

## Review articles

# Effects of using the stimulating palatal plate in combination with orofacial stimulation on the habitual tongue and lip posture in children with trisomy 21: an integrative literature review

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**ABSTRACT**

**Purpose:** to verify, in the literature, the effects of using the stimulating palatal plate on lip and tongue posture in children with trisomy 21.

**Methods:** a search was conducted in Medline, LILACS, CINAHL, EMBASE, Scopus, Web of Science, and Cochrane. Original articles designed as clinical trials, longitudinal studies, or case-control studies, approaching stimulating palatal plate in the treatment of children with trisomy 21 and assessing habitual lip and tongue posture as an outcome, were included.

**Literature Review:** a total of 376 studies were found, of which 10 met the selection criteria. They were published between 1996 and 2007 and carried out mostly in Europe, with small samples. The age when they began wearing the plate ranged from 1 month to 5 years, and intervention lasted from 4 to 58 months; in most cases, it was combined with orofacial muscle stimulation. Use frequency ranged from two to four times a day, each period lasting from 30 minutes to 2 hours. The children's tongue and lip posture improved in most pieces of research.

**Conclusion:** studies suggest that using the stimulating palatal plate in combination with orofacial muscle stimulation brings benefits to tongue and lip posture in children presented with trisomy 21.

**Keywords:** Down Syndrome; Rehabilitation; Stomatognathic System; Speech, Language and Hearing Sciences; Dentistry

## INTRODUCTION

Trisomy 21 (T21), also known as Down syndrome, is a genetic disorder characterized by the presence of three copies of chromosome 21 in the cells<sup>1</sup>. In Brazil, its estimated incidence is one in every 600 births<sup>1</sup>. It occurs independently of geographic region, sex, or race<sup>2</sup>. The typical phenotype of people with this syndrome includes low stature, microcephalia, upslanting palpebral fissures, small low-set ears, small hands, and so forth<sup>1</sup>. T21 is usually associated with muscle hypotonia, immune impairment, hypothyroidism, congenital cardiopathies, and intellectual disability<sup>3</sup>.

Specifically, craniofacial morphology and oral cavity characteristics include orofacial muscle hypotonia, lack of lip closure, usually low and forward tongue posture, retruded maxilla with a deep atretic palate, and a predominance of skeletal Class III malocclusion<sup>1,3-5</sup>.

The literature points to positive speech-language-hearing therapy results in patients with T21, with improvements in articulatory organs<sup>6,7</sup>. Orofacial Regulation Therapy, proposed by Castillo-Morales, is one of the various therapeutic approaches speech-language-hearing therapists can use with this population<sup>8</sup>. This approach is based on a neuromuscular stimulation program, which can be combined with the use of an oral orthopedic device called stimulating palatal plate (SPP)<sup>8-10</sup> – which is made by the dentist based on the patient's upper arch model. SPPs have a stimulating button for the tongue and vestibular knobs for the lips<sup>8</sup>. It has been indicated in cases of a hypotonic tongue, habitually positioned between the teeth or lips, and a hypotonic upper lip without closure<sup>8</sup>. This approach requires coordinated work between a dentist and a speech-language-hearing therapist<sup>11</sup>.

Various authors report the benefits of SPP treatment in combination with orofacial stimulation for children with T21. They mainly include improvements in habitual tongue and lip posture<sup>12-18</sup>, although improved facial muscle tone<sup>15,19</sup>, decreased sialorrhea<sup>19-21</sup>, eliminated deleterious oral habits<sup>21</sup>, and improved performance of stomatognathic functions such as breathing<sup>15</sup>, sucking<sup>20,22</sup>, masticating, swallowing<sup>22</sup>, and speech articulation<sup>15,19</sup> are also reported.

However, some questions on this therapeutic approach remain unclear, especially regarding benefits to tongue and lip posture. Likewise, ideal plate use frequency, intervention duration, age at treatment initiation, and combination with other therapeutic approaches need further clarification. A more detailed

survey of information in the literature is important to guide professionals who work with this type of intervention.

Hence, the aim of this study was to verify, in the literature, the effects of SPP on tongue and lip posture in children presented with T21. A secondary goal was to analyze the age at intervention initiation, intervention duration, plate use frequency, assessment methods used in the studies, and the use of other combined interventions.

## METHODS

An integrative review of the literature was conducted in the following stages: defining the research question, establishing keywords and inclusion/exclusion criteria for the articles, selecting the articles, and critically evaluating them.

The research question was defined as follows: “Does SPP use improve habitual tongue and lip posture in children with T21?”. The question was defined based on the PICO strategy, in which P (participants) refers to children with T21; I (intervention) refers to SPP; C (comparison) refers to the group not submitted to plate intervention; and O (outcome) refers to tongue and lip posture.

The national and international literature was surveyed to select the articles, with no restriction of language or year of publication. The search was conducted in Medline/PubMed, CINAHL, Scopus, EMBASE, Web of Science, LILACS, and Cochrane, as well as in bibliographical references in the selected articles.

