



Effects of Midface Hypoplasia and Facial Hypotonia at Rest and During Clenching on Drooling in Down syndrome Children

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Abstract

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BACKGROUND: Down syndrome is a chromosome 21 disorder and the most common cause of physical abnormalities including midface hypoplasia, facial hypotonia, and also drooling. Drooling is unintentional anterior salivary flow from the mouth.

AIM: The objectives of the study are to determine and analyze the effects of midfacial hypoplasia and facial hypotonia on drooling in Down syndrome children.

METHODS: Of the research is analytic correlational. Sample retrievement using purposive sampling technique and obtained 20 samples that fulfills the inclusive criteria, consisting of 13 boys and 7 girls with an age range of 6-16 years old.

RESULTS AND DISCUSSION: The results were tested statistically by Kendall Coefficient of Concordance Test and Spearman Coefficient of Rank Correlation Test. The results showed that the effect of midfacial hypoplasia, facial hypotonia at rest, and during clenching on drooling is very significant (p = 0.0002) with Kendall Coefficient of Concordance. Spearman Coefficient of Rank Correlation test results show correlation of midface hypoplasia on drooling is not significant (p = 0.1265). Facial hypotonia at rest has a very significant correlation on drooling (p = 0.0000) and during clenching also has a very significant correlation (p = 0.0000).

CONCLUSION: Conclusion of the research is there are effects of midface hypoplasia, facial hypotonia at rest, and facial hypotonia during clenching on drooling, also facial hypotonia at rest and facial hypotonia during clenching on drooling, but no effect of midface hypoplasia on drooling in Down syndrome children.

Introduction

Down syndrome is a chromosome 21 disorder which has typical craniofacial features such as overall reduction in head size and brachycephaly with flattened occipital bone, narrower, less deep, and shorter face than normal individual [1], [2], [3]. The upper part of the face seems broad with decreased interorbital width, the palpebra fissures are reduced with slanted eyelids, and prominent forehead [2]. The middle part of the face (maxillary region) is hypoplastic or also referred as midface hypoplasia, with reduced vertical, lateral, and anteroposterior dimensions marked by underdevelopment in the maxillary bones and depressed nasal bridge [2], [4]. The lower part of the face has normal size of mandible (pseudoprogeny) or slightly smaller [2], [3], [5], [6], with more acute gonial angles and a more prominent position [2].

Deficient development of the midface and normal size of mandible leading Down syndrome children tend to have class III malocclusion [2], [3], [5], [7]. This causes the tongue to have downward and lingual position (relative macroglossia) since the maxilla is less developed [3], [5].

Craniofacial characteristics are strongly influenced by facial hypotonia specifically weakness of orbicularis oris, masseter, and temporalis muscles which cause significant facial changes, such as the angle of the lower mouth and open bite [8]. Relative macroglossia is also caused by facial hypotonia which lead the tongue anteriorly and downward in oral cavity [1], [3], [7], [9]. This tongue posture causes imbalance in muscle strength between lips and tongue resulting in an open anterior bite which has an impact on incompetent lip seal so that Down syndrome children tend to breathe through the mouth [3], [7], [10].

Due to an openbite anterior and incompetent lip closure, the tongue protrudes to form an oral seal which will affect the swallowing action. The swallowing action can be compromised further if the tongue is used to stabilize the mandible against the maxilla [7]. Both the protruding tongue and facial hypotonia induce hyperflexible joints and saliva flows from the mouth resulting drooling at the labial commissure [3], [7].

Drooling is the uncontrolled leakage of saliva outside the mouth [11], [12]. Drooling is normal until 18–24 months of age, [12], [13], [14], [15] though in some cases can persist up to 4 years of age, [12], [13], [16] This condition can occur as a result of neurological disorders such as cerebral palsy, Down syndrome, and mental retardation. Salivary flow is seen in about 10–38% of patients with cerebral palsy and in significant number of children with Down syndrome [11], [17].

Drooling can have a negative impact on the welfare of the patient's life, family, and caregivers. Physical and psychosocial complications that occur are maceration around the mouth, secondary bacterial infection, halitosis, dehydration, and social stigmatization. Other problems with drooling include the incidence of saliva, food, or fluid entering the lungs due to disruption of the reflex mechanism of choking and coughing [13].

