Effects of voice therapy in children with vocal fold nodules: A systematic review

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ABSTRACT

Background: Vocal fold nodules (VFNs) are the main cause of paediatric dysphonia. Voice therapy is recommended as the preferable treatment option for VFNs in children.

Aim: The aim of this systematic review is to provide an overview of the existing literature concerning the effects of voice therapy in children with VFNs.

Methods & Procedures: This systematic literature review was developed following the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) guidelines. The Cochrane Central Register of Controlled Trials (CENTRAL), MEDLINE (via PubMed) and Embase were searched and the grey literature was checked. The search strategy was based on three concepts: VFNs, voice therapy and children. Two examiners independently determined article eligibility and extracted all relevant data from the included studies. The methodological quality of the included study was assessed using the QualSyst tool.

Main contributions: By identifying, evaluating and summarizing the results of all relevant studies about voice therapy in paediatric VFNs, this systematic review makes the available evidence more accessible to voice therapists, otolaryngologists and other relevant stakeholders.

Conclusions & Implications: 24 studies were included in this systematic review. Eight studies (8/24) reported a significant improvement for at least one outcome parameter after voice therapy. However, five papers (5/24) could not demonstrate significant changes after voice therapy. All studies that did not test for significance (11/24) found improvements for one or more outcome parameters. The overall quality of the included studies is adequate (55%). In sum, there is some evidence that voice therapy is effective in children with VFNs, but further well-designed research, especially randomized controlled trials, is necessary to confirm these results.

WHAT THIS PAPER ADDS

What is already known on the subject

- Voice therapy is preferable in children with VFNs because of the phonotraumatic nature of the nodules and the associated high recurrence rate after phonosurgery. Most voice therapists in clinical practice offer an eclectic voice therapy programme, consisting of direct and indirect voice therapy techniques.

What this study adds to existing knowledge

- This systematic review provides a clear overview of the available evidence concerning the effects of voice therapy in paediatric VFNs. There is some evidence that voice therapy is an effective treatment option in children with VFNs, but well-designed research is scarce on this subject.

What are the potential or actual clinical implications of this work?

- This review shows that effectiveness studies with strong designs are very scarce in children with VFNs. Clinicians should be aware that few therapy techniques have been thoroughly investigated in this population. However, this review may guide voice therapists when creating a treatment plan for a child with VFNs because it identifies, evaluates and summarizes the results of all relevant individual studies about voice therapy in paediatric VFNs. Voice therapy seems to be effective in treating paediatric patients with VFNs, given the fact that a considerable number of included studies report significant improvements after voice therapy. Both direct and indirect therapy approaches appear to have a positive effect on the phonation of children with VFNs.

INTRODUCTION

Vocal fold nodules (VFNs) are bilateral benign lesions of the membranous vocal folds. They usually occur at the junction between the anterior one-third and two-thirds of the vocal folds (Mudd & Noelke, 2018). VFNs are the main cause of dysphonia in children, responsible for 35–78% of all cases (Hartnick et al., 2018; Martins et al., 2012; Mudd & Noelke, 2018; Van Houtte et al., 2010). They are related to vocal abuse, which includes yelling, screaming and throat-clearing, leading to phonotrauma (Hron et al., 2019; Martins et al., 2013; Middendorf, 2007). The voice of VFNs patients is often perceived as rough, strained and breathy with a low pitch (Hron et al., 2019; Nuss et al., 2010). Paediatric dysphonia may have a negative impact on different aspects and life domains, such as children's general health, communicative effectiveness, social and educational development, self-esteem, self-image, and participation in school group activities (Connor et al., 2008; Ma et al., 2007). Especially in school-aged children and adolescents, a dysphonic voice can cause emotional stress (Connor et al., 2008).

Akif Kilic et al. (2004) stated that the prevalence of VFNs in school-aged children is 16.9%. A general, non-treatment-seeking group of children was examined using rigid

laryngoscopy. Overall, 30.2% of these children were diagnosed with minor vocal lesions or (im)mature nodules. Because the minor vocal lesion group showed similar pitch and amplitude perturbation quotients with the group without vocal lesions, children in the minor lesion group were reclassified into the no lesion group leading to a prevalence of 16.9%. It can be debated whether it is appropriate to reclassify children with minor lesions and whether the used acoustic parameters are diagnostically valid. Furthermore, it is generally noticed that VFNs are more common in boys during childhood with a male-to-female ratio of 2:1, which may be explained by the typical social and physical activities of boys, such as sportive team games, that demand a loud and intense voice use (Akif Kilic et al., 2004; Carding et al., 2006; Martins et al., 2013; Mudd & Noelke, 2018; Nagata et al., 1983). There is evidence that the prognosis of VFNs in boys is more positive compared with girls. De Bodt et al. (2007) pointed out that girls, diagnosed with VFNs before the age of 12, have a greater risk to experience voice problems after puberty as compared with boys, despite enrolling in voice therapy. Moreover, this study revealed that 47% of the post-pubescent girls could still be diagnosed with VFNs, whereas VFNs are only seen in 7% of the male participants. Also Sander (1989) asserted that VFNs tend to disappear during puberty in boys.

Several risk factors for developing VFNs are described in the literature. First, children with siblings are more likely to develop VFNs, potentially because of the noisy home environment (Tuzuner et al., 2017). A diagnosis of ADHD and an extraverted personality are other possible risk factors for the development of VFNs (D'Alatri et al., 2015; Roy et al., 2007; Verduyckt et al., 2019).Gastroesophageal reflux and laryngopharyngeal reflux, the retrograde flow of gastric contents into the oesophagus and laryngopharynx, respectively, are also associated with paediatric VFNs (Saniasiaya & Kulasegarah, 2020; Singendonk et al., 2019). Although gastroesophageal reflux is a normal physiological process, it can cause troublesome symptoms such as heartburn and regurgitation. Gastroesophageal reflux symptoms are present on a weekly basis in more than 10% of children older than 18 months (Martigne et al., 2012; Saniasiaya & Kulasegarah, 2020; Singendonk et al., 2019). In a systematic review of Saniasiaya and Kulasegarah (2020), VFNs could be found in 28% of the pooled participants with a diagnosis of reflux. Lastly, asthma and allergic diseases are also a risk factor to develop VFNs during childhood (Ercan et al., 2020).

A debated question is whether children with VFNs really need to be treated. On the one hand, it is generally recommended to treat voice disorders in children (Hooper, 2004). Powell et al. (1989) stated that one-fourth to one-third of untreated dysphonic children still show voice problems 5 years after the initial diagnosis, indicating that maturation is not sufficient to clear up voice disorders. It is noteworthy that the exact diagnosis of the participants was not known since no laryngoscopic evaluation was performed, making it unclear whether these patients had VFNs. In addition, the gender distribution of the participants was not reported in this study. Consequently, it is unclear which participants, in terms of laryngeal diagnosis or gender, experienced persistent voice problems. On the other hand, some authors cast doubt on the idea that VFNs always need to be treated. Hakansson and Kitzing (1984), who specifically focused on the long-term evolution of VFNs, claimed that 88% of children with VFNs improved spontaneously. Also Martins et al. (2013) consider the possibility of not providing treatment in paediatric patients with VFNs, based on the favourable

evolution of VFNs after adolescence in a large number of children, especially among boys.

Recently, Hron et al. (2019) adopted another point of view: treatment of VFNs in children must be guided by their vocal needs, just as in adults. Additionally, two important questions must be raised before starting voice therapy sessions in children: (1) is the child failing to meet their needs? and (2) Is the child able to cooperate with treatment? If a child is not bothered by the voice and shows no motivation to change vocal habits or no maturity to participate in voice therapy, it is not necessary to provide an intervention.

Lastly, the question can be raised whether counselling parents is sufficient and effective to treat VFNs. Only one study investigated the effects of a parent counselling programme in children with nonorganic voice disorders, among which the authors include VFNs (Kollbrunner & Seifert, 2013). During two sessions, parents were counselled on psychodynamics and family dynamics. A significant improvement in vocal quality was reported by the parents after 1 year. However, there was no objective or perceptual evaluation of the actual vocal quality of the children by a clinician and the content of the parent counselling therapy was not described. Research on the effects of parent counselling on vocal hygiene recommendations in dysphonic children is currently missing.

Treatment options for paediatric VFNs are based on adult therapies and include direct and indirect voice treatment, phonosurgery, and modulation of associated factors such as allergy or reflux (Mudd & Noelke, 2018; Nuss et al., 2010; Ongkasuwan & Friedman, 2013). Voice therapy is preferable in children because of the phonotraumatic nature of VFNs and the associated high recurrence rate after phonosurgery (Martins et al., 2013, 2020; Mudd & Noelke, 2018). Immaturity of the laryngeal structures and the frequent failure to respect postoperative vocal rest also make phonosurgery a controversial choice in children with VFNs (Martins et al., 2020). Direct voice therapy techniques modify vocal behaviour through motor execution, somatosensory feedback, or auditory feedback. Indirect therapy consider the cognitive, behavioural, and psychological aspects of voicing or change the physical environment in which voicing occur (Van Stan et al., 2015).

Recently, Feinstein and Abbott (2021) published a systematic review concerning behavioural treatment for benign vocal fold lesions in children. The scope of this review was broader and included children with vocal oedema, unspecified hoarseness, and hyperfunctional voice disorders. The authors stated that the most commonly used voice therapy was eclectic direct voice treatment, including different vocal exercises or facilitating techniques. Moreover, they concluded that behavioural voice therapy is generally effective in treating benign vocal fold lesions in children, with an advantage of direct voice therapy over indirect voice therapy. A long-term treatment was also seen to be more effective than a short intervention. It is noteworthy that the methodological quality of the included studies was not examined using a formalized quality assessment tool. However, the authors generally described some important issues regarding methodological quality of the included studies, such as poor use of statistical analyses and the lack of strong research designs.

To our knowledge, this is the first systematic review about effectiveness of voice therapy specifically for VFNs in children. The aim of this systematic review is to summarize the evidence of the effects of direct and indirect voice therapy in paediatric VFNs. An overview of the existing literature will help voice therapists in selecting appropriate vocal techniques and exercises while working with children with VFNs.

METHODS

This systematic literature review was developed following the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) guidelines (Moher et al., 2009). The recommendations from the Cochrane Collaboration were also taken into account (Higgins et al., 2018). The research protocol was published in the PROSPERO register (registration number: CRD42021232315).

The systematic literature search was conducted in three different electronic databases in order to detect studies investigating the effects of voice therapy in children with VFNs. The databases were searched on 10 February 2021, including Medline (using the PubMed interface), Embase, and Cochrane Central Register of Controlled Trials (CENTRAL). Additionally, Open Access Theses and Dissertations (oatd.org), Open Grey (opengrey.eu), and ProQuest Dissertations and Theses (proquest.com) were checked for grey literature. Web of Science was also searched to identify relevant meeting abstracts. The eligibility criteria were described using the PICO structure. For an overview of the criteria, see Table 1. No publication date restrictions were used. In order to avoid language bias, only English articles were included. Narrative and systematic literature reviews were excluded, all other study designs were considered for inclusion.

The search strategies for PubMed, Embase and CENTRAL are shown in Tables 2–4, respectively. The Boolean operator AND was used between three different concepts, namely ‘vocal nodules’, ‘voice therapy’, and ‘children’. In PubMed and CENTRAL, relevant MeSH terms (Medical Subject Headings) were identified and the corresponding Emtree terms were selected in Embase. All MeSH terms (or Emtree terms) and free text words were combined with the Boolean operator OR. No filters or search blocks were used. The search strategy was checked by an information specialist of the faculty of Medicine and Health Sciences of Ghent University (NP).