The following terms were used: “*Down Syndrome* (DeCS/Mesh/Emtree)”, “*trisomy 21* (Emtree)”, “*mongolism* (free term)”, “*meiotic nondisjunction*” (free term), “*mitotic nondisjunction*” (free term), “*partial trisomy 21* (Emtree)”, in combination with “*orthotic devices* (DeCS/Mesh)”, “*palatal plate*” (free term), “*orofacial regulation therapy*” (free term), “*Castillo-Morales*” (free term), “*stimulating plate*” (free term), “*functional appliances*” (free term), “*palatal plates*” (free term), “*modified palatal plate*” (free term), “*orofacial stimulation*” (free term), “*Castillo-Morales stimulating plate*” (free term), “*Castillo-Morales plate*” (free term), “*palatal plate therapy*” (free term), “*Castillo-Morales concept*” (free term), and “*stimulating palatal plate*” (free term). The free terms were obtained from descriptors in articles on the topic, retrieved in a pilot search.

Chart 1 shows the search strategies used in each portal or database.

**Chart 1.** Search strategies used in each database

| Source           | Strategy  |
|------------------|---|
| PubMed           | ("Down Syndrome"[MeSH Terms] OR "trisomy 21" OR mongolism OR "meiotic nondisjunction" OR "mitotic nondisjunction" OR "partial trisomy 21") AND ("orthotic devices"[MeSH Terms] OR "palatal plate" OR "Orofacial regulation therapy" OR "Castillo-Morales" OR "stimulating plate" OR "Functional appliances" OR "Palatal plates" OR "Modified Palatal Plate" OR "Orofacial Stimulation" OR "Castillo-Morales stimulating plate" OR "Castillo-Morales plate" OR "Palatal plate therapy" OR "Castillo-Morales concept" OR "Stimulating palatal plate")   |
| CINAHL           | ((("Down Syndrome" OR "trisomy 21" OR mongolism OR "meiotic nondisjunction" OR "mitotic nondisjunction" OR "partial trisomy 21") AND ("orthotic devices" OR "palatal plate" OR "Orofacial regulation therapy" OR "Castillo-Morales" OR "stimulating plate" OR "Functional appliances" OR "Palatal plates" OR "Modified Palatal Plate" OR "Orofacial Stimulation" OR "Castillo-Morales stimulating plate" OR "Castillo-Morales plate" OR "Palatal plate therapy" OR "Castillo-Morales concept" OR "Stimulating palatal plate"))  |
| Scopus           | (ALL("Down Syndrome") OR ALL ("trisomy 21") OR ALL (mongolism) OR ALL ("meiotic nondisjunction") OR ALL ("mitotic nondisjunction") OR ALL ("partial trisomy 21")) AND (ALL ("orthotic devices") OR ALL ("palatal plate") OR ALL ("Orofacial regulation therapy") OR ALL ("Castillo-Morales") OR ALL ("stimulating plate") OR ALL ("Functional appliances") OR ALL ("Palatal plates") OR ALL ("Modified Palatal Plate") OR ALL ("Orofacial Stimulation") OR ALL("Castillo-Morales stimulating plate") OR ALL ("Castillo-Morales plate") OR ALL ("Palatal plate therapy") OR ALL ("Castillo-Morales concept") OR ALL ("Stimulating palatal plate")) |
| EMBASE           | ("Down Syndrome" OR "trisomy 21" OR "partial trisomy 21") AND ("orthotic devices")  |
| Web of Science   | ALL=((("Down Syndrome" OR "trisomy 21" OR mongolism OR "meiotic nondisjunction" OR "mitotic nondisjunction" OR "partial trisomy 21") AND ("orthotic devices" OR "palatal plate" OR "Orofacial regulation therapy" OR "Castillo-Morales" OR "stimulating plate" OR "Functional appliances" OR "Palatal plates" OR "Modified Palatal Plate" OR "Orofacial Stimulation" OR "Castillo-Morales stimulating plate" OR "Castillo-Morales plate" OR "Palatal plate therapy" OR "Castillo-Morales concept" OR "Stimulating palatal plate"))  |
| Cochrane Library | ((("Down Syndrome" OR "trisomy 21" OR mongolism OR "meiotic nondisjunction" OR "mitotic nondisjunction" OR "partial trisomy 21") AND ("orthotic devices" OR "palatal plate" OR "Orofacial regulation therapy" OR "Castillo-Morales" OR "stimulating plate" OR "Functional appliances" OR "Palatal plates" OR "Modified Palatal Plate" OR "Orofacial Stimulation" OR "Castillo-Morales stimulating plate" OR "Castillo-Morales plate" OR "Palatal plate therapy" OR "Castillo-Morales concept" OR "Stimulating palatal plate")) in Title Abstract Keyword - (Word variations have been searched)   |
| LILACS           | ("Down Syndrome" OR "trisomy 21" OR mongolism OR "meiotic nondisjunction" OR "mitotic nondisjunction" OR "partial trisomy 21") and ("orthotic devices" OR "palatal plate" OR "Orofacial regulation therapy" OR "Castillo-Morales" OR "stimulating plate" OR "Functional appliances" OR "Palatal plates" OR "Modified Palatal Plate" OR "Orofacial Stimulation" OR "Castillo-Morales stimulating plate" OR "Castillo-Morales plate" OR "Palatal plate therapy" OR "Castillo-Morales concept" OR "Stimulating palatal plate")   |

The eligibility criteria were defined with PICOT: participants (individuals with T21); intervention (SPP use); comparison (control group comprising individuals with T21 who did not use SPP or used it for less time); outcomes (habitual lip and tongue posture); type of study (experimental, quasi-experimental, longitudinal observational, or case-control studies).

