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Children with Down syndrome: oral development and morphology after use of palatal plates between 6 and 48 months of age

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Objective. The aim of this study was to describe the number of erupted teeth, sucking habits, tongue morphology, facial expression and speech in children with Down syndrome (DS) aged 48 ± 6 months and treated from 6 months of age with palatal plates in combination with speech and language therapy.

Methods. The research took the form of a multicentre, multidisciplinary, longitudinal study of children with DS followed from the age of 6 months. A total of 37 children with DS were included. One child could not cooperate at all and was excluded from the evaluations. In combination with speech and language intervention provided by speech and language therapists, the children used palatal plates provided by dentists from 6 months of age. In the evaluation, the children in the sample ($n = 36$) were compared with two similarly aged control groups: one group of children with DS who never had used palatal plates ($n = 31$) and one group of children with normal development ($n = 36$). The evaluation of oral parameters was performed by dentists after calibration. Registration of facial expression and speech was done by a speech and language therapist, and the evaluation was done by two speech and language therapists and one phonetician who were calibrated in joint discussions.

Results. In contrast to the children with DS in the control group, the subjects in the study were found to have as many erupted teeth as the children with normal development. The prevalence of sucking habits did not differ between the three groups. Only children with DS sucked their tongue, a toy or other things in addition to a thumb or dummy. The prevalence of tongue diastase in the study group with DS was of the same magnitude as in the evaluation at the age of 18 ± 3 months. The palatal plates were used by 57–65% of the children without any larger problems. In the study sample, the possible beneficial effects of palatal plate therapy were a lower prevalence of posterior cross-bite, a higher prevalence of frontal cusp-to-cusp relation and a lower prevalence of frontal open bite. Evaluation of facial expression and speech showed a higher score for facial expression and a better communicative capacity in the children in the study group than in the control children with DS.

Conclusions. In children with DS, palatal plate therapy between 6 and 48 months of age in connection with speech and language intervention had a positive effect on occlusion, oral motor function, facial expression and speech. No harmful effects were observed. Although this is a valuable method, however, it must be emphasized that palatal plate therapy puts additional demands on already burdened children and their caretakers.

Introduction

Generalized growth deficiency and intellectual disability characterize Down syndrome (DS) (trisomy 21)[1]. Among many other symptoms, orofacial hypotonia is an obvious feature[2]. To improve oral motor function, palatal plates designed according to Castillo-Morales[3] have been recommended. Many studies on the effect of this therapy, have not involved untreated control groups[4–9], but the results have shown a beneficial effect on oral motor function in longitudinal evaluations. It has also been noted, however, that palatal plate therapy alone is inadequate to obtain good results and needs to be complemented with physiotherapeutic programmes. Carlstedt *et al.*[10,11] drew the same

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conclusion after having performed a longitudinal randomized control study of nine children with DS treated with palatal plates since infancy.

In 1995, a longitudinal, multicentre, multidisciplinary study on 6-month-old children with DS was started by a group of paediatric dentists, and speech and language therapists. The aim of the study is to evaluate palatal plate therapy in combination with speech and language intervention between the ages of 6 months and 8 years. The design of the palatal plates was to be based on the progression of sound development in similarly aged children with normal development. Only the plate used between 6 and 10 months of age was designed according to Castillo-Morales[3].

The hypothesis was that the occlusion anomalies[12–15] and the poor speech skills[16–18] often found in children with DS are worsened by their deficiencies in oral motor and sensory function, and consequently, that the children in the study will have a lower prevalence of malocclusion, as well as improved oral motor performance, articulation and speech.

After use of palatal plates between 6 and 18 months of age, there were no differences in oral development and morphology between the 42 children with DS in the study and a group of age-matched controls with DS[19]. The children with DS in the study group, however, seemed to have better prerequisites for articulation than the children with DS in the control group, as shown by better control of motor performance, more appropriate patterns of upper and lower articulators, and sound production, as well as getting better results on the tests and performing them with more maturity than the children with DS in the control group[20].

