

## Palatal Plate Therapy In Children with Down Syndrome: A Systematic Review Of Literature

Review Article

Farah Chouchene<sup>1\*</sup>, Fatma Masmoudi<sup>2</sup>, Ahlem Baaziz<sup>3</sup>, Fethi Maatouk<sup>4</sup>, Hichem Ghedira<sup>5</sup>

<sup>1</sup> Pediatric and Preventive Dentistry Department, Faculty of Dental Medicine of Monastir, ABCD F Laboratory of Biological, Clinical and Dento-Facial Approach, University of Monastir, Monastir, Tunisia, Rue HediChekir. Hiboun 5111 Mahdia.

<sup>2</sup> Pediatric and Preventive Dentistry Department, Faculty of Dental Medicine of Monastir, Tunisia, Monastir 5119 Tunisia.

<sup>3</sup> Professor in Pediatric Dentistry, Pediatric and Preventive Dentistry Department, Faculty of Dental Medicine of Monastir, Monastir, Tunisia, Monastir 5119 Tunisia.

<sup>4</sup> Pediatric and Preventive Dentistry Department, Faculty of Dental Medicine of Monastir, Monastir, Tunisia, Monastir 5119 Tunisia.

<sup>5</sup> Pediatric and Preventive Dentistry Department, Faculty of Dental Medicine of Monastir, Monastir, Tunisia, Monastir 5119 Tunisia.

### Abstract

**Background:** The oro-facial abnormalities in children with Down syndrome (DS) can be managed through oro-facial therapy using a palatal plate. The present review aimed to investigate the frequency, duration, and type of palatal plates with their different stimulation elements in palatal plates therapy used in early infancy in children with DS.

**Methods:** Electronic databases including Medline (via PubMed), The Cochrane Library (CENTRAL) and Scopus were searched. Only studies published in English during the last twenty years describing the effects of palatal plate therapy (PPT) on oral motor function were included. The ROBINS-I tool was used to assess the quality of the methodology of the included studies.

**Results:** Six studies were retained, included a total of 300 children with DS with a mean age ranged between 2 months and 13 years. All children in the PPT reported a significant improvement of the mimetic muscles, tongue retraction and significant longer lip closure. The children in the palatal plate therapy group were treated with modified palatal plates according to Castillo Morales. Different palatal plates were used, and the PPT frequencies were ranged between 2 and 3 times daily for 5 to 60 minutes with a duration ranged from 12 to 48 months.

**Conclusion:** All the included studies in the present review, reported that palatal plates designed in accordance with the shape of Castillo-Morales basic plate when used before 3 months of age and for several minutes a day combination with orofacial physiotherapy, improved the orofacial disorders in children with DS.

**Keywords:** Down Syndrome; Trisomy 21; Palatal Plate Therapy; Orofacial Regulation Therapy.

### Introduction

Children with Down syndrome (DS) exhibit peculiar orofacial features. These orofacial characteristics have a considerable negative impact on the quality of life, causing problems related to the performance of daily activities [1-3].

To improve oral motor function of children with DS, a therapeutic concept, orofacial regulation therapy (OFRT), was developed and introduced by Castillo-Morales [4, 5].

This therapy aimed to eliminate tongue dysfunctions and improve the function of the orbicularis oris and mimetic muscles, leading to improvements in sucking, articulation, swallowing and nasal breathing [6, 7].

OFRT includes the functional diagnosis of oral sensorimotor dysfunctions, a special manual facial stimulation program, and treatment of oral functions with removable activating palatal plates and other orthodontic appliances [7].

#### \*Corresponding Author:

Farah Chouchene,

Pediatric and Preventive Dentistry Department, Faculty of Dental Medicine of Monastir, ABCD F Laboratory of Biological, Clinical and Dento-Facial Approach, University of Monastir, Monastir, Tunisia, Rue HediChekir. Hiboun 5111 Mahdia.

Tel: 0021622821412

E-mail: Farah.pedo@gmail.com

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The palatal plate therapy (PPT) was designed to stimulate tongue movements, to increase mobility of the upper lip improving the facial musculature tonus and consists of two simultaneous steps: the first one is the insertion of a palatal plate device and the second one is a simultaneous orofacial therapy provided by a physiotherapist or speech-language pathologist [7, 8].

The aim of the present review was to investigate the frequency, duration, and type of palatal plates with their different stimulation elements in palatal plates therapy used in early infancy in children with DS.

