A four-year longitudinal study of palatal plate therapy in children with Down syndrome: effects on oral motor function, articulation and communication preferences

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The orofacial function in 20 children with Down syndrome was evaluated after 4 years of palatal plate therapy in 9 of the children (PPG); the remaining 11 were untreated age-matched controls (CG). All 20 children had received continuous orofacial physical therapy from their speech therapist during the treatment period. A clinical extra- and intraoral examination was performed, including oral motor function, facial expression, the occurrence of malocclusions, and hypertrophic tonsils. A questionnaire requesting data on breathing patterns, drooling, eating problems, and communicative preferences was answered by the parents. An articulation assessment was performed by two speech and language pathologists blinded to the treatment status of the children in order to find out whether the palatal plate had stimulated to improved oral speech behavior. The results for oral motor function showed significant differences between the groups in favor of the PPG for the summary variables for: visible tongue ($P < 0.01$), visible tongue during non-speech periods ($P < 0.05$), and lip-rounding during spontaneous speech ($P < 0.01$). During non-speech time, the PPG had their mouths open significantly less than the CG ($P < 0.05$). Expressivity of facial expression on a visual analog scale in the PPG scored 75.6 ± 13.3 compared to 51.8 ± 25.7 in the CG ($P < 0.05$). The intraoral examination showed that 6/9 children in the PPG and 7/11 in the CG had enlarged tonsils, resulting in more than 50% inter-tonsillary space reduction. Despite these findings, and no significant differences between the groups with respect to mouth/nose breathing, nocturnal snoring was significantly less in the PPG than in the CG ($P < 0.05$), according to the parental questionnaire. After 4 years of palatal plate therapy, orofacial function had improved significantly in the 9 PPG children and specifically in terms of tongue position and lip activity. □ *Down syndrome; functional appliances; oral dysfunction; speech; tonsils*

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Down syndrome (DS), a genetic anomaly related to trisomy of chromosome 21 (1), is associated with several functional disorders (2, 3). General muscle hypotonia, including the musculature of the face, tongue, upper lip and the ligamentary apparatus of the temporomandibular joint are common features, combined with underdevelopment of the maxilla (4–6). Movements of the lips, cheeks and tongue in swallowing and in articulation are abnormal, and may affect the basis for the articulation of speech sounds (6, 7).

It is important that continuous orofacial stimulation is introduced at an early age in children with DS. This consists of an exercise program for the orofacial muscles performed and initiated by a speech and language pathologist (SLP) (8). Most Swedish DS children are given a program for language stimulation based on adapted manual signs (signed main words, often both signs and speech in the same sentence) (7). If the child has a general hypotonia in the orofacial muscles, the program can be combined with an individually designed palatal plate. The aim of plate therapy is to alter the resting position of the tongue, to stimulate specific tongue movements, to increase mobility of the upper lip, and to increase tonus of the facial musculature (6, 9–11).

In our first study, the results from a 1-year treatment period with palatal plate therapy (PPT) in DS children with a mean age of 24 ± 6 months showed improved mouth closure and reduced tongue protrusion compared to untreated DS children evaluated by video registration (12).

After 4 years of treatment with PPT, we reported an improved oral-motor function in the treated DS group compared to the untreated DS group (13).

The aim of the present study was to further evaluate the effects of PPT on oral motor function, articulation, and communicative preferences after 4 years of therapy in a group of DS children by comparing the results from an extra- and intraoral examination, a speech assessment test, and a parental questionnaire.

Materials and methods

Patients

The patients participating in the study were 20 children with DS referred to the Department of Pediatric Dentistry, Karolinska Institutet. The parents were offered the opportunity to participate in a study in which their children would start PPT at that time or after 1 year. The treatment group comprised 9 children with DS (6 M, 3 F) who were randomized for PPT (12–14). PPT was started at between 3 and 33 months of age; 4 children started therapy during their 1st year of life, 3 during their 2nd year, and 2 during their 3rd year. All children had been using the plate for at least 4 years (range 49–58 months) at the time of follow-up. Only 4 children in the CG started PPT after 1 year according to their parents wish and were excluded. The CG consisted of 11 age-matched children with DS (6 M, 5 F). The mean age of both groups was 5.6 ± 1.5 years. All children lived with their biological parents, and none suffered from any congenital malformations of the heart. Permission was obtained from the ethics board of Karolinska Institutet and Huddinge University Hospital.