Assessment on incidence of droolina needs valid and reliable measurement tool of saliva control [18], [19]. One of the methods is to give questionnaire that could either be self-administered by parents/carers or allow an interviewer to record the parents/carers' answer. Calculation of variations in drooling records from time to time can be subjectively measured by caregivers every day for a week related to drooling. To show the validity of the contents of the questionnaire, the impact of drooling was measured by the Drooling Impact Scale which had been tested for validity in accordance with the relevant statement according to the caregiver [19].

Based on the previous studies, drooling in Down syndrome children is still not clearly known the etiology purely only by midface hypoplasia or by facial hypotonia or either both so the authors are interested in examining the influence of midface hypoplasia and facial hypotonia on drooling in Down syndrome children who came to Dental Hospital of Padjadjaran University.

Subjects and Methods

Population of this study are children who have been diagnosed with Down syndrome and came to Dental Hospital of Padjadjaran University from March until May 2018. Sampling was carried out by purposive sampling with research subjects selected based on the inclusion criteria, namely Down syndrome children aged 5–16 years who have midface hypoplasia and/or facial hypotonia. The exclusion criteria were Down syndrome children who were unable to follow the instructions given and did not cooperate, has neurological disorders, and apparent craniofacial deformities.

The researcher instructed the subjects to sit upright with an occlusal line parallel to the floor

craniofacial anthropometry for measurements. Measurement of vertical facial thirds is done using a digital caliper: the upper third measured from trichion to alabella (tr-a), the middle third from alabella to subnasale (q-sn), and the lower third from subnasal to menton (sn-me), the measurements are in millimeter (mm) units (Figure 1) [20], [21], [22]. Measurements were made twice by two different people. The measurement results of each face size are summed and divided into two to obtain the average value, then the comparison of height of the upper, middle, and lower third is calculated. The measurement standard is according to the standard of East Asians, namely, the middle third is much greater than the upper third and has the same size as the lower third, and the upper third is less than the lower third [22]. Faces with the middle third less than the upper third and also the lower third of the face are evaluated as midface hypoplasia because the middle third of the face has a smaller size ratio [22], [23].

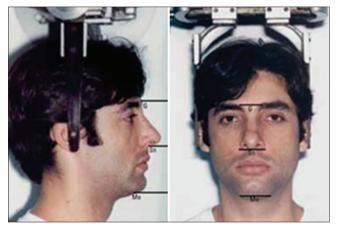


Figure 1: Vertical facial thirds [24]

Measurement of facial muscle tone was carried out using surface electromyogram/sEMG device (Electromiography Shield Arduino) programmed by Arduino 1.8.2 Program in microvolt (μ V), each surface electrode disc placed on the masseter and temporal muscles bilaterally during resting position and centric occlusion. The researcher cleansed the subject's face using a cotton pad that had been moistened with 70% alcohol. The patient was instructed to clench the teeth when the researcher palpates the masseter and temporal muscles so that the muscle contraction could be felt. The first electrode was attached on the anterior temporal muscle, second electrode on the masseter muscle, and a neutral reference electrode on the bony area of the subject's hand (Figure 2).

The electrode of the anterior temporal muscle was attached on the temple region that contract approximately 3 cm above the zygomatic arch, which is precisely on the lateral eyebrow, the electrode of the masseter muscle was attached to the middle part of the muscles that contract along the cheek bone and the corner of the mandible [25]. The right and left sides were examined separately. Patient was instructed to rest his jaw while the researcher observes the



Figure 2: Placement of surface electrode discs on masseter and anterior temporal muscles [25]

electromyogram computer screen in the measurement of facial muscle tone at rest. After the electromyogram evaluation showed the most relaxed and stable state, the researcher records the results of measurements of the right and left masseter and temporal muscles. Measurement of facial muscle tone during clenching was done by instructing the patient to bite a cotton roll on the posterior teeth as strongly as possible and the researchers records the results of measurements of the right and left masseter and temporal muscles displayed on the electromyogram computer screen. The results of recording the right and left masseter and temporal muscle tone at rest are summed and divided by four to get the mean. The same ways were done during clenching. The average measurement results are called: [26]

Normal: If the average measurement at rest is in the range of 1–2 μ V; If the average measurement during clenching is ≥100 μ V.

