

Agreement between perceptual tests and videofluoroscopy in the diagnosis of velopharyngeal dysfunction

Concordância entre os testes perceptivos e a videofluoroscopia no diagnóstico da disfunção velofaríngea

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ABSTRACT

Purpose: To determine the agreement between the results of the Nasal Air Emission and Hypernasality tests and the videofluoroscopy findings in the diagnosis of velopharyngeal dysfunction in individuals with cleft lip and palate. **Methods:** The sample consisted of 89 scores of Nasal Air Emission and Hypernasality tests and 89 judgments of videofluoroscopy recordings, which were interpreted as *consistent velopharyngeal closure*, or as *inconsistent velopharyngeal closure*, or as *non velopharyngeal closure*. The sensitivity, specificity and agreement between the interpretation of the results of the perceptual tests and the findings of the videofluoroscopy were calculated. **Results:** The rates found for sensitivity of Nasal Air Emission and Hypernasality tests were 98% and 96%, respectively, and the rates for specificity of Nasal Air Emission and Hypernasality tests were 37% and 63%, respectively. Regarding the percentages of agreement between the Nasal Air Emission test scores and the videofluoroscopy judgments, it was found an agreement of 62% for the *consistent velopharyngeal closure* condition, 43% for the *inconsistent velopharyngeal closure*, and 68% for the *non velopharyngeal closure*. Between the scores of Hypernasality test and videofluoroscopy judgments the agreement found was 70% for the *consistent velopharyngeal closure* condition, 47% for the *inconsistent velopharyngeal closure* and 77% for the *non velopharyngeal closure*. **Conclusion:** There was a good level of agreement between the perceptual tests and the videofluoroscopy judgments for the *consistent velopharyngeal closure* and *non velopharyngeal closure* conditions, but not for the *inconsistent velopharyngeal closure*.

Keywords: Cleft palate; Fluoroscopy; Velopharyngeal insufficiency; Diagnosis; Speech

RESUMO

Objetivo: Verificar a concordância entre os resultados dos Testes de Emissão de Ar Nasal e de Hipernasalidade e os achados do exame de videofluoroscopia no diagnóstico da disfunção velofaríngea, em indivíduos com fissura labiopalatina. **Métodos:** A amostra foi constituída por 89 exames de videofluoroscopia e 89 escores dos Testes de Emissão de Ar Nasal e de Hipernasalidade, interpretados como *fechamento velofaríngeo consistente*, ou como *fechamento velofaríngeo inconsistente*, ou ainda, como *não fechamento velofaríngeo*. Foram calculadas a sensibilidade, a especificidade e a concordância entre a interpretação dos achados dos testes perceptivos e os achados da videofluoroscopia. **Resultados:** Foram encontrados índices de sensibilidade e especificidade de 98% e 37%, respectivamente, para o Teste de Emissão de Ar Nasal e de 96% e 63%, respectivamente, para o Teste de Hipernasalidade. As porcentagens de concordância entre os escores do Teste de Emissão de Ar Nasal e os exames de videofluoroscopia e entre os escores do Teste de Hipernasalidade e os exames de videofluoroscopia, para a categoria *fechamento velofaríngeo consistente*, foram de 62% e 70%, respectivamente, de 43% e 47%, para a de *fechamento velofaríngeo inconsistente*, respectivamente, e de 68% e 77%, para a de *não fechamento velofaríngeo*, respectivamente. **Conclusão:** Houve um bom nível de concordância entre os testes perceptivos e os exames de videofluoroscopia para as categorias *fechamento velofaríngeo consistente* e *não fechamento velofaríngeo*, mas não para a de *fechamento velofaríngeo inconsistente*.

Descritores: Fissura palatina; Fluoroscopia; Insuficiência velofaríngea; Diagnóstico; Fala

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INTRODUCTION

The velopharyngeal insufficiency and incompetence are types of velopharyngeal dysfunction (VPD) which require different kinds of treatment. For cases with velopharyngeal insufficiency, a physical procedure (surgery or palatal prosthesis) is always indicated, and for those with velopharyngeal incompetence, the procedure to be indicated is speech therapy. However, it is quite common to find both types of VPD in the same patient. In this case, a physical procedure combined with speech therapy should be indicated⁽¹⁾.

