Avaliação perceptiva e instrumental da função velofaríngea na fissura de palato submucosa assintomática****

Perceptual and instrumental assessment of velopharyngeal function in asymptomatic submucous cleft palate

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Abstract

Background: submucous cleft palate (SMCP) may or may not be associated to symptoms of velopharyngeal dysfunction (VPD). Aim: to verify whether patients with SMCP, who were considered as asymptomatic on a perceptual speech evaluation, present absence of hypernasality and adequate velopharyngeal closure on an instrumental assessment. Method: 22 patients with SMCP and with no VPD symptoms, of both genders, aged 6 to 46 years, underwent speech acoustic assessment (nasometry) - to determine nasalance scores - and speech aerodynamic assessment (pressure-flow technique) - to verify velopharyngeal closure. The complete agreement between the findings on the perceptual and instrumental assessments was the tested null hypothesis. Results: the pressure-flow technique confirmed the perceptual findings, that is, all patients were diagnosed as having adequate velopharyngeal function on both types of assessment. Nasometry results confirmed the perceptual findings in 15 out of the 20 analyzed patients (75% of the cases). The remaining 5 patients (25%) presented nasalance scores that suggest hypernasality (nasometry). This symptom was not identified by the perceptual evaluation, yielding to the rejection of the null hypothesis. Conclusion: results indicate the importance of the combined use of perceptual and instrumental assessments for the diagnosis of VPD in SMCP cases. The follow-up of patients diagnosed as asymptomatic by a perceptual speech evaluation and presenting evidences of VPD on instrumental assessment (e.g. nasometry) is recommended, particularly when dealing with children who are more prone to develop symptoms with aging.
Key Words: Cleft Palate; Velopharyngeal Insufficiency; Rhinomanometry.

Resumo

Tema: a fissura de palato submucosa (FPSM) pode estar associada, ou não, a sintomas de disfunção velofaríngea (DVF). Objetivo: o presente estudo teve por propósito verificar se pacientes com FPSM diagnosticados como assintomáticos em uma avaliação perceptiva da fala apresentam ausência de hipernasalidade e fechamento velofaríngeo adequado em exame instrumental. Método: vinte pacientes com FPSM e sem sintomas de DVF, de ambos os gêneros, com idade entre 6 e 46 anos, foram submetidos à avaliação acústica da fala (nasometria), para a determinação da nasalância, o correlato acústico da nasalidade, e, à avaliação aerodinâmica da fala (técnica fluxo-pressão), para a determinação do fechamento velofaríngeo. A total concordância entre os resultados aferidos na avaliação perceptiva e nas avaliações instrumentais foi a hipótese de nulidade testada. Resultados: a avaliação aerodinâmica confirmou integralmente as observações da avaliação perceptiva, ou seja, todos os 20 pacientes foram diagnosticados como tendo fechamento velofaríngeo adequado em ambas as modalidades de avaliação. Os resultados da nasometria, por sua vez, concordaram com os da avaliação perceptiva em apenas 15 dos 20 pacientes analisados (75% dos casos). Os 5 pacientes restantes (25%) apresentaram escores de nasalância sugestivos de hipernasalidade na nasometria, não constatada na avaliação perceptiva, levando, neste caso, à rejeição da hipótese de nulidade. Conclusão: os resultados mostram a importância do uso combinado de avaliação perceptiva e instrumental para o diagnóstico da DVF em casos de FPSM. Com base nos achados recomenda-se o acompanhamento periódico dos casos considerados assintomáticos em avaliação perceptiva da fala e que apresentem evidências de DVF em uma avaliação instrumental, como a nasometria, particularmente em se tratando de crianças, mais sujeitas ao desenvolvimento de sintomas com o avanço da idade.
Palavras-Chave: Fissura Palatina; Disfunção Velofaríngea; Rinomanometria.

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Introduction

Submucous cleft palate (SMCP) is a congenital malformation characterized by the lack of fusion of muscle and/or bone structures along the midline of the palate, with an intact oral mucosa (Genaro et al., 2004). It is in this last feature that SMCP differs from overt cleft palate that which also involves the mucosal plane.

