Speech-5

by

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. Down syndrome 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. Children with DS demonstrate patterns of relative strengths and weaknesses in development which can be characterized as the DS behavioural phenotype (Fidler, Hepburn, & Rogers, 2006). Evidence suggests that individuals with DS tend to demonstrate a specific behavioural phenotype (Dykens, 1995; Walz & Benson, 2002; Hodapp, 2004), which begins to emerge at an early age (Fidler, Hepburn, & Rogers, 2006). Theoretically, it is important to know which aspects of the behavioral phenotype are specific to DS in order to learn more about the genetic profile of the syndrome (Abbeduto et al. 2001).

Research suggests that DS commonly display poor oral-motor skills, although little is known about specific deficits of vocal tract structure. 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. Clinically, some children with Down syndrome demonstrate difficulties with oral motor skills whereas some demonstrate difficulties with oral motor planning, and some exhibit symptoms of both (Kumin & Adams, 2000; Kumin, 2001, 2002a, 2002b, 2003a, 2003b, 2004).

Studies have suggested various aetiological factors that contribute to the oral motor difficulties in children with Down syndrome. 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). The oral motor difficulties in this population include feeding problems and low muscle tone, in addition to motor planning difficulties (Kumin, 2002). Research in DS has documented 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, any of which may have an impact on speech production in this population (Dodd & Thompson, 2001; Dykens et al., 2000; Miller & Leddy, 1998; Smith & Stoel-Gammon, 1983; Spender et al., 1995, 1996; Stoel-Gammon, 1997, 2001). Children with DS have smaller skull, missing or poorly developed midfacial bones, and a small but wide mandible (Sommers et al., 1988; Miller & Leddy, 1998; Stoel-Gammon, 2001). These structural deviations in the oral motor structures may result in delay in development of the oral motor skills in children with DS.

Most children with DS are reported to exhibit hypotonia or low muscle tone, decreased strength, and hyperextendable joints (Miller & Leddy, 1998; Kumin & Bahr, 1999; Dykens, Hodapp, & Finucane, 2000). Share & French (1993) reported that the incidence of hypotonia is over 95% in children with DS. Hypotonia is also reported to lead to many other functional problems such as open moth 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 (Frazer & Friedman, 1996; Spender at al., 1996; Desai, 1997). Due to low muscle tone and difficulties with neuromotor control, muscles of the oral mechanism are reported to be poorly dissociated.

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 et al., 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 (Kavanaugh, 1995). There may be poor jaw closure resulting in open mouth posture, jaw thrust and/or tonic jaw closure (Morris & Klein, 2000).

The speech production of many 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). Articulation movements can be negatively influenced by several factors such as smaller oral cavity than normal, hypotonic muscles around the mouth, joint lip muscles, and excessive amount of lip muscles. In individuals with DS the tongue thrust 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). Lip control allows for a strong lip seal which is affected in children with DS and difficulties with jaw control affect lip and tongue movements. General hypotonicity affects lip and tongue movements involved in all aspects of speech production. 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 Down syndrome.

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 found that the children with DS demonstrated more oral structure and oral-motor dysfunction, such as excessive tongue protrusion, inadequate lip closure, and poorly controlled jaw function. Another study by Spender and colleagues (Spender et al., 1996) compared the oral-motor development of 14 children with DS (ages 11 to 34 months) to that of 58 mental-age matched TD children (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 and oral motor control in DS, there is a lack of literature exploring the development of oral motor function 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 deviations in oral structures and the oral motor function 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.

Method

Participants

The 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 mental retardation

without DS Thirty two children with DS and twenty three children with mental retardation without DS participated in the study. Participants were diagnosed as having DS by a qualified paediatrician or by a genetician after undergoing karyotyping. Children with MR were diagnosed by a qualified Psychologist. The participants were children in the age group of 6 to 60 months. The DS group comprised of 15 males and 17 females while the group with MR comprised of 15 males and 8 females. The two groups represent a more homogeneous group as both the groups exhibit developmental delays which is associated with limitations in a child's communicative and intellectual functioning. The present 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. 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.