No effective treatment can be performed if the precise differential diagnosis of the type of VPD is not made and to achieve it, it is necessary to perform a perceptual and instrumental evaluation. There are some procedures to evaluate the changes of the velopharyngeal mechanism (VPM) and thus diagnose the presence of VPD. Clinically, the speech pathologist has the perceptual assessment as an important tool in the diagnosis of VPD, and therefore indicators of the clinical significance of the changes in speech related to VPD can be performed^(2,3). Instrumental methods such as videofluoroscopy and nasoendoscopy can provide information on the functional anatomy of the VPM, as well as nasometry and the pressure-flow technique can respectively measure the acoustic and aerodynamic parameters of the velopharyngeal function⁽⁴⁾. They may also provide the perceptual critical finding trial, enable the diagnosis of the velopharyngeal function in cases where the clinical diagnosis by itself was not possible, and also determine the effectiveness of the proposed treatment⁽⁵⁻⁹⁾.

Among the various methods for perceptual evaluation described in the literature, few of them provide the differential diagnosis of VPD, as the Tests of Hypernasality (THYPER) and Nasal Air Emission (TNAE)⁽⁵⁻¹⁰⁾, with the aim of assisting assessment of velopharyngeal function and dysfunction. The use of these tests is very practical and simple and can be performed in children younger than 4 years. The tests are standardized on a set of 10 words, each with a base of 10 indicators of the frequency of occurrence of nasal air emission and hypernasality. The interpretation of each test is made so that the indicators 0/10, 1/10 or 2/10 indicate velopharyngeal closure and 3/10 to 10/10 indicate presence of VPD. From the analysis of these tests, it is possible to set a default for each patient. The score of 10/10 for both tests in a given patient might mean presence of velopharyngeal insufficiency by indicating the absence of consistent velopharyngeal closure. Scores between 1/10 and 9/10 in both tests might mean velopharyngeal incompetence, since these scores indicate the possibility of inconsistent velopharyngeal closure⁽¹⁰⁾.

Videofluoroscopy is a videorecording instrumental direct technique that allows the dynamic evaluation of the VPM structures during speech. Although it is a technique which makes use of radiation, it is performed for a short time. With

the visualization of the anatomical and physiological characteristics of such structures, it is possible to identify the cause of VPD and the best treatment for the patient.

The perceptual tests of TNAE and THYPER have great value for professionals who do not have instrumental techniques in their routine, and even to large centers, which can better select patients for instrumental techniques. However, there are no studies that prove the indication of these tests as an option for the differential of VPD diagnosis⁽¹⁰⁾.

Seeking to prove the validity of TNAE and THYPER tests in clinical assessment of patients with VPD, the aim of this study was to verify the correlation between the results of TNAE and THYPER tests and the videofluoroscopy findings in the diagnosis of VPD in individuals with cleft lip and palate.

METHODS

This project was approved by the Ethics Committee (CEP) of the Hospital for Rehabilitation of Craniofacial Anomalies, Universidade de São Paulo (HRAC/USP), N° 377/2011 and 5/2013- SVAPEPE -CEP. Data were collected from speech assessment and videofluoroscopy protocols pre-existing in the patients records.

Sample

The sample was formed by means of the scores obtained in the Tests of Nasal Air Emission (TEAN) and Hypernasality (THYPER) and the results of videofluoroscopy exams contained in patient charts. As inclusion criteria, both perceptual tests and videofluoroscopy should have been performed on the same patient visit to the HRAC/USP.