Hypotonia: If the average measurement at rest is <1 μV ; If the average measurement during clenching is <100 $\mu\text{V}.$

Hypertonia: If the average measurement at rest is >2 μ V. The results of measuring the facial tone on clenching were 9 (45%) hypotonia and 11 (55%) normal.

Drooling can be measured using а measurement tool of saliva control, one of which is the Drooling Impact Scale which is a subjective measurement scale regarding the frequency and severity of drooling for a week. End-anchored semantic differential scales with ten steps numbered 1-10, were chosen for each response in the form of a questionnaire that will be filled in by the parents or caregivers of the research sample. Parent or caregiver rate using a score of 1-10 which was the caregiver's assessment of the incidence rate globally, that is, score 1 if none at all, score 2-4 if occasionally and not every day, scale 5-7 if every day but not continuously, and a scale of 8-10 if every day and continuously. This scale was summed

and interpreted based on the criteria set by Cohen, that is, a value of 10 as no drooling, 11–41 as mild drooling, 42–72 as moderate drooling, and 73–100 as severe drooling [19], [27].

Researchers tested the validity and reliability of measurement results (intra examiner error). Calculation data were managed by statistical methods. Kendall Coefficient of Concordance test as a non-parametric test to find the correlation between drooling conditions on all independent variables in Down syndrome patients. Spearman Coefficient of Rank Correlation Test as a non-parametric test to find the correlation between drooling conditions on each independent variable in Down syndrome children. The significance level for the entire test is p < 0.05.

Results

Table 1 shows the description of subjects based on gender and age. Table 2 shows the description of subjects based on the midface size, facial tone at rest, facial tone during clenching, and drooling.

Characteristics	Number of subjects	Percentage
Gender		
Male	13	65
Female	7	35
Age (year-old)		
6	1	5
7	2	10
8	6	30
10	4	20
11	4	20
12	2	10
16	1	5
Total	20	100

Table 2 shows that 15 individuals (75%) presented midface hypoplasia and 5 individuals (25%) presented normal size of midface. The results of measuring the facial tone at rest were that 10 (50%) were hypotonia, 6 (30%) were normal, and 4 (20%) were hypertonia. The results of measuring the facial tone on clenching were 9 (45%) hypotonia and 11 (55%) normal.

A correlative study was done on all variables using Kendall Coefficient of Concordance (midface

Table 2: The	description (of subjects	based on	midface size,
facial tone at	rest, facial to	ne during c	lenching, a	nd drooling

Characteristics	Number of subjects	Percentage	
Midface hypoplasia	15	75	
Normal facial tone at rest	5	25	
Hypotonia	10	50	
Normal	6	30	
Hypertonia	4	20	
Facial tone during clenching			
Hypotonia	9	45	
Normal	11	55	
Drooling			
None	11	55	
Mild	4	20	
Moderate	5	25	
Severe	0	0	
Total	20	100	

hypoplasia, facial tone at rest, and facial tone on clenching) on drooling, and the result is shown in Table 3.

Table 3: Kendall coefficient of concordance analysis on midfac hypoplasia, hypotonia at rest, hypotonia on clenching on drooling

N	Df	p-value	W (%)
20	3	0.0002**)	0.333 (33.3)
o) : Not significant if p > 0.05, *): Significant if p < 0.05, **): Very significant if p < 0.01.			

Table 3 shows the results of Kendall Coefficient of Concordance analysis of the influence of midface hypoplasia, hypotonia at rest, hypotonia during clenching on drooling with low percentage linkage, 33.3% (W = 0.333) with p = 0.0002 that means very/ highly significant. The analysis on the relation between each variable on drooling using Spearman Coefficient of Rank is shown in Table 4.

Table 4: Spearman coefficient of rank analysis on midfacehypoplasia, hypotonia at rest, hypotonia during clenching ondrooling

Variable- grievance	rs	t-value	p-value	W (%)
Midface hypoplasia	-0.267	-1,17	0.12650)	7.11
Hipotonia at rest	-0.818	-6,04	0.0000**)	66.96
Hipotonia during clenching	-0.957	-14,07	0.0000**)	91.67
o) : Not significant if p > 0.05, *): Sign	nificant if p < 0.05,	**): Very significar	it if p < 0.01.	

