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Critical Review: Continuous Positive Airway Pressure (CPAP) as a Treatment for Hypernasality

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This critical review examines the effects of using Continuous Positive Airway Pressure (CPAP) to reduce hypernasality for individuals with velopharyngeal inadequacy. Study designs include: single subject ABA treatment design, within groups repeated measures design, a non-randomized clinical trial and a series of non experimental case studies. Overall, research suggests that CPAP therapy can be useful for some individuals with hypernasality; however, the optimal treatment program as well as the differential effects that the treatment may have on the severity of hypernasality is yet to be determined. The findings of this review have implications for further research and clinical practice in the field of Speech-Language Pathology.

Introduction

Treatment for individuals with hypernasality has traditionally utilized non-speech oral motor exercises (NSOME). Research shows that although these exercises may be helpful for some individuals with hypernasality, there is insufficient evidence to support the use of NSOME as an effective means for reducing hypernasality (Ruscello, 2008). It could be that although NSOME work to strengthen the velum, the increase in velar movement may not transfer to speech tasks. Therefore hypernasality in speech is not reduced although the muscles of the velum may be stronger. Kuehn (1991) proposed a new treatment for hypernasality. He used the same strengthening principals as for NSOME except that he applied these strengthening exercises to speech tasks. This would theoretically increase the transfer of velar strength to an individual's conversational speech. He proposed that by providing a continuous resistance to the velar muscles during speech, the muscles could be strengthened therefore increasing the ability of the individual to maintain velopharyngeal closure when needed for speech. Continuous Positive Airway Pressure (CPAP) machines have been used traditionally to treat individuals with sleep apnea by putting resistance on the velum, making a larger velopharyngeal opening for air to pass through (Schmidt-Nowara, 1984). By using this machine during speech exercises, the pressure creates a resistance that the velum must contract more forcefully to overcome, thus strengthening its muscles. By using the CPAP machine during speech exercises, it eliminates the need to transfer this increase in velar strength and movement to conversational speech as was needed for the NSOME (Kuehn 1991).

Objectives

The primary objective of this paper is to critically evaluate the existing literature regarding the use of CPAP to treat individuals with hypernasality. The secondary objective is to propose evidence-based practice recommendations about using CPAP treatment in clinical practice to help reduce hypernasality for individuals with velopharyngeal inadequacy

Methods

Search Strategy

The computerized databases, PubMed and Medline, were searched using the following search strategy:

> ((hypernasality) OR (resonance disorders)) AND ((continuous positive airway pressure) OR (CPAP))

The search was limited to articles written in English between 1991 and 2004.

Selection Criteria

Studies selected for inclusion in this critical review were required to investigate the effects of treating individuals with hypernasality, with a treatment regime involving CPAP. No limits were set on the demographics of research participants or outcome measures.

Data Collection

Results of the literature search yielded the following types of articles congruent with the aforementioned selection criteria: single subject ABA design (1), mixed non-randomized clinical trial (1), within groups repeated measures pre and post test design (1), and non experimental case studies (1).

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Results

Study #1: Using a mixed non-randomized clinical trial, Kuehn, Moon and Folkins (1993) examined how increased positive airway pressure affected EMG activity of the levator palatini (LP) muscle in individuals with and without cleft palate. It was assumed that since the LP muscle primarily moves the velum, an increase in EMG activity in response to increased pressure would create resistance on the muscle. With that being true, this resistance could be used to theoretically strengthen the LP in individuals with hypernasality due to velopharyngeal inadequacy. Five normally speaking adults and four adults with cleft palate and a history of mild to moderate hypernasality were fit with CPAP masks. Electrodes were placed into the LP muscle of each subject and the subjects completed three tasks. In the first task, EMG activity was recorded while the subjects repeated the non-word 'ansi' (VNCV format) ten times with the CPAP machine set at four different pressure levels (0, 7.5, 10 and 12.5 cmH20). The order of the pressure levels was randomly selected and inter and intra word EMG levels were recorded. In the second task, EMG activity levels were recorded while the subjects repeated the syllable 'sa' continuously during which the pressure from the CPAP machine increased gradually from 5 to 12.5 cmH20 and then back down again (ascending-descending condition). In the third task, EMG activity was recorded while each person swallowed. EMG levels were averaged for the 'ansi' condition as well as between the 'ansi' repetitions to see if the muscle would adjust its tonic level of activity in response to changing pressures. Peak levels of activity during 'ansi' productions were compared with peak levels from the swallowing task to determine the relative level of muscle activity during speech compared to non speech tasks.

