

Case reports

The therapeutic effects of a modified palatal memory plate on oral motor function in children with Trisomy 21 at age 3 and 4 years: Case Reports

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ABSTRACT

People with Trisomy 21 (T21) have generalized hypotonia, also manifesting in oral structures. The palatal memory plate (PMP) is a removable appliance to improve tongue and lip posture. The evidence of research elucidates an improvement in oral motor function in children with T21 up to the age of 2 who received PMP-based therapy, with only limited, scanty reports in the literature concerning older children. Therefore, the aim of this study is to evaluate the effects of modified PMP on an oral motor function in patients over 2 years old. Two patients with T21, aged 4 and 3, with absence of lip seal and tongue malposition, were subjected to PMP-based therapy for 6 months. The patients were evaluated for an extra and intra oral exam; their parents answered a questionnaire about their children's habitual tongue and lip position. Two-minute footage to record orofacial motor functions was performed monthly, during follow-up appointments to assess the changes. The substantial improvement of tongue and lip posture was observed after 6 months of plate regular use. PMP has proved to be clinically effective in children over 2 years old, presented with T21, leading to improvement of lip and tongue posture, observed by parents and professionals.

Keywords: Down Syndrome; Muscle Hypotonia; Orthotic Devices; Myofunctional Therapy; Facial Muscles



INTRODUCTION

Trisomy 21 (T21), is the most common chromosomal genetic alteration in newborns, caused by the complete trisomy of chromosome 21¹. People with T21 have characteristics, features, well-described phenotype, usually presenting with short stature, obesity, epicanthus, oblique palpebral fissure, shortening of the extremities, and a single palmar crease². Regarding the face and craniofacial development, they present underdevelopment of the bones in the middle third of the face, brachycephalic profile, maxillary atresia, absence of lip seal, tongue interposition, posterior crossbite, tendency to mandibular prognathism, and oral structures hypotonia². The tongue appears abnormally large with a more anterior and low posture, characterized as relative macroglossia². These factors generate oromotor problems, observed during swallowing, chewing, speaking, and breathing².

In the search for early intervention in the 70's, the Argentine physician Castillo-Morales developed the "Orofacial Regulation Therapy (ORT)". An interdisciplinary therapeutic approach that consists of performing exercises to improve muscle tone associated with the use of palatal memory plate (PMP)^{3,4}. The PMP, a self-curing acrylic device from a model of the upper arch, aims to reduce tongue dysfunctions, improve the function of the orbicular muscle of the mouth and mimetic muscles, leading to improvements in sucking, articulation, swallowing, and nasal breathing^{5,6}.

In the rehabilitation process of T21 using a PMP, speech therapists play a crucial role. Speech therapy employs Orofacial Regulation Therapy (ORT) as one of its therapeutic strategies and oversees the use of the PMP⁷. Additionally, speech therapists provide specific exercises to strengthen the muscles involved in the functions of the stomatognathic system, including respiration, suction, swallowing, and speech processes⁸. Consequently, patients with T21 using the PMP can experience an improvement in readaptation of strength, tonus, sensitivity, and mobility of the speech organs following these interventions.

Studies indicate a significant improvement in oral motor functions in children with T21 up to 2 years of age who undergo PMP therapy, with few reports in the literature in children with T21 over 2 years of age⁹. However, the presence of children with T21 over 2 years of age who require an orofacial stimulation intervention is observed, and thus, the use of modified PMPs with a palatal expander may be considered as a primary clinical modality. Therefore, the aim of this

study is to evaluate the effect on oral motor function of the modified PMP in patients over 2 years of age diagnosed with Down Syndrome.

CASE REPORT

The present study is a clinical case, associated with the research project approved by the Ethics Committee of the Federal Fluminense University, Brazil, reference number: 5.603.099 and CAAE: 60151022.0.0000.5626. The study has been conducted in full accordance with ethical principles (Declaration of Helsinki). The informed consent was obtained from all subjects involved in the study.

The writing of these clinical case reports follows the guidelines recommended by The CARE Guidelines: Consensus-based Clinical Case Reporting Guideline Development¹⁰.