The aim of this study is to describe results including the number of erupted teeth, sucking habits, tongue morphology, facial expression and speech after use of palatal plates in combination with speech and language intervention between 6 and 48 months of age.

Subjects and methods

Subjects

The children with DS taking part in the study have been followed since the age of ≤ 6 months. Five of the original 42 participants have left the study since their evaluation at 18 ± 3 months; three of them were medically healthy, one had a minor cardiac problem and one child had developed sleep apnoea. Of the remaining 37 children, 11 live in Stockholm (south-east Sweden), nine in Umeå (northern Sweden), 11 in Karlstad (south-west Sweden) and six in Skövde (southern Sweden). The health of all the study children has been accounted for previously[19]. Two children have been successfully treated for leukaemia since the evaluation at 18 ± 3 months and 59% are healthy. The 31 age-matched controls with DS were selected via their special needs clinical support teams in Stockholm, Umeå, Karlstad, Skövde and Gothenburg. Only children with trisomy 21 who had never been treated with palatal plates were included. Like all children with DS in Sweden, they were included in the special medical care programme for children with DS which also includes speech and communication training[21,22]. Palatal plate therapy had not been instituted by their special needs clinical support teams, but the children were offered such treatment after having been evaluated in this study. Of the controls with DS, two had had cardiac surgery, one had a heart condition that was incurable and one had had a minor heart condition that did not require surgery. In addition, one child had been treated for leukaemia, three had allergic conditions and two children had autistic traits. The remaining controls were healthy (68%).

Thirty-six age-matched healthy controls with normal development were selected from child health centres and the Public Dental Health Service in Stockholm and Umeå.

The evaluations were performed when the children were 48 ± 6 months old.

The number of children evaluated in the study is shown in Table 1.

Methods

In this study, speech and language therapists provide language intervention and oral motor and sensory stimulation in close cooperation with parents, according to methods described by Johansson[22]. Dentists provide palatal plates. The plates are to be used two to three times

Table 1. Number of children evaluated in the study (number not evaluated listed in the footnotes). Causes of not being evaluated included illnesses, missing appointments and technical problems connected with video recording.

		Evaluation		
Group	**Mean age months (range)**	**Number of erupted teeth, sucking habits, tongue morphology and occlusion**	**Facial expression**	**Speech**
Normal controls	49.3 (42–55)	36 (15 F, 21 M)	36 (15 F, 21 M)	30 (14 F, 16 M)
Controls with DS	49.8 (42–56)	31 (13 F, 18 M)	29 (12 F, 17 M)*	30 (13 F, 17 M)†
Study subjects with DS	50.6 (44–57)	36 (17 F, 20 M)	33 (15 F, 18 M)‡	32 (17 F, 15 M)§

(F) female; (M) male; and (DS) Down syndrome. *Two missing. †One missing. ‡Three missing. §Four missing.

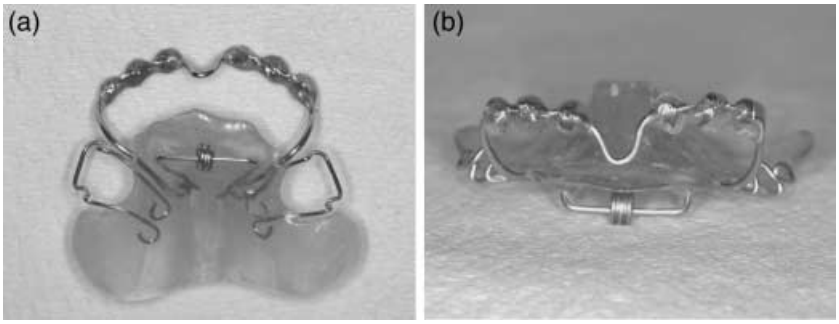

Fig. 1. Two views of the palatal plate used between 18 and 30 months of age.