An ANCOVA revealed that as positive airway pressure was introduced there was a significant increase in LP muscle EMG activity compared to the zero pressure condition. This was true for inter as well as intra word measurements and was consistent across cleft palate and non cleft palate groups. The data also suggested that as the pressure was increased past 10 cmH20, EMG activity decreased for 9 of the 12 subjects. No clear difference emerged in the swallowing and speech data for the two groups. The researchers concluded that the LP muscle contracted more forcefully when positive airway pressure acted as a resistance. Because of this, they proposed that the muscles could be strengthened in an exercise situation and this increased strength could act to more effectively close the velum in individuals with velopharyngeal inadequacy due to cleft palate. They also suggested that during therapy, pressure levels past 10 cmH20 should not be used as these may decrease the force that the LP muscle contracts. Finally, they suggested that the LP muscle could be postured in a 'ready' position to overcome increased intranasal air pressure that would be introduced. This could be an added benefit to CPAP therapy as the motor units would be constantly active while the CPAP machine is in use.

Caution should be taken when interpreting the results of this study since the sample size was small. The study design utilized is one level below the 'gold standard' of research (randomized clinical trial/single subject ABA designs). This means that although we can have fairly good confidence in the result of this study, it is still not considered to be within the highest level of evidence that research studies can achieve. The major limitation of this study is that there is no evidence to suggest that strengthening the LP muscle during resistance training as is done in CPAP therapy, will transfer to conversational speech. It may be that individuals with velopharyngeal inadequacy are able to forcefully contract the LP muscle when resistance is added. However, behavioural training would be needed to apply this forceful contraction automatically during conversational speech when an increase in resistance is not being applied.

The results of this study suggest that that the velum can be strengthened by applying resistance through air pressure. Therefore treatments for hypernasality that apply resistance, such as CPAP therapy, could be used to reduce hypernasality by increasing the ability of the velum to contract more forcefully to close the velopharyngeal opening during non nasal sounds.

Study #2: Kuehn (1991) introduced CPAP as a new therapy for treating hypernasality. For therapy, each subject was given a CPAP machine to borrow for an 8 week period with which they conducted sessions at home 6 days per week. They were each given a schedule to follow telling them how long and at what pressure the CPAP machine was to be set to. Pressure levels ranged from 3 to 7 cmH20 and time per session ranged from 10 to 24 minutes. The pressure setting and time per session gradually increased over the course of the 8 weeks of therapy. During each session, the subject was given sets of 50 VNCV (vowel, noun, pressure consonant, vowel) words and non words, and sets of 10 short sentences containing nasal and non-nasal sounds to read aloud. The subject read the lists of words and sentences until the session was over. The VNCV pattern was used for the words because the lowered velar position on the nasal sound and then the quick elevation for the pressure consonant created a need for the muscles of the velum to contract forcefully to elevate.