## Methods

### Protocol

The present review was reported according to the principles of the Preferred Reporting Items for Systematic Reviews and Meta-analysis (PRISMA) statement [9] and the Cochrane Handbook [10].

### Review question

The review question was established based on the Participants, Interventions, Control, and Outcomes (PICO) principles: “In children with Down’s syndrome, is the PPT effective, and what are the frequency, duration and type of stimulation palatal plates used?” The detailed PICO principles were as follows: 1. Participants: Children with Down syndrome who received palatal plate. 2. Interventions: Palatal plate therapy. 3. Control: Children with Down syndrome not having received palatal plate. 4. Outcome: oral motor function improvement.

### Eligibility criteria

Studies were considered eligible if they met the following criteria: Longitudinal/observational studies or controlled clinical trials comparing palatal plates treatment in children with down syndrome (under the age of 18 years) with a control group for at least 12-month follow-up period describing the type of palatal plate used.

Evaluating the treatment effect of palatal plates by at least two assessment methods: a clinical examination or a parental questionnaire and video recording.

Case series, case reports, studies focusing on adults were excluded.

### Search strategy

An electronic literature search was conducted independently by two authors (FC and FM) using MEDLINE (via PubMed), The Cochrane Library (CENTRAL) and Scopus databases.

The following search terms and combinations of Medical Subject Heading terms (MeSh) were used and adapted for each database: (Down Syndrome OR Trisomy 21) AND (Stimulation plate OR Palatal plate OR Orthodontic Appliances, Removable OR Dental appliance) AND (Orofacial OR Orofacial regulation therapy) AND (Child).

Only articles published in English from the year January 1, 2000, to March 31, 2020, were included.

The last research was conducted in April 2020.

The two authors (FC and FM) supplemented the electronic search by a manual research.

The research was also supplemented by tracking citations of the relevant studies via Google Scholar. A research of the gray literature was also carried out by the authors to identify any additional unpublished articles.

### Studies selection

Two authors (FC and FM) independently screened the titles and abstracts of all the records selected from the different databases. Then, to select the articles that meet the inclusion criteria the two authors independently screened all the selected full texts and all the references the included studies. Agreement and discrepancies between the two authors were resolved by discussion.

### Data collection

Two authors (FC and FM) independently collected data from the collected studies using a data sheet extraction.

The following variables were included: publication details (author and year), study design, number and age of children, sample size of test and control group, frequency, duration, and palatal plate design assessment methods, follow-up period and main outcomes.

All the studies were subject to qualitative analyses.

In the present systematic review, the palatal plate therapy success was defined on the accomplishment in oral motor function improvement: longer closed mouth, improved tongue position and improved muscle function.

### Quality assessment

Each included article was assessed independently by the authors (FC and FM) using the ROBINS-I tool for assessing risk of bias in non-randomized studies of interventions for seven domains [11].

Domains one and two covered confounding and selection of participants into the study, address issues before the start of the interventions. Domain three covered the classification of the interventions. The other four domains covered: biases due to deviations from intended interventions, missing data, measurement of outcomes and selection of the reported result.

Each domain was divided into three categories as low risk of bias, unclear and high risk of bias.

If one or all domains were evaluated to be of low risk; the study was classified as low risk of bias, if one or more domains were evaluated to be unknown risk; the study was classified as moderate risk.

If one or more domains were evaluated to be a high risk; the study

was classified as high risk of bias.

## Results

### Study selection

About 40 potentially related titles were derived from the electronic research (Figure 1).

After removing the duplicates and reviewing the abstract, the full text of nine studies were retained and compared by the authors with the inclusion criteria. Of these nine studies, three studies did not meet the inclusion criteria were excluded and at the final stage of selection, only six articles were retained for qualitative analyses.

### Study characteristics

In the present review, six longitudinal prospective studies were retained (Table 1).

The studies were published between 2001 and 2014. A total of 300 children with DS with a mean age ranged between two months and 13 years were included. The test group in all the included studies was treated with palatal plates therapy when the control group were children treated only by speech therapy and physiotherapy. The follow-up period ranged between 12 and 58 months.

Figure 1. Prisma flow diagram.

