

Research Article

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The Castillo-Morales Approach to Orofacial Pathology in Down Syndrome

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Infants with Down syndrome often present with a familiar orofacial disorder which exists at birth or becomes more pronounced by the end of the first year. The primary pathology includes hypotonicity of the perioral muscles, lips, and masticatory muscles and a protruding tongue, later followed by active tongue protrusion. This results in problems with sucking, swallowing, drooling and dentition.

Early intervention methods employing the combination of Castillo-Morales Manual Orofacial Therapy and his specially designed palatal plate, can improve orofacial function, facial appearance and prevent secondary conditions like pseudopognathism, dental diseases, malocclusions, open mouth habit and pseudomacroglossia.

This retrospective study examines the outcome of therapy, as prescribed by Castillo-Morales, in 39 children with Down syndrome. Normally, the average age to begin oral therapy is between six to eight months. The children were treated with the Castillo-Morales Manual Orofacial Therapy and his palatal plate for an average of 17.9 months. In this study, clinical evaluations at the beginning and the end of therapy focused only on open mouth posture and tongue protrusion. In addition, the direct stimulating effect of the palatal plate on tongue protrusion was evaluated. Significant positive results were observed in all three areas.

INTRODUCTION

The orofacial morphology and associate motor dysfunction found in newborns with Down syndrome have a significant impact on the eventual facial appearance and resulting oromotor problems that these children experience as they mature. Abnormalities in the oral-musculature can result in an open mouth habit and inappropriate intraoral air pressures. Proper intraoral air pressure is necessary for the coordination of velar movements with the lips and cheeks for speech articulation. Mouthbreathing is associated with an increased incidence of respiratory infections.

The characteristic oral pathology of infants with Down syndrome (Castillo-Morales, 1982; Castillo-Morales, 1985) includes open mouth breathing and hypotonicity of the tongue, resulting in tongue protrusion (Figs. 1 & 2).

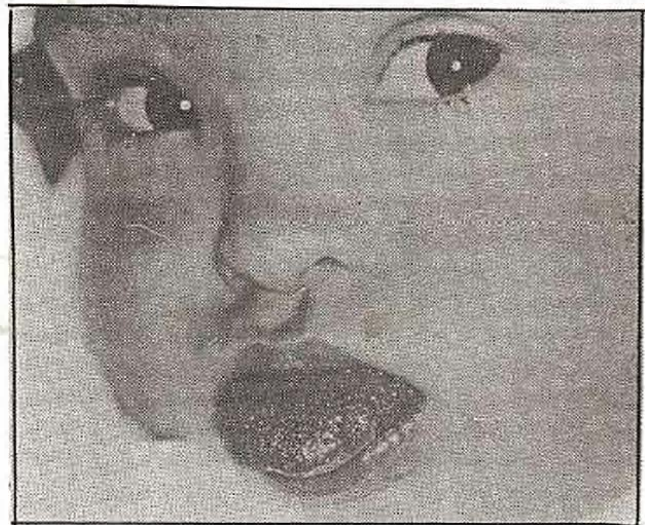


Fig 1. Twelve-month-old boy with Down syndrome. Hypotonic tongue protrudes outside the lip. Everted lower lip.

The tongue is often actively protruding during the infant's first year of development, noted particularly when crawling begins. Both factors give the impression of macroglossia, but in fact the tongue size is not increased. The tongue can easily be retracted into the oral cavity. The musculature of the tongue is not hypertrophied and the tissue consistency is not augmented, as occurs in primary macroglossia syndromes such as Beckwith-Wiedemann or EMG (Exomphalos-Macroglossia-Gigantism) syndrome (Fig. 3).