All references collected by the screening process were saved in a first EndNote X9 (Clarivate Analytics) library. Afterwards, three other libraries were created: (1) records after removing reduplicated articles, (2) included records based on title and abstract screening, and (3) included records based on full-text screening.

Following the initial search, duplicates of references were automatically detected in EndNote X9 (Clarivate Analytics). The highlighted records were compared and one was manually removed in case they were identical. Afterwards, the full list of references was checked by hand for remaining duplicates. The whole screening procedure was accomplished by two independent researchers (first author AA and IM). Title-abstract and full-text screening were performed using Rayyan (Ouzzani et al., 2016). The authors determined whether the articles were appropriate according to the eligibility criteria (Table 1). Any discrepancies after full-text screening were discussed and a third independent researcher (ED) was asked to reach a decision in

TABLE 1 Eligibility criteria

	Inclusion criteria	Exclusion criteria
Population	Children (0–18 years at the time of diagnosis VFNs) All ethnicities Diagnosis of VFNs: •At least 50% of the study group has a diagnosis of VFNs OR •The results of participants with VFNs are shown separately	Comorbidities: •Severe neurological disorders •Hearing loss •Mental retardation
Intervention	Direct or indirect voice therapy	Only surgical intervention Only psychological intervention
Comparison	Control group without intervention Sham therapy Another variant of voice therapy Surgical intervention No control group	
Outcome		
	Objective vocal quality measures	
	Aerodynamic and acoustic voice parameters	
	Assessment of laryngeal anatomy and function using imaging techniques	
	Perceptual voice assessments	
	Psychosocial well-being	
	Patient's or parental perception of the voice	

TABLE 2 Search strategy for MEDLINE (PubMed)

Research question: What are the effects of voice therapy in children with vocal fold nodules in terms of objective and subjective vocal outcome measures?		
DATABASE: PubMed		
Concept	**Line**	**Search strategy**
Concept 1: Vocal fold nodules	1	**MeSH terms**: 'Vocal Cords'[Mesh] OR 'Laryngeal Diseases'[Mesh] OR **Free text words**: 'vocal nod*'[TIAB] OR 'voice nod*'[TIAB] OR 'vocal fold nod*'[TIAB] OR 'vocal cord nod*'[TIAB] OR 'singing nod*'[TIAB] OR 'singer's nod*'[TIAB] OR 'speaking nod*'[TIAB] OR 'speaker's nod*'[TIAB] OR 'screaming nod*'[TIAB] OR 'screamer's nod*'[TIAB] OR 'nodulus vocalis'[TIAB] OR 'noduli vocalis'[TIAB] OR 'nodulus plica vocalis'[TIAB] OR 'noduli plica vocalis'[TIAB] OR 'nodul*'[TIAB] OR 'dysphon*'[TIAB] OR 'vocal fold lesion*'[TIAB] OR 'hoarse*'[TIAB]
Concept 2: Voice therapy	2	**MeSH terms**: 'Rehabilitation of Speech and Language Disorders'[Mesh] OR 'Speech–Language Pathology'[Mesh] OR **Free text words**: 'voice therap*'[TIAB] OR 'vocal therap*'[TIAB] OR 'voice training*'[TIAB] OR 'vocal training*'[TIAB] OR 'speech therap*'[TIAB] OR 'speech training'[TIAB] OR 'therapeutics'[TIAB] OR 'vocal exercise*'[TIAB] OR 'voice exercise*'[TIAB] OR 'exercise therap*'[TIAB] OR 'vocal rehabilitation'[TIAB] OR 'voice rehabilitation'[TIAB] OR 'speech rehabilitation'[TIAB] OR 'logopedic*'[TIAB] OR 'logopaedic*'[TIAB] OR 'logotherap*'[TIAB] OR 'language patholog*'[TIAB] OR 'speech–language'[TIAB] OR 'vocal hygiene'[TIAB] OR 'vocal function exercise*'[TIAB] OR 'vocal function therap*'[TIAB] OR 'voice production therap*'[TIAB] OR 'phonation exercise*'[TIAB] OR 'phonation therap*'[TIAB] OR 'voice treatment'[TIAB] OR 'vocal treatment'[TIAB] OR 'vocal reeducation'[TIAB] OR 'vocal re-education'[TIAB] OR 'voice reeducation'[TIAB] OR 'voice re-education'[TIAB]
Concept 3: Children	3	**MeSH terms**: 'Child'[Mesh] OR 'Adolescent'[Mesh] OR 'Infant'[Mesh] OR **Free text words**: 'child*'[TIAB] OR 'pediatric*'[TIAB] OR 'paediatric*'[TIAB] OR 'adolescen*'[TIAB] OR 'preadolescen*'[TIAB] OR 'pre-adolescen*'[TIAB] OR 'juvenile'[TIAB] OR 'youth*'[TIAB] OR 'boy'[TIAB] OR 'boys'[TIAB] OR 'girl*'[TIAB] OR 'toddler*'[TIAB] OR 'infan*'[TIAB] OR 'young*'[TIAB] OR 'teen*'[TIAB] OR 'preschool*'[TIAB] OR 'school*'[TIAB] OR 'highschool*'[TIAB] OR 'high school'[TIAB] OR 'kid'[TIAB] OR 'kids'[TIAB] OR 'teenage*'[TIAB] OR 'pube*'[TIAB])

TABLE 3 Search strategy for Embase

Research question: What are the effects of voice therapy in children with vocal fold nodules in terms of objective and subjective vocal outcome measures?		
DATABASE: Embase		
Concept	**Line**	**Search strategy**
Concept 1: Vocal fold nodules	1	**Emtree terms**: 'vocal cord'/exp OR 'larynx disorder'/exp OR **Free text words**: 'vocal nod*':ti,ab OR 'voice nod*':ti,ab OR 'vocal fold nod*':ti,ab OR 'vocal cord nod*':ti,ab OR 'sing* nod*':ti,ab OR 'speak* nod*':ti,ab OR 'scream* nod*':ti,ab OR 'nodulus vocalis':ti,ab OR 'noduli vocalis':ti,ab OR 'nodulus plica vocalis':ti,ab OR 'noduli plica vocalis':ti,ab OR 'nodul*':ti,ab OR 'dysphon*':ti,ab OR 'vocal fold lesion*':ti,ab OR 'hoarse*':ti,ab
Concept 2: Voice therapy	2	**Emtree terms**: 'speech and language rehabilitation'/exp OR 'speech disorder'/exp OR 'speech analysis'/exp OR **Free text words**: 'voice therap*':ti,ab OR 'vocal therap*':ti,ab OR 'voice training*':ti,ab OR 'vocal training*':ti,ab OR 'speech therap*':ti,ab OR 'speech training':ti,ab OR 'therapeutics':ti,ab OR 'vocal exercise*':ti,ab OR 'voice exercise*':ti,ab OR 'exercise therap*':ti,ab OR 'vocal rehabilitation':ti,ab OR 'voice rehabilitation':ti,ab OR 'logopedic*':ti,ab OR 'logopaedic*':ti,ab OR 'logotherap*':ti,ab OR 'language patholog*':ti,ab OR 'speech–language':ti,ab OR 'vocal hygiene':ti,ab OR 'vocal function exercise*':ti,ab OR 'vocal function therap*':ti,ab OR 'voice production therap*':ti,ab OR 'phonation exercise*':ti,ab OR 'phonation therap*':ti,ab OR 'voice treatment':ti,ab OR 'vocal treatment':ti,ab OR 'vocal reeducation':ti,ab OR 'vocal re-education':ti,ab OR 'voice reeducation':ti,ab OR 'voice re-education':ti,ab
Concept 3: Children	3	**Emtree terms**: 'child'/exp OR 'adolescent'/exp OR **Free text words**: 'child*':ti,ab OR 'pediatric*':ti,ab OR 'paediatric*':ti,ab OR 'adolescen*':ti,ab OR 'preadolescen*':ti,ab OR 'pre-adolescen*':ti,ab OR 'juvenile':ti,ab OR 'youth*':ti,ab OR 'boy':ti,ab OR 'boys':ti,ab OR 'girl*':ti,ab OR 'toddler*':ti,ab OR 'infan*':ti,ab OR 'young*':ti,ab OR 'teen*':ti,ab OR 'preschool*':ti,ab OR 'school*':ti,ab OR 'highschool*':ti,ab OR 'high school':ti,ab OR 'kid':ti,ab OR 'kids':ti,ab OR 'teenage*':ti,ab OR 'pube*':ti,ab

TABLE 4 Search strategy for Cochrane central register of controlled trials

Research question: What are the effects of voice therapy in children with vocal fold nodules in terms of objective and subjective vocal outcome measures?		
DATABASE: Cochrane Central Register of Controlled Trials		
Concept	**Line**	**Search strategy**
Concept 1: vocal fold nodules	1	**MeSH terms**: [mh 'Vocal Cords'] OR [mh 'Laryngeal Diseases'] OR **Free text words**: vocal NEXT nod*:ti,ab OR voice NEXT nod*:ti,ab OR vocal NEXT fold NEXT nod*:ti,ab OR vocal NEXT cord NEXT nod*:ti,ab OR singing NEXT nod*:ti,ab OR singer's NEXT nod*:ti,ab OR speaking NEXT nod*:ti,ab OR speaker's NEXT nod*:ti,ab OR screaming NEXT nod*:ti,ab OR screamer's NEXT nod*:ti,ab OR 'nodulus vocalis':ti,ab OR 'noduli vocalis':ti,ab OR 'nodulus plica vocalis':ti,ab OR 'noduli plica vocalis':ti,ab OR nodul*:ti,ab OR dysphon*:ti,ab OR vocal NEXT fold NEXT lesion*:ti,ab OR hoarse*:ti,ab
Concept 2: voice therapy	2	**MeSH terms**: ([mh 'Rehabilitation of Speech and Language Disorders'] OR [mh 'Speech–Language Pathology'] OR **Free text words**: voice NEXT therap*:ti,ab OR vocal NEXT therap*:ti,ab OR voice NEXT training*:ti,ab OR vocal NEXT training*:ti,ab OR speech NEXT therap*:ti,ab OR 'speech training':ti,ab OR therapeutics:ti,ab OR vocal NEXT exercise*:ti,ab OR voice NEXT exercise*:ti,ab OR exercise NEXT therap*:ti,ab OR 'vocal rehabilitation':ti,ab OR 'voice rehabilitation':ti,ab OR 'speech rehabilitation':ti,ab OR logopedic*:ti,ab OR logopaedic*:ti,ab OR logotherap*:ti,ab OR language NEXT patholog*:ti,ab OR 'speech–language':ti,ab OR 'vocal hygiene':ti,ab OR vocal NEXT function NEXT exercise*:ti,ab OR vocal NEXT function NEXT therap*:ti,ab OR voice NEXT production NEXT therap*:ti,ab OR phonation NEXT exercise*:ti,ab OR phonation NEXT therap*:ti,ab OR 'voice treatment':ti,ab OR 'vocal treatment':ti,ab OR 'vocal reeducation':ti,ab OR 'vocal re-education':ti,ab OR 'voice reeducation':ti,ab OR 'voice re-education':ti,ab
Concept 3: children	3	**MeSH terms**: [mh 'Child'] OR [mh 'Adolescent'] OR [mh 'Infant'] OR **Free text words**: child*:ti,ab OR pediatric*:ti,ab OR paediatric*:ti,ab OR adolescen*:ti,ab OR preadolescen*:ti,ab OR pre-adolescen*:ti,ab OR juvenile:ti,ab OR youth*:ti,ab OR boy*:ti,ab OR girl*:ti,ab OR toddler*:ti,ab OR infan*:ti,ab OR young*:ti,ab OR teen*:ti,ab OR preschool*:ti,ab OR school*:ti,ab OR highschool*:ti,ab OR 'high school':ti,ab OR kid*:ti,ab OR teenage*:ti,ab OR pube*:ti,ab)

case of disagreement. At the end of the study selection process, backward reference tracking was undertaken for the included articles.

