

Long-term evaluation of orofacial function in children with Down syndrome after treatment with a stimulating plate according to Castillo Morales

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The aim of this investigation was to evaluate the long-term orofacial development of Down children who received plate therapy according to Castillo Morales in their early childhood. The orofacial development of 27 Down children was documented before and after plate therapy and at a follow-up examination 13 years \pm 6 months after initiation of therapy. The orofacial appearance significantly improved during therapy ($p=0.00$). During the follow-up, mouth posture remained stable ($p=0.259$), whereas tongue position further improved ($p=0.034$). A better long-term development was documented in children with initial severe orofacial dysfunctions.

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INTRODUCTION

The general muscle hypotonia in children with Down syndrome affects the orofacial appearance and development. Characteristic orofacial features encompass a reduced orofacial and masticatory muscle tonus with an open mouth posture and an active, protrusive, often extraoral tongue position, the hypotonic ligamentary apparatus of the temporomandibular joint combined with an underdevelopment of the nasomaxillary complex. These orofacial dysfunctions are described as primary pathologies.¹ In the case of their persisting, secondary pathologies such as drooling, dental and skeletal malocclusions, airway infections, retarded and decreased bite function and the development of oral stereotypes establish and impede the physiological development. In a sense, abnormal oral motor functions negatively affect speech and mastication.¹

In the seventies, Castillo Morales developed the "orofacial regulation therapy" in order to improve the facial appearance during early childhood.² This interdisciplinary therapy approach consists of functional exercises and a specific manual program of neuromuscular stimulation based on the different motor zones on body and

face.^{3,4} By stimulating these zones with defined directions of pressure, traction and/ or vibration a higher orofacial muscle tonicity and activity can be established. For a daily exercise, parents are taught to conduct a small program of orofacial stimulation every day at home. Only as a supplement subject to a strict indication, a stimulating plate is inserted, to position the tongue up and back and to stimulate the upper lip.⁵ According to Castillo Morales, a stimulating plate is indicated when a broad hypotonic tongue with habitually interdental or interalveolar tongue position with hypotonic inactive upper lip is present.² The palatal plate (Figure 1) is produced using mini-plast or acrylic techniques and is extended to the vestibule. In order to exert the stimulation effect repeatedly, the plates are inserted three times a day for 60 minutes each.

Within the last decade, interdisciplinary consultation hours have been established for children with Down syndrome.^{5,9} Stimulating plate therapy has been shown to improve orofacial appearance and function^{3,5-10} and subsequently to have a positive influence on the development of speech and mastication.^{11,12} Most evaluations are conducted directly after the end of therapy. But how stable are the improved orofacial findings after plate therapy was discontinued? The aim of this follow-up study was to investigate whether the improved orofacial function remained stable after an intensive period of growth without wearing the stimulating plate, and whether there were certain factors that could indicate a good candidate for the type of therapeutic approach on the long-term.

MATERIAL AND METHODS

Another 102 children with Down syndrome who had been treated with a palatal plate therapy according to Castillo Morales in their early childhood at the children's rehabilitation center from 1984 - 1991 were invited to attend a follow-up examination.

During the initial examination (T), the orofacial region was examined by an interdisciplinary team including a neuropsychiatrist, a dentist and an orthodontist. Eating habits, speech development, mouth posture and tongue position were recorded by a standardized questionnaire. In the case of treatment need, the children were re-

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ferred to the dentist who produced the plates and controlled the stimulating plate therapy in regular sessions. The mean age of the children to begin with stimulating plate therapy was 13 months \pm 4 months. According to the records, all examined patients showed a good acceptance and regular attendance at the control sessions. In addition to the stimulating plate, an orofacial regulation therapy was carried out in physiotherapeutic practices. At the end of the stimulating plate therapy (R1), the orofacial symptoms were documented by the same neuropsychiatrician who saw initially the child at the first visit (T). At R1 the mean age was 33 months \pm 6 months. The mean duration of plate therapy was 19 months \pm 4 months.

R2 describes the follow-up examination after a long period without wearing plate therapy. At this stage eleven girls and 16 boys could be examined. At R2 the mean age was 170 months \pm 7 months. The follow-up period (T_R2) was 156 months \pm 6 months.