Palatal plate stimulation

The palatal plates used were thin acrylic plates formed as a full denture base in patients with edentulous jaws; if the patients had teeth, spring retention elements were made. The plates were designed with stimulation areas as knobs, pearls, and bowls (6). The first and most frequently used plates were designed with a bowl-shaped depression at the A-line and vestibular knobs for the upper lip according to Castillo-Morales (6). For a limited period of time (4 and 5 months, respectively) a palatal plate with additional stimulating knobs on the lateral alveolar ridges to stimulate the side borders of the tongue was used by 2 of the patients. A plate design with a metal cube moveable on a wire in the midline of the palate to train the articulatory motor precision in the tip of the tongue was used by the 2 oldest children. These plates were used when the patients had good lip closure and a normal resting position of the tongue, which is of main importance, and also for a limited time of 5 months (13).

The parents were instructed that the children should use the plates for at least 1 h twice a day. To create space for erupting teeth, it was necessary to adjust the acrylic base, and in 4 children there was a short interruption for a couple of months during the most intense eruption of deciduous teeth due to retention problems.

All the children in both groups had followed a special physiotherapy program for orofacial stimulation from birth (7, 8), which was one of the inclusion criteria for participation in the study (14), and had been continuously checked during visits to the pediatric dentist. The children also received adequate support from their parents in the therapy.

Orofacial structure appearance

An extra- and intraoral examination form was developed in order to observe anatomical and functional conditions (Table 1).

Articulation assessment

A speech assessment form was developed to investigate the influence of PPT on (a) the correct place of articulation (bilabial place for the bilabials /p, b, m/, dental place for the dentals /t, d, n, s/ and velar place for the velars /k, g, ng/); and (b) the correct manner of articulation of stops, fricatives (only /s/), and nasals.

The target consonants were assessed as: (a) correct articulation, (b) deviant articulation: linguo-interlabial, interdental, apicolabial, alveolar, other, (c) sound missing, or (d) could not be judged. Moreover, the auditory perceptual impression of velopharyngeal function was assessed as: (a) normal, (b) low intraoral pressure, and (c) nasal emission. For nasal consonants: (a) normal, (b) presence of hyponasality. Finally, the presence of vowels articulated in the front, mid, and posterior part of the mouth (front, mid, and back vowels) was noted as well as lip-rounding of rounded vowels in the words assessed. No disagreement of assessing place and manner of articulation was found between the two SLPs.

Table 1. Extra-oral and intraoral examination variables of DS children

Variables	Explanation
Facial expression	Facial expression was observed during the non-speech periods using a VAS scale: lack of vivid features – lively features.
Lip activity	(a) judged visually as anatomical short upper lip: yes/no, (b) during non-speech: active upper lip: yes/no, (c) during non-speech: active lower lip: yes/no, (d) lip-rounding when blowing a flute: yes/no, (e) lip-rounding in spontaneous speech as part of the clinical examination: present/absent.
Mouth opening	During the non-speech part of the examination: (a) mouth closed, (b) open mouth during less than half of the time, (c) open mouth during more than half of the time.
Tongue behavior	(1) During the non-speech part of the examination (a) tongue inside mouth, (b) tongue intermittently visible, (c) tongue constantly visible. (2) During spontaneous speech: tongue visible: yes/no.
Tongue anatomy	(a) normal, (b) lingual diastasis, (c) flat, (d) other.
Anatomy of hard palate	(a) staired, (b) narrow, (c) high, (d) pointed, (e) flat, (f) round.
Appearance of soft palate	Evaluated visually (light blue slit in the midline or bifid uvulae) and by palpation (notch in the midline) in order to exclude or confirm presence of submucous cleft palate.
Tonsils	Tonsil size estimated as percent of the pharyngeal space: (a) tonsils not visible or <50%, (b) 50–75%, (c) >75%, (d) tonsillectomized.
Nose breathing	Tested with the mouth shut: (a) air passage with no audible air turbulence, (b) decreased = audible air passage, (c) no air passage, (d) cannot be judged.
Malocclusions	(a) normal, (b) unilateral cross-bite, (c) bilateral cross-bite, (d) open bite, (e) prenormal tendency, (f) crowding in upper jaw, (g) crowding in lower jaw.

