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Relationship between velopharyngeal closure, hypernasality, nasal air emission and nasal rustle in subjects with repaired cleft palate

Correlação entre o fechamento velofaríngeo, hipernasalidade, emissão de ar nasal e ronco nasal em indivíduos com fissura de palato reparada

ABSTRACT

Purpose: To investigate the correlation among velopharyngeal closure, hypernasality, audible nasal air emission (NAE) and nasal rustle (NR), in individuals with repaired cleft palate. **Methods:** One hundred patients with repaired cleft palate and lip, submitted to pressure-flow study for measurement of velopharyngeal orifice area (velopharyngeal area) and speech sample recordings. Velopharyngeal area was estimated during the production of the sound /p/ inserted in a sentence, and the velopharyngeal closure was classified as adequate, borderline or inadequate. Hypernasality was rated using a 4-point scale, NAE and NR were rated as absent or present, by three speech language pathologists, using recorded speech samples. Inter and intra-judge agreements were established. Statistical analysis was performed using the Spearman correlation coefficient considering p<0.05. An ordinal logistic regression model was developed to investigate whether the characteristics of speech can predict velopharyngeal closure. For this, the speech samples included in this analysis were those that obtained 100% agreement among raters as to the degree of hypernasality (43 out of 100). **Results:** Significant correlation was found between hypernasality and velopharyngeal area; audible NAE and velopharyngeal area. A negative correlation was observed between the NR and velopharyngeal area. The regression analysis showed that the perceptual speech characteristics contributed significantly to predict the velopharyngeal closure. **Conclusion:** There is significant correlation between velopharyngeal closure and hypernasality, NAE and NR. It suggests that the perceptual speech characteristics can predict velopharyngeal closure, favoring the diagnosis and the definition of treatment conduct of velopharyngeal dysfunction.

RESUMO

Objetivo: Investigar a correlação entre as dimensões do orifício velofaríngeo, hipernasalidade, emissão de ar nasal (EAN) audível e ronco nasal (RN), em indivíduos com fissura palatina reparada. **Métodos:** Foram avaliados cem pacientes com fissura labiopalatina reparada, submetidos à medida da área do orifício velofaríngeo (área velofaríngea) por meio da técnica fluxo-pressão e à gravação de fala. A partir da área velofaríngea, determinada durante a produção de /p/ inserido numa frase, o fechamento velofaríngeo foi classificado em adequado, marginal e inadequado. A hipernasalidade foi classificada em escala de quatro pontos, EAN e RN em presente-ausente, por três fonoaudiólogas utilizando amostra de fala gravada. A concordância inter e intra-avaliadores foi estabelecida e a correlação entre as variáveis foi analisada por meio do coeficiente de correlação de Spearman, considerando p<0,05. Um modelo de regressão logística ordinal foi elaborado para investigar se as características da fala podem predizer o

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fechamento velofaríngeo. Para tanto, foram incluídas somente 43 amostras de fala que obtiveram 100% de concordância quanto ao grau de hipernasalidade entre as avaliadoras. **Resultados:** Correlação significativa entre hipernasalidade e área velofaríngea; EAN audível e área velofaríngea. Correlação negativa foi verificada entre RN e área velofaríngea. O modelo logístico mostrou que as características da fala contribuíram significativamente para a previsão do fechamento velofaríngeo. **Conclusão:** Existe correlação entre dimensões do orifício velofaríngeo e hipernasalidade, EAN e RN, sugerindo que as características perceptivas da fala podem predizer o fechamento velofaríngeo, favorecendo o diagnóstico e a definição de conduta de tratamento da disfunção velofaríngea.

INTRODUCTION

Most speech disorders observed in individuals with repaired cleft palate are directly or indirectly related to velopharyngeal dysfunction (VPD). Symptoms such as hypernasality, nasal air emission (NAE) (audible or not), and weak intraoral pressure are direct consequences of the failure of velopharyngeal closure. VPD is diagnosed by the auditory-perceptual assessment of speech[1-5] and by instrumental evaluations[5-7].