Kuehn (199) presented 6 cases studies in this article. Two of the case studies (subjects 1 and 2) were individuals who received CPAP therapy only once per week. No improvements were seen thus, treatment was abandoned in these two cases. Four case studies involved individuals who had received the full regimen of CPAP therapy as described previously. For each of the case studies, subjective ratings by the researcher were the bases of comparison for pre and post treatment hypernasality. Subject 3, an 8 year old girl with moderate hypernasality due to Klippel-Feil syndrome, showed perceptual improvements in hypernasality as noted by the researcher. Five expert judges rated this subject's speech samples at pre-treatment, one month into treatment, after 8 weeks of treatment and 2 months following the completion of treatment. The judges rated sentences and an overall level of significance across the five judges was determined using a chi-squared goodness of fit test. Results revealed that the judges were able to detect a decrease in hypernasality ($p<0.01$). Subject 4 was an 8 year old girl with cleft lip and palate causing moderate hypernasality. The researcher determined that her hypernasality improved from a rating of 4/7 to 2/7 on an equal interval scale (1= normal speech, 7= severely hypernasal speech). Subject 5 was a 14 year old girl with moderate/severe hypernasality due to a deep pharynx. No improvements in hypernasality were noted after one month of therapy and thus treatment was terminated. Subject 6 was a 20 year old man with a sustained closed head injury resulting in speech dysarthria with moderate hypernasality. Kuehn (1991) determined that his hypernasality rating improved from 4/7 to 2/7 pre to post treatment. Another month of treatment was conducted after completion of the 8 weeks, but no more improvements were made and thus treatment was terminated.

Because of the low level of evidence provided in case studies, as well as the small sample size used for this study, the results should be treated with caution. The results are subjective to experimenter bias because perceptual measurements were made only by the researcher on all but one of the cases (subject 3). Also, within the small sample size, each of the subjects was different in how they presented with hypernasality. This limits the ecological validity of the findings since no single population is represented sufficiently to generalize the findings to a specific population. Despite the limitations of this study, the information gained through the case studies can help to fuel further research looking at the usefulness of CPAP therapy for treating individuals with hypernasality. The researcher also suggested that patients with severe hypernasality may not benefit from this therapy as these patients will likely require physical management. Finally, Kuehn (1991) suggested that intensive therapy should be done over a two month period since it is likely that gains in treatment would happen in this time frame and that no further gains would be made after this.

Study #3: Kuehn, Imrey, Tomes, Jones, O'Gara, Seaver et al (2002) followed up on the case studies previously mentioned (i.e. Kuehn (1991)) with a within-groups repeated measures, pre and post treatment design to determine if using CPAP therapy would reduce hypernasality for individuals with cleft palate. Twenty-four male and 19 female subjects born with cleft palate and ranging in age from 3 to 25 years, were recruited through eight treatment sites in the United States. The 43 subjects were identified as hypernasal by perceptual evaluation from local speech pathologists. The CPAP training and treatment took place at each of the subjects respective treatment sites as well as at the home of the participating subjects. The CPAP treatment followed the same protocol as described by Kuehn (1991). For children who were too young to read, parents read the words and sentences to the child and the child repeated them back during treatment. Each subject received a schedule showing the time and pressure level for each session (time ranged from10-24 min and pressure levels ranged from 4 – 8cmH20). Hypernasality measurements were made before treatment, midway through, immediately following treatment, and between 8 and 21 months after treatment was concluded. Measurements included blinded judges' and un-blinded clinician-investigator ratings of speech samples obtained at each measurement point on an equal interval scale of 1-8 (1= normal speech, 8= extremely hypernasal speech). For the blinded judges ratings, standard reference scales and replicate evaluations were used to reduce inter-judge variability. As well, average nasalance scores were obtained using a nasometer during three readings of 'the zoo passage". The subjects were given a video camera to record each session which helped to ensure that the subjects were following the CPAP therapy protocol properly.

A Wald-type F statistic was used to determine if a change in blinded hypernasality scores occurred from pre to post treatment. Changes in nasalance scores were analyzed by a randomized block analysis of variance. Un-blinded nasality ratings were analyzed using Wald statistics for comparison of marginal mean scores in a categorical data mixed model. On average, mean nasality scores as determined by the blinded judges' ratings, declined 0.2 rating points from pre to post treatment ($p=0.016$). The researchers noted that most of the change in nasality occurred in the last month of treatment when the therapy was more intense. Nasalance scores were consistent with the observed improvements in perceptual nasality scores but did not reach statistical significance. Follow up ratings for 25 of the subjects showed that the mean change in nasalance scores were unchanged at follow-up for subjects whose hypernasality had improved post treatment indicating possible stability of improvements made through CPAP therapy. Un-blinded ratings of hypernasality correlated modestly with blinded ratings although un-blinded ratings tended to exaggerate the benefit of the CPAP therapy.