The diagnosis of SMCP is usually made on the basisbased on of the observation of the following anatomical signals by visual and tactile inspection of the oral cavity: bifid uvula, muscular diastasis of the soft palate and notching of the posterior border of the hard palate (Sommerlad et al., 2004). These signs may appear isolately or combined . In a study developed by Oliveira (2002) in 50 children with SMCP, aged 20 to 40 months, selected during a period of one year These signs may appear isolately or combined. Oliveira (2002), in a study of 50 children with SMCP, aged between 20 and 40 months and selected by order of scheduling over a 1one-year period among the patients seen at of the Hospital for the Rehabilitation of Craniofacial Anomalies (HRCA-USP) of the University of Sao Paulo - , Bauru, Brazil, found that 50% of the cases presented the classical triad, 30% exhibited two of the clinical signs and the remaining 20% showed muscular diastasis only.

SMCP frequently courses with speech disorders, such as hypernasality, nasal air escapeemission, and compensatory articulations, which are related to velopharyngeal inadequacy dysfunction (VPIVPD) caused by the anatomical palate defect, such as hypernasality, nasal air escape, and compensatory articulations (Gosain et al 1999, Senders 2001Park et al., 2000; Kummer, 2001; Senders, 2001; , Ysunza et al 2001; Sommerlad et al., 2002; Yamashita et al., 2002; Johns et al., 2003; Bataiola e Yamashita, 2005). In general, these are the symptoms that lead patients to seek treatment centers where the cleft is identified as the etiological factor and where treatment is provided.

For reasons not yet fully clarified, the same type of anatomical defect of the palate may not cause evident speech symptoms (McWilliams 1991). In this case, when the cleft is identified, at most services including our hospital, the absence of VPI is confirmed by perceptual speech evaluation. The patient is informed about the speech symptoms that might eventually develop and instructed to seek the service in case they do occur. The patient is then excluded from the standard treatment given to cleft invididuals.

For reasons still unclear, SMCP may not cause to VPD symptoms. McWilliams (1991), in a retrospective speech analysis of 130 patients with SMCP seen at the Cleft Palate-Craniofacial Center of the University of Pittsburgh-USA, reported that 44% remained asymptomatic into adulthood requiring no surgical intervention. Of all the cases assessed by Oliveira (2002) at HRCA-USP, 80% were classified as asymptomatic. The difference in the proportion of asymptomatic cases between these studies is likely to be due to the diagnostic criteria used. Regardless of the actual prevalence, the fact is that a considerable number of the patients with SMCP are asymptomatic. Attempting to explain VPD occurrence, or absence, Park (2002) compared the craniofacial morphology of symptomatic children with SMCP (n=22) with that of asymptomatic children (n=24) using cephalometry. Significant differences were found for some variables. However, cephalometric data were not useful for predicting velopharyngeal function in the patients studied.

At HRCA, once SMCP is diagnosed by a multidisciplinary team, a perceptual evaluation of speech is performed (Genaro et al., 2004). If the absence of VPD symptoms is confirmed, the patient is informed about the speech symptoms that might eventually develop and instructed to return to our center in case they do occur. The patient is then discharged from the standard treatment given to cleft individuals.Considering the known limitations of perceptual evaluation the following question arises: have the patients considered asymptomatic been inappropriately discharged from treatment? This questioning is supported by the fact that the perceptual evaluation of velopharyngeal function is influenced by factors such as phonetic context, patient's articulatory pattern and voice quality, examiner's previous experience and expectations (Kummer 2001; Dalston 2004).

Therefore, the objective of this study was to determine whether patients with SMCP considered asymptomatic upon perceptual speech evaluation actually display adequate velopharyngeal closure and no hypernasality upon instrumental evaluation. To answer that question, nasometry and the pressure-flow technique were used.

