

Case Report

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Early oral functional therapy in Down syndrome: Need for evidence

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Abstract

Early oral functional therapy in toddlers with Down Syndrome was first described by Castillo Morales in the 1970s. Management of facial manifestations remains essential to gain social integration of these children. Oral functional therapy by palatal plates is a complement to the early multidisciplinary management by speech therapists and physiotherapists. Different recent reviews suggested that palatal plates seem to be efficient on oral motor function, but due to low quality of the evidence, they have highlighted the real need for evidence of this therapy. The aim of this short commentary is to address key points of methods and suggest leads for further in-depths studies.

Keywords: Down syndrome; Stimulation; Physiotherapist; Dentist; Palatal plate.

Introduction

Down Syndrome (DS), also known as trisomy 21, is the most common chromosomal disorder. Its prevalence at birth is estimated between 1/650 and 1/1000 worldwide [1]. Life expectancy of individuals with Down Syndrome has tremendously increased in the past decades, as their quality of life, by dint of progress in medical care and prevention. In addition to cognitive and developmental impairments, children with Down Syndrome have general and orofacial hypotonia, characterized by labial hypotonia, labial open bite and low tongue position often protruding, as synthesized in a recent review [2]. Oral ventilation and muscle dysfunction result in maxilla hypodevelopment and mandible prognathia [3]. Drooling may also occur in some individuals due to lack of lip tone. In babies, early multidisciplinary care including physiotherapist and speech therapist helps children with Down Syndrome overcome general hypotonia and

more specifically orofacial hypotonia and aims to improve overall development and limit the onset of specific dysfunctions related to the syndrome. Functional orofacial therapy is a part of the early process of therapeutic education. It is conducted by the dentist, with significant parental involvement, and includes palatal plates, as described by Castillo Morales [4]. Two recent reviews aimed to assess the effect of palatal plates on oral function, one was based on studies among children aged up to 12 years and the second one related to DS and cerebral palsy [5,6]. Both reviews pointed out the methodological shortcomings of the studies which made it difficult to draw conclusions. Moreover, the effect of the early orofacial functional therapy, which focuses on babies and includes palatal plates as well as massages, as Castillo Morales described it, has not been specifically studied. Some recent studies, not included in the reviews, were also inconclusive [7,8].

Recent publications from different countries highlight the fact that early functional therapy using palatal plates is a technique probably used worldwide, and that researchers are struggling to carry out high-quality studies to demonstrate its benefits [2,5-8]. There are therefore methodological barriers to conducting such studies that have never been mentioned or discussed.

The aim of this paper is (1) to expose and discuss the methodological barriers to high-quality studies of the early orofacial functional therapy in babies with DS, and (2) to suggest concrete leads for further studies.

Early oral functional therapy

Children with Down Syndrome have reduced extra- and intra-oral proprioception, vague localization of stimuli, and altered pain expression [9]. In the 1970's, Castillo-Morales described the myofunctional therapy using external and internal orofacial stimulation to help the babies with DS become aware of their orofacial region, including the ora-facial cavity. External stimulation of the face and neck muscles consists in specific anatomical point pressures followed by massage [4,10]. Mechanical internal stimulation is achieved through a palatal plate designed by the dentist. By stimulation, the palatal plate aims to reduce labial hypotonia, decrease lingual protrusion and stimulate mouth closure, which could help prevent respiratory infections favored by a continually open mouth. It also aims to stimulate the lingual mobility, enhance its high position, enabling correct development of the maxilla (transverse expansion) and limiting the onset of a mandibular prognathia.

The palatal plate can be performed as early as 3 months of age [4]. The studies suggest that the earlier the palatal plate therapy begins, the better. The goal is to intervene as early as possible before teeth erupt, before phonation and mastication are established, and before malocclusion appears. Thus, mastication and phonation could set up in the best conditions without disorder. In order to vary the type of stimulation and follow the growth, the plate should be renewed every 3 months as previously suggested in a multicentre, multidisciplinary project in Sweden [11]. The resin plate includes several stimulation elements: anterior labial stimulators such as buccal pads and metal arches with resin beads and lingual stimulators to make the child put the tongue on the palate.