Although the sample size in this study was sufficient, the addition of a control group and randomization into groups would have increased the level of evidence to the 'gold standard'. This increased level evidence would have provided the reader with more confidence in the interpretations that could be made from the results. Since the treatment was conducted at eight different treatment sites, there is the possibility that variability in clinician-investigator training and follow-through therapy could have affected the results. Also, non-compliance by the subjects for the intense therapy regime could have affected the results. One of the major limitations of this study was that the inter-judge reliability for the blinded judges was only adequate (0.50) despite the precautions taken to limit this variability. Since this was the primary measurement taken, caution must be used when interpreting the results of this study given that the measurements may have been invalid. The researchers do not address this limitation and the effect it might have on the interpretation of their results in this article. Despite these limitations, the results suggest that CPAP therapy can be useful for treating hypernasality resulting from cleft palate.

Study #4: Cahill, Turner Stabler, Addis Theodoros and Murdoch (2004) studied three individuals with moderate to severe hypernasality following a Traumatic Brain Injury (TBI). They used a single subject ABA treatment design to determine the effects of CPAP therapy on hypernasality caused by TBI. Each subject underwent CPAP therapy, 4 days per week for 4 consecutive weeks. The subjects were taught how to use the CPAP machine and given lists of words and sentences to say during each session. They were given a schedule to follow regarding the length and pressure intensity of the sessions (time of sessions ranged from 10-24 minute and pressures ranged from 4-8 cmH2O). Subjects read lists of VNCV words as well as short sentences containing nasal and non-nasal sounds until the allotted time for the session was over. Perceptual and instrumental measurements were made before treatment, mid-way through, immediately after treatment and one month post-treatment. Perceptual measurements included ratings on the Frenchay Dysarthria Assessment (FDA) (Enderby, 1983), the Assessment of Intelligibility of Dysarthric Speech (AIDS) (Yorkston & Beukelman, 1981) sentence level, and analysis of a speech sample rated on a seven point equal interval scale for 5 dimensions of speech including; hypernasality, precision of consonants, length of phonemes, precision of vowels and overall intelligibility. Instrumental assessments included nasalance measurements made using a nasometer while the subjects read "the zoo passage". Analysis included qualitative comparison of FDA and speech sample analysis scores. A reliable change index was calculated based on nasalance scores from 20 non-neurologically impaired individuals to determine if statistically significant changes in the subject's nasalance scores occurred between test periods. Finally, a percentage change calculation was used to compare each subject's sentence intelligibility scores on the AIDS between test periods.

All three of the subjects in this study showed significant decreases in nasalance scores one month following the conclusion of treatment. For two of the three subjects, significant improvements in sentence intelligibility as measured by the AIDS were observed. One subject did not show any improvement throughout and after treatment on the perceptual measures even though the instrumental measures did show some improvement in nasalance scores one month post treatment. It was predicted that the severity of this subject's hypernasality may have been a factor. The improvement in the nasalance scores may not have been picked up by the perceptual measurements because the improvements made were not significant enough to be heard.

Limitations of this study affect how it can be interpreted. Results of this study must be interpreted with caution due to the small sample size. Despite this, single subject ABA treatment designs are considered to have a high level of evidence since the subjects act as their own controls, and thus some confidence can be taken when interpreting the result. Information about how the participants were recruited is not given by the researchers and thus a bias in the selection process could have been present. Also, no information was given on whether or not the subjects continued with speech treatment of any kind during the one month after treatment. If they were involved in any treatment during this month, this could have affected the measurements made post-treatment thus exaggerating the overall results of the study. Researcher bias was limited by recruiting blind judges whose inter-judge reliability was over 0.8 but it was not stated who made the FDA measurements. If the researchers of the study performed these measurements, then this could have been subjected to researcher bias. Perceptual measurements, although useful and important to provide information on how speech might be heard in real life situations, may not provide a sensitive enough measurement to be used alone when measuring improvements in hypernasality. Instrumental assessment of nasalance could show statistically significant improvements for very severely hypernasal subjects, where perceptual measurements fail to show these same significant improvements. This may give

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some insight into how using only perceptual measurements for assessment improvements in hypernasality may be misleading for clients with very severe hypernasality. Perceptual measurements may miss smaller improvements that are being made but may not yet be perceived.