Nasometry allows the indirect assessment of the velopharyngeal function by measuring nasalance, which corresponds to the relative acoustic energy emerging from the nose during speech. Nasalance is the acoustic correlate of nasality. The method is based on the assumption that an increase in nasalance during the production of a speech sample containing only oral sounds is suggestive of hypernasality, one of the VPD signs. The pressure-flow technique, on its turn, allows the indirect assessment of velopharyngeal function by simultaneously measuring nasal airflow and oral-nasal differential pressure during speech. It is the only method that quantifies the velopharyngeal orifice size, that is, its cross-sectional area (Dalston, 2004, Genaro et al, 2004; Warren, 2004). In this study, nasometry was used to investigate the acoustic repercussions of an eventual VPD associated to SMCP, while the pressure-flow technique was used to assess aerodynamic outcomes.

Method

Casuistics

This study was conducted at the HRCA Laboratory of Physiology with the approval of the institutional Research Ethics Committee (file #103/96- CEP-UEP), and written informed consent was secured from all participants.

Subjects were selected among the patients with SMCP registered at HRCA, from the Bauru region, or identified among the new cases admitted to the hospital, on the basis of the following criteria:

Inclusion criteria:

1. Submucous cleft palate with or without cleft lip.
2. Presence of at least two signs of the classical SMCP triad (bifid uvula, muscle diastasis and hard palate notch).
3. Absence of VPD symptoms, i.e., adequate velopharyngeal function according to perceptual evaluation performed by an experienced speech therapist as described below.
4. Age above 4 years.

Exclusion criteria:

1. Unrepaired cleft lip.
2. Obvious syndromes.
3. Previous palatal surgery.
4. Respiratory or allergic symptoms that could cause nasal congestion upon examination.
5. Physical and mental disability to undergo examination.

Based on these criteria, the sample included a total of 20 individuals (11 females and 9 males) aged 6 to 46 years, of whom 8 had isolated SMCP and 12 had SMCP with cleft lip previously repaired. All cases presented the classical triad of SMCP clinical signs. Having the absence of VPI symptoms been confirmed at speech perceptual evaluation, all patients underwent both acoustic and aerodynamic evaluations of the velopharyngeal function.

Perceptual assessment of velopharyngeal function

The patients were submitted to the perceptual speech assessment routinely performed at HRCA Laboratory of Physiology (Trindade and Trindade Junior, 1996). Velopharyngeal function was rated according to the classification system proposed by Trindade et al.(2005), which is based on scores attributed to nasality, nasal air emission and compensatory articulation.

Hypernasality was scored on a 6-point scale, with 1 = absent, 2 = mild, 3 = mild to moderate, 4 = moderate, 5 = moderate to severe, and 6 = severe. Scores of 2 or more were considered to be significant. Nasal air emission was scored according to the amount of air escape detected in the mirror test during blowing, blowing with a protruding tongue, prolonged emission of the /i/, /u/, /f/, /s/ and / / phonemes and of words and phrases with plosive and fricative phonemes, using a 6-point scale, with 1 = absent, 2 = mild, 3 = mild to moderate, 4 = moderate, 5 = moderate to severe, and 6 = severe. Scores of 3 or more were considered to be clinically significant. The presence of compensatory articulations was analyzed during the emission of plosive, fricative and affricative phonemes using a 2-point scale, with 1 = absent and 2 = present in one or more phonemes. Velopharyngeal function was classified as "adequate", "borderline", or "inadequate" on the basis of the set of scores attributed to the speech characteristics mentioned above (Table 1).

Data concerning the patients studied are listed in Table 2. All patients presented no hypernasality (score 1), nasal air emission either absent (score 2) or mild (score 2), absence of compensatory

articulations (score 1), and were, therefore, considered to be asymptomatic.

Acoustic assessment of velopharyngeal function: nasometry

Velopharyngeal function was estimated by measuring nasalance with an IBM Kay Elemetrics nasometer model 6200-2, software version 30-02-1.7. The system consists of two microphones positioned on each side of a sound separation plate placed above the upper lip of the patient and held in place with a helmet. The upper microphone senses the signals of the nasal component of speech and the lower microphone senses the signals of the oral components, which are filtered, digitalized by electronic modules and processed in a microcomputer using a specific software. Nasalance (expressed as %) corresponds to the relative amount of nasal acoustic energy in speech, i.e., to the numerical ratio between the amount of nasal acoustic energy and the total acoustic energy (sum of the nasal and oral acoustic energy) multiplied by 100.

