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REVIEW ARTICLE

Effectiveness of oral motor appliances on oral motor function and speech in children: a systematic review

Anna-Maria Pelkonen[a], Linnea Närhi[a,b], Sanna Häkli[b,c,d], Anna-Maria Raatikainen[b,c,d], Pertti Pirttiniemi[a,b] and Anna-Sofia Silvola[a,b]

[a]Research Unit of Population Health, University of Oulu, Oulu, Finland; [b]Medical Research Center Oulu, Oulu University Hospital, University of Oulu, Oulu, Finland; [c]Department of Otorhinolaryngology and Phoniatrics, Oulu University Hospital, Oulu, Finland; [d]PEDEGO Research Unit, University of Oulu, Oulu, Finland

ABSTRACT

Background: Different oral motor appliances have been used in connection with speech therapy to improve oral motor function and speech development, but no consensus has been reached on the effectiveness of the appliances. The objective was to systematically review the effectiveness of oral motor appliances on oral motor function and speech in children with speech sound disorders (SSDs) or oral motor dysfunctions.

Methods: A systematic search was conducted up to February 2023 in the PubMed, Scopus, and Cochrane databases. Inclusion criteria were prospective randomized or case-control clinical trials investigating the effect of intraoral appliances on orofacial function and/or speech. The risk of bias was evaluated by the Cochrane Collaboration's Robins-I tool.

Results: Nine publications of three individual studies met the inclusion and search criteria. Six of the publications were conducted in children with Down Syndrome (DS) and three publications were conducted in children with Cerebral Palsy (CP). No meta-analysis was made due to the limitations of the publications. Selected studies reported some beneficial effects of intraoral appliances on oral motor function in children with DS and CP, although the evidence is low. Due to the study design in selected studies and confounding factors, the overall risk of bias was categorized as moderate or high.

Discussion: Intraoral appliances may improve oral motor function in children with DS and CP. Due to lack of studies this review limited to children with DS and CP. The initial question concerning SSDs was not answered. Well-designed RCTs with larger sample sizes are needed, especially among non-syndromic children with SSDs. The level of evidence was considered very low.

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Introduction

Problems in oral motor function and speech sound disorders (SSDs) are prevalent in children. Oral motor problems and SSDs are often associated with each other [1–3]. The prevalence of SSDs in school-aged children has been reported to range between 2.3–24.6%, depending on study population and criteria [2,4,5]. Difficulties in oral sensory-motor skills may affect eating and saliva control, which can lead to social implications and other complications [1]. SSDs have a negative effect on self-esteem and self-confidence, and therefore, may also be considered as a public health problem [6].

Oral motor dysfunctions and SSDs often occur as a part of problems in general motor skills, being more prevalent in children with neurologic disorders and developmental disability [1,7,8]. Typical conditions requiring oral motor rehabilitation are Down syndrome (DS) and cerebral palsy (CP). Common orofacial features in both DS and CP are muscle hypotonicity, a protruding tongue and open mouth posture, causing difficulties in swallowing, eating, and speech [9,10].

Different oral motor appliances have been developed to improve oral motor function and articulation in connection with speech and language therapy [11–14]. The most often used custom-made oral motor appliances include palatal plates (a.k.a palatal training appliances) [13,15,16], Castillo-Morales plates [10,17–25], and the Innsbruck sensorimotor activator and regulator (ISMAR) [12,26–28].

The palatal plates used in oral motor appliance therapy typically contain movable pearl/pearls in the alveolar ridge or knots in the dentoalveolar or distopalatal part which stimulate tongue movements and improve articulation [13]. Palatal plate therapy includes a variety of exercises to increase awareness, to develop coordination and movements of the tongue, and to improve differentiation of the tongue and lower jaw. Castillo-Morales plates have been designed to stimulate the tongue and upper lip in children with DS [29]. They usually include a hollow cylinder at the dorsal edge to stimulate tongue retraction and a ridged frontal vestibular area to stimulate the upper lip [10]. ISMAR has been

CONTACT Anna-Maria Pelkonen anna.pelkonen@oulu.fi Research Unit of Population Health, University of Oulu, Oulu, Finland
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developed to achieve mandibular stability, better lip closure, tongue position, and swallowing in children with CP [12,30]. It has been modified from the Andresen monoblock activator by adding maxillary and mandibular vestibular pads and tongue shields [31]. Stimulating areas or beads may also be attached and can be modified during therapy. Oral motor appliances differ not only in their structure but also in their methods of use; palatal plates are usually used for shorter periods daily, while the ISMAR is mostly worn during the night [31,32].

In Nordic countries, speech therapy with oral motor appliances is typically coordinated by multidisciplinary teams including speech therapists, phoniatrists and dentists, who assess the need and type of appliances [13]. Despite the use of intraoral appliances in clinical practice, there is a lack of consensus of the effectiveness of palatal plate therapy or the use of other intraoral appliances. Especially custom-made appliances cause additional costs and require commitment from children and parents. A previous systematic review focusing on Down children concluded that palatal plate therapy (in connection with physiotherapy/orofacial regulation therapy) seems to be effective in improving orofacial disorders [33]. To date, no systematic review has also included non-syndromatic children and different intraoral appliance types.

The aim of this study was to systematically review the effectiveness of different oral motor appliances on oral motor function and speech in children with SSD or oral motor dysfunctions.

Materials and methods

Protocol and registration

The review protocol was written in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-analysis (PRISMA 2020) [34]. The systematic review was registered in an international database of prospectively registered systematic reviews (PROSPERO), and the ID number of this review is CRD42021230340 (https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=230340).