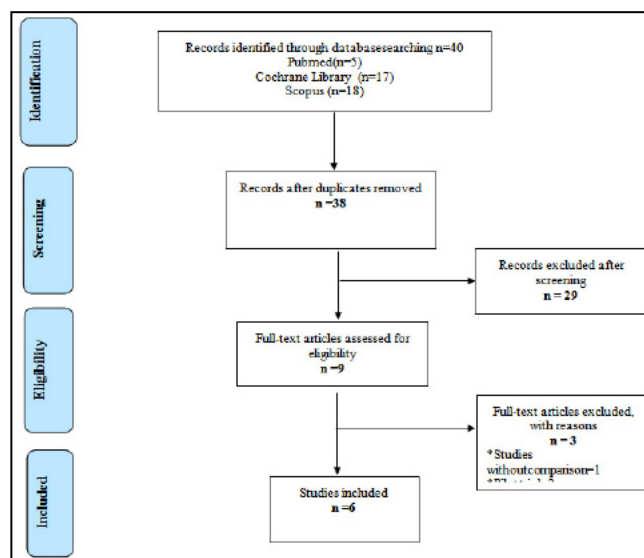


Table 1. Characteristics of the included studies.

Authors/ Year	Study design	Total number of patients		Age/ Mean age	Frequency/ Duration of palatal plate therapy	Follow up (months)	Assessment methods	Main outcomes
		Test	Control					
Carlsedt et al 2001[12]	Longitudinal prospective	9	11	24±6 months	2 times/day; 60 min/ 48 months	Up to 49-58	Clinical examination, video recording	Significant longer « closed mouth » and shorter “inactive protrusion of the tongue”.
Carlsedt et al 2003[14]	Longitudinal prospective	9	11	24±6 months	1 time/day; 60 min/ 48 months	Up to 49-58	Video recording, parental questionnaire	Significant improvement in oral motor function.
Bäckman et al 2003[16]	Longitudinal prospective	42	31	6 months	2-3 times/day; 15 min/ 12 months	Up to 12-48	Clinical examination, video registration, parental questionnaire	Significant improvement in oral motor function.
Carlsedt et al 2007[13]	Longitudinal prospective	9	11	24±6 months	2 times/day; 60 min/ 48 months	Up to 49-58	Video recording, parental questionnaire	Significantly less “inactive muscle function”.
Bäckman et al 2007[6]	Longitudinal prospective	36	31	6 months	2-3 times/day; 30 min/ 42 months	Up to 12-48	Clinical examination, video recording	Significant improvement in oral motor function.
Matthews-Brzozowska et al 2014[17]	Longitudinal prospective	50	50	2 months	Nm	Up to 24	Clinical examination, parental questionnaire	Improved lingual retraction and lip closure.

The different palatal plates designs used in the included studies are summarized in Table 2.

The palatal plate therapy frequencies ranged between two and three times daily for 5 to 60 minutes with a duration ranged from 12 to 48 months.

The outcomes assessed in all the studies included oral parameters, oral motor function, facial expression, tongue position, lip activity and speech.

The treatment outcomes were evaluated by clinical examination and video recording in two studies [6, 12] by video recording and parental questionnaire in two studies, [13, 14] by Clinical examination and parental questionnaire in one study [15] and by clinical examination, video registration and parental questionnaire in one study [16].

**Main outcomes**

All the included studies reported that the palatal plate therapy improved orofacial disorders in children with DS.

In the study by Carlsedt et al. conducted in 2001, [12] the PPT was initiated between the age of 3 and 33 months, and all children had used the plate for at least 4 years for approximately 1h twice or three times a day. The palatal plates used in this study were designed with knobs and/or bowls stimulation areas to enhance the orofacial function [12].

The first and principally used plate in Carlsedt et al. study [12] was designed in accordance with Castillo-Morales.

After 4 years of PPT, extraoral examination and video registrations showed that children in the palatal plate group had significantly more rounding lips during speech (P < 0.05). [12]

