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Emma Finch, Anna F. Rumbach & Stacie Park

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REVIEW ARTICLE

Speech pathology management of non-progressive dysarthria: a systematic review of the literature

Emma Finch[a,b,c], Anna F. Rumbach[a] and Stacie Park[a]

[a]School of Health and Rehabilitation Sciences, The University of Queensland, Brisbane, Australia; [b]Centre for Functioning and Health Research (CFAHR), Metro South Hospital and Health Service, Brisbane, Australia; [c]Department of Speech Pathology, Princess Alexandra Hospital, Brisbane, Australia

ABSTRACT

Purpose: The purpose of this study is to conduct a systematic review of interventions for the treatment of non-progressive dysarthria in adults.

Materials and methods: Five electronic databases (PubMed, Embase, CINAHL, PSYCINFO, Cochrane Collaboration) were searched for all studies that described and evaluated treatment used for non-progressive dysarthria in adults. Studies were included if (1) participants were adults (18+ years) with a confirmed diagnosis of non-progressive dysarthria, (2) participants received intervention with pre-post outcome data, and (3) the article was published between 2006 and 2017 (including early online publications). Data extracted included the number of participants; etiology; dysarthria type and severity; age; gender; presence of a control group; intervention tasks, frequency and duration; outcome measures; and conclusions. Data extraction was completed by a member of the research team independently and cross-checked by another team member.

Results: Of the 6728 articles identified, 21 met the inclusion criteria. The predominant study design was a case study or case series. The methodological quality of the studies varied. Typically, the interventions included impairment-based and activity level tasks targeting conversation. Approximately half of the interventions adhered to a treatment manual.

Conclusions: The evidence base to guide treatment for non-progressive dysarthria is increasing, with interventions showing promise in results, participant numbers, and positive participant feedback.

➤ IMPLICATIONS FOR REHABILITATION

- The evidence base to guide treatment for non-progressive dysarthria is increasing, but remains limited.
- The majority of evidence is of moderate methodological quality.
- The emergence of new research indicates that health professionals need to be continuously aware and critically appraise new literature in the area.

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Dysarthria; non-progressive; speech pathology; stroke; traumatic brain injury; intervention; management

Introduction

Dysarthria is a motor speech disorder characterized by weakness, spasticity, incoordination, and/or imprecision of the speech musculature and may involve respiratory, phonatory, resonatory, articulatory, and prosodic aspects of speech production [1,2]. Dysarthria is considered to be the most commonly acquired primary communication disorder, representing 53% of diagnosed acquired neurogenic communication disorders in clinical practice [1]. A large proportion of research to date has focused on dysarthria as a symptom of progressive disorders (e.g., Parkinson's disease) and its treatment, while dysarthria resulting from non-progressive disease or injury of the nervous system has received less specific attention. Estimates of dysarthria following traumatic brain injury (TBI) range from 10% to 60% [3–5], while estimates of dysarthria following stroke range from 4.5% to 69.5% of people who have a stroke [6,7].

In terms of the management of non-progressive dysarthria, a recent survey of speech-language pathologists in the Republic of Ireland reported that approaches to dysarthria management varied greatly, a finding that the authors attributed in part to the limited research evidence available to guide clinicians [8]. Earlier this year, the Cochrane Database of Systematic Reviews published a systematic review of interventions for dysarthria due to stroke and other adult-acquired, non-progressive brain injuries [9]. A search of the literature up to May 2016 identified five Randomized Control Trials (RCTs) that met the criteria for the study. This result represents an improvement from the earlier review published by the Cochrane Library on the topic of non-progressive dysarthria management which found no studies that met the review inclusion criteria of being a RCT [10]. Meta-analyses in the 2017 review failed to identify any significant changes on the majority of outcome measures, including the primary outcome measure of persistent improvement in everyday

CONTACT Emma Finch e.finch@uq.edu.au School of Health and Rehabilitation Sciences, Building 84A, The University of Queensland, St. Lucia,Brisbane, Australia

Supplemental data for this article can be accessed here.

speech relative to a control [9]. Although there was a statistically significant immediate effect of intervention at an impairment level (secondary measure), the authors cautioned that there was insufficient evidence to determine whether intervention was any better than general support or no treatment [9].

The dearth of adequately powered RCTs means that speech-language pathologists may have to rely on the results of less rigorous study designs to guide their clinical decision making around the selection of traditional dysarthria treatment techniques. As a result, there is a need to examine the current state of evidence in the treatment of non-progressive dysarthria that encompasses all types of study designs. The inclusion of all study designs rather than including only RCTs in a systematic review into dysarthria management is particularly important given that Yorkston and Baylor [11] suggested that the high level of heterogeneity in people with dysarthria renders RCTs less than ideal due to a number of factors. These factors include difficulty recruiting participants with identical dysarthria profiles, questions over the suitability of using only a single intervention technique with all participants in a trial despite heterogeneous dysarthria profiles, and limited ability to apply the results of RCT interventions to real-life clinical situations as patients may present with different dysarthria profiles to participants in the RCT [11]. Furthermore, the typical progression in the research process is to conduct a small proof-of-principal study at the basic science and exploratory level followed by a RCT [12]. It would, therefore, be expected that there would be a greater number of small proof-of-principal studies exploring different dysarthria interventions, only some of which may have led to subsequent RCTs. Despite the potential clinical utility of conducting a systematic review that includes case series and small cohort trials, few attempts to review these data has been made.

