

Exploratory Study on the Developmental Pattern of Oral-motor Function in Children with Down's syndrome

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Abstract

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Key Words

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Children with Down's syndrome (DS) most commonly exhibit poor oral motor skills. These deficits in oral motor functioning may be attributed to abnormalities in the anatomy and physiology of the oral mechanism. The deviations in oral structures affect the development and functioning of the oral motor mechanism in this population. The present study examined the deviations in the oral motor skills in children with DS in the age group of 6 months to 60 months and compared with age matched children with developmental delay and intellectual disability (ID) without DS. An adaptation of the Scales for Oral-Motor Assessment (Rupela, 2008) was administered to the participants which consisted of rating of oral structures at rest and on non-speech tasks. The results showed that participants with DS exhibited lower scores on oral motor structures at rest. The findings indicated a developmental trend for participants with DS on oral motor function tasks in contrast to the ID group without DS. Further results of correlation analysis revealed a significant correlation between the oral motor structures at rest and oral motor function for participants with DS. These results suggest that although participants with DS and participants with ID without DS display atypical oral motor skills, they differ in oral-motor development patterns and exhibit heterogeneity within the group.

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Introduction

Down's syndrome (DS) is one of the most common genetic condition which is found to be associated with intellectual impairment and developmental disability in infants and children. The syndrome results most commonly from trisomy 21, which results from a failure in disjunction of the 21st chromosome during meiosis of embryonic development. DS is a relatively common and easily identifiable syndrome because of its unique physical and mental characteristics which facilitates in the clinical diagnosis of the condition. It is characterized by delays in all areas of development including oral motor skills. Oral motor skills develop within a system that changes rapidly both in structural growth and neurological control during the first three years of life (Arvedson & Lefton Greif, 1996; Bosma, 1986). During this period, children engage in a great variety of oral motor experiences as they satisfy their basic needs for food and comfort and begin to explore their world.

Research suggests that DS commonly display poor oral-motor skills (Spender at al. 1995). Clinically, some children with DS demonstrate difficulties with oral motor skills whereas some demonstrate difficulties with oral motor planning, and some exhibit symptoms of both (Kumin & Adams, 2000). The oral motor difficulties in this population include feeding problems and low muscle tone, in addition to motor planning difficulties (Kumin, 2002).

Studies have suggested various aetiological factors that contribute to the oral motor difficulties in children with DS.Individuals with DS have skeletal and muscular systems that differ from those without DS (Miller & Leddy, 1998; Leddy, 1999) and these differences are reported to affect speech production (Yarter, 1980; Rast & Harris, 1985; Miller, 1988). Research suggest that the speech production deficits in DS may be attributed to abnormalities in the anatomy and physiology of the oral mechanism, such as deficient growth in the bones of the head and face, hypotonia of the speech muscles, and reduced space in the oral cavity(Dodd & Thompson, 2001; Dykens, Hodapp, & Finucane, 2000; Miller & Leddy, 1998; Smith & Stoel-Gammon, 1983; Spender et al., 1995, 1996; Stoel-Gammon, 1997, 2001).

Most investigators noted tongue protrusion in children with DS, but there are differing opinions about whether the tongue is enlarged. While some have reported that the tongue is enlarged (i.e., true macroglossia) (Sommers,Patterson, & Wildgen 1988; Stoel-Gammon, 2001), others have found an average sized tongue. However, it was observed that combination of a small cavity and a normal sized tongue (i.e., relative macroglossia) may limit the distance and range of movement of the tongue (Miller & Leddy, 1998).

Open mouth posture another common feature noted in DS may result due to the presence of small oral cavity, normal-sized tongue, hypotonic lip and jaw muscles and lax ligaments (Rynders & Horrobin, 1996). Other factors that are reported to contribute to open mouth posture are frequent upper respiratory tract infections, enlarged tonsils and adenoids that may cause the need to breathe through the mouth. There may be poor jaw closure resulting in open mouth posture, jaw thrust and/or tonic jaw closure (Morris & Klein, 2000).

Hypotonia is reported to lead to functional problems such as open mouth posture, drooling, difficulties with lip closure, angle of mouth pulled down, tongue protrusion at rest, aspiration related to hypotonia of the pharyngeal musculature, and pharyngeal incoordination in DS population (Frazer & Friedman, 1996; Spender at al., 1996; Desai, 1997). General hypotonicity affects lip and tongue movements involved in all aspects of speech production.