Babies being edentulous, plate retention is achieved by a suction effect. When the primary teeth erupt, plates lose their retention. Thus, plates need to be adjusted and sometimes the treatment is interrupted until molars erupt [11]. When primary teeth have erupted, treatment may consist in a stimulation plate as previously described with a median ram added to support growth.

There is no consensus in the literature about the rhythm and length of daily use of the plates. Previous published papers mention using the plates at least two to four times a day with stimulation sessions lasting 5 minutes to 1 or 2 hours [11-13]. Stimulation should be done when the child is awake and supervised by one of his parents or caregivers.

Previous studies and reviews

The lacks and weaknesses of the studies [13-17] could be grouped in 2 categories: study design and outcomes.

Study design

The study design did not involve a comparable control group in all studies. In one study, there was none [16]; in another, the children in the control group had only mild oral dysfunction while those in the "plate" group had severe oral dysfunction [15], thus leading to a high indication bias. Some studies included children from general population in the control group [4,18], which make the results difficult to interpret.

Javed et al. concluded their review suggesting trials for a proper assessment of the therapy [5] and Pelkonen et al. recommended longitudinal recent studies [6]. Trials seem to be difficult to carry out, for ethical reasons. It would be indeed difficult not to suggest the orofacial therapy to the children who come for consultation. Observational studies seem therefore more appropriate than trials to assess the effect of early orofacial functional therapy. However, the comparison with a control group is crucial. Children eligible for the control group should not have received early orofacial therapy but should be as comparable as possible to the therapy group: same syndrome and same age at the time of assessment of oral motor function. In previous studies, children's age at inclusion ranged from 2 months to 12 years [5], which is very broad for assessing the benefit of early oral functional therapy. Some children are delayed in being taken into care for various reasons, such as difficulties in accessing special dental care. Children in the control group could be older at the time of the first consultation. We suggest an assessment the outcomes when the temporary dentition is stable (after plates for those treated and before plates for those not yet treated), followed by subsequent assessments to monitor the evolution of oral motor function. According to previous studies and our clinical experience, when babies are treated with palatal plates, they are also cared for by physiotherapists and speech therapists, as recommended by Castillo-Morales. It is probably not possible to study the effect of plates alone. Thus, children receiving an early orofacial functional therapy, i.e., including physiotherapy, speech therapy and plates within the child's first year, should be compared with children receiving early physiotherapy and speech therapy but no early plate, and with children not receiving physiotherapy and speech therapy in the first year.

In addition, the number of children included (9 to 36) was very small and could not represent the heterogeneity of the Down Syndrome population.

Outcomes

The outcomes used to assess the benefit of oral functional therapy were mouth closure, tongue protrusion or/and position, speech, and facial expression. Assessment methods are heterogeneous, some relying on clinical examination and some on video recordings, as in one recent case report, where the length of time the child's lips are open, closed or semi-closed were measured. It appears, however, that while authors of all studies struggle with the assessment method, a consensus seems to be emerging on the main outcomes, which should be

lip closure and tongue posture. We suggest considering a simple assessment by the clinicians (lip closure and tongue posture) during the consultation. To limit the subjectivity of the method, it should be carried out by 2 clinicians who could compare their opinion at the end of the consultation. This could also be based on a video recording. As a secondary outcome, parents' appraisal of their child's oral motor function should be considered. It would also be relevant to assess the child's cooperation in accepting the plates throughout early orofacial functional therapy, as well as the impact of the therapy itself on the child's behavior in the dental chair, and even on his or her cooperation in tooth brushing by parents.