In 2007, Palmer and Enderby published a systematic review including a greater range of study designs. The authors noted that of the 23 articles included in the review, the majority of studies had small participant numbers and addressed widely varying treatment techniques [13]. Consequently, the authors concluded that it was not possible to draw conclusions about the efficacy of the different treatment techniques. It is also worth noting that the authors did not evaluate the methodological quality of the studies, so, therefore, information was unavailable to moderate the findings based on the rigor of the included studies [14].

A more recent review paper by Mackenzie [15] explored the current state of treatment approaches for dysarthria in stroke between January 2007 and May 2010 and yet again concluded that there was a low level of evidence to guide dysarthria treatment after stroke. However, the project involved only a narrative review and did not evaluate methodological quality. It is also unknown whether a similar evidence base exists for non-progressive dysarthria stemming from etiologies other than stroke.

The lack of an evaluation of methodological quality of the papers included in the reviews by Palmer and Enderby [13] and Mackenzie [15] limits the ability of clinicians to apply these findings to the clinical decision making process, thus limiting the contribution of these reviews to evidence-based practice. Therefore, the present investigation sought to determine the current evidence for specific dysarthria treatment techniques and the methodological quality of studies examining the techniques with all study designs. Specifically, the review sought to answer the question "What is the current evidence base to guide interventions for patients with non-progressive dysarthria?" The overarching goal of our systematic review was to guide clinical practice and determine directions for future research.

Methods

Protocol and registration

The protocol for this systematic review was registered prospectively with PROSPERO (CRD42016038546). This systematic review was conducted in accordance with PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) guidelines [16].

Search strategy

Five electronic databases (PubMed, Embase, CINAHL, PSYCINFO and the Cochrane Collaboration) were searched in May 2016 and January 2018 using the terms (dysarthria) AND (manag* OR treat* OR exercis* OR therapy) to identify potentially relevant studies. Search filters were used to limit the results to those that were human studies that used adult participants, written in English, published between 2006 and 2017 (including early online publications), and accessible to members of the public. Reference lists of included publications were searched for additional literature.

Selection criteria

Studies were included if (1) participants were adults (18+ years) with a confirmed diagnosis of non-progressive dysarthria, (2) participants received intervention with pre-post outcome data, and (3) the article was published between 2006 and 2017 (including early online publications). The following articles were excluded (1) editorials and review articles, (2) studies involving children and adolescents, (3) studies reporting outcomes in the treatment of progressive dysarthria, (4) studies reporting data pertaining to mixed cohorts (i.e., participants with non-progressive and progressive dysarthria reported together), and (5) studies that addressed stimulation (i.e., the technique was used in a single session and required an immediate response) rather than treatment (i.e., repeated use of the technique over more than one session).

Identification of studies

The electronic database searches uncovered 6728 articles which were imported into Endnote. Hand searches of the reference lists of suitable articles led to the inclusion of one additional article (see Figure 1). Duplicate articles were excluded using Endnote, giving a total of 6728 articles which were imported into Covidence systematic review software, Veritas Health Innovation, Melbourne, Australia (available at www.covidence.org) for review by the authors. Two review authors (E. F. and A. R.) independently screened for suitability based on title and abstract. In the case of any disagreement between the two authors, the third review author (S. P.) made a decision regarding the suitability of the article for inclusion. Screening yielded 23 articles for full-text review. The authors independently reviewed the full text articles against the defined inclusion and exclusion criteria. Following the review process, 21 articles were deemed to have satisfactorily met the inclusion criteria and were included in the data extraction and methodological appraisal stages. Two articles were excluded from the earlier 23 articles due to (1) a mixed dysarthria etiology (and being unable to separate the non-progressive dysarthria patients from the progressive dysarthria patients) and (2) a qualitative study design reporting participant perceptions of a treatment program rather than quantitative outcome measures.

