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REVIEW ARTICLE

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Outcome of orthodontic palatal plate therapy for orofacial dysfunction in children with Down syndrome: A systematic review

F. Javed[1] | Z. Akram[2] | A. P. Barillas[1] | S. V. Kellesarian[1] | H. B. Ahmed[3] | J. Khan[4] | K. Almas[5]

[1]Department of General Dentistry, Eastman Institute for Oral Health, University of Rochester, Rochester, NY, USA

[2]Faculty of Dentistry, Department of Periodontology, Ziauddin University, Karachi, Pakistan

[3]Private Dental Practitioner, Doha, Qatar

[4]Department of Orofacial Pain and Temporomandibular Joint Disorders, Eastman Institute for Oral Health, University of Rochester, Rochester, NY, USA

[5]Department of Preventive Dental Sciences, College of Dentistry, Imam Abdulrahman Bin Faisal University, Dammam, Kingdom of Saudi Arabia

Correspondence
Fawad Javed, Department of General Dentistry, Eastman Institute for Oral Health, University of Rochester, Rochester, NY, USA.
Email: fawad_javed@urmc.rochester.edu

Abstract

To evaluate the effects of orthodontic palatal plate therapy (OPPT) in the treatment of orofacial dysfunction in children with Down syndrome (DS). Indexed databases were searched. Clinical trials in DS allocated to test (treatment with palatal plates) versus control group (without palatal plates/special physiotherapy for orofacial stimulation) with follow-up of any time duration and assessing mouth closure, tongue position, active and inactive muscle function as outcomes. Study designs, subject demographics, frequency and duration of palatal plate therapy, method for assessment, follow-up period and outcomes were reported according to the PRISMA guidelines. Eight clinical studies were included. The risk of bias was considered high in three studies and moderate in 5 studies. The number of children with DS ranged between 9 and 42. The mean age of children with DS at the start of the study ranged between 2 months and 12 years. The duration of palatal plate therapy ranged between 4 months and 48 months. The follow-up period in all studies ranged from 12 to 58 months. All studies reported OPPT to be effective in improving orofacial disorders in children with DS. Most of the included studies suggest that palatal plate therapy in combination with physiotherapy/orofacial regulation therapy according to Castillo Morales/speech and language intervention seems to be effective in improving orofacial disorders in children with DS. However, the risk of bias of the included studies was high to moderate. Longitudinal trials with standardized evaluation methods, age of children at treatment initiation, treatment duration and standard orofacial outcomes are recommended.

KEYWORDS

Down syndrome, orofacial regulation therapy, palatal plate, systematic review

1 | INTRODUCTION

Orthodontic treatment through orofacial regulation therapy (ORT) from the early childhood can monitor and rectify functional abnormalities.[1] The objective of ORT is a dorsal cranial shift of the tongue, combined with automatic training of the muscles and stimulation of the inactive upper lip.[2] Orthodontic palatal plate therapy (OPPT) is an integral component of ORT that serves two main purposes, that is (i) improvement in oral motor function; and (ii) improving articulation.[3] However, palatal plate therapy alone may be inadequate and may require adjunct treatment such as oral motor stimulation/physiotherapy.[4]

Down syndrome (DS) is a chromosomal disorder caused by the presence of a third copy of chromosome 21. This developmental disability occurs in almost 1:700-1000 births and poses a greater risk of medical problems for the child.[5,6] Several cranial and orofacial dysmorphic features have been also described in children with DS, including small cranium, flattened face, slanted eyes, sloping under chin and low muscle tone in the orofacial region.[7,8] Lip closure is mostly poor, and the

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mouth is often kept open with a protruded tongue resting inactively between the lips.[9] The palate of children with DS is often described as high arched and constricted or narrow, but research results are contradictory; new data showed that the hard palate of infants with DS is of normal shape in the first 6-9 months of age but considerably smaller in all three dimensions compared with healthy normals.[10] Due to oral motor dysfunction, children with DS often show severe articulation disorders.[11,12] Class III malocclusion is found in most children with DS because of the underdevelopment of the maxilla.[13,14] Hypodontia is common among children with DS, and its occurrence resembles that of the general population with respect to type and localization, but it is considerably more frequent among individuals with DS.[15] Moreover, due to narrow nasal meatus, nasal breathing is often difficult.[16] Due to these complex orofacial disorders, children with DS require multidisciplinary medical-dental attention, including the treatment by orthodontists.[17]

A number of studies have evaluated the outcomes of OPPT in the management of orofacial dysfunction in children with DS and showed conflicting results.[18,19] In a clinical trial by Bäckman et al,[18] children with DS treated with OPPT showed significant improvement in oral motor function, facial expression and speech as compared to children with DS without OPPT. Similar results were reported by Carlstedt et al[19] To our knowledge from the indexed literature, there has been no study that have evaluated the effect of OPPT in the management of orofacial function. Therefore, the aim of this study was to systematically review the effects of OPPT in the treatment of orofacial dysfunction in children with DS.