The inclusion criteria were as follows: original research articles designed as experimental, quasi-experimental, longitudinal observational, or case-control studies, approaching SSP therapy in the treatment of children with T21 and assessing habitual lip and tongue posture as an outcome. Studies without a control group or that were not available in full-text were excluded.

The material was analyzed in stages. In the first one, duplicate references in the databases were excluded. Abstracts were read in the second stage, and articles that did not meet the inclusion criteria were excluded, while studies that met them were retrieved in full text.

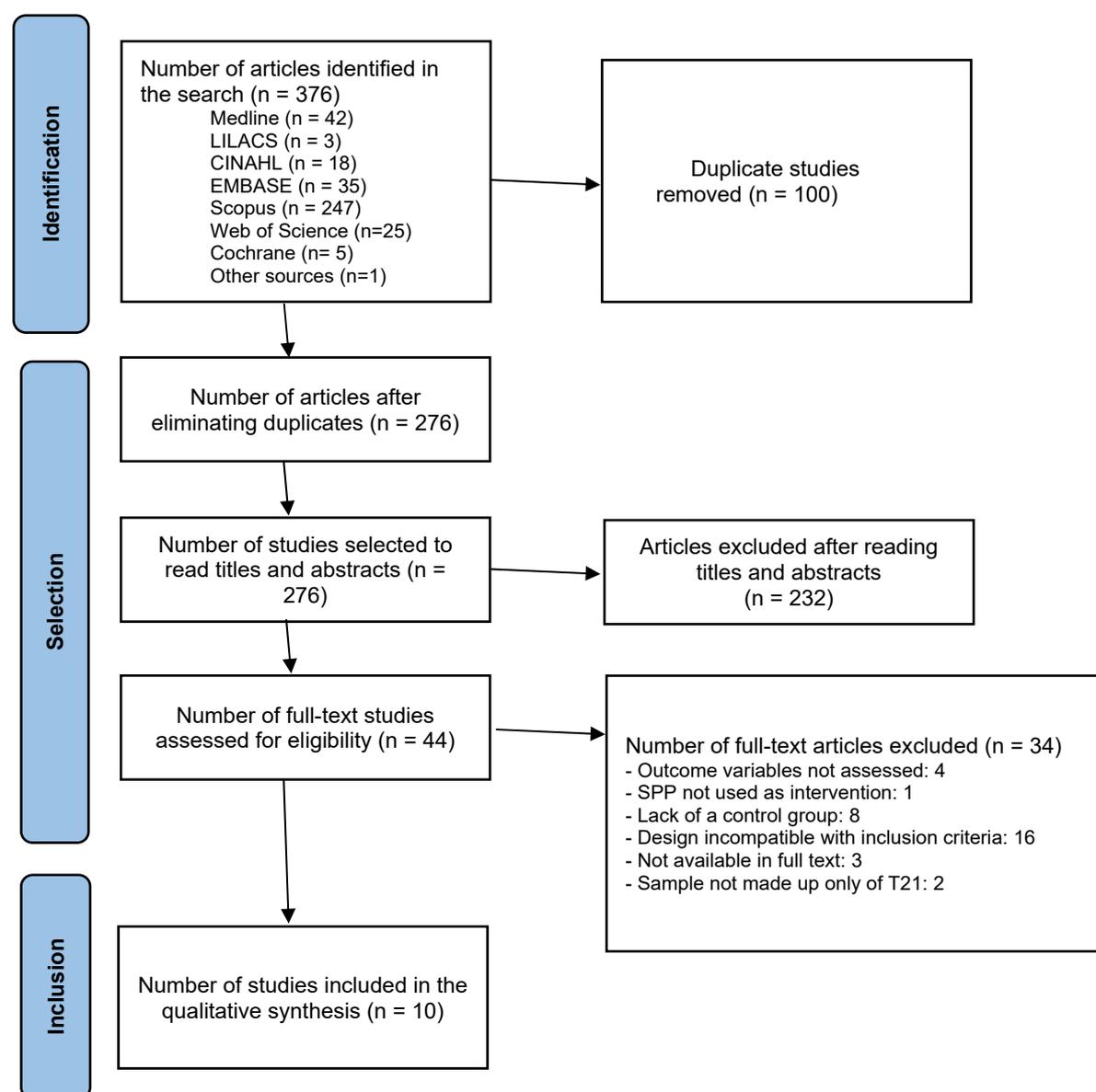
In the third stage, articles potentially relevant to the review were analyzed in full text. The following data were collected in a protocol developed by the researchers: author, year of publication, the country where the study was conducted, type of study, sample characteristics, age at intervention initiation, intervention duration, frequency of SPP use, other combined orofacial stimulations, the method used to assess habitual tongue and lip posture, and results regarding tongue and lip posture.

