Critical Review:
Is videonasopharyngoscopy biofeedback therapy effective in improving velopharyngeal closure in patients with cleft palate?

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This critical review examines whether videonasopharyngoscopy biofeedback is an effective therapy tool in improving velopharyngeal closure in patients with cleft palate. Study designs in this review include: randomized clinical trial, single subject 'n-of-1', single group or case series pre-posttest study, and systematic review. Results of these studies provide suggestive evidence for the clinical use of videonasopharyngoscopy biofeedback therapy. Clinical implications and recommendations for further research are discussed.

Introduction

Velopharyngeal dysfunction (VPD) is a condition in which the velopharyngeal (VP) port does not completely or consistently close during the production of oral phonemes (Kummer, 2008). This is often perceived as hypernasal speech because air escapes through the nasal cavity instead of only resonating through the oral cavity. VPD commonly occurs in patients with cleft palate for a variety of reasons. Abnormal palatal anatomy or impaired physiology of the VP port can impact complete closure. As well, some patients with cleft palate have the anatomical and physiological ability to close their VP port, but have not learned to do so adequately (Kummer, 2008). As a result, even after surgical repair of the cleft, VPD still remains in 10-20% of cases (Ysunza, Pamplona, Femat, Mayer & Garcia-Velasco, 1997).

Videonasopharyngoscopy is a tool regularly used as part of cleft palate assessments in order to evaluate VPD and the extent of VP closure (D'Antonio, Achauer, Vander Kam, 1993). In this approach, a small camera is inserted through the nares in order to obtain a video image of the VP port. This enables examiners to observe the function of the VP mechanism in real-time during speech and non-speech tasks (such as blowing and swallowing). Videonasopharyngoscopy can help assess the cause, size, and location of VPD (Kummer, 2008). Videonasopharyngoscopy also enables a clear image of VP function without inhibiting speech production (Witzel, Tobe & Sayler, 1989).

Traditional speech therapy is often administered to correct VPD in cleft palate patients with persistent impairments in VP closure after structural and functional deficits are surgically repaired (Brunner et al., 2005). However, it can be difficult to change this behavioural response if the patient does not have a clear understanding of the mechanism they are attempting to change (Brunner et al., 2005). Since videonasopharyngoscopy provides direct, real-time, functional information, it has been used as a therapy tool to provide visual biofeedback information to cleft palate patients with VPD.

Objectives

The primary objective of this review was to critically evaluate existing literature regarding the effects of videonasopharyngoscopy biofeedback therapy on VP closure in patients with cleft palate. The secondary objective of this paper was to propose clinical implications for professional practice and suggest areas for future research.

Methods

Search Strategy

Electronic databases CINAHL, EMBASE Proquest – Nursing and Allied Health, PubMed, and SCOPUS, were searched using the following terms: (cleft palate) AND (biofeedback OR visual feedback OR video feedback). The reference lists of relevant studies were also manually reviewed for additional articles meeting the selection criteria.

Selection Criteria

Studies selected for inclusion were required to examine videonasopharyngoscopy as a biofeedback therapy tool for patients with repaired cleft palate. All studies were required to assess VP movement or closure as an outcome measure. Only studies written in English were considered.

Data Collection

Search of electronic databases yielded five articles that met inclusion criteria. Manual search of reference lists generated one article. Results included the following study designs: randomized clinical trial (1), single

subject 'n-of-1' (2), single group or case series pre-posttest study (2), systematic review (1).

Results

Results are reported based on experimental design.

Randomized control trial (RCT)

RCTs are designated as level 1 experimental designs based on an evaluation system by Archibald (2009) (where level 1 represents the strongest level of evidence and level 4 represents the weakest level). RCTs consist of a treatment group and a control group, which enable the direct comparison of the intervention to no intervention. This increases the reliability of the study. RCTs also enable randomization of participants into control and treatment groups. This reduces the likelihood that participant characteristics would differ at baseline and, therefore, limits the risk of including confounding variables. These strengths were considered when critically reviewing the following study.

Ysunza et al. (1997) conducted an RCT in order to examine whether videonasopharyngoscopy could be used as a biofeedback tool to correct outward / negative pharyngeal wall movements in association with compensatory articulation. For the purposes of this review, only elements of the study involving VP closure and pharyngeal wall movements will be discussed further.

The study included 17 participants with VPD (mean age 11y;10m, age range not reported). Patients were randomly selected based on inclusion criteria explicitly outlined by the authors.

