A longitudinal study of palatal plate therapy in children with Down syndrome. Effects on oral motor function

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Abstract
Aim: The aim of the present investigation was to study the effects of palatal plate therapy on oral motor function during a 4-year treatment period. Study design: The effect of palatal plate therapy on oral motor function in children with Down syndrome (DS) was studied over a 4-year period in a prospective randomised clinical study. Subjects: Twenty children with a mean age of 24 months were randomly assigned to a palatal plate group (n=9), or to a control group (n=11). The children in the palatal plate group were treated with modified palatal plates according to Castillo Morales for a treatment period of 4 years. All children in both groups had been enrolled in the same orofacial physiotherapy programme from birth. Orofacial muscle function was documented using video registrations of the face at baseline, after 1 year, and after 4 years. The parents’ perception of the treatment method was investigated using a questionnaire. Results: After 1 year of palatal plate therapy a significant increase in the variable ‘closed mouth’ (p < 0.001) and a significant decrease in the variable ‘inactive tongue protrusion’ (p<0.01) were found. A significant decrease in the summary variables for inactive muscle function was found after 4 years of palatal plate therapy (p<0.05). The summary variables for active muscle function were increased after 4 years but not significantly. Conclusion: The results indicate that palatal plate therapy in combination with orofacial physiotherapy may have beneficial long-term effects on oral motor function in DS children.

Keywords: Children, Down syndrome, functional appliances, oral dysfunction, video

Introduction
Down syndrome (DS), is a genetic disorder caused by trisomy of chromosome 21 (Lejeune et al., 1959). The syndrome is associated with several physical stigmata and functional disorders including general muscle hypotonia (Cohen and Cohen, 1971). The hypotonia in the orofacial region includes the musculature of the face, tongue, and upper lip and the ligamentary apparatus of the temporomandibular joint (Castillo-Morales, 1978; 1991). Underdevelopment of the cranial base and the maxilla also occur (Fischer-Brandies et al., 1986; Limbrock et al., 1990).

Orofacial muscle function during rest and activities like breathing, swallowing, eating, and articulation is abnormal in these children and may affect oral health, craniofacial and dentoalveolar development, and the basis for articulation of speech sounds (Cohen and Cohen, 1971; Castillo-Morales, 1978; Limbrock et al., 1991; Van Borsel, 1996).

A therapeutic concept for children with neuromuscular dysfunction, orofacial regulation therapy, was developed to improve oral motor function (Castillo-Morales, 1991). The therapy includes functional diagnostics of oral sensorimotor dysfunctions, a special manual stimulation and facilitation programme, and treatment with removable activating palatal plates and other orthodontic appliances (Castillo-Morales et al., 1982; Castillo-Morales, 1991; Limbrock et al., 1993). Continuous orofacial stimulation at an early age is important for DS children (Fischer-Brandies, 1988; Hoyer and Limbrock, 1990). In Sweden this is initiated and performed by speech and language pathologists in habilitation centres (Johansson, 1990). We have previously reported the effects of combining this programme in early treatment with a palatal plate modified after Castillo-Morales (Carlstedt et al., 1996; 2001; 2003). We found significant improvements in the group treated with palatal plates compared to a control group in variables describing facial expression, habitual tongue position, lip activity, and mouth closure.

The aim of the present investigation was to study the
effects of palatal plate therapy on oral motor function during a 4-year treatment period. The hypothesis was that, oral motor function in DS children, who had been treated with a stimulation programme and palatal plates, was better than that of DS children who had only undergone the stimulation programme. A secondary aim was to investigate the parents’s perceptions of the treatment method.

