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ORIGINAL ARTICLE

Effectiveness of nasopharyngoscopic biofeedback in clients with cleft palate speech—a systematic review

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Abstract

Objective. To conduct a systematic review analyzing the effectiveness of nasopharyngoscopic biofeedback in clients with cleft lip and palate and velopharyngeal dysfunction. *Method.* Extensive electronic search and analysis of the databases of Cochrane Library, MEDLINE, EMBASE, ERIC, PsycInfo, CINAHL, AMED, Journals@Ovid, and German Databases, including all papers published since 1970 plus a manual search of the *Cleft Palate-Craniofacial Journal* (1970-3/2010). *Results.* Six studies met the inclusion criteria. Their analysis reflects a low level of evidence and a broad heterogeneity concerning age range, intervention methods, and outcome measurement. *Conclusion.* The analyzed studies show that nasopharyngoscopy may be effective only in combination with traditional speech therapy in helping patients with cleft palate speech optimize their velopharyngeal closure in articulation, but the quantity and quality of studies are limited.

Key words: *Biofeedback, cleft lip and palate, cleft palate speech, cleft palate, compensatory articulation, nasopharyngoscopy, speech therapy, velopharyngeal dysfunction, velopharyngeal insufficiency*

Introduction

Quality management and evidence-based practice are crucial to therapy and research of speech-language pathology (SLP). However, surveying the publications on SLP reveals a continued lack of systematic reviews on the effectiveness of therapies for specific speech and language disorders, particularly cleft palate speech.

Velopharyngeal insufficiency (VPI) in patients with repaired cleft lip and palate (CLP) is generally treated by secondary surgical methods like pharyngeal flap or sphincter pharyngoplasty (1). Also prosthetic treatment in the form of speech bulbs and palatal lifts is used to aid velopharyngeal closure. In cases of phoneme-specific VPI, compensatory articulation, and/or nasal fricatives, speech-language therapy is conducted to improve speech intelligibility (2–5). In a recent systematic review (2) concerning therapy methods and their efficiency in patients with repaired CLP the following speech-language intervention methods could be detected: early intervention by inclusion of parents, articulation therapy, phonological intervention, intensive speech-language therapy (summer schools), biofeedback therapy (Nasometer®, KayPentax, USA, videofluoroscopy), continuous positive airway pressure (CPAP), and prosthetic treatment in combination to traditional speech therapy (2).

Speech therapy for VPI in patients with repaired CLP and/or where VPI persists in phoneme-specific or intermittent forms is often regarded as difficult or unending (3–5). Many cases are cited where velopharyngeal closure (VPC) is adequate (or borderline) in swallowing or blowing, but insufficient in articulation due to faulty or maladaptive sound learning (3,4). Another difficulty found among cleft palate patients is the phenomenon of outward displacement of the lateral pharyngeal wall (LPW) during speech production in association with compensatory articulation (6), e.g. 'None of the muscles of the velopharyngeal sphincter appears to be responsible for this aberrant displacement. A possible explanation is a passive displacement in response to air pressure at the vocal tract' (6, p. 297).

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For many years, speech therapy treatment of VPI has generally focused on improving velopharyngeal muscle function during sound production (7). However, as Brunner et al. (8) point out, improving such muscle function is difficult when clients lack the 'inner percept' to do so. Like Siegel-Sadewitz and Shprintzen (9), they describe visual therapy methods—such as nasopharyngoscopy—as providing an appropriate source of feedback (8,9).

Thus, not only does nasopharyngoscopy with a flexible endoscope represent an effective tool for assessing the function of the velopharyngeal sphincter in clients with a CLP condition, it may also be used as a supplemental therapy intervention, in addition to traditional speech therapy (10). As an intervention tool, nasopharyngoscopy is conducted only in clinical settings by a speech-language pathologist or phoniatrician using a flexible fiberoptic nasopharyngoscope with a small diameter (3–4 mm). Patients are generally older than 9 years, because of better compliance and ability to use the visual cues. The subjects are requested to articulate target sounds while seeing their velopharyngeal sphincter on a monitor. By this visual feedback they try to modulate their VPC to adequate closure in articulation supported by the SLP (5).

Various handbooks and textbooks describe this biofeedback therapy method as providing the possibility of improving VPC in connected speech, minimizing compensatory articulation, and activating lateral wall movement following pharyngeal flap surgery (3,5). In particular, it may be particularly useful among patients who have problems with auditory feedback, providing them visual feedback that allows them to learn and eventually achieve sufficient VPC for articulation (5). Nevertheless, despite the recommendations of various authors, the evidence base does not immediately appear strong enough to support this special biofeedback method.

Thus, a systematic review of journals was undertaken to determine the strength of evidence in support of nasopharyngoscopic biofeedback (NPB) among patients—of any age—with CLP and VPI/articulation disorders.

Methods

Eligibility criteria

All studies were included that concerned cleft palate speech and nasopharyngoscopic treatment and were published in international journals from 1970 to the present. No restrictions were made based on the language of the publication. In order to gain an overview of results and the quality of studies available according to evidence-level criteria, no restrictions were made based on study design (11). Subjects of any age/sex with repaired CLP (irrespective of type of cleft) and resistant VPI, hypernasality, accompanying nasal emission or nasal turbulence, and compensatory articulation were considered for inclusion. Only studies involving nasopharyngoscopy as a therapy were included, but restrictions were not made regarding the exact intervention type. The types of outcome measures included were nasopharyngoscopy, videofluoroscopy, nasometer, articulation testing, and self-perception questionnaires, depending on the focus of the study. The primary outcomes included activation of LPW and VPC in articulation, reduction of hypernasality, nasal emission, or nasal turbulence, as well as improvement of articulation or intelligibility in connected speech.