Participants in the treatment group (n=8) received traditional speech therapy three times per week and videonasopharyngoscopy biofeedback treatment twice per week. Control group participants (n=9) only received the traditional speech therapy. Authors reported that after twelve weeks, negative lateral pharyngeal wall movements had been modified in all participants in the treatment group and in 1/9 participants in the control group. When biofeedback therapy was later introduced in the control group, all remaining participants were able to modify lateral pharyngeal wall movement during speech.

Participant selection was a strength of this study. Inclusion criteria were stated clearly and participants were randomly selected. All participants were included throughout the duration of the study and were accounted for at its conclusion.

In terms of methodology, therapy techniques were not clearly described. This decreases the possibility of replication of this study. Additionally, due to the nature of intervention, neither participants nor examiners were blinded to the treatment. As a result, biases may have influenced observed results. As well, although similar in other respects, treatment and control groups differed in the number of hours of therapy they received. As a result, the *frequency* of treatment may reduce the likelihood that improvements can be attributed to the *type* of treatment alone.

Additionally, the results in this study were not clearly stated. The authors described outcomes in terms of "modifications" of lateral pharyngeal wall movements. However, no objective data or statistical results were presented. Therefore, the significance of the results is unclear.

Despite the methodological limitations of this study, given the strengths of RCTs, the results are evaluated to have suggestive clinical implications.

Single subject 'n-of-1'

Single subject 'n-of-1' type studies are evaluated as level 1 experimental design (Archibald, 2009). They enable greater sensitivity to individual improvements. A small number of participants were included in the following studies, so results should be cautiously applied to other populations.

Witzel, Trobe and Salyer (1988) evaluated whether using videonasopharyngoscopy as a feedback tool could help correct inadequate VP closure.

The one participant in this study was a ten-year-old girl with repaired bilateral cleft lip and palate. At baseline, the subject's speech was characterized by distorted sibilant fricatives and nasal air emissions during the production of /s/. She had previously received four years of speech therapy at school and continued to maintain phoneme-specific VPD.

The subject received one biofeedback treatment session using videonasopharyngoscopy. She was instructed to observe her VP movement during the productions of /s/ when shaped from /t/. Afterward, traditional speech therapy was recommended and the subject also received maxillary advancement surgery. VP function was reassessed six months post-surgery. Authors reported that after the biofeedback training, the subject was able to close her VP port during connected speech. VP closure was also maintained during the follow-up assessment.

The methodology and treatment design were clearly reported in this study. However, there are a number of limitations which may have affected the results. The introduction of traditional speech therapy and surgery on the maxilla after biofeedback training may have had a significant impact on the VP closure observed at follow-up. Though the patient maintained consistent VP closure immediately post-biofeedback therapy, it cannot be concluded these that improvements were maintained long term due to the presence of these confounding variables.

Since no statistical analyses were conducted, the significance of observed improvements cannot be assessed in this study.

Given the methodological limitations, the findings in this study are equivocal.

Witzel et al. (1989) assessed the consistent closure of VP ports using videonasopharyngoscopy as a visual feedback therapy tool.

Three participants were included in this study (age range 35-50, mean age 40). All patients had persistent hypernasiliaty, nasal air emission, and VPD.

Subjects participated in thirty minute therapy sessions using videonasopharyngoscopy for biofeedback. Phonemes produced with the best VP closure were targeted first in therapy. Other treatment protocol was also described. Two subjects received traditional speech therapy between biofeedback sessions. Authors reported that one subject was able to produce complete and consistent VP closure after four biofeedback therapy sessions. Improvements were maintained at fourteen month follow-up. Another participant achieved complete VP closure and was discharged for therapy after two sessions. No follow-up data was reported. The final participant dropped out of treatment after five therapy sessions. The authors noted that this patient was making improvements in VP closure but dropped out due to "frustrations" with her progress.

This study provided sufficient detail in terms of the methodology and treatment procedures to allow for replication. One limitation is that traditional speech therapy was also provided to some participants in the study. Therefore, outcomes many not result from the effects of the biofeedback therapy alone. As well, follow-up reassessment was not conducted for all subjects. Therefore, conclusions about long term treatment effects are limited.

Authors did not report any statistical analyses in their results. Therefore, the level of significance of the outcomes cannot be assessed.

Despite some methodological concerns, the results of this study are suggestive of the effectiveness of videonasopharyngoscopy biofeedback on VP closure.

