

Intensive speech therapy in a patient with submucosal cleft palate - case report

Fonoterapia intensiva em paciente com
fissura de palato submucosa – relato de caso

Fonoterapia intensiva en um paciente con
fisura palatina submucosa - reporte de caso

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Abstract

Introduction: Submucosal cleft palate is a craniofacial malformation that can cause speech disorders and intensive speech therapy is an alternative treatment. **Objective:** report the case of a patient with submucous cleft who participated in an intensive speech therapy program, describe the therapeutic processes and compare speech production before and after therapy. **Methods:** the intensive therapy program was conducted with a 13-year-old female patient with submucosal cleft and consisted of three daily speech therapy sessions over a period of 4 weeks, resulting in 60 therapy sessions. Before the first session and after the last session, clinical and instrumental assessments of velopharyngeal function were performed. **Results:** systematization of velopharyngeal closure, improvement of speech intelligibility, elimination of compensatory articulations, weak intraoral pressure and hypernasal resonance in monitored speech. **Conclusion:** The intensive speech therapy program provided positive short-term results in speech rehabilitation of a patient with submucosal cleft.

Keywords: Speech-Language Pathology and Hearing Sciences; Speech therapy; Cleft palate; Speech; Speech disorders.

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Resumo

Introdução: A fissura de palato submucosa é uma malformação craniofacial, que pode ocasionar alterações de fala, e a fonoterapia intensiva é uma possibilidade de tratamento. **Objetivo:** relatar o caso de uma paciente com fissura submucosa de palato sintomática não operada que participou de um programa de fonoterapia intensiva para reabilitação de fala, descrever os processos terapêuticos e comparar a produção da fala antes e depois da terapia. **Metodologia:** o programa de fonoterapia intensiva foi realizado com uma paciente do gênero feminino de 13 anos de idade, com fissura submucosa de palato sintomática, não operada; e constituiu de três sessões diárias de terapia fonoaudiológica durante um período de 4 semanas, resultando em um total de 60 sessões de terapia. Antes da primeira sessão, e após a última, foi realizada avaliação clínica da fala e instrumental da função velofaríngea. **Resultados:** sistematização do fechamento velofaríngeo, melhora na inteligibilidade de fala, correção das articulações compensatórias, da fraca pressão intraoral e hipernasalidade em fala dirigida. **Conclusão:** o programa de fonoterapia intensiva proporcionou resultados positivos em período curto na reabilitação da fala da paciente.

Palavras-chave: Fonoaudiologia; Fonoterapia; Fissura palatina; Fala; Distúrbios da fala.

Resumen

Introducción: el fissura palatina submucosa es una malformación craneofacial, por esta razón puede causar trastornos del habla y la terapia intensiva del habla es un tratamiento alternativo. **Objetivo:** relatar el caso de una paciente con fisura palatina submucosa que participó en un programa intensivo de terapia fonoaudiológica para la rehabilitación del habla, describir los procesos terapéuticos y comparar la producción del habla antes y después de la terapia. **Metodos:** el programa de cuidados intensivos se llevó a cabo con una paciente del género femenino de 13 años de edad con fisura palatina submucosa y consistió en tres sesiones diarias de terapia fonoaudiológica durante un período de 4 semanas, lo que resultó en un total de 60 sesiones de terapia. Antes de la primera sesión y después de la última, se realizó una evaluación clínica e instrumental de la función velofaríngea. **Resultados:** sistematización del cierre velofaríngeo, mejor inteligibilidad del habla, eliminación de las articulaciones compensatorias, de la presión intraoral débil y de la resonancia hipernasal en el habla dirigida. **Conclusión:** el programa de cuidados intensivos proporcionó resultados positivos en un corto período de tiempo en la rehabilitación del habla de una paciente con fisura submucosa.

Palabras clave: Fonoaudiología; Logoterapia; Fisura del Paladar; Habla; Trastornos del habla.

Introduction

Submucous palate cleft (SMCP) is a congenital craniofacial malformation¹. The etiology is multifactorial, as it encompasses a combination of environmental, genetic and epigenetic factors^{2,3}. The diagnosis of SPF is often late, and the lack of early diagnosis of evident anatomical characteristics may be one of the reasons that justify this³. It is essential that speech therapists, doctors, dentists and other health professionals be made aware of the signs and symptoms of SMCP, in order to enable early diagnosis and monitoring of the condition⁴.