**Table 2. The different palatal plates designs used in the included studies.**

Authors/ Year	Palatal plate design
Carlsedt et al 2001[12]	Thin acrylic plates.
	Patients with edentulous jaws: full denture base.
	Patients with teeth: with spring retention elements.
	First palatal plate: with a bowl-shaped depression, a stimulating button anterior to the A-line, and vestibular knobs.
	Second palatal plate: with additional stimulating knobs on the lateral alveolar ridges.
	Third palatal plate: with a metal cube moveable on a wire in the midline of the palate to train the articulatory motor precision in the tip of the tongue.
Carlsedt et al 2003[14].	Thin acrylic plates.
	Patients with edentulous jaws: full denture base.
	Patients with teeth: with spring retention elements.
	First palatal plate: with a bowl-shaped depression at the A-line/ vestibular knobs for the upper lip.
	Second palatal plate: with additional stimulating knobs on the lateral alveolar ridges.
	Third palatal plate: with a metal cube moveable on a wire in the midline of the palate to train the articulatory motor precision in the tip of the tongue.
Bäckman et al 2003[16]	Thin acrylic plates.
	First palatal plate: with a bowl-shaped elevation at the borderline between the hard palate and the velum + a ridged frontal area facing the inside of the upper lip.
	Second palatal plate: with a stainless-steel wire with a movable ball in the premaxillary region.
	Third palatal plate: with several plastic knobs on both sides of the alveolar ridges
Carlsedt et al 2007[13]	Thin acrylic plates.
	Patients with edentulous jaws: full denture base.
	Patients with teeth: with spring retention elements.
	First palatal plate: with a palatal stimulator at the A- line and vestibular ‘knobs’ for a child without teeth.
	Second palatal plate: with a palatal stimulator at the A- line and vestibular pearls for a child with teeth.
Bäckman et al 2007[6]	Thin acrylic plates.
	A first palatal plate: with a ball on a stainless-steel wire behind the maxillary incisors/ a facial arch placed as high as possible in the mouth + a bowl-shaped elevation at the border between the hard palate and the velum.
	A second palatal plate: with plastic bands attached to the facebow, which extends laterally into the mouth crease + a transverse steel wire with three bends at the border between the hard palate and the velum + a moving pearl.
Matthews-Brzozowska et al 2014 [17]	Thin acrylic plates.
	First palatal plate: with a stimulator in the form of a cylinder with a “roller” and wire “whiskers”
	Second palatal plate: with a stimulator in the form of a movable bead and wire “whiskers”.

The mean normal muscle tension recorded was 81.0% ± 11.0% in the palatal plate group, whereas it was 68.2% ± 22.5% in the control group [12].

In this study, a statically significant difference was also reported between the groups in the duration of mouth opening and tongue protrusion ( $P < 0.01$ ) [12].

In the study of Carlsedt et al. conducted in 2003, [14] a significant difference between the groups; in visible tongue ( $P < 0.01$ ), visible tongue during non-speech time ( $P < 0.05$ ) and lip-rounding during spontaneous speech ( $P < 0.01$ ) were reported.

The palatal plate group showed, during non-speech time, significantly longer period of “closed mouth” ( $P < 0.05$ ) [14].

In this study, PPT was started in children aged between 3 and 33 months with a four-years follow-up (49-58 months) [14]. The plates were designed with pearls, knobs, and bowls and were used at least 1 hour twice a day [14].

In this study the most frequently used plates were designed with a bowl-shaped depression at the line A associated with vestibular buttons to stimulate the upper lip [14].

After 4 and 5 months, a second palatal plate with additional stimulation buttons on the lateral alveolar ridges to stimulate the lateral edges of the tongue was used. For older children for a period of five months a third plate with a movable metal cube on a wire at the level of the palate to stimulate the tip of the tongue was used [14].

In Carlsedt et al. study conducted in 2007, [13] the palatal plate therapy was initiated in children aged between 3 and 33 months with a follow-up of at least four years.

The palatal plates used were designed in accordance with the shape of Castillo-Morales basic plates, as full denture base for children with edentulous jaws and with spring retentions when children had teeth [13].

The plates used for approximately one hour twice a day aimed to increase the tongue activity and to stimulate the upper lip [13]. The results of this study, showed that after one year of using the palatal plate, a significant increase of mouth closure ( $P < 0.01$ ), and tongue protrusion ( $P < 0.01$ ) were found [13].

In the study of Bäckman et al. [6, 16] results showed that PPT improved the oral motor performance and prerequisites for articulation.

In Bäckman et al. study, [16] the palatal plates were used two to three times daily for 15 minutes in addition but not during speech therapy exercises.

Three different palatal plates were used. Although the plates were used for only an average of 15 minutes per day, the repeated short-time stimulation seems to have been beneficial [16].

The first palatal plate used between 6 and 10 months of age, was designed to stimulate normalized position of the tongue and lips

[16].

The second palatal plate used between the age of 10 and 14 months, in addition to the stimulation areas in the first plate, had a stainless-steel wire with a movable ball to stimulate lip closure, retraction and lateral movements of the tongue [16].

