the ability to examine cause–effect relationships between treatments and outcomes. They are unique from any other experimental designs and considered to be the gold standard for driving meaningful change in practice (Hariton & Locascio, 2018; Tarnow-Mordi et al., 2017). It is acknowledged that RCTs require considerable resources and, in cases of rare disorders, are not always feasible to conduct. Furthermore, we understand the need to begin studying the effects of treatments using quasi-experimental designs to determine the potential impact of an expensive RCT. In response, we suggest that researchers consider employing alternative designs such as the (multiple) single-subject design (Tate et al., 2014, 2016). Single-subject research offers a very high level of experimental control and is particularly suited to rare/unique patient populations.

Also related to study quality was a lack of standardization in outcome measurement (due, in part, to a lack of standardized tools available), resulting in an overreliance on subjective measures of intelligibility and speech naturalness. When studies use unique/unvalidated tools or operational definitions for outcome measurement, it is difficult to compare findings across studies and adequately measure treatment effectiveness. We support the suggestion made 20 years ago by Yorkston et al. (2003) that a comprehensive set of measures with demonstrated psychometric properties be developed, with the goal of improving the ability to measure treatment effectiveness. We further recommend that researchers comply to the minimal data set recommendations and measures provided for disorders at the National Institutes of Health (like those for PD and amyotrophic lateral sclerosis [ALS] advocated for by the National Institute of Neurological Disorders and Stroke).

Furthermore, some of the outcomes reported in the studies reviewed here are not reliable or valid measures of speech production. For example, the validity of MPT as an index of respiratory or laryngeal function has not been established, despite its wide use. Furthermore, MPT is highly impacted by effort and cueing, and most studies do not detail how the task is completed. Additionally, jitter and shimmer have been shown to be less reliable than CPP due to issues of identifying vibratory periods in dysphonic voices (Patel et al., 2018). We encourage researchers and clinicians to choose valid and reliable measures that have been shown to relate to respiratory and laryngeal function when indexing changes to function during speech production. There is also substantial work to be done in the development/validation of outcome measures in non-English speakers. As an example, a recent literature search by one of the authors (D.B.) revealed no validated tools for measuring intelligibility in Spanish speakers.

Measures of treatment effectiveness, such as effect sizes, were noticeably missing from most included studies. Of the 88 studies included in this review, only 10 included effect size estimates. Including effect size estimates helps to determine whether a statistically significant effect is clinically meaningful, informs sample size calculations for future studies, and facilitates comparison between different studies (Aarts et al., 2014).

Effect size estimates are one aspect of best practice guidelines for scientific research that were not consistently followed in the studies included in this review. Other concerns about study reporting were noted with regard to recruitment methods, participant characteristics (e.g., dysarthria subtype, medical diagnosis), inclusion/exclusion criteria, and attrition (including number of patients screened to obtain sample). This may be due, in part, to overly restrictive word limits for scientific journals or oversights on behalf of the authors. Importantly, these reporting details are critical elements of scientific communication for treatment studies. They provide data to understand the application of research findings to clinical practice (e.g., Is a given treatment effective for people with a given dysarthria subtype?) and predicting treatment success/failure (e.g., Is attrition higher in those with dyskinesias, reduced cognition, or more severe disease?), to name but a few. One way to ensure such details are included is for authors to use the CONSORT flow diagram (Butcher et al., 2022; Schultz, 2010)—or, for non-RCTs, the TREND statement (Des Jarlais, 2014)—to describe participant flow through studies.

As a result of the lack of clear data regarding participant flow through the study, it was hard to tell in some cases whether intent-to-treat principles were followed, and in others, it was clear that intent-to-treat was not followed. This is a substantial weakness in the treatment efficacy literature reviewed here. Intent-to-treat principles ensure that every participant who was allocated to a treatment is included in the analysis, using the last data point obtained from participants who leave the study at any point. This is critical since there may be systematic differences between participants who complete the study and those who do not. Only including people who complete the treatment paradigm results in an overestimate of the treatment effects.