During the data extraction phase, the reviewers were not masked to authors, institution, or publication source. A data extraction form was developed and piloted. The characteristics of the articles were extracted by the two independent researchers and any discrepancies were discussed. Following data were collected: authors, publication year, DOI, country, study design, sample size, gender of participants, age, pathology, intervention groups, therapy content, duration and frequency of voice therapy, outcome parameters, timing of measurements, and results per parameter. If the study design was not explicitly stated, it was deciphered from the method section of the article.

The methodological quality of the included study was assessed using the QualSyst assessment tool from the 'Standard Quality Assessment Criteria for Evaluating Primary Research Papers from a Variety of Fields' (Kmet et al., 2004). The QualSyst tool for quantitative and qualitative research consists of 14 and 10 different scoring items respectively. Scores ('yes' = 2, 'partial' = 1, 'no' = 0) could be assigned depending on the degree to which the different criteria were met or reported. An item is marked n.a. (not applicable) in case it is impossible to assign a score due to the nature of the intervention. A QualSyst score was calculated for each included study using the following formula: [(number of 'yes' * 2) + (number of 'partials' * 1)]/[28 – (number of 'n.a.' * 2)]. The creators of this tool suggested to take 75% (or more liberal: 55%) as the threshold for allowing an article in the review. For this review, it was decided to not remove any articles even if the score was below the threshold. The quality of the included studies was assessed by the first author of this review. No quality assessment was carried out in case of included conference abstracts. The calculated QualSyst score was interpreted following the classification of Lee et al. (2008): strong (score of > 80%), good (70–80%), adequate (50–70%) or limited quality (< 50%).

RESULTS

Study selection

The study selection process is reported in a PRISMA flow diagram (Figure **1**) (Moher et al., 2009). Overall, 4254 records were identified after searching the three databases and 21 records were selected through other sources. A total of 3689 records remained after removing reduplicated articles and 3579 records were excluded during the title-abstract screening process, resulting in 110 articles eligible for full-text screening. Other language than English was the main reason for exclusion (n = 33), followed by background article (no intervention) (n = 25), and population (n = 12) and outcome measure (n = 8) not meeting the selection criteria. Both researchers agreed on 90% of the articles screened on full text. Disagreement on the remaining 10% could be resolved by discussion with the exception of one article for which the opinion of the third researcher was sought. A total of 23 papers were included after full-text screening and one article was added after backward reference tracking, resulting in a total of 24 included studies.

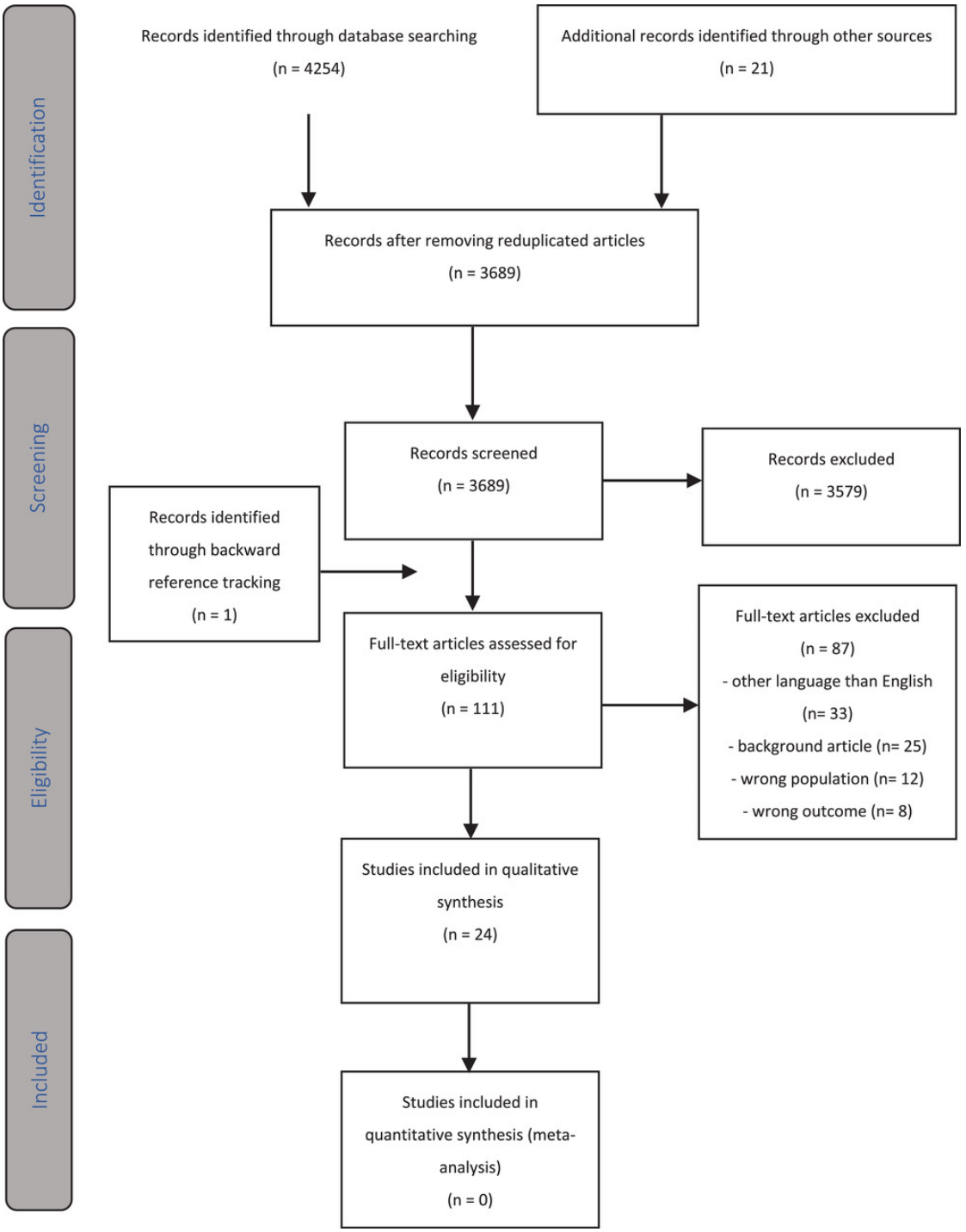

FIGURE 1. Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flow diagram

Study characteristics

The descriptive characteristics and total QualSyst score of the included studies are shown in Table 5. The following information is provided for each article: authors and publication year, country where study was conducted, calculated QualSyst score (%), study design, number of participants, mean age of the participants, gender distribution, and distribution of pathology. In determining the study design, a distinction was made between observational (cohort study, case series and cross-sectional study) and experimental studies. Information about the intervention and the results is shown in Table 6. A brief summary of the voice therapy content is provided, as well as details about the duration and frequency of the voice therapy. Most importantly, the (changes in) outcome characteristics are shown, divided into objective, perceptual, and patient-reported outcome measures. Results that were marked significantly in the study are shown in bold.

Half of the included studies (*n* = 12) were conducted in North America, five in Asia, four in Europe and three in South America (Allen, 1991; Braden & Thibeault, 2020; Braden et al., 2018; Deal et al., 1976; Filter & Poynor, 1982; Hartnick et al., 2018; Mosby, 1970; Nardone et al., 2014; Song et al., 2017; Song & Schloegel, 2016; Valadez et al., 2012; Wilson, 1962; Akin Senkal & Ciyiltepe, 2013; Akin Senkal & Özer, 2015; Lee & Son, 2005; Mori, 1999; Tezcaner et al., 2009; De Bodt et al., 2007; Kay, 1982; Mackiewicz-Nartowicz et al., 2014; Trani et al., 2007; Martins et al., 2017; Nemr et al., 2015; Ramos & Gama, 2017). Seven papers were published before 2000, four between 2001 and 2010, and 13 were published from 2011 onwards (Allen, 1991; Deal et al., 1976; Filter & Poynor, 1982; Kay, 1982; Mori, 1999; Mosby, 1970; Wilson, 1962; De Bodt et al., 2007; Lee & Son, 2005; Tezcaner et al., 2009; Trani et al., 2007; Akin Senkal & Ciyiltepe, 2013; Akin Senkal & Özer, 2015; Braden & Thibeault, 2020; Braden et al., 2018; Hartnick et al., 2018; Mackiewicz-Nartowicz et al., 2014; Martins et al., 2017; Nardone et al., 2014; Nemr et al., 2015; Ramos & Gama, 2017; Song et al., 2017; Song & Schloegel, 2016; Valadez et al., 2012).

Risk of bias

The total QualSyst score ranged from 6% for Kay (1982) to 93% for Hartnick et al. (2018) with a mean score of 55%. For a presentation of the results on each criterion for quantitative studies, see Figure 2. According to the classification of Lee et al (2008), the overall quality of the included studies in this review is adequate. Three papers (14%) have a strong (score > 80%) methodological quality (Hartnick et al., 2018; Ramos & Gama, 2017; Tezcaner et al., 2009), four (18%) have a good (score 70–80%) quality (Braden & Thibeault, 2020; De Bodt et al., 2007; Song et al., 2017; Valadez et al., 2012), seven (32%) have an adequate (score 50–70%) quality (Akin Senkal & Ciyiltepe, 2013; Allen, 1991; Lee & Son, 2005; Mackiewicz-Nartowicz et al., 2014; Martins et al., 2017; Nardone et al., 2014; Trani et al., 2007), and eight (36%) have a limited (score < 50%) quality (Akin Senkal & Özer, 2015; Braden et al., 2018; Deal et al., 1976; Filter & Poynor, 1982; Kay, 1982; Mori, 1999; Mosby, 1970; Wilson, 1962). Frequent shortcomings were an inappropriate study design to answer the research question, insufficiently reported results, and lack of estimates of variance.