Parental questionnaire

A questionnaire designed for this study by two of the authors was mailed to the parents to document their experiences of their child's early and present communication habits and preferences, extent of speech training and the intelligibility of speech both to parents and to strangers. In addition, their estimations of tongue protrusion, breathing patterns, eating habit, and their children's susceptibility to infections, particularly otitis media, were asked for. The questions were to be answered on a VAS scale (visual analog scale ranging from 0 to 100, but also by multiple choice and as yes/no answers.

Clinical examination

Duration of the clinical examination was 1.5 h and was in 4 parts: an extra- and intraoral examination including oral motor function, an articulation assessment, and an estimation of communication level in terms of spoken language and manual sign language. Two independent SLPs unaware of which group the children belonged to and a pediatric dentist (KC) known to the children were involved in the clinical examination.

Articulation assessment was performed by two SLPs, one of whom also checked the parental questionnaire. Pictures and toys were used to stimulate the child to produce the specific speech sounds according to the form. Each target sound had to be produced at least twice, both assessed in the form. The 2 SLPs each filled in the articulation assessment form and compared results after the session. A tape-recording of this speech session was made and used in the event of disagreement between them. The other SLP, along with the pediatric dentist, was placed behind a one-way screen where she could listen to the speech production through earphones; she completed the articulation assessment and communication level forms. Consensus discussion for agreement was done after the clinical examination.

The extraoral variable 'facial expression' was estimated separately in terms of visual appearance of muscle tone on a VAS scale by authors KC, GH, and a blinded SLP, and presented as mean values. Consensus discussion was used for all the other extra- and intraoral parameters. The results from the blinded investigator were used in cases of disagreement (Table 1).

Oral motor function

Since overall tongue and lip behavior was of special interest, but observed during various conditions and evaluated in different ways, the results were transformed into z-scores and presented as summary variables (Figs 1–3) from the different examinations: (1) 'visible tongue' (yes/no) at the clinical examination: at articulation of target sounds in words, during spontaneous speech, at home from the parental reports, and finally during non-speech (noted as constantly, intermittently, or not visible); (2) 'lip-rounding' (yes/no) at the clinical examination during three activities: (a) when blowing a flute, (b) when articulating rounded vowels during the articulation assessment, and (c) during spontaneous speech. For 'yes', both upper and lower lip had to be noted as active.

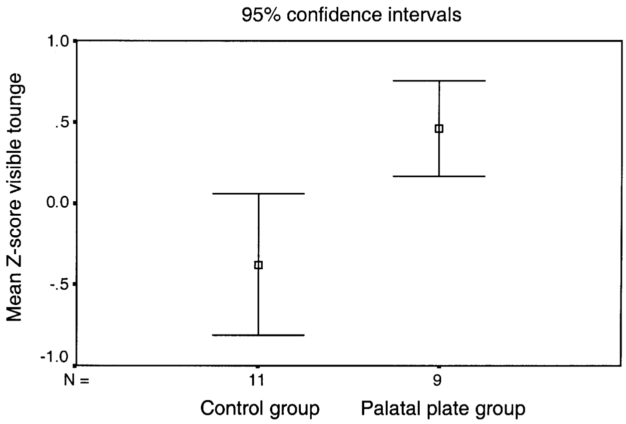

Fig. 1. All variables for the tongue visible during the clinical examination at non-speech, at articulation of target sounds in single words, during spontaneous speech and at home from the parental reports were z-transformed and summarized to the variable 'visible tongue', and compared between the palatal plate group (PPG, $n = 9$) and the control group (CG, $n = 11$). Mann-Whitney t test was used. The summarized z-score for 'visible tongue' showed a strong significance in favor of PPG ($P < 0.01$).

Communicative preferences

Many children in this study communicated using both speech and adapted manual signs. In order to study whether