Nasometry was performed during the reading of a text consisting of 5 sentences in Brazilian Portuguese containing exclusively oral sounds: Papai caiu da escada. Fábio pegou o gelo. O palhaço chutou a bola. Teresa fez pastel. A árvore dá frutos e flores (Daddy fell off the ladder. Fábio picked up the ice. The clown kicked the ball. Teresa made pastry. The tree gives fruits and flowers) (Trindade et al., 1997). For analysis, we considered the recording of the first technically acceptable utterance of the text, i.e., produced without errors and within the acceptable intensity limit of the instrument.

Aerodynamic assessment of velopharyngeal function: pressure-flow technique

Velopharyngeal function was also estimated by measuring velopharyngeal orifice area during speech with a PERCI-SARS computerized system (Microtronics Corporation). The technique is based on the principle that the cross-sectional area of a constriction (velopharyngeal orifice) can be estimated by simultaneous measurement of the differential pressure existing between the two sides of the constriction (oral pressure and nasal pressure) and the airflow that crosses through it (nasal flow). Velopharyngeal orifice area is calculated at the point where oral pressure reaches its maximum value during the production of a plosive consonant. The oral and nasal pressures are sensed by polyethylene catheters connected to differential pressure transducers. To record oral pressure, a catheter is positioned into the mouth and to record nasal pressure, a second catheter is inserted into a cork which is adjusted to the nostril of lower airflow, creating a static air column. Nasal airflow is sensed by a plastic tube adjusted to the other nostril. The tube is connected to a heated pneumotachograph attached to a differential pressure transducer. The signals of the three transducers are then processed by pre-amplifiers, fed into the microcomputer, and analyzed with a specific software.

The cross-sectional area of the velopharyngeal orifice was calculated during the repeated production of the /p/ phoneme inserted in the word "rampa", using the equation: $A = V / k [2 P/ d]1/2$, where A = velopharyngeal area, in cm2; V = nasal air flow, in ml/s; k = 0.65; P = difference between oral pressure and nasal pressure, in dyn/ cm2; d = 0.001 g/cm3 (air density). The mean values of 4 to 6 productions were used for analysis.

Data analysis

Nasalance scores were compared to normal scores established in a study carried out in our laboratory on subjects speaking Brazilian Portuguese (Trindade et al 1997). Theoretically, nasalance scores can range from 0%, which corresponds to absence of nasal sounds in the speech, to 100%, which corresponds to sound entirely emerging from the nose. A 27% score was considered as the upper normal limit for the text used. In other words, values higher than 27% were considered to indicate an excessive quantity of nasal acoustic energy in speech, suggesting hypernasality. That cut-off score corresponds to the value which maximized sensitivity and specificity of nasometry in identifying, respectively, the presence or absence of hypernasality in previous study carried out in our laboratory (Trindade et al., 2003).

In the aerodynamic assessment, velopharyngeal function was classified according to the criteria proposed by Warren (2004): areas of 0 to 4.9 mm2 = adequate closure, 5 to 9.9 mm2 = adequate to bordeline closure, 10 to 19.9 mm2 = borderline to inadequate closure, and 20 mm2 or larger = inadequate closure.

This study tested the hypothesis that, in aggreement with speech perceptual evaluation (indicative of adequate velopharyngeal function in all study patients), instrumental evaluation

(nasometry and pressure-flow assessment) would also suggest adequate velopharyngeal function, that is, nasalance values inferior or equal to 27% (suggesting no hypernasality) and velopharyngeal areas up to 4.9mm2 (indicating adequate velopharyngeal closure), in all patients.

Results

As previously mentioned, perceptual evaluation showed that all the individuals included in this study (Table 2) had adequate velopharyngeal function, which was characterized by no hypernasality (n=20), nasal air emission either absent (n=7) or mild (n=13), and absence of compensatory articulations (n=20), and were thus considered asymptomatic.