The overall speech production of individuals with DS is affected by anatomic features specific to individuals with DS (Roberts, Price, & Malkin, 2007; Spender et al. 1995; Miller, Leddy & Leavitt, 1999; Abbeduto, Warren, & Conners, 2007). Orofacial abnormalities that may interfere with normal speech development include a small oral cavity (Benda 1949; Engler, 1949), abnormal occlusion and agenesia (McMillian & Kashgarian, 1961; Zisk & Bialer, 1967), and hypotonicity of the tongue, cheeks, and lips (Crome, Cowie, & Slater, 1966). For instance the tongue thrust in individuals with DS could possibly reduce lingual motility for speech production, while hypotonic facial muscles could limit lip movements necessary for consonant and vowel production (Miller & Leddy, 1998; Stoel-Gammon, 1997). Any one of these factors is likely to influence motor movements associated with speech, and negatively impact the articulatory and phonatory abilities of children with DS.

Spender and colleagues (1995) studied 3 twin pairs (1 child with DS and 1 non-affected child in each twin pair) between the ages of 11 and 27 months and reported oral-motor dysfunction in children with DS, such as excessive tongue protrusion, inadequate lip closure, and poorly controlled jaw function. Another study by Spender and colleagues (1996) compared the oral-motor development of 14 children with DS (ages 11 to 34 months)

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to that of 58 mental-age matched typically developing children (TDC) (ages 12 to 17 months). Similar to the first study, the authors found that the children with DS had poor jaw control and intermittent lip closure, but in this study they also noted arrhythmic tongue movements. Although research has reported abnormalities in the oral motor mechanism in DS, there is a dearth of literature exploring the development of oral motor skills in children with DS. Children with DS have been reported to demonstrate delays in oral motor development which can affect feeding and speech development. The present study aims to explore the developmental patterns of oral motor skills in children with DS across the age group of 6 months to 60 months. The study included comparison of two groups of subjects namely children with DS and children with intellectual disability without DS on the oral motor structures at rest and during non-speech tasks. The two groups represent a more homogeneous group as both the groups exhibit developmental delays which is associated with delays in the child's communicative and cognitive skills. The study expected to find atypical oral structure as well as impaired function in individual oral structures in children with DS as indicated by previous research.

 Table 1: No. of participants in each age group for

 Group I and Group II

Age Groups ^(in months)	No. of participants		
1160 oroupo	Group I	Group II	
6-12	7	2	
13-18	2	2	
19-24	2	3	
25-30	2	4	
31-36	2	3	
37-42	3	3	
43-48	5	1	
49-54	3	3	
55-60	6	2	

Method

Participants

Participants in the present study were divided into two groups namely Group I consisting of children with DS and Group II consisting of children with intellectual disability without DS. 32 participants with DS and 23 participants with intellectual disability without DS participated in the study. The participants in both the groups were divided into nine age intervals with each age interval ranging over a duration of 6 months across the age range of 6 months to 60 months. Table 1 indicates the number of participants in each age interval for the two groups. Inclusionary criteria for Group I

- Participants diagnosed with DS by a qualified paediatrician or by a Geneticist after undergoing karyotyping.
- Participants with DS in the age group of 6 to 60 months

Exclusionary criteria for Group I

Children in the DS group were excluded if any of the following criteria applied:

- Children with IQ level <50 i.e., children with severe and profound intellectual disability associated with DS were excluded from the study. For the younger age groups (below 3 years of age) the developmental quotient (DQ) was considered and children with DQ scores representing significant developmental delay were excluded from the study
- if associated physical and sensory problems (visual or auditory deficits) were present
- if there is a history of long term hospitalization due to heart diseases and major respiratory airway infections that may interfere in the child's global development.

Inclusionary criteria for Group II:

- Children with intellectual disability diagnosed by a qualified Psychologist.
- Participants were children in the age group of 6 to 60 months.

Exclusionary criteria for Group II

Children with intellectual disability were excluded if any of the following criteria applied:

- Children with IQ level <50 i.e., children with severe and profound intellectual disability were excluded from the study. For the younger age groups (below 3 years of age) the developmental quotient (DQ) was considered and children with DQ scores representing significant developmental delay were excluded from the study
- Children exhibiting recurrent episodes of seizure with regression in skills
- Children diagnosed with neuro-developmental disorders such as cerebral palsy
- Children with associated sensory problems (visual or auditory deficits).