Fewer than half of the studies included in this review described PROs. PROs provide information that is unique from standard clinical measures and reflect the value of an intervention to patients. While some tools—such as the VHI (Jacobson et al., 1997)—specifically focus on voice-related outcomes, there are also tools to measure the global communicative impact of interventions with a focus beyond *impairment* as per the International Classification of Functioning, Disability and Health, such as the Communicative Participation Item Bank (Baylor et al., 2013). Global tools such as this may make a valuable addition to respiratory/phonatory intervention trials, to ensure that the impact of interventions is comprehensively examined, including from the patients' perspectives.

There is currently a lack of diversity in the patient populations served by the current literature. This review reflects research completed in predominantly native English speakers, with a disproportionate number of studies completed in people with PD and/or hypokinetic dysarthria, and dysarthria severity skewed toward the mild end of the spectrum. Part of the reason for this imbalance, as mentioned above, is the current lack of validated tools for measuring dysarthria outcomes in nonnative English speakers. This may also reflect a lack of diversity within the field, where native language listeners are needed to make ratings of native language speakers.

Another issue is participant recruitment, particularly for disorders that are rare, or for which increased dysarthria severity is related to a decrease in other areas—such as cognition and mobility—that become a barrier to research participation. Patient populations notably missing from this review include various types of motor neuron diseases (including ALS and primary lateral sclerosis), myasthenia gravis, various types of muscular dystrophy, and postpolio syndrome. Some of these, such as ALS, are represented in the literature with approaches such as AAC. However, recent evidence across both the limb and bulbar literature suggests that mild–moderate intensity exercise, undertaken in the early stages of the disease, may have beneficial effects (Park et al., 2020; Plowman et al., 2016, 2019). It is therefore worthwhile including populations such as this in future research efforts.

An additional issue was the infrequency with which participant characteristics were reported. Reporting of participants' sex, age, or dysarthria type was infrequent, despite growing evidence supporting the idea that neurological diseases are experienced differently between the biological sexes, for example, Cerri et al. (2019). This was recently highlighted by the Parkinson's Foundation in the United States, which created a national agenda to identify research and management practices that better support the needs of women. We hope that, by highlighting the lack of heterogeneity in the research supporting the field of neurogenic dysarthria, future researchers might be encouraged to make efforts to contribute to a more diverse—including culturally and ethnically diverse—evidence base and develop tailored interventions that meet the distinct requirements of the patient populations we serve.

Based on the findings of this review, we have several recommendations for future treatment efficacy research in this field. First, it is important that replication of studies takes place by research groups unaffiliated with the design of the treatment paradigm. This not only increases the validity of a treatment but also determines if treatment effects are still possible in the hands of other clinicians. This has started to happen with the LSVT LOUD treatment, but many other published treatments in the field are supported by only a single scientific study. As noted by Yorkston et al. (2003), partial replication studies are also immensely valuable, particularly when they seek to better define treatment parameters such as optimal timing, treatment dosage, termination, and the usefulness of prophylactic therapy. Furthermore, implementation studies are almost nonexistent in the dysarthria literature, so knowledge about the translation, feasibility, and impact of treatments in clinical practice is limited.

Second, there is an urgent need for research documenting the economics of speech rehabilitation. In the context of continuing funding cuts, this information will be crucial for guiding resource allocation decisions. Some examples of economic analyses that are needed are device-driven versus clinician-led therapy, individual versus group therapy, low- versus high-intensity therapy, in-person therapy versus telehealth, and comparing different treatment doses. Related to this is the potential refinement of existing treatment parameters. For example, are all elements of LSVT LOUD required for the treatment to be successful, or is there potential to systematically evaluate each component of the treatment based on principles of motor learning (Kleim & Jones, 2008)? As noted by Yorkston et al. (2003), do these principles apply to other dysarthrias/populations? These questions warrant further, systematic investigation.

Third, little is known about changes to respiration as a result of dysarthria treatment. Only a few studies have been conducted using gold-standard measures of respiration including respiratory kinematics, pausing characteristics, spirometry, and respiratory strength. The respiratory system is critical to the development of pressure for speech, and inefficiency in the respiratory system can impact effort, vocal intensity, and naturalness. More studies need to directly examine the function of the respiratory subsystem to ensure that our treatments do not exacerbate fatigue.