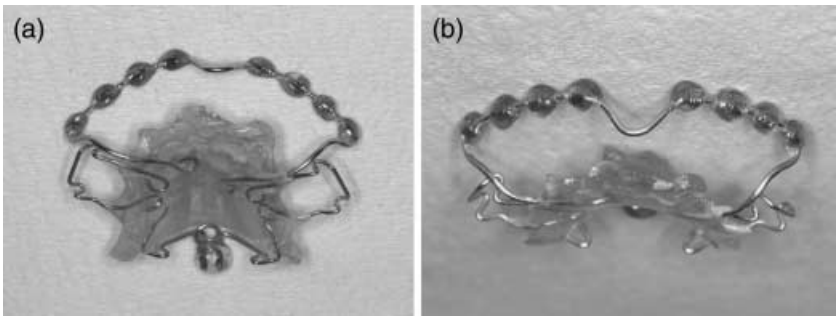

Fig. 2. Two views of the palatal plate used between 30 and 48 months of age.

daily for short time periods not exceeding 30 min and in connection with speech therapy exercises. The plates are similarly designed for all children.

The palatal plate intended for use between 18 and 30 months of age is shown in Fig. 1.

In order to stabilize articulatory patterns of dento-alveolar consonant production, the mobility of the anterior part of the tongue is stimulated by a movable ball on a stainless-steel wire behind the maxillary incisors. The facial bow is placed as high as possible in the buccal fold to improve lip closure by stimulating the orbicularis oris muscle in order to promote the various labial articulation patterns. If further training to retract the tongue body is necessary, this can be achieved through a bowl-shaped elevation at the borderline between the hard palate and the velum.

The palatal plate intended for use between 30 and 48 months of age, or until the child has outgrown the device, is shown in Fig. 2.

Plastic bulbs are attached to the facial bow, which is extended laterally in the buccal fold to stimulate lip closure. To stimulate the mobility of the lateral and dorsal parts of the

tongue, the plate has a transversal steel wire with three bends at the borderline between the hard palate and the velum. A pearl is moved along the wire and placed in a new bend every time the plate is used so that the tongue is stimulated both medially and laterally. The aim is to promote palato-velar articulation patterns.

Parents register how many times the palatal plate is used every day. When the plate is going to be exchanged, it is evaluated by the parents and by the dentist with regard to compliance, problems and benefits.

Oral parameters

The parameters evaluated are shown in Box 1. All dentists involved in the examinations are experienced practitioners who are calibrated in joint meetings by discussing and assessing clinical illustrations of the evaluated parameters. No radiographs were taken.

Evaluation of facial expression and speech

The children were videotaped during play and interaction with one of the parents and a test leader, a speech and language therapist. Each child was filmed for 30–45 min. Two speech and language therapists, and one phonetician qualitatively evaluated facial expression, speech sounds and spontaneous sound production. Cases of deviance were discussed until a consensus was reached.

Facial expression was classified in five groups ranging between 'active, coordinated' and 'very low level of coordination'. The evaluation was based on the parameters shown in Box 2.

Box 1. Evaluated oral parameters.

1. Number of erupted teeth; established by clinical examination.
2. Morphology of the tongue; established by clinical examination and classified as:
   - **a** normal;
   - **b** diastase, i.e. the midline of the tongue is elevated, with fissures on each side along the midline;
   - **c** horizontal sulcus in the anterior part of the tongue;
   - **d** lingua plicata; and
   - **e** others, i.e. combinations of the above-mentioned traits.
3. Type of sucking habit; established by interviewing the parents.
4. Occlusion; established by clinical examination and dental casts.
5. Compliance with the use of the palatal plates; established by interviewing the parents and daycare staff.

Box 2. Evaluated parameters for facial expression.

1. Position of the jaw is based on estimation of the jaw angle. The angle is judged as correct or low. A low angle means that the mandible is lowered compared to a normal rest position.
2. Mouth closure judged as closed, half-open or open.
3. Tongue position judged as retracted, visible or outside the mouth.
4. Morphology of the upper and lower lip, respectively. Judged as straight, bent upward or protruding.
5. Musculature under the eyes judged as normal, somewhat loose or loose.
6. Cheek musculature judged as having good tonus or not possible to judge.
7. Smile judged in interaction with a grown-up as genuine or not.