This retrospective study was designed to determine whether the Castillo-Morales Manual Orofacial Therapy, including his special palatal plate inserts, can improve mouth closure and tongue protrusion commonly seen in infants with Down syndrome. The ultimate goal of this type of therapy in children with Down syndrome is to



Fig. 2. Twenty-month-old girl with Down syndrome. Hypotonic, inactive and elevated upper lip. Everted lower lip. Tongue protrusion. Open mouth posture.

improve the facial appearance and to prevent the secondary orofacial pathology associated with chronic tongue protrusion. This may also include



Fig. 3. Sixteen-month-old girl with Beckwith-Wiedemann syndrome. Real macroglossia with augmented tissue consistency. The tongue can be retracted into the mouth, but with effort.

pseudoprognathism, teeth protrusion, dental diseases (Fischer-Grandies, 1988; Limbrock, 1988), frequent respiratory infections and motor speech problems (Wilken, 1989) (Fig. 4).

METHOD

Subjects

During a five year period, 104 children with Down syndrome (57 males and 47 females) were screened as candidates for treatment with the Castillo-Morales therapy approach, including palatal plate inserts. Of those screened, only 39 children were included in this retrospective study; the remainder were excluded due to a variety of reasons that precluded adequate analysis consistent with the study protocol.

Criteria for Inclusion in the Study

A confirmed diagnosis of Down syndrome associated with: a) tongue protrusion; b) hypotonia of the upper lip; and c) open mouth and protruding low lip.

The above characteristics occasionally occur during the first month of life, but most will become obvious by the sixth month. Therapy was begun when all three malfunctions became obvious. Ideally, it is best to begin therapy as soon as indicated.

Castillo-Morales Manual Orofacial Therapy Program

All 39 patients received the Castillo-Morales Manual Orofacial Therapy from a physiotherapist. The patients were treated for an average of 17.9 months with a range of three to forty months. The average age to begin therapy was under twelve months.

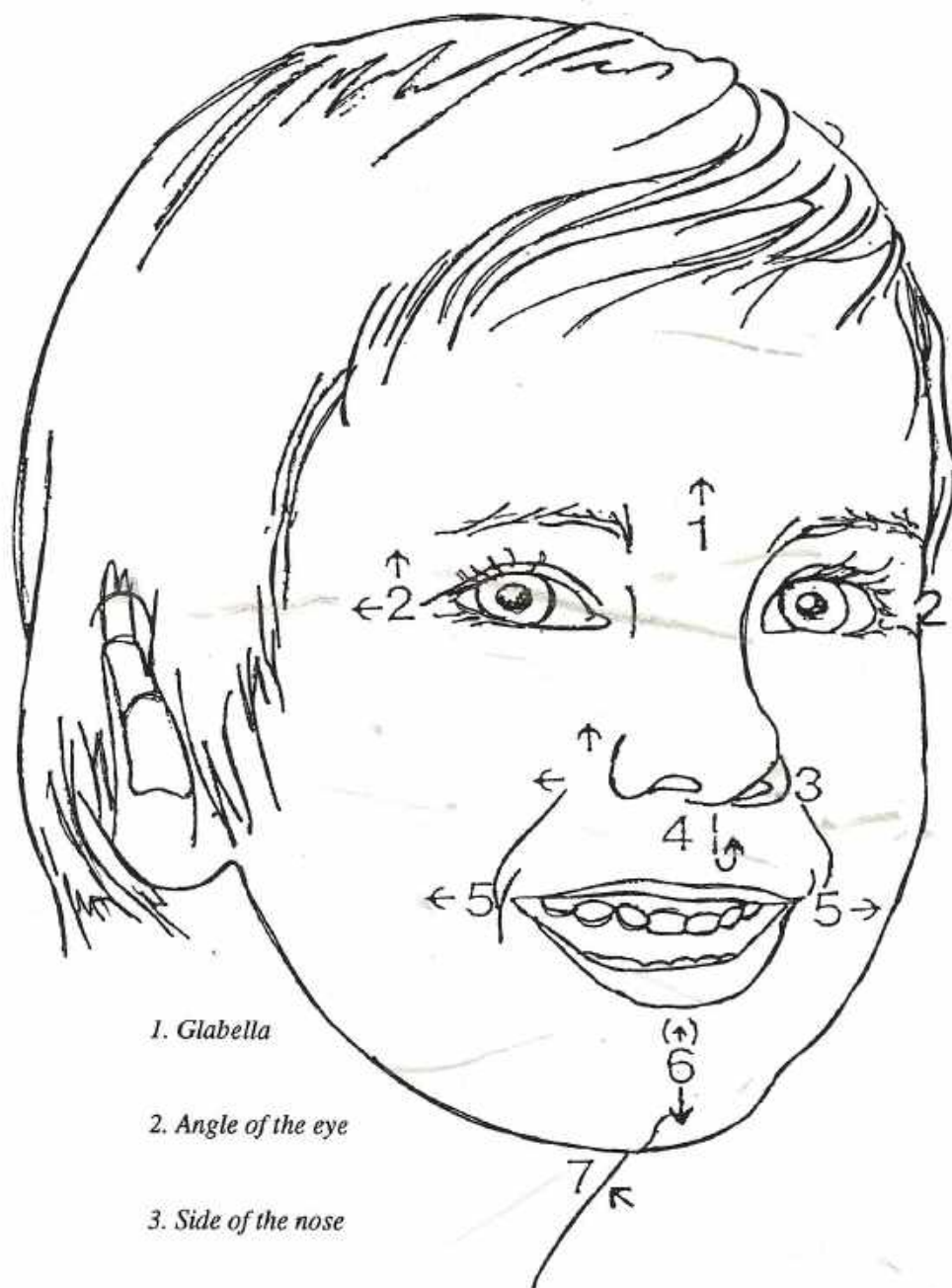
The Castillo-Morales Manual Orofacial Therapy consists of several components. Two important ones are described here: the first is called a basic muscle activation exercise and the second consists of stimulating pinpoint anatomical areas on the face. In order to obtain the greatest influence on oral function during this therapy, it is critical to maintain the position of the head and spine in a straight line, regardless of whether the child is upright or supine.