Search methods for identification of studies

Studies were identified by searching electronic databases, scanning reference lists of articles, and by hand searching, without any language restrictions. Papers were identified from the following sources: Cochrane databases, AMED (1985-04/2010), BIOSIS (1985-04/2010), EMBASE (1960-04/2010), ISTPB & ISTP/ISSHP (1998-04/2010), MEDLINE (1960-04/2010), PsycInfo (1960-04/2010), Psyndex (1977-04/2010), SciSearch (1974-04/2010), SocialSciSearch (1973-04/2010); and German publication databases Hogrefe (1999-04/2010), Karger (1998-04/2010), Thieme (1981-04/2010), and Krause and Pachernegg (1998-04/2010). Search terms covered the key words cleft lip and cleft palate, biofeedback, nasendosc*, nasopharyngosc*, videonasopharyngosc*, speech therapy, treatment, and intervention. The search terms were used individually and in combination. An example of a search strategy is illustrated in Figure 1.

The *Cleft Palate-Craniofacial Journal* (1970-3/2010) was searched by hand. The reference lists of relevant articles were also reviewed to identify other published and unpublished studies that might have been overlooked. No translations were necessary since only English and German studies were found.

Data collection and analysis

Studies' eligibility for inclusion was independently assessed by two reviewers in an unblinded, standardized manner. A data extraction database was developed, pilot-tested on three randomly selected studies, and accordingly refined. One reviewer extracted the data and the other reviewer checked the data. Allocation of evidence level was determined by consensus agreement between the two reviewers.

Search strategy: MEDLINE (20.10.2010)

1. cleft palate
2. cleft lip
3. 1 OR 2
4. speech therapy
5. speech treatment
6. speech intervention
7. 4 OR 5 OR 6
8. 3 AND 7
9. biofeedback
10. 8 AND 9
11. nasopharyngosc*
12. nasoendosc*
13. videonasopharyngosc*
14. 11 OR 12 OR 13
15. 10 AND 14

Figure 1. Example of search strategy.

The articles identified were analyzed considering 153 data items arranged according to a PICOS scheme (12), providing structured information about participants (P), intervention (I), comparators (C), outcomes (O), and study design (S): level of evidence (9); specific biofeedback goal (e.g. articulation improvement); inclusion/exclusion criteria; number, age, and type of participants; control group (+/–, number and age of controls); treatment design (time, material, procedure); outcome assessment; and limitations/risk of bias. Predictive Analysis SoftWare (PASW 18, IBM) was used for both quantitative and qualitative analyses of data. Preferred reporting items for systematic reviews and meta-analyses (PRISMA) checklist (12) criteria were used to summarize the results.

Results

Study selection

A total of 296 studies were initially identified for inclusion in the review: A search of databases provided a total of 286 citations, while a manual search of reference lists identified 10 more studies. After adjusting for duplicates, 79 studies remained and were screened further. Of these, 63 were excluded after a review of their abstracts revealed that they were not therapy studies. The full texts of the remaining 16 citations were examined in greater detail, at which point 10 more studies were excluded. In the end, only six studies met the criteria for inclusion and were subjected to further analysis (Figure 2).

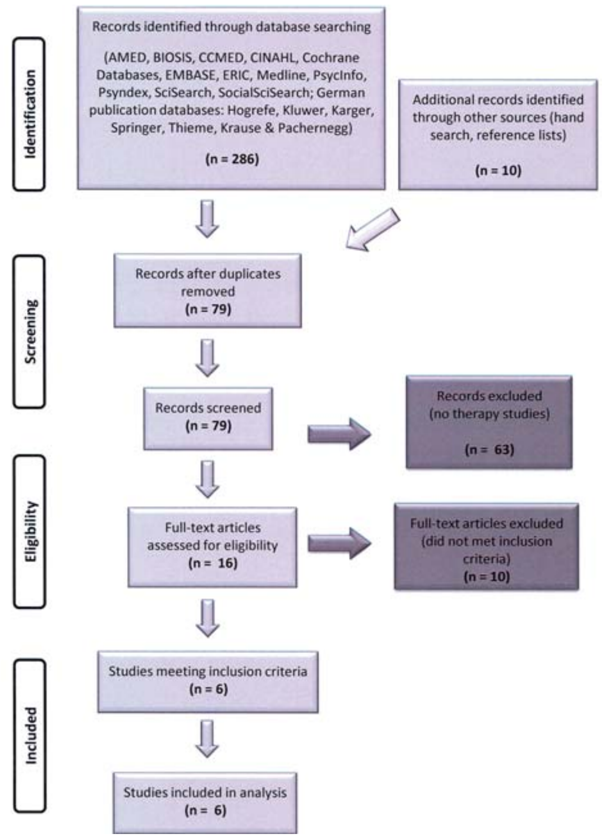

Figure 2. Flow chart of study selection.