Single group or case series pre-posttest

By their nature, single group or case series designs do not contain control groups for comparison. As a result, these studies are limited in their ability to control for the potential effects of participant characteristics. Therefore, statements of causality need to be carefully considered. Accordingly, single group or case series pre-posttest designs provide level 3 experimental evidence (Archibald, 2009). These limitations were taken into account when critically evaluating the following studies.

Yamaoka, Matsuya, Miyazaki, Nishio and Ibuki (1983) investigated the longitudinal effects of self-training using videonasopharyngoscopy on VP closure.

Participants included 59 individuals with cleft palate and persistent VPD (age range 8-45, mean not reported). All participants were included based on criteria stated by the authors. In the present study, VP closure was examined during: blowing, vowel production, consonant production and swallowing. Subjects were divided into five groups based on tasks in which complete closure was observed. All participants displayed complete VP closure during swallowing.

Subjects participated in biweekly, one hour experimental sessions in which they completed the above tasks with the videonasopharyngoscopy in situ. Participants were instructed to try to close their VP port during tasks in which they were unable to achieve complete closure previously. Traditional speech therapy was also given to some patients in order to correct misarticulations. Two examiners evaluated VP closure as either complete or incomplete. Results indicate that 59.3% of subjects achieved improvements in VP closure (had complete closure on more tasks than at baseline). Authors also reported the percentage of participants in each of the five groups who showed improvements.

A limitation of this study relates to the methods of subject selection. Though the sample size is relatively high compared to other studies on this topic, it is unclear how subjects were selected for the study. Randomized selection was not reported, which limits the generalizability of the results.

The treatment protocol was clearly outlined in this study, which increases the ability to replicate its design. However, the duration of treatment was not reported. As well, some patients received additional speech therapy, which may have had an uncontrolled and important effect on the results. Another methodological weakness is the evaluation of VP closure. Outcomes were measured with a binary system of either complete or incomplete closure, so improvements in *degree* of closure were not noted. As a result, partial improvements in VP closure were not considered. As well, two raters evaluated VP closure; however interrater and intrarater reliability were not discussed.

Only descriptive statistics were reported in this study. The lack of statistical analyses limits the ability to infer significant effects of this treatment and is an important weakness in this study.

Due to the study design and methodological limitations, the findings in this study are evaluated to be suggestive.

Brunner et al. (2005) aimed to evaluate immediate, long-term and carry-over effects of videonasopharyngoscopy biofeedback therapy in cleft palate patients with VPD. Mean VP closure during speech sounds and patient self-perception were assessed. For the purposes of this review, only the outcomes on VP closure will be discussed further.

Eleven subjects with VPD (age range 7-30; mean age 14y;2m) participated in this study. Inclusion criteria were clearly outlined.

All phonemes in which VP closure was not complete were treated using biofeedback therapy. Authors implemented a multiple baseline methodological design in which VP closure was assessed pre-treatment, post-treatment, and at six month follow-up. The authors described the methodology for the treatment intervention. Subjects previously receiving traditional speech therapy were encouraged to continue with therapy during the course of the study. Other participants were instructed to practice learned therapy techniques between biofeedback sessions. Three independent raters evaluated VP closure as either complete or incomplete. Interrater reliability was evaluated to be high (91%) among the raters. Descriptive and statistical analyses were conducted. The data was evaluated to be non-normally distributed and parameter-free tests were performed. It was determined that a significant increase in mean VP closure was observed post-treatment ($Z=-5.64$, $p=0.00$) and at six month follow up ($Z=-5.433$, $p=0.0$). One subject dropped out of the study after the training of one speech sound. As a result, there was one set of missing data.

Authors did not report how participants were selected for this study. As a result, randomized selection likely did not occur. Therefore, participant characteristics at baseline were not controlled and may have influenced the results.

The rational and objectives of this study were explicitly reported. Similarly, the authors clearly reported the methodology of the intervention, such that it could be easily replicated. Confounding variables were well controlled in this study. Participants receiving traditional speech therapy continued to be given therapy during baseline, for the duration of the study, and at reassessment. Throughout the study, traditional speech therapy was never introduced to subjects not receiving traditional therapy at baseline. As a result, improvements in VP closure can likely be attributed to the biofeedback intervention. One methodological flaw is the assessment of outcomes. Like some of the other studies discussed, results relate to a binary score that did not account for partial increases in VP closure.

Brunner et al. (2005) used appropriate nonparametric statistical analyses. This strengthens the interpretation of their significant treatment outcome. However, one limitation with the stated results is that one subject dropped out of the study early and reasons for drop out were not reported.