The articles were independently searched and selected by two researchers – a speech-language-hearing therapist specialized in oral-motor function and a pediatric dentist. Data was handled in a Microsoft Excel spreadsheet with two answer options (yes or no) for researchers in the selection process. In the second stage, the raters decided whether to include each study. Those that obtained a “yes” from both researchers were included to be read in full text, while a “no” from both meant exclusion from the paper. Initial calibration was made with 20% of the studies, resulting in a very good level of agreement (kappa: 0.83). After this stage, the researchers independently read the remaining studies. It was established that when the

raters’ answers diverged, a consensus meeting would be held; if the situation persisted, a third person would be consulted.

## LITERATURE REVIEW

Initially, 376 references were found. After an additional search in references in the articles, one more study was included. After the first stage (removal of duplicates), 276 articles remained; after the second stage (exclusion by abstract reading), 44 remained; and after the third stage (analysis of full-text articles), only 10 articles remained in this study, as shown in the flowchart developed according to PRISMA protocol guidelines<sup>23</sup> (Figure 1).



Source: Flow Diagram (Page et al., 2021)<sup>23</sup>

**Figure 1.** Flowchart with the different phases of the review, based on the PRISMA protocol guidelines.

Tables 1 and 2 present a summary of the main findings in the selected references.

The studies analyzed in this literature review were published between 1996 and 2007. More recent

studies on the topic were retrieved in the search, but they did not have control groups; therefore, they were not included in the analysis. Most research was conducted in Europe, especially in Germany and

**Table 1.** Data on the methods used in research on the use of stimulating palatal plate in children with trisomy 21

| Author (year of publication)<br>Country – Design  | Sample  | Age when the plate started being worn           | Intervention duration  | Frequency of plate use                                  | Combined orofacial stimulation? | Data collection method   |
|---|---|---|--|---|---------------------------------|--|
| Carlstedt et al. (1996) <sup>24</sup><br>Sweden – Experimental study                              | - SG: 14 children with T21 (used SPP)<br>- CG: 15 children with T21 (did not use SPP)   | From 3 months to 5 years, a mean of 24±6 months | 12 months  | 30 minutes to 1 hour, 2x a day                          | Yes                             | Analysis of 10-minute video recording without SPP                          |
| Hohoff, Ehmer (1997) <sup>25</sup><br>Germany – Retrospective longitudinal observational study    | - SG: 20 children with T21 (used SPP)<br>- CG: 18 children with T21 (used SPP for a maximum of 1 month)   | 4.8 months on average                           | 10.8 months on average   | 2 hours, 2x a day                                       | Yes                             | Clinical observation and parental questionnaire                            |
| Hohoff, Ehmer (1999) <sup>26</sup><br>Germany – Retrospective longitudinal observational study    | - Group 1: 38 children with T21 (used SPP for 4 months on average)<br>- Group 2: 18 children with T21 (used SPP for 9.9 months on average)  | 6.5 months on average                           | - Group 1: 4 months on average<br>- Grupo 2: 9.9 months on average | 1 hour, 4x a day  | Yes                             | Clinical observation and parental questionnaire                            |
| Carlstedt et al. (2001) <sup>27</sup><br>Sweden – Experimental study                              | - SG: 9 children with T21 (used SPP)<br>- CG: 11 children with T21 (did not use SPP)  | From 3 to 33 months                             | 48 months  | 1 hour, 2 to 3x a day (minimum 2hr a day)               | Yes                             | Clinical observation, videos, and parental interview                       |
| Schuster, Giese, (2001) <sup>28</sup><br>Germany – Retrospective longitudinal observational study | - SG: 20 children with T21, lack of lip closure, and habitual protruded tongue posture (used SPP), with a mean of 8.8±2.3 years.<br>- CG: 13 children with T21, slightly changed tongue and lip posture (did not use SPP), with a mean of 8.9±3.0 years.      | 8±5 months                                      | 2 years  | 30 minutes, 4x a day                                    | Yes                             | Clinical observation, photos, and parental questionnaire                   |
| Rincón et al. (2003) <sup>29</sup><br>Colombia – Quasi-experimental study                         | - SG: 31 children with T21 (used SPP) with a mean of 6 months (1-11 months)<br>- CG: 13 children with T21 (did not use SPP) with a mean of 5 months (1-12 months)   | From 1 to 11 months                             | 6 months   | 4 hours a day   | Not reported                    | Analysis of 10-minute video and parental questionnaire                     |
| Carlstedt et al. (2003) <sup>30</sup><br>Sweden – Experimental study                              | - SG: 9 children with T21 (used SPP)<br>- CG: 11 children with T21 (did not use SPP)  | From 3 to 33 months                             | 48 months  | 1 hour, 2x a day  | Yes                             | Clinical observation, videos, speech recording, and parental questionnaire |
| Zavaglia et al. (2003) <sup>31</sup><br>Italy – Retrospective longitudinal observational study    | - SG: 38 children with T21 (used SPP)<br>- CG: 30 children with T21 (did not use SPP)   | 15 months on average                            | Minimum of 3 years; maximum duration not reported                  | Initially, 1 hour a day, increasing to 1 hour, 3x a day | Yes                             | Clinical observation   |
| Backman et al. (2007) <sup>32</sup><br>Sweden – Retrospective longitudinal observational study    | - SG: 37 children with T21 (used SPP) with a mean age of 50.6 months (44–57)<br>- CG: 31 children with T21 (did not use SPP) with a mean age of 49.8 months (42–56)<br>- CG: 36 children without T21 (did not use SPP) with a mean age of 49.3 months (42–55) | < 6 months                                      | 42 months  | 5 to 30 minutes, 2 to 3x a day                          | Yes                             | Clinical observation, videos, and parental interview                       |
| Carlstedt et al. (2007) <sup>33</sup><br>Sweden – Experimental study                              | - SG: 9 children with T21 (used SPP)<br>- CG: 11 children with T21 (did not use SPP)<br>Mean age at the end of treatment: 5.6±11.5 years  | From 3 to 33 months, a mean of 24 months        | 49 to 58 months  | 1 hour, 2x a day  | Yes                             | Clinical observation, videos, and parental questionnaire                   |