**Material and methods**

The patients participating in the study were 20 children from an initial group of 50 with DS referred to the Department of Pediatric Dentistry, Karolinska Institutet. The parents were offered the opportunity for their children to participate in a study. They were to be randomly assigned to begin palatal plate therapy immediately or after one year. Twenty-nine accepted and were available for this study. Eleven children were excluded since their parents wanted treatment, and the children could therefore not be randomised in the study; seven failed to attend appointments; three had severe medical disorders: two children had heart disease, of which one child also had intestinal malformation with several operations planned, and another child had asthma and breathing difficulties. Four children started therapy after one year, and five children did not continue the therapy after one year; those nine were excluded from the study. The patients were randomly assigned to the palatal plate group (PPG) (n=9, 6 boys, 3 girls) and the control group (CG) (n=11, 6 boys, 5 girls). Palatal plate therapy was started between 3 and 33 months of age: four children started therapy during their 1st year of life, three during their 2nd year, and two during their 3rd year. All children used the plate for at least 4 years (range 49 –58 months). The control group comprised 11 children with DS (six males, five females) who were offered palatal plate therapy after 1 year. The mean age after 4 years of treatment was 5.6 ± 1.5 years in both groups. All children lived with their biological parents, and none suffered from any congenital malformations of the heart or had any severe medical complication. Permission was obtained from the ethical boards of Karolinska Institutet and Huddinge University Hospital.

**Palatal plate therapy**

The children in both groups had been enrolled in the orofacial physiotherapy programme from birth and were monitored by speech and language therapists throughout the four years. If the patient had teeth the acrylic plates were designed with spring retentions (Figure 1); otherwise, the plates were formed like a full denture base (Figure 2). They were designed mainly in accordance with the shape of Castillo-Morales basic plates, with a stimulating button anterior to the A-line to influence tongue position and increase tongue activity, and with vestibular knobs for stimulation of the upper lip. The stimulation elements were designed to improve oral motor function by changing the resting position and increasing the activity of the tongue, further improving sucking, swallowing, and eating patterns and enhance breathing through the nose (Castillo-Morales, 1982; Hoyer and Limbrock, 1990; Castillo-Morales 1991). The children used the plates for approximately 1 hour, twice a day. All children had the same design of the plates throughout the 4 years except for two children who, for a limited period (4 and 5 months), had plates with additional stimulating features. These features were knobs placed on the lateral alveolar ridges, and were designed to further stimulate the side borders on the tongue. This was initiated first when the patients had a good lip closure and the tongue had a normal resting position. We found it important to limit the variations in the designs of the plates so as not to create evaluation problems; however, several oral motor teams in Sweden have used various stimulators in the plates. Specific articulatory targets can be stimulated using different movable stimulators in the palate of a child with good lip closure and a normal resting position of the tongue.
Table 1. Variables for evaluating active and inactive muscle function on the video registration at baseline, after 12 months, and after 48 months.

<table>
<thead>
<tr>
<th>Variables</th>
<th>Definition</th>
<th>1-year exam</th>
<th>4-year exam</th>
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<tbody>
<tr>
<td>I</td>
<td>Closed mouth</td>
<td></td>
<td></td>
</tr>
<tr>
<td>II</td>
<td>Tip of tongue visible, tongue on lower lip, closed mouth</td>
<td></td>
<td></td>
</tr>
<tr>
<td>III</td>
<td>Open mouth a) and b)</td>
<td></td>
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<tr>
<td>IV</td>
<td>Inactive protrusion of the tongue</td>
<td></td>
<td></td>
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<tr>
<td>V</td>
<td>Active protrusion of the tongue</td>
<td></td>
<td></td>
</tr>
<tr>
<td>VI</td>
<td>Lips pouting/Pull of lower lip</td>
<td></td>
<td></td>
</tr>
<tr>
<td>VII</td>
<td>Conversation</td>
<td></td>
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Inactive variables for tongue and muscle function: III b, IV
Active variables for tongue and muscle function: I, II, III a, V