Conclusion

Management of facial manifestations remains essential to gain social integration of children with Down Syndrome. Oral functional therapy by palatal plates is a complement to the early multidisciplinary management by speech therapists and physiotherapists. High-quality studies on the benefits of this therapy and specifically the impact of palatal plates within the overall management would be tremendously useful. It would enable this comprehensive care to be promoted to dentists, orthodontists, pediatricians, and new parents of babies with DS. This paper addressed key points of methods enabling researchers to demonstrate the benefits of early functional therapy.

Declarations

Disclaimers: None.

Author contribution statement: All authors drafted or critically revised the manuscript. All authors approved of the final version of the manuscript.

Conflict of interest. None.

References

1. Roizen NJ, Patterson D. Down's syndrome. *Lancet*. 2003; 361: 1281-9.
2. Kaczorowska N, Kaczorowski K, Laskowska J, Mikulewicz M. Down syndrome as a cause of abnormalities in the craniofacial region: A systematic literature review. *Adv Clin Exp Med*. 2019; 28: 1587-92.
3. Sixou JL. Oral cavity in Down syndrome. *Arch Pediatr*. 2008; 15: 852-4.
4. Limbrock GJ, Castillo-Morales R, Hoyer H, Stöver B, Onufer CN. The Castillo-Morales approach to orofacial pathology in Down syndrome. *Int J Orofacial Myology*. 1993; 19: 30-7.
5. Javed F, Akram Z, Barillas AP, Kellesarian SV, Ahmed HB, et al. Outcome of orthodontic palatal plate therapy for orofacial dysfunction in children with Down syndrome: A systematic review. *Orthod Craniofac Res*. févr 2018; 21: 20-6.
6. Pelkonen AM, Närhi L, Häkli S, Raatikainen AM, Pirttiniemi P, et al. Effectiveness of oral motor appliances on oral motor function and speech in children: A systematic review. *Acta Odontol Scand*. 2023; 1-9.
7. Xepapadeas AB, Weise C, Frank K, Spintzyk S, Poets CF, et al. Technical note on introducing a digital workflow for newborns with craniofacial anomalies based on intraoral scans - part I: 3D printed and milled palatal stimulation plate for trisomy 21. *BMC Oral Health*. 2020; 20: 20.
8. Ferreira JE de A, Almeida BRS de, Deps TD, Pretti H, Furlan RMMM. Orofacial myofunctional therapy associated with the use of the stimulating palatal plate in children with trisomy 21: Case studies. *Codas*. 2023; 35: e20210231.
9. Hennequin M, Morin C, Feine JS. Pain expression and stimulus localisation in individuals with Down's syndrome. *Lancet*. 2000; 356: 1882-7.
10. Hoyer H, Limbrock GJ. Orofacial regulation therapy in children with Down syndrome, using the methods and appliances of Castillo-Morales. *ASDC J Dent Child*. 1990; 57: 442-4.
11. Bäckman B, Grevér-Sjölander AC, Holm AK, Johansson I. Children with Down Syndrome: Oral development and morphology after use of palatal plates between 6 and 18 months of age. *Int J Paediatr Dent*. sept 2003; 13: 327-35.
12. 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: 39-46.
13. Carlstedt K, Henningsson G, Dahllöf G. A longitudinal study of palatal plate therapy in children with down syndrome. Effects on motor function. *Journal of Disability and Oral Health*. 2007; 8: 13.
14. Bäckman B, Grevér-Sjölander AC, Bengtsson K, Persson J, Johansson I. Children with Down syndrome: Oral development and morphology after use of palatal plates between 6 and 48 months of age. *Int J Paediatr Dent*. 2007; 17: 19-28.
15. 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: 255-63.
16. 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: 60-73.
17. Zavaglia V, Nori A, Mansour NM. Long term effects of the palatal plate therapy for the orofacial regulation in children with Down syndrome. *J Clin Pediatr Dent*. 2003; 28: 89-93.
18. Walasz J, Matthews-Brzozowska T, Matthews-Kozanecka M, Cudzilo D. Types and positioning of palatal plate stimulation elements in children with Down syndrome. *Jour of Med Sc & Tech*. 2014; 1-6.