Captions: T21 – trisomy 21; SG – study group; CG –control group, SPP – stimulating palatal plate, x – times..

**Table 2.** Variables analyzed, results obtained, and difficulties found in the studies regarding the use of stimulating palatal plates

| Author (year)                         | Assessment method for habitual tongue posture  | Assessment method for habitual lip posture   | Results  |
|---------------------------------------|--|--|--|
| Carlstedt et al. (1996) <sup>24</sup> | <u>Duration of the following postures in the video:</u><br>- Visible tip of the tongue<br>- Active (moving) protruded tongue<br>- Inactive (still) protruded tongue  | <u>Duration of the following postures in the video:</u><br>- Mouth closed (lips fully touching each other)<br>- Mouth open (lips not touching each other, and tongue inside the mouth) | - After 9 months of treatment, the mouth was closed for longer ( $P < 0.01$ ), and the tongue was inactively protruded for less time ( $P < 0.01$ ) in SG, in comparison with CG.<br>- After 12 months, the mouth was closed for longer ( $p < 0.001$ ), and the tongue was inactively protruded ( $p < 0.001$ ) and actively protruded for less time ( $p < 0.05$ ) in SG, in comparison with CG. |
| Hohoff, Ehmer (1997) <sup>25</sup>    | Parental opinion on improved or worsened tongue posture on average 39.5 months after treatment.  | Parental opinion on improved or worsened lip posture on average 39.5 months after treatment.   | - In SG, 80% of the patients improve tongue and lip posture (parental opinion).  |
| Hohoff, Ehmer (1999) <sup>26</sup>    | Parental opinion on tongue posture.<br><u>Clinical assessment:</u><br>- Tongue much protruded in relation to the lips<br>- Tongue slightly protruded in relation to the lips<br>- Tongue inside the oral cavity<br>Group 1: data collected soon after the treatment. Group 2 – data was collected 53 months after the treatment. | Parental opinion on lip posture.<br><u>Clinical assessment:</u><br>- Mouth wide open<br>- Mouth partly open<br>- Mouth closed  | - Group 1: 65.8% of the children improved lip and tongue posture, according to parental report.<br>- Group 2: 88.9% improved lip posture, and 77.7% improved tongue posture, according to parental report.<br>- In clinical assessment, most cases improved tongue and lip posture.<br>- The analysis was qualitative, without comparison between the groups.                                      |
| Carlstedt et al. (2001) <sup>27</sup> | <u>Clinical assessment:</u><br>- Tongue on the lower lip, mouth closed<br>- Inactive protruded tongue, mouth open<br>- Active protruded tongue   | <u>Clinical assessment:</u><br>- Lips fully closed<br>- Lips partly closed<br>- Inactive open mouth<br>- Active open mouth   | - CG had more periods with inactive lips and tongue ( $p < 0.05$ ) when interacting.<br>- No difference in tongue posture and lip closure between the groups.  |
| Schuster, Giese (2001) <sup>28</sup>  | <u>Clinical assessment:</u><br>- Inside the oral cavity<br>- On the alveolar ridge<br>- On the lower lip<br>- Eversion<br>- Slightly protruded over the lower lip<br>- Much protruded over the lower lip   | <u>Clinical assessment:</u><br>- Mouth closed<br>- Mouth open  | - Tongue posture and lip closure improved in both groups – greater improvement in SG.<br>- Better results were obtained in cases of greater adherence.<br>- Positive parental feedback on tongue posture and decreased sialorrhea.<br>The analysis was only descriptive.   |
| Rincón et al. (2003) <sup>29</sup>    | Clinical assessment of tongue posture  | Clinical assessment of lip posture   | 100% of SG children improved tongue posture, and 13 children progressed to lip closure. The analysis was qualitative, without comparison between the groups.   |
| Carlstedt et al. (2003) <sup>30</sup> | <u>Duration of the following postures in the video:</u><br>- Inside the oral cavity<br>- Intermittently visible<br>- Constantly visible  | <u>Duration of the following postures in the video:</u><br>- Mouth closed<br>- Mouth open less than half the time<br>- Mouth open more than half the time                              | - Better lip closure and tongue posture ( $p < 0.05$ ) in SG, in comparison with CG ( $p < 0.05$ ).  |
| Zavaglia et al. (2003) <sup>31</sup>  | <u>Clinical assessment:</u><br>- Tongue occasionally protruded, passing the lower lip<br>- Tongue slightly protruded, but continuously passing the lower lip<br>- Tongue significantly protruded, passing the lower lip  | Not reported   | - Improved tongue posture and lip closure in most SG cases. No comparison was made between the groups.   |
| Backman et al. (2007) <sup>32</sup>   | <u>Clinical assessment:</u><br>- Retracted<br>- Visible<br>- Out of the mouth  | Clinical assessment:<br>- Mouth closed<br>- Mouth partly open<br>- Mouth open  | - Better facial expressions in children with T21 in SG than in CG. In SG with T21, 66.7% of the children had active facial expressions, in contrast with only 34.5% in CG with T21.<br>The study does not specify in what terms the muscles were considered active or inactive; however, it mentions lip closure and tongue posture as parameters used in this classification.                     |
| Carlstedt et al. (2007) <sup>33</sup> | <u>Duration of the following postures in the video:</u><br>- Tip of the tongue visible, and tongue over the lower lip<br>- Inactive protruded tongue (still tongue, outside the oral cavity)<br>- Active protruded tongue (moving tongue, outside the oral cavity).  | Duration of the following postures in the video:<br>- Mouth closed (total or partial lip closure)<br>- Mouth open (active or inactive when open > 15 to 20 mm).                        | - SG had lip closure for longer ( $p < 0.05$ ) and the tongue was inactively protruded for less time, in comparison with CG ( $p < 0.05$ ).  |