Video registration of oral motor function

Video recordings of oral motor function were made of all children in separate sessions at baseline; after 3, 6, 9 and 12 months; and after 4 years of therapy. Recordings of the PPG were made without their palatal plates. The methods used in the video registration have been previously described (Carlstedt et al., 1996; 2001). A 10-minute section of the video recording of each child without the plate, when the mouth of the child was clearly visible, was selected for the evaluation. The videos were made when the children were sitting and playing with a toy or looking in a book. They were not supposed to imitate speech, answer questions, or be in any way encouraged to speak. At the 4-year registration, the children were even told not to speak. In the first study, the videos were evaluated using five variables modified after Limbrock et al. (1991). The video time was evaluated by two of the authors, and the five variables were registered in percentage of the total video time. In the second study, four complementary variables for evaluation of the oral motor function were developed, owing to the increase in complexity and change in oral function patterns that occur with increasing age and development. Three of the active variables; ‘conversation’, ‘pouting the lips’ and ‘pulling the lips’ were not possible to include in the original five variables from baseline (Table 1). The film sequence for each variable was divided into 4-second units, and each unit was classified in accordance with one of the variables. Two types of summary variables were used, to describe active and inactive muscle function. The five variables that were recorded for the two groups at baseline, during the first year and throughout the 4 years including the extended versions of the variables, are presented in this study (Table 1). At baseline and throughout the first year, the variable ‘open mouth’ was registered as an inactive variable. At the follow-up, this variable was divided into an active and an inactive part. The results are expressed as the time for each variable in percentage of the total video time.

All films were evaluated by two of the authors, one of whom did not know which group the children belonged to. In the first study, all films were evaluated separately by two of the authors, one blinded. Each evaluator measured the time the child spent in each activity. A third evaluation was made by the two evaluators if the difference in time between the two evaluations of variables exceeded 10 seconds (Carlstedt et al., 1996). In the second study each 4-second unit was evaluated separately by two of the authors, one blinded. If there was disagreement, a consensus was made. The evaluation was repeated one month later. Agreement between the two evaluators was tested using four randomised films (r= 0.94). The results from the first evaluation were used (Carlstedt et al., 2001).

Parental questionnaire

The parents of the children in the palatal plate group answered a questionnaire after 4 years of therapy. The aim of the questionnaire was to document the parents’ opinions of treatment effects on their children, during and after use of the plate. How much time the parents had spent handling the plate each day and how they handled problems in cooperation during the different activities were additional questions on the questionnaire, as were the time spent using the plate each day and the number of plates each child used.

Statistical analyses

Differences between the two groups in the variables describing oral motor function were evaluated using the Mann-Whitney U-test. To describe differences within the groups after 1 year and 4 years compared to baseline, the Wilcoxon signed rank test was used.
Results

Video registration of oral function

The longitudinal alterations in the five variables documented from the video registration are shown in Figures 3 and 4. At baseline, no statistically significant differences between the two groups were found in these variables. After 12 months, the time for the variable ‘closed mouth’ was significantly longer ($p<0.001$) (Figure 3a), and the time for the variable ‘inactive protrusion of tongue’ was significantly shorter ($p<0.01$) (Figure 3b) in the palatal plate group than in the control group. These changes were most rapid during the first year of treatment: during the subsequent 3 years of treatment, differences between the palatal plate group and the control group were less. After 4 years of treatment, there were no significant differences between the groups regarding these two variables, although the time for ‘closed mouth’ was longer, and the time for ‘inactive tongue protrusion’ was shorter, in the palatal plate group compared to the control group. Neither were there significant differences in the variables ‘tip of the tongue visible’ (Figure 3c), ‘open mouth’ (Figure 3d), and ‘active protrusion of the tongue’ (Figure 3e). Of the total time, the time for the variable ‘open mouth’ was reduced by nearly 50% after 4 years compared to baseline in both the palatal plate group and the control group (Figure 3d).

Regarding changes in the individual parameters over time, it was found that compared to baseline, the variable ‘closed mouth’ accounted for significantly longer time in the palatal plate group after 1 year ($p<0.001$). This change was not seen in the control group during this time. After 4 years of treatment, the time for the variable ‘closed mouth’ was significantly longer ($p<0.05$) compared to baseline in both the palatal plate group and the control group (Figure 3a).