Material

In the present study the oral motor assessment protocol included two sections namely observation of oral structures at rest and assessing function of oral mechanism during non-speech tasks. The test 'Scales for Oral-Motor Assessment' used by Rupela, V. (2008) in her unpublished doctoral thesis,was adapted for development of the oral motor assessment protocol. Modifications were incorporated to the original test to develop the oral motor assessment protocol in the present study.

The section of oral motor structures at rest consisted of eight questions based on the appearance of oral structures at rest. Oral structures of all participants were assessed for (a) placement of jaws, lips, tongue at rest, (b) presence of hypotonia, and (c) other behaviors such as drooling and involuntary movements. The second section assessed the function of oral mechanism namely the lip, jaw and tongue functions. All children were required to complete a total of 25 oral motor tasks involving isolated movements of the lips, tongue and jaw to assess the oral motor function.

Procedure

The oral structures at rest were analyzed by observing the participants when the assessment was being carried out. To assess the oral motor function the participants were instructed to imitate or spontaneously make movements involving the different oral structures as demonstrated by the examiner. Since the oral-motor assessment requires participants to follow verbal instructions, impaired cognition may adversely affect a child's performance on the assessment. Therefore, the two groups of participants were matched in their IQ levels.For participants in the younger age groups between 6-24 months the parents/ caretakers help was sought to elicit the different oral movements. If the participant was not able to copy the oral movement through visual cues, the examiner provided auditory cues with regard to the placement and movement of oral structures. If the child fails to imitate with both visual and auditory cues, the examiner provided tactile cues by physically prompting the participant to imitate the oral movement. The participant was given two trials to elicit appropriate response for each of the non-speech task under the oral motor function subsection of the protocol.

Scoring and/ or Analysis

As indicated in Table 2, a three point rating scale from 0 to 2, where response 'a' corresponded to a score of '2', 'b' a score of '1' and 'c' a score of '0' was used to assess each of the eight items on oral motor structures at rest. The scoring of observed behaviors was done by the examiner and the raw scores were tabulated. The maximum possible score for the oral motor structures subsection of the test was 16. For the oral motor function assessment, scoring was based on a four point rating scale. Each oral motor task was scored as either adult-like with only visual cues (3 points), adult-like with visual and auditory cues (2 points), approximating adult

4-point rating scale	Behaviour	Cues
3	Adult-like	Visual
2	Adult-like	Visual and auditory
1	Approximating adult behaviour	Visual, auditory and tactile
0	Deviant totally	All the three above cues

Table 2: Scoring for oral motor function domain

Table 3: Mean and SD of percentage scores for oral structures at rest across age groups for Group I and Group II

Age groups (in months) Mean		Group I	I	Age groups (in months)	Group II		
	Mean	SD	р		Mean	SD	р
6-12	76.79	12.87)	6-12	93.75	8.84)
13-18	68.75	0.00		13-18	93.75	8.84	
19-24	87.50	8.84		19-24	100.00	0.00	
25-30	68.75	0.00		25-30	96.88	6.25	
31-36	75.00	0.00	0.101	31-36	89.58	9.55	0.833
37-42	87.50	16.54		37-42	87.50	21.6	
43-48	83.75	8.39		43-48	93.75	-	
49-54	89.58	9.55		49-54	91.67	14.43	
55-60	88.54	7.31	J	55-60	93.75	0.00	J

behavior with visual, auditory and tactile cues (1 point) or deviant totally with all the cues (0 point). For the oral motor function subsection of the test the maximum score was 78. The total score obtained by the participants on the oral motor structures at rest and oral motor function domains of the test was converted into percentage score.

The data obtained was tabulated and subjected to statistical analysis using SPSS software (version 16.0). The total raw scores were calculated for the oral motor structures at rest and oral motor function subsections for participants in the two groups. The total raw scores were then converted to percentage scores and then used to obtain the mean and standard deviation. Non-parametric tests were employed to analyze the data. The Kruskal Wallis test was used to determine if there is a significant difference in performance across the age groups for participants in Group I and Group II.

Results

Oral motor Structures at Rest

For the oral motor structures at rest, the comparison across age groups in children with DS revealed no significant difference across age groups with p<0.05 level of significance. Similar results were obtained for participants with intellectual disability without DS as indicated in Table 3. The mean percentage score was highest for children with DS in the age group of 49-54 months, followed by participants in the age group of 19-24 months and 37-42 months who showed similar performance. **For** the age groups of 13-18 months and 25-30 months exhibited the lowest percentage scores. From Table 3 it is evident that participants with DS in the older age groups exhibited better percentage scores compared to younger age groups although there was variability in scores across the age groups.