For the study, two male children diagnosed with Trisomy 21, aged 3 and 4 years, were selected. The children were selected because they had undergone treatment with PMP for six months. Besides, they had never used PMP or any other device, and they underwent speech therapy. Both presented with lingual hypotonia and absence of lip sealing. Prior to treatment and after 6 months, the children were evaluated by an orthodontist and a specialist in special care dentistry. In addition to this, parents answered a questionnaire with questions related to the child's acceptance of the plate and their perception of the child's habitual tongue and lip position. Regarding the habitual tongue position, parents were asked to indicate in which position the child's tongue was most of the time: 'inside the mouth,' 'over the lower lip,' and 'outside the mouth (severe protrusion).' As for the habitual lip position, parents were asked to indicate whether the lips were most often: 'open,' 'semi-open,' or 'closed.' The criteria for lip position considered 'closed' as total lip sealing, 'semi-open' as the absence of sealing but with lip touch at the corners of the mouth, and 'open' as the total absence of lip touch. Video recordings lasting 2 minutes and photographs for recording orofacial motor functions were performed monthly during follow-up appointments to assess changes in the habitual tongue and lip positions. Recordings of the child without the plate were made, prioritizing moments when the view of the mouth area was clear. It was asked the child to sit on the floor or in the lap of one of the caregivers without interaction. During the video evaluation, the time when the lip and tongue positions followed the classification in the parents' questionnaire was observed. The clinical

progress of the treatment was obtained by comparing the videos recorded at the first appointment and after 6 months of treatment. The patients underwent speech therapy only once a week; however, this modality was not specific to PMP. The cases reported did not undergo speech therapy in the university due to the distance between their place of residence and the location of therapy. However, both received speech therapy guidance during adaptation consultations with the dentists and the speech therapist of the university⁸.

RESULTS

Case 1

A 4-year-old male patient diagnosed with T21 was referred to the Orthopedics and Orthodontics in Patients with Special Needs Service at the Health Institute of Nova Friburgo, Fluminense Federal University with a complaint of crooked teeth. During the anamnesis, the mother reported that the patient had intellectual disability, hearing loss, recurrent throat infections, and speech difficulty. He also had a medical and speech therapy follow-up and denied other comorbidities. The mother also reported that the patient had difficulty chewing, awake bruxism, drooling, digital sucking, snoring, and apnea during sleep. Extraoral and intraoral examination revealed proportional facial thirds, convex profile, facial pattern I, a normal position of the lower lip, and mesofacial. Lack of passive lip seal; hypotonicity in the lips, masseter, and chin; tongue in a low position and projected outward passing the lower

lip were also observed. In addition, the patient was a mouth breather. Besides, the patient had complete deciduous dentition, bilateral Class I, and anterior open bite. Given this situation, it was used the modified PMP with a palatal expander, grooves in the lip stimulation, and an oval button behind the expander for lingual stimulation (Look at the figure 1). Prior to the start of treatment, the mother answered a questionnaire with questions related to her perception of the child's tongue position and lip seal. Regarding the position of the tongue, the mother responded "inside the mouth", and regarding the lips, the mother responded "open". At the same time, a photo and video recording of the child's face were taken. Table 1 shows the duration of the participant's habitual lip and tongue positions before and after 6 months of treatment. At the beginning of the treatment, the patient aged 4 years had his mouth molded and the plate was installed. The mother was instructed that the patient should use the PMP four times a day for 30 min at 2-h intervals and monthly return for follow-up. In the sixth month of treatment, the guardian answered the follow-up questionnaire for PMP therapy. When asked about the tongue position, the mother responded, "inside the mouth", and regarding the lip position, the mother responded "semi-open", indicating that the child adopted a posture where the lips remained less open (Figure 2. A–B). The mother reported satisfaction with the treatment. A quarter of activation of the palatal expander was performed monthly to monitor jaw growth.