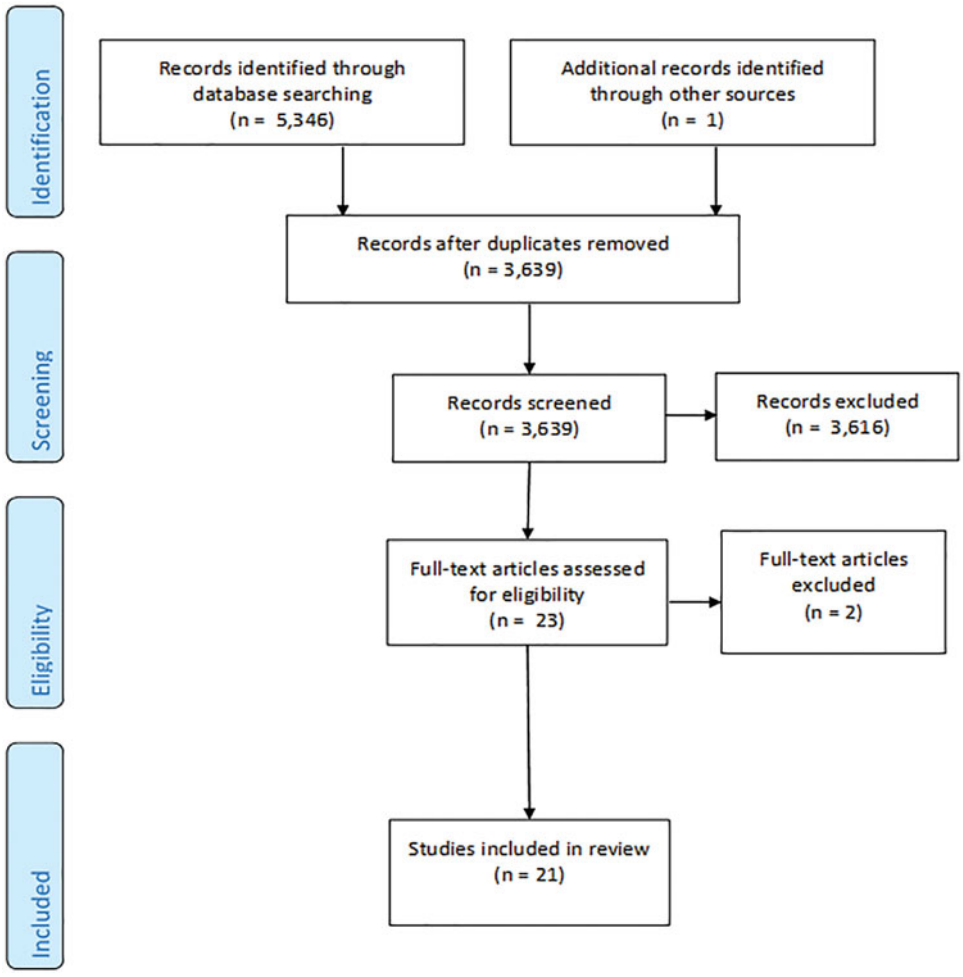

Figure 1. Study selection flow chart.

Data extraction

Data extraction was managed with Microsoft Office Excel. For each study, the following data (if available) were extracted to aid in critical appraisal: number of participants; etiology; dysarthria type and severity; age; gender; presence of a control group; intervention tasks, frequency and duration; outcome measures; and conclusions. Data extraction was completed by a member of the research team independently for each article with a second member of the team crosschecking the extracted data.

Study appraisal

Two tools were used to appraise the current evidence for management of non-progressive dysarthria: (1) National Health and Medical Research Council framework for levels of evidence [17]; (2) McMaster critical review form for quantitative studies [18]. One study included in this systematic review was authored by two of the current authors (E. F. and S. P.). For this study, A. R. completed the critical appraisal, with a fourth reviewer not involved in the research team, providing the second consensus review. Outcome measures were classified according to the World Health Organization International Classification of Functioning, Disability and Health model [19] into impairment-based (a deviation or loss in an anatomical part of the body or in a physiological function of the body, such sound pressure level) or an activity limitation (problems with performing activities, such as a rating of communicative effectiveness using equal-appearing interval scaling), or a participation restriction (difficulties with involvement in everyday life, such as communication partner ratings of the individual's everyday communication abilities).

NHMRC levels of evidence

An evaluation of research quality was conducted by two reviewers (E. R. and A. R.) using the National Health and Medical Research Council Levels of Evidence [17]. Any conflicts were resolved by discussion with the third reviewer (S. P.). The National Health and Medical Research Council Levels of Evidence hierarchy classifies studies according to the robustness of the study design, ranging from Level IV (case series) through to Level II (Randomized controlled trial, Prospective cohort study) through to Level I (Systematic review of Level II studies) [17].

McMaster critical review for quantitative studies

The McMaster University Critical Review form for quantitative studies [18] was used to assess the methodological quality of the 21 included studies. Two authors independently assessed each included article, evaluating the study design, study purpose, potential bias (stemming from participant selection, measurement and intervention domains), sample characteristics, intervention details, analysis and reporting of results, description of participant drop-outs, identification of clinical implications, and conclusions reached. With the exception of study design, responses were categorized as "yes", "no", "not addressed", or "not applicable". When evaluating the participant sample in each study, a "no" rating was

assigned to the sample being described in detail if information regarding method of recruitment was not provided. To obtain a "yes" for reliable and valid outcomes measures, the study had to include at least one measure with these psychometric properties and explicitly state this in the manuscript (where psychometric properties refer to reliability or validity). Following independent completion of ratings, the team met to obtain rating consensus, with a third member of the team available to resolve any disagreements. To allow comparison between studies, a numerical value was calculated based on McMaster critical review form scores. With the exception of the question relating to study design, items that scored "yes" were given a value of 1, while items that were scored as "no" or "not addressed" were scored as 0. Items that were not applicable ("N/A") were given a value of 1 so that the study was not penalized for items that were not relevant in the context of the study design. The maximum value a study could receive was 14.