To begin the basic exercise, the child is placed on the mother's lap or on a mattress, depending on age. The head is held at the occiput by one hand, which gives a constant slight traction impulse in the mentioned straight line of the spine. In this position, the head is slightly bent, so that the cervical spine tends to be a little elongated. We call this a constant neck stretch. It is maintained during the passive bending and lateral turning of the head. While the one hand remains at the occiput, the other hand of the therapist is placed on the child's chin and rotates the head towards the shoulder. After a few seconds, up to a minute, the head is turned back to the midline. This can be repeated several times, each time turning the head in the opposite direction. In cases of hypotonic oral musculature, such as in Down syndrome, the therapist gradually waits for or provokes a slight

PRIMARY PATHOLOGIES	SECONDARY PATHOLOGIES
<p>muscle hypotonia of orbicularis, zygomatic, masseter, and temporalis muscles</p> <p>reduced tonus of muscles controlling mandibular joint</p> <p>disorders in the immune system</p> <p>lower lip everted, later even prominent</p>	<p>angle of the mouth pulled down</p> <p>upper lip inactive and pulled up, with hypoplastic lateral aspects and short frenulum</p> <p>open mouth breathing, drying; chronic periodontitis</p> <p>drooling, chapping in lower lip and angles</p> <p>respiratory infections</p>
<p>hypotonic tongue protrusion and later active tongue protrusion</p> <p>lingual diastases, bowl of the lingual blade, feeble lingual frenulum</p>	<p>tongue protrusion, or thrust, during drinking, sucking the pacifier, eating and speaking; tongue sucking</p> <p>later</p> <ul style="list-style-type: none"> • relative macroglossia, rarely true macroglossia • dry tongue surface, chapped • protrusion of upper and lower front teeth • indistinct pronunciation/articulation
<p>hypoplastic middle face</p> <p>hypoplasia of maxilla in sagittal and transverse directions</p> <p>reduced palatal height, but not a narrow palate, measurable through cephalometry</p> <p>"stair palate," eminent and remaining palatal prominences</p> <p>hypotonic velum, sometimes submucous cleft of palate and/or velum</p>	<p>tongue protrusion due to small oral cavity</p> <p>maxillary sagittal growth and palatal height remain reduced</p> <p>maxillary transverse growth reduces progressively</p> <p>later sometimes V-shaped palate, which seems to be high</p> <p>velar insufficiency, sometimes contraction in a bowl shape</p>
<p>reduced jaw angle</p> <p>reduced total length of the mandible</p> <p>retarded dentition, microdontia, anodontia, hypodontia; aberrant teeth</p>	<p>progressive reduction of jaw angle, but less reduction of mandibular growth</p> <p>open bite with dentoalveolar components; protrusion of front teeth</p> <p>"pseudoprogathia," Angle class III</p> <p>bad habit of subluxation of the jaw</p>