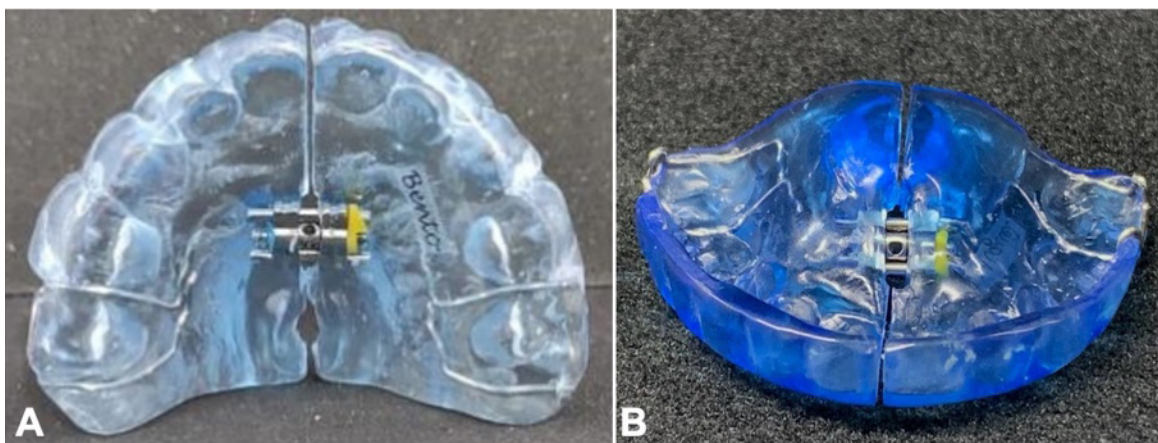


Figure 1. Modified Palatal Memory Plate with expander (Case 1 and 2): A. Occlusal view showing the suction button, palatal expander, occlusal covering of all teeth and buccal undulations for proprioceptive stimulation of the lip. B. View of the anterior region showing the grooves in the buccal region.

Table 1. The impact in seconds observed in the recordings on the position of the patients' lips and tongues, after a six-month-treatment

Evaluation Moment	Cases	Lips Posture (time in seconds)			Tongue Posture (time in seconds)		
		Open	Semi open	Closed	Inside the mouth	On the lower lip	Outside the mouth
Before Treatment	1	44	15	0	47	12	0
	2	56	17	0	62	12	0
After 6 months	1	12	38	0	67	7	0
	2	28	48	0	78	4	0

**Figure 2.** Clinical Aspects (Case 1): A. Before treatment, an open lip posture and the tongue inside the oral cavity, was observed. B. After 6 months of treatment, a reduction in mouth opening with lip contact at the corners of the mouth and the tongue inside the oral cavity, was observed.

Case 2

A 3-year-old male patient diagnosed with T21 was referred to the Orthopedics and Orthodontics in Patients with Special Needs Service at the Health Institute of Nova Friburgo, Fluminense Federal University with the complaint of lack of lip seal. In the anamnesis, the mother reported mild leukopenia and lymphocytopenia, hyperopia, and astigmatism, and claimed that the patient has the habit of biting his fingers. The patient is being monitored by an occupational therapist, speech therapist, and physical therapist. Extraoral and intraoral examinations revealed an enlarged lower third, convex profile, facial pattern I, and a tendency toward

vertical growth. The patient does not perform passive lip sealing, keeping the tongue in a low protruding position resting on top of the lip. Additionally, it was observed swallowing, drooling, hypotonicity of the lips, masseter, and chin as well as mixed breathing. The patient has bilateral deciduous Class I dentition and functional anterior crossbite due to a low tongue posture and absence of lip seal. Given this situation, it was used the modified Palatal Memory Plate with a palatal expander with grooves for lip stimulation and an oval button behind the vice for tongue stimulation, as used in case 1. Before starting the treatment, the mother responded to the questionnaire regarding the perception of lip and tongue positions. When asked

about the lip position, she selected the option “open”, and regarding the tongue position, she related “inside the mouth”. Video recordings and photos were also taken to document the lip and tongue positions. Table 1 shows the duration of the participant’s habitual lip and tongue positions before and after 6 months of treatment. The mother was instructed that the patient should use the PMP four times a day for 30 minutes at 2-hour intervals and was also advised to return

for monthly follow-ups. After 6 months of follow-up, when responding to the perception questionnaire, the mother reported improvement in the tongue position, selecting the option “inside the mouth”, and regarding the lip position, she observed improvement and chose the option “semi-open” (Figure 3. A–B). The mother is satisfied with the results, and the patient is being followed-up.



Figure 3. Clinical Aspects (Case 2): A. Before treatment, an open lip posture and the tongue inside the oral cavity, was observed. B. After 6 months of treatment, an improvement in lip sealing and tongue position, can be observed.

DISCUSSION

In T21, while generalized muscle hypotonia is present in 100% of the cases, early diagnosis, and treatment of lip and tongue hypotonia is considered as essential^{5-7,9}. The impairment of oral motor skills due to the absence of lip sealing and improper tongue movements hinders the development of basic functions such as chewing and swallowing¹¹. The PMP has been reported to be a highly effective device for the treatment of these issues; however, there are few reports in older children. This study aimed to report two cases in older children aged 3 and 4. The documentation of the treatment progress through audiovisual records has been reported in previous studies, proving a solid method of analysis. Similarly, questionnaires for recording parental perception have been utilized accordingly¹².

In this report, it was observed that the 4-year-old participant showed improvement in habitual lip posture,

adopting a reduction in mouth opening and tongue protrusion movements after 6 months of follow-up. Castillo-Morales recommends the early use of the PMP, starting from 2 months of age, as it is the period of greatest development of the central nervous system and the phase of greater acceptance and tolerability of the device¹³. However, a clinical study with a 3-year-old child showed positive results in 4 months of treatment, indicating that it is possible to achieve beneficial outcomes in older children¹⁴.