Results

The aim of the present study was to systematically investigate the current state of evidence in the treatment of non-progressive dysarthria. The review process uncovered 21 articles in total. Five articles were RCTs [20–24], with the remaining 16 studies involving case series [17] (Supplementary Table S1). It should be noted that Wenke et al. [22–24] reported different elements of the data from a single RCT. According to the National Health and Medical Research Council levels of evidence, the majority of studies ($n = 16$) were Level IV studies which is the lowest level of evidence [17] (Supplementary Table S1). The highest level of evidence of the included studies was the five RCTs at Level II [20–24]. It is worth noting that the highest level of evidence (Level I) consists of systematic reviews, which the design of our review specifically excluded.

Participant details

The total number of participants from the studies included in this review was 274, with no one study contributing more than 14% of participants. Eight studies did not specify participants' dysarthria type (Table 1). The remaining 13 articles included participants with a range of dysarthria types (Table 1). Across the articles, the dysarthria severity varied greatly from mild to severe. Three articles did not specify dysarthria severity [20,25,26]. Overall, more male participants ($n = 122$) were included than female participants ($n = 69$). The dysarthria etiologies varied, but were primarily stemmed from a CVA or TBI (Table 1). There was a variety of ages included in the articles, ranging from 18 to 93 years. Only two studies [21,27] included adults above the age of 85 years. In contrast, six studies included very young adults at 18 or 19 years of age. Kwon et al. [20] reported age in terms of a mean ± standard deviation rather than reporting the actual age range of participants. With the exception of Mahler and Jones [28] and Palmer et al. [29] who included participants with Down syndrome and cerebral palsy, respectively, the time the intervention was administered in relation to the onset of the dysarthria varied from 26 d to 21 years post onset.

The recruitment source was not stated for 10 studies (see Table 1). Two studies recruited from medical centers [20,26], two studies recruited from health boards [21,27], three studies recruited from hospitals [30–32], one study recruited from national health service units [33], one study recruited from an outpatient medical speech-language practice [25], one study recruited from the author's client files [34], and one study reported recruiting from both a hospital and a community-based rehabilitation service [35]. Five studies explicitly excluded individuals with concomitant cognitive or other communication disorders. Four studies stated that individuals with significant other communication disorders were excluded but did not indicate whether individuals with milder forms of communication disorders were included (see Table 1). Of these studies, in the case of Wenke et al. [22–24,36] individuals with cognitive impairments were included. Two studies reported that some participants also had aphasia [21,37]. Mahler and Jones [28] reported that participants possessed sufficient cognitive linguistic abilities to participate in therapy, but did not provide further details about participants' actual communication and cognitive abilities. Jones et al. [25] reported that participants had no cognitive impairments, but did not report upon concomitant communication disorders.

Outcome measures

All of the articles reviewed used a multi-dimensional approach to the assessment of intervention outcomes. There was, however, a lack of consistency across the studies with respect to outcome measures, with no one outcome measure/s being used by the majority of studies (see Table 2). Outcome measures typically addressed an impairment level of functioning, rather than activity or participation levels (see Table 2). The most common approach for measuring treatment outcomes was at an activity level in the form of intelligibility, either through a formal assessment such the Assessment of Intelligibility in Dysarthric Speech (ASSIDS or AIDS) or listener ratings at a word or sentence level (see Table 2). The next most commonly used approach was acoustic analysis with 12 studies using at least one acoustic measure (see Table 2). Ten studies explicitly reported the inter-rater reliability of their outcome measures [21–24,29,32–34,36–38] and 12 studies reported the intra-rater reliability of their measures [22–24,29,32,34,36–39].

Intervention details

None of the included studies compared an intervention with no intervention. With respect to the interventions examined in the articles, half of the studies involved a manualized treatment program (i.e., followed a manual outlining the treatment method), including LSVT LOUD® [22–24,28,36,37,40], Living with dysarthria [27], Be Clear [35], and the La Trobe University Smooth Speech Programme [34] (see Supplementary Table S11 for further details about the intervention content). The remaining 11 articles encompassed a range of non-manualized tasks (see Supplementary Table S1). Often, but not always, the interventions included impairment-based and activity-level tasks targeting conversation. Only one study [27] included group sessions. The remaining 20 studies involved individualized treatment sessions only. Thirteen studies included home practice as part of the intervention program. Typically, this home practice occurred for approximately 10–15 min per day (see Supplementary Table S1). Seventeen studies included sufficient detail of the intervention to enable replication (see Table 1). Of the RCTs that reported using traditional dysarthria therapy techniques (using a multiple speech-subsystem approach) with a range of tasks as a control treatment, the selection of traditional dysarthria therapy techniques was typically based on the expert opinion of a group of speech-language pathologists [21–24] and in some cases a review of current literature [22–24]. Kwon et al. [20] did not specify the basis for their traditional dysarthria treatment tasks. Palmer et al. [29] used an ABAC or ACAB design to compare computer therapy using the Ortho-Logo-Paedia program to traditional dysarthria therapy. No details

Table 1. Participant details.