Characteristics of included studies

Methods. Of the six included studies, only one was a randomized control trial (RCT); the others were single-case studies or case-series studies of 1 to 59 participants, without control groups. As a result, the evidence level was very low among the therapy studies identified (level 4) (Table I) (11).

Participants. The included studies comprised 83 participants—aged 7 to 50 years—with repaired CLP. All participants showed VPI in articulation. Inclusion criteria ranged from occurrence hypernasality, nasal emission, and/or compensatory articulation to reduced intelligibility (Table II).

Interventions. None of the studies analyzed was multicenter. The nasopharyngoscopic biofeedback therapy was generally conducted by a speech-language therapist (SLT) ($n = 4$), except for one case where it was done by a psychologist and another case where it was done by a surgeon; all who conducted the treatment were members of a regular cleft palate team. Treatment methods differed in terms of the number of therapy stages and the overall objective. The length of individual therapy sessions varied from 20 minutes to 1 hour, with some interventions lasting only one session and others encompassing multiple

Table I. Levels of evidence by Centre for Evidence-based Medicine (Phillips et al. 2011) (11).

Evidence level	Therapy
1a	SR (with homogeneity [a]) of RCTs
1b	Individual RCT (with narrow confidence interval)
1c	All or none [b]
2a	SR (with homogeneity [a]) of cohort studies
2b	Individual cohort study (including low-quality RCT; e.g. $<80\%$ follow-up)
2c	'Outcomes' research; ecological studies
3a	SR (with homogeneity [a]) of case-control studies
3b	Individual case-control study
4	Case series (and poor-quality cohort and case-control studies [c])
5	Expert opinion without explicit critical appraisal, or based on physiology, bench research, or 'first principles'

SR = systematic review; RCT = randomized controlled trial.

[a]By homogeneity we mean a systematic review that is free of worrisome variations (heterogeneity) in the directions and degrees of results between individual studies. Not all systematic reviews with statistically significant heterogeneity need be worrisome, and not all worrisome heterogeneity need be statistically significant. Studies displaying worrisome heterogeneity should be tagged with '–' at the end of their designated level.

[b]Met when all patients died before the Rx became available, but some now survive on it; or when some patients died before the Rx became available, but none now die on it.

[c]By poor quality cohort study we mean one that failed to clearly define comparison groups and/or failed to measure exposures and outcomes in the same (preferably blinded) objective way in both exposed and non-exposed individuals and/or failed to identify or appropriately control known confounders and/or failed to carry out a sufficiently long and complete follow-up of patients. By poor-quality case-control study we mean one that failed to clearly define comparison groups and/or failed to measure exposures and outcomes in the same (preferably blinded) objective way in both cases and controls and/or failed to identify or appropriately control known confounders.

sessions spread out over many months. The frequency of biofeedback sessions reported ranged from a single session, to one session a week for over one year, to twice a week for six months. Thus, there was a great deal of heterogeneity in this area.

Outcomes. Outcome measurements included flexible fiberoptic nasopharyngoscopy (6,8,13,14,16,17,18, 19) itself, multiview videofluoroscopy (6), self-perception sheets (8), articulation proficiency tests (19), perceptual analysis (14), nasometer (14), and mirror test (14). Blinding of data collectors was not reported in any studies. Only one study (8) reported blinding of outcome assessors. Assessment by nasopharyngoscopy was performed according to the standardization for the reporting of nasopharyngoscopy and multiview videofluoroscopy by Golding-Kushner et al. (15) in only one study (6). The timing of outcome measures varied, including assessments before and after each training term (19,16) or after ten sessions (14). Only half of the studies ($n = 3$) reported a follow-up evaluation to determine the stability of results over time: the reported follow-ups occurred 12 weeks after the final session (14), six months after the final session (8), one year after the final session (17), and after an unknown period of time (6).

Results of individual studies

In the following section, the results of individual studies are summarized according to a PICOS scheme (12), providing structured information about participants (P), intervention (I), comparators (C), outcomes (O), study design (S), and biofeedback method. The studies are presented chronologically.

Yamaoka et al. (1983)

Objective(s). The objectives of this study were to determine the efficiency of NPB as a self-training method for activating velopharyngeal movement in patients with cleft palate speech and to demonstrate longitudinal changes in VPC.

Study design. It was a case-series study, which is allocated an evidence level of 4 (Table I) (11).

Participants. The therapy study included 59 Japanese hypernasal speakers (aged 8–45 y) with repaired (V-Y push back < 2 y) CLP (39 CP, 16 CSP, 4 SMCP). They all displayed persistent VPI despite long-term speech therapy (exact duration unknown) in infancy, and they displayed VPC during swallowing. To compare results, the patients were divided into five groups (Ib–IV) (19,18) according to occurrence of VPI in different tasks (Table III). These experimental tasks to assess VPC or VPI visually were defined as follows: swallowing, blowing a carnival blower continuously (blowing), producing Japanese vowels /i, w, e, o/ alone in a sustained manner (phonation of vowels), producing Japanese consonants /p, b, t, d, k, g, s, dz/ in CV syllables (phonation of consonants) (Table III).

Table II. Patient-inclusion criteria and study design of detected studies.