Eligibility criteria

The criteria for considering studies for this review were developed according to the PICOS framework.

The eligibility criteria were:

1. Population: children between ages 0–18 years with speech problems/oral motor dysfunctions/orofacial disorders
2. Intervention: trials investigating the effects of custom-made or prefabricated intraoral appliances
3. Control: case–control, comparison between different therapies
4. Outcome: improvement in orofacial function and/or speech (articulation, tongue, lip closure, drooling)
5. Study design: prospective controlled clinical trials and RCTs

The exclusion criteria in this systematic review were:

1. Case reports and case series involving the use of intraoral appliance in less than 8 participants
2. Any study not involving the use of intraoral appliance as a therapy
3. Studies using intraoral appliances or other orthodontic appliances primarily for orthodontic reasons
4. Retrospective studies and studies with no control group
5. Oral appliances having contact only with the lips (i.e., face former)
6. Children with clefts or obstructive sleep apnoea

Information sources and search strategy

The search strategy was designed, tested, and developed in collaboration with a medical information specialist. The searches were made in three electronic databases: MEDLINE (*via* PubMed), SCOPUS, and Cochrane Library. The search strategy was adapted to each database. The PubMed search included relevant MeSH terms. The SCOPUS search was performed using the standard search, and in the Cochrane search, the search manager was used. The language restrictions were English, Spanish, German, Swedish, and Finnish. Electronic database searches were limited to studies published from 1st January 1980 to 28th February 2023.

The search terms were divided into three categories and combined in the final searches.

1. The main search terms (A)
2. The search terms that describe the problem (B)
3. The search terms that describe treatment/appliance (C)

In all three databases, the same search phrases were used (Table 1). Relevant MeSH terms (Medical Subject Headings) were used in the PubMed and Cochrane Library searches (Figure 1).

Table 1. Search strategy using different databases.

	Search strategy
Database	Key words
PubMed/Medline	Oral motor *function* OR speech OR orofacial *function* OR dyspraxia* OR apraxia* OR chewing problem* OR chewing disorder* OR swallowing problem* OR swallowing disorder* OR dysarthria OR Down OR Cerebral palsy AND Palatal plate* OR palatal training appliance* OR stimulat* plate* OR prefabricated appliance* OR oral appliance* OR intraoral appliance* OR functional appliance* OR orofacial regulation therap* OR Castillo-Morales AND Child OR infant OR pediatric* OR paediatric* OR adolescent*
Scopus	(Similar to PubMed/Medline search)
Cochrane	(Similar to PubMed/Medline search)

"Speech", "Articulation Disorders", "Apraxias", "Mastication", "Dysarthria", "Down Syndrome", "Cerebral Palsy", "Myofunctional Therapy"

Figure 1. The MeSH terms used in the study.

Study selection

Two authors (A-M.P. and A-S.S.) screened independently the titles and abstracts according to predefined eligibility criteria, and the articles not meeting the eligibility criteria were excluded. Remaining articles were evaluated by full text, and the reasons for exclusion are presented in (Supplementary Table 1). Reference lists were also checked to identify additional relevant studies. Disagreements were solved by using a third investigator (P.P.). The PRISMA flow chart for study screening can be seen in Figure 2.

Data collection process and data items

Two authors (A-M.P. & A-S.S.) collected information independently from the included studies. Studies that did not include all required information were excluded (Figure 2). The following details were collected:

1. Study characteristics (authors, year of publication, country, study design)
2. Characteristics of participants (sample size, gender, age range, problems, patient group)
3. Type of intervention (therapy, appliance)
4. Evaluation characteristics (methods for assessment, follow-up time)
5. Main results concerning the original research question

Risk of bias

All included studies were assessed independently for risk of bias by two authors. In case of disagreement a third investigator (P.P.) was consulted. The risk of bias was evaluated using the Cochrane Collaboration's tool Robins I [35], and the included studies were categorized as 'low', 'moderaté or 'serious' (Figure 3).

Quality of evidence

The overall quality of evidence was rated using the Grades of Recommendations, Assessment, Development and Evaluation (GRADE) approach [37].

Results

Study characteristics

Nine publications pertaining to three unique studies were finally included in this systematic review:

1. Bäckman [38,39]
2. Carlstedt et al. [32,40–42]
3. Gisel [26,27] and Haberfellner [28]

All included studies were prospective controlled clinical trials with long follow-up. There was major variation in sample sizes ($n = 17 - 106$). The included trials were conducted in two countries: Sweden [32,38–42] and Canada [26–28]. Mean age at the baseline varied from ≤ 6 months to 10.2 ± 3.0 years. Key characteristics of the studies are summarized in Table 1 (children with DS) and Table 2 (children with CP).

Carlstedt et al. [32,40–42] used a group of age-matched children with DS as controls, and Bäckman et al. [38,39] utilized two different age-matched control groups (controls with DS and normally developed controls). In the studies of Gisel et al. [26,27] and Haberfellner [28], children with CP were divided into two groups (immediate treatment group and delayed onset treatment group acting as a control group).

Intervention

Intraoral appliances

Two types of intraoral appliances were used in the included studies: palatal plates in children with DS [32,38–42] and ISMAR treatment in children with CP [26–28] (Tables 2 and 3).