Participants with intellectual disability without DS showed variable performance across the age groups with participants in the age group of 19-24 months showing the highest percentage score of 100% followed by participants in the age group of 25-30 months with a score of 96.88. The age groups of 6-12 months, 13-18 months, 43-48 months and 55-60 months showed similar mean percentage score was exhibited by participants in the age group of 37-42 months.

Oral-Motor Function

The comparison of participants with DS across age groups showed a clear developmental trend with improvement in performance with increasing age as indicated in Table 4. The results of Kruskal-Wallis test showed that there was a significant difference in performance across age groups, with p < 0.05 level of significance. Participants with intellectual disability without DS exhibited variable performance with improvement in mean percentage scores up to the age group of 25-30 months followed by a regression in the scores for the age groups from 31-48 months as shown in Table 4. There was a decline in score for the age group of 43 to 48 months in group II and this indicates the performance of a single participant in this age group. The performance of participants in the older age groups of 49-54 months and 55-60 months were similar for the two groups

Age groups (in months)M		Group I		Age groups (in months)	Group II		
	Mean	SD	р		Mean	SD	р
6-12	7.74	5.72)	6-12	4.17	1.96)
13-18	18.06	3.93		13-18	9.03	2.95	
19-24	19.44	7.86		19-24	17.13	5.78	
25-30	27.08	2.95		25-30	35.07	16.60	
31-36	40.28	5.89	0.001**	31-36	25.00	12.11	0.092
37-42	46.30	11.23		37-42	33.80	14.46	
43-48	46.39	10.96		43-48	15.28	-	
49-54	58.33	8.67		49-54	43.52	19.95	
55-60	60.42	3.90	J	55-60	43.75	34.37	J





Figure 1: Mean percentage scores across age groups for oral structures at rest for participants of Group I and Group II.

showing better mean percentage scores compared to younger age groups. However, contrary to the DS group, participants with intellectual disability without DS showed no significant difference in performance across age groups, with p<0.05 level of significance.

Correlational Analysis

Pearson's correlation co-efficient (r) was computed for participants with DS irrespective of age, to assess the relationship between oral motor structure and oral motor function domains. A significant positive correlation was found between the oral motor structures at rest and oral motor function(r= 0.545^{**} , p<0.01).

Discussion

The results of the present study revealed that there was no significant difference on oral motor structures at rest across the age groups for participants in both the groups. Participants with DS failed to achieve maximum scores on oral structure at rest which demonstrates that the oral motor structures were deviant in this population. This may be attributed to the structural deviations in oral mechanism that was consistently observed across the age groups in participants with DS. A study carried out by Barnes et al. (2006) reported that individuals with DS exhibited lower scores on oral structure than individuals with Fragile X Syndrome and TDC. Their results showed that boys with DS had the most atypical oral structures with respect to lips, tongue and velopharyngeal structure compared to individuals with Fragile X Syndrome and TDC.

It was observed that children with DS exhibited deviations in the oral structures at rest which included open jaw at rest, protracted jaw, protruded lips and open mouth posture with tongue thrust. Hypotonia of the oral musculature in participants with DS was indicated by position of the tongue (placed outside mouth) and jaw (in open position) at rest and the presence of drooling. These deviations in the oral structure in participants with DS resulted in lower than maximum overall percentage scores for the subsection of oral structures at rest. The deviations in oral structure were absent or rarely found in children with developmental delay without DS thereby exhibiting better scores compared to DS population. The participants in the age group of 19-24 months attained a maximum percentage score of 100%. However for the other age groups the percentage scores ranged between 91.67% to 96.88%. This may be attributed to the jaw position at rest, with a slightly open jaw observed in some children with intellectual disability without DS.



Figure 2: Mean percentage scores across age groups for oral structures at rest for participants of Group I and Group II.

On the oral-motor function domain participants with DS exhibited a developmental trend with an improvement in performance across age groups. However they failed to achieve maximum score which indicates that they have not acquired adult functional levels of oral motor skill. The poor performance of participants with DS on the oral motor function tasks may be supported by a vast body of literature which reports impaired oral-motor functioning in children with DS, and this may stem from hypotonia of the oral musculature and may also involve some degree of dyspraxia (Spender et al., 1995; Kumin & Bahr, 1999; Kumin & Adams, 2000). Due to the various anatomical deviations, individuals with DS generally exhibit weak motor function performances (Spender et al., 1995; Frith & Frith, 1974; Span, et al., 1999).