Source	Study design	No. of patients and etiology	Age and range (years)	Inclusion criteria	Evidence level
Brunner et al. 1994	Single-case study	1 UCLP	25	Long-term conventional speech therapy, VPI, CA, reduced intelligibility	4
Brunner et al. 2005	Case series	11 CP: $n = 4$ CLP: $n = 6$ BCLP: $n = 1$	7;0–30;0 (mean 14;02)	VPI, VPC possible in at least 1 speech sound, tolerance of endoscopic procedure, positive stimulability testing during endoscopy, closure of soft palate: 1–3 y, hard palate: 4 y, previously conventional speech therapy without showing significant improvement	4
Witzel et al. 1988	Single-case study	1 BCLP	10	Inconsistent VPI in articulation after 4 years of conventional speech therapy	4
Witzel et al. 1989	Case series	3 BCLP: $n = 2$ CP: $n = 1$	34–50	Persistent hypernasality, nasal emission, and VPI after pharyngeal flap surgery/revision in adulthood	4
Yamaoka et al. 1983	Case series	59 CP: $n = 39$ CSP: $n = 16$ SMCP: $n = 4$	8–45	Normal hearing, persistent VPI despite long-term conventional speech therapy in infancy, VPC in swallowing	4
Ysunza et al. 1997	RCT	8 UCLP 9 UCLP (control group)	mean 11;11 mean 11;09	Repaired UCLP, non-syndromic, CP width I–II, VPI, NMLPW, no fistula, consistent CA, normal hearing and language development	2-b

BCLP = bilateral cleft lip and palate; CA = compensatory articulation; CLP = cleft lip and palate; CP = cleft palate; CSP = cleft soft palate; NMLPW = negative movement of the lateral pharyngeal walls; SMCP = submucous cleft palate; UCLP = unilateral cleft lip and palate; VPC = velopharyngeal closure; VPI = velopharyngeal incompetence.

Interventions. Each group underwent the same NPB self-training, with sessions lasting 1 hour and occurring every two weeks for over a year. Occasionally some patients ($n = ?$)—but not all—received traditional speech therapy parallel to the biofeedback sessions to correct misarticulation.

Biofeedback method. The participants were asked to try to close the velopharyngeal sphincter during sound articulation while observing their sphincter on a TV monitor. If the patient showed incomplete VPC the SLP assisted him/her during the exercises by giving some instructions.

Table III. Categories of participant groups according to the amount of velopharyngeal closure or VPI achieved in four kinds of activity (17, p. 192).

Category	n	Phonation of vowels	Phonation of consonants	Blowing	Swallowing
I-b	8	−	+	+	+
I-c	3	−	±	+	+
II	12	−	−	+	+
III	4	−	±	−	+
IV	32	−	−	−	+

+ indicates complete closure; ± indicates complete closure in phonation of some of the pressure consonants or some of the vowels; − indicates incomplete closure.

Comparators. The study design did not include any comparators.

Outcomes. As an outcome measurement (before and after each self-training term), videorecorded nasopharyngoscopy (using a nasopharyngoscope with a diameter of 3.4 mm) was used to identify VPC or VPI, and the results were defined based on the consensus decision of two authors. Articulation proficiency tests were also used.

The study authors summarized the results (grouped as described above in Table III) as follows: In group IV an improvement of VPC could be seen in 37.5% of patients; 50% of patients showed improvement in group III; and 83.3% of patients showed improvement in group II. In group I-b and group I-c a complete VPC was detected in 100% of the patients. Articulation proficiency tests revealed that 59% of the patients displayed improvements characterized by a general decrease in nasality when articulating vowels and a general decrease in distortion of non-nasal consonant production.

Witzel et al. (1988)

Objective(s). The stated objective of the study was to determine whether use of nasopharyngoscopic biofeedback as a visual feedback tool helps correct inadequate VPC in production of sibilant-fricative phonemes.

Study design. It was a single-case study, which is allocated an evidence level of 4 (11).

Participant. One 10-year-old girl with repaired bilateral cleft lip and palate. She exhibited a severe mid-face hypoplasia and an anterior cross-bite. Concerning articulation she had complete VPC for all phonemes except for sibilant-fricatives, especially for /s/ (inconsistent phoneme-specific VPI), with accompanying nasal emission after four years of speech therapy at school.

Intervention. The girl was offered one session (duration unknown) of nasopharyngoscopic biofeedback during articulation of /s/, using a flexible fiberoptic nasopharyngoscope (ENF-P Olympus, USA) with a diameter of 3.7 mm and a bending radius of 180 degrees; she was instructed to use the /t/ phoneme before producing /s/ and to focus on VPC. After this biofeedback session, traditional speech therapy was conducted to improve the manner and place of /s/ production, because the girl continued to have an oral sibilant-fricative distortion related to dental malocclusion.

Comparators. The study design did not include any comparators.

Outcomes. As an outcome measurement, nasopharyngoscopy was used at the end of the session and also six months after surgical repositioning of maxilla. The patient progressed from no velopharyngeal movement during production of sibilant-fricative phonemes to consistent VPC during connected speech after one session of NPB.

Witzel et al. (1989)

Objective(s). The objective of this study was to determine the effectiveness of NPB for improvement of VPC by movement of lateral pharyngeal walls (LPW) in articulation and connected speech.

Study design. It was conducted as a case-series study, which is allocated an evidence level of 4 (11).