In the follow-up study of Bäckman et al. [38,39], the first plate (at 6–10 months of age) was used to stimulate a normalized position of the tongue and lips at rest. The second plate (at 10–14 months of age) was used to stimulate lip closure and movements of the tongue. The third plate (at 14–18 months of age) was used to stimulate lip closure and tongue retraction. The fourth plate (at 18–30 months of age) was designed to stabilize articulatory patterns of dento-alveolar consonant production and also had facial bows to improve lip closure. The last plate (at 30–48 months of age or until the children had outgrown the device) was made to stimulate lip closure and the dorsal parts of the tongue with a transversal steel wire. The instruction for the use of the plates was 2–3 times a day for periods of 5–30 min [38,39].

The palatal plates used in the follow-up study of Carlstedt [32, 40–42] had stimulating knobs, pearls, and bowls, and were modified depending on individual needs. There was a short interruption in the use of palatal plate during the most intense eruption of deciduous teeth because of retention problems. The parents had been instructed to make their children use the palatal plate for at least one hour twice a day [32,40–42].

In children with CP, in phase I (6 months), night-time wear of the ISMAR appliance aimed to stabilize the mandible. In phase II (6 months), the appliance was modified by adding

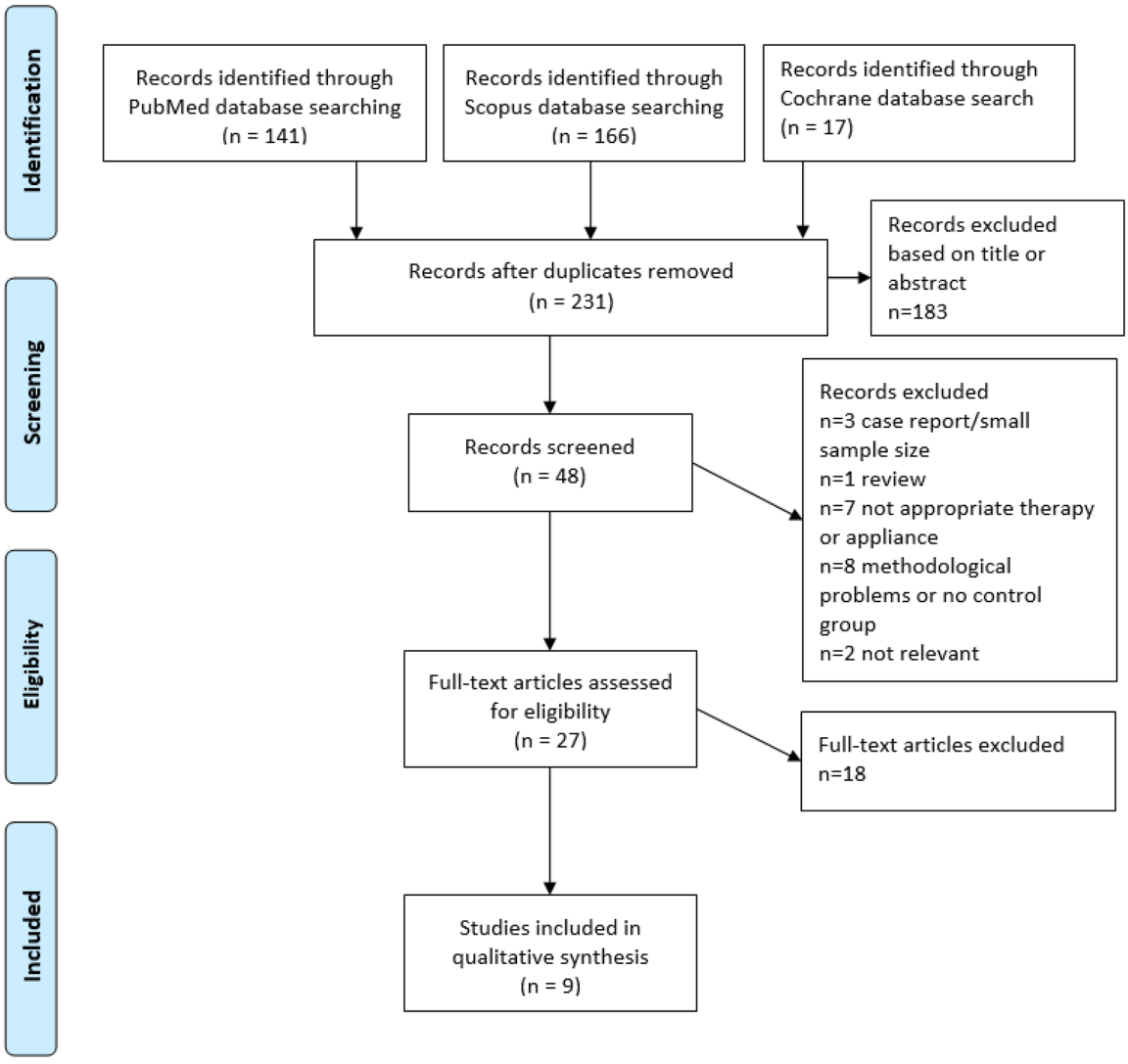

Figure 2. PRISMA flow diagram.

beads or drilled grooves to elicit tongue movements, aiming to facilitate ingestive skills [26,28]. Gisel et al. [27] continued to follow up these children for one year after 12 months of ISMAR therapy. One group continued to wear the intraoral appliance while the control group no longer wore the appliance.

Compliance

In all studies, the compliance of the children was evaluated by caregiver's logbook or parental questionnaire. In palatal plate therapy, most of the children had no or minor problems in compliance, the most frequent problems being palatal plate retention difficulties and recurrent infections [32,39]. In ISMAR therapy, the compliance varied significantly, the main problems being illnesses and other problems in daily life preventing the children from wearing the appliances for the required amount of time [27,28].

