



Comparing the Tongue and Lip Strength and Endurance of Children with Down Syndrome with Their Typical Peers Using IOPI

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Abstract

Muscular hypotonia is a feature of Down syndrome (DS), and it affects the tongue and lips. A study on oral dysfunction in children with DS concluded that most of them did not have the tongue strength for completing the oral phase of swallowing. Recognizing the weakness of the oral muscles and improving its motor function positively affects mastication and swallowing, and prevents complications. This cross-sectional study aimed to measure the lip and tongue strength and endurance of children with DS and their typical peers, and compare these two groups with each other using Iowa Oral Performance Instrument (IOPI). Eight children with DS and 33 typical children aged 8–13 years were enrolled in this study. To examine the effect of age on the tongue strength and endurance, we divided the children into three groups of 8–9, 10–11, and 12–13 years old. The results showed that both anterior and posterior tongue strength were significantly lower in children with DS ($p=0.004$ and 0.003). But, it was not the case with tongue endurance. Also, in 10–11 years old age group, the mean posterior tongue strength and in 12–13 years old age group the lip endurance was significantly lower in children with DS ($p=0.05$ for both). Lips strength and endurance were both remarkably lower in children with DS ($p=0.004$ and 0.02). In this study, tongue, and lip strength and endurance in both children with DS and typical ones were measured with IOPI for the first time. Moreover, it provided quantitative data on the strength and endurance of the muscles of the tongue and lips, which can contribute to future studies.

Keywords Down syndrome · Iowa Oral Performance Instrument (IOPI) · Tongue · Lip · Muscle strength · Deglutition · Deglutition disorders

Introduction

Down syndrome (DS) or trisomy 21 is the most common survivable autosomal aneuploidy among newborns. The prevalence of DS varies from 0.14 to 1.66 per 1000 live births [1, 2].

Muscular hypotonia is a feature of trisomy, and it affects the tongue and lips, which is noticeable on physical examinations [3]. Many children with DS have functional problems during mastication and swallowing caused by anatomical and neuromotor dysfunction. These abnormalities can lead to aspiration, recurrent pneumonia, chronic respiratory

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illness or lung disease, poor nutrition, and failure-to-thrive, FTT [3–5].

Research shows that the prevalence of swallowing problems in people with DS varies between 31 and 80% [2, 6]. Tongue pressure is crucial for bolus clearance in the oral phase and indirectly in the pharyngeal phase of swallowing [7]. Evaluating tongue function is crucial in the assessment and rehabilitation of chewing and swallowing disorders in adults and children. A study on oral dysfunction in children with DS concluded that most of them did not have the tongue strength for completing the oral phase of swallowing [8]. Also, a study showed that the absence of jaw closure after food intake was common in the oral phase of feeding, accompanied by the difficulty in lateral movements of the tongue and reduced awareness of food on lips due to oral hyposensitivity [9].

Cross-sectional studies have shown that early recognition and rehabilitation interventions on the oral muscle strength in patients with DS promote the oral shape and the resting position of the tongue [10, 11]. A study conducted by Hashimoto et al. using pressure sensors to assess the tongue strength and pressure in adults with DS, suggested that the duration and maximum tongue pressure in participants with DS were significantly shorter and lower than the typical group [6]. To the best of our knowledge, no quantitative study has studied this issue in children with DS using IOPI. Besides, there is minimal data on the strength and endurance of lips and endurance of the tongue in children with DS. Therefore, we hypothesized that the tongue and lip strength and endurance are lower in this group. This study aimed to measure the lip and tongue strength and endurance of children with DS and their typical peers, and compare these two groups with each other using the IOPI device.

Methods

Study Design and Population

In this cross-sectional study, eight students with DS were recruited from a school established for children with special needs in Shiraz, Iran, and their typical peers ($n = 33$) were selected from typical schools.

Inclusion and Exclusion Criteria

Participants with DS who were able to follow the instructions and children without DS who did not have any other health problems such as speech, hearing, or vision problems (as tested and reported by their schools annually) and were able to follow the instructions were included in this study. Children who did not have the consent of their families, or

those with disabilities other than DS, including autism, cerebral palsy, or hearing loss were excluded.

Tools and Processes of Data Collection

Before the study started, all participants were trained individually on how to perform the test correctly. Training for children with DS took place over a course of three days. They were taught the meaning of the strength and the meaning of the endurance. Strength was defined as pressing the speech therapist's hand with their hand as hard as they could. Endurance was defined as pressing the therapist's hand as long as they could. They were taught not to be afraid of the device and/or not to bite the pulp. They were also taught how to perform each of the tests correctly. The typical children received the same training, however it took them a shorter time to learn. All tests were performed the day after the trainings for all groups were completed. The tests were performed in each of participants' school in a designated room for healthcare-related practices.