Participants. Three subjects with cleft lip and palate, persistent hypernasality, nasal emission, and VPI following velopharyngeal flap surgery/revision in adulthood. Subject 1 was a 50-year-old man with repaired bilateral cleft lip and palate who had undergone pharyngeal flap surgery nine weeks before the start of the biofeedback therapy. Subject 2 was a 37-year-old woman with repaired bilateral cleft lip and palate who had undergone pharyngeal flap revision seven months before the start of the biofeedback therapy. Subject 3 was a 34-year-old woman with repaired cleft palate who had undergone pharyngeal flap revision two months before the start of the biofeedback therapy. The final subject dropped out after five sessions for personal reasons.

Interventions. The three subjects differed regarding the duration and frequency of their NPB. Subject 1 received NPB once a month for four months with each session lasting 30 minutes, in addition to traditional speech therapy on a weekly basis. Subject 2 only received NPB on two occasions with each session lasting 30 minutes; the lapse of time between the two sessions was not reported. Subject 3 received NPB five times—with one-to-two-month intervals—in addition to traditional speech therapy sessions; the exact frequency of the sessions was not reported.

Biofeedback method. The method used was described as follows: 1) Identification of sounds with most velopharyngeal sphincter motion or closure; 2) visual feedback of VPC in blowing and cueing; and 3) production of isolated sounds to consonant-vowel combinations and cueing.

Comparators. The study design did not include any comparators.

Outcomes. The outcome measure and biofeedback tool used was an end-viewing flexible fiberoptic nasopharyngoscope (Olympus ENF-P) with a diameter of 3.7 mm and a bending radius of 180 degrees. The sessions were videorecorded using a high-resolution camera (DXC 1850 Sony, Germany). The authors summarized the outcomes as follows:

Subject 1 consistently demonstrated complete closure of velopharyngeal ports during connected speech as well as elimination of nasal emission and hypernasality, assessed in follow-up nasopharyngoscopies two-and-a-half months and one year after the final session.

Subject 2 consistently displayed complete VPC during connected speech as well as elimination of hypernasality and extensive nasal air emission.

Subject 3 began to control medial movement of the lateral pharyngeal walls (LPW) to close the velopharyngeal sphincter during her first NPB session. In the course of the next four sessions, she achieved good LPW movement by swallowing. After five sessions, she displayed increased control and coordination of the velopharyngeal mechanism. Her articulation was improved by correcting her tongue position for /k/ (previously: glottal stop) and in the production of sibilant-fricative sounds (previously: pharyngeal fricatives).

Brunner et al. (1994)

Objective(s). The objective of this study was to evaluate whether NPB—infravelar view—would improve

VPC during articulation in a male subject with repaired unilateral cleft lip and palate.

Study design. It was conducted as a single-case study, which is allocated an evidence level of 4 (11).

Participant. The participant was a 25-year-old man with repaired unilateral cleft lip and palate, a long history of speech therapy, but no velopharyngeal flap surgery. He displayed pharyngeal articulations for sibilant fricatives and palatal articulation for velars with accompanying VPI, reducing his intelligibility and therefore restricting his social participation (Table II).

Interventions. The participant received ten 30-minute NPB sessions (frequency unknown) using a flexible nasopharyngoscope (type unknown) according to a three-stage process: 1) Experimentation phase by infravelar view (mesopharyngeal level) to get a view on non-oral articulation, with coupling of visual and proprioceptive perception of appropriate articulation; 2) Anchoring phase using the primary perceptual learning channel; and 3) Automatization phase transferring accurate sounds into spontaneous speech at home using specially developed documentation sheets.

Comparators. The study design did not include any comparators.

Outcomes. The outcome measurements used were nasopharyngoscopy, perceptual analysis, nasometer (type unknown), and a mirror test directly following ten therapy sessions. An additional follow-up evaluation was conducted after three months. The outcomes were summarized as follows: The participant displayed complete VPC in connected speech, but not in spontaneous speech. In the three-month follow-up, improved articulation of fricatives or sibilants and correct tongue placement were observed.

Ysunza et al. (1997)

Objective(s). The objective of this study was to find out whether nasopharyngoscopy is effective in correcting negative lateral pharyngeal wall movements (NMLPW) and compensatory articulation (CA) during speech in children with repaired unilateral cleft lip and palate (UCLP).

Study design. It was a low-quality randomized controlled trial (RCT), because there was no blinding of data collectors and outcome assessors, and there were missing data concerning the intervention method. This might be considered an evidence level 2b (Table I) (11).

Participants. The experimental group included eight participants with a median age of 11;11 years. Inclusion criteria were described as follows: repaired UCLP without fistula; non-syndromic, width of cleft palate: I–II, VPI, NMLPW; consistent compensatory articulation (CA); normal hearing and language development (Table II).

Comparators. The comparator group included nine children with a median age of 11;09 years. The inclusion criteria were the same as those for the experimental group. The participants were randomly selected.

Interventions. Participants: The experimental group received 25-minute NPB sessions twice a week for 12 weeks using phoneme samples. In addition, they received three 60-minute sessions of traditional speech therapy each week for a year to correct CA. After six months, all participants underwent a tailor-made pharyngeal flap surgery.

Comparators. Initially, the comparator group solely received three 60-minute sessions of traditional speech therapy per week to correct CA (over the course of a year). However, after 12 weeks, the authors incorporated NPB into their therapy too. The comparator group received speech therapy from the same speech-language pathologist and according to the same methods as the experimental group. Participants in the comparator group also underwent a tailor-made pharyngeal flap surgery six months after the start of therapy.