Study	*N*	Aetiology	Dysarthria type	Dysarthria severity	Age range	Gender	Recruitment source	Patients included with other cognitive/communication disorders?
Wenke et al. [22]	28	TBI; CVA	Varied	Mild to moderate	19–85	16 M; 10 F	N/A	Excluded significant communication disorders. Included cognitive impairments
Wenke et al. [23]	10	TBI; CVA	Varied	Mild to moderate-severe	19–85	3 F; 7 M	N/A	Excluded significant communication disorders. Included cognitive impairments
Wenke et al. [24]	26	TBI; CVA	Varied	Mild to moderate-severe	19–85	16 M; 10 F	N/A	Excluded significant communication disorders. Included cognitive impairments
Mackenzie et al. [21]	39	CVA	N/A	Mild/moderate to severe/profound	30–91	13 F; 26 M	Health boards	Some of the included participants had minimal aphasia
Kwon et al. [20]	25 recruited (20 finished)	CVA	N/A	Not specified	rTMS mean-= 69.4+11.8 Sham mean = 68.8+9.8	17 M; 3 F	Medical center	No – specifically excluded participants with other cognitive/communication disorders
Mackenzie et al. [27]	12 recruited (9 finished)	CVA	N/A	Mild to moderate	50–93	7 M; 5 F	Health boards	Excluded significant communication disorders. Included cognitive impairments.
Wenke et al. [36]	10	CVA; TBI	Varied	Mild to moderate-severe	19–80	5 F; 5 M	N/A	Excluded significant communication disorders. Included cognitive impairments.
Lee and McCann [39]	2	CVA	Flaccid	1 mild; 1 severe	61–79	1 F; 1 M	N/A	No – specifically excluded participants with other cognitive/communication disorders
Mahler and Jones [28]	2	Down syndrome	Flaccid	1 moderate; 1 mild	34; 33	1 F; 1 M	N/A	Stated that participants had sufficient cognitive linguistic abilities to participate in treatment.
Mahler and Ramig [40]	4	CVA	N/A	Mild to moderate	50–74	1 F; 3 M	N/A	N/A
Palmer et al. [29]	10 recruited (7 finished)	CVA; TBI; CP	Varied	Mild to very severe	36–80	3 M; 4 F	N/A	No – specifically excluded participants with other cognitive/communication disorders
Tamplin [32]	4	CVA; TBI	N/A	Mild to severe	19–51	3 F; 1 M	Hospital	Excluded participants if other major cognitive or communication disorders were present.
Stocks et al. [34]	1	Anoxic encephalopathy	Ataxic	Mildly unintelligible and moderately unnatural	49	F	Author's client files	N/A
McGhee et al. [31]	2	TBI	Flaccid-ataxic	1 Mild; 1 Moderate	33; 45	1 F; 1 M	Hospital	N/A
Mahler et al. [37]	2	CVA	N/A	1 Not specified; 1 Moderate	37; 74	M	N/A	Both participants had expressive aphasia
Kumar et al. [30]	1	Anoxic encephalopathy	Flaccid	Severe	26	M	Hospital	No – specifically excluded participants with other cognitive/communication disorders
Mackenzie and Lowit [33]	8	CVA	N/A	Rated from 1 (low intelligibility) to 7 (high intelligibility) – ranged from 1 to 6	47–75	2 F; 6 M	National health service units	N/A

(*continued*)

Table 1. Continued.

Study	*N*	Aetiology	Dysarthria type	Dysarthria severity	Age range	Gender	Recruitment source	Patients included with other cognitive/communication disorders?
Mackenzie and Lowit [38]	1	CVA	N/A	Severe	69	M	N/A	N/A
Kim and Jo [26]	6	CVA	Mixed dysarthria with spastic component	Not specified	52–65	2 F; 4 M	Rehabilitation medical center	N/A
Park et al. [35]	8	TBI; CVA	Varied	Mild-moderate to severe	18–51	3 F; 5 M	Hospital and community-based rehabilitation center	Excluded participants with aphasia and apraxia. Included patients with cognitive impairments, with the exception of dementia and post-traumatic amnesia
Jones et al. [25]	1	Lance-Adams syndrome and TBI	Mixed ataxic-hyperkinetic-spastic	Not specified	26	F	Outpatient medical speech-language practice	No cognitive disorders; status regarding presence of other communication disorders unknown

TBI: traumatic brain injury; CVA: cerebrovascular accident; CP: cerebral palsy; UUMN: unilateral upper motor neuron; rTMS: repetitive transcranial magnetic stimulation.

were provided about the basis of the traditional dysarthria therapy regime.

All 21 studies included a baseline and post-intervention assessment time point (see Supplementary Table S1). Eleven of the studies included a follow-up assessment time point. The timing of the follow-up assessments varied from 1 to 6 months, with just over half of the studies reporting that beneficial changes were observed in participants at the follow-up assessment time point. The total amount of therapy was highly variable between studies and ranged from 5 h to 59 h (see Supplementary Table S1). The duration of therapy ranged from 1 week to 6 months. The average therapy session length varied from 30 min to 9 h per day, while the weekly number of sessions per week ranged from 1 to 5. Some studies delivered the intervention at high intensity, such as Kim and Jo [26] who delivered therapy via 5×30 min sessions per week for 2 weeks. Other studies used a more distributed approach to intervention, such as Mackenzie and Lowit [33] and Mackenzie and Lowit [38] who used 16×45 min sessions over 8 weeks. Some researchers used both approaches such as Stocks et al. [34] who administered 9 h of therapy per day for 5 d followed by 1×2 h therapy session per week for 7 weeks. Nineteen studies reported that the intervention was delivered by a speech-language pathologist (see Supplementary Table S1). Tamplin [32] reported that the intervention was delivered by a music therapist, while Kumar et al. [30] did not specify the discipline of the treating therapist.