Participants in Group I and Group II in the younger age groups between 6 months to 24 months failed to perform the oral-motor tasks with auditory, visual and tactile cues. Hence the oral motor function was assessed through observation of spontaneous oral movements and interviewing parents about the participants oral motor functioning. However the poor performance of participants in the younger age groups on the oral motor function tasks may be attributed to the normal developmental pattern, as the oral motor skills are in the developmental stage. The other probable reason for the poor performance could be because of the impaired ability to follow instructions/ commands due to developmental delay in this population.

The variability in performance across the age groups in participants with DS may be attributed to the normal developmental trend of oral motor skills. The difference in performance may also be contributed by the variability in occurrence of different oral-structural deficits and the extent of impairment of the oral motor skills in this population irrespective of the age. These findings may be supported by studies which indicate variable percentage scores across the oral motor deviations in individuals with DS. A study by Kumin and Bahr (1999) reported open mouth posture in 71 %,

ncoral structures at rest and function indicate that the poor performance in oral motor function tasks may be due to the structural variations in oral mechanism in individuals with DS. The ability to create precise movements of the articulators for the oral function task may be influenced by the anatomical differences in the oral mechanism and hypo-

Conclusions

tion.

Children with DS present with anatomical deviations in oral motor structure which was demonstrated by lower scores on the domain of oral motor structures at rest. The function of the oral structures which include lips, tongue and jaw as assessed by the oral motor function domain were affected as a result of oral structural deviation and hypotonicity of the oral mechanism seen in participants with DS. The persisting difficulties with oral motor function in relation to the deviant oral motor structure may eventually affect their speech intelligibility and also result in speech errors. Children with DS exhibited a developmental trend with an improvement in performance on the oral motor function tasks with increasing age which may be linked to the typical process of oral motor development. However the development of oral motor skills in children with DS may be influenced by the type and severity oral motor difficulties/ deviations.

tongue thrust in 52% and hypotonia of lips and

tongue in 44% and 80% of the children. Share and

French (1993) reported a 95% occurrence of hypotonia in children with DS. Kumin and Bahr (1999)

reported drooling in 41% of the children with DS

tonicity of oral musculature seen in this popula-

The significant correlation obtained between the

in the age range of 8 months to 4.11 years.

In future, an extensive study could be carried out on a larger sample size to increase the validity of the findings of the present study. A comparison of the oral motor function between males and females could be carried out to determine if there are any gender differences in the oral motor function. Future studies could explore the oral motor function in relation to speech in DS population and investigate if oral motor function early in life could be a predictive factor for the development of speech.

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Appendix-A

I. Oral structures at rest

- 1. The child's jaw is:
 - In normal alignment
 - Slightly protracted or retracted
 - Noticeably protracted or retracted

2. The child's jaw at rest is:

- Closed
- Slightly open
- Noticeably open

3. The child is lips are:

- In a normal position
- Slightly protruded or retracted
- Obviously protruded or retracted

4. The child:

- Does not drool
- Drools, but tries to swallow it
- Drools and does not use any strategy to clear it

5. The child's tongue is:

- Placed appropriately inside the mouth
- On the bottom of the lower lip
- Outside the mouth

6. Based on the interpretation from the five items above, the oral structures seems to show

- Normal tone
- Mildly abnormal tone
- Moderately abnormal

7. Involuntary movements are:

- Absent
- Present but rarely noticeable
- Apparently present

8. When the child moves his/her oral structures:

- Other parts of the body do not move
- Other parts of the body move minimally

• Other parts of the body move noticeably and hinder in speech production

II. Function of the oral mechanism

- 1. Lip functions:
 - Rounding
 - Retraction
 - Protrusion
 - Alternate protrusion and retraction
 - Closing lips from open position
 - Pausing
 - Side to side movement of lips which is closed
 - Opening and closing lips with clenched teeth
 - Upper teeth on lower lips
 - Lower teeth on upper lips
 - Tongue out of mouth test

2. Jaw functions:

- Elevate mandible (open wide)
- Depress mandible
- Elevate and depress
- Chewing

3. Tongue functions:

- Volitional extension/stick out tongue
- Apex to left corner of mouth
- Apex to right corner of mouth
- Retracting
- Alternate retracting and protrusion
- Lateral movement (left to right corner of mouth and vise versa)
- Place the tongue in the medial position between the teeth
- Elevate the tongue to touch the upper lip
- Touch the lower lip with the tongue
- Elevate the tip of the tongue to alveolus as in producing /ta/
- Elevate the back of the tongue as in producing /ka/