2 | MATERIALS AND METHODS

2.1 | Protocol and focused question

This review was registered at the National Institute for Health Research PROSPERO (Registration Number: CRD42017072525).[20] This study was carried out using the Preferred Reporting Items for Systematic Review and Meta-Analysis (PRISMA) guidelines.[21] The addressed focused question was "Does OPPT improve orofacial functions in children with DS?"

2.2 | Search strategy

Electronic and manual literature searches were conducted in the following databases: MEDLINE/PubMed, EMBASE, CINAHL, Cochrane Central Register of Controlled Trials and Cochrane Oral Health Group Trials Register, up to August 2017 for articles addressing the focused question. For the PubMed library, combinations of following MeSH terms were used: ([stimulation plate] OR [orthodontic appliance] OR [palatal plate] OR [dental appliance] AND ([orofacial] OR [orofacial regulation therapy] AND [Down syndrome] OR [Trisomy 21]).

2.3 | Eligibility

The following eligibility criteria were entailed: longitudinal/observational studies or controlled clinical trials; follow-up in children with DS of any time duration; mouth closure, tongue position, active and inactive muscle function as outcome; and articles published only in English language. Case series, case reports, letters to the editor, abstracts and unpublished articles were excluded.

Three reviewers independently screened titles and abstracts for eligible papers. Interobserver's agreement was assessed by means of kappa scores. Full-text papers that fulfilled the eligibility criteria were identified and included in the review. Reference lists of original studies were handsearched to identify articles that could have been missed during the electronic search. A manual search of journals including *Int J Pediatr Dent* and *J Clin Pediatr Dent* was also performed. Figure 1 describes the screening process according to PRISMA guidelines.[22]

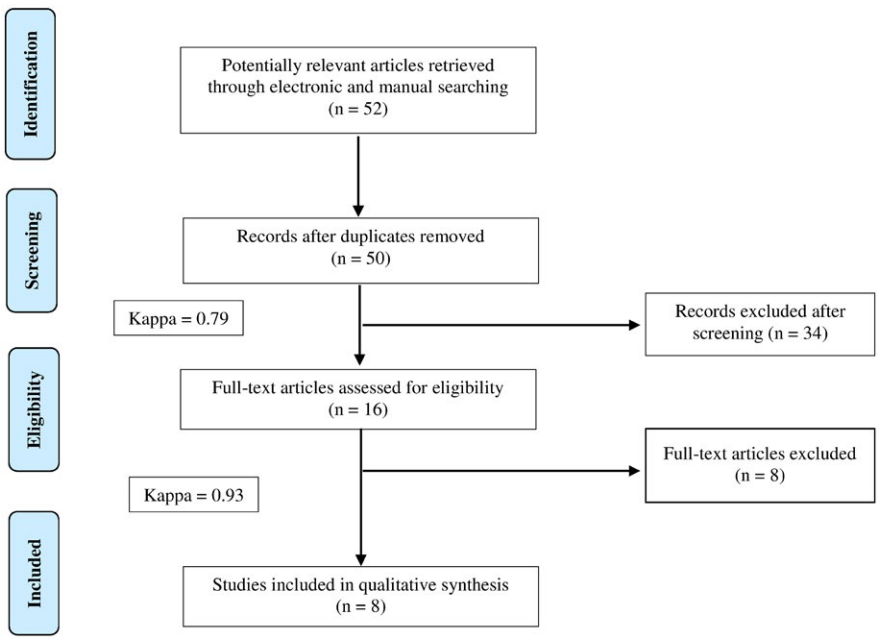

FIGURE 1 PRISMA flow diagram for studies retrieved through the searching and selection process

2.4 | Quality assessment

Quality assessment of the included studies was performed using Swedish Agency for Health Technology Assessment and Assessment of Social Services assessment tool.[23] With this tool, each article was assessed independently by the authors in six different sections. If the bias in a particular section was unclear, it was discussed with a third investigator, and a decision was reached as to whether the section should be classified as having a low, moderate or high risk of bias. The following criteria were used to determine the overall risk of bias: to get a low risk of bias in total, it was required low risk of bias in the majority of the sections (≥4 sections). The article was classified as a high risk of bias in total if half of the sections were rated "high risk of bias" (≥3 sections). The article was assessed as moderate risk of bias, if it did not reach the criteria for low or high risk of bias.