Outcomes. All participants were followed until CA had been corrected during isolated speech using nasopharyngoscopy and multiview videofluoroscopy as outcome measurements. The authors described the results as follows: All participants in the experimental group displayed significant improvement in LPW movement after 12 weeks. At the same point in time, outward displacement of LPW was still present in the comparator group (in eight of nine children), despite partial improvement of CA.

After six months, all 17 children (both groups) had completely corrected CA during isolated speech. All participants received continued speech therapy following their pharyngeal flap surgery. According to the authors, more than a year later (exact point in time unclear) all patients displayed completely normal articulation in connected speech.

Brunner et al. (2005)

Objective(s). The objective of this study was to evaluate the immediate, long-term, and carry-over effects on words and sentences of NPB in children and adults with repaired cleft palate and VPI. It also sought specifically to determine whether NPB differs in its effectiveness among patients with general VPI versus those with phoneme-specific VPI.

Study design. It was a case-series study, which is allocated an evidence level of 4 (Table I) (11). It implemented pre- versus post-treatment and follow-up comparisons.

Participants. The study comprised 11 patients (4 with cleft palate, 6 with unilateral cleft lip and palate, and 1 with bilateral cleft lip and palate) with VPI when articulating speech sounds and words. Each had received conventional speech therapy without showing significant improvement. They ranged in age from 7;0 to 30;0 years, with a mean age of 14;02 years. They had received closure of the soft palate between 1 and 3 years and closure of the hard palate at about age 4 years. Selection criteria included tolerance of endoscopic procedure, positive result of stimulability testing, and the ability to achieve VPC in at least one speech sound. Prior to NPB therapy, the mean occurrence of VPC in articulation was only 5% among all patients. One patient dropped out after the second session of the first target sound.

Interventions. For the biofeedback therapy, a flexible nasopharyngoscope (Olympus ENF Type P3, Lake Success, NY) with diameter of 3.5 mm was used together with a video recorder and two monitors. Participants received two biofeedback sessions—spaced six weeks apart—per target sound. The number of sessions completed by participants ranged from 2 to 16 sessions (Figure 3). The average duration of visual feedback sessions was 20 minutes.

The biofeedback therapy was conducted according to a standardized four-stage process, emphasizing training of target sounds in isolation and on the syllable level. This four-stage process comprised the following areas (20): 1) Introduction: learning velopharyngeal structures and functions; 2) Experimentation: self-monitoring of articulation movements guided by a speech-language pathologist; 3) Coupling: anchoring (psychological) motor perception and auditory perception by guided relaxation; and 4) Automatization: overlearning by means of a diary sheet (8).

The authors recommended the patients to continue their traditional speech therapy to practice what they learned in the biofeedback training, but this was not part of the study.

Comparators. The study design did not include any comparators.

Outcomes. The study implemented two outcome measures: patients' self-perception and nasopharyngoscopy. The latter displayed an inter-rater reliability ($n = 3$) of 91%. The outcomes were as follows: The mean occurrence of VPC was 91% after two sessions ($P < 0.001$) and 86% at six-month follow-up ($P = 0.000$). A significant stability of transfer to word and sentence level ($P < 0.001$) was found at six-month follow-up. Aside from that, no significant improvement ($P = 0.566$) between phoneme-specific VPI (improvement of 87%) and generalized VPI (improvement of 95%) could be detected. The improvement in transfer of articulation to words and sentences was a success. Concerning sounds in initial word position as well as final position, the improvement of VPC was more than 90% at six-month follow-up, displaying statistical significance when compared to the pre-therapy assessment ($P < 0.0001$). The result was the same in terms of transfer to sentences. Analyzing patients' self-perception sheets, nine of ten patients

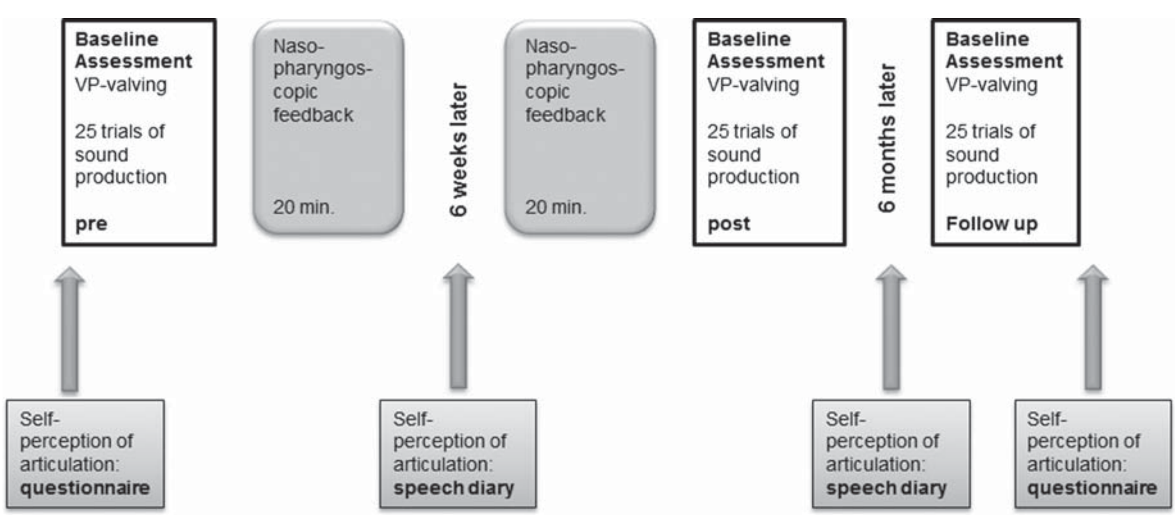

Figure 3. Time line Brunner et al. (2005) (8).

considered themselves capable of controlling their speech production after the NPB therapy.