Fig. 4 Development of orofacial pathology in Down syndrome infants (Castillo-Morales et al. 1982)

FACIAL MOTOR POINTS of Castillo-Morales therapy



1. Glabella

2. Angle of the eye

3. Side of the nose

4. Upper lip

5. Corner of the mouth

6. Chin

7. Floor of the mouth

Fig. 5

resistance of the head movement against the hand which is placed on the child's chin. Maintaining the traction at the neck and turning the head against resistance activates the hypotonic muscles of the lips, buccinator and the tongue, thus increasing their tone and facilitating normal mobility.

The second component of therapy is based on seven facial motor points (Fig. 5): the forehead, angle of the eyes, lateral nares, middle of the upper lip, corners of the mouth, chin just below the lower lip, and the floor of the mouth. These anatomical focal points are stimulated by pressure traction, and vibration in specifically defined directions. In the beginning, they are stimulated in isolation and later in combination with the basic exercises described above.

Description of the Palatal Prosthesis

The palatal plate is a removable orthodontic device that was created by Castillo-Morales and designed to be strategically placed against the hard palate. It is specifically fabricated with two stimulatory areas that act as intraoral foreign bodies, thus eliciting the Weiffenbach Reflex (Weiffenbach, 1972).

The first area is the lingual stimulator which is located on the lingual surface of the plate. It contains a "button" that resembles a hollow concave cylinder with an external diameter of 7-8 mm. Ninety percent of Down syndrome children have a tongue diastasis which requires the use of an oval-shaped stimulator (Figs. 6 and 8). In the other 10% without tongue diastasis, a round-shaped stimulator is used.

The second area is the vestibular stimulator. It is

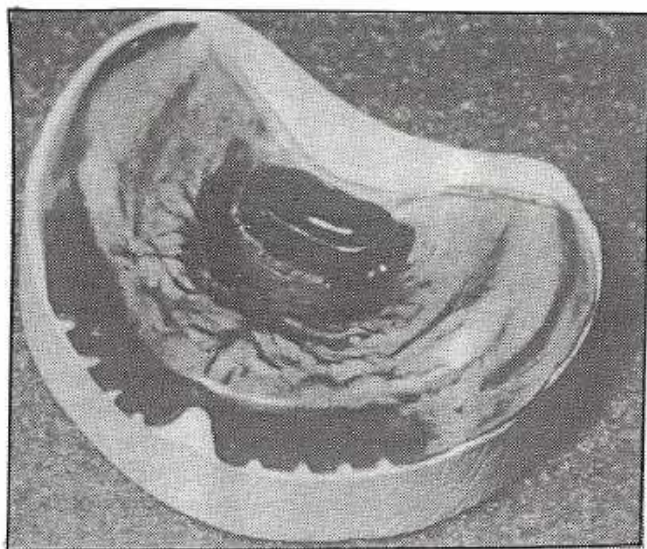


Fig. 6. Acrylic plate according to Castillo-Morales for an infant's toothless palate. The hollow cylinder at the dorsal edge stimulates tongue retraction. Oval shape accommodates the lingual diastasis of a Down syndrome infant. The ridged frontal vestibular area stimulates the upper lip.