SMCP is subdivided into two types, classic and occult. The anatomical manifestations of the classic SMCP are the bifid uvula, muscle diastasis and bone notch in the posterior region of the hard

palate^{5,6}. In the occult SMCP there is diastasis of the muscles of the soft palate, however there are no anatomically visible signs of the presence of the cleft, which makes the diagnosis more difficult after clinical assessment⁶. Anatomical manifestations may appear concomitantly or isolated. The standard finding in classical SMCP is the presence of muscle diastasis in the soft palate^{1,3,4}. A relevant factor of SMCP is that it differs from other cleft palates due to the integrity of the oral mucosa of the palate¹.

According to the anatomical manifestations of SMCP, velopharyngeal closure may be altered⁷. Velopharyngeal closure occurs through the elevation and posteriorization of the soft palate, medialization of the lateral pharyngeal walls and anteriorization of the posterior pharyngeal wall⁸. When the velopharyngeal mechanism does not

adequately perform its function, there is a gap between the aforementioned structures, characterizing velopharyngeal dysfunction (VPD)^{7,8}.

VPD may be related to velopharyngeal insufficiency, i.e. when there is a change in the physical/anatomical structure, such as lack of tissue, absence of velar muscles, short palate, or increased nasopharyngeal space⁷. It can also be related to velopharyngeal incompetence, in this case, the physical/anatomical structure is sufficient to allow proper functioning; however, velopharyngeal closure does not occur due to neuro-sensory-motor determinants or even due to learning error of velopharyngeal functioning in production of speech during its acquisition⁷.

Studies have shown that the speech of a considerable number of patients with SMCP does not present characteristics of the presence of submucous cleft, that is they are asymptomatic¹; however, when there is a change in speech, hypernasality occurs which is usually associated with the escape of nasal air and weak intraoral pressure. In this case, they are called obligatory disorders related to SMCP due to the impossibility of performing velopharyngeal closure^{4,9}.

In addition to the obligatory disorders, SMCP favors compensatory articulations, which are characterized by the use of an atypical production site in the articulation of speech sounds⁷. In cases of SMCP with velopharyngeal insufficiency and speech disorders, surgical correction of the palatal structures or the use of a speech bulb might be recommended, and in both cases speech therapy will be required⁴.

Intensive speech therapy is an alternative before commencing conventional speech therapy this gives the therapist the advantage of being closer to the patient to understand his or her needs, and closely monitor the gradual modification of speech, provide rapid improvement and keep the patient motivated to continue the treatment¹⁰⁻¹². It is worth mentioning that the success of speech therapy is directly associated with the frequency of activities proposed during treatment^{10,12}.

Intensive therapy has shown successful results in other studies^{7,11,12}. It is a facilitator and with regu-

lar sessions, the aims are for quick improvements and greater patient satisfaction. The aim of this study is to analyze and describe the case of a patient with symptomatic submucous cleft palate who participated in an intensive speech therapy program for speech rehabilitation, describing the therapeutic processes and comparing speech production before and after the speech therapy treatment.

Case report

This study was approved by the Research Ethics Committee of the Ceilândia School of the University of Brasília (Protocol: CAAE 03333118.4.0000.8093). It is a speech rehabilitation case report of a patient with symptomatic classic submucous cleft who had not undergone any surgical treatment, and demonstrated the possibility of velopharyngeal closure by means of intensive speech therapy performed at the University Hospital of Brasília (HUB).

The patient in the case study was a 13-year-old female teenager with SMCP, who lives in Brasília - DF. After agreeing to participate in the therapy program, both the girl's guardian and the patient herself signed a consent form, in the case of the patient it was a minor's consent form. The guardian responsible also signed and consented to the use of imaging and audio for research purposes.

Patient history

The patient's prior history was retrieved from documentation sent by the service of origin, in addition to the interview carried out in the initial consultation at the outpatient's clinic of HUB by an experienced speech therapist responsible for the case.

The patient had a history of nasal reflux as a baby, delay of speech and language development and underwent speech therapy from 3 to 8 years of age. Because of her speech delay she was referred to the Regional Hospital of Asa Norte (HRAN) when she was 8 years old and was diagnosed with submucous cleft palate (Figure 1) by the medical team of the cleft lip and palate clinic.