Statistical methods

To compare variables measured on a nominal scale, the chi-square and the exact chi-square test were used. The exact McNemar test was used to analyse the change in the morphology of the tongue in the children in the study between 18 and 48 months. *P*-values of < 0.05 were set as an indication of a significant difference. No statistical differences were calculated for the qualitative data of facial expression and speech.

Ethical approval

This study has been approved by the ethical boards of the medical/odontological faculties in Umeå, Gothenburg and Stockholm, and by the ethical research committee in Örebro. All parents of the children in the study group as well as in the two control groups have given their informed consent to participation.

Results

Compliance in use of the palatal plates

The plates were used according to instructions.

The palatal plates used between 18 to 30 months of age (Fig. 1) were made for all but one child, who refused. The plates were used without problems by 24 children (65%). For two of these children, adhesive was needed to improve retention. Three children used the plates sporadically, four had problems with

retention as tooth eruption proceeded and recurrent infections in three children made cooperation difficult.

The palatal plates used between 30 and 48 months of age (Fig. 2) were used by all but two children: one who had not use the previous plate either and one who left the study because of practical problems. For 20 children (57%), there were no or only minor problems with the plates; three children outgrew their plates and received new ones, one child suffered from regular infections, and in two children, erupting teeth interfered with use. Leukaemia was diagnosed in one child and autism in a second child, interrupting use of the plate.

The child who did not use any of the two plates during the period between 18 and 48 months was excluded from the evaluations.

At the evaluations, three children in each of the groups with DS and two children in the group with normal development were 1–2 months older than 48 ± 6 months because of problems interfering with appointment times. Since they were found not to interfere with the results, they were not excluded from the comparisons.

No statistically significant differences were found in the comparison of the evaluated parameters between girls and boys, which is why the results are presented for each group as a whole.

Number of erupted teeth

The number of primary teeth in the three groups is shown in Fig. 3. In the group with normal development, the mean number of erupted teeth was 19.9 (standard deviation 0.4), in the control group with DS this was 19.4 (standard deviation 0.9) and in the study group with DS it was 19.7 (standard deviation 0.7).

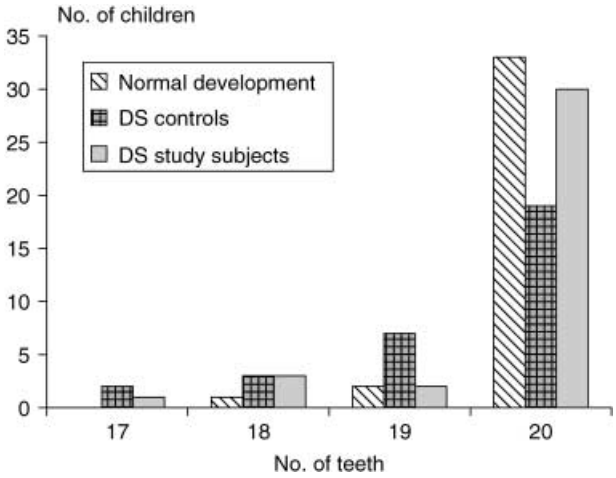

Fig. 3. Number of teeth in the three groups.

The difference in the number of erupted teeth between the children with normal development and the controls with DS was statistically significant ($P = 0.01$). There was no difference in the number of erupted teeth between the children with normal development and the children with DS in the study group.

Table 2 shows the number of children with unerupted teeth and the reasons for missing teeth. All of the unerupted teeth were incisors. One primary second molar had been extracted because of caries.

Double-formation of 71–72 and 82–83, respectively, was seen in two children in the study. In one child in each of the two groups of children with DS, maxillary canines were erupting as the last primary teeth.

Sucking habits

The number of children in the three groups with different sucking habits is shown in Fig. 4.

Table 2. Number of children, number of unerupted teeth and causes of missing teeth in the three groups.