This review is not without limitations. We acknowledge that use of the PEDro scale in the context of very few RCTs is not ideal, as it was impossible for some well-designed studies to satisfy all scale items. It is also worth acknowledging that high PEDro scores do not necessarily mean that a given treatment is clinically useful or cost-effective. However, to our knowledge, a well-established alternative scale for evaluating treatment evidence has not yet been developed. Due to sparse and inconsistent reporting, study designs and methodologies were not always clear, leading to a relatively large number of initial discrepancies between our raters regarding inclusion/exclusion criteria. Furthermore, most of the studies included in this review used null hypothesis significance testing (i.e., *p* values) as the only measure of treatment effectiveness. Without information regarding statistical power or effect size estimates, we acknowledge that our analysis of these studies is flawed. In addition, owing to the large number of articles, this review excluded those focused on nondegenerative populations and populations requiring tracheostomy and/or any form of mechanical ventilation. There is a need for future work to investigate the state of the evidence base regarding these groups. Finally, as this review was limited to studies published in English, we cannot rule out the possibility that treatment evidence published in other languages was missed from this review.

In summary, this literature review reflects the expanding literature on the effects of treatment on respiratory/phonatory function in neurodegenerative diseases. The largest number of studies examined LSVT LOUD, potentially due to its long-standing presence in the field. It is important to remember that a large literature does not mean a treatment is effective for all people with a particular disorder or that a treatment will work with another disorder. Several reporting and methodological weaknesses were identified in the literature including lack of reporting of participant characteristics and flow through the study, lack of consistent outcomes, little to no research on disorders other than PD, a lack of RCTs and strong multiple single-subject designs, and a lack of reporting of effect size estimates. It is recommended that clinicians consider the research evidence and the physiologic impact of the disorder, along with clinical experience and patient preferences, following evidence-based practice guidelines in making treatment decisions.

Data Availability Statement

All data generated or analyzed during this study are included in this published article and supplemental materials.

Author Contributions

Sarah E. Perry: Formal analysis (Lead), Investigation (Lead), Validation (Lead), Visualization, Writing – original draft (Lead), Writing – reviewing & editing (Lead). **Michelle Troche:** Conceptualization (Lead), Data curation (Lead), Formal analysis (Lead), Investigation (Lead), Methodology (Lead), Project administration (Lead), Resources (Lead), Software (Lead), Supervision (Lead), Validation (Lead), Visualization (Lead), Writing – reviewing & editing (Lead). **Jessica E. Huber:** Conceptualization (Lead), Data curation (Lead), Formal analysis (Lead), Investigation (Lead), Methodology (Lead), Project administration (Lead), Resources, Supervision (Lead), Validation (Lead), Visualization (Lead), Writing – original draft (Lead), Writing – reviewing & editing (Lead). **James Curtis:** Data curation (Supporting), Investigation (Supporting), Software (Supporting), Writing – reviewing & editing (Supporting). **Brianna Kiefer:** Investigation (Supporting). **Jordanna Sevitz:** Investigation (Supporting), Writing – reviewing & editing (Supporting). **Qiana Dennard:** Data curation (Supporting), Investigation (Supporting). **James Borders:** Investigation (Supporting), Writing – reviewing & editing (Supporting). **Jillian River Browy:** Data curation (Supporting), Investigation (Supporting). **Avery Dakin:** Investigation (Supporting), Writing – reviewing & editing (Supporting). **Victoria Gonzalez:** Investigation (Supporting). **Julianna Chapman:** Investigation (Supporting). **Tiffany Wu:** Investigation (Supporting). **Lily Katz:** Investigation (Supporting). **Deanna Britton:** Conceptualization (Lead), Data curation (Lead), Formal analysis (Lead), Investigation (Lead), Methodology (Lead), Project administration (Lead), Resources (Lead), Supervision (Lead), Validation (Lead), Writing – reviewing & editing (Lead).

Acknowledgments

This work was organized through the Dysarthria Working Group, which is part of the Evidence-Based Clinical Research Committee of the Academy of Neurologic Communication Disorders and Sciences. The authors wish to thank Ann Alvar, Jake Cahn, Julie Bergman, Andrew Exner, Mehak Noorani, Eve Ngo, and McKayla Schloemer for their assistance in screening and/or reviewing articles for this systematic review.

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