The third palatal plate used by Bäckman et al. [16] between 14 and 18 months of age was designed to stimulate tongue retraction and lip closure.

During PPT, no negative effects of the plates were noticed by the authors, but some of the palatal plates had to be adjusted to enhance retention either by rebasing or by using adhesive and in minority of cases by making a new plate [6, 12-14, 16, 18].

A special problem in the design of the second plate was noticed by Bäckman et al [16]. Indeed, the arch wire facilitated the removal of the plate by the child.

In the second study of Bäckman et al. [6] children with DS were treated with palatal plate from the age of 6 months, the palatal plates were used two to three times daily for 30 minutes not during speech therapy exercises. Only two different types of palatal plates were used by Bäckman et al [6].

The first palatal plate, intended for use until the age of 30 months, was designed to stimulate the tongue and to improve lip closure [6].

The second palatal plate intended for use between 30 and 48 months, was designed to stimulate lip closure, and mobility of the lateral and dorsal parts of the tongue [6].

In the study of Matthews-Brzozowska et al. [15] children were assessed by clinical examination and parental questionnaire and reported an improvement of the masticatory muscles, tongue retraction and lip closure.

Two types of palatal plates were used one with a stimulator in the form of a cylinder with a “roller” and wire “whiskers” and one with a stimulator in the form of a movable bead and wire “whiskers” [15].

The results showed that the plate with a “roller” produced best results in lip closure, masticatory muscles, and tongue retraction [15].

### Quality assessment

The risk of bias assessment summarized in Figures 2 and 3 were generated by the robvis (visualization tool) which is a web application designed for visualizing risk-of-bias assessments [19].

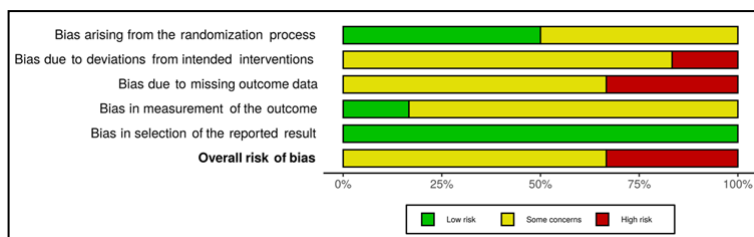
The quality of the included articles in the present review was overall low. Although all the included studies described an improvement in oral motor functions as well as a satisfactory effect of the therapies followed, no clear consensus describing the evaluation methods of these therapies has been well explained and described. Assessment methods, plate designs, and treatment times differ considerably in each study. Different variables of orofacial functions were evaluated in each selected study.



Figure 2. Risk of bias graph. (Green indicates “Low risk of bias, yellow indicates “some concerns of bias”).



Figure 3. Risk-of bias summary (Green indicates “Low risk of bias, yellow indicates “some concerns of bias”).



Four included studies [6, 12-14] had moderate risk of bias as the patients were randomized into control and treatment groups.

However, in these studies a non-standardized method was used, and a large individual variation in the groups was identified. One study [16] had high risk of bias, in this study no drop out informations and a large variation in the study sample size were reported.

Discussion

Children with DS can present some orofacial features that, when not treated and corrected, may interfere with their physical, psychological, and social development [8].

The most common rehabilitation method for orofacial disorders in patients with DS is the orofacial regulation therapy created by Castillo Morales. The Castillo Morales method, combining elements of sensorial rehabilitation, speech therapy exercises, activation of mimic muscles and orthopedic treatment, has shown its effectiveness in the treatment of orofacial neuromotor dysfunctions from the first days after birth [20-22].

This method consists of modeling exercises and therapeutic exercises that prepare for good swallowing, stimulate neuromotor trigger points of the face, activate mimic muscles and evoke movements related to swallowing, chewing, articulation, closing lips and tongue retraction [22-24].

The dentist’s role in orofacial therapy is the provision of palatal plate therapy, which aims to improve oral musculature function and hypotonia by stimulating the lip, tongue muscles and muscles of mastication. Castillo-Morales designed the original palatal plate. These appliances made of thin acrylic usually includes two stimulators; posterior or lingual stimulators and a bowl-shaped elevation at the border line between the hard palate and the velum and it can be oval or round. This type of stimulator is usually added to the first plate used in the youngest children aged 3 months and with the third plate used in children aged from 9 to 13 months.