Generally, the velopharyngeal function is classified based on the quality of resonance, assessed by the auditory-perceptual assessment and by the direct observation of velopharyngeal structures using methods such as nasendoscopy and videofluoroscopy[8]. Besides these, indirect methods, such as aerodynamic evaluations, provide quantitative data and, therefore, objective information about the velopharyngeal function. The pressure-flow technique in particular allows to determine the velopharyngeal closure by measuring the area of the velopharyngeal orifice.

The perceptual characteristics of the velopharyngeal function are used to make inferences on the adequacy of the velopharyngeal mechanism. Studies suggest, for example, that there is a correlation between the degree of hypernasality, the presence of audible NAE and nasal rustle (NR), and velopharyngeal gap size[3-10].

Once more and more the subjectivity and the reliability of the auditory-perceptual assessment of speech have been investigated in literature, this study analyzed the following question: can the perceptual judgment of speech parameters predict the velopharyngeal gap size? Therefore, this study was carried out with the objective of investigating the correlation between the velopharyngeal gap size, the degree of hypernasality, and the presence of NAE and NR.

METHODS

Subjects

This study was approved by the Research Ethics Committee of the Hospital for the Rehabilitation of Craniofacial Anomalies, Universidade de São Paulo, and all of the participants signed the informed consent. One hundred patients with repaired cleft palate were assessed, being 37 with isolated cleft palate and 67 with lip and cleft palate; 36 male and 68 female participants, aged between 6 and 47 years

old. Participants were selected consecutively, for 1 year and 3 months. Have not been included in the study individuals who presented syndromes; neurological problems; physical and/or mental incapacity to undergo the tests; acute or chronic allergic respiratory symptoms that could lead to nasal congestion during the examination; subnormal values of nasal area, according to the result of the rhinomanometry performed on the same day; residual palatal fistulae difficult to sealing during the evaluation; pharyngeal flap; compensatory articulation in the production of the consonant "p", since this sound is used to measure the velopharyngeal orifice area.

Procedures

Aerodynamic Assessment of Speech (pressure-flow technique)

The area of the velopharyngeal orifice was assessed by the pressure-flow technique, using the PERCI-SARS system (version 3.50, Microtronics, Chapel Hill, NC, USA). The method is based on the aerodynamic principle that the area of an orifice can be estimated by simultaneously measuring the differential pressure between the two sides of the orifice and the airflow that passes through it (Figure 1).

The equation $A = V/k(2\Delta P/d)^{1/2}$, where A is the area of the orifice in cm^2; V is the nasal flow in cm^3/s; k is 0.65; ΔP is the oral-nasal pressure in $dynes/cm^2$; and d is the air density $(0.001\ g/cm^3)$ is used to measure the minimum cross-sectional area of the velopharynx during the production of the consonant
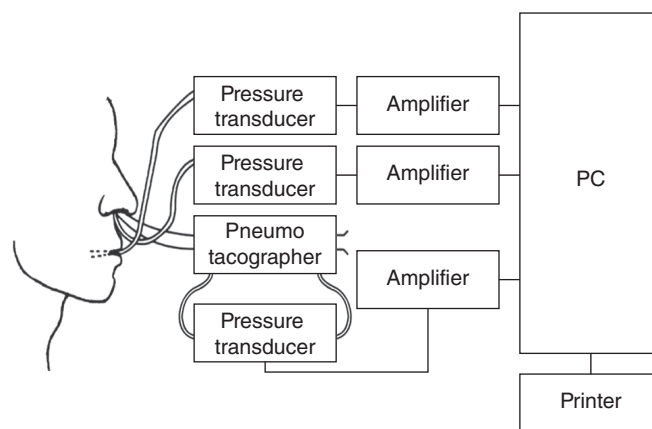
Figure 1. Instrumentation to determine the area of the velopharyngeal orifice[11] (PERCI-SARS System, Microtronics, Chapel Hill, NC, USA)

/p/ inserted in the word "rampa", in the sentence "Papai pintou a rampa", as aforementioned[11,12]. The velopharyngeal closure was classified according to the velopharyngeal orifice area based on criterion adapted from literature[13]: 0–4.9 mm² = adequate velopharyngeal closure; 5.0–19.9 mm² = borderline; and ≥20 mm² = inadequate.