TABLE 5 Descriptive characteristics of included studies

Article	Country	QualSyst score	Study design	Sample size	Age (years)	Gender	Pathology
Akin Şenkal and Çiyiltepe (2013)	Turkey	50%	Observational: cohort study	99	10.56 (SD[a]: 2.55; range = 7–15)	62 boys 37 girls	62 VFNs (62.6%) 9 muscle tension dysphonia 2 vocal polyp 8 mutational falsetto 6 vocal fold oedema 9 laryngopharyngeal reflux 1 vocal fold paralysis 2 normal larynx
Akin Şenkal and Özer (2015)	Turkey	43%	Observational: case series	53	10.86 (SD = 2.51; range = 7–14)	39 boys 14 girls	29 VFNs (54.7%) 9 muscle spasms 5 laryngopharyngeal reflux 4 mutational falsetto 2 oedema 1 vocal polyp 1 vocal paralysis 2 normal laryngeal examination
Allen (1991)	USA	58%	Observational: case study (multiple baseline design)	1	9	1 boy	Hyperfunctional dysphonia and VFNs (100%)
Braden and Thibeault (2020)	USA	72%	Observational: cohort study	28	9.92 (SD = 4.25; range = 2.95–17.58)	14 boys 14 girls	19 VFNs (67.9%) 2 cyst and reactive lesion 1 questionable cyst and reactive lesion or VFNs 6 bilateral mid-membranous lesions (not specified)
Braden et al. (2018)	USA	40%	Qualitative study using semi-structured interviews	34 patients 34 parents 5 speech–language pathologists	Patients: 8.82 (range = 4–17)	Patients: 18 boys; 16 girls	28 VFNs (82.3%) 5 laryngeal oedema 1 polyp

(Continues)

TABLE 5 (Continued)

Article	Country	QualSyst score	Study design	Sample size	Age (years)	Gender	Pathology
De Bodt et al. (2007)	Belgium	80%	Observational: cohort study	92	Premutation boys: 7.5 (range = 4–11.6) Premutation girls: 8 (range = 4–10) Post-mutation boys: 16 (range = 14–19) Post-mutation girls: 16 (range = 13–20)	52 boys 40 girls	92 VFNs (100%)
Deal et al. (1976)	USA	44%	Observational: cohort study	31	8.5 (range = 5.5–13.58)	19 boys 12 girls	31 VFNs (100%)
Filter and Poynor (1982)	USA	25%	Observational: cohort study	64	9 (range = 5–15)	37 boys 27 girls	36 VFNs (56%) 28 no VFNs
Hartnick et al. (2018)	USA	93%	Experimental: multicentre randomized clinical trial	114	8 (SD = 1.4; range = 6–10)	83 boys 31 girls	114 VFNs (100%)
Kay (1982)	UK	6%	Observational: cross-sectional study	42	Boys: 6.58 Girls: 6.5 Range = 10–10.83 days	32 boys 10 girls	42 VFNs (100%)
Lee and Son (2005)	Korea	57%	Observational: case series	8	7.5 (range = 4.2–12.2)	8 boys	7 VFNs and muscle tension dysphonia (87.5%) 1 muscle tension dysphonia
Mackiewicz-Nartowicz et al. (2014)	Poland	65%	Observational: cohort study	29	During therapy: 5–12 During study: 15–20	14 boys 15 girls	29 soft VFNs and hyperfunctional dysphonia (100%) 2 additional allergies to inhalatory allergens 1 gastroesophageal reflux
Martins et al. (2017)	Brazil	65%	Observational: cross-sectional study	21	First evaluation: 9 (range = 5–11) End of treatment: 14 (range = 13–18)	21 boys	21 VFNs (100%) 2 microweb at follow-up
Mori (1999)	Japan	25%	Observational: cross-sectional study	259	9 (range = 2–18)	176 boys 83 girls	259 VFNs (100%)

(Continues)

TABLE 5 (Continued)

Article	Country	QualSyst score	Study design	Sample size	Age (years)	Gender	Pathology
Mosby (1970)	USA	16%	Observational: case study	1	10–11	1 boy	1 VFNs (100%) (+ psychologic diagnosis)
Nardone et al. (2014)	USA	65%	Observational: cohort study	67	6.0 (range = 3.8–20.6)	B/G[b] = 2.35/1	67 VFNs (100%)
Nemr, Zemari, Nalom, Oliveira and Silva (2015), poster abstract	Brazil	n.a.	Observational: cohort study	3	Not specified	Not specified	3 VFNs and vocal hyperfunction (100%)
Ramos and Gama (2017)	Brazil	85%	Experimental: randomized, comparative intra-subject study	27	8.18 (SD = 2.03; range = 5–10)	21 boys 6 girls	25 VFNs (95.5%) 2 vocal cyst
Song and Schloegel (2016), presentation abstract	USA	n.a.	Observational: cohort study	155	At diagnosis: 3–12	Not specified	155 VFNs (100%)
Song et al. (2017)	USA	80%	Observational: cohort study	155	At evaluation: 21.4 (SD = 2.6; range = 18–29)	B/G = 2.3/1	155 VFNs (100%)
Tezcaner et al. (2009)	Turkey	90%	Observational: cohort study	39	10 (SD = 2.03; range = 7–14)	20 boys 19 girls	39 VFNs (100%)
Trani et al. (2007)	Italy	60%	Observational: cohort study	16	9 (range = 6–11)	10 boys 6 girls	11 VFNs (72%) 1 VFNs and intracordal cyst 1 fusiform mucous thickening 1 fusiform mucous thickening with a sulcus 1 fusiform mucous thickening with a pseudocyst
Valadez et al. (2012)	Mexico	75%	Observational: cohort study	20	Experimental group: 8.25 (range = 6–10) Control group: 8.42	15 boys 5 girls	20 VFNs (100%)
Wilson (1962)	USA	14%	Observational: case studies	5	5–6	4 boys 1 girl	5 VFNs (100%)

[a]*Notes*: Standard deviation.
[b]Male-to-female ratio.

TABLE 6 Characteristics of interventions and outcomes

Article	Voice therapy content	Duration and frequency intervention	Outcomes of objective voice-related parameters	Outcomes of perceptual voice-related parameters	Outcomes of patient-reported outcome measures
Akin Şenkal and Çiyiltepe (2013)	Type of therapy planned according to children's needs: 1. Physiological VT[a]: optimizing voice production. 2. Hygienic VT: improving behaviours that can lead to injury of the vocal folds. 3. Symptomatic VT: treating abnormal vocal quality	1–2 per week 30 min each 1–11 sessions (mean = 2.9)	Physiological VT: **MPT[b]: pre: 6.18 s; post: 11.9 s*** *s/z*: pre: 0.72; post: 0.84 Hygienic VT: **MPT: pre: 6.8 s; post: 11.87 s*** *s/z*: pre: 0.86; post: 0.91 Symptomatic VT: **MPT: pre: 6.97 s; post: 12.57 s*** ***s/z*: pre: 0.84; post: 1*** Physiological VT: **G[c]: pre: 2; post: 0.6*R[d]: pre: 1.7; post: 0.81***B[e]: pre: 1.11; post: 0.44 A[f]: pre: 0; post: 0 **S[g]: pre: 0.88; post: 0.4***Hygienic VT: **G: pre: 1.31; post: 0.71*R: pre: 1.7; post: 0.56*B: pre: 0.9; post: 0.28***A: pre: 0; post: 0 S: pre: 0.78; post: 0.31 Symptomatic VT: **G: pre: 1.42; post: 0.65*R: pre: 1.3; post: 0.57*B: pre: 1.3; post: 0.27***A: pre: 0; post: 0 **S: pre: 1.258; post: 0.52***	n.a.
Akin Şenkal and Özer (2015)	Type of therapy planned according to children's needs: 1. Physiological VT: optimizing voice production 2. Hygienic VT: improving behaviours that can lead to injury of the vocal folds 3. Symptomatic VT: treating abnormal vocal quality	1–2 per week 30 min each	MPT: pre: 6.2 s; post: 10.86 s ***s/z*[h]: significant change**	**G, R and B: significant change**A and S: no significant change	Frequency of complaints of 'body structures/functions': pre: 163; post: 98 Frequency of complaints of 'activity and limitations': pre: 144; post: 85 Frequency of complaints of 'environmental factors': pre: 57; post: 57

(Continues)

TABLE 6 (Continued)

Article	Voice therapy content	Duration and frequency intervention	Outcomes of objective voice-related parameters	Outcomes of perceptual voice-related parameters	Outcomes of patient-reported outcome measures
Allen (1991)	EMG[i] biofeedback training during speech tasks	Biweekly 30 min each	EMG resting phase: baseline: 7.5 μV; post-treatment: 3.2 μV EMG speaking phase: baseline: 49.5 μV; post-treatment: 10 μV Low levels maintained at 3- and 6-month FU[j] Subglottal pressure: reduced to levels equal to or below established norms for this age group	Voice quality, pathologists: baseline: severely impaired; post-treatment and 6-month FU: normal Voice quality, parents: baseline: 4; post-treatment: 2; 6-month FU: 2 (rated using Buffalo II voice profile) Endoscopic evaluation: decrease in size throughout treatment and at 3-month FU. Gone at 6-month FU	n.a.
Braden and Thibeault (2020)	All participants received some component of direct therapy, 25/28 some component of indirect therapy. Direct VT: Semi-occluded vocal tract exercises Resonant voice/forward focus Abdominal breathing flow mode Vocal projection/healthier yelling Laryngeal reposturing/massage Stretching vocal function exercises Indirect VT: Pedagogical: educating patients and families on the voice and on vocal health recommendations Counselling: discussing reasons for behaviours, counselling the family on behaviour change, and developing strategies for child and family behaviour change	2–11 sessions (mean = 5.79)	Jitter%: pre: 2.19; post: 1.72 Shimmer%: pre: 5.19; post: 5.05 **TP[k]: 5.99 cm H_2O; post: 4.71 cm H_2O***Mean peak pressure: pre: 12.75 cm H_2O; post: 12.0 cm H_2O Resistance: pre: 89.62; post: 60.68	**CAPE-V[l] overall rating: pre: 35.5; post: 27.7***	n.a.

(Continues)

TABLE 6 (Continued)

Article	Voice therapy content	Duration and frequency intervention	Outcomes of objective voice-related parameters	Outcomes of perceptual voice-related parameters	Outcomes of patient-reported outcome measures
Braden et al (2018)	Not specified	Not specified	n.a.	n.a.	Experiences and perceptions of voice therapy and reflections on barriers to and facilitators of adherence: 7 primary themes were identified: 1. Voice therapy is fun 2. Voice therapy is easy 3. Voice therapy is worthwhile 4. The clinician–patient match matters 5. Support systems are helpful 6. Fitting it in 7. Changing behaviour is hard
De Bodt et al. (2007)	1. VT ($n = 90$): not specified 2. Vocal fold surgery ($n = 6$): not specified	Not specified	DSI[m] is worse in patients with voice complaints (1.6 ± 0.7) compared with patients without voice complaints (2.22 ± 0.3) DSI is worse in pre- and post-mutation girls compared with boys	Post-mutational vocal pathology 29% vocal nodules 15% inflammation 12% scaring 65% incomplete closure 44% normal	Post-mutational voice complaints are present in 21% in post-pubescent participants **More voice complaints in post-pubescent girls*** **VHI[n] worse in patients with voice complaints***

(Continues)

TABLE 6 (Continued)

Article	Voice therapy content	Duration and frequency intervention	Outcomes of objective voice-related parameters	Outcomes of perceptual voice-related parameters	Outcomes of patient-reported outcome measures
Deal et al. (1976)	Development of voluntary vocal management Reduction of amount of talking Reduction of vocal loudness Reduction of tension in the laryngeal musculature Increasing breath flow on phonation Encouraging and maintaining gentle adduction of the vocal folds Auditory monitoring of good voice production	2–3 per week 30 min each	Mean nodule rating: 2 months FU: 2.57; 4 months FU: 1.68; 6 months FU: 1.42	n.a.	n.a.
Filter and Poynor (1982)	1. VT ($n = 14$) Decrease vocal abuse and misuse Establishing easy, less forced phonatory patterns Reduction of vocal loudness following Deal et al. (1976) Improving monitoring of the voice through the P-T-K approach 2. Voice rest ($n = 3$) 3. No VT ($n = 4$) 4. Voice rest and VT ($n = 6$)	During 1 year	n.a.	18/27 children with VFNs improved: lower ratings on both vocal tension and severity indices or absence of nodules. Higher percentage of children showing improvement were enrolled in voice rehabilitation. Half of children receiving no voice therapy improved	n.a.