3 | RESULTS

3.1 | Study selection

A total of 12 studies in EMBASE, 19 studies in CINAHL, 8 studies in Cochrane Central Register of Controlled Trials and 17 studies in Cochrane Oral Health Group Trials Register were identified. The studies from these databases were matched with PubMed/MEDLINE. A total of 52 study titles and abstracts were identified in PubMed and considered. After removal of the duplicates, 50 articles were retrieved. Thirty-four records were excluded as irrelevant to the focus question (κ score at initial screening kappa = 0.79). A total of 16 papers were selected for full-text reading. Of these 16 studies, 8 studies were further excluded. After the final stage of selection, 8 studies[18,19,24-29] were included and processed for data extraction (κ score at full-text eligibility kappa = 0.93). Figure 1 shows the study identification flow chart according to PRISMA[22] with the reasons for exclusion of articles.

3.2 | General characteristics of included studies

Eight longitudinal prospective studies[18,19,24-29] were included in this review. These primary studies were carried out in Sweden[18,19,24,25,27,29], Italy[26] and Germany[28]. In all studies[18,19,24-29], number of children with DS ranged between 9 and 42. Altogether, 241 children with DS were included in these studies. Their mean age at the start of the study ranged between 2 months and 12 years. In all studies,[18,19,24-29] the follow-up period ranged from 5 to 58 months (Table 1).

The frequency of palatal plate therapy ranged between 1 and 3 times daily for 5 minutes to 120 minutes. The duration of OPPT ranged from 4 months to 48 months. The treatment effect was evaluated by clinical examination in 4 studies,[18,26,27,29] video recording/registration in 7 studies[18,19,24,25,27-29] and parental questionnaire in 3 studies.[19,27,29] The outcomes assessed in all the studies[18,19,24-29] included mouth closure, inactive protrusion, position of the tongue, lip activity, speech, oral parameters/motor function, facial expression, breathing mode and upper lip tone (Table 2).

Evaluation and assessment of the treatment effects were sought by speech and language pathologist in the studies by Carlstedt et al[19,24,25,27], whereas in the studies by Bäckman et al[18,29] and Zavaglia et al[26], the treatment effects were evaluated by speech and language therapist/phonetician and speech therapist, physiotherapist, otorhinolaryngologist and psychologist, respectively.

TABLE 1 General characteristics of the included studies

Author et al	Country	Study design	Total number of children with Down syndrome		Mean age of children at the start of study (months)	Follow-up (months)
			Test	Control		
Bäckman et al[29]	Sweden	Longitudinal prospective	42 DS + OPPT	31 DS + no plate + ST/PT	6	Up to 12-48
Bäckman et al[18]	Sweden	Longitudinal prospective	36 DS + OPPT	31 DS + no plate + ST/PT	6	Up to 12-48
Carlstedt et al[27]	Sweden	Longitudinal prospective	9 DS + OPPT	11 DS + no plate + ST/PT	24 ± 6	Up to 49-58
Carlstedt et al[19]	Sweden	Longitudinal prospective	9 DS + OPPT	11 DS + no plate + ST/PT	24 ± 6	Up to 49-58
Carlstedt et al[25]	Sweden	Longitudinal prospective	9 DS + OPPT	11 DS + no plate + ST/PT	24 ± 6	Up to 49-58
Carlstedt et al[24]	Sweden	Longitudinal prospective	14 DS + OPPT	15 DS + no plate + ST/PT	24 ± 6	Up to 12
Glatz-Noll and Berg[28]	Germany	Longitudinal prospective	24 DS + OPPT	19 DS + no plate + ST/PT	2-144	Up to 5-20 after finished treatment
Zavaglia et al[26]	Italy	Longitudinal prospective	38 DS + OPPT	30 DS + no plate + ST/PT	15	NA

OPPT, orthodontic palatal plate therapy; ST, speech therapy; PT, physiotherapy; NA, not available.