Speech sample recording

All of the audio-digital recordings were recorded in a soundproof booth with the software Wave Studio (Creative Labs) and a headset microphone PRA-30 XLR (Superlux) laterally placed 5 cm away from the patient's mouth. The speech sample was comprised of a 10-sentence set mostly containing words with the consonant /p/ because this is the target sound used in the aerodynamic evaluation. The patient was oriented to read the sentences. Those who were incapable of reading repeated the sentences after the evaluator. Speech sample recording and aerodynamic evaluation were performed on the same day.

Analysis of the perceptual parameters of speech

The speech samples were analyzed by three speech language pathologists experienced in the diagnosis of VPD, who individually classified hypernasality in a 4-point scale: 1=absent hypernasality (normal resonance); 2=mild; 3=moderate; and 4=severe. Audible NAE and NR were classified as follows: 1=absent or 2=present. The samples were recorded in three CDs and 20% of the sample was duplicated for the intra-rater analysis. The CDs also had speech samples representative of the four degrees of hypernasality, in order to be used as reference in the resonance classification. In total, each rater analyzed 120 speech samples.

Data analysis

The final score of hypernasality was established as score with more occurrences among the three raters. The percentage of inter- and intra-rater agreement was established using the Kappa coefficient[14]. The correlation between the perceptual aspects of speech (absent, mild, moderate, and severe degree of hypernasality, audible NAE, and present or absent NR) and the velopharyngeal closure was analyzed using the Spearman correlation coefficient. The Fisher's exact test and the χ^2-test were used to verify the association between these same variables, when expressed qualitatively. An ordinal logistic regression model was established to predict the scoring of velopharyngeal closure according to these same speech parameters. For all these tests, a 5% significance level was considered ($p < 0.05$).

RESULTS

Perceptual analysis of speech characteristics

The agreement between the raters in the analysis of the 100 speech samples was moderate for hypernasality (0.41) and audible NAE (0.59), and substantial for NR (0.72). In 43 speech samples, there was total agreement between the evaluators,

with Kappa index of 1.0, therefore being interpreted as almost perfect. The intra-rater agreement was almost perfect for the three analyzed speech characteristics.

Correlation between velopharyngeal orifice area and speech parameters

Hypernasality

The Spearman correlation coefficient showed significant correlation ($p < 0.000$; $r = 0.581$) between hypernasality and velopharyngeal area. Likewise, the Fisher's exact test indicated significant association ($p < 0.000$) between the level of hypernasality and the velopharyngeal closure, as shown in Table 1.

Audible nasal air emission

The Spearman correlation coefficient showed significant correlation ($p < 0.000$; $r = 0.547$) between audible NAE and the measurement of velopharyngeal area. The χ^2-test results showed significant association ($p < 0.000$) between the variables (Table 2).

Nasal rustle

The Spearman correlation coefficient showed significant correlation and the χ^2-test showed significant association between the presence of NR and velopharyngeal closure ($p = 0.005$). However, in this case, the Spearman correlation was negative ($p = 0.004$; $r = -0.287$), as shown in Table 3.

Table 1. Distribution of patients according to degree of hypernasality, perceptually assessed, and velopharyngeal closure, measured by velopharyngeal area

Hypernasality	Velopharyngeal closure			Total
	Adequate	Borderline	Inadequate	
	0–4.9 mm²	5.0–19.9 mm²	>20 mm²	
Absent	25	3	5	33
Mild	20	14	14	48
Moderate	0	3	14	17
Severe	0	0	2	2
Total	45	20	35	100

Fisher's exact test ($p < 0.000$)

Table 2. Distribution of patients according to the presence and the absence of audible nasal air emission, perceptually measured, and velopharyngeal closure, measured by the velopharyngeal measurement