(Continues)

TABLE 6 (Continued)

Article	Voice therapy content	Duration and frequency intervention	Outcomes of objective voice-related parameters	Outcomes of perceptual voice-related parameters	Outcomes of patient-reported outcome measures
Hartnick et al. (2018)	1. Direct therapy arm ($n = 58$): Establishing new voice patterns through motor learning principles, including resonance training and behavioural modeling and/or shaping, while simultaneously overriding existing phonotraumatic vocal patterns. Generalizing new motor control patterns to settings outside of the clinic in varying environments, via homework and at home practice. 2. Indirect therapy arm ($n = 58$) Basic education of the vocal mechanism, normal function, and general care. Identifying phonotraumatic behaviours, as well as environments and/or situations and ways to avoid or modify behaviours. Discussion of 'undesired' and 'desired' voice quality and production	Direct therapy arm: 8 therapy modules 1 per week 8–12 weeks (8 weeks is target) Indirect therapy arm: 6 sessions 8–12 weeks (8 weeks is target, 12 maximum)	NHR[o]: Direct therapy arm: decreased from baseline to 1-month post-therapy to 0.007 (95% CI = −0.021 to 0.007) Indirect therapy arm: increased by 0.005 (95% CI = −0.001 to 0.011) -> difference 0.012 (95% CI = −0.003 to 0.027) PTP: Direct therapy arm: increased by 0.6 Indirect therapy arm: increased by 0.5 -> difference 0.0; 95% CI = −1.8 to 1.7)	CAPE-V: Direct therapy arm: CAPE-V overall severity ratings decreased by 6.2 points Indirect therapy arm: CAPE-V overall severity ratings decreased by 6.7 points -> difference 0.5 (95% CI −5.3 to 4.4) Nodule rating: Those who improved: 31.4% (22/70) had diminished nodules, four participants from each group ($n = 8$ [11%]) had no vocal nodules post-treatment Nodule size improved more in direct therapy arm (39% vs 24%) 4 participants had worsened nodules	PVRQOL[p]: Direct therapy arm: mean change score of 19.2 (95% CI = 19.4 57.9). Only completers: 20.1 (95% CI = 13.9 to 26.3) 27 (61%) achieved a clinically meaningful improvement in PVRQOL scores Indirect therapy arm: mean change score of 14.7 (95% CI = 13.8 43.3). only completers: 15.4 (95% CI = 11.3 to 19.8) 26 (53%) achieved a clinically meaningful improvement in PVRQOL scores -> Mean change score difference of 4.5 (95% CI = 10.8 19.8) -> Clinically meaningful improvement: difference of 8 percentage points (95% CI = 12 28)
Kay (1982)	1. Surgical removal (with or without voice therapy) 2. VT 3. No treatment	Not specified	n.a.	2/9 surgical removal and voice therapy: hoarseness cured 3/5 only voice therapy: hoarseness cured	n.a.

(Continues)

TABLE 6 (Continued)

Article	Voice therapy content	Duration and frequency intervention	Outcomes of objective voice-related parameters	Outcomes of perceptual voice-related parameters	Outcomes of patient-reported outcome measures
Lee and Son (2005)	Three basic phases of behavioural modification: Awareness of voice problem and laryngeal tension Vocal hygiene education Producing easy onset voice: Manual circumlaryngeal massage and respiration training Humming Practicing the vowel beginning with the glottal sound /h/ Self-monitoring and carry over	1–2 per week 30 min each 1–2.5 months	Jitter: pre: 4.06%; post: 2.23% Shimmer: pre: 8.47%; post: 6.61% NHR: pre: 0.27%; post: 0.14%	GRBAS: improvement, especially grade and strained scales A-P[q] contraction of the supraglottis and incomplete glottal closure were relieved into a relaxed normal configuration	n.a.
Mackiewicz-Nartowicz et al. (2014)	Individual training in proper breathing patterns, vocal exercises, psychological counselling and parental education	2–7 years	n.a.	GRBAS: **G: pre: 1.52 ± 0.509; post mut: 1.07 ± 0.267*R: pre: 1.92 ± 0.267; post mut: 1.22 ± 0.424***B: pre 1.03 ± 0.192; post mut: 1.03 ± 0.192 A: pre: 1.07 ± 0.267; post mut: 1.03 ± 0.192 **S: pre: 1.96 ± 0.192; post mut: 1.33 ± 0.480***Complete regression of voice disorders in all boys and 8 out of 15 girls Symmetry: pre: 1.44 ± 0.506; post-mutation: 1.00 ± 0.00 **Amplitude: pre: 1.92 ± 0.267 post-mutation: 1.37 ± 0.492*Mucosal wave: pre: 1.81 ± 0.396; post-mutation: 1.29 ± 0.465*Vocal fold closure: pre: 1.96 ± 0.192; post-mutation: 1.37 ± 0.492***Amplitude improved by 100% and 33.3% in the male and female subjects, respectively ($p < 0.0001$), while the vocal fold closure improved by 91.7% and 42.7% in the male and female subjects, respectively ($p < 0.01$)	n.a.

(Continues)

TABLE 6 (Continued)

Article	Voice therapy content	Duration and frequency intervention	Outcomes of objective voice-related parameters	Outcomes of perceptual voice-related parameters	Outcomes of patient-reported outcome measures
Martins et al. (2017)	Regular VT, not specified	1 per week at least one year	n.a.	15/21 (71.4%) complete improvement (complete remission of symptoms and total reabsorption of lesions) 4/21 (19%) partial improvement (sporadic symptoms and/or partial reabsorption of lesions) 2/21 (9.6%) without improvement (permanent symptoms and/or lesions)	n.a.
Mori (1999)	1. Vocal hygiene ($n = 47$) 2. VT: accent method ($n = 122$) 3. Surgery ($n = 43$) 4. Reassurance and follow-up (no treatment) ($n = 47$)	Vocal hygiene: not specified Voice therapy: 1–2 sessions ($n = 54$), 3–6 sessions ($n = 42$), > 6 sessions ($n = 26$)	n.a.	Perceptual evaluation (not specified): Vocal hygiene: Normal (16%) Improved (0%) No change (80%) Deteriorated (4%) Not available ($n = 22$) VT: Normal (22%) Improved (30%) No change (48%) Not available ($n = 32$) Number of sessions: 1–2: 17% improved voice 3–6: 48% improved voice > 6: 69% improved voice Surgery: Normal (47%) Improved (42%) No change (12%) FU: not available	n.a.

(Continues)

TABLE 6 (Continued)

Article	Voice therapy content	Duration and frequency intervention	Outcomes of objective voice-related parameters	Outcomes of perceptual voice-related parameters	Outcomes of patient-reported outcome measures
Mosby (1970)	Counselling: Identifying situations Attempting systematically to reduce bad phonation in these situations Counselling on how to change his voice quality Vocal manipulation: certain techniques of chewing, yawning and singing psychotherapy	6 weeks VT: 20 sessions psychotherapy: 3 months VT: 10 sessions psychotherapy	n.a.	After VT: nodules still present	n.a.
Nardone et al. (2014)	1. No treatment or behavioural modification only ($n = 19$) 2. Targeted VT with or without treatment of associated factors ($n = 45$) 3. surgical intervention ($n = 3$)	Not specified	n.a.	Nodule grade, two-step method described by Feldman: Mean slope (change in grade/time [months]): −0.03 (0.12) median slope: −0.01 (range = −0.94 to 0.06) Slope was significantly associated with baseline VFNs size ($p < 0.001$), with an increased rate of improvement in VFNs size observed for those children with larger baseline VFNs size Rate of change in VFNs size was significantly associated with treatment, with a greater rate of improvement seen in those children receiving VT with or without the management of associated conditions or those undergoing surgery No treatment: median (range) slope of 0.00 (−0.08 to 0.06) **VT: median (range) slope of −0.03 (−0.94 to 0.05)***Surgical intervention: median (range) slope of −0.08 (−0.09 to 0.00)	n.a.

(Continues)

TABLE 6 (Continued)

Article	Voice therapy content	Duration and frequency intervention	Outcomes of objective voice-related parameters	Outcomes of perceptual voice-related parameters	Outcomes of patient-reported outcome measures
Nemr et al. (2015), poster abstract	Infantile Speech–Language Pathology programme for vocal hyperfunctioning: breathing, glottis activity and resonance/articulation	6 sessions	Improvements in acoustic analysis	Improvements in perception Visual analogic scale: one child kept the same initial score and two presented improvement	n.a.
Ramos and Gama (2017)	1. Control group (CG): complete vocal rest 2. Experimental group (EG): straw phonation exercises	1 session	EG: no differences in acoustic analysis between time points CG: no differences in acoustic analysis between time points Increased GNE[r] after third minute of straw phonation: **m3 GNE: mean CG: 0.69; mean EG: 0.80***decreased noise in dB after third minute of straw phonation: **m3 noise: mean CG: 1.53; mean EG: 1.05***	CAPE-V measurements after 1, 3, 5 and 7 min of straw phonation/rest: EG: no differences in auditory perceptual evaluation between time points CG: no differences in auditory perceptual evaluation between time points Improved vocal quality was observed after 3 and 5 min of straw phonation: **m0–m3 Improved EG: 14 (51.85%); CG: 2 (7.41%)*m0–m5 Improved EG: 14 (51.85%); CG: 6 (22.22%)***	n.a.

(Continues)

TABLE 6 (Continued)

Article	Voice therapy content	Duration and frequency intervention	Outcomes of objective voice-related parameters	Outcomes of perceptual voice-related parameters	Outcomes of patient-reported outcome measures
Song and Schloegel (2016), presentation abstract	Not specified	Not specified	n.a.	n.a.	VHI-30 scores did not significantly differ between patients who underwent voice therapy and those who did not
Song et al. (2017)	1. VT ($n = 87$) 2. No VT ($n = 68$)	1–5 sessions ($n = 60$) > 5 sessions ($n = 27$)	n.a.	n.a.	Mean VHI-10 scores did not significantly differ between those who received speech therapy (6.1) and those who did not (4.5)

(Continues)

TABLE 6 (Continued)

Article	Voice therapy content	Duration and frequency intervention	Outcomes of objective voice-related parameters	Outcomes of perceptual voice-related parameters	Outcomes of patient-reported outcome measures
Tezcaner et al. (2009)	Vocal hygiene and voice abuse reduction Diaphragmatic breathing and coordination of breathing with phonation Relaxation exercises including laryngeal massage, yawn/sign exercise, and chewing method Accent method, stretching exercises and resonant voice therapy	45 min per week 8 weeks 4 × 10 min home exercises per day	fo^{s} -> pre: 275.3 ± 30.9; 6 months: 269.8 ± 34.3 **Jitter: pre: 2.51 ± 2.31; 6 months FU: 1.55 ± 1.11*** **Shimmer: pre: 6.39 ± 4.15; 6 months FU: 4.55 ± 3.32*** **NHR: pre: 0.20 ± 0.20, 6 months FU: 0.15 ± 0.08***	**G: pre: 1.8 ± 0.5; post: 0.7 ± 0.6*** **R: pre: 0.8 ± 0.7; post: 0.2 ± 0.4*** **B: pre: 1.4 ± 0.5; post: 0.6 ± 0.4*** A: pre: 0; post: 0 **S: pre: 0.2 ± 0.4; post: 0***	n.a.
Trani et al. (2006)	Individual treatment of Borragan associated with the counselling according to Dr Magnani	15 sessions (10 voice therapy, 5 of maintenance)	Jitter, shimmer, NHR, vFo^{t}, fo, MPT + evaluation of spectrograms and phonetograms -> improvement trend of all the considered parameters but no one reached statistical significance	GRBASI: nearly universal improvement of all parameters but most notably in the parameters G, R, B and less so for I^{u} and S. There was a little change for A ratings	n.a.