Table 4 shows that there is no effects between midface hypoplasia and drooling but there are effects of hypotonia at rest and hypotonia during clenching on drooling. The result of Spearman Coefficient of Rank correlation test showed that midface hypoplasia had a very low linkage on drooling, only 7.11% (rs = -0.267) with p = 0.1265 that means not significant. Hypotonia at rest has a high correlation on drooling, 66.96% (rs = -0.818) with p = 0.0000 that means highly significant. Hypotonia during clenching has a strong linkage, 91.57% (rs = -0.957) with p = 0.0000 that also means highly significant.

Discussion

Table 1 shows the total number of Down syndrome children as the subject of this study; males are more than females with the youngest age of 6 years and the oldest age 16 years (most of the subjects were Down syndrome children aged 8 years, 6 of the total 20 subjects).

Table 2 shows that the total of Down syndrome children with midface hypotonia were more than those with normal face. This is similar with the study by Sujatha and Akshita [5] and Alio *et al.* [10] that found Down syndrome children generally had retarded growth caused by the smaller size of midface. A significant result was found in the study by Ferrario *et al.* [28] on 28 Italian subjects with Down syndrome.

A study by Bertelli *et al.* [29] at the Educational Hospital Sao Jose do Rio Preto Brazil found 59% Down syndrome children had hypotonia. This is nearly similar with those presented in Table 2 that 45–50% Down syndrome children had facial hypotonia. Usually, the measuring of facial tone is done at the same time (deputized) with body tone as it might occur centrally.

Midface hypoplasia can be measured by cephalometric analysis and craniofacial anthropometry. Cephalometric analysis is the main diagnostic tool with accurate method in studying and scoring the changes in growth as well as treatment of different craniofacial structure including midface [10], but it has several limitation such as in radiation that might be dangerous for health, skeletal configuration scoring that has only two dimensional, and unable in detecting most of the soft tissues, and unable to project the whole structure on one plain (usually only on midsagital plain) [28]. In contrast, craniofacial anthropometry is a noninvasive three-dimension technique that gives more complete result of the patient's [28], [32]. Craniofacial anthropometry is oftenly used on Down syndrome patients because of the simple method on quantitative scoring of anatomic changes of head and face surfaces (craniofacial) [2], [30], [31].

The midface is said to have a midface hypoplasia if the craniofacial anthropometric analysis of one-third of the face height showed less than the upper or lower part of the face as had been stated by Prendergast [22]. A study by Dagklis *et al.* [33] stated that most of the causes of midface hypoplasia on Down syndrome individuals were caused by retardation of craniofacial growth and development that might cause disorders of nasal bone ossification and genetical factor. Genes which have a role on most of the phenotypes of Down syndrome including midface hypoplasia are Hsa21 genes located in the critical region of 21q22 [34].

Other causes were shown in the study by Pacheco *et al.* [35] that stated the habit of mouth breathing during growth and development might result in vertical rise of lower part of the face, causing the individual to have smaller comparison of midface. The habit of mouth-breathing is often found on Down syndrome individual that was caused by systemic anomaly on the respiratory system [1], [3], [5].

Midface hypoplasia on Down syndrome individuals tend to cause the Grade III of jaw joint and dental maloclusion including anterior open bites, protrusion of the teeth, and imperfect mouth closing [3], [7]. Anterior open bites and imperfect lip closing cause the tongue struggling to form oral seal that will influence the action of swallowing [3], [7]. The struggling position of the tongue makes the saliva flow out from the mouth cavity through the lip's "komisura" that is called drooling [3], [7].

The study result in Table 4 shows that the statistical result of midface hypoplasia has an insignificant value on drooling with p = 0.1265. This might be as a result of the cause of drooling is multifactorial, not only

one factor, namely, midface hypoplasia. This statement was stated by Kilpatric *et al.* [36], Kumar *et al.* [37], and Bavikatte *et al.* [13] that the incorresponding position of the head and neck, and sitting position, emotional status, ability to concentrate, position and size of the tongue, diseases of mouth cavity, decreased of oral sensoric awareness, habit of mouth breathing, respiratory obstruction, the use of anticonvulsant drugs such as nitrazepam, disorders of gastric acid, rabies infection, and neuromuscular dycfunction, might cause/ trigger the drooling.