Children in the DS group were excluded if any of the following criteria applied: (1) if associated physical and sensory problems (visual or auditory deficits) were present (2) 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. The exclusionary criteria for participants in Group II were: (1) children with IQ level greater than moderate retardation (2) recurrent episodes of seizure exhibiting regression in skills (3) children diagnosed with neuro-developmental disorders such as cerebral palsy (d) children with associated sensory problems (visual or auditory deficits).

Development of the oral motor assessment protocol

The oral motor assessment protocol was based on previously developed test Scales for Oral-Motor Assessment used by Rupela, V. (2008) in her unpublished doctoral thesis. This test

served as the source for development of the oral motor assessment protocol for 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. Because 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. 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. The examiner will instruct the child to imitate or spontaneously make movements involving the different oral structures.

Scoring and/ or analysis:

A three point rating scale from 0 to 2 (where 2 indicated better scores) was used to assess each of the items on oral motor structures at rest and the raw scores were tabulated.

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 behavior with visual, auditory and tactile cues (1 point) or deviant totally with all the cues (0).

Table 1: *Scoring for oral motor function domain*

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

Results and discussion

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 mental retardation without DS as indicated in Table 2. 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. Participants in the age groups of 13-18 months and 25-30 months exhibited the lowest percentage scores. The DS participants in the older age groups exhibited better scores compared to younger age groups although there was a variability in scores across the age groups.

There was no specific developmental trend noted for participants with mental retardation without DS 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. Participants in the age groups of 6-12 months, 13-18 months, 43-48 months and 55-60 months showed similar mean percentage scores of 93.75. The lowest mean percentage score was exhibited by participants in the age group of 37-42 months.

Table 2: *Mean and SD of percentage scores for oral structures at rest across age groups for Group I and Group II*

Group I				Group II			
Age groups (in months)	**Mean**	**SD**	**Sig.**	**Age groups (in months)**	**Mean**	**SD**	**Sig.**
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		55-60	93.75	0.00	

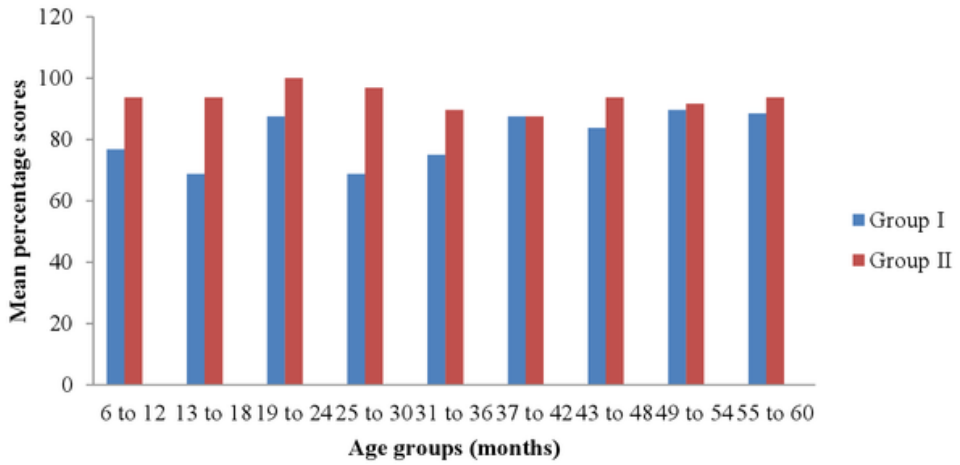

Figure 1: *Mean percentage scores across age groups for oral structures at rest for participants of Group I and Group II*

From Table 2 and Figure 1 it was noted that participants with DS did not achieve maximum scores on oral structure at rest which demonstrates that the oral motor structures were deviant compared to TDC. Similar findings were reported by Barnes et al. (2006) with individuals with DS exhibiting lower scores on oral structure than individuals with Fragile X

Syndrome and TDC. It was also observed that participants in Group II exhibited better scores on oral structures at rest compared to Group I. The difference in oral structures at rest may be attributed to the differences in skeletal and muscular systems in DS population (Miller & Leddy, 1998; Leddy, 1999). These structural variations were absent or rarely found in children with developmental delay without DS thereby exhibiting better scores compared to DS population. However the percentage scores were below 100% and ranged between 91.67% to 100%in MR without DS except for the age group of 19-24 months. The below maximum scores may be attributed to the jaw position in children with MR without DS with a slightly open jaw observed in some children.