Captions: T21 – Trisomy 21; SG – study group; CG – control group, SPP – stimulating palatal plate.

Sweden. Moreover, some research conducted in these two countries was from the same group of researchers, possibly using the same sample, though analyzed in different treatment moments.

Four of the studies were experimental<sup>24,27,30,33</sup>, one was quasi-experimental<sup>29</sup>, and the others were retrospective longitudinal observational ones. The samples in the studies had 20 to 104 participants. The study with the most children with T21 using SPP had 38 such participants<sup>31</sup>. The comparison groups comprised children with T21 who did not use SPP<sup>24,27-33</sup>, usually matched for age. Nonetheless, they either received orofacial stimulation or used SPP for less time than the study group<sup>25,26</sup>. No article had a control group with a different therapy. In the four experimental studies, the participants were randomly allocated into groups; all of them were published by the same group of researchers<sup>24,27,30,33</sup>. The authors pointed out the difficulty of conducting randomized studies, due to parental non-consent, despite offering SPP treatment 1 year after beginning the research<sup>24,27,30</sup>.

SPP began being used in the studies at 1 month<sup>29</sup> to 5 years old<sup>24</sup>; generally, the studies began the approach in their first year of life. Hohoff and Ehmer<sup>26</sup> suggest that treatment begin in the first weeks of life, the time of the central nervous system's greatest development. Furthermore, when their teeth begin erupting, adaptation becomes harder<sup>26</sup> – sometimes even requiring an interruption in the treatment.

The intervention duration in the studies ranged from 4<sup>26</sup> to 58 months<sup>33</sup>. There was no consensus on the minimum time necessary to change habitual posture. The articles suggest that each person has different needs; some children may achieve the desired result sooner than other ones. The study whose intervention lasted only 4 months reported improvements in tongue posture and lip closure<sup>26</sup>, which remained in the long run (assessment made 53 months after the treatment).

Most articles indicate SPP use for 30 minutes<sup>24,28</sup> up to 2 hours<sup>25</sup>, two<sup>24,25,30,33</sup> to four times a day<sup>28</sup>. Continuous SPP use is not indicated; otherwise, the child might get used to the sensory stimuli and stop paying attention to them<sup>31</sup>. Some authors recommend gradually increasing the time of SPP use<sup>31</sup>. No study researched the influence of daily time of SPP use on the results. Castillo-Morales<sup>9</sup> recommends that the plate be removed during meals so it will not negatively influence the sensory experience while eating.

SPP was used in combination with other forms of orofacial muscle stimulation, except in one study<sup>29</sup>.

According to the literature, SPP is an auxiliary resource in the orofacial treatment of children with T21; hence, it should not be used alone<sup>8</sup>. Castillo-Morales presents SPP as part of the Orofacial Regulation Therapy, conducting both approaches together<sup>8</sup>. In the studies, SPP was usually used in combination with some type of orofacial stimulation, though not specifying whether the Orofacial Regulation Therapy, Myofunctional Therapy, or another therapeutic approach was used. The studies in general did not report either the therapeutic strategies the professionals used or their frequency. Since most children underwent different treatments in combination, no conclusion can be drawn on which specific treatment was responsible for the muscle changes verified in the results. Therefore, the benefits pointed out in this literature review must be ascribed to SPP use in combination with orofacial muscle stimulation.

Study methodologies were heterogeneous, with varying intervention duration, time until reassessment, and SPP characteristics. These ranged from traditional ones, with a stimulating button in the center of the palate and vestibular knobs, to modified varieties, with accessories.