Risk of bias within the studies

Analysis of the selected studies revealed many limitations in terms of their underlying methodology, study design, elaboration of the biofeedback intervention, documentation of results, and blinding of outcome assessors.

Table IV provides an overview of bias risk among the studies analyzed. On balance, the studies were good in terms of completeness in reporting outcomes (free of selective reporting) and absence of dropouts ($n = 4$) due to study design. However, data collectors were not blinded in the studies, and blinding of outcome assessors only occurred in one (8) of the six studies. All studies analyzed showed missing or unclear data with respect to study design. The following section describes the limitations of the individual studies in greater detail.

The case-series study of *Yamaoka et al. (1983)* provided no information on the gender or exact age of participants (only age groups), the age of participants according to groups, or the type of VPI relating to articulatory distortions. Further, it did not specify the exact training method of NPB used and did not account for the effect of additional traditional speech therapy in some patients. Regarding outcome measures, no information was provided regarding timing, and no follow-up data were reported concerning transfer of sound improvement into word level or connected speech (19).

In their first paper, *Witzel et al. (1988)* did not provide any information on the number or duration of therapy sessions. They also failed to account for the effect of additional traditional speech therapy in their results (16).

The same was true of *Witzel et al. (1989)*, namely, the effect of additional speech therapy was not discussed. Further, no standardized therapy regimen was used, and there were missing data due to one participant ($n = 1$) dropping out (7).

No information about the frequency of therapy sessions could be found in the paper of *Brunner et al. (1994)* (14).

The RCT of *Ysunza et al. (1997)* also displayed limitations. Though randomization was performed, no information was provided on the manner used. Data collectors and outcome assessors were not blinded. The speech-language pathologist was the same for both the experimental and the comparator group. The study design was not precisely described, leaving the dates of surgery and duration of therapy unclear (6).

The study of *Brunner et al. (2005)* was well described, but the sample of patients was small, and generalization effects were not systematically controlled. The authors themselves criticized the lack of systematic assessment of perceptual data for each sound in the documentation of motor control of VPC. Here too, data were missing due to one participant ($n = 1$) dropping out (8).

Discussion

Synthesis of results

In 1983, *Yamaoka et al.* found a latent ability of achieving VPC in 60% of patients aged 8 or older following NPB. The authors suggested that NPB self-training provides a strong neuromuscular signal and establishes a sense of optico-muscle awareness for VPC. They describe NPB as a useful tool for improving VPC in speech-language therapy by way of visual feedback control (19).

Table IV. Risk of bias in individual studies.

Source	Concealment of randomization	Blinding: Data collectors	Blinding: Outcome assessors	Complete outcome data (no drop-out)	Complete outcome reporting (no selection)	Complete methodical documentation
Brunner et al. 1994	✗	✗	✗	✓	✓	✗
Brunner et al. 2005	✗	✗	✓	✗	✗	?
Witzel et al. 1988	✗	✗	✗	✓	✓	✗
Witzel et al. 1989	✗	✗	✗	✗	✓	?
Yamaoka et al. 1983	✗	✗	✗	✓	✓	✗
Ysunza et al. 1997	?	✗	✗	✓	✓	✗

✗ = high risk of bias; ? = risk of bias unclear; ✓ = low risk of bias.

Five years later, *Witzel et al. (1988)* found that NPB therapy was helpful in correcting inconsistent VPI. They confirmed NPB as a valuable method for revising the sound-learning process by providing *visual* cues that are missing in traditional techniques. They suggested that NPB could be useful in helping children with CLP achieve consistent VPC during production of sibilant-fricative phonemes, including connected speech (16).

In their study concerning adults with cleft lip and palate, *Witzel et al. (1989)* summed up by stating that NPB is useful in motivating patients to achieve better VPC during connected speech following pharyngeal flap surgery or revision. They point to the advantage of NPB in enabling visual input into therapies as a complement to auditory feedback, particularly useful among older patients with long-standing and ingrained compensatory speech patterns (17).

Brunner et al. came to the same conclusion in 1994, claiming that NPB appears to be an appropriate therapy method to complement traditional speech therapy, especially in patients with CLP and poor auditory perception (14).

Concerning modification of aberrant lateral pharyngeal wall motion associated with compensatory articulation, *Ysunza et al. (1997)* found NPB to be a safe and reliable procedure for helping cleft palate patients (6).

The most recent study by *Brunner et al. (2005)* found NPB to be a quick and effective method for changing VPI, one that displays stable results and carry-over effects. The stability of therapy effects at six-month follow-up suggested implementation of new and effective self-monitoring techniques among participants (8).

Limitations of the review

The authors of the present review did not implement any book chapters, oral presentations on NPB, or papers without an English abstract in the present review, possibly limiting the summary of results.