Figure 1. Oral cavity showing the hypoplastic uvula and muscle diastasis.

Initially there was a recommendation for primary palatoplasty to be carried out, but it was not possible due to preoperative complications. After this, she had sporadic speech-therapy sessions from 8 to 13 years of age. According to the legal guardian, she was not particularly meticulous about her appointments and did not adhere to the proposed treatment. When she was 13, she was referred to the Speech Therapy Service at HUB to take part in

the intensive speech therapy program according to recommendations by the service of origin.

Intensive speech program therapy proposal

Initially, the patient was evaluated by the speech therapist responsible for the care of patients with cleft lip and palate at HUB, to see if there was any recommendation for her participation in the intensive speech therapy program. In other words, checks were carried out to ascertain whether the patient had functional velopharyngeal closure which would make her eligible to participate in the program.

The evaluation took place in one session before the start of the actual intensive speech therapy program and a one session reevaluation at the end of the program.

To evaluate velopharyngeal function at the beginning and at the end of the program, a nasofibroscope was performed with a flexible nasofibroscope made by Machida®, model ENT-30PIII.

The examination was conducted by an otorhinolaryngologist at the hospital and accompanied by the speech therapist responsible for directing speech assessment during the examination. In the therapeutic test, the velopharynx (Figure 2) was observed at rest (Figure 2A) and during the emission of the syllable /pa/ (Figure 2B). During the emission, the possibility of velopharyngeal closure in

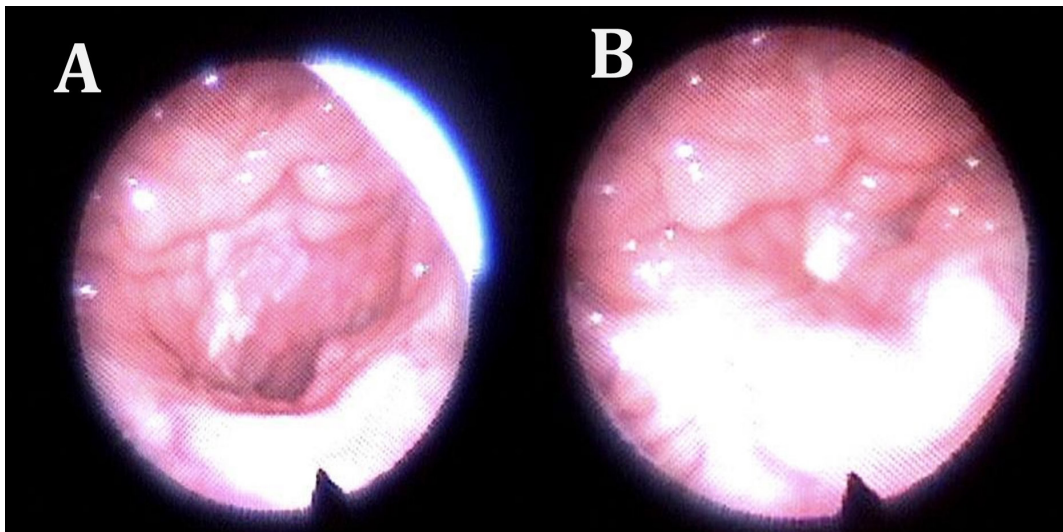


Figure 2. Top view of the velopharynx during nasofibrolaryngoscopic evaluation before the intensive speech therapy program. (A) Velopharynx at rest. (B) Velopharynx during the emission of the syllable /pa/.

speech was found, which suggested velopharyngeal dysfunction related to learning errors.

In the clinical evaluation, a specific evaluation protocol was used in the speech therapy outpatients' clinic of the hospital. Directed speech was filmed and recorded which included counting from one to twenty, repetition of sentences and spontaneous speech¹³ for later evaluation and comparison. The video was recorded on a Sony Cybershot DSC-W120 camera and the audio was recorded on a HP Compac Pro 6305 desktop computer with

a Samson USB Go Mic portable microphone and Audacity 2.4.2. During the intraoral clinical inspection, grooved and hypoplastic uvula, muscle diastasis and intact mucosa were observed. In speech, the calculation of the percentage index of correct consonants (PCC)¹⁴ of fragments of spontaneous speech, connected speech (counting from one to twenty) and standardized phrases were also performed. The data from the initial assessment are shown in Table 1.