Data collection in the studies was based on three main procedures: clinical assessment, in which participants were observed by the professional in 80% of studies<sup>25-28,30-33</sup>; opinion of parents or guardians, obtained with questionnaires or interviews, also in 80% of studies<sup>25-30,32,33</sup>; and video recording<sup>24,27,29,30,32,33</sup>, in 60% of studies. One study was based on photographic records<sup>28</sup>. Each type of assessment has positive and negative aspects. Clinical observation is subjective and depends on professional experience; also, as it is conducted at a single moment, in an environment other than what the child is used to, it may not be reliable. In two studies<sup>27,30</sup>, clinical assessments were made by two professionals blinded to the group to which the child belonged, which made their results more reliable. The family reports, on the other hand, are influenced by their desires and expectations. Videos and photographs may minimize these problems. Frontal face photographs may not record the child's most frequent posture<sup>28</sup>, whereas video recording – though the most reliable analysis method – may be influenced by the child's fatigue and environmental conditions. Therefore, the most adequate is to use different assessment methods in combination.

Most studies reported positive results regarding improved tongue posture and lip closure. Only one study<sup>27</sup> verified no difference in tongue posture

between the study and control groups after 48 months of treatment. However, its sample was rather small, with only nine children undergoing treatment.

The studies that reassessed participants years after finishing the treatment<sup>25,26</sup> verified that the results had remained in the long run. One of these studies reassessed the participants 39.5 months after the treatment and verified better habitual tongue posture, tongue mobility, and lip closure<sup>25</sup>. Another study assessed participants 53 months after the treatment and likewise verified that improvements in tongue posture and lip closure had remained in most cases<sup>26</sup>.

SPP use and therapeutic strategies were biased in the studies because they were assessed only with parental reports. Participants' withdrawal from the research was recurrent in the studies. However, many of them neither explored the reasons for withdrawal nor established strategies to address discontinued follow-up. Some studies did not present assessment data on variables investigated at the beginning of the research. Confounding factors – such as the participants' age, other combined treatments, and degree of severity of orofacial characteristics at the beginning of the research – were not always controlled. Some study results were based only on clinical observation by a single professional, which is considered non-reliable. Some measures may be taken to improve assessment reliability, namely: assessments independently made by more than one professional, blind assessments, and video recordings. Furthermore, some studies presented only descriptive statistical analyses in the comparison between groups.

## CONCLUSION

Results reported in the studies suggest that combined treatments, SPP use and orofacial stimulation, improve tongue posture and lip closure in children presented with T21.

## REFERENCES

1. Brandão IM, Fonseca V, Madi RR. Prevalence of people with Down syndrome in Brazil. *Scientia Plena*. 2012;8(3):037501.
2. Kruszka P, Porras AR, Sobering AK, Ikolo FA, La Qua S, Shotelersuk V. Down syndrome in diverse populations. *Am J Med Genet A*. 2017;173(1):42-53.
3. World Health Organization. Human Genomics in Global Health [Homepage on the Internet] [accessed 2020 April 3]. Available at: <https://www.who.int/genomics/public/geneticdiseases/en/index1.html>
4. Doriguêto PVT, Carrada CF, Scalioni FAR, Abreu LG, Devito KL, Paiva SM et al. Malocclusion in children and adolescents with Down syndrome: a systematic review and meta-analysis. *Int J Paediatr Dent*. 2019;29(4):524-41.
5. Kaczorowska N, Kaczorowski K, Laskowska J, Mikulewicz M. Down syndrome as a cause of abnormalities in the craniofacial region: A systematic literature review. *Adv Clin Exp Med*. 2019;28(11):1587-92.
6. Giacchini V, Tonial A, Bolli-Mota H. Aspects of language and oral motor observed in children treated at an early stimulation sector. *Disturb Comun*. 2013;25(2):253-65.
7. Pinheiro DLSA, Alves GAS, Fausto FMM, Pessoa LSF, Silva LA, Pereira SMF et al. Effects of electrostimulation associated with masticatory training in individuals with down syndrome. *CoDAS*. 2018;30(3):e20170074.
8. Morales RC. *Terapia de regulación orofacial*. São Paulo: Memnon, 2002.
9. Xepapadeas AB, Weise C, Frank K, Spintzyk S, Poets CF, Wiechers C et al. Technical note on introducing a digital workflow for newborns with craniofacial anomalies based on intraoral scans - part I: 3D printed and milled palatal stimulation plate for trisomy 21. *BMC Oral Health*. 2020;20(171):1-7.
10. Javed F, Akram Z, Barillas AP, Kellesarian SV, Ahmed HB, Khan J et al. Outcome of orthodontic palatal plate therapy for orofacial dysfunction in children with Down syndrome: a systematic review. *Orthod Craniofac Res*. 2018;21(1):20-6.
11. Furlan RMMM, Pretti H, Almeida TDD. Terapia miofuncional orofacial associada ao uso da placa palatina de memória em crianças com Síndrome de Down – discutindo uma série de casos clínicos. In: Silva HJ, Tessitore A, Motta AR, Cunha DA, Berretin-Felix G, Marchesan IQ et al., editors. *Discutindo casos clínicos em Motricidade Orofacial*. São José dos Campos; Pulso Editorial: 2020. p.59-65.
12. Limbrock GJ, Castillo-Morales R, Hoyer H, Stover B, Onufer CN. The Castillo-Morales approach to orofacial pathology in Down syndrome. *Int J Orofacial Myology*. 1993;19:30-7.