At all three examinations (T, R1, R2), the tongue position and mouth posture was judged by the same neuropsychiatrician using the following grading systems:

-Habitual mouth posture:

- habitual open mouth posture with a lip distance of more than 10 mm
- habitual open mouth posture with a lip distance of less than 10 mm
- primarily incompetent lip closure
- competent lip closure (except when suffering from oropharyngeal infections)

- Habitual tongue position:

- extraoral tongue position
- interdental/ interalveolar tongue position
- caudal tongue position

PARENTAL QUESTIONNAIRE

In addition to the clinical examination, a standardized questionnaire covering an assessment of the orofacial development according to the above mentioned grading system and additional therapy approaches was sent to the parents who were asked to fill it in and bring it along to the examinations. The questions were to be answered by multiple choice and as yes/ no answers.

STATISTICAL ANALYSIS

Statistical analysis was done by SPSS 10.0.7 (SPSS Inc. Chicago, IL, USA). Level of significance was set at 0.05 for all tests. To compare mean changes in mouth and tongue posture before and after treatment as well as at R2, Chi-square test was used. The Mann-Whitney U test was applied to test the differences in the long-term orofacial development in dependence to mouth posture and tongue position at baseline.

RESULTS

The subsequent comparison of the clinical examination and the parents questionnaire revealed a high level of conformity (24 children were assessed the same). In the three deviating cases with different assessments, the base line mouth and tongue position before treatment and the poorer clinical appearance after treatment was determined.

Data concerning adjuvant therapies was taken from the parental questionnaire only.

Habitual mouth posture

The overall improvement from T to R2 was statistically highly significant ($p = 0.000$). The changes of the mouth posture observed during plate therapy (T_R1) were significant ($p = 0.038$), whereas the changes after completion of plate therapy to the follow up examination (R1 to R2) were not ($p = 0.283$).

Taking the different initial findings into account (habitual open mouth posture with a lip distance of more than 10mm versus an open mouth posture with a lip distance of less than 10 mm at baseline), the Mann Whitney U test revealed statistically highly significant differences in the overall development ($p = 0.001$). Within the group with a lip distance of more than 10 mm at baseline, a 100% success rate was recorded: an improvement by three grades in four children, by two grades in five, and by one grade in three children [Table 1]. Of the group of children with an open mouth posture with a lip distance of less than 10 mm, six children improved mouth posture (three by two and three by one degree), in eight children mouth posture remained unchanged, and deteriorated in one child.

Habitual tongue position

From T to R2 the development of the tongue position was positively influenced. The improvements were statistically significant during the overall observation period ($p = 0.004$). A statistically significant amelioration was achieved during plate therapy ($p = 0.004$). Tongue position showed a statistically significant improvement during the observation time without the plate ($p = 0.031$).

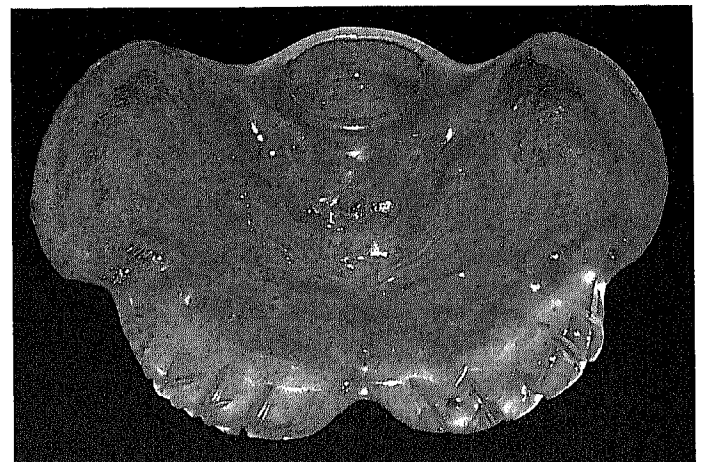


Figure1. Castillo Morales stimulating plate.

Changes [degree]	Lip distance >10 mm [n=12]	Lip distance < 10 mm [n=15]	
-1	0	1	$p = 0.001$
no change	0	8	
1	3	3	
2	5	3	
3	4	0	

Table 1. Overall improvement (T_R2) in mouth posture for the different postures at baseline. -1 = deterioration by one degree; 0 unchanged mouth posture, 1 = improvement by one degree; 2 = improvement by two degrees; 3 = improvement by three degrees.

Changes [degree]	Lip distance >10 mm [n=17]	Lip distance < 10 mm [n=10]	
-1	0	0	p = 0.018
no change	2	3	
1	6	7	
2	9	0	
3	0	0	

Table 2. Overall changes (T_R2) in tongue position for children with different positions at baseline. -1 = deterioration by one degree; 0 unchanged tongue position, 1 = improvement by one degree; 2 = improvement by two degrees; 3 = improvement by three degrees.