Concerning nasometric evaluation, Table 3 shows that the mean nasalance of the group of patients analyzed was 20±10%. Individual scores ranged from 7% to 46%. Normal nasalance scores, that is, nasalance less or equal to 27%, were observed in 15 of the 20 patients (75% of the cases). In the remaining 5 patients (15%), identified by an asterisk in Table 3, nasalance values were higher than 27%, suggesting hypernasality. Table 3 also shows that in 3 of these patients nasalance scores were very close to the normality cut-off score (nasalance of 28% in patients 4 and 9; and 30% in patient 11). However, in the other 2 patients, nasalance was way above the normality threshold (39% in patient 5, and 46% in patient 12). Since these 5 (15%) patients were diagnosed as symptomatic by nasometry and asymptomatic by perceptual evaluation, the null hypothesis was rejected in this case and no further statistical treatment of data was applicable.

Concerning aerodynamic assessment, Table 3 shows that the group exhibited a mean velopharyngeal area of 0.9±1.3mm2 . Individual values ranged from 0mm2 to 4.8mm2 . According to Warren (2004), these values are all indicative of adequate velopharyngeal closure. Thus, all patients were classified as asymptomatic both in the aerodynamic and in the perceptual evaluations

The outcomes of the three types of assessments performed, i.e., perceptual evaluation, nasometry and pressure-flow technique, are compared in Table 4.

TABLE 1. Classification system adopted in the assessment of velopharyngeal function according to scores attributed to hypernasality, nasal air emission and compensatory articulations (Trindade et al., 2005).

SPEECH CHARACTERISTICS			FUNCTION
Hypernasality *	Nasal Air Emission *	Compensatory Articulations #	VELOPHARYNGEAL
1	1	1	ADEQUATE
1	2	1	
1	3	1-2	BORDERLINE
2	1-3	1-2	
3	1-3	1-2	
2	4-6	1-2	INADEQUATE
3	4-6	1-2	
4-6	2-6	1-2	

* 1 absent; 2 mild ; 3 mild to moderate; 4 moderate; 5 moderate to severe; 6 severe.
\# 1 absent; 2 present

TABLE 2. Age, sex, and perceptual speech evaluation scores of the patients with submucous cleft palate analyzed in the study

PATIENT	SEX	AGE (years)	PERCEPTUAL SPEECH EVALUATION			
			COMPENSATORY ARTICULATION	NASAL AIR EMISSION	HYPERNASALITY	VELOPHARYNGEAL FUNCTION
1	M	6	1	2	1	Adequate
2	M	7	1	2	1	Adequate
3	M	7	1	2	1	Adequate
4	M	10	1	2	1	Adequate
5	M	10	1	1	1	Adequate
6	M	12	1	1	1	Adequate
7	M	16	1	1	1	Adequate
8	M	18	1	2	1	Adequate
9	M	22	1	1	1	Adequate
10	F	6	1	2	1	Adequate
11	F	7	1	2	1	Adequate
12	F	9	1	1	1	Adequate
13	F	10	1	2	1	Adequate
14	F	12	1	2	1	Adequate
15	F	12	1	1	1	Adequate
16	F	14	1	2	1	Adequate
17	F	18	1	2	1	Adequate
18	F	26	1	2	1	Adequate
19	F	32	1	1	1	Adequate
20	F	46	1	2	1	Adequate

1 absent; 2 mild

TABLE 3. Individual and mean nasalance scores (%), and velopharyngeal cross-sectional areas (mm^2) upon nasometric and aerodynamic evaluations, respectively.

PATIENT	NASALANCE (%)	VELOPHARYNGEAL AREA (mm^2)
1	8	0,1
2	8	0,4
3	18	1,5
4	28 *	0,7
5	39 *	0,8
6	24	0,4
7	20	0,2
8	22	0,2
9	28 *	4,8
10	21	2,9
11	30 *	3,0
12	46 *	0,1
13	17	0,1
14	16	0,1
15	7	0
16	11	0,9
17	15	0,6
18	25	0,3
19	9	0,1
20	16	0,2
Minimum	7	0
Maximum	46	4.8
Mean±standard deviation	20 ± 10	0.9 ± 1.3

* abnormal values suggestive of hypernasality (nasalance > 27%)

TABLE 4. Number (percentage) of patients with adequate and inadequate velopharyngeal function on perceptual and instrumental evaluation (nasometry and pressure-flow technique).