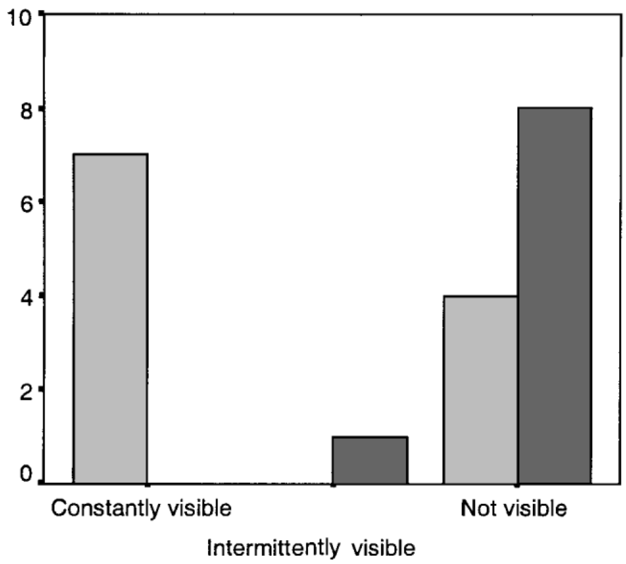

Fig. 2. 'Visible tongue' during non-speech: (a) constantly visible tongue, (b) intermittently visible tongue, and (c) not visible tongue, observed during the clinical examination, and compared between the palatal plate group (PPG, $n = 9$) and the control group (CG, $n = 11$). Fisher's exact test was used. Tongue constantly visible during non-speech time was never found in the PPG compared to 7/11 in CG ($P < 0.05$). (□ Control group; ■ palatal plate group).

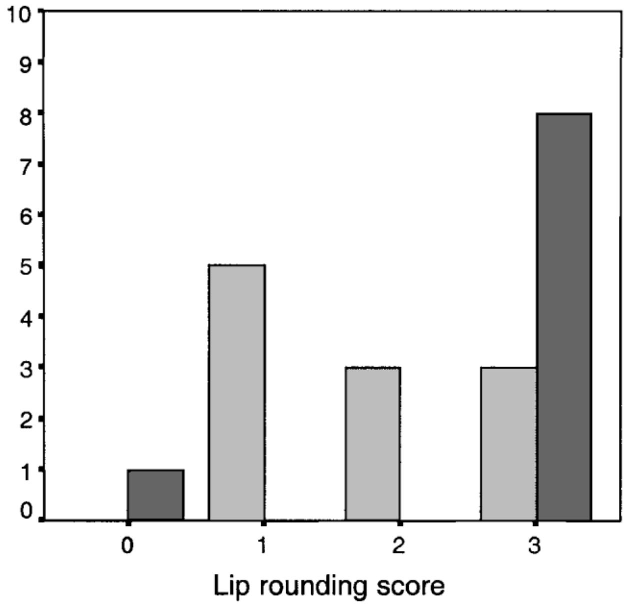

Fig. 3. Lip-rounding (both upper and lower lip activity) observed at the clinical examination for the three activities: (a) articulation of rounded vowels in single words, (b) spontaneous speech, and (c) when blowing a flute for the palatal plate group (PPG, $n = 9$) and the control group (CG, $n = 11$). Lip-rounding score: 0 = no lip-rounding, 1 = lip-rounding during one activity, 2 = lip-rounding during two activities, 3 = lip-rounding during all three activities. Fisher's exact test was used. The summary variables showed significant differences between the groups ($P < 0.01$). (□ Control group; ■ palatal plate group).

early oral speech habits (amount of sounding/babbling) and the amount of orally produced speech at the examination could be related to use of the palatal plate, the following information was collected and analyzed. (1) During the clinical examination: length of spoken sentences as well as manual sign sentences (1 word, 2 words, >2 word sentences); (2) from the parental questionnaire, communication habits at home: (a) spoken language frequency (VAS scale): 'never talking' – 'always talking', (b) preferred communication manner: spoken language/manual sign language/equal use of the two, (c) number of used words (spoken and manually signed), (d) 0–19, 20–100, and >100 words.

Age differences in the patient material

To find out if age differences could have any major influence on speech and language performance, all children were evaluated by one of the blinded SLPs as follows: without knowing age or group (PPG or CG), data (VAS scale) from the parental questionnaire (see point 2 '*Communicative preferences*'), babbling frequency and speech intelligibility within and outside the family were each scored subjectively: 1 = low, 2 = middle, and 3 = high performance as regards whole group performance. A median score (1, 2, 3) was calculated for each child. Then age and finally group identity were included.

Statistical analyses

The chi-square test was used for comparison of distributions between groups. The Fisher exact test was used in the event of fewer than 5 cases in individual cells. When more than 2 categories were compared, Fisher's trend test was used. For comparison of continuous variables between groups, the Mann-Whitney U-test was used. Student's *t* test was used for the summary variables for 'visible tongue', where the variables from all 4 examinations were transformed into z-scores in order to get one scale in common to describe the mean values. To study correlation between the summary variables for visible tongue and lip-rounding in the PPG and CG, the Spearman rank-correlation test was used. The level of significance * $P < 0.05$.