Other therapies

In children with DS, a combination therapy of palatal plate and speech and language intervention (oral motor and sensory stimulation) was used [32,38–42]. In the study of Carlstedt et al. [32,40–42], a special physiotherapy programme for orofacial stimulation was used from birth in the palatal plate and control groups. The details of other therapies were not reported.

In studies among children with CP, the delayed onset treatment group received standard oral motor rehabilitation at school during the first 6 months [26–28]. The content and amount of standard rehabilitation was not reported.

Data measurement

Video recordings were used for analysing orofacial muscle function or oral motor function [40–42], lip and tongue position [26,39] and speech [39]. The analyses based on video recordings in the included studies were done by a dentist, SLT [41] and phonetician [39]. Articulation assessments [42] were done by SLPs. Functional feeding skills in children with CP were observed by the child's regular feeder [27,28]. A parental questionnaire was used for asking caregiver's experiences of their child's oral motor capabilities [32,42] and sucking habits [38,39].

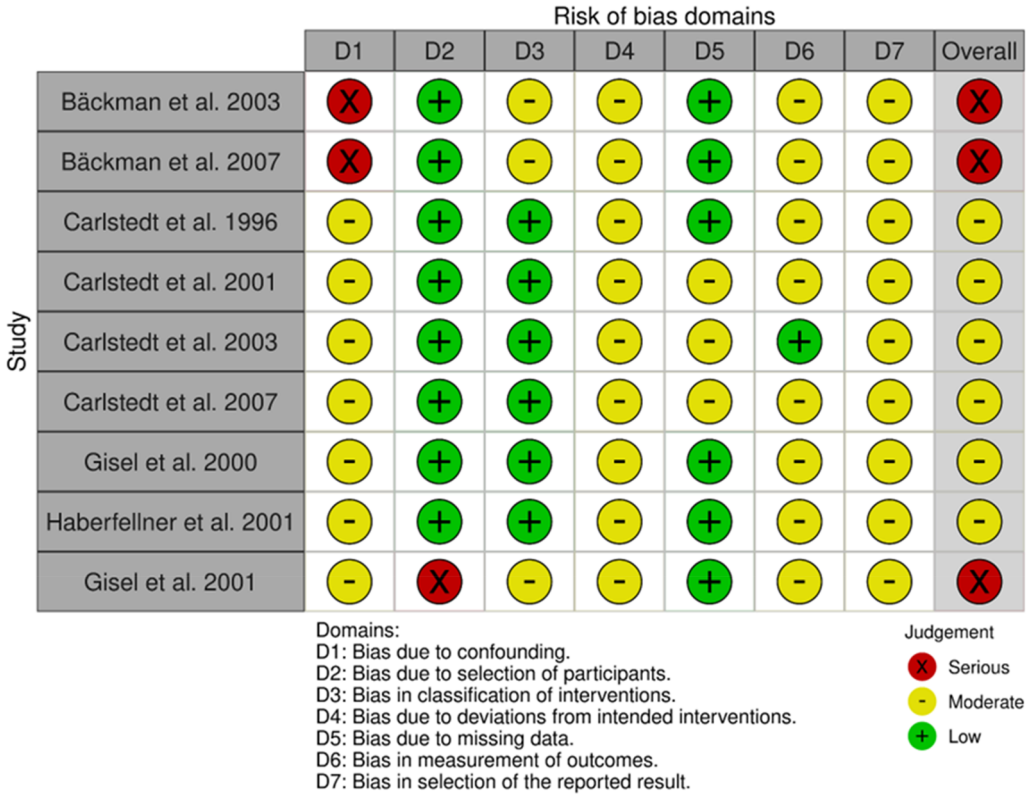

Figure 3. Risk of bias assessment using Robins I tool [36].

Table 2. Characteristics of the studies with children with down syndrome (DS).

References	Study design	Participants	Mean age at the baseline	Intervention	Follow-up
[39]	Controlled clinical study	SG: children with DS ($n=42$, 24 M, 18 F). CG: age-matched 1) children with DS ($n=33$, 20 M, 13 F) 2) children with normal development ($n=31$, 16 M, 15 F)	≤ 6 months	Palatal plate therapy with structured communication and speech training	12 months, from ≤ 6 m to 18 ± 3 m
[39]	Controlled clinical study	SG: children with DS ($n=37$, 20 M, 17 F) CG: age-matched 1) children with DS ($n=31$, 18 M, 13 F) 2) children with normal development ($n=36$, 21 M, 15 F)	≤ 6 months	Palatal plate therapy	3.5 years, from ≤ 6 m to 48 ± 6 m
[40]	Controlled clinical study	SG: children with DS ($n=14$, 10 M, 4 F) CG: age-matched children with DS ($n=15$, 6 M, 9 F)	24 ± 6 months	Palatal plate therapy	12 months
[40]	Controlled clinical study	SG: children with DS ($n=9$, 6 M, 3 F) CG: age-matched children with DS ($n=11$, 6 M, 5 F)	24 ± 6 months	Palatal plate therapy	4 years
[42]	Controlled clinical study	SG: children with DS ($n=9$, 6 M, 3 F) CG: age-matched children with DS ($n=11$, 6 M, 5 F)	24 ± 6 months	Palatal plate therapy	4 years
[32]	Controlled clinical study	SG: children with DS ($n=9$, 6 M, 3 F) CG: age-matched children with DS ($n=11$, 6 M, 5 F)	24 ± 6 months.	Palatal plate therapy	1 and 4 years

SG: study group; CG: control group; M: male; F: female.