The results of this study suggest that CPAP treatment may be useful for treating individuals with hypernasality following a TBI.

Discussion

All of the studies examined in this systematic review looked at the reasons for and the implications of using CPAP therapy to treat hypernasality in individuals with VP inadequacy. Overall, the evidence that this treatment improves hypernasality is suggestive. Small sample sizes as well as study designs that fall short of the 'gold standard' of research cause the findings and implications of the studies to be interpreted with caution.

There were some common limitations that were present throughout the research. First, all of the studies utilized perceptual judgment measures based on equal interval scales. Research shows that using direct magnitude estimation with modulus may be a more valid and reliable method for perceptually measuring hypernasality (Whitehill, Lee & Chun, 2002). In this method, investigators assign a number to a standard speech sample. The judges are then required to rate all test samples compared to this standard sample. By using this method of measurement, systematic bias associated with interval scales is eliminated and statistical analysis is still applicable (Stevens, 1975 as cited in Whitehill et al, 2002). Another common limitation is that none of the studies investigated the source of hypernasality. The possible sources of hypernasality are VP insufficiency (characterized by structural differences that causing an increased VP port) or VP incompetence (characterized by intact and normal structure but abnormal use that increases the size of the VP port) (Kummer, 2008). It would be important for researchers to distinguish between these causes of hypernasality before using CPAP treatment that is designed to strengthen the muscles of the velum. If the subject has a velum that is too short or a deep pharynx, it may be that VP closure is not possible no matter how much the VP muscles are strengthened. It is more likely that if this treatment is going to help anyone it would be the subject with VP incompetence. Examining what type of VP closure an individual exhibits would also be beneficial. For some individuals, VP closure is achieved by moving the lateral pharyngeal walls medially to meet the midline of the velum, and little posterior velar movement is used (Kummer, 2008). For these individuals, CPAP therapy may not be effective as there would be little need to strengthen the velar muscles. Finally, each of these studies assumes that the source of hypernasality for all of the individuals is decreased strength of the muscles of the velum. It is also plausible that instead of a problem with strength, hypernasality is caused by a decrease in coordination of the velum during connected speech. It would be important for researchers to consider this possibility when determining possible treatments for hypernasality

Future Research Implications

It is clear from the previous literature on the effectiveness of CPAP therapy, that more research is needed before common use of the therapy can be applied to individuals with hypernasality. The extensive time commitment in addition to the expense of buying and maintaining a CPAP machine warrants more compelling evidence of positive treatment effects before the therapy can be used regularly in clinical practice. It is recommended that future research focus on larger sample sizes as well as study designs that utilize randomized control and treatment groups. Consideration for the limitations of the studies, as previously mentioned, should be given. This would ensure that treatment is only being tested on groups of individuals that could potentially benefit from its effects. It is important that future research follow to determine possible effective treatments for hypernasality as it is a disorder that can adversely affect overall intelligibility of speech and therefore can have a significant impact on an individual's life.

Clinical Implications

Although the research reviewed here is not without limitations, there is suggestive evidence that CPAP therapy is an effective means of reducing hypernasality. If clinicians were to adopt this treatment as part of their practice, the following should be considered:

a) The cause of hypernasality should be considered to ensure that the client's hypernasality is due to VP incompetence rather than VP insufficiency.

b) Treatment should not exceed 8 weeks as effects appear to occur within this time frame and not after.

c) Treatment should be intense and occur no less than six days per week.

d) Pressure levels should not exceed 10cmH2O as no further benefit was seen past this pressure.

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