After defining the inclusion criteria it was found the number of patients undergoing the videofluoroscopy and how many of them presented the protocol of this exam in their respective charts. Two hundred and twenty one videofluoroscopic recordings were found from a total of 112 patients. The second step was to verify which of them had also undergone clinical evaluation of speech, including the THYPER and TNAE tests at the same visit at the hospital. That resulted in 187 exams. Subsequently it was found that 98 cases had not a complete videofluoroscopy protocol, according to the interests of this study. Thus, the sample consisted of 89 videofluoroscopy protocols and 89 judgments of THYPER and TNAE scores from a series of 73 patients of both genders, with age between 5 and 15 years (mean = 9 years and 2 months).

Procedures

After the sample selection, the collection of the perceptual tests and the videofluoroscopic data from each patient chart was performed.

Test of Nasal Air Emission (TNAE)

To perform this test, a mirror was positioned directly under the patient's nose, while he repeats ten words (*papai, papel, piupiu, pepê, popô, babá, bebê, bobi, boba, bibi*). This technique is based on viewing the presence or absence of the nasal air emission during the repetition of each word by condensation of air in the mirror. The score obtained reflects the number of words that presented nasal air emission (Figure 1).



Figure 1. Procedures for Nasal Air Emission Test

Test of Hypernasality (THYPER)

To perform this test, the patient was asked to repeat 10 words (*babá, bebê, bibi, bobó, bubu, baba, bebe, bobi, boba, buba*) twice, once with the nostrils occluded and again with the nostril non-occluded. This technique is based on the difference in the quality of audible resonance, characterized by the presence of nasalization. Under the velopharyngeal closure conditions, in which there is no acoustic energy passing through the velopharyngeal mechanism, there should be no noticeable change in the quality of resonance, not even with the nostrils occluded, nor with the same left open. The score obtained reflects the number of words in which there was a difference in audible resonance (Figure 2).



Figure 2. Procedure for Hypernasality Test

For the analysis of the scores of THYPER and TNAE tests the following classification suggested by Bzoch (2004)⁽¹⁰⁾

was established: scores between 0/10 and 2/10 were classified as *consistent velopharyngeal closure* (CC); between 3/10 and 7/10 as *inconsistent velopharyngeal closure* (IC), and scores between 8/10 and 10/10 as *non velopharyngeal closure* (NC).

Videofluoroscopy

The equipment used for videofluoroscopy was the fluoroscopic type, composed of a closed circuit television, one X-ray machine with an image intensifier and a videorecording system (Arch Arm BV - Pound Philips®). One cephalostat for fixing the individual's head in a constant position was also used (Figure 3).



Figure 3. Videofluoroscopy system: TV monitors (1), video-recording system (2), X ray unit (3) ray image intensifier (4) and cephalostat (5)

Videofluoroscopy was conducted by an experienced speech pathologist in performing this procedure with the aid of a radiographer in handling the equipment. Before each exam, each patient ingested 5 mL of barium sulfate and 2 mL of this contrast was also applied in each nostril. The lateral view was taken during the emission of syllables, words and sentences with oral and nasal phonemes (Figure 4).

Although the videofluoroscopy protocol (Appendix 1) contains several information about the velopharyngeal mechanism structures and its function, it was of interest for this study to know whether or not the patient had a touch of the soft palate in the posterior pharyngeal wall (or in the pharyngeal tonsil when present). For this study it was established that an exam would be considered indicative of *consistent velopharyngeal closure* (CC) when touching the soft palate in the posterior pharyngeal wall during all emissions; indicative of *inconsistent velopharyngeal closure* (IC), when the touch occurred in at least one emission, and indicative of *non velopharyngeal closure* (NC), if the touch never happened.



Figure 4. Videofluoroscopy lateral view showing velopharyngeal closure during speech

Efficacy of TNAE and THYPER Perceptual Tests

The clinical applicability of an instrument can be defined by its level of efficiency, which is determined by the rates of Sensitivity and Specificity. In this analysis it was investigated the ability of the TNAE and THYPER Tests to distinguish between the presence and absence of *velopharyngeal closure* and to provide an estimate of the value of the perceptual tests to confirm the findings of videofluoroscopy.