Nasal air emission (audible)	Velopharyngeal closure			Total
	Adequate	Borderline	Inadequate	
	0–4.9 mm²	5.0–19.9 mm²	>20 mm²	
Absent	42	13	12	61
Present	3	7	23	39
Total	45	20	35	100

χ^2-test ($p < 0.000$)

Ordinal logistic regression model

The χ^2-test showed that the model presented a statistically significant adjustment (p=0.000). Considering the reduced number of participants classified with severe hypernasality (2/100), it was decided to group the scores moderate and severe in this analysis. Besides, considering that the inter-rater regarding the degree of hypernasality was moderate, it was also chosen to use only the samples that obtained 100% agreement to improve the reliability of the results of this analysis. Table 4 shows the percentage of predicted patients according to the logistic model related to the real classification obtained in the sample. According to this model, of the 43 (100%) assessed individuals, 72% (31) were predicted in the correct category.

Table 3. Distribution of patients according to the presence and the absence of nasal rustle, perceptually measured, and velopharyngeal closure, measured by velopharyngeal area

Nasal rustle	Velopharyngeal closure			Total
	Adequate 0–4.9 mm²	Borderline 5.0–19.9 mm²	Inadequate >20 mm²	
Absent	22	14	29	65
Present	23	6	6	35
Total	45	20	35	100

χ^2-test (p=0.005)

Table 4. Distribution of 43 samples that presented total agreement between raters as to the degree of hypernasality and the prediction of velopharyngeal closure related to hypernasality, audible nasal air emission, and nasal rustle

Real VPC	Prediction of VPC according to the model			Total (%)
	Adequate VPC (%)	Borderline VPC (%)	Inadequate VPC (%)	
Adequate VPC	96	4	0	100
Marginal VPC	36	46	18	100
Inadequate VPC	20	30	50	100
Total	63	21	16	100

Caption: VPC = velopharyngeal closure

DISCUSSION

The difficulty to perceptually rate the speech symptoms and the need to achieve good agreement between the judgments of different raters make it legitimate to correlate these findings with instrumental methods because obtaining a significant correlation will lead to more reliable results in studies and clinical practice. The results of this study indicate a linear and clinically relevant relationship between speech symptoms (hypernasality, NAE, and NR) and the velopharyngeal orifice size (velopharyngeal closure).

The individual analysis of hypernasality results showed a stronger association between the degree of hypernasality and velopharyngeal closure in the two extremes of both modalities of evaluation. These results corroborate the findings of authors who verified that the characteristics of hypernasality can predict the velopharyngeal gap size in cases of small and large gaps assessed by nasendoscopy[3]. According to literature, the categories of hypernasality between both extremes (normal and severe) are usually associated with borderline VPD[15-18]. Therefore, the results in this study lead to the agreement that the attempt to explore the mild degree of hypernasality is probably the key to understand borderline velopharyngeal insufficiency[18].

Data analysis showed a significant correlation between these two measurements, hypernasality and velopharyngeal gap, thus indicating that the velopharyngeal area increased when the degree of hypernasality increased. These results corroborate previous studies[3,7,10,19] that also showed a significant correlation between hypernasality and velopharyngeal gap. However, most of these studies used direct instrumental methods; however, they also kept their subjective aspect to analyze the correlation between different aspects of the velopharyngeal function. Studies comparing the performance of nasendoscopy and videofluoroscopy in the judgment of velopharyngeal closure showed that nasendoscopy tends to underestimate the size of velopharyngeal opening in comparison with videofluoroscopy[20,21]. However, better results in the nasendoscopy concerning the agreement between the perceptual judgment of the velopharyngeal function and the function of the velopharyngeal mechanism assessed by images were found in another study[22]. These findings show the limitations of this type of evaluation and reinforce the importance of studies that use objective and quantitative methods of the velopharyngeal functioning to investigate the correlation with speech features.