The second participant started treatment at 3 years old, and it was observed significant improvement in habitual lip posture, with a considerable reduction in mouth opening. These observations may be related to the fact that the caregiver used the PMP for a longer daily period than recommended, and the child did not exhibit severe hypotonia.

The application of the questionnaire to the parents was carried out with the aim of documenting the use of the PMP during the treatment period, recording

their perceptions of the case's evolution, especially since this therapy's success is directly related with family involvement and collaboration. In addition to the improvement in lip and tongue positions, the caregivers also noted an increase in sucking movements and improvement in phonation^{15,16}.

The PMP, proposed by *Castillo de Morales*, employed as integrated part of with orofacial therapy, is indicated to positively contribute to the development and neuromotor rehabilitation of patients, as an adjuvant mean^{5,7,9}. Although research commonly addresses patients at an earlier age, the results obtained in this report are consistent with previous reports, since in both described patients a decrease in tongue protrusion, and elevation on the palate, reduced lip opening, and improvements in phonation and chewing were observed^{6,8,17-19}.

Furthermore, studies have revealed that PMP therapy vastly improve the function of the orbicular muscle of the mouth and mimetic muscles, leading to enhancement of sucking, articulation, swallowing and nasal breathing^{6,17,19}. The modification of the standard traditional PMP proved to be beneficial when used in this group of patients, appearing to be effective in older children. It is common to find articles describing the effectiveness of PMP therapy if it is implemented as early as possible, preferably in the first year of life^{8,17,19}.

Consequently, the search for research on the proposed treatment consistent with the aim of study was difficult because of age-related constraint associated with available data. Seemingly the desired, optimal effect of PMP therapy is only achieved as part of multidisciplinary team, including physical therapy, speech therapy, and/or Orofacial Regulation Therapy, and that treatment outcome depends firmly on the level of cooperation and involvement of the caregivers^{6,7,9,15}.

In the described cases, patients were undergoing speech therapy; however, the speech therapist did not know the PMP. It is known that the benefits of PMP treatment can be significantly reduced if orofacial regulation therapy is absent, which composes a limitation of this study^{8,20}.

Another limitation could be attributed to the 2-minute video recordings, which, due to their short duration, may not capture the actual posture that children adopt for most of the day. Additionally, children were in a various, different environment during the recording, which may influence their behaviors. Thus, perceiving the different characteristics of the patients and, in the context of the family members, understanding that for the success of

the treatment it is indispensable that several nuances are in tune, the comparison between cases of this type of therapy becomes more complex. In fact, it is worth noting, that the two patients reported were able to wear the plate initially without the use of a fixator, which may be associated with the good adaptation to the palate and the motivation of caregivers in general. Further, observational, robust prospective research is required to validate the clinical usefulness of PMPs in children with complex special needs, particularly those presented with disabilities.

CONCLUSION

The palatal memory plate appliance has been shown to be effective in children presented with T21 at older ages, generating substantial improvement in lip and tongue posture. The clinical outcome depends on patient-centered, highly individualized management, with an unique plate design adequate to the patient's age and clinical needs. In addition, other factors contribute to therapy success such as regular monthly follow-ups, caregivers' primary motivation, and involvement of other professionals in the therapy process, such as speech therapists.

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Authors' contributions:

NFRN: Conceptualization; Methodology; Investigation; Resources; Data curation; Validation; Writing - Original draft; Writing - Review & editing.

LAPL: Investigation; Resources; Data curation; Writing - Original draft; Writing - Review & editing.

AAS: Data curation; Investigation; Resources; Writing - Original draft; Writing - Review & editing.

PL, FW: Software; Writing - Review & editing.

AD: Visualization; Writing - Review & editing.

DBR: Conceptualization; Data Curation; Methodology; Investigation; Resources; Supervision; Writing - Original draft; Writing - Review & editing.

MJSM: Formal Analysis; Funding Acquisition; Project Administration; Supervision; Validation; Writing - Review & editing.

BLSP: Conceptualization; Data Curation; Formal Analysis; Methodology; Funding Acquisition; Investigation; Resources; Project Administration; Visualization; Supervision; Validation; Writing - Original draft; Writing - Review & editing.

Data Sharing Statements:

All of the individual deidentified participant data can be shared, as informed consent form and clinical study case report, immediately following publication, anyone who wishes to access the data indefinitely. Proposals should be directed to brunapicciani@gmail.com