TYPE OF EVALUATION	ADEQUATE VELOPHARYNGEAL FUNCTION	INADEQUATE VELOPHARYNGEAL FUNCTION
Perceptual evaluation	20 (100%)	0 (0%)
Nasometry	15 (75%) *	5 (15%) *
Pressure-flow technique	20 (100%)	0 (0%)

* null hypothesis rejected (absence of total agreement between nasometry and perceptual evaluation)

Discussion

The question asked for the development of the present study was whether patients with SMCP might be unduly considered asymptomatic on the basis of a perceptual speech assessment and thus not receive appropriate treatment. In view of the results obtained for the 20 patients studied, we may infer that the answer was positive for five of the cases analyzed. Confirming our initial hypotheses, there was a complete agreement between perceptual judgment and aerodynamic measurements. In some cases, however, no agreement was observed between perceptual judgment and nasometric measurements, a finding that may be of clinical significance.

Among the 20 patients included in this study, as they were considered to be asymptomatic on perceptual evaluation, abnormal nasalance scores were observed in five patients, which corresponds to 15% of the total number of cases analyzed. Therefore, the hypothesis that all of them would present normal nasalance was rejected. This may mean that in those five patients there was faulty perceptual judgment or that nasometry possibly detected resonance deviations not audible by the human ear. As shown in Table 3, three patients presented nasalance scores (28%, 28% and 30%) quite close to the normal threshold of 27% established in our laboratory (Trindade et al., 2003). The possibility of measurements contamination by transpalatine acoustic transmission (Gildersleeve-Neumann and Dalston, 2001), especially in cases of SMCP, cannot be excluded. In the remaining patients, however, the scores observed were clearly abnormal, suggesting the presence of hypernasality. It is important to point out that the scores observed in these two patients (39% and 46%) were close to the mean observed in cleft palate patients with VPD (42±8%) in another study conducted at our laboratory (Zuiani et al., 1998). The fact that evidences of VPD were detected in five (or even 2) patients by nasometry, but not by perceptual assessment, may indicate the need for changing the approach in the treatment offered to these patients.

On the other hand, the velopharyngeal orifice areas were in all cases below the normal limit of 5 mm2 established by Warren (2004), confirming findings of the perceptual that velopharyngeal function was adequate in these patients . The coincidence between the aerodynamic and perceptual assessments seems to invalidate, per se, the results of nasometric assessment. However, two aspects support the observations made with nasometry. First, on aerodynamic assessment, velopharyngeal function is estimated by measuring orifice size during the production of a single consonant sound, whereas nasalance is measured during the production of a speech sample that contains different vowel and consonant sounds, and, therefore, closer to spontaneous speech. Second, the five patients with increased nasalance could be experiencing problems in the timing of velopharyngeal closure (Dotevall et al., 2002; Zajac and Hackett, 2002; Warren, 2004), causing hypernasality in the presence of adequate velopharyngeal closure, what could explain the discrepancies observed between the two methods.

The identification of evidences of hypernasality on nasometric testing in at least two of the patients with SMCP studied emphasizes the importance of the combined use of perceptual and instrumental methods and, furthermore, recommends the careful and regular follow-up of the cases in which instrumental evaluation reveals signs of dysfunction of the velopharyngeal mechanism in opposition to clinical impression. This is particularly true for children whose vocal tract structures and mechanisms are not yet fully developed, and who, therefore, are more prone to developing symptoms typical of VPD with aging. Ideally, these symptoms should be identified as early as possible so that more adequate therapeutic-surgical techniques might be used, leading to more favorable results and preventing the development of speech disorders. Since the use of nasometry and pressure-flow technique is limited to older children, it is necessary to develop instrumental methods that permit the evaluation of VPD symptoms as early as infancy, in a similar way to what was done in the present study.

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