Taking the different tongue positions at baseline into account, the further development differentiated significantly (p = 0.018): Of the group with extraoral tongue position: improvement was seen in 15 children, nine by two degrees, six about one degree. In two children, the tongue position was not affected. Of the group with inter-dental tongue position at baseline, seven children improved by one degree. Tongue position was unaltered in three children [Table 2].

Adjuvant therapies

All children received orofacial regulation therapy in addition to the stimulation plate therapy. For all children, the physiotherapy initiated during early childhood was continued after the end of the stimulating plate therapy.

DISCUSSION:

Palatal plate therapy has lasting effects on orofacial development: Mouth and tongue position could be affected significantly during plate therapy. After the end of therapy, mouth posture remained stable, whereas tongue position further improved.

Since earlier studies were based on a short observation time after the end of plate therapy,^{5,7,13,15} only a limited interpretation of these clinical observations was suggested.⁹ Therefore, definite statements as to whether the treatment has lasting effects on the orofacial function are limited. However, the results of this study after thirteen years after initiation of plate therapy show, that the achieved improvements remain stable even without the mechanical stimulation by the plate.

The results underline the conclusion of Zavaglia *et al.*⁶ who stressed the importance of intervening with the muscular component from the early childhood in Down children in order to re-establish an orofacial equilibration. The different developments of mouth and tongue position after plate therapy can be explained by the complex otorhinolaryngologic symptoms in children with Down syndrome with often unsuccessful treatment options.¹⁶

The orofacial status at baseline seems to be a predictive value for the success of long-term improvement: Children with initially extreme orofacial dysfunctions exhibited a statistically significant improved orofacial development during the follow-up than children with more moderate orofacial findings at baseline. Within a shorter

observation time, these tendencies were confirmed in literature.^{8,15} The different developments were explained as a result of varying good compliances on the part of the parents and the patients. Parents of children with extreme orofacial dysfunction tend towards a better compliance supported by motivated physiotherapists and driven by an intensified suffering level.¹⁵ Since good compliance was one of the criteria of the study protocol, the observed changes during the long-term follow-up without mechanical stimulation cannot only be explained by different motivations during the therapy at early childhood. As earlier discussed by Hohoff and Ehmer,⁸ the impressive improvements in children with the worst initial orofacial symptoms could be a hint that the neuromuscular training process evoked by the mechanical stimulus of the plate can only be automated in cases with extreme initial dysfunctions.

Over the time, a therapy-independent decrease in the open mouth posture has been shown in healthy children.¹⁷ This positive development can only be transferred to the development of Down children to a limited extend: children with Down syndrome, extreme orofacial dysfunctions and good compliance during stimulating plate therapy reveal a positive development of the orofacial symptoms when growing up. Whereas children with Down syndrome and mild orofacial dysfunctions during early childhood hardly improve during growth even though compliance is good.

The lack of a control group with untreated Down children with similar orofacial findings at baseline must be criticized. To take a group of children without treatment needs that serves as a control group¹⁵ is rather critical, because children with an obviously better orofacial situation at baseline undergo a different craniofacial development than those with initially severe dysfunctions.¹⁸

Furthermore, the results of this study cannot be reduced to the impact of plate therapy only. Growth and development have an influence on the orofacial development as well.

The difficulty in being objective in the mouth and tongue posture should be mentioned too. The investigator can evaluate only during the examination. The combined evaluation covering the investigator and parental assessment should reduce this problem. The precision of the variables is low. More accurate non-invasive methods for examination and evaluation remain to be developed to reproduce and measure muscle tone and function.

High dropout rates are typical of such patients, and reveal the difficulties associated with a follow-up of children with Down syndrome. To improve the power of the study, the authors offered a second date for the follow-up examination one year later. Due to the high drop out rate it must be assumed that the examined children represent a subgroup of Down children with patients who are particularly interested.

Within these limitations, the results suggest that early plate therapy in Down children yields favorable results that remain stable or further improve even after plate therapy is discontinued. On the short and long term, extreme orofacial dysfunction at baseline seem to improve more than initial mild dysfunction.

Further studies are of clinical interest in order to evaluate the different developments in children with Down syndrome and to specify the prognosis of the long-term treatment outcome.

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