	Normal controls (*n* = 36)		Controls with DS (*n* = 31)		Study subjects with DS (*n* = 36)	
Variable	**Children (*n*)**	**Teeth (*n*)**	**Children (*n*)**	**Teeth (*n*)**	**Children (*n*)**	**Teeth (*n*)**
Unerupted teeth	1 (2.8%)	2	10 (32.2%)	16	6 (16.2%)	9
Trauma	2	2	2	3	1	1
Caries	0	0	0	0	0	1

(DS) Down syndrome.

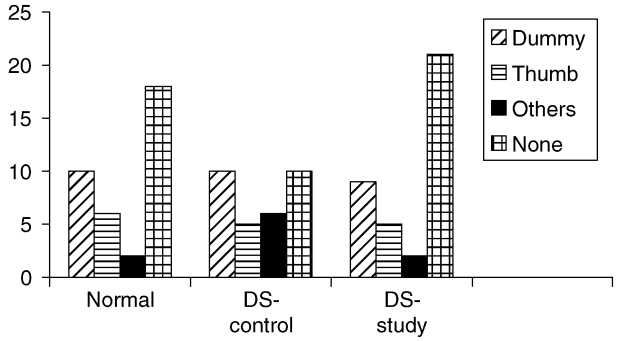

Fig. 4. Sucking habits in the three groups.

Of the children with normal development, two still used a nursing bottle.

Of the control children with DS, six sucked something else than dummy or thumb; for example, their tongue or a toy. Two of the children with DS in the study had similar habits. There was no statistically significant difference in the frequency of sucking habits between the three groups.

Tongue morphology

Tongue morphology in the three groups is shown in Table 3.

There was no statistically significant difference in tongue morphology between the two groups with DS. The children with DS had a statistically significant greater number of deviations from normal morphology than those with normal development ($P < 0.001$). There was no statistically significant difference in tongue morphology between children in the evaluation at 18 ± 3 months and those aged 48 ± 6 months in this study. In this comparison, the children with DS in the study were compared with themselves at the age of 18 ± 3 months.

Occlusion

Occlusal relations in the three groups are shown in Table 4. Judgement of occlusion was possible in all children with normal development and in most of those with DS.

Angle Class III occlusion was statistically significantly ($P < 0.001$) more prevalent in each of the two groups with DS compared with children with normal development. A posterior lateral cross-bite was statistically significantly more prevalent in the controls with DS than in the study subjects with DS ($P = 0.02$) or the children with normal development ($P < 0.007$). Comparisons between the groups concerning frontal vertical relations showed no statistically significant differences between the two groups with DS, but a higher prevalence of frontal cusp-to-cusp relation and a lower prevalence of frontal open bite in the study subjects with DS compared with the children with normal development ($P = 0.011$). Concerning frontal horizontal relations, all children with normal development had positive values for overjet. All children with DS in the two groups had a higher prevalence of negative values for frontal overjet and cusp-to-cusp relation ($P < 0.001$) than the children with normal development.

Facial expression

As a result of illness, missing appointments, bad quality of the videotapes or other technical problems, facial expression was not evaluated in all children. Table 1 shows the number of children evaluated.

Classification of facial expression in the three groups is shown in Table 5.

In all but one child with normal development, facial expression was classified as 'active,

Table 3. Number of children with different tongue morphologies in the three groups.

	Tongue morphology					
Group	**Normal**	**Diastase**	**Lingua plicata**	**Horizontal sulcus present**	**Others**	**Total number of children**
Normal controls	35 (97.2%)	1	0	0	0	36
Controls with DS	18 (58.1%)	9	4	0	0	31
Study subjects with DS	19 (52.8%)	8	6	2	1	36

(DS) Down syndrome.

Table 4. Occlusal relations in the three groups and statistically significant *P*-values of group comparisons.