Figure 3a. Duration of the variable ‘closed mouth’ from baseline through 4 years expressed as per cent of total time for the palatal plate group (solid circles) and the control group (open circles). The Wilcoxon signed rank test was used to describe differences within the groups after 1 year and 4 years compared to baseline. The Mann-Whitney U-test was used to make comparisons between the groups, $p<0.05$ *, $p<0.001$ ***.

Figure 3b. Duration of the variable ‘inactive protrusion of tongue’ from baseline through 4 years expressed as per cent of total time for the palatal plate group (solid circles) and the control group (open circles). The Wilcoxon signed rank test was used to describe differences within the groups after 1 year and 4 years compared to baseline. The Mann-Whitney U-test was used to make comparisons between the groups, $p<0.05$ *, $p<0.01$ **.

Figure 3c. Duration of the variable ‘tip of tongue visible’ from baseline through 4 years expressed as per cent of total time for the palatal plate group (solid circles) and the control group (open circles).

Figure 3d. Duration of the variable ‘open mouth’ from baseline through 4 years expressed as per cent of total time for the palatal plate group (solid circles) and the control group (open circles).
time for the variable ‘inactive protrusion of tongue’ was significantly shorter after 1 year ($p<0.05$), and 4 years ($p<0.05$) of treatment compared to baseline in the PPG. In the CG the time for this variable was significantly longer after 1 year ($p<0.05$) compared to baseline (Figure 3b).

Regarding the summary variables for inactive muscle function, the time in percentage of total time was significantly shorter after 1 year ($p<0.01$), and after 4 years ($p<0.05$), in the palatal plate group compared to the control group (Figure 4a). The time for active variables in percentage of total time was significantly longer ($p<0.001$) after 1 year in the palatal plate group compared to the control group; after 4 years these times were still longer in the PPG, but differences were non-significant (Figure 4b).

**Questionnaire**

Eight out of nine parents reported positive effects of the palatal plate therapy in their children, such as more activity in the face and a changed facial expression, both while using the plates, and after using them. Furthermore, five of the nine parents reported improved mouth closure and more talk, both with and without the plates. The mean number of palatal plates designed for each child was six (range four to seven). Regarding problems with cooperation, six of the nine parents had no or few problems most of the time. Cooperation problems were often associated with tooth eruption, in a few cases during illnesses such as upper airway infections, and when the child’s ability to say ‘no’ increased. All children used the plates twice a day, for approximately 30-60 minutes except during illness. Most of the parents agreed upon the importance of creating daily routines in the use of the plate; thus storytelling and television were favourite occupations. None of the parents considered the time they spent, or problems with their child’s cooperation while using the plate, to be too much trouble or not worthwhile.

**Discussion**

Orofacial physiotherapy for children with different neuro- muscular deficiencies including treatment with palatal plates was introduced more than 20 years ago (Castillo-Morales, 1978). It is still difficult to find prospective clinical studies that evaluate treatment with palatal plates using a randomised design and an untreated DS control group. In previous reports on this study, we have shown positive effects of treatment with palatal plate therapy performed according to the methods of Castillo-Morales (Carlstedt et al., 1996; 2001; 2003). In those studies, we found that treatment of DS children for 4 years with palatal plate therapy had positive effects on some aspects of oral motor function in comparison with untreated controls.

Longitudinal studies on children with learning disabilities are difficult, time consuming and have high drop-out rates (Leonard and Wen, 2002; Seltzer et al., 2004). The strengths of this study are its longitudinal design, the inclu-
sion of an untreated control group from the start, and that few participants drop out.

Evaluating oral motor function objectively is difficult, and one problem is the precision of the variables used since the methods available are mostly based on visual and audible estimation (Limbrook et al., 1991). Video registration has been used in previous studies for evaluation of treatment effects on oral dysfunction in DS children (Glatz-Noll and Berg, 1991). The agreement between the investigators in this study was tested and found to be high, which is important for reliability, and the results are also strengthened by one blinded investigator.