In a single study that correlated hypernasality with the velopharyngeal gap size using the pressure-flow technique[19], the authors observed moderate correlation between the degree of hypernasality and velopharyngeal closure. These authors pointed out as a limitation of this study the fact that the velopharyngeal area was measured based on the emission of a single word (hamper). However, according to international parameters of perceptual analysis of speech[23,24], it is also possible to consider as a limitation of this study the fact that hypernasality was only judged by a single rater. In the present study, it was important to make the perceptual judgment as reliable as possible, submitting the speech samples to the analysis of three experienced raters. Besides, the aforementioned authors included individuals with reduced nasal area (subnormal values) to verify if the values of nasal area could influence the speech resonance. This study only included individuals with good nasal permeability confirmed by posterior rhinomanometry, that is, with nasal area values within the limits of normality. This is because a decrease in nasal airway patency resulting from the temporary nasal

congestion, for example, could NR and interfere with the perceptual judgment of audible NAE and NR, besides hypernasality itself[5].

Concerning audible NAE, the results also showed significant correlation with the velopharyngeal area, thus indicating that, at the presence of the symptom, the velopharyngeal gap tends to be larger. Studies using nasendoscopy and videofluoroscopy also showed correlation between these two variables[3,10]. However, other authors did not find a significant correlation between NAE and the velopharyngeal gap size[7]. However, these authors used a specific scale in the perceptual analysis of speech, called Pittsburgh Weighted Speech Scale, in which velopharyngeal insufficiency is rated based on the combination of nasality, NAE, facial grimacing, vocal characteristics, and compensatory articulation[25]. One can speculate that that the use of a scale containing more levels can increase the possibilities of variations, so it is more difficult to correlate the analyzed variables.

The presence of NR also revealed a statistically significant correlation with velopharyngeal closure. However, in this case, it is curious fact that the correlation was negative, which means that the presence of NR is related to smaller values of velopharyngeal area. These results confirm what can be seen by using nasendoscopic evaluations: NR occurs in the presence of a small velopharyngeal opening[26] as a consequence of the increasing orifice resistance to airflow, and is a speech aspect that significantly contributes with the prediction of a small velopharyngeal gap[5].

Considering that the correlation between the speech parameters and the velopharyngeal closure was statistically significant, a logistic regression model was used to analyze if the degree of hypernasality and the presence of audible nasal air emission or nasal rustle could estimate the scoring of velopharyngeal closure. It is worth mentioning that the fact that the correlation has been more evident among the extreme categories, both in the classification of hypernasality and in velopharyngeal closure, led us to consider the reliability of the results obtained from the moderate agreement between the different raters as to the degree of hypernasality in the 100 speech samples. Therefore, to obtain more reliable results, the logistic regression model was elaborated using the speech samples that obtained 100% agreement between raters in the judgment of hypernasality, that is, 43 samples whose classification of the degree of hypernasality was unanimous among the three raters. This model classified 72% samples in the correct category of velopharyngeal closure, so it was possible to predict the intermediate category, the borderline velopharyngeal closure.

So, this study showed that even though the correlation between the perceptual aspects of speech and the objective measurement of velopharyngeal closure is not total, this correlation is significant and, according to the ordinal logistic regression, the model strongly suggests that the speech features can help and predict velopharyngeal closure. One can speculate that these results might have been even better if we had selected only the samples with 100% agreement between the raters for the other speech categories, NAE and NR.

In the future, the intention is to propose a protocol of velopharyngeal closure based on the most representative perceptual parameters of speech of individuals with cleft palate and on the results obtained by the objective assessment of velopharyngeal functioning. It is suggested that the development of a protocol that establishes the correspondence between the perceptual aspects of speech and the possible results obtained in the objective evaluation may provide more reliable data and, therefore, assist the diagnosis and the definition of treatment for patients with VPD.

CONCLUSION

There is a significant correlation between the velopharyngeal gap size and the degree of hypernasality, and the presence of audible NAE and NR. These results reinforce the importance of using pressure-flow technique as a complementary objective method in the diagnosis of velopharyngeal dysfunction, and suggest that perceptual aspects of speech can predict velopharyngeal closure, therefore assisting the speech-language pathologists to define a more effective diagnosis and treatment for velopharyngeal dysfunction.

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