3.3 | Main outcomes

All studies[18,19,24-29] reported OPPT to be effective in improving orofacial disorders in children with DS at follow-up (Table 2). In the study by Bäckman et al,[29] children were assessed by clinical examination, video registration and parental questionnaire and reported that OPPT in combination with oral motor and sensory stimulation had a positive effect on oral motor performance in children with DS (66.7%) as compared to children with DS in control group (34.5%). In the study by Carlstedt et al,[24] the treatment effect was evaluated by video registration and showed that the OPPT group had significantly longer period of "closed mouth" ($P < .001$) and significantly shorter "inactive protrusion of the tongue" ($P < .001$) in children with DS. The active variables were diagnosed to constitute 81.0% ± 11.0% of the registered video time in the palatal plate group, compared with 68.2% ± 22.5% in the control group. The percentage of "inactive tongue protrusion" of total time decreased in both groups compared with the 1-year follow-up (palatal plate group, 3.9% ± 10.1%; control group, 8.8% ± 9.7%). This study[24] was longitudinally followed up in 2001 and 2003 and reported significantly shorter "inactive protrusion of the tongue" ($P < .01$)[19,25] and significantly more "closed mouth" during non-speech time ($P < .05$).[19]

In the follow-up study by Carlstedt et al,[27] the treatment effect was evaluated by a parental questionnaire and video recording after 49-58 months of treatment. The study[27] showed that the OPPT group had significantly less "inactive muscle function" even after 4 years and found no significant difference between the groups for "inactive protrusion of tongue" and "closed mouth" at follow-up. Almost 55% of the children in the OPPT group showed improved "tongue position" compared to 24% improvement in the control group.

The study by Glatz-Noll and Berg[28], who evaluated the treatment by video registration, showed 50% children with DS reached normalization of tongue function, without any improvement in mouth closure after OPPT treatment.

3.4 | Quality of the included studies

A total of five studies[18,19,24,25,27] had moderate risk of bias as the patients were randomized into control and treatment groups (Table 3). However, non-standardized methods were used, and the groups had large individual variations. Three studies[26,28,29] had high risk of bias as no information about dropout was reported, and the study groups had variations in study sample size.

4 | DISCUSSION

The present systematic review was based on the hypothesis that OPPT improves orofacial functions in children with DS. Overall, the studies[18,19,24-29] included in the present systematic review showed that OPPT showed significant improvement in the oral motor function including mouth closure, inactive protrusion and position of the tongue. This suggests that OPPT is a potential treatment strategy for the improvement of orofacial disorders in children with DS. However, it is important to interpret these findings with caution due to a number of factors.

Down syndrome is characterized by large individual variations with diverse dysmorphic features and clinical signs in varying individuals.[30,31] Some individuals with DS may show inconsistent features, while others may have multiple illnesses and severe cognitive disorders that could affect individuals with treatment by palatal plates.[32] From the studies included in this review, none of the studies report the degree of disfigurement or other variations among children with DS. Furthermore, the methods used to evaluate the treatment effect in the included studies consisted of clinical examination, video recording/registration and parental questionnaire. It is interesting to note that none of these methods are standardized. Clinical examination and video registration seem to be more reliable methods for assessing therapeutic progress, in contrast to parental interview that relies on subjective estimation from the guardians. These methods would significantly be more reliable if the examiners were trained, calibrated and blinded. Moreover, a standardized protocol for parental interview could be developed for parents that often tend to exaggerate the improvements in their own child. Therefore, it is essential to develop a precise, reproducible and reliable techniques for examination and assessment of orofacial function.

It is noteworthy from the studies[18,19,24-29] included that there was a significant heterogeneity in the mean age of children at the start of palatal plate therapy and treatment duration. It is well known that Castillo-Morales approach emphasizes on early treatment to achieve normal oral motor function in children with DS.[33] It can be noted from the studies that the mean age of children at the start of study ranged from 2 months to 12 years. Moreover, the duration of palatal plate therapy ranged from only 4 to 48 months. Therefore, it is difficult to interpret the findings for ideal treatment age and duration. Future studies should be performed with standardized treatment duration and standardized age group children to verify optimal time for initiating the therapy and its duration.

It seems that the effect of palatal plate therapy is nebulized by the additional physio/speech/orofacial regulation therapy that most of the included children got. Therefore, it is impossible to distinguish which is the effect of the one or the other.

Furthermore, different palatal plate designs were used, that is some with mobile pearls/beads, and some with fixed buttons, which may lead to further bias. It is indicated in several studies by Castillo Morales that in patients with DS, only fixed stimulating objects should be used. In the study by Bäckman et al,[18] up to approximately one-third of the children had problems in wearing the plates.