This study is judged to provide suggestive evidence of treatment efficacy.

Systematic Review

Neumann and Romonath (2012) conducted a systematic review of the literature analyzing the "effectiveness of nasopharyngoscopic biofeedback in clients with cleft lip and palate and velopharyngeal dysfunction". Multiple outcome measures were considered in this review, but for the purpose of this report, only information on VP closure will be discussed further.

Authors conducted a rigorous search method of electronic databases, reference lists and publications in the Cleft Palate-Craniofacial Journal. Articles written in English and German were considered. Six studies were evaluated to meet criteria for inclusion and each was summarized according to their participants, intervention, comparators, outcomes, study design and level of evidence (as assessed by consensus agreement between two reviewers). The risk of bias in the individual studies was also evaluated.

Authors concluded that there were no strong results in terms of a high level of evidence available in the current literature. They also noted that there were a number of limitations in terms of methodological design, participants, and outcome measures in the analyzed studies. However, the authors conclude that based on existing data, nasopharyngoscopy biofeedback therapy with the support of traditional speech therapy may be effective for optimizing VP closure in individuals with cleft palate speech.

This is a strong systematic review paper that explicitly stated its purpose, objectives, and methods. Many sources were consulted during the location and selection of studies for inclusion. As well, numerous relevant search terms were used. The authors also included all articles in English and German which expanded the number of applicable studies that were analyzed. Both published and unpublished literature was considered as well. As a result, all studies that met inclusion criteria were likely located for this study. However, current searches of electronic databases reveal that some articles written in languages other than German and English may have also been relevant. Since they did not meet the language requirements for inclusion, these studies were not assessed.

The critical appraisal of the articles was completed by two reviewers. Although reviewers were not blinded by the ratings of one another, the inclusion of multiple assessors increases the validity of the appraisal. Additionally, authors provided sound rational for their critiques and clearly outlined the implications of their analysis.

As a result, opinions from this article have valid clinical implications.

Discussion

A number of concerns should be considered when summarizing the overall results of these studies. Firstly, most studies reviewed included designs that limited their levels of evidence and restricted their sample sizes. For example, articles by Witzel, Trobe and Salyer (1989), Witzel et al. (1989) and Brunner et al. (2005) all included less than 15 participants. As a result, the effects observed in these studies are not necessarily representative of the general population, so overall conclusions should be evaluated cautiously.

Another notable limitation of the reviewed studies is an overall weakness in the reported results. In many articles, it was unclear how the evaluation of outcomes was conducted and vague descriptions of results were provided. As well, most studies (i.e. Ysunza et al., 1997; Witzel, Trobe and Salyer, 1988; Witzel et al., 1989, Yamaoka et al., 1983) did not conduct statistical analyses of their results. Therefore, it is unclear whether observed effects were significant.

As well, this review is limited because treatment outcomes were only assessed with regards to VP closure. Other potential outcome measures, such as nasalance score, perceptions of voice quality, and intelligibility, were not considered. These types of outcomes were rarely evaluated in the literature, and as a result, were not included in the present review. Nonetheless, it may be reasonable to infer that with greater VP closure, improvements in nasalance score, voice quality and intelligibility would also be expected.

These considerations should be noted when judging the clinical implications based on the conclusions of this review.

Conclusions, Clinical Implications and Recommendations

This critical review suggests that biofeedback therapy using videonasopharyngoscopy may be effective in improving VP closure in patients with cleft palate. To date, there is only a limited amount of available literature on the topic with few strong experimental designs. Taken together, these studies provide suggestive evidence for the clinical use of this treatment approach.

Before videonasopharyngoscopy biofeedback therapy is regularly used clinically, a number of factors need to be considered. Videonasopharyngoscopy treatment is financially expensive to administer and may be uncomfortable for some patients (Peterson-Falzone, 2006). As a result, not all centres would have the capacity to administer this treatment method. Therefore, it is important to consider whether the benefits of this therapy approach outweigh these reported disadvantages. In order to better address these concerns, further research is required. In particular, suggestions for future research include:

- Study designs that lend to stronger levels of evidence and greater sample sizes in order to improve the confidence of results.
- Exploration of the optimal frequency, duration and method of intervention in order to enhance clinical implications.
- Additional outcome measures (e.g. naslance scores, intelligibility, perceptions of voice quality) in order to provide functional effects of treatment.
- Statistical analyses of results in order to assess significance of outcomes.

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