Results

Orofacial structure appearance

Nineteen of the 20 children were estimated as having a short upper lip (the exception was 1 child in the CG; Table 2). The intraoral examination showed that 16/20 children appeared with normal tongue anatomy, while 4 children, 2 in each group, demonstrated lingual diastasis (n.s.). No child was found with submucous cleft palate. Six of 9 children in the PPG and 7/11 of those in the CG had enlarged tonsils resulting in more than 50% space obstruction of the pharyngeal space, and 5 of these children in both groups had tonsils obstructing more than 75% of the space. No significant differences were found between the groups, which also included malocclusions and crowding (Table 2).

Articulation assessment

There were no significant differences between the groups

Table 2. Appearance of orofacial structures in DS children (clinical examination)

Parameters	Palatal plate group $n = 9$	Control group $n = 11$	Sign*
Upper lip short	9/9	10/11	$P > 0.9999$
Tongue normal	7/9	9/11	$P > 0.9999$
Lingual diastasis	2/9	2/11	$P > 0.9999$
Palate anatomy			
Staired	7/9	6/11	$P = 0.3742$
Pointed	3/9	5/11	$P = 0.6699$
Flat, round	3/9	2/11	$P = 0.6169$
Size of tonsils			
Non-obstructive	1/9	1/11	$P > 0.9999$
Hypertrophic >50% obstruction	6/9	7/11	$P > 0.9999$
Tonsillectomy	2/9	3/11	$P > 0.9999$
Crowding in upper jaw	4/9	8/11	$P = 0.3618$
Crowding in lower jaw	5/9	7/11	$P > 0.9999$
Unilateral cross-bite	1/9	2/11	$P > 0.9999$
Open bite	0/9	4/11	$P = 0.0941$
Prenormal tendency	3/9	5/11	$P = 0.6699$

* Fisher's exact test * $P < 0.05$.

in relation to single speech variables. All 20 children could produce stops and nasals and 16/20 (7/9 in the PPG and 9/11 in the CG) also fricative production of /s/. The /s/-sound was missing in these 4 children, 2 in each group, and in the remaining 16 children incorrectly produced, such as linguo-interlabial articulation (visible tongue, 3/9 in the PPG and 7/11 in the CG) and in a few children palatal articulation (4/9 in the PPG and 2/11 in the CG). Velar place was missing or was dentally placed in 4 children, 2 in each group. Fourteen of the 20 children (6/9 PPG, 8/11 CG) used bilabial and dental place, respectively; however, not always consistently within each target sound. Visible tongue of both the bilabial and dental speech sounds, i.e. apicolabial or linguo-interlabial articulation, was intermittently noted in 1/9 (PPG) and 3/11 (CG) and consistently noted in 2/9 in the PPG and 4/11 in the CG (n.s.)

In 3/9 children in the PPG and in 7/11 in the CG, velopharyngeal friction sounds or perceptually low intraoral pressure were noticed intermittently (n.s.). Neither hyponasal nor hypernasal speech quality was noted in any child. Nineteen of the 20 children had front, mid, and back vowels (back vowels were missing in 1 child; PPG). Rounded lips were used in 8/9 in the PPG and in 7/11 children in the CG. There were no significant differences between the groups in the separate variables.

Parental questionnaire

A visual analog scale was used to evaluate most of the answers of parents of DS children to the questionnaire. Snoring during the night was found significantly more often for children in the CG than in children treated with a palatal plate ($P < 0.05$), and fewer children in the PPG had sleep apneas and drooling during the night than had the children in the CG (n.s. Table 3). However, although not significant, a tendency in favor of PPG was found in the variable 'visible tongue' at home, with only 1 child in the PPG compared to 6 in the CG (answered by multiple choice; Fig. 1). The parents of 5 children who were tonsillectomized (Table 1) reported fewer upper airway infections, fewer sleep apneas, and less mouth breathing in their children than before tonsillectomy.