The following limitations should be taken into account when considering the conclusions of the present review: (i) approximately 75% of the included studies were from the same research group (Carlstedt et al[19,24,25,27] and Bäckman et al[18,29]). There is also a high probability that the children investigated in the study from Carlsted et al[19,24,25,27] probably were also the same with respect to patients group that may be considered a selection bias. (ii) Relatively small sample sizes with different age groups were noted from the included studies, ranging

TABLE 2 Treatment characteristics of the included studies

Author et al	Frequency of palatal plate therapy; minutes	Duration of palatal plate therapy (months)	Method for assessment	Outcomes assessed	Main outcomes compared with the control group
Bäckman et al[29]	2-3 times daily; 5-30 mins	12 ± 3	Clinical examination; video registration; parental questionnaire	Oral parameters, oral motor function, speech	Significant improvement in oral motor function in test group.
Bäckman et al[18]	2-3 times daily; ≤30 mins	42 ± 6	Clinical examination; video registration	Oral parameters, facial expression, speech	Significant improvement in oral motor function in test group.
Carlstedt et al[27]	2 times daily; 60 mins	48	Video recording; parental questionnaire	Articulation, oral motor function, communicative preferences	Significantly less "inactive muscle function" in the test group.
Carlstedt et al[19]	2 times daily; 60 mins	48	Video recording; parental questionnaire	Tongue position, lip activity, speech, articulation	Significant improvement in oral motor function in test group.
Carlstedt et al[25]	2 times daily; 60 mins	48	Clinical examination; video recording	Tongue position, lip activity, facial expression	Significant longer "closed mouth" and shorter "inactive protrusion of the tongue" in the test group.
Carlstedt et al[24]	2 times daily; 60 mins	12	Video recording	Mouth closure, tongue position and protrusion	Significant longer "closed mouth" and shorter "inactive protrusion of the tongue" in the test group.
Glatz-Noll and Berg[28]	≥2 times daily; 60-120 mins	4-11	Video registration	Mouth and lip closure, tongue position and protrusion	Significant improvement in oral motor function except mouth closure in test group.
Zavaglia et al[26]	Once daily and then 3 times daily; ≥60 mins	24	Clinical examination	Lip tone, mouth closure, tongue protrusion	Significant improvement in oral motor function in test group.

TABLE 3 Risk of bias of included studies

Author et al	Selection bias A1	Performance bias A2	Detection bias A3	Attrition bias A4	Reporting bias A5	Conflict of interest A6	Risk of bias (Summed)
Bäckman et al[29]							
Bäckman et al[18]							
Carlstedt et al[27]							
Carlstedt et al[19]							
Carlstedt et al[25]							
Carlstedt et al[24]							
Glatz-Noll and Berg[28]							
Zavaglia et al[26]							

, Low risk; , Moderate risk; , High risk.

9-42 participants per test/control group and 6-24 months, respectively, in only 3 European countries. (iii) The children in control group did not receive any palatal plate therapy. This may also have introduced bias and that palatal plate therapy had to be given without activation. (iv) Due to significant heterogeneity in the outcomes of data presented, we were unable to perform the meta-analysis. Although, the authors were contacted to obtain the numerical data, none of the authors responded. Furthermore, the presentation of outcomes significantly differed in the included studies; that is, some studies are presenting data in the form of box plots, some percentages, while others in the form of bar graphs. Most importantly, all the studies reported a positive effect of treatment, but there was no consensus regarding evaluation methods for the treatment with palatal plates, treatment times or which orofacial outcome variables that should be investigated. No meta-analysis was made due to this lack of consensus. (v) All of the studies[18,19,24-29] included had a high to moderate risk of bias with low to moderate quality studies. (vi) The present systematic review only considered studies in English language which may have resulted in publication bias with potentially relevant studies published in other language being missed.[34] (vii) The findings of the included studies have presented data with short-term follow-up. Future studies are warranted with long follow-up periods to investigate whether the effects obtained by palatal plate therapy persists in DS children. Therefore, these methodological shortcomings should be cautiously considered when interpreting the findings of the present study.

The outcomes of the present study indicate that all studies showed significant improvement in oral motor functions in children with DS. However, it must be noted that the effect of palatal plate therapy is achieved only in addition to physiotherapy, speech therapy and/or orofacial regulation therapy.

5 | CONCLUSION

- The effect of OPPT is achieved only in addition to physiotherapy, speech therapy and/or orofacial regulation therapy.
- Further longitudinal trials with standard evaluation methods, age of children for treatment initiation, treatment duration and standard orofacial variable outcomes are recommended.

CONFLICT OF INTEREST

The authors have stated explicitly that there is no conflict of interests in connection with this article.

ORCID

Z. Akram http://orcid.org/0000-0001-9618-8818

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