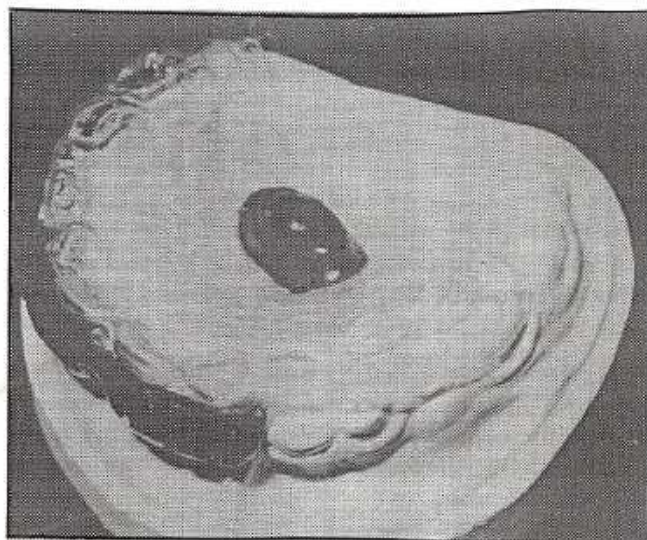


Fig. 8. The vacuum-produced palatal plate according to Castillo-Morales. In many cases, it helps to continue the palatal plate treatment in critical periods of dentition when the deciduous teeth are too short for clamp fixation.

formed by a thickening and ridging of the frontal alveolar-labial area of the plate. The tongue reacts to the plate by pressing against the hollow "button." Consequently, it retracts into the mouth and exerts a "desired" direction of force upwards and backwards. In contrast to the "pathological" direction of forward and downward. This is called the primary reaction (Hoyer, 1984) (Fig. 7).

Parents must be trained to observe whether the plate is exerting a positive effect on the tongue. It is rare for the tongue to avoid contact with the plate. However, if this occurs, it should be removed and the position of the stimulators modified. Initially, the palatal plate is inserted for a short time each day (i.e. five minutes, twice a day). As the child accommodates to the palatal insert, it is left in place for longer periods of time, typically for one hour three times per day. The average time per day in this study was 3.9 hours. Normally, the palatal plate must be refitted every three to four months to accommodate the natural growth of the mouth.

Indications for Discontinuing Treatment

Treatment should be discontinued 1) when the mouthbreathing stops and the mouth remains closed during non-verbal or feeding activities; 2) when the tongue remains inside the oral cavity; or 3) when other factors prohibit the continued use of the plate, such as the appearance of three to five upper teeth, or if the teeth are too short for clamp fixation of the palatal plate. In many situations this problem can be rectified with a pressure-formed plate that also covers the teeth (Fig. 8).

RESULTS

The combined effects of orofacial therapy and a palatal plate insert on mouth posture and tongue position were evaluated by comparing the final neuropsychiatric