Nineteen studies reported a positive effect of the intervention on at least one outcome measure. A positive effect was where the intervention resulted beneficial change on at least one outcome measure. This was sometimes but not always based on statistical analysis. A single case study by Kumar et al. [30] did not report a beneficial effect, while an RCT examining Non-Speech Oromotor Exercises [21] found statistically significant gains across the intervention period but no additional benefit with the addition of Non-Speech Oro-motor Exercises. Studies without statistical analysis did not always include multiple baseline measurements prior to intervention, nor if multiple baseline measurements were included did all patients demonstrate stability. In terms of the effects of the interventions, the greatest volume of evidence existed for LSVT LOUD with three papers reporting the results of a RCT [22–24] and four Level IV case series studies [28,36,37,40] (note: three of the LSVT LOUD studies were derived from one trial). These studies suggested that the LSVT led to positive changes in areas such as intelligibility, loudness, hypernasality, vowel space area, and articulatory precision (see Supplementary Table S1). The other highest level of evidence in the form of a RCT included in the systematic review was by Mackenzie et al. [21], which found that the addition of Non-Speech Oromotor Exercises provided no additional benefit in the treatment of non-progressive dysarthria, and in a RCT by Kwon et al. [20] which found support for the use of low frequency rTMS combined with dysarthria therapy (see Supplementary Table S1). The remaining 12 studies provided Level IV evidence (that was case series designs including before-after designs) for a range of treatment techniques that were typically impairment-based (see Supplementary Table S1). Two studies favored a more activity and participation focused approach with the Be Clear program [35] targeting the use of clear speech in functional phrases and speech tasks, and the Living with Dysarthria Program [27] targeting support, education, and everyday communication. The Level IV studies reported beneficial effects for both Be Clear [35] and the Living with Dysarthria Program [27], along with beneficial effects for expiratory muscle strength training [25], accent-based music speech [26], smooth speech with reading and

Table 2. Summary of outcome measures.

Outcome measures	Wenke et al. [22]	Wenke et al. [23]	Wenke et al. [24]	Mackenzie et al. [21]	Kwon et al. [20]	Mackenzie et al. [27]	Wenke et al. [36]	Lee and McCann [39]	Mahler and Jones [28]	Mahler and Ramig [40]	Palmer et al. [29]	Tamplin [32]	Stocks et al. [34]	McGhee et al. [31]	Mahler et al. [37]	Kumar et al. [30]	Mackenzie and Lowit [33]	Mackenzie and Lowit [38]	Kim and Jo [26]	Park et al. [35]	Jones et al. [25]
Impairment-based measures																					
Perceptual rating using DME (e.g., articulatory precision, hypernasality)	X	X	X				X										X	X			
Acoustic analysis (e.g., fundamental frequency, articulation rate, vowel space area, SPL in dB, phonation time)	X	X			X		X	X	X	X					X	X		X	X		X
Expert listener judgment of articulatory precision											X										
Perceptual Speech Analysis Scale													X	X							
Westmead post-traumatic amnesia scale														X							
Motor speech examination tool														X							
Frenchay Dysarthria Assessment				X				X			X		X			X					
Mean length of utterance																		X			
Diadochokinesis (alternating and sequential motion rates)					X														X		
Urimal Test of Articulation and Phonology					X																
Activity-based measures																					
ASSIDS/AIDS			X				X	X		X										X	X
SIT						X						X									
Word Intelligibility Test																	X	X			
Listener intelligibility ratings			X	X					X			X			X					X	
Self-report questionnaire			X				X														
Communication partner questionnaire			X				X													X	
Ratings of communication effectiveness				X		X											X	X			X
Participation-based measures																					
Quality of life communication scale						X															
Dysarthria Impact Profile																	X			X	
Measures encompassing all three domains																					
AusTOMs speech scale			X				X						X								
Goal attainment scaling						X															
N/A																					
Short general health questionnaire						X															
Stroke and speech knowledge test						X															

ICF: international classification of functioning, Disability and Health (WHO, 2001); I: impairment; A: activity; P: participation; DME: direct magnitude estimation; AusTOMS: Australian Therapy Outcome Measures; AIDS or ASSIDS: Assessment of Intelligibility in Dysarthric Speech; SPL: sound pressure level; dB: decibels; SIT: sentence intelligibility test.

conversational activities [34], computer therapy [29], and diaphragmatic breathing with phonation of non-words and changes in pitch level [39].

Methodological quality

In terms of methodological ratings using the McMaster Critical Review Form for Quantitative Studies [18], no study achieved the maximum rating of 14 (Table 3). Three articles achieved ratings of 12 [21,35,38]. Interestingly, all three articles were published recently. The lowest rating [4] was obtained by a single case study by Kumar et al. [30]. The main areas of methodological strength were in the study purpose ($n = 19$), literature review ($n = 20$), drop outs (attrition bias, all studies = yes or N/A), appropriateness of analysis ($n = 19$), intervention described in detail ($n = 17$), reliability of measures ($n = 17$), clinical importance ($n = 19$), and appropriateness of the study conclusion ($n = 19$). The main areas of weakness were in the sample size justification (selection bias, $n = 3$), validity of measures ($n = 3$), and the avoidance of co-intervention (performance bias, $n = 2$; see Table 3). Approximately half of the studies ($n = 11$) provided adequate information about sample description, potentially exposing the studies to selection bias. The majority of studies did not include measures of effect size.