Oral motor function

Oral motor function was expressed as summary variables as well as individual variables. The summary variables for 'visible tongue' observed during the clinical examination at non-speech, at articulation of target sounds in single words, during spontaneous speech, and at home from the parental reports showed a strong significance in favor of PPG ($P < 0.01$, Fig. 1). Fig. 2 shows the distribution of visible tongue (constantly, intermittently, and not visible) for the PPG and CG during non-speech time. No child in the PPG had constantly visible tongue compared to 7/11 in the CG ($P < 0.05$). Rather, 8/9 in the PPG had no visible tongue compared to 4/11 in the CG, and only 1 PPG child had intermittently visible tongue. The summary variables for lip-rounding during articulation of rounded vowels, at spontaneous speech, and when blowing a flute showed significant differences in favor of the PPG ($P < 0.01$, Fig. 3). The only individual significant variable found for lip activity was rounding of the lips during spontaneous sentence speech, which was observed in 8/9 in the PPG and in 3/11 in the control group ($P < 0.01$). Upper and lower lip 'activity' was estimated visually during non-speech. Both upper and lower lips in 7/9 children in the PPG and in 6/11 in the CG appeared to have muscle tone (n.s.). One child from each group showed inactivity in one of the lips, while the remaining 4 in the CG were estimated to have bilabial inactivity. Since the plates were designed with pearls (for the lip) and a button for the tongue position, we correlated only lip-rounding and tongue position. Consequently, the summary variables for visible tongue and lip-rounding, respectively, were tested for correlation with each other in the PPG as well as in the CG. A positive significant correlation between the variables was found in the PPG ($r_s = 0.58$, $P < 0.01$), but not in the CG ($r_s = 0.16$ n.s.).

Regarding the variable 'mouth opening', 2/9 children in the PPG and 9/11 in the CG held the mouth open during more than 50% of the non-speech part of the examination ($P < 0.05$). Five children in both groups lacked nose breathing (no air passage through the nose), 2 in the CG had decreased nose passage, and thus 12/20 children of the whole group were mouth breathing. There were no

Table 3. Results from questionnaire to parents of DS children participating in the study. All values obtained from a visual analog scale (0–100)

Parameters	Palatal plate group (n = 9)		Control group (n = 11)		Significance#
	x	SD	x	SD	
Sleeps with open mouth	81.1	25.2	77.7	33.6	$P = 0.8616$
Snores during night	22.7	19.9	49.1	19.7	$P = 0.0321$*
Sleep apnea	13.9	23.4	36.4	37.3	$P = 0.0959$
Drooling during night	16.8	31.8	31.4	33.2	$P = 0.2146$
Mouth open during day	24.1	20.7	60.0	36.0	$P = 0.0242$*
Drooling during day	14.1	25.3	19.6	30.9	$P = 0.5820$
Susceptibility to infection	37.2	25.6	48.6	27.1	$P = 0.5581$
Susceptibility to otitis media	8.7	23.8	15.0	29.9	$P = 0.5444$
Amount of babbling/sounding	93.1	8.6	88.1	15.2	$P = 0.3145$
Speech understandable to family	59.4	27.4	72.2	18.8	$P = 0.1651$
Speech understandable to strangers	32.8	26.5	33.2	22.6	$P = 0.8738$

Mann-Whitney; # $P < 0.05$.

significant differences concerning nose breathing between the groups. Part of the clinical examination was assessment by the examiners of facial expression on a visual analog scale (Table 1). The PPG scored 75.6 ± 13.3 compared to 51.8 ± 25.7 in the CG ($P < 0.05$).

Communicative preferences

Regarding preferences of communication system, 3/9 children in the PPG and 5/11 in the CG preferred spoken language, while 4 PPG children and 2 CG children preferred adapted manual signs. Two children in the PPG and 4 in the CG used both communication systems equally. The estimated number of used signs/spoken words demonstrated no statistically significant differences (Table 4). The length of spoken and signed sentences (1-word sentences and more than 1-word sentences) was tested as part of the communication analysis. There were more children in both groups using 1-word/1-sign sentences. No statistically significant differences between the groups were found (Table 4).

Age differences in the patient material

The age range was found to be nearly equally distributed among three performance levels (1, 2, 3) in both groups.