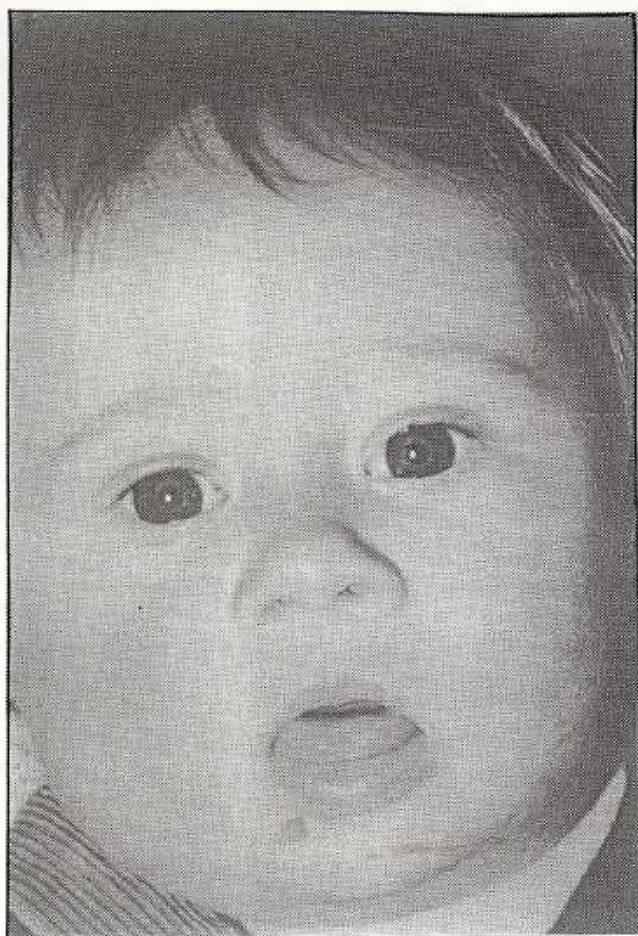


Fig. 7(a) and 7(b). So-called primary reaction. (a) Spontaneous open mouth posture, lip hypotonicity and tongue protrusion in a six-month-old girl with Down syndrome.

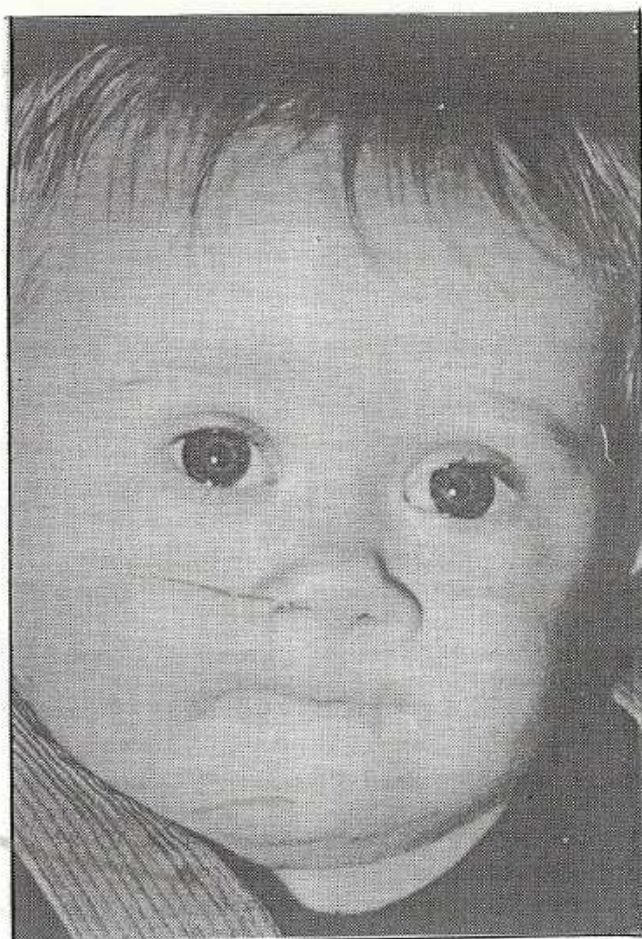


Fig 7(b) Active oral movements with retracted tongue after insertion of the palatal plate.

and orthodontic assessments with the status at the time of initial assessment. For each of the two areas of observation, the 39 cases were initially divided into two groups: mouth posture was widely open in group A and slightly open in group B. Tongue position was interlabial in group A and interalveolar in group B.

Mouth Posture

At the beginning of therapy the mouth posture was widely open in 17 cases (group A) and slightly open in 22 cases (group B). Eleven children in group A (65%) improved to a slightly open mouth posture, while six (35%) improved to complete mouth closure. This corresponds to a one degree amelioration in the first subgroup and to a two degree amelioration in the latter one. Eighteen patients (82%) of group B also showed a closed mouth posture at the end of therapy, and four patients (18%) did not change. This means a one degree amelioration in the first subgroup of group B. Both groups together showed a one degree amelioration in 74% and a two degree amelioration in 15.4% of the cases.