Risk of bias across studies

Figure 3 shows the risk of bias of the included studies. Six studies [26,28,32,40–42] were classified as having a moderate risk of bias and three studies [27,38,39] a high risk of bias, confounding factors and selection of participants increasing the risk of bias. The most important confounding factors were SLT and other therapies, age, differences in developmental level and observation time. In the study of Gisel et al. [27], children changing study groups increased the risk of bias.

Effect of the interventions on outcomes

Children with down syndrome. The results of studies concerning children with DS are summarized in Table 4. In the studies of Bäckman et al. [38,39], the effect of palatal plates on oral motor function and speech were studied. After one-year

Table 3. The characteristics of the studies with children with cerebral palsy (CP).

References	Study design	Study groups	Mean age at the baseline	Intervention	Follow-up
[26]	Prospective longitudinal study	Children with CP (9 M, 11 F) *Immediate treatment group* (ISMAR treatment, $n = 10$) *Delayed onset treatment group* ($n = 10$)	8.3 ± 0.9 years	Intraoral appliance (ISMAR) on oral-motor, postural and ambulatory control	12 months
[28]	Prospective longitudinal study	Children with CP (9 M, 11 F) *Immediate treatment group ($n = 10$)Delayed onset treatment group ($n = 10$)*	8.3 ± 0.9 years	Intraoral appliance (ISMAR)	12 months
[27]	Prospective longitudinal study	Children with CP, who had received one year ISMAR treatment (7 M, 10 F) SG: ($n = 9$) continued to wear the appliance CG: ($n = 8$) no longer wear the appliance	10.2 ± 3.0 years (after 1st year of ISMAR treatment)	Intraoral appliance (ISMAR) on functional feeding skills and growth	12 months

Table 4. The results of the studies in children with down syndrome (DS).

References	Outcome assessed and measurement	Results
[38]	*Oral motor function and speech* (Video recordings)	Improvement in oral motor parameters and prerequisites of articulation
[39]	*Facial expression and speech* (Video recordings)	Improvement in facial expression and communicative capacity
[40]	*Orofacial muscle function* (Video recordings)	Improvement in mouth closure ($p < 0.001$) and tongue position ($p < 0.001$)
[41]	*Oral motor function* (Extraoral examination and video registration)	Improvement in mouth closure and tongue position ($p < 0.05$), and lip activity ($p < 0.01$).
[42]	*Oral motor function, facial expression, and articulation* (Clinical examination)	Improvement in lip activity and tongue position ($p < 0.01$), mouth closure ($p < 0.05$), decrease in visible tongue during non-speech periods ($p < 0.05$).
[39]	*Oral motor function* (Video recordings and parental questionnaire)	Improvement in mouth closure ($p < 0.001$) and tongue position ($p < 0.01$) after one year of therapy. Improvement in muscle function after one- and four-years follow-up ($p < 0.01$, $p < 0.05$, respectively)

Table 5. The results of the studies in children with cerebral palsy (CP).

References	Outcome assessed and measurement	Results
[26]	*Lip and tongue position* (Video recordings)	No statistically significant difference in lip or tongue position between groups
[28]	*Functional feeding skills* (Observation of the regular feeder)	In both groups, improvement in spoon feeding, biting, chewing, and cup-drinking during the first 6 months of therapy ($p = 0.0003$, $p = 0.047$, $p = 0.001$, $p = 0.015$), after 6 months only minor changes in immediate treatment group
[27]	*Functional feeding skills* (Observation of the regular feeder)	No significant differences in functional feeding skills between those who continued and those who discontinued the ISMAR therapy after one year of treatment

follow-up, the palatal plate therapy had a positive effect on oral motor parameters and prerequisites for articulation [38]. No statistical information of these findings was reported. After 3.5 years of follow-up, better communicative capacity and a higher score for facial expression was found in the study group compared to the control group with DS. In this study, there was no statistical analyses of the data concerning speech. The prevalence of sucking habits did not differ between the study group and the control group [39].

Carlstedt et al. [40] reported a positive effect on mouth closure and tongue position in the study group compared to the control group after one year of palatal plate therapy. Palatal plate therapy of four years was found to improve orofacial function, especially in terms of tongue position, lip activity, and mouth closure [32,41,42]. Children with palatal plate therapy had their mouths open significantly less often during non-speech time and had higher scores in facial expression compared to the control group. The palatal plate therapy was concluded to have a positive long-term effect on oral motor function [32,41,42].

Children with cerebral palsy. The results of the follow-up study in children with CP are presented in Table 5. No changes were found in tongue position during the 12-month ISMAR therapy [26]. Functional feeding skills improved significantly during the first 6 months, and improvement in chewing occurred also between 6 and 12 months of ISMAR therapy [28]. After 24 months of follow-up, there were no significant differences in functional feeding skills between the children who continued the use of ISMAR after 12 months and those whose ISMAR therapy was discontinued [27]. In both immediate and delayed onset treatment groups, at least half of the children showed clinically meaningful improvement in lip posture, although the finding was not statistically significant. During ISMAR therapy, noticeable changes occurred in facial expression [26].

Quality of evidence

Quality of available evidence regarding to the original study question was evaluated as very low using the GRADE rating.

There was lack of studies, risk of bias was considered moderate or high, sample sizes were generally low, and the statistical data was missing in the included studies.

Discussion

The present systematic review aimed to determine the effectiveness of oral motor appliances in improving speech articulation and/or oral motor function in children with oral motor dysfunctions. This review also aimed to investigate possible variety in treatment response between different oral appliances and patient groups (i.e. non-syndromatic children, DS, CP).