Discussion

The principal factor of interest in this study was the influence of PPT on oral motor function, including tongue position and lip activity in children with DS. The summary variables for visible tongue in all examined activities – in non-speech, during articulation assessment, as reported from parents – showed significant differences between the groups in favor of the PPG. The effect on the variable 'constantly visible tongue during non-speech' was particularly evident, with no child found in the PPG compared to 7 in the CG. Summarizing the variables from the clinicians' and parents' assessments strengthens the results, since bias could affect the results of the clinicians, who assessed the children only during a small time corridor, whereas the parents could "assess" them all the time. On the other hand, the data given by the parents may have been influenced by wishful thinking, and therefore were less objective. A retrospective study of Hohoff & Ehmer supports the presented results of reduced tongue protrusion in DS children treated with palatal plates (15). It is a well-known fact that tongue protrusion influences the distinction of articulation negatively, as part of disordered articulation (16).

During articulation of dental and bilabial target sounds in single words, a deviant pattern with visible tongue was found, especially in CG, and tended to increase during spontaneous speech. During longer speech sequences there is a greater demand on the motor coordination of the tongue muscles, with continuous change of tongue position, than during articulation of well-known single words (16).

The summary variables for lip-rounding during the 3 activities blowing a flute, articulating rounded vowels in single words, and spontaneous speech showed significant differences between the groups in favor of PPG. Lip-rounding could be performed by most of the children during shorter sequences, such as rounding of vowels in single words or when blowing a flute, but only the PPG managed to keep the lips rounded for a longer period, such as during spontaneous speech, possibly an effect of palatal plate on articulation after 4 years of therapy. This may imply that the PPG have obtained increased muscle tone in the lips.

In the PPG, but not in the CG, there was also a positive significant correlation between the variables for lip-rounding and tongue position. The children in the PPG having the highest score for lip-rounding also had the least tongue protrusion, which we interpret as an effect of the PPT on oral motor function in this study.

An anatomical short upper lip seemed to have no importance for lip-rounding. In this study, 19/20 had a short upper lip; however, 8/9 children in PPG had an active upper lip, and there were 7/9 with activity also in the lower lip in this group. The plates equipped with vestibular knobs on the buccal surface and designed to stimulate the upper lip muscles may have had an effect on the upper lip (9–11). Effects on oral motor function after 4 years of PPT in this study can be concluded as a significant increase in the variables for lip activity, tongue position, and facial expression. More children in the PPG had the ability to articulate without VPI (velopharyngeal insufficiency), and perhaps this was due to less hypotonicity in this group, since there were no submucous clefts in either group.

There were significant differences in facial expression between the groups, which was interpreted as better muscle tone in the whole mimic musculature in the PPG,

Table 4. Communicative preferences among children with DS (from the parental questionnaire) and length of used sentences among children with DS (during the clinical examination estimated by the two speech and language pathologists)

Variables	Palatal plate group ($n = 9$)	Control group ($n = 11$)	Significance#
Number of spoken words			
0–19	4	2	$P = 0.3009$
20–99	1	4	
>100	4	5	
Number of signed words			
0–19	0	1	$P = 0.3960$
20–99	6	4	
>100	3	5	
Spoken sentences			
1-word sentences	5	6	$P = 0.4954$
2-word sentences	1	0	
>2 word sentences	3	5	
Signed sentences			
1-word sentences	5	5	$P = 0.6312$
2-word sentences	4	5	
>2-word sentences	0	1	

Chi-square trend test * $P < 0.05$.

including mm. orbicularis oris, zygomaticus major, and levator anguli oris. Children with DS have an aberrant facial appearance with lack of vivid expression due to slower motor development, hypotonic muscles, and difficulties with motor coordination compared to children without DS (9, 11). Development of the facial expression is closely connected with the total (motor, cognitive, and social) development of the child. Children with DS have a slower motor development than normal children which is known to be more delayed than mental development during the first 2 years (17). The slow motor development in DS children is related to the general hypotonia including the orofacial muscles (6, 9, 11, 17). The position of the lips has been used in studies of facial expression. The lips are the most mobile parts of the facial musculature and cooperate with the other mimic face muscles in functional units, contralaterally innervated, so that both sides can move independently of each other (18). Most methods used in research to describe and quantify facial expressions rely on the subjective impressions of an observer (19). Visual assessment by three independent observers in this study made the judgement less subjective. The upper and lower lip position and the impression of 'lively features' where the muscles seemed coordinated was important in making the judgement.