Neuromuscular dysfunction is the most general cause of drooling in the form of disorders in maintaining the saliva flowing out from the mouth cavity. In this condition, the production of saliva is normal but there is other difficulty such as to close the mouth, and less frequency of swallowing caused by hypotonia of the face tone and neck [13], [37].

The generally used examination of the muscle tone is observations, filming, and palpation; but this procedure is incompact, less corresponding the counting because the subjectivity of the professionals that may be inaccurate in counting the tone strength [38]. Electromiogram is a measuring tool with high accuracy in diagnosing motoric disorders including orofacial motoric weakness [38], [39]. In this study we used the surface electromiogram, similar with that used in the study by O'Kane [40]. which stated this tool used non-invasive method, easy to conduct, and able to evaluate the electrical activity of the muscle fibers, even though there is a limitation that it could only detect the tone which positioned near/next to the skin and not covered by other tone [25], [38], [39].

The face tone examination using surface electromyogram can only be done on the masseter and temporalis tones as the tone of mouth closing. The masseter and temporalis tones are the most touchable/ fingered facial tones, strong, and precisely positioned under the skin. Many studies have been conducted on the strength of this tone on individuals with various characteristics of vertical face. The weakness of the facial tone that is called facial hypotonus is determined by measuring the tone on rest (not in contraction) that show the minimal limit of its activity, and at the time of clenching (maximum contraction) as the maximum limit [26], [38], [42].

Table 4 shows the facial tone hypotonia in rest as well as during clenching, 66.96% and 91.67%, correlates to drooling with p = 0.0000, that it could be concluded that facial hypotonia might cause the drooling. This is similar with a statement by Kowash [7] that drooling might be caused by facial tone hypotonia that resulted as imperfect closing of the lips. This is in accordance with the study by Kumar *et al.* [37] stating the motoric therapy of orofacial tone on an individual with special needs is significantly decreased the frequency and severity of drooling.

The closing of the lips is supported by the mouth closing tones namely masseler tone, temporalis tone, and lateralis pterygoid tone, superior and inferior orbicularis oris. The lateralis pterygoid tone and orbicularis oris have weaker strength compared to masseter and temporalis, so in this study we did not measure those tones, [41] and other consideration that those tones could not be examined using the surface electromiogram tools [25], [39].

Studies by Damasceno [43], Macho *et al.* [3] revealed that midface hypoplasia and the weakness of facial tones cause the Down syndrome individuals tend to have droolings. This is similar with the result of Kendall Coefficient of Concordance in Table 3 that there was a correlation between midface hypoplasia, facial tone hypotonia in rest and during clenching to the complaint of drooling with percentage of correlation 33.3% (W = 0.333) with p = 0.0002 that indicated significant. This result was supported/strengthened by the result of the study by Vianna-Lara *et al.* [41] which stated that in those with shorter facial size, the strength activity of their facial tone was smaller.

The limitation of this study was that the total amount of sample who fulfilled the criteria was less than the total children examined, so it is less representative to the general amount of Down syndrome population. Determination of midface hypoplasia using several kinds of techniques could not give uniformed results of the studies [31]. Besides, the examination on the muscle tones were only done around the face area, meanwhile the drooling might also cause by dysphagia/ difficulty in swallowing that is related to the neck tones around the suprahyroid and infrahyroid [13], [40] The examination of the neck tone muscles need to use needle electromicrogram tools to get a more accurate information compared to the surface electromicrograms which is invasive and traumatic [44].

Conclusion

Based on the results of this study, the findings of this study show that there are effects of midface hypoplasia, facial hypotonia at rest and facial hypotonia during clenching on drooling in Down syndrome children, also facial hypotonia at rest and facial hypotonia during clenching on drooling in Down syndrome children but there is no effect of midface hypoplasia on drooling in Down syndrome children.

Suggestions given by researchers including further research on the effect of midface hypoplasia, facial hypotonia at rest, and facial hypotonia during clenching on drooling in Down syndrome children combined with cephalometric examination and measurement of neck muscle tone to obtain more thoroughly results, needs to be done on more Down syndrome children, and pediatric dentists should pay attention to the etiology of drooling so that optimal treatment planning on Down syndrome children can be obtained.

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