Critical discussion of results

Current empirical support for the effectiveness of NPB is not strong in terms of high-level evidence. Only one low-quality RCT could be found, in addition to several case studies and case series without control groups. The studies analyzed display a number of limitations with respect to their methodological design, including the lack of adequate descriptions of VPI and speech presentations, incomplete description of the intervention, mix of therapy methods, heterogeneity of participants, and absence of outcome measures.

Nevertheless, the analyzed studies describe NPB, usually supported with additional traditional speech therapy, as an effective therapy method for helping people with cleft palate speech optimize their VPC in articulation. Only one study (14) implemented NPB exclusively as therapy method; the others included or accepted traditional speech therapy going on in parallel. All authors described that their patients had had dissatisfying conventional speech therapy before, some for years. Then, when adding NPB as biofeedback therapy, there was a change in VPI in articulation. While these preliminary results point to NPB's promise as a therapy method, the current state of knowledge remains limited. It is not possible to attribute effectiveness solely on NPB when there is a mixture of therapy methods. The RCT by Ysunza et al. (6) stopped effectively after 12 weeks, because after that the same treatments were given to both the study and the control groups. So, the results gained were a combination of effectiveness of NPB, conventional speech therapy, and secondary surgery—and cannot be separated.

Also there is not enough known about the stability of treatment effects or their transfer into spontaneous speech. There is insufficient knowledge about NPB's suitability for different types of VPI; for example, standardization concerning specific treatment methods is lacking (8).

Further, the connection of auditory and kinesthetic perception to visual biofeedback therapy is not clear. Brunner and colleagues (14) suggest that auditory self-perception can improve after visual feedback. Brunner et al. (8) observed: 'once the patients had managed to interrupt their dysfunctional motor pattern of tongue retraction under feedback control, they were immediately able to reproduce this result without visual control. Thus, visual cues exert a most powerful stimulus to change an automated articulatory pattern' (8, p. 655).

With NPB, patients gain important insights into the correlation of articulation and VPC. Afterwards, their extended knowledge and improved sensory control can enlarge the effectiveness of traditional speech therapy (8). Thus, NPB should be reserved for patients who are not responding to conventional speech therapy in correcting compensatory articulation with VPI or display very little progress (6). Brunner et al. (14) identified the following indications for NPB, which promise to get good results: 1) Compensatory articulation, but general adequate VPC; 2) Functional, limited tension of muscles after pharyngeal flap surgery; and 3) Partial VPI, VPI only in some phonemes.

Further, it appears possible to change negative movements of lateral pharyngeal walls (NMLPW) with NPB shortly before patients undergo pharyngeal flap surgery as a means of improving their prognosis (6). Following surgery, patients may be successfully treated with NPB, increasing LPW motion postoperatively (6).

Concerning the levels of nasopharyngoscopic view, the infravelar level and the supravelar level were both described as showing promise (14).

This unique viewing method has its pros and cons: The strongest advantage of NPB is that both the patient and the speech-language pathologist are able to monitor velopharyngeal activities during on-going articulation (21). It is easily tolerated and can be repeated on single sessions. There is no risk of exposure to radiation, and articulation is not disturbed (17).

But the treatment method is not free of disadvantages: Flexible fiberoptic nasopharyngoscopes are expensive and therefore are not available to therapists working in schools or home settings. It is an invasive method and not suitable with small children. It requires patients' motivation and compliance. In general, a speech pathologist and a physician must be present during its use (17).

Miyazaki's (21) summary from 1989 holds true today. It stated that future studies should document in detail the way in which the following factors impact treatment results: size, position, and shape of VPI (10,15,17); the type and extent of previous surgery; traditional speech therapy (and its definition) used in parallel or prior to treatment; and any associated difficulties, such as hearing loss, that participants may have. There is also a need for consistent descriptions of the perceptual speech characteristics using standardized and accepted terminology (22,23). Ysunza et al. (6) introduce the prospect that speech therapy combined with NPB as visual feedback may correct VPI without surgery, but more information is needed about the types of VPI, articulation characteristics, and their consistency. Brunner et al. (8) point out that more investigation is needed to evaluate the effectiveness of NPB for phoneme-specific VPI and generalized VPI not associated with compensatory articulation disorder.

In particular, future study designs must incorporate standardization for the reporting of nasopharyngoscopy and multiview videofluoroscopy (15) as well as the standards published by Royal College of Speech and Language Therapists' position paper entitled 'Speech and language therapy and nasopharyngoscopy for patients with velopharyngeal dysfunction' (10).

Conclusions

Preliminary results show effectiveness for visual feedback by flexible nasopharyngoscopy in helping older children and adults improve their VPC during articulation, but only in combination with conventional speech therapy. It seems to give the clients an idea or inner percept of the sphincter closure mechanism—one that cannot be obtained solely on the basis of auditory feedback. Actually there are no studies published measuring the effectiveness of NPB without additional treatments like secondary surgery or speech-language therapy. So no evidence of effectiveness of NPB as unique therapy method can be stated. In order to verify these preliminary results and establish a strong evidence base for high-quality interventions in cleft palate speech, more research is needed that features well designed single case (-control) studies and controlled case series of larger patient groups, taking into account that it might be difficult to get enough patients for a high-quality RCT.

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