13. Limbrock GJ, Castillo-Morales REC, Hoyer H, Stover B, Onufer CCN. The Castillo-Morales therapy in 39 children with Down syndrome. *J US Army Med Dep.* 1994;8:6-12.
14. Mello CRS, Gugisch RC, Fraiz, FC, Lopes MN. Orofacial regulation therapy in Down Syndrome. Case Report. *J Bras Odontoped Odontol Bebê.* 1998;1(1):34-43.
15. Carneiro VL, Sullcahuamán JAG, Fraiz FC. Utilización de la placa palatina de memoria y desarrollo orofacial en infante con Síndrome de Down. *Revista Cubana Estomatología.* 2012;49(4):305-11.
16. Matthews-Brzozowska T, Walasz J, Matthews-Kozanecka M, Matthews Z, Kopczyński P. The role of the orthodontist in the early simulating plate rehabilitation of children with Down syndrome. *J Med Sci.* 2014;2(83):145-51.
17. Walasz J, Matthews-Brzozowska T, Matthews-Kozanecka M, Cudzilo D. Types and positioning of palatal plate stimulation elements in children with down syndrome. *Jour of Med Sc & Tech.* 2014;3(1):1-6.
18. De la Cruz-Campos SB, Cárdenas-Flores CM. Palatine plate use for improving the closing mouth and tongue position in patients with down syndrome: clinical case report. *Revista Científica Odontológica.* 2016;4(1):464-70.
19. Padró SMJ, Barraza VE, Brucher SC, Concha TE, Delagado V. Efectividad del uso de placas palatinas y de la estimulación orofacial en el desarrollo oral en niños con síndrome de Down. *Rev Chil Pediatr.* 2010;81(1):46-52.
20. Limbrock GJ, Fischer-Brandies H, Avasse C. Castillo-Morales' orofacial therapy: treatment of 67 children with Down syndrome. *Dev Med Child Neurol.* 1991;33(4):296-303.
21. Andrade MER, Calvillo JN, Díaz MAS. Terapia de regulación orofacial mediante la placa Castillo-Morales modificada en el síndrome de Down. Informe preliminar. *Rev ADM.* 1993;50(2):85-8.
22. Hernández-Antonio A, Sánchez- Sánchez M, Azamar-Cruz E, Díaz-Arellano M, Velásquez-Paz AL, Ángeles-Castellanos M. Regulación orofacial Castillo-Morales y placa palatina modificada en niños con síndrome de Down. *Avan C Salud Med.* 2015;3(2):40-5.
23. Page MJ, McKenzie JE, Bossuyt PM, Boutron I, Hoffmann TC, Mulrow CD, et al. The PRISMA 2020 statement: an updated guideline for reporting systematic reviews *BMJ.* 2021;372(71).
24. Carlstedt K, Dahllöf G, Nilsson B, Modéer T. Effect of palatal plate therapy in children with Down syndrome. A 1-year study. *Acta Odontol Scand.* 1996;54(2):122-5.
25. Hohoff A, Ehmer U. Effects of the Castillo-Morales Stimulating Plate on speech development of children with Down's syndrome - a retrospective study. *Orofac Orthop/Fortschr Kieferorthop.* 1997;58(6):330-9.
26. Hohoff A, Ehmer U. Short-term and long-term results after early treatment with the Castillo-Morales Stimulating Plate. A longitudinal study. *J Orofac Orthop/Fortschr Kieferorthop.* 1999;60(1):2-12.
27. Carlstedt K, Henningsson G, McAllister A, Dahllöf G. Long-term effects of palatal plate therapy on oral motor function in children with Down syndrome evaluated by video registration. *Acta Odontol Scand.* 2001;59(2):63-8.
28. Schuster G, Giese R. Retrospective clinical investigation of the impact of early treatment of children with Down's syndrome according to Castillo-Morales. *J Orofac Orthop/Fortschr Kieferorthop.* 2001;62(4):255-63.
29. Rincón RR, Jiménez BJ, Duque EO, Chaurra MRC. Evaluation after early maxillary orthopedic stimulation in Down syndrome children. *Rev Fac Odontol Univ Antioq.* 2002;14(1):45-53.
30. Carlstedt K, Henningsson G, Dahllöf G. A four-year longitudinal study of palatal plate therapy in children with Down Syndrome: effects on oral motor function, articulation and communication preferences. *Acta Odontol Scand.* 2003;61(1):39-46.
31. Zavaglia V, Nori A, Mansour NM. Long term effects of the palatal plate therapy for the orofacial regulation in children with Down syndrome. *J Clin Pediatr Dent.* 2003;28(1):89-93.
32. Bäckman B, Grevér-Sjölander AC, Bengtsson K, Persson J, Johansson I. Children with Down syndrome: oral development and morphology after use of palatal plates between 6 and 48 months of age. *Int J Paediatr Dent.* 2007;17:19-28.
33. Carlstedt K, Henningsson G, Dahllöf G. A longitudinal study of palatal plate therapy in children with Down syndrome. Effects on oral motor function. *J Disabil Oral Health.* 2007;8(1):13-9.