A Bead-type activator located in a more anterior position is generally added to the second plate used in children aged from 6 to 9 months. Anterior or vestibular stimulators (ridges, knobs...) positioned at the frontal-alveolar labial aspect of the plate ridges varying in number, depth and thickness can be included in these plates. Complementary activators can also be included in palatal plates and most often with the third plates. These activators can be pips or/and granulations located unilaterally or bilaterally. In this third plate, a crater can also serve as activator [22, 24, 25].

Carlstedt et al. [12, 13] noted a significant improvement in mouth closure after four years of treatment with PPT and a significant reduction in tongue protrusion values during speech and non-speech activities.

Carlstedt et al. [14] reported also a statistically significant improvement in facial expression with the palatal plate therapy group.

In the study of Carlstedt et al., the PPT was started between the age 3 and 33 for 4 years for at least 1 hour twice a day, with palatal plates designed with stimulation areas as knobs, pearls, and bowls [14].

Treatment with PPT according to Castillo-Morales in Carlstedt et al. studies [12-14] reported normalized position of the tongue, muscle conditions and mouth closure, also language training and articulation were more developed in children treated with PPT.

Bäckman et al. [6,16] reported also a significant improvement in facial expression and pre-requisites for speech.

In Bäckman et al. studies [6,16] children with DS have been also treated from the age of 6 months, with palatal plate used two to three times a day for 30 minutes and in addition to speech therapy exercises.

Matthews-Brzozowska et al. [15] reported a very important improvement of the not only in tongue retraction and lip closure but also in memetic muscles.

Two different types of palatal plate were used and the results showed that the plate with a cylinder with a “roller” produced the best results about lip closure, and memetic muscles. The tongue retraction was more pronounced after the use of a movable bead but the duration and frequency of PPT was not specified.

In all the include studies in the present review, no negative effects of the plaques were noted, but since the children are young and growing, it was essential to adjust the used palatal plaques to improve their retention and for some children making new adapted plates was necessary.

All the included studies showed a visible improvement in oral motor functions in children with DS. However, the results showed that the effect of palatal plate therapy was achieved only in addition to speech therapy and/or orofacial regulation therapy and physiotherapy.

Indeed, to be effective, PPT must start as soon as possible from the age of 3 months (before the age of 6 months) and until the age of 4 years.

Different types of plates can be used depending on the child's age and dentition.

For best results, the palatal plate should be used at least twice a day for a few minutes (at least 30 min).

The palatal plates should be designed in accordance with the shape of Castillo-Morales basic plates. Different type of simulators can be integrated into the plates: ridges, knobs or bowls.

Complementary activators can also be included in palatal plates, these activators can be substantial pips and granulations.

The included studies reported that the PPT was effective in improving orofacial disorders in children with DS, but, the non-standardized studies design used and the large individual variation in the groups identified using different palatal plate design led to some bias risks.

The mean age of DS children at the start of studies ranged from 2 months to 13 years and the duration of palatal plate therapy ranged from 12 to 58 months which make difficult to interpret the findings for ideal treatment age and duration.

The treatment outcomes were evaluated by non-standardized methods such as clinical examination, video recording and parental questionnaire. These methods would be more reliable if the examiners were calibrated and blinded.

Within these limitations, results suggest that early PPT in children with DS implied favorable results but further trials with standardized evaluation methods are recommended.

## Limitations

No consensus regarding the methods of evaluation in palatal plates treatment was described in the included studies. Different methods of evaluation such as clinical examination, video registration and/or parental interview were used in the studies

but none of these used methods are standardized. In addition, children with this syndrome may present individual variations, specific characteristics, and various clinical symptoms, although no included study mentioned the aspect of individual variation between the different children.

The non-randomized clinical studies included in the present review did not allow to answer the complex question regarding the start of treatment or the treatment duration, for this reason further research is required.

## Conclusion

Various techniques of myofunctional stimulation and device therapy have been proposed over the past 25 years to prevent orofacial dysfunctions in infants and children with SD.

Among these techniques, palatal plate therapy has been suggested and has been found to be effective in improving these orofacial disorders if it is provided at a very young age and in collaboration between speech therapists, physiotherapists and dentists.

Numerous types of plates designed according to the shape of the Castillo-Morales base plates have been shown to be effective when used before the age of 3 months for several minutes per day in addition to speech therapy exercises.

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