Table 1. Results of the evaluation before and after the intensive speech therapy program.

Clinical Evaluation	Pre-speech therapy	Post-speech therapy
Resonance	Mild hypernasal resonance - nonacceptable	Balanced
Hypernasality Test	7 of 10 words and vowel /i/	0 of 10 words and 0 vowels
Nasal Air Emission Test	Nasal Air Emission present in: /b/, /f/, /s/, /ʃ/, /l/ and vowel /i/	Nasal Air Emission absent
Intraoral Pressure	Weak intraoral pressure when emitting the phoneme /s/	Adequate intraoral pressure
Compensatory articulations	Glottal stop in the phonemes / p / and / k /, and unsystematic pharyngeal stop in the phoneme / t /	Absence of compensatory articulations
Phoneme substitutions	/tʃ/ by /ʃ/, /dʒ/ by /ʒ/ and /l/ by /r/	No substitution
Lisping	phonemes /z/ e /s/	Absence of lisping
Distortions	/l/ - isolated and in consonant blends	Absence of distortions
Facial Mime	Present	Present
Speech intelligibility	Moderately intense alteration	Mild alteration
PCC	Numbers from 1 to 20: 64% Spontaneous conversation: 40% Phrases: 68%	Numbers from 1 to 20: 94% Spontaneous conversation: 91,5% Phrases 93%

PCC: percentage of correct consonants.

After evaluation the patient was recommended to participate in the program which consisted of 60 sessions of speech therapy, carried out consecutively over four weeks between the months of January and February 2019. There were three daily sessions with a minimum duration of 30 minutes and a maximum of 45 minutes, with intervals of an hour and a half between sessions. The patient was instructed to practice at home every day, and the person responsible for her was instructed to assist her. The advice was to practice at home twice for 15 minutes from Monday to Friday, totaling 30 minutes, and four times for 15 minutes on Saturdays and Sundays totaling one hour of practice. The number of therapeutic sessions was determined by an intensive speech therapy study⁷ and an increase in the number of sessions was proposed in order to achieve correct production of speech sounds and

automation of the speech pattern acquired during the intensive speech therapy program. The main objectives of intensive speech therapy were to increase intraoral pressure, correct speech nasality and reduce compensatory articulations^{4,12}.

At the beginning of the program, the phoneme to be worked on was selected. This is called the target sound and installed correctly without producing compensatory articulation, with the air directed to the oral cavity and without any nasal air emission. Then the phoneme was associated with seven oral vowels, followed by syllables, words without meanings, words and phrases. After the target sound was determined, it became a vehicle for the generalization of velopharyngeal closure for the other sounds, which were worked according to a speech hierarchy. After determining all the target sounds, without compensatory articulation and with

the air directed to the oral cavity and without any nasal air emission, practice was carried out with texts containing oral and nasal phonemes, followed by directed speech. For automation, spontaneous conversations were made with self-monitoring by the patient^{7,8}. The stages of the intensive speech therapy program took place in line with the patient's progress.

To promote self-monitoring of the speech pattern, cues were used, such as: Scape-Scope, Glatzel mirror, cup with Styrofoam balls, fringed paper, air paddle and auditory biofeedback^{7,10,12}. The guardian and the patient were instructed to use the terms "new speech" (speech without compensatory articulations) and "old speech" (speech with compensatory articulations)⁸ as a strategy to contrast the speech patterns used¹². To assist in self-monitoring and automation of "new speech" a visual communication signal was established to be used by the mother to self-correct the emission of "old speech". In the post-speech therapy evaluation, the patient was performing self-monitoring of speech.

The result of the nasal air emission test after the intensive speech therapy program revealed that velopharyngeal closure was systematized during the production of oral sounds and confirmed that the VPD of the patient in the case studied was due to a learning error.

Despite the fact that in some sessions the patient was inattentive and hyperactive, at the end of the 60 sessions, she was able to fix all phonemes, eliminate compensatory articulation, and adjust intraoral pressure and nasal air emission (NAE) (Table 1). However, it was necessary to continue with conventional weekly speech therapy to automate the new standard in spontaneous speech.

Discussion

The late diagnosis of SMCP described in this case report and in prior studies⁴ highlights the importance of making academics and health care professionals aware of the signs of SMCP. Early diagnosis is important so that patients and their families are informed about the signs, symptoms of VPD and the need for treatment^{1,4}.