(Continues)

TABLE 6 (Continued)

Article	Voice therapy content	Duration and frequency intervention	Outcomes of objective voice-related parameters	Outcomes of perceptual voice-related parameters	Outcomes of patient-reported outcome measures
Valadez et al. (2012)	Vocal hygiene education Awareness of voice problem and laryngeal tension Producing easy onset voice self-monitoring and carry over	2 sessions per week 45 min each 20 weeks	**f_o: pre: 234.18 (40.90); post: 254.38 (57.22)*Jitter: pre: 2.06 (1.23); post: 0.37 (0.25)*Shimmer: pre: 9.94 (4.80); post: 4.66 (2.92)***	Perceptual assessment: improvement in all cases Videonasolaryngostroboscopy: no vocal nodules discernible	n.a.
Wilson (1962)	Voice re-education: Reduction or elimination of vocal abuse Training for a correct pitch level and an adequate loudness level Production of a voice free from hoarseness	Case 2: 16 sessions during 3 months Case 3: 25 sessions during 5 months Case 4: 24 sessions during 14 weeks Case 5: 17 sessions during 3 months, additional nine sessions over 4 months, bimonthly voice check-up during 14 months	n.a.	Case 2: nodules reduced in size, hoarseness eliminated Case 3: lowered habitual pitch level, vocal abuses reduced or eliminated, tones being easily initiated Case 4: nodules reduced in size, reduced hoarseness, eliminated vocal abuse, lowered pitch Case 5: after 3 months: reduction of nodule size but beyond expectation; after 4 months: vocal abuse almost eliminated, pitch lower and voice almost free from hoarseness, nodules very small; after 14 months: elimination of nodules, clear voice	n.a.

[a]*Notes*: Voice therapy.
[b]Maximum phonation time.
[c]Grade.
[d]Roughness.
[e]Asthenicity.
[f]Breathiness.
[g]Strain.
[h]*s/z* ratio.
[I]Electromyography.
[j]Follow-up.
[k]Phonation threshold pressure.
[l]Consensus auditory–perceptual evaluation of voice.
[m]Dysphonia severity index.
[n]Voice handicap index.
[o]Noise-to-harmonic ratio.
[p]Pediatric voice-related quality-of-life survey.
[q]Anterior–posterior.
[r]Glottal-to-noise excitation.
[s]Fundamental frequency.
[t]Variation in fundamental frequency.
[u]Instability.
*Results which were marked significantly in the study.

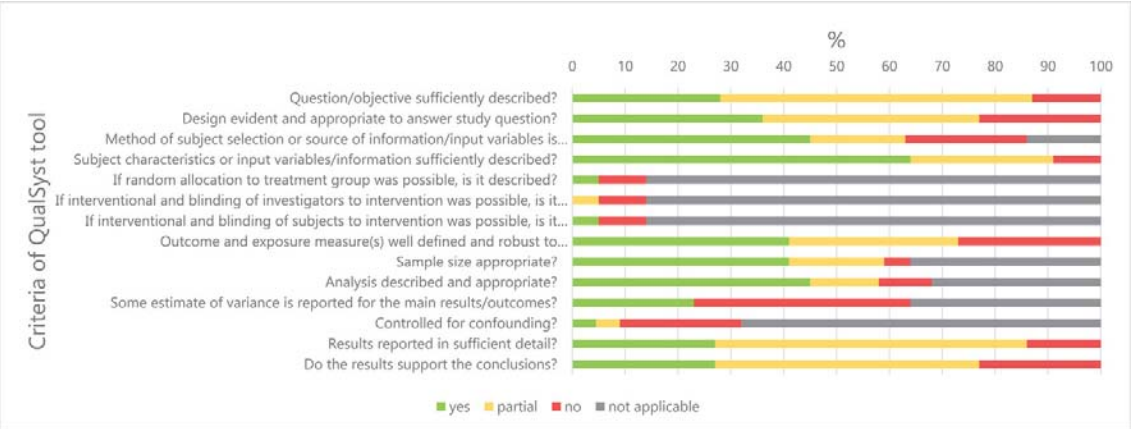

FIGURE 2. Methodological quality of the individual studies: results of the QUALSYST Tool

Study design

One qualitative study was included in this review (Braden et al.,2018). Only two quantitative studies had an experimental nature (Hartnick et al., 2018; Ramos & Gama, 2017), all other papers were observational (Table 5).

Participant characteristics

In sum, 1363 participants were included across all different studies in the systematic review. Sample sizes ranged from one in the case studies by Allen (1991) and Mosby (1970) to 259 in the retrospective study of Mori (1999), with a median sample size of 33 children. Mean age of the participants is 8.69 years (SD = 1.89, range = 10 days–20.6 years at the end of treatment). All studies with multiple participants, except for Martins et al. (2017), included both boys and girls, with a mean male-to-female ratio of 2.02/1. In 16 studies, only children with VFNs were included, whereas eight papers described a mix of laryngeal pathologies (Akin Senkal & Ciyiltepe, 2013; Akin Senkal & Özer, 2015; Braden & Thibeault, 2020; Braden et al., 2018; Filter & Poynor, 1982; Lee & Son, 2005; Ramos & Gama, 2017; Trani et al., 2007). Overall, 91% of all included participants had a diagnosis of VFNs.

Intervention characteristics

Six studies did not provide any specifications about the content of offered voice therapy (Braden et al., 2018; De Bodt et al., 2007; Kay, 1982; Martins et al., 2017; Song et al., 2017; Song & Schloegel, 2016). Between the other included studies, the content of the intervention sessions widely differed. Five multigroup studies made a comparison between direct voice therapy techniques and an indirect voice therapy approach (Akin Senkal & Ciyiltepe, 2013; Akin Senkal & Özer, 2015; Hartnick et al., 2018; Mori, 1999; Nardone et al., 2014). A combination of direct and indirect therapy is offered in 10 studies (Braden & Thibeault, 2020; Deal et al., 1976; Filter & Poynor, 1982; Lee & Son, 2005; Mackiewicz-Nartowicz et al., 2014; Mosby, 1970; Tezcaner et al., 2009; Trani et al., 2007; Valadez et al., 2012; Wilson, 1962). For indirect therapy, the following techniques were mentioned: identifying and decreasing vocal abuse and misuse, increasing awareness of voice problem and laryngeal tension, psychological counselling, and parental education. For direct therapy, the

following techniques were used: breathing exercises, semi-occluded vocal tract exercises, vocal function exercises, resonant voice and forward focus, accent method, tension reduction in laryngeal musculature, laryngeal reposturing, laryngeal massage, easy onset exercises, flow mode, stretching, humming, chewing, yawning, singing techniques, and improving monitoring of voice. Allen (1991), Nemr et al. (2015) and Ramos and Gama (2017) examined exclusively the effects of direct therapy, respectively EMG biofeedback training, the Infantile Speech–Language Pathology Program for Vocal Hyperfunctioning, and straw phonation exercises. Breathing exercises were included in five studies (Braden & Thibeault, 2020; Lee & Son, 2005; Mackiewicz-Nartowicz et al., 2014; Nemr et al., 2015; Tezcaner et al., 2009).

Total number, duration and frequency of therapy sessions were not mentioned in each paper. Reported or calculated (by combining frequency and therapy duration) number of sessions ranged from one session (Ramos & Gama, 2017) to more than 52 sessions (Martins et al., 2017), with a mean of 14.67 sessions (SD = 12.3). In three studies, information was given on how the total number of sessions varied among participants (Mori, 1999; Song et al., 2017; Wilson, 1962).

Five studies described a session duration of 30 min (Akin Senkal & Ciyiltepe, 2013; Akin Senkal & Özer, 2015; Allen, 1991; Deal et al., 1976; Lee & Son, 2005), only Valadez et al. (2012) and Tezcaner et al. (2009) provided 45 min sessions. Frequency of voice therapy was reported in eight studies: once a week (Hartnick et al., 2018; Martins et al., 2017; Tezcaner et al., 2009), one or two times a week (Akin Senkal & Ciyiltepe, 2013; Akin Senkal & Özer, 2015; Lee & Son, 2005), two times a week (Valadez et al., 2012), and two or three times a week (Deal et al., 1976). Allen (1991) mentioned ‘biweekly’ sessions, which is unclear because it can mean both ‘twice a week’ as ‘every 2 weeks’.

Outcome characteristics

Objective voice-related parameters

Percentage jitter

Six studies investigated percentage jitter. Two studies reported a significant improvement in post-treatment jitter: Tezcaner et al. (2009) found a decrease of 0.96% at 6 months’ follow-up and Valadez et al. (2012) described a drop of 1.69% after 20 weeks of voice therapy. Lee and Son (2005) also reported a decrease in jitter (pre: 4.06%; post: 2.23%), but no statistical test is performed making it unclear whether the difference is significant. Braden and Thibeault (2020) (pre: 2.19%; post: 1.72%) and Trani et al. (2007) showed a non-significant improvement in post-treatment jitter. Lastly, Ramos and Gama (2017) did not find significant differences in jitter between the experimental or control group at any time point.

Percentage shimmer

Similar results were found for percentage shimmer. Tezcaner et al. (2009) and Valadez et al. (2012) reported significant improvements of 1.84% at 6 months’ follow-up and 4.88% after 20 weeks of voice therapy, respectively. Lee and Son (2005) described a decrease from 8.47% to 6.61%, not tested for significance. Shimmer non-significantly changed post-treatment in Braden and Thibeault (2020) (pre: 5.19%; post:

5.05%) and in Trani et al. (2007). In Ramos and Gama (2017), no significant differences were reported between the experimental or control group at any time point.

Noise-to-harmonic ratio (NHR)—noise in dB—glottal-to-noise excitation (GNE)

Four papers examined the NHR. A significant improvement (decrease) in NHR (pre: 0.20%; post: 0.15%) was reported at the 6 months' follow-up by Tezcaner et al. (2009). In contrast, the three other papers did not find a significant change in NHR after voice therapy (Hartnick et al., 2018; Lee & Son, 2005; Trani et al., 2007). Ramos and Gama (2017) reported a significant decrease in noise in dB between 3 min of straw phonation and 3 min of vocal rest and a significant increase in GNE at the same time point.

s/z ratio

The *s/z* ratio was investigated in two studies that both found a significant change after therapy. Akin Senkal and Ciyiltepe (2013) reported a significant increase in *s/z* ratio, while Akin Senkal and Özer (2015) did not mention the direction of change. This parameter was not examined in other studies.

Fundamental frequency (f_o)

Four studies investigated effects on f_o after voice therapy. Valadez et al. (2012) reported a significant increase of 20.2 Hz. Differences in f_o reported in other studies were not significant (Ramos & Gama, 2017; Tezcaner et al., 2009; Trani et al., 2007).

Maximum phonation time (MPT)

Three studies investigated the effect of voice therapy on MPT. Akin Senkal and Ciyiltepe (2013) reported a significant increase in each group, regardless of voice therapy type (physiological, hygienic or symptomatic). MPT improved with approximately 5 s (range = 5.07–5.71 s) post-therapy. Akin Senkal and Özer (2015) and Trani et al. (2007) also described an increase in MPT but the results did not reach statistical significance.

Subglottal pressure

Subglottal pressure measures were included in the protocol of Allen (1991). Before EMG biofeedback training, subglottal pressure levels were significantly higher than norm values for children. After therapy, subglottal pressure dropped to levels equal or below the norms and these levels were maintained at 6 months' follow-up. Hartnick et al. (2018) investigated changes in phonation threshold pressure (PTP), defined as the minimum subglottal pressure required to initiate vocal fold oscillation (Titze, 2009), following a direct and indirect voice therapy programme. No significant difference in PTP was found between both therapy approaches. In contrast, Braden and Thibeault (2020) described a significant decrease (improvement) in PTP after therapy, going from 5.99 cm H_2O to 4.71 cm H_2O.