The occurrence of malocclusions in DS children increases with increasing age, owing to a combination of craniofacial growth retardation and oral motor dysfunction with poor swallowing capacity, uncoordinated, rigid, and slow muscle movements (20, 21). After 4 years, no measurable orthopedic effects on the occlusion were registered in the PPG. The small number of children in each group, sometimes less than 5, can partly explain this, and perhaps also the length of time the plates were used during the day; however, no child in the PPG had an open bite. A majority of the children, 7/9 (PPG) and 6/11 (CG), had a staired palate, which is known to be frequent in DS children, and a possible effect of the hypotonic tongue with the protruded position in the lower jaw (22).

The clinical intraoral examination revealed a high prevalence of enlarged tonsils in both groups of DS children. Eighteen of the 20 children had had enlarged tonsils, and at the time of this study 13/20 still had, while 5 children had been tonsillectomized. This coincides with habitual mouth breathing for these children. The parents of the tonsillectomized children in this study reported less sleep apneas and decreased mouth breathing during sleep after tonsillectomy. There is still resistance to adeno-tonsillectomy in DS children due both to risk of complications and the complexity of upper airway obstruction (23–26). Enlarged adenoid vegetation (not evaluated in this study) may also contribute to the breathing pattern and the open mouth habit (27). Enlarged tonsils/adenoids may limit the effect of PPT, i.e. maintain anterior tongue position and an open mouth so that airway passage is sufficient. It is possible that we would have had an even better effect on oral motor function if all children in this study had been tonsillectomized. In order to help these children develop both nose-breathing and a proper oral motor function, examination of tonsillar size (hypertrophic tonsils) should be included regularly in medical care. The 6 children in the CG who had enlarged tonsils also exhibited decreased values for facial expression, in contrast to the 7 children in the PPG with enlarged tonsils, where no correlation was found to decreased values for facial expression, a possible treatment effect. This needs to be further investigated.

One of the aims of this study was to find out whether PPT might have influenced choice of communication method, i.e. spoken or manual sign language and level of language. At articulation assessment, all children could express themselves in sentences, usually at a 1-word level, with no significant differences between spoken language and adapted manual signs. Hohoff et al. (28) showed a faster speech development of 1, 2 and multi-word sentences in a group of DS children treated with PPT, although not statistically verified, but their study did not comment on the use of other communication systems. Even though there is a desire and also a social demand for use of the spoken language, many of these children experience not being properly understood by strangers when they speak. The children communicate in the way they do best; manual signs/spoken words, a dissociation between verbal comprehension and production is apparent, since they are severely delayed in reaching the developmental stages in speech (29). The preferred form of communication may not change with better oral motor function due to plate therapy. In addition to motor ability and hearing, the social environment, motivation, and general physical condition play decisive roles (30). In this study, PPT started with a wide age range, even at the time of the examination, and this might have had an effect on various parameters of the results, especially on the outcome of speech development. However, age range did not influence the results regarding speech or language performance. Marcell et al. (31) have shown that DS children, despite their language delay, will name familiar pictures and sounds. Knowing this difficulty of repeating words, the naming ability was used successfully in this study and we could thus get good control over the tested target sounds.

The results from the parental questionnaire showed differences between the groups with regard to sleeping habits, snoring, open mouth habits and sleep apneas. It is possible that less snoring in the PPG could be dependent on an altered tongue position during sleep. Whether PPT has positive effects on breathing patterns during sleep in the long-term perspective needs to be further investigated, and also the earlier mentioned problem with enlarged tonsils/adenoids.

Articulation and language training seemed to be well developed among the children in both groups in this study, which is important for DS children (7), and because the training program was carefully followed even in CG, both groups developed successfully. Most parents in the PPG claimed that the plate therapy had facilitated eating procedures for their children, and helped develop a more vital facial expression and a higher degree of mouth closure even during the time when the children did not use the plates.

We found that after 4 years of PPT Castillo-Morales goals

(6) were partly reached with a better tongue position, activation of the contractors in the upper lip, better mouth closure and facial expression, although some results are based on fewer than 5 patients, which might influence the representativity of the results. Randomized clinical studies on mentally retarded children are difficult and time-consuming regarding compliance and cooperation, especially during certain ages and developmental stages. Furthermore, there are other medical reasons to consider in many cases. More accurate non-invasive methods for examination and evaluation remain to be developed to reproduce and measure muscle tone.

Cooperation with the parents is fundamental and a prerequisite for the treatment. It is important to bear in mind that the palatal plate is only one part in the treatment program and a complement to the basic training program for hypotonous children with DS.

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