During the study we noticed that the legal guardian did not understand the anatomical alterations and what the patient's therapeutic needs were. It was necessary, therefore, to provide information

about the alterations and rehabilitation needs, in addition to reinforcing the importance of the family's support. Due to the poor knowledge level and emotional factors of the legal guardian, simple and repeated explanations were used throughout the therapeutic process¹⁵. We emphasize that the support of family members during the intensive speech therapy program is essential for the acquisition of the new speech pattern¹⁰.

The patient in this study had VPD which resulted in weak intraoral pressure, hypernasality and compensatory articulations (Table 1). Despite the anatomical alteration, she presented with the possibility of achieving velopharyngeal closure for speech without the necessity of surgical correction of the cleft (Figure 1), in contrast to cases in which the malposition of the palate muscles can result in velopharyngeal insufficiency³.

At the end of speech therapy program, the systematization of velopharyngeal closure was verified, confirmed by the evaluation of NAE after the intensive speech therapy program (Table 1). This case was recommended for isolated speech therapy in the treatment of VPD due to learning errors⁷. The use of facilitating cues contributed to the production of sounds in an appropriate way, with the absence of NAE and compensatory articulations. Through these clues, the patient achieved better self-monitoring, as described in other studies^{8,10,12}.

The patient and the therapist met daily for the therapy sessions, allowing effective direction of the treatment and monitoring of the patient's progress^{11,12}. This was evident in the case studied, since at the end of the first week of speech therapy the patient and the guardian noticed changes in the speech of the patient, which consequently made them more motivated and committed to the program. The percentage of correct consonants (PCC)¹⁴ before and after intensive speech therapy program (Table 1) confirmed the satisfactory results of intensive speech therapy.

During the study, the patient was hyperactive and had problems paying attention which may have consequently interfered with her therapeutic performance. A study showed that the discrimination of auditory stimuli in people with ADHD is compromised as well as auditory processing¹⁶. This fact may have contributed to the patient's difficulty in understanding some of the target sounds and automation of the new speech pattern in spontaneous conversation. Despite these changes, it was not pos-

sible to say how detrimental it was to the patient's progress because her results were satisfactory.

At the end of the intensive speech therapy program the patient showed evident improvements in her speech (Table 1), but did not manage to automate this pattern in spontaneous conversation, corroborating the study by Lima¹¹ that addressed this fact as a challenge in intensive speech therapy. In this case, it was found that the family's lack of encouragement and help correcting the patient's speech were detrimental to the automation process. Patients in another study reported that family members did not correct them, because they were already able to understand them¹¹. Because of the lack of automation of the new speech pattern during the intensive speech therapy program, it was recommended that the patient should continue with conventional speech therapy after the program had finished.

The elimination of compensatory articulation and the systematization of velopharyngeal closure in a short period of time may not have been achievable in many years of conventional speech therapy treatment due to the infrequency of the sessions. In conventional speech therapy, in addition to the sessions taking place infrequently, the patient did not achieve good adherence, which resulted in poor attendance of the therapeutic sessions, and even dropouts.

Instrumental assessment of velopharyngeal function in conjunction with the clinical assessment of speech is extremely important. The patient's positive results after the intensive speech therapy program showed good progress, and systematization of velopharyngeal closure was verified. After she returned for reassessment at the institution of origin, it was considered that the patient no longer needed surgical treatment. The need for longitudinal monitoring is highlighted to ensure the automation of the new speech pattern.

The report found that intensive speech therapy resulted in quick and satisfactory results for the patient; however, there are few studies of this type which explore the stages of its application. In addition, the case studied referred to a submucous cleft palate, and there are also very few studies that refer to this type of cleft. A peculiarity of this case report is that the patient had VPD due to learning errors and most cases of submucous cleft that present with speech disorders related to velopharyngeal insufficiency³.

Conclusion

In this case study the patient's anatomical structure allowed for velopharyngeal closure, even with the alterations of the submucous cleft palate, which made intensive speech therapy an option. The therapeutic process described, and the strategies used, provided therapeutic success when comparing speech production before and after the program, leaving only a few aspects to be worked on to finalize the therapeutic process in conventional speech therapy.

We conclude that intensive speech therapy provided a fast and effective solution for the patient of this case study.

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