Occlusal relation	(a) Normal controls (*n* = 36)	(b) Controls with DS (*n* = 31)	(c) Study subjects with DS (*n* = 36)	*P*-value
Sagittal relation				
Angle Class:				
I	29	25	32	
II	7	1	0	
III	0	4	3	< 0.001 (a/b, a/c)
Judgement not possible	0	1	1	
Transversal relation				
Normal	34 RS, 30 LS	19 RS, 22 LS	32 RS, 30 LS	
Posterior cross-bite	2 RS, 5 LS	9 RS, 6 LS	3 RS, 5 LS	0.02 (b/c)
Cusp-to-cusp	1 LS	0	0	< 0.007 (b/a)
Judgement not possible	0	3	1	
Frontal vertical relation (overbite)				
Cusp-to-cusp	5	8	15	0.011 (c/a)
Open bite	12	12	6	0.011 (c/a)
Deep bite	19	9	14	
Judgement not possible	0	2	1	
Frontal horizontal relations (overjet)				
Cusp-to-cusp	0	7	9	< 0.001 (a/b, a/c)
Negative	0	4	5	< 0.001 (a/b, a/c)
Positive	36	18	20	
Judgement not possible	0	0	0	

(DS) Down syndrome; (RS) right side; and (LS) left side.

Table 5. Classification of facial expression in the three groups.

	Facial expression					
Group	Active, coordinated	Active, almost coordinated	Active, partly coordinated	Low level of coordination	Very low level of coordination	Total number of children
Normal controls	35	1	0	0	0	36
Controls with DS	0	1	9	15	4	29
Study subjects with DS	0	6	16	9	2	33

(DS) Down syndrome.

coordinated', but none of the children with DS were so categorized. Of the study subjects with DS, 22 (66.7%) children had an 'active, almost coordinated' or 'active, partly coordinated' facial expression compared with 10 (34.5%) of the controls with DS.

Speech

The number of children evaluated is shown in Table 1. The reasons for not participating in the evaluation were illness, missing appointments, inferior quality of the videotapes or other technical problems. The three groups differed in answering behaviour. The children with normal development answered 93% of the test items ($n = 1440$), the controls with DS 30% and the study subjects with DS answered 84% of the test items.

The only answering mode of the children with normal development was speech. They pronounced the target consonants correctly and approximately correctly in 85% and 7% of the items, respectively.

The children with DS in the control group met the criteria of correct pronunciation and approximately correct pronunciation for 8% and 6% of the items, respectively. Of the answers, 8% were pure imitations, onomatopoetic sounds or strings of sounds without phonological structure. In 3% of the answers, the children signed and spoke the words simultaneously, and in 5%, they signed the word silently.

The study subjects with DS pronounced 23% and 9% of the consonants correctly and approximately correctly, respectively. In 4% of their answers, they used imitations, onomatopoetic sounds, gestalts or strings of sounds. Signs were used in 38% of all answers, in 15% of the answers in which the children signed and spoke the word simultaneously, and in 23% of the answers they signed silently.

No statistical evaluation was made of these data.

Discussion

This describes an evaluation made after 4 years of palatal plate therapy in combination with speech and language intervention in a group of similarly aged children with DS.

Comparisons with previous studies of palatal plate therapy for children with DS are difficult because of differences in age, treatment methods, treatment time and type of evaluation.

One significant result from this study is the difficulties involved in the use of palatal plates, which are in addition to all the other demands on carers considering the health problems affecting many children with DS. Furthermore, the plates are used during periods of tooth eruption, which interferes with the adhesion of the plates. Nevertheless, the plates were used by 57–65% of the children without any major problems. Use of the plates was interrupted mainly because of health problems, and the children who exhibited less-than-perfect compliance still used one or both plates during varying time periods. These subjects were not excluded from the evaluations since they represent ordinary problems connected with use of palatal plates in very young children with DS. Only the child who never used either of the two plates was excluded. Previous studies have not accounted explicitly for compliance, but the number of dropouts reported indicate that compliance could have been of the same magnitude as in this study[5,6,8,23].

The study subjects with DS in this study have been treated with palatal plates since the age of 6 months, in combination with oral motor and sensory training. The plates are specially designed to stimulate places of articulation, and tongue and lip movements for speech sounds. Therefore, the design differs from the recommendation by Castillo-Morales[3] that only non-mobile stimulators should be used for children with DS.