Most of the screened studies were conducted decades ago and their quality was considered low according to the present methodological demands. In the present review, only few studies met the inclusion criteria [26–28,32,38–42]. Included nine publications are derived from three major studies and all of them had syndromic children (DS, CP) as a study group. Unfortunately, no studies with a study population of non-syndromic children fulfilled the inclusion criteria. In addition, there were no high-quality studies that could have answered the initial research question about the responses of oral motor appliances on speech.

Investigating the efficacy of oral motor appliances can be complex due to the multifactorial entity of oral motor function. When studying this field, the importance of growth and development must be emphasized. Appropriate control groups are crucial when trying to separate the effects of therapies and the effect of normal development of children. Bäckman et al. [38,39] and Carlstedt et al. [32, 40–42] used age-matched control groups. Gisel et al. [26, 27] and Haberfellner [28] compared two groups with different timing of treatment. The weakness of the study of Gisel et al. [27] was that although the groups were randomized, some children switched from the study group to the control group and vice versa due to parent's wishes. In addition, the selection of children into the studies caused a selection bias in some studies. The caregivers had to provide consent before participation, causing the first selection of sample. It can be assumed that the selected caregivers had a favourable attitude towards the intervention, were motivated to adhere to the intervention and might have overrated the effects of therapies. Drop-out rates were rather high in some studies.

There were generally large age ranges and relatively small sample sizes in the included studies. The severity of the oral motor dysfunction varied from mild to severe. In addition, the cognitive level of the children could possibly influence the co-operation. Evaluation methods were also considered to increase the risk of bias. Most of the studies used video registrations; however, some of the assessors were aware of the children's intervention. Also, self-reporting by the caregivers may not be considered an objective measure.

The limitations of the included studies also included insufficient reporting of other therapies. In general, syndromic children receive different kinds of therapies (physical and occupational therapy, SLT) which can affect oral motor function as well. It should also be noted that in the study of Carlstedt et al. [32,40–42], the children represented different stages of development according to chronological age. All included studies used an oral motor appliance combined with other therapies. There is also a lack of knowledge of the optimal time and duration of treatment. Some references recommend early intervention when children have better ability to adapt to the use of oral appliances [41], and positive treatment results are found to be more significant after one year of early treatment [32]. These positive impacts on orofacial muscle function also have an influence on children's appearance, which is meaningful for the parents at a psychological level.

In the included studies, some positive effects of oral motor appliance combined with SLT on oral motor function were observed. The results of the long follow-up studies suggest that the beneficial effects of palatal plate therapy on oral motor function may be long-term [41]. In children with CP, ISMAR therapy seems to be beneficial in terms of its impact on feeding skills and lip posture [26–28]. Only part of the children were able to communicate either verbally or through augmentative and alternative communication; therefore, evaluating articulation may not be relevant.

In addition to the included studies, some retrospective studies have been conducted among syndromic and non-syndromic children. Those studies have reported positive effects of oral motor appliances in children with DS, i.e. improved orofacial appearance, tongue position and faster initial speech development (i.a. [21–23,43,44]). The studies with children with CP concluded that the oral motor therapy may have positive effects on drooling by enhancing the function of the lips and tongue, chewing, and swallowing and speech intelligibility of words [12 14,17,19,45]. Retrospective studies in non-syndromic children indicate beneficial effects of oral motor appliance in oral motor function, tongue position, and mouth closure [46,47].

In the Nordic countries, palatal plate therapy has been used in children with SSDs to improve articulation and oral motor function [13,15,16]. In a retrospective study among children with mild or moderate problems in speech articulation or oral motor skills, palatal plate therapy improved speech articulation and tongue movements in half of the children [13]. According to Haapanen et al. [15], oral motor appliance may speed up and enhance the treatment of uncomplicated speech and language difficulties. In children with severe oral dysfunctions, palatal plate training seems to improve speech articulation more effectively than speech therapy alone [16].

The strength of the present systematic review was that it screened a large variety of studies, not only those with children with certain syndromes. This systematic review used standardized protocols and tools. The limitation of this review is that due to lack of studies, it could not answer the initial question concerning non-syndromic children. There was heterogeneity between the studies in the type of intraoral appliance used, the use of the appliance, and treatment schedule. Because of the large variety of outcome assessments used, unstandardized methods, differences in treatment times and measuring variables and a low number of included studies, a meta-analysis could not be performed.

Studies were conducted in only two countries (Sweden and Canada) which must be noted when generalizing the results.

Cost-effectiveness must be taken into consideration when making recommendations or planning therapy protocols. In all included studies, palatal plates were used in connection with speech and language or oral motor therapy. However, long-lasting appliance therapies require commitment and resources. It must be emphasized that caregivers may feel the burden of different therapies of children with special needs. On the other hand, the use of oral motor appliance may also have effects on cost-effectiveness by shortening the time of SLT. For these reasons, the benefits and disadvantages of the recommended therapies must be carefully considered. Limited resources underline the importance of the efficiency of rehabilitation. In the future, health care funders are expected to demand increased evidence of cost efficiency.

In general, quality of evidence was considered very low. No exact recommendations can be made based on the results of the present systematic review due to the lack of high-quality studies. The use of oral motor appliances may have a role in clinical practice in well-selected patient groups. All the included studies were conducted 10–30 years ago, and it seems that there has not been continuous research interest in this area. There is still a need for high-quality research in different study populations (syndromic and non-syndromic patients) to get clinical recommendations of the use of oral motor appliances. The future studies should be well designed and methodologically adequate, with larger sample sizes and RCT study design.