Treatment effect decreased over time in the palatal plate group. After one year of treatment, there were significant improvements in the variables for mouth closure and inactive tongue protrusion, which illustrates the ability of this treatment to facilitate normalised tongue position, mouth closure, and facial expression at an early age. The reasons for the diminishing difference between the two groups after 4 years of therapy e.g. the decreasing effect of the plates are four-fold:

1) Children increase their oral motor repertoire and as a consequence more variables compete for the observed time. To overcome this problem we constructed the summary variables.
2) Developmental effects which always affects treatment results.
3) Oral motor function in the children in the control group also improved since they had followed the same special orofacial physiotherapy programme from birth, with manual stimulation as well as speech training, as had the children in the palatal plate group.
4) The use of more complicated plates could have improved the results, but would have made interpretation of results more difficult.

Several studies report positive results with palatal plate therapy, although most are not randomised, prospective studies and no untreated control groups are included, which makes treatment effects difficult to evaluate (Castillo-Morales, 1982; Fischer-Brandies, 1988; Hoyer and Limbrock, 1990; Glatz-Noll and Berg, 1991; Hohoff and Ehmer, 1997, 1999; Schuster and Giese, 2001; Zavaglia et al., 2003; Korbmacher et al., 2004). Because we found the positive treatment results to be most pronounced after one year, we must consider the importance of the treatment effects-improved facial expression, improved breathing and swallowing patterns, and a changed tongue position during the first period of the child’s life. Greater muscle tone in the orofacial muscles has a crucial influence on the appearance and functional aspects of a child during his or her early life. These aspects are also psychologically important for the parents and the child in attitudes of others. Moreover, there were significant differences between the two groups in the summary variables describing inactive oral muscle function, in the variables describing whether the tongue was visible during speech and non-speech, and whether the lip was rounded at the articulation of rounded vowels during spontaneous speech after 4 years (Carlstedt et al., 2001, 2003).

Korbmacher et al. (2004) stress the importance of a manual stimulation programme in combination with PPT, and in their study, 55% of the children received additional speech therapy. The authors point out that early diagnostic findings, which identify children with extreme orofacial dysfunction are important. Such children exhibited the best long-term improvement. However, without an untreated control group, it is difficult to distinguish treatment effects from developmental effects.

Since the support and interest of the parents is a crucial factor throughout treatment, we wanted to find out about their experiences of the efforts and time spent with their children’s plates, and also their appraisal of their children’s improvement. How their children cooperated was also one of the questions. In this study, most parents had no, or minor, problems most of the time. Cooperation and retention problems were often associated with tooth eruption. During tooth eruption, retention problems are common, which has been found by other authors (Fischer-Brandies, 1988; Hohoff and Ehmer, 1997, 1999). Illnesses such as upper airway infections and being in an obstinate stage of development were other explanations in a few cases. As discussed in previous studies, parental questionnaires are subjective since parents often tend to observe improvements because they correspond to their wishes (Hohoff and Ehmer, 1997; Schuster and Giese, 2001). Despite this, we found the parents’ experiences and appraisal to be a valuable complement to the results since they provided information about efforts and benefits, aspects that need to be considered in the overall conclusion on treatment advantages. It is of great importance that parents are involved in the training of their children and include them in improved development under the circumstances. From our results it therefore seems reasonable to recommend this treatment as early as possible.

Conclusions

In conclusion, the result of this study is that palatal plate therapy has a positive effect on oral motor function, particularly during the first year of therapy. However, variables describing inactive muscle function were significantly reduced in the palatal plate group also after 4 years of treatment compared to the control group. The parents’ perception of the treatment effect was favourable. More activity in speech and more active facial expressions were found. Despite these positive results, the incorporation of palatal plate therapy into routine treatment of oral motor dysfunction remains to be further investigated.
Acknowledgements

This study was supported by grants from the Institute of Odontology, Karolinska Institute and the May Day Flower Annual Campaign for Children’s Health. Special thanks to Bo Nilsson for statistical assistance.

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