Sensitivity of the TNAE and THYPER refers to the frequency which each test identified the *absence of velopharyngeal closure*, when this absence is also been observed in videofluoroscopy. Specificity refers to the frequency which each test identified the *presence of velopharyngeal closure*, when this presence is also observed in videofluoroscopy. To perform this analysis, it was necessary to define only two categories, the *velopharyngeal closure* and *non velopharyngeal closure*. The *velopharyngeal closure* category consisted of the CC and IC groups and the *non velopharyngeal closure* category remained with the NC group.

Statistical analysis

Once the clinical sample did not allow a homogeneous distribution of the exams in the three categories of closure (CC, IC, NC), the Kappa test was not considered appropriate to establish the correlation in this study. Statistical analysis was then performed by calculating the percentage of agreement and through the establishment of efficiency levels of the tests (Sensitivity and Specificity). This analysis was performed considering the two tests separately, ie, the percentage of agreement was calculated between the interpretation of the findings of TNAE test and the videofluoroscopic findings, and between the interpretation of the THYPER test and videofluoroscopic findings.

RESULTS

Efficiency indices of the TNAE and THYPER tests

The indices of sensitivity and specificity were 98% and 37% for TEAN, and 96% and 63% for THYPER, respectively.

Agreement between the TNAE test scores and the videofluoroscopy findings

Out of the 8 patients with scores between 0/10 and 2/10 (interpreted as CC) in the TNAE test, 5 (62%) were considered to be CC for the videofluoroscopy and 3 (38%) as IC, showing a 62% of agreement between the results. Out of the 7 patients with scores between 3/10 and 7/10 (interpreted as IC), 3 (43%) were considered by videofluoroscopy as CC, 3 (43%) as IC, and 1 (14%) as NC, showing an agreement of 43% between the results. Out of the 74 cases with scores between 8/10 and 10/10 (interpreted as NC), 10 (13%) were considered CC by videofluoroscopy, 14 (19%) as IC, and 50 (68%) as NC, showing an agreement of 68% between the results (Table 1).

Table 1. Distribution of scores obtained in the TNAE test

| Velopharyngeal condition judged by videofluoroscopy | TNAE Scores | | | Total |
|---|-------------|----------|-----------|-----------|
| | CC | CI | NC | |
| CC | 5 | 3 | 10 | 18 |
| CI | 3 | 3 | 14 | 20 |
| NC | 0 | 1 | 50 | 51 |
| Total | 8 | 7 | 74 | 89 |

Note: TNAE = Nasal Air Emission Test; CC = Consistent velopharyngeal closure; IC = Inconsistent velopharyngeal closure; NC = Non velopharyngeal closure

Agreement between the THYPER results and the videofluoroscopy findings

Out of the 13 patients with scores between 0/10 and 2/10 (interpreted as CC) in the THYPER test, 9 (70%) were also considered CC by videofluoroscopy, 2 (15%) as IC, and 2 (15%) as NC, showing an agreement of 70% between the results. Out of the 15 patients with scores between 3/10 and 7/10 (interpreted as IC), 6 (40%) were considered CC by videofluoroscopy, 7 (47%) as IC, and 2 (13%) as NC, showing an agreement of 47%. Out of the 61 patients with scores between 8/10 and 10/10 (interpreted as NC), 3 (5%) were considered CC by videofluoroscopy, 11 (18%) as IC, and 47 (77%) as NC, showing an agreement 77% between the results (Table 2).

DISCUSSION

The results of this study have shown that there was a poor agreement between the cases with scores of 3/10 and 7/10 for

Table 2. Distribution of scores obtained in the THYPER test

| Velopharyngeal condition judged by videofluoroscopy | THYPER Scores | | | N |
|---|---------------|----|----|----|
| | CC | IC | NC | |
| CC | 9 | 6 | 3 | 18 |
| IC | 2 | 7 | 11 | 20 |
| NC | 2 | 2 | 47 | 51 |
| Total | 13 | 15 | 61 | 89 |

Note: THYPER = Test of Hypernasality; CC = Consistent velopharyngeal closure; IC = Inconsistent velopharyngeal closure; NC = Non velopharyngeal closure

the TNAE, as well as the THYPER and judgment of velopharyngeal condition by videofluoroscopy, unlike the cases with scores between 8/10 and 10/10, most of whom had agreement with the videofluoroscopic findings. The results agree with another study⁽⁹⁾ which tested the agreement between perceptual tests (TNAE and THYPER) and nasoendoscopy for the diagnosis of VPD, using the same criteria adopted in this study.