Conclusion

Intraoral appliances seem to have beneficial effects on oral motor function in connection with speech therapy in children with DS and CP. Overall, the quality of evidence was very low. Well-designed RCTs are needed to achieve evidence of the effectiveness of palatal plate therapy in children with SSDs.

Disclosure statement

The authors report no conflict of interest.

References

[1] Mogren Å, Sjögreen L, Barr Agholme M, et al. Orofacial function in children with speech sound disorders persisting after the age of six years. Int J Speech Lang Pathol. 2020;22(5):526–536. doi: 10.1080/17549507.2019.1701081.

[2] Shriberg LD, Tomblin JB, McSweeny JL. Prevalence of speech delay in 6-year-old children and comorbidity with language impairment. J Speech Lang Hear Res. 1999;42(6):1461–1481. doi: 10.1044/jslhr.4206.1461.

[3] Wren Y, Miller LL, Peters TJ, et al. Prevalence and predictors of persistent speech sound disorder at eight years old: findings from a population cohort study. J Speech Lang Hear Res. 2016;59(4):647–673. doi: 10.1044/2015_JSLHR-S-14-0282.

[4] Law J, Boyle J, Harris F, et al. Prevalence and natural history of primary speech and language delay: findings from a systematic review of the literature. Int J Lang Comm Dis. 2000;35(2):165–188.

[5] Qvarnström MJ, Laine MT, Jaroma SM. Prevalence of articulatory disorders of different sounds in a group of finnish first-graders. J Commun Disord. 1991;24(5-6):381–392. doi: 10.1016/0021-9924(91)90010-g.

[6] Lewis BA, Freebairn L, Tag J, et al. Adolescent outcomes of children with early speech sound disorders with and without language impairment. Am J Speech Lang Pathol. 2015;24(2):150–163. doi: 10.1044/2014_AJSLP-14-0075.

[7] Bishop DVM. Motor immaturity and specific speech and language impairment: evidence for a common genetic basis. Am J Med Genet. 2002;114(1):56–63. doi: 10.1002/ajmg.1630.

[8] Kaczorowska N, Kaczorowski K, Laskowska J, et al. Down syndrome as a cause of abnormalities in the craniofacial region: a systematic literature review. Adv Clin Exp Med. 2019;28(11):1587–1592. doi: 10.17219/acem/112785.

[9] Kiran E, Üstüner Atik G, Dursun N, et al. Serebral palsīlī hastalarda dīl ve konuşma bozukluklari [speech and language disorders in patients with cerebral palsy]. J Rheumatol Med Rehab. 2004;15(3):166–173. Turkish.

[10] Limbrock GJ, Castillo-Morales R, Hoyer H, et al. The Castillo-Morales approach to orofacial pathology in down syndrome. Int J Orofacial Myology. 1993;19(1):30–38. doi: 10.52010/ijom.1993.19.1.6.

[11] Castillo-Morales R. Die orofaziale regulationstherapie [orofacial regulation therapy]. München: pflaum; 1998. German.

[12] Johnson H, Reid S, Hazard C, et al. Effectiveness of the innsbruck sensorimotor activator and regulator in improving saliva control in children with cerebral palsy. Dev Med Child Neurol. 2004;46(1):39–45. doi: 10.1111/j.1469-8749.2004.tb00432.x.

[13] Koskimies M, Pahkala R, Myllykangas R. Palatal training appliances in children with mild to moderate oral dysfunctions. J Clin Pediatr Dent. 2011;36(2):149–153. doi: 10.17796/jcpd.36.2.37532478138 03702.

[14] Limbrock GJ, Hoyer H, Scheying H. Drooling, chewing and swallowing dysfunctions in children with cerebral palsy: treatment according to Castillo-Morales. ASDC J Dent Child. 1990;57(6):445–451.

[15] Haapanen ML. Suunsisäisen irtokojeen käytön aiheet ja hoitovaste puhehäiriöissä [reasons for using an intraoral removable appliance and response to treatment in speech disorders]. Suom Lääkäril. 2003;58(39):3877–3881. Finnish.

[16] Toivanen S, Raveikko L, Qvarnström M, et al. ORA-kojeet puheterapian apuna [ORA devices as a speech therapy aid]. Suom Hammaslaakaril. 2013;20(7):22–27.

[17] Avalle C, Fischer-Brandies H, Schmid R. Orofacial dysfunctions and its therapy in cerebral palsy – preliminary report on the treatment according to Castillo-Morales. Monatsschrift Kinderheilkunde. 1986;134(1):32–36.

[18] Alacam A, Kolcuoğlu N. Effects of two types of appliances on orofacial dysfunctions of disabled children. Br J Dev Disabil. 2007;53(105):111–123. doi: 10.1179/096979507799103397.

[19] Fischer-Brandies H, Avalle C, Limbrock GJ. Therapy of orofacial dysfunctions in cerebral palsy according to Castillo-Morales: first results of a new treatment concept. Eur J Orthod. 1987;9(2):139–143. doi: 10.1093/ejo/9.2.139.

[20] Gerek M, Ciyiltepe M. Yutma bozukluklarının rehabilitasyon yöntemleri ile tedavisi [the management of swallowing disorders through rehabilitation methods]. KBB. 2005;14(1-2):10–17. Turkish.

[21] Glatz-Noll E, Berg R. Oral dysfunction in children with downs' syndrome: an evaluation of treatment effects by means of video-registration. Eur J Orthod. 1991;13(6):446–451. doi: 10.1093/ejo/13.6.446.