It was found that children with DS exhibited deviations in the oral structures at rest which include open jaw at rest, protracted jaw, protruded lips, open mouth posture with tongue thrust. Hypotonia of the tongue and jaw was found in participants with DS indicated by position of tongue (placed outside mouth) drooling and jaw (in open position) at rest. These findings are in consonance with literature which reports open mouth posture (Frazer & Friedman, 1996; Desai, 1997; Kumin & Bahr, 1999; Bahr & Hillis, 2001), tongue thrust (Spender et al., 1996, Desai, 1997; Kumin & Bahr, 1999, Bahr & Hillis, 2001; Kumin et al., 2001), and hypotonia (Share & French, 1993; Kumin & Bahr, 1999) in DS population.

There was no significant difference in performance across the age groups for children with DS as structural deviations were noted in all the age groups. However there is wide variability in the results across the age groups in DS participants with participants in the older age groups exhibiting better scores. The variability in performance may be supported by the findings of the study by Kumin and Bahr (1999) which reported open mouth posture in 71 %, 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. Children with DS are often reported to exhibit drooling (Desai, 1997; Kumin & Bahr, 1999; Morris & Klein, 2000). Kumin and Bahr (1999) reported drooling in 41% of the children with DS in the age range of 8 months to 4.11 years.

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 1. 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 MR without DS exhibited an improvement in mean percentage scores up to the age of 25-30 months. However the scores were variable across age groups and the mean percentage scores reduced with variable scores between the age groups of 31 to 48 months as shown in Table 3. Similar to that of children with DS, participants in the age group of 55-60 months exhibited the highest mean percentage score. Contrary to the DS group, children with MR (without DS) showed no significant difference in performance across age groups, with $p<0.05$ level of significance.

Table 3: *Mean and SD of percentage scores for oral-motor function across age groups for Group I and Group II*

Group I				Group II			
Age groups (in months)	**Mean**	**SD**	**Sig.**	**Age groups (in months)**	**Mean**	**SD**	**Sig.**
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		55-60	43.75	34.37	

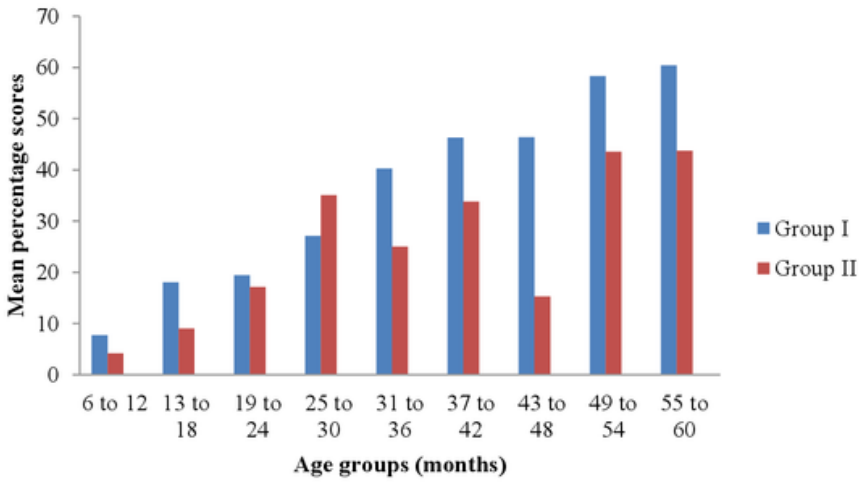

Figure 2: *Mean percentage scores across age groups for oral motor function for participants with DS and MR without DS*

From Figure 2 it may be noted that although participants with DS exhibited a developmental trend across age groups they failed to attain the maximum score. The poor performance of participants with DS on the oral motor function tasks may be supported by a vast body of literature that 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 anatomical condition, individuals with DS generally exhibit weak motor function performances (Spender et al., 1995; Frith & Frith, 1974; Spanò et al., 1999). Barnes et al (2006) found that boys with Down's syndrome showed significantly lower levels of lip, tongue, velopharynx, larynx and coordinated speech function than typically developing boys and lower levels of coordinated speech movements than boys with Fragile X syndrome.