Tongue Position

The tongue position at the beginning of therapy was interlabial in 26 children (group A) and interalveolar in thirteen children (group B). Thirteen patients in group A (50%) improved by two degrees; their tongue remained inside the oral cavity. Twelve patients (46%) improved by one degree, i.e. to an interalveolar tongue position. Eleven patients of group B (85%) retracted their tongue into the mouth; two patients demonstrated no change. Both groups together showed a one degree amelioration in 59% and a two degree amelioration in 33%.

Additionally, eight patients were randomly selected and the time of tongue protrusion was measured during a 20 minute observation period with and without the plate. The average time of tongue protrusion with the plate was reduced by 60%; it ranged from 40% minimum to 80% maximum (see Fig. 9). This represents a high level of statistical significance ($p < 0.0005$; $t = 5.8$; $df = 7$).

DISCUSSION

Improvements in the pathologic development of oral structures in children with Down syndrome during the

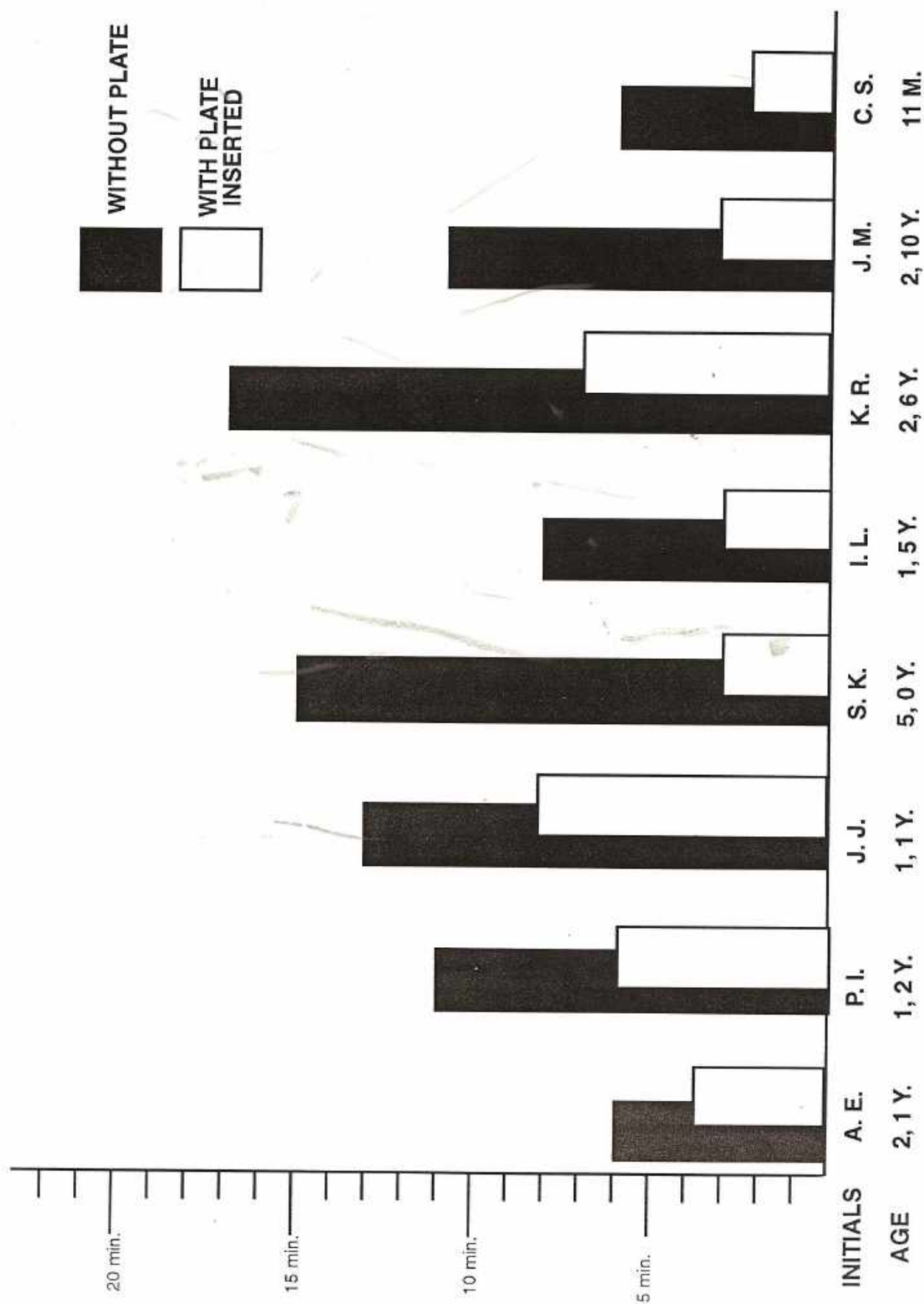


Fig. 9. Time of tongue protrusion in eight Down children in a 20 minute observation period.

course of other rehabilitation therapies are described by Bobath (1980), Vojta (1988), Garliner (1976), Haberfellner (1981) and Purdy (1987). The advantage of the Castillo-Morales therapy is the combination of a manual stimulation and facilitation program (important for the general muscular hypotonicity of children with Down syndrome) with the use of a palatal plate insert. This dual treatment approach enhances the effect of manual exercise programs. None of the rehabilitation therapies (with the exception of ISMAR, which is used only for cerebral palsy) can or even should be implemented for long periods per day. The palatal plate insert, in contrast, can be used for extended or repeated periods. The positive effect of the palatal plate on the oral motor function of children with Down syndrome is illustrated by the primary reaction after inserting the plate. Furthermore, an interruption in therapy often causes a temporary deterioration...even for short periods while a new plate is being constructed.