Dysphonia severity index (DSI)

This multiparameter vocal quality index was only investigated by De Bodt et al. (2007). DSI in adolescents with a history of paediatric VFNs is worse in patients with current voice complaints (DSI = 1.6) compared with patients without voice complaints (DSI = 2.22). The number of sessions of voice therapy did not influence the outcome.

Electromyographic (EMG) activity measures

The biofeedback study of Allen (1991) reported EMG activity measures. During the training, stepwise reductions in the average levels of muscle tension were observed in both resting and speaking mode. These low levels of EMG activity were maintained at 3 and 6 months' follow-up.

Perceptual voice-related parameters

GRBAS(I)

Six papers used the GRBAS(I) scale for perceptual evaluation of the voice. G (Grade) and R (Roughness) improved significantly in four studies (Akin Senkal & Ciyiltepe, 2013; Akin Senkal & Özer, 2015; Mackiewicz-Nartowicz et al., 2014; Tezcaner et al., 2009). These parameters also improved in the other two studies, but there was no testing for significance (Lee & Son, 2005; Trani et al., 2007). B (Breathiness) significantly improved in two papers (Akin Senkal & Özer, 2015; Tezcaner et al., 2009) and in the hygienic and symptomatic voice therapy groups of Akin Senkal and Ciyiltepe (2013). B also improved in the studies of Lee and Son (2005) and Trani et al. (2007), but they did not test for significance. A non-significant change was found in Mackiewicz-Nartowicz et al. (2014) and in the physiological voice therapy group of Akin Senkal and Ciyiltepe (2013) Concerning S (strained), a significant improvement was found in two studies (Mackiewicz-Nartowicz et al., 2014; Tezcaner et al., 2009) and in the physiological and symptomatic voice therapy groups of Akin Senkal and Ciyiltepe (2013). Two other studies found an improvement without testing for significance (Lee & Son, 2005; Trani et al., 2007). Akin Senkal and Özer (2015) and the hygienic voice therapy group in Akin Senkal and Ciyiltepe (2013) found a non-significant improvement of S. Only Trani et al. (2007) included the parameter I (Instability), which improved non-significantly. None of the studies reported a significant change in A (asthenicity). No study examined inter- or intra-rater reliability.

Consensus auditory perceptual evaluation of voice (CAPE-V)

CAPE-V was administered in three papers. The overall severity rating improved significantly in Braden and Thibeault (2020) after therapy (pre: 35.5; post: 27.7). In contrast, Hartnick et al. (2018) reported no significant change in overall severity after direct and indirect therapy. Agreement between the two raters for overall severity was 0.63. No significant differences could be demonstrated in CAPE-V measurements after 1, 3, 5 and 7 min of straw phonation (Ramos & Gama, 2017), but when comparing the straw phonation group and the vocal rest group, R and B were significantly lower after 3 min of straw phonation and B and G were lower after 5 min. One of the five raters had an intra-rater agreement of 0.80. Agreement scores of the other raters were not reported.

Other perceptual voice-quality ratings

Speech–language pathologists and parents used the Buffalo II Voice Profile, containing five-point Likert scales, to evaluate the voice perceptually in the case study by Allen (1991). According to speech–language pathologists, vocal quality of the boy was severely impaired at baseline and near normal at 6 months' follow-up. The parents also reported an improvement in vocal quality (pre: 6–6 months FU: 2). Filter and Poynor (1982) used a modification of the pitch, vocal tension, and severity of

hoarseness scale of the Jewish Hospital voice profile, which included seven-point rating scales. Mean intra-rater reliability, calculated for the eight raters, was 0.42 for pitch, 0.90 for vocal tension and 0.97 for severity of hoarseness. A higher percentage of children enrolled in voice therapy showed improvement compared with children not enrolled in voice therapy. But interestingly, half of children without voice therapy also improved.

Endoscopic laryngeal evaluation

In 11 studies, an endoscopic examination was carried out to evaluate the vocal folds. Mackiewicz-Nartowicz et al. (2014) described the NAPZ scale proposed by Wiskirska-Woznica (2002) and found a significant improvement for the parameters amplitude, mucosal wave and vocal fold closure. Nardone et al. (2014) used the two-step method described by Feldman and Nuss (2007). VFNs size decreased significantly faster in children receiving targeted voice therapy or surgery compared with children receiving no treatment or only behavioural modification. VFNs were completely disappeared after 40 voice therapy sessions by Valadez et al. (2012), at 6 months' follow-up in Allen (1991), and in one case study by Wilson (1962). VFNs reduced in size in the other case studies by Wilson (1962) and Deal et al. (1976), while there were still present in the case study by Mosby (1970). Hartnick et al. (2018) concluded that nodule size decreased more in the direct therapy group. Endoscopic evaluation showed De Bodt et al. (2007) that 29% of treated participants still suffer from VFNs in adolescence. Additionally, 65% have incomplete vocal fold closure. Lastly, a more relaxed vocal fold configuration was seen by Lee and Son (2005) after voice therapy in all patients who tolerated stroboscopy.

Patient-reported outcome measures

Voice handicap index (VHI)

VHI was administered in three studies examining vocal outcomes of post-mutational adolescents or adults after voice therapy during childhood. VHI was completed by the adolescent or adult patients. VHI-30 was reported in the presentation abstract of Song and Schloegel (2016) while Song et al. (2017) used the VHI-10. VHI scores did not significantly differ between patients who received voice therapy during childhood and those who did not, nor between patients with more or less than five sessions of voice therapy. According to De Bodt et al. (2007), the number of sessions of voice therapy did not influence the VHI outcome.

Paediatric voice-related quality of life (PVRQOL)

In Hartnick et al. (2018), the PVRQOL was completed by the caregivers of the children. It was found that 61% of children in the direct therapy group and 53% in the indirect therapy group achieved a clinically meaningful improvement in PVRQOL scores.

Complaints about hoarseness

Akin Senkal and Özer (2015) obtained information about voice complaints by using a parent-proxy questionnaire based on the ICF model. After voice therapy, the frequency of complaints related to 'body structures or functions' and 'activity and limitations' decreased.

DISCUSSION

The aim of this systematic literature review was to provide an overview of the evidence concerning the effects of voice therapy in paediatric VFNs. Different treatment options are available, including direct and indirect voice therapy, phonosurgery, modulation of associated factors, and watchful waiting. In order to properly inform, advise and treat patients with VFNs, clinicians must know the effects of each possible treatment method. This systematic review is important because it identifies, evaluates, and summarizes the results of all relevant individual studies about voice therapy in paediatric VFNs, making the available evidence more accessible to voice therapists, otolaryngologists, and other stakeholders (Gopalakrishnan & Ganeshkumar, 2013).

After the study selection process, 24 articles were included in this review. Most studies were conducted in North America (50%), followed by Asia (21%), Europe (17%) and South America (12%). This reflects a broad international spread of studies, however, Africa and Oceania are not represented. According to the country classification of The World Bank for 2022, all studies were carried out in high-income economies (67%; Belgium, Japan, Italy, Poland, the UK and USA) or upper middle-income economies (29%; Brazil, Japan and Turkey), except for the paper of Lee and Son (2005), which was conducted in the low-income economy of Korea (The World Bank, 2021). This highlights the need for more research in low-income countries because evidence from these regions is currently missing. Regarding the publication date, the oldest relevant article was published by Wilson (1962). Several publications followed in the 1970s, 1980s and 1990s, indicating that researchers have been interested in paediatric VFNs for several decades. Recently, the focus on this subject has increased which is reflected in the fact that 54% of the studies were published from 2011 onwards.

The methodological quality of the included studies was assessed using the QualSyst tool. Fourteen aspects, such as research question, study design, subject characteristics, blinding of investigators and subjects, sample size and analytical methods, were evaluated in order to check the risk of bias (Kmet et al., 2004). For this review, a mean QualSyst score of 55% was calculated, ranging from 6% to 93%. According to the classification of Lee et al. (2008), the overall quality of the included studies in this review is adequate. Some extremely low QualSyst scores are seen by Kay (1982) (6%), Wilson (1962) (14%) and Mosby (1970) (16%), followed by Mori (1999) (25%) and Filter and Poynor (1982) (25%). All these low-quality articles were published more than 20 years ago, suggesting that attention for a strong research methodology was lacking in the 20th century. Considering these weak methodologies and the rather low mean QualSyst score, the results must be interpreted with caution.

Almost all included quantitative studies used an observational study design (92%), except for the multicentre randomized clinical trial of Hartnick et al. (2018) and the randomized, comparative intra-subject study of Ramos and Gama (2017). According to Sackett (1989), the highest levels of evidence are given to randomized controlled trials (RCTs) since they are designed to avoid biases and systematic errors. To our knowledge, no RCTs are carried out to test the effectiveness of voice therapy in paediatric VFNs. The control group of a well-designed RCT receives a placebo therapy, a standard treatment or no intervention at all (Bhide et al., 2018). In the experimental study of Hartnick et al. (2018), direct and indirect voice therapy programmes were compared, but a true placebo control group was missing due to

ethical reasons. In other words, there is a great lack of well-designed experimental studies.

Participant inclusion criteria in this review were aged under 18 years, a diagnosis of VFNs and no comorbidities. In total, 1363 participants were included in this review, whereas the median number of participants in a Cochrane review is 945 with an interquartile range from 313 to 2511 (Mallett & Clarke, 2002). The calculated male-to-female ratio in this review is 2.02/1, which can be explained by the difference in prevalence of VFNs between boys and girls. It is generally noticed that VFNs are more common in school-aged boys with a male-to-female ratio of 2/1 (Akif Kilic et al., 2004), which is also reflected in this review. The mean age of the participants is 8.69 years with a range from 2 to 20.6 years at the end of treatment, with the bulk of the studies mainly including school-aged children. Children younger than 5 and older than 14 years are underrepresented in the studies. This can be explained by the fact that preschoolers have difficulties cooperating with voice therapy and children older than 12 years can undergo pubertal voice changes, making it difficult for researchers to examine the effects of voice therapy during this life period. Few authors uniquely focus on older teenagers, possibly because a post-mutational adolescent's voice does not differ much from an adult voice. Moreover, the typical age to be diagnosed with VFNs is 9 years (Cornut & Troillet-Cornut, 1995), indicating the clinical importance of research in school-aged children. However, more research about voice therapy options in young children and in adolescents is necessary.

The content of voice therapy shows a wide variation through the different studies. There seems to be no consensus about the aspects that constitute effective voice therapy for children. Most papers provide an eclectic voice therapy programme, which is consistent with current clinical practice. First, indirect techniques are often part of the therapy programme, educating the patient on their voice, voice problem and vocal hygiene. Based on the fact that paediatric VFNs are often caused by vocal misuse or abuse (Middendorf, 2007), patient and parent counselling can be assumed to be an important topic in voice therapy for children. It is proven that school-aged children are mature enough to learn from an indirect vocal hygiene programme and that this voice care knowledge can be maintained for at least 1 year (Ma & Leung, 2021). In three included studies comparing direct and indirect voice therapy, the participants of both groups improved without a significant difference between them (Akin Senkal & Ciyiltepe, 2013; Akin Senkal & Özer, 2015; Hartnick et al., 2018). These results suggest that children benefit from an indirect therapy programme. In contrast, Mori (1999) stated that 16% of the participants improved after a vocal hygiene programme whereas 52% improved after voice therapy using the accent method. Nardone et al. (2014) found no significant decrease in VFNs size after indirect therapy or observation. In sum, it is not yet clear whether voice care knowledge affects the vocal behaviour of children with VFNs. Further research should be undertaken to investigate the effects of indirect therapy in children with VFNs.