Discussion

The aim of the present study was to systematically investigate the current state of evidence in the treatment of non-progressive dysarthria. The present study differed from the recent Cochrane review by encompassing study designs beyond RCTs, while also differing from earlier reviews that included non-RCT studies [13,15] by including a methodological evaluation of article quality.

Consistent with previous reviews [9,13,15] the current systematic review found that there was limited evidence to guide the management of patients with non-progressive dysarthria. However, as reflected in the recent Cochrane review [9], there has been a noticeable increase in the number of studies investigating interventions for non-progressive dysarthria over recent years. This is particularly positive as it suggests that the evidence base to guide speech-language pathologists when managing patients with non-progressive dysarthria is slowly, but steadily increasing.

In the current review, 19 studies reported a positive effect of the intervention on at least one outcome measure. A single case study by Kumar et al. [30] did not report a beneficial effect, while an RCT examining Non-Speech Oromotor Exercises [21] found statistically significant gains across the intervention period but no additional benefit with the addition of Non-Speech Oro-motor Exercises. The current review revealed that the greatest evidence, both in the strength of the study design and volume of studies, exists for LSVT LOUD®. Other invention approaches had less evidence supporting their intervention approach. Specifically, for a number of techniques such as Be Clear, Smooth Speech and Expiratory Muscle Strength Training, there was only one study examining the effects of the technique. Furthermore, in the majority of articles, the studies involved Level IV evidence. This may be at least partially reflective of the research process, given that typically intervention research starts with a feasibility study that then progresses to a more rigorous RCT if the results of the feasibility study are considered promising [12]. It is possible that the predominance of Level IV evidence in the form of case series and before–after study designs is due in part to research into interventions for non-progressive dysarthria being in the relative infancy phase of the research process. Over time, it would, therefore, be expected that this would change with more interventions of the early feasibility studies progressing to RCTs. This hypothesis is supported by the change from no RCTs in the initial Cochrane review [10] to the inclusion of 5 RCTS in the more recent Cochrane review [9]. Furthermore, it has been suggested that the high level of heterogeneity in people with dysarthria renders RCTs a less than ideal study design for this patient population [11].

In terms of methodological ratings, in general, the studies included in the current review achieved moderate ratings. On a positive note, one of the highly scoring areas was in the description of the interventions, suggesting that the majority of the intervention programs were capable of being replicated by readers. The main areas of weakness were in the sample size justification, which was most likely a reflection of the Level IV study designs employed, the validity of measures, and the absence of co-intervention being stated. The inclusion of co-intervention, even in the RCTs, leaves open the potential for factors other than the targeted treatment to have produced the changes recorded with the outcome measures.

With the exception of LSVT LOUD®, the frequency and intensity of the interventions differed greatly between studies. This is consistent with Mitchell et al. [9] who reported that no studies compared the timing, dose or intensity of the treatment with the same intervention. This finding highlights the imperative need for further research into timing, dose and intensity for specific intervention approaches for non-progressive dysarthria. There did not appear to be an association between the total amount of the intervention and beneficial results in the current review as the two studies that reported limited additional benefit for their dysarthria intervention [21,30] provided a similar total amount of intervention to other studies which reported beneficial effects.

The recent systematic review by Mitchell et al. [9] concluded that there was a significant immediate effect of interventions on impairment-based measures, but no effect on immediate measures of activity or participation, or on long-term measures in any of the three domains. In the current systematic review, the most commonly used outcome measures were activity level measures in the form of speech intelligibility were also frequently used with positive changes reported by a number of studies. The frequent occurrence of intelligibility as an outcome measure, suggests that the studies strove to use an ecologically valid outcome to evaluate their interventions [41]. Interestingly, in the present review, measures at an impairment level in the form of an acoustic measure were also frequently used. The high use of acoustic measures may have reflected a desire to provide an objective measure of changes in speech function. However, several studies did not use outcome measures with published validity and reliability data. In the case of listener perception intelligibility ratings, the ratings may have also been susceptible to rater bias. In particular, Walshe et al. [42] suggested that listener perceptual judgments of intelligibility may be influenced by a range of factors including acceptability, bizarreness, and naturalness.

It is possible that the lack of consistency in the use of outcome measures between the studies may have influenced the different findings between the studies. As such, there is a need for a consistent set of outcome measures for use in non-progressive dysarthria research. It is also worth noting that improvements between individuals varied within studies. It is possible that not all intervention approaches work equally for all patients. It is also possible that the presence of co-morbidities, in particular aphasia and cognitive impairments, may have at least partially contribute to this observation.

Table 3. McMaster University Critical Review Form for quantitative studies ratings.