[22] Hohoff A, Ehmer U. Effects of the Castillo-Morales stimulating plate on speech development of children with down's syndrome. A retrospective study. J Orofac Orthop. 1997;58(6):330–339. doi: 10.1007/BF02682673.

[23] Korbmacher H, Limbrock J, Kahl-Nieke B. Long-term evaluation of orofacial function in children with down syndrome after treatment with

a stimulating plate according to castillo morales. J Clin Pediatr Dent. 2006;30(4):325–328. doi: 10.17796/jcpd.30.4.60q6841412763771.

[24] Marinone S, Gaynor W, Johnston J, et al. Castillo morales appliance therapy in the treatment of drooling children. Int J Pediatr Otorhinolaryngol. 2017;103:129–132. doi: 10.1016/j.ijporl.2017.10.020.

[25] 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. 2001;62(4):255–263. doi: 10.1007/pl00001933.

[26] Gisel EG, Schwartz S, Petryk A, et al. Whole body" mobility after one year of intraoral appliance therapy in children with cerebral palsy and moderate eating impairment. Dysphagia. 2000;15(4):226–235. doi: 10.1007/s004550000032.

[27] Gisel EG, Haberfellner H, Schwartz S. Impact of oral appliance therapy: are oral skills and growth maintained one year after termination of therapy? Dysphagia. 2001;16(4):296–307. doi: 10.1007/s00455-001-0088-2.

[28] Haberfellner H, Schwartz S, Gisel EG. Feeding skills and growth after one year of intraoral appliance therapy in moderately dysphagic children with cerebral palsy. Dysphagia. 2001;16(2):83–96. doi: 10.1007/PL00021293.

[29] Castillo-Morales R, Grotti E, Avalle C, et al. Orofaziale regulation beim down-Syndrom durch gaumenplatte [orofacial regulation in down syndrome by palatal plate]. Sozialpadiatrie. 1982;4:10–17. German.

[30] Bavikatte G, Sit PL, Hassoon A. Management of drooling of saliva. Br J Med Pract. 2012;5:507.

[31] Gisel EG, Schwartz S, Haberfellner H. The innsbruck sensorimotor activator and regulator (ISMAR): construction of an intraoral appliance to facilitate ingestive functions. ASDC J Dent Child. 1999;66(3):180–187, 154.

[32] Carlstedt K, Henningsson G, Dahllof G. A longitudinal study of palatal plate therapy in children with down syndrome. Effect Mot Funct. 2007;8(1):13.

[33] Javed F, Akram Z, Barillas AP, 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):20–26. doi: 10.1111/ocr.12211.

[34] Page MJ, McKenzie JE, Bossuyt PM, et al. The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. BMJ. 2021;372:n71.

[35] Sterne JAC, Hernán MA, Reeves BC, et al. ROBINS-I: a tool for assessing risk of bias in non-randomized studies of interventions. BMJ. 2016;355:i4919.

[36] McGuinness LA, Higgins JPT. Risk-of-bias VISualization (robvis): an R package and shiny web app for visualizing risk-of-bias assessments. Res Syn Meth. 2021;12(1):55–61. doi: 10.1002/jrsm.1411.

[37] Schunemann HJ, Brennan S, Akl EA, et al. The development methods of official GRADE articles and requirements for claiming the use of GRADE - a statement by the GRADE guidance group. J Clin Epidemiol. 2023;159:79–84. doi: 10.1016/j.jclinepi.2023.05.010.

[38] Bäckman B, Grevér-Sjölander AC, Holm AK, et al. Children with down syndrome: oral development and morphology after use of palatal plates between 6 and 18 months of age. Int J Paediatr Dent. 2003;13(5):327–335. doi: 10.1046/j.1365-263x.2003.00485.x.

[39] Bäckman B, Grevér-Sjölander AC, Bengtsson K, et al. 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(1):19–28. doi: 10.1111/j.1365-263X.2006.00781.x.

[40] Carlstedt K, Dahllöf G, Nilsson B, et al. Effect of palatal plate therapy in children with down syndrome a 1-year study. Acta Odontol Scand. 1996;54(2):122–125. doi: 10.3109/00016359609006017.

[41] Carlstedt K, Henningsson G, McAllister A, et al. 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):63–68. doi: 10.1080/000163501750157117.

[42] 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. doi: 10.1080/ode.61.1.39.46.

[43] Hohoff A, Ehmer U. Short-term and long-term results after early treatment with the castillo morales stimulating plate. J Orofac Orthop. 1999;60(1):2–12. doi: 10.1007/BF01358711.

[44] Fischer-Brandies H, Avalle C, Stahl A. Die auswirkung der systematischen kieferorthopädischen frühbehandlung bei trisomie 21 [the impact of systematic early orthodontic treatment in trisomy 21]. Fortschr Kieferorthop. 1988;49(3):252–262. German. doi: 10.1007/BF02164448.

[45] Ray J; Washington State University Department of Speech & Hearing Services. Functional outcomes of orofacial myofunctional therapy in children with cerebral palsy. Int J Orof Myol. 2001;27(1):5–17. doi: 10.52010/ijom.2001.27.1.1.

[46] Korbmacher H, Limbrock J, Kahl-Nieke B. Orofacial development in children with down's syndrome 12 years after early intervention with a stimulating plate. J Orofac Orthop. 2004;65(1):60–73. doi: 10.1007/s00056-004-0229-y.

[47] McAllister A. Voice disorders in children with oral motor dysfunction: perceptual evaluation pre and post oral motor therapy. Log Phon Vocol. 2003;28(3):117–125. doi: 10.1080/4302002000014.