Second, a variety of direct voice techniques are represented in the different studies. Van Stan et al. (2015) proposed a taxonomy that subdivides direct voice techniques into five different categories: auditory, respiratory, somatosensory, musculoskeletal, and vocal function. It seems that voice therapy for children with VFNs does not focus on musculoskeletal aspects such as neck and orofacial modification, posture, and stretching. Most reported direct techniques for paediatric VFNs require a certain

degree of somatosensory or auditory discrimination and respiratory coordination. Some papers also pay attention to glottal contact, such as easy onset exercises, and pitch modifications.

In general, the description of the voice therapy offered is rather vague, with six studies even not providing any specifications about the content (Braden & Thibeault, 2020; De Bodt et al., 2007; Kay, 1982; Martins et al., 2017; Song et al., 2017; Song & Schloegel, 2016). Most other studies give a brief overview of the therapy topics but details about time spent on each topic or specific exercises are usually lacking, only Hartnick et al. (2018) describe in depth the principles and concepts for each therapy session. There are two explanations for this methodological shortcoming. First, a lot of studies are retrospective, which implies that no predefined therapy plan was formulated. Second, the specific techniques and exercises are normally adapted to the individual's need. In a clinical setting, voice therapy is not a static and fixed plan that can be implemented unchanged for each patient. However, the lack of a thorough description of the delivered voice therapy increases the risk for intervention bias and poses a problem concerning the reproducibility of the study. This limitation is also seen in the review of Feinstein and Abbott (2021) and Alegria et al. (2020) about voice therapy in adults with VFNs.

There is a large spread in the total number of voice therapy sessions, ranging from one (Ramos & Gama, 2017) to more than 52 (Martins et al., 2017), with a mean of 14.67 sessions. The mean number of sessions could only be calculated on the papers reporting the exact number of sessions or the frequency and therapy duration, so not all studies are represented. For example, Martins et al. (2017) described the highest number of sessions (one session a week for at least 1 year), but this was not used in the calculation as the exact number could not be determined. In a review of De Bodt et al. (2015), an average of 10.87 sessions distributed over 9.25 weeks was found. They did not focus on a specific target population or treated voice disorder. Compared with the findings of De Bodt et al. (2015) about general voice therapy, this systematic review shows an increase with 35% for mean number of voice therapy sessions in children with VFNs. Children may need more time to learn the voice exercises, control their voices or implement the techniques during therapy sessions and spontaneous talking. Geographical differences may also contribute to the discrepancy in number of sessions. In this review, half of the studies are conducted in North America, where the average number of voice therapy sessions is higher (12.52 sessions) compared with other regions (De Bodt et al., 2015). Duration of one session ranged between 30 and 45 min, while De Bodt et al. (2015) described 30 and 60 min sessions. It can be assumed that therapy sessions of 60 min are too demanding for children given their limited attention span (Huh et al., 2019). The majority of the included studies provided therapy sessions one or two times a week, which corresponds to the review of De Bodt et al. (2015).

Another important temporal aspect is the optimal dosage for voice therapy in children. Two included studies examined the relation between temporal aspects of voice therapy and therapy outcomes. Akin Senkal and Özer (2015) found a moderate degree significance between the total number of voice therapy sessions and *s/z* ratio in the physiological voice therapy group, and they concluded that ‘physiological voice therapy is a very long-term application requiring active attendance of both parents and children’. They did not find any other significant relationship between session

frequency and therapy modalities. Also De Bodt et al. (2007) noted that duration and frequency of therapy sessions seemed not to influence the long-term outcome. The optimal dosage for voice therapy in an adult population has been extensively studied by Meerschman et al. (2019). They stated that 'therapy frequency and duration used today depends on medical prescription, rules of reimbursement, the specific vocal pathology and its severity, the type of therapy or training, the client's limitations and expectations, and upcoming vocal performances'. Moreover, they proved that massed practice, which means that voice therapy sessions are organized in a short time frame, is at least equally effective as spaced practice. However, the optimal dosage for paediatric voice therapy is still unknown and research on intensive voice therapy programmes for children with VFNs is completely lacking. Future research is necessary to investigate the effects of therapy duration and frequency, and especially the influence of massed practice, in dysphonic children.

The recurrence of VFNs, even after voice therapy or phonosurgery, is an inevitable risk (Mansuri et al.,2018). It is therefore important to investigate the longer term effects of voice therapy as well. Twelve studies (50%) included at least one follow-up, defined as a measurement that is not carried out immediately following the treatment but after a certain period without intervention (Allen, 1991; De Bodt et al., 2007; Filter & Poynor, 1982; Hartnick et al., 2018; Mackiewicz-Nartowicz et al., 2014; Martins et al., 2017; Mosby, 1970; Nardone et al., 2014; Song et al., 2017; Song & Schloegel, 2016; Tezcaner et al., 2009; Wilson, 1962). Most studies (9/12) reported positive longer term changes after voice therapy. The follow-up period ranges from 1 month (Hartnick et al., 2018) to 3 years (Nardone et al., 2014). Some authors showed interest in the outcomes of post-mutational adolescents or adults after voice therapy during childhood (De Bodt et al., 2007; Mackiewicz-Nartowicz et al., 2014; Martins et al., 2017; Song et al., 2017; Song & Schloegel, 2016). De Bodt et al. (2007) found more voice problems in post-pubescent girls as compared with boys. This was confirmed by the results of Mackiewicz-Nartowicz et al. (2014) who reported complete regression of the voice problems in all boys and in 8/15 girls. As mentioned above, the long-term expectation for VFNs is better in boys than in girls and girls are at risk for persistent vocal complaints.

The need for long-term results is generally known in the field of paediatric VFNs, but not all studies include follow-up measures. A frequently reported problem is the loss to follow-up, especially in long-term outcome studies. For example, De Bodt et al. (2007) contacted 126 patients and only 35 (28%) of them agreed to participate.

A wide variation of outcome characteristics is shown in the different studies. Perceptual voice-related parameters are the most described characteristics as at least one perceptual parameter is investigated in 83% of the included studies. Auditory–perceptual evaluation is considered as the 'gold standard' in a clinical voice assessment because of the perceptual nature of vocal quality and limitations in validity and reliability of objective measures (Kreiman et al., 1993; Yamasaki et al., 2017). However, perceptual parameters also show limitations in listener reliability, so it is advised to combine both perceptual and objective measures when assessing vocal quality (Barsties & De Bodt, 2015; Oates, 2009), which was reported in 11 included studies (46%). Information on listener reliability is scarce in the included articles. Only three studies reported inter- or intra-rater reliability, suggesting that future research should pay more attention to reporting measures of listener reliability.

Recently, the importance of patient-reported outcome measures has been recognized as patients can provide valuable information that cannot be obtained in any other way (Deshpande et al., 2011). This includes details about the medical condition, such as presence and severity of invisible symptoms, as well as insight into patient's quality of life. In this systematic review, patient-reported outcome measures are used in only six papers (25%). Strictly, one of them (Hartnick et al., 2018) used an observer reported outcome measure as the PVRQOL is completed by a parent of the dysphonic child. In the field of paediatric voice disorders, the question can be raised if children are capable of reporting information about their own health and voice. According to a report of the Oxford PROM group, children older than 5 years can self-report their health with assistance of an adult and children older than 8 years are competent to complete health related questionnaires (Morris et al., 2009).The majority of the validated tools to assess children's voice-related quality of life are parent-proxied in nature, but some self-reported questionnaires are already available such as the child version of the Pediatric Voice Symptom Questionnaire (Verduyckt et al., 2012) and the Children's Voice Handicap Index—10 (Ricci-Maccarini et al., 2013). Future research should focus on the development and validation of self-reported assessment tools for dysphonic children. In this way, patient-reported outcome measures can be used complementary to perceptual and objective parameters when investigating paediatric dysphonia.

The results of this systematic review suggest that there is some evidence for a positive effect of voice therapy in children with VFNs. A considerable amount of the included studies demonstrate a significant improvement for at least one outcome measure after voice therapy. Moreover, it is noteworthy that no study reports a significant deterioration of a single voice parameter after voice therapy. However, some included studies could not indicate a significant effect of voice therapy and not all authors tested for significance in their paper.

Unfortunately, it is impossible to determine the active ingredients of a successful intervention due to the eclectic nature of the provided therapy programmes, the lack of a comprehensive and accurate description of the therapy content, the adaptation of voice therapy to the abilities and specific needs of individual patients, methodological differences between the included studies, and methodological shortcomings of the individual studies. The difficult linkage between changes in voice characteristics and therapy components is also described in the systematic review of Alegria et al. (2020) and Feinstein and Abbott (2021). This is a widely recognized problem in behavioural treatment studies (Hart et al., 2014).

Voice therapy results may not only depend on the provided therapy techniques. Filter and Poynor (1982) note that improvements of patients are related to the abilities, skills, and competencies of the clinician. In the qualitative research of Braden et al. (2018), parents underline the importance of the clinician being an expert and being capable to manage the behaviour of their child. Moreover, the results of this study suggest that ‘given the child's propensity for learning, treatment success appears largely driven by the clinician's ability to adapt treatment to the child's developmental level and interests and the parent's understanding and support of the process’. The importance of the practitioners’ expertise is also emphasized in the evidence based practice model (Sackett et al., 1996). Another frequently discussed factor is the degree of adherence limited to the extent in which patients hold on to clinicians’ recommendations (van Leer

& Connor, 2010). Verdolini Abbott (2013) notes that many children are at risk for poor voice therapy compliance. However, Hartnick et al. (2018) reported rather high compliance rates (85.4% in the direct therapy group and 91.2% in the indirect therapy group) and did not find a significant relationship between compliance and outcomes for voice therapy in children with VFNs.to voice therapy because it can be assumed that effectiveness of voice therapy is

Some limitations must be taken into account while interpreting the results. First, this review is at risk for language bias because only English articles are included. Relevant articles published in other languages may be missing. Second, the methodological quality of the included study was assessed by only one evaluator. It had been better if a second researcher was involved in this quality assessment. Third, it is remarkable that all included studies report positive changes after voice therapy, which causes concern about a possible publication bias. It is generally known that negative results are less likely to get published (DeVito & Goldacre, 2019). Lastly, there is great heterogeneity across the different studies in terms of voice therapy content and outcome characteristics hampering comparisons between the papers and preventing the authors to perform a meta-analysis.

CONCLUSIONS

In conclusion, the effect of voice therapy in children with VFNs has been studied several times in the last 60 years, although more attention has been paid to this topic lately. Voice therapy seems to be effective in treating paediatric patients with VFNs, given the fact that a considerable number of included studies report significant improvements after voice therapy. However, not all studies find significant effects of voice therapy. Based on this systematic literature review, it is not yet clear which therapy techniques and therapy dosage achieve the best results and are therefore preferable. Quality assessment using the QualSyst tool showed that the overall quality of the included studies is adequate (55%). Experimental research on effects of voice therapy in children with VFNs is still scarce. In sum, there is an urgent need for well-designed effectiveness studies, especially RCTs, to determine the active ingredients and preferred dosage of a successful intervention for paediatric VFNs.

CONFLICT OF INTEREST

The authors declare that there is no conflict of interest.

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