The Castillo-Morales therapy shares one drawback with all rehabilitation therapies: orofacial disorders are difficult to measure objectively. An interesting approach to quantify hypersalivation has been presented by the Hugh McMillan team in Toronto (Sochaniwskyj, 1982), but it requires a sophisticated technique and a cooperative patient. Garliner's semi-quantitative method to measure forces of tongue stretching forward, lip closure and muscles of mastication also require a cooperative patient. Furthermore, the utility and reliability of these measurements are often in question. Therefore, the acceptable method to document treatment outcome continues to be clinical observation (Haberfellner, 1981; Stratton, 1981). The method presented in this study excludes many biases; however, it does lack a control group. All children for whom palatal plate therapy was indicated were treated because their parents would not consent to enrollment in a non-treatment control group.

For the methodological problem of lacking a control group, Purdy et al. (1987) used an intraindividual evalua-

tion assessment as a practical alternative. The authors described two different therapeutic approaches to reduce tongue protrusion in children with Down syndrome. They treated five children, aged 21 to 31 months. Three were subjected to oral motor treatment, such as cheek and tongue tapping, according to H.A. Müller (1972). Two underwent behavior modification according to Numata (1978). In five minute periods per day the percentage of time observed with tongue protrusion was compared with a baseline assessment. With this intraindividual research design, the authors noted success in all three patients of the first group and in one child of the second group. The evaluation procedure in this study does not allow a direct comparison to Purdy et al. Fig. 9 provides data to demonstrate time measurement of the baseline tongue protrusion compared with the "primary reaction" to the inserted palatal plate. Further modifications in intraindividual evaluation assessments are anticipated in future studies.

As mentioned in the introduction, the ultimate goal of Castillo-Morales therapy in children with Down syndrome is to improve facial appearance and to prevent secondary orofacial pathology associated with chronic tongue protrusion. Until now, the requirements of such a prospective study exceed our current capabilities. Perhaps the observations from this study will encourage other centers to continue similar investigations.

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