Since none study that has compared findings of TNAE and THYPER with the findings of videofluoroscopy has been found in the literature, the comparison of the obtained results with other studies was limited. However, one study found 91% of agreement between the THYPER and the aerodynamic measures of 10 normal subjects⁽¹¹⁾. It was found 100% of agreement between the presence of nasal air emission, evaluated by a five-point scale and the presence of velopharyngeal gap, identified by videofluoroscopy, in a study that investigated whether the speech symptoms were predictive of the velopharyngeal condition⁽¹²⁾. Other authors compared the nasoendoscopy, videofluoroscopy and perceptual assessment for the diagnosis of VPD and found a strong relationship between velopharyngeal gap size and degree of hypernasality, with a better relation with nasoendoscopy⁽¹³⁾. One study investigated the relationship between perceptual characteristics of hypernasality, assessed by a three-point scale (mild, moderate, severe) and nasal air emission, measured by a dichotomous scale of presence and absence, and the size of the velopharyngeal gap, estimated by videofluoroscopy and nasoendoscopy. The findings have shown a higher association between severe hypernasality and velopharyngeal large gap⁽¹⁴⁾. Another study used videofluoroscopy, nasoendoscopy and perceptual assessment to evaluate the results of the secondary palatal surgery for correction of VPD. The results have shown that the perceptual assessment was confirmed by videofluoroscopy and it was compatible with the nasoendoscopy⁽¹⁵⁾.

Similar to the NC velopharyngeal condition, the CC category also showed good levels of agreement for both the TNAE and THYPER tests, unlike the IC category, which showed lower levels of agreement. The vocal literature extensively discusses the fact that the tests and perceptual assessment tools have good agreement at the extremes, where the ear has good accuracy, and fails in the points between them, where the ear

has difficulty. In some studies it was found that voice judges disagreed about what constituted normality or severity, ie the extremes, while disagreed on the scores between them⁽¹⁶⁻¹⁸⁾. In some voice studies it was found that judges agreed more about what constituted normality or severity, ie the extremes, while disagreed on the scores between them⁽¹⁶⁻¹⁸⁾.

Some hypotheses could explain the results found in this study for the IC velopharyngeal condition, one of them may be the limited number of cases in the sample representative of IC, as well as for the CC condition, which also had a small number of cases in the sample. On the other hand, it is not expected that a case that presents suggestive CC scores in listening tests are referred to videofluoroscopy, since the clinical examination would be enough to perform the diagnosis, with no need to expose the patient to radiation without a real need. The cases in this study that present CC in the perceptual tests were referred to videofluoroscopy for other reasons than for the diagnosis of VPD, such as to evaluate the position of the tongue during velopharyngeal closure and for post-operative evaluation of tonsillectomy.

Another hypothesis that could explain the low agreement for the IC condition would be the possibility that some cases could have shown some degree of nasal obstruction, which could influence the results of TNAE. Patients with cleft lip and palate have deformities in the nasal cavity, which tend to reduce the size of the airways^(19,20). Thus, if a patient from this study presented with nasal obstruction during TNAE, this could mask the presence of the nasal air emission and interfere with the test results.

Another aspect that may have influenced the results of this study is the possibility of the occurrence of compensatory articulation (CA), even with the effort of the speech pathologist to eliminate it by means of diagnostic therapy. The CA usually occur in the pharynx or larynx, with an attempt of generating pressure to produce a plosive or fricative sound. When this happens, the individual does not make use of the velopharyngeal mechanism, and therefore presents hypernasality and/or nasal air emission, but actually has anatomical conditions for velopharyngeal closure⁽²¹⁾.