Participants in Group I in the younger age groups across 6-12 months and 19-24 months failed to perform the oral-motor tasks with auditory, visual and tactile cues and thereby information was obtained through observation and interviewing the 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 similar to that of TDC. The other probable reason could be because of the impaired ability to follow instructions/ commands at a young age in these children.

Correlational analysis

Pearson's correlation co-efficient (r) was calculated for the performance of participants with DS irrespective of age for the oral motor structure and oral motor function domains. A significant correlation (r= 0.545**) was found between the oral motor structures at rest and oral motor function. These results of the present study indicate that children with DS exhibited impaired performance in oral motor function due to structural variations in the oral motor structures and hypotonicity of the oral muscles. This finding may be supported by the study carried out by Barnes et al. (2006) comparing the oral structure and oral motor functioning of boys with DS, Fragile-X syndrome and TDC. Both groups of boys with disorders scored lower than typically developing boys. However boys with DS had the most atypical oral structures with respect to lips, tongue and velopharyngeal structure amongst all three groups.

Conclusions:

Children with DS present with greater deviations in oral motor structure which was demonstrated by lower scores on the domain of oral motor structures at rest when compared to

children with MR without DS. The function of the oral structures of lip, tongue and jaw were affected as a result of the oral motor deviation and hypotonicity of oral musculature. There may be persisting difficulties with oral motor function in relation to the deviant oral motor structure which eventually affect their speech intelligibility and also results 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 children with DS exhibited heterogeneity within the group indicating that not all children exhibited difficulties/ deviations in the oral motor tasks and appearance of oral structures.

Future directions:

1. An extensive study could be carried out on a larger sample size to increase the validity of the findings of the present study
2. 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
3. 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:
    a) In normal alignment
    b) Slightly protracted or retracted
    c) Noticeably protracted or retracted

2. The child's jaw at rest is:
    a) Closed
    b) Slightly open
    c) Noticeably open

3. The child is lips are:
    a) In a normal position
    b) Slightly protruded or retracted
    c) Obviously protruded or retracted

4. The child
    a) Does not drool
    b) Drools, but tries to swallow it
    c) Drools and does not use any strategy to clear it

5. The child's tongue is:
    a) Placed appropriately inside the mouth
    b) On the bottom of the lower lip
    c) Outside the mouth

6. Based on the interpretation from the five items above, the oral structures seems to show
    a) Normal tone
    b) Mildly abnormal tone
    c) Moderately abnormal

7. Involuntary movements are:
    a) Absent
    b) Present but rarely noticeable
    c) Apparently present

8. When the child moves his/her oral structures:
    a) Other parts of the body do not move
    b) Other parts of the body move minimally
    c) Other parts of the body move noticeably and hinder in speech production

II. Function of the oral mechanism

1. Lip functions:

a) Rounding
b) Retraction
c) Protrusion
d) Alternate protrusion and retraction
e) Closing lips from open position
f) Pausing
g) Side to side movement of lips which is closed
h) Opening and closing lips with clenched teeth
i) Upper teeth on lower lips
j) Lower teeth on upper lips
k) Tongue out of mouth test

2. Jaw functions:

a) Elevate mandible (open wide)
b) Depress mandible
c) Elevate and depress
d) Chewing

3. Tongue functions:

a) Volition[24]xtension/stick out tongue
b) Apex to left corner of mouth
c) Apex to right corner of mouth
d) Retracting
e) Alternate retracting and protrusion
f) Lateral movement (left to right corner of mouth and vise versa)
g) Place the tongue in the medial position between the teeth
h) Elevate the tongue to touch the upper lip
i) Touch the lower lip with the tongue
j) Elevate the tip of the tongue to alveolus as in producing /ta/
k) Elevate the back of the tongue as in producing /ka/

Speech-5

ORIGINALITY REPORT

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