In this study, the controls with DS had fewer erupted teeth than both the children with normal development and the study subjects with DS. These results are in accordance with Fischer-Brandies[4], and could possibly be attributed to the stimulation of the oral mucosa by the palatal plates, which might have hastened tooth eruption in the study subjects with DS. Even though late eruption characterizes children with DS, this study showed that about 62% of the controls with DS and 84% of the study subjects with DS had all 20 primary teeth erupted at the age of 48 ± 6 months. Since the unerupted teeth are incisors, it is possible that they could be absent as a result of hypodontia. The prevalence of hypodontia of primary teeth in children with DS has been reported to be 11–17%, and the incisors are primarily affected[24,25]. Since no radiographs were taken in this study, it is not possible to verify the diagnosis. The number of teeth lost because of trauma was lower than previously reported[26] and did not differ between the groups. Double-formations were observed in two of the study subjects with DS, as previously reported[24].

The prevalence of sucking habits did not differ between the three groups. As in the previous evaluation[19], only children with DS sucked their tongue, a toy or other things in addition to a dummy or thumb. As previously reported[27], about 50% of the children with normal development and 57% of the study subjects with DS had no sucking habit. The corresponding figure for the controls with DS was 26%. One explanation for the difference between the two groups with DS could be the effect of the training with palatal plates, which could have interfered with the sucking habits in the study subjects.

Oral motor and sensory training, with or without combination with palatal plates, does not seem to influence tongue morphology. Since tongue diastase is considered to be a sign of hypotonia, it was hypothesized that oral motor training would decrease its prevalence[28]. This

was not the case in this study, although the prevalence was found to be considerably lower than previously reported.

One aim of the palatal plates is to increase the mobility of the tongue and to stimulate tongue retraction and lip closure. A beneficial effect on occlusion of the training could be a lower prevalence of posterior cross-bite in the study subjects than in the controls with DS. Another positive effect could a higher prevalence of cusp-to-cusp relation and a lower prevalence of frontal open bite in comparison with children with normal development. Since the prevalence of sucking habits was the same in children with normal development and in the study subjects with DS, the difference is most probably a result of the training with palatal plates. The consequences of the growth pattern in DS[4] resulting in Angle Class III occlusion with a negative frontal overjet cannot be hindered with palatal plate therapy.

Active facial expression is important for non-verbal communication, in conveying messages and illustrating shades of meaning. There was a large difference in facial expression between both groups of children with DS and those with normal development, however, the study subjects scored better than the controls with DS. In evaluating facial expression, underlying parameters like tongue and jaw position and lip closure are influenced by training with palatal plates, which should have contributed to the higher scores in the study subjects. These results are in accordance with Carlstedt *et al*.[29].

The evaluation of speech showed a large difference between the children with normal development and those with DS. Although there was also a variation among the children with normal development, they are more confident as a group in the role of speaker than those with DS. Among the two groups with DS there, was one obvious difference: the children in the study group answered the majority of the test items, whereas the children in the control group left the majority of the test items unanswered. The study subjects with DS had a more advanced communicative capacity than the controls with DS; their pronunciation was more correct and they had a lower prevalence of sounds without phonology.

What this paper adds

- Previous studies on the use of palatal plates in children with Down syndrome are mostly case reports based on the concept formulated by Castillo-Morales.
- The aim with the present longitudinal, case-control study is to evaluate use of palatal plates in combination with speech and language intervention.

Why this paper is important to paediatric dentists

- Palatal plate therapy puts great demands on both children and caretakers.
- It is important that the dentist is highly competent and that the decision to perform the treatment is based on due analyse by a multi professional team.
- In the present study, palatal plate therapy is scientifically evaluated. Benefits and drawbacks with the therapy are accounted for.

Conclusion

Speech and language intervention using palatal plate therapy between 6 and 48 months of age has a positive effect on occlusion, oral motor function, facial expression and speech. No harmful effects were observed. Although palatal plate therapy is a valuable method, it must be emphasized, however, that it puts additional demands on already burdened children and caretakers.

The next evaluation of this study will describe the results for the subjects at 8 years of age.

Acknowledgements

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