	Purpose	Literature	Sample description (selection bias)	Sample size justification (selection bias)	Drop-outs (attrition bias)	Measures reliable	Measures valid	Intervention described in detail	Contamination avoided	Co-intervention avoided (performance bias)	Statistical significance	Analysis appropriate	Clinical importance	Conclusions	SCORE (/14)
Level II studies (randomized controlled trials)															
Wenke et al. [22]	●	●	○	○	●	●	○	●	○	○	●	●	●	●	9
Wenke et al. [23]	●	●	○	○	●	●	○	○	●	○	○	●	●	●	8
Wenke et al. [24]	●	●	○	○	●	●	○	●	○	○	●	●	●	●	9
Mackenzie et al. [21]	●	●	●	●	●	●	●	●	○	○	●	●	●	●	12
Kwon et al. [20]	●	●	●	○	●	○	○	○	○	○	●	●	●	●	9
Level IV studies (case series)															
Mackenzie et al. [27]	●	●	●	○	●	○	○	●	N/A	○	●	●	●	●	10
Wenke et al. [36]	●	●	○	○	●	●	○	●	N/A	○	●	●	●	●	10
Lee and McCann [39]	●	●	○	○	●	●	○	●	N/A	○	●	○	●	●	9
Mahler and Jones [28]	●	●	○	○	●	●	○	●	N/A	○	●	●	●	●	10
Mahler and Ramig [40]	●	●	○	○	●	●	○	●	●	●	●	●	●	●	11
Palmer et al. [29]	●	●	●	○	●	●	○	○	N/A	○	●	●	●	●	10
Tamplin [32]	○	●	●	○	●	●	○	○	N/A	○	●	●	●	●	9
Stocks et al. [34]	●	●	●	○	●	●	○	●	N/A	○	○	●	●	●	10
McGhee et al. [31]	●	●	●	○	●	●	○	●	N/A	○	○	●	●	●	10
Mahler et al. [37]	●	●	○	●	●	●	○	●	N/A	○	○	●	●	●	10
Kumar et al. [30]	○	○	●	○	●	○	○	●	N/A	○	○	○	○	○	4
Mackenzie and Lowit [33]	●	●	○	○	●	●	○	●	N/A	●	●	●	●	●	11
Mackenzie and Lowit [38]	●	●	●	●	●	●	○	●	●	○	●	●	●	●	12
Kim and Jo [26]	●	●	○	○	●	○	○	●	N/A	○	●	●	○	○	7
Park et al. [35]	●	●	●	○	●	●	●	●	N/A	○	●	●	●	●	12
Jones et al. [25]	●	●	●	○	●	●	●	●	N/A	○	○	●	●	●	11

●: yes; ○: no; N/A: not applicable.

In the current systematic review, the majority of dysarthria etiologies were CVA or TBI, suggesting in aggregate that there is evidence to indicate that behavioral interventions can produce beneficial improvements in patients with non-progressive dysarthria stemming from CVA or TBI. However, the highly heterogeneous nature of the participants across studies in terms of dysarthria severity, subtype, and chronicity meant that it was not possible to determine whether a specific intervention approach was better suited for a particular dysarthria profile. Additionally, there were a variety of ages included in the articles, ranging from 18 to 93 years, with only two studies [21,27] including adults above the age of 85 years. This lack of information about the effects of dysarthria interventions on older adults suggests that further research is required.

Directions for future research

The systematic review uncovered a number of areas requiring future research. Most notably, further research utilizing more rigorous study designs (including RCTs and single case experimental designs with multiple baselines) to evaluate the short and long-term efficacy of specific dysarthria treatment techniques is required to aid speech-language pathologists in providing evidence-based practice in the management of non-progressive dysarthria. Questions also remain unanswered about whether individual or group therapy is the more effective method of delivering therapy. Furthermore, studies exploring optimal treatment intensity and duration are required. There is also a need for a minimum standard of reliable and valid outcome measures for evaluating dysarthria treatment effects to be established and utilized in both clinical practice and in the researching of new treatment approaches. Activity and participation level outcome measures should be incorporated alongside impairment-based measures to ensure the effects of interventions across all ICF domains can be adequately recorded.

Study limitations

The results of the current systematic review were hindered by a number of limitations. Despite the authors' best efforts to conduct thorough searches, there is the potential that some relevant articles were inadvertently missed. The authors were not blinded to the authors of each study due to all three authors being involved in database searching, data extraction and critical appraisals. The heterogeneity of study populations and methodology, alongside lack of sufficient data, used meant that meta-analysis could not be conducted. It is also worth noting that the majority of studies did not include measures of effect size, which hindered comparison of the results across studies. On a final note, as per the recent Cochrane review, our current review focused on non-progressive dysarthria as a group rather than specific etiologies.

Conclusion

The majority of studies in the current systematic review reported that the interventions produced beneficial effects on at least one outcome measure, typically at an impairment or activity level. While the evidence base to guide treatment for non-progressive dysarthria is increasing, there is currently only a limited body of evidence to guide therapy, particularly when it comes to the use of specific treatment techniques. Furthermore, the majority of evidence is of moderate methodological quality with only Level IV study designs. The emergence of new research over recent years since earlier systematic reviews highlights the need for speech-language pathologists to continually be aware and critically appraise new literature as it emerges in the field of non-progressive dysarthria.

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ORCID

Emma Finch http://orcid.org/0000-0002-9690-8165

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