Some cases that were judged to present NC in the TNAE and/or in the THYPER presented judgement of CC or IC condition in the videofluoroscopy. One aspect that might have influenced this result is the presence of fistula in some case samples. Even with the attempt to seal that during evaluation, it is possible that air and/or acoustic energy detected in the tests went through the fistula⁽²²⁾. This finding may also be explained by the fact that the videofluoroscopic analysis of the present study was performed only through the lateral view, which may have precluded the identification of cases that presented asymmetric velopharyngeal closure, in which only a portion of the soft palate touches the posterior pharyngeal wall, causing a velopharyngeal gap⁽²³⁾. Moreover, it appears that videofluoroscopy may have indicated velopharyngeal closure, when in fact,

there was not the touch of the soft palate in the posterior wall of the pharynx or tonsil. A study found that videofluoroscopy in lateral view, often underestimates the degree of velopharyngeal insufficiency when compared with basal vision and nasoendoscopy⁽²⁴⁾. Another study found similar results, in which videofluoroscopy overestimated velopharyngeal closure when compared with nasoendoscopy and perceptual assessment⁽¹³⁾.

The efficacy of the tests in this study reported rates of 98% of sensitivity and 37% of specificity for the TNAE and 96% of sensitivity and 63% of specificity for the THYPER, indicating that the efficacy was good only for identifying the condition of non velopharyngeal closure.

Low levels of specificity can be justified by the need to include the IC condition in one of the closure or non-closure conditions for efficacy calculation. Arbitrary criterion for joining the CC and IC into velopharyngeal closure was adopted. Thus, a good part of individuals who had IC on videofluoroscopy and were judged by perceptual tests as NC caused the specificity levels to be low, especially for TNAE.

CONCLUSION

There was a good level of agreement between the perceptual tests and the videofluoroscopy for the *consistent velopharyngeal closure* (CC) and *non-closure* (NC), but not for the *inconsistent closure* (IC).

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Appendix 1. Speech Videofluoroscopy Protocol, the Hospital of Craniofacial Anomalies of Universidade de São Paulo

Date: ____ / ____ / ____ RG: _____
 Patient' name: _____ Age : _____
 Reason for referral: _____
 Cleft type: _____ tape nº: _____

Barium sulfate

no yes
 mouth nose

Cephalostat

não sim

Duration: _____

Kv: _____ mA: _____

Soft palate

NA no yes
 knee at palatal plane movement
 knee above palatal plane
 knee below plane
 knee absence

Passavant' pad

NA No Yes
 palatal plane
 below palatal plane

OBS: _____

Posture of tongue

NA
 normal function
 tongue retraction against soft palate
 tongue retraction towards posterior pharyngeal wall
 tongue contact with posterior pharyngeal wall

Front view: () NA

Movement of lateral walls

no yes
 symmetric up tp 50 % of the midline
 asymmetric beyond 50 % of the midline
 touching in the midline

Cephalometric measurements: () NA

| Measures at rest | Patient | Min | Max |
|------------------|---------|-----|-----|
| VP Extension | | | |
| VP Thickness | | | |
| NF Depth | | | |
| PNF/EV | | | |

Suggestive of VP closure

NA no yes soft palate contact:
 ppw
 adenoid
 passavant's pad
 palatal prosthesis

Adenoid

NA no yes
 about 1/3 of np space
 about 2/3 of np space
 more than 2/3 of np space

Palatine tonsils

- NA not present not visible
- present
- occupy large space of oropharynx
- makes contact with the ppw
- compromise velar elevation

Basal view: NA

Velopharyngeal closure

- no yes _____

In speech (better performance):

Gap size : _____ mm

Gap height: _____ mm

Velar angle: _____ degree

Conduct

- nasoendoscopy
- surgical correction
- tonsillectomy may favor speech
- assessment of respiratory function
- palatal prosthesis speech therapy
- adenoidectomy may facilitate breathing and / or speech
- adenoidectomy may impair speech
- other _____

Observations: _____