Initially data were collected on demographic information, as well as baseline information on the tongue and lip. This included data on the ability to move the tongue up, down, in, out, left, right, and rotate it as well as data on lip movements, including saying the letters “U” and “E”, kissing, smiling, and sucking. Furthermore, to examine the effect of age on the tongue strength and endurance, the children were divided into three groups, including 8–9, 10–11, and 12–13 years old ones.

The Iowa Oral Performance Instrument (IOPI) (IOPI Medical, Redmond, WA) was used [12] to measure children's anterior and posterior tongue strength. The device measure the maximum pressure that an individual can produce in a standard-sized air-filled bulb by squeezing a compliant bulb against the hard palate with the tongue for about two seconds. The units were displayed with kilopascals (kPa). All measurements were performed by a speech therapist.

In order to measure lip strength, the bulb was placed inside the cheek just lateral to the corner of the mouth and the patient squeezed the IOPI bulb against the buccal surface of the teeth by pressing the lips as hard as possible. The maximum pressure was recorded when a patient pressed their lips as hard as they could. After 10 min of rest, lip endurance was measured by quantifying the length of time that a patient could maintain 50% of his/her maximum pressure. This procedure was conducted by setting the target value in the target mode to 50% of the patient's maximum pressure and the length of the time the patient could hold the green light on. Endurance was measured three times for all anterior and posterior tongue as well as lip, with an interval

of 10 min. The mean of three measurements was considered as the maximum endurances.

Ethical Considerations

This study was performed in accordance with the ethical principles of Shiraz University of Medical Sciences (IR.SUMS.MED.REC.1398.134). Written consent was collected from parents of all participants. Participants were allowed to withdraw from the study at any stage. Parents and participants were assured of the confidentiality of their information and parents were informed of the results of the study.

Statistical Methods

Data were collected and analyzed using SPSS software version 22. We showed descriptive information in frequency and mean. Quantitative data are shown as means and standard deviations. χ^2 , Independent *T*-test, exact Fisher, Mann–Whitney, Kruskal–Wallis, and ANOVA were used to compare the groups.

Results

Demographic Review

This study included 41 children aged 8 to 13 years, with a mean age of 10.62 (± 1.51) years. In total, 8 of them (19.5%) had DS, and there were 12 boys (29.3%) and 29 girls (70.7%) participated in the study. 16.7% of the boys and 20.7% of the girls had DS. The overall mean age was 10.20 (± 1.92) years in boys and 10.79 (± 1.31) in girls. In the group with DS, the mean age 10.37 (± 1.68) years, and 2 were boys (25%) and the rest were girls (75%). In the typical group, the mean age in the typical group was 10.68 (± 1.49) years, and there were 23 girls (69.7%) and 10 boys (30.30%). There was no significant difference between the two groups in terms of age ($p = 0.58$).

Tongue and Lip Strength and Endurance

All participants with DS were able to perform lip movements, including saying letters “U” and “E”, kissing, smiling, and sucking. In addition, all had the ability to move their tongue up, down, in, out, left, right, and rotate it, except for one participant who was unable to rotate the tongue. Results showed that both anterior and posterior strength of the tongue were significantly lower in children with DS in comparison with their typical peers ($p < 0.001$ for both). Also, the mean lip strength between the two groups was significantly higher in the typical group ($p < 0.001$). Furthermore, lip endurance was significantly higher in typical

Table 1 Comparison of variables between children with Down syndrome and their typical peers

Variable (mean)	Group	<i>n</i>	Mean \pm SD	<i>p</i> value
LS ^b	DS	8	11.91 \pm 8.03	0.00
	T	33	24.15 \pm 10.08	
LE ^a	DS	5	9.00 \pm 7.31	0.02
	T	28	34.17 \pm 21.35	
ATE ^a	DS	5	6.60 \pm 5.12	0.13
	T	30	11.50 \pm 7.66	
PTE ^a	DS	5	4.20 \pm 2.16	0.20
	T	28	8.03 \pm 5.97	
ATS ^b	DS	8	13.29 \pm 7.46	0.00
	T	33	31.43 \pm 15.39	
PTS ^b	DS	8	13.48 \pm 7.47	0.00
	T	33	34.73 \pm 17.20	

LS lip strength, LE lip endurance, ATE anterior tongue endurance, PTE posterior tongue endurance, ATS anterior tongue strength, PTS posterior tongue strength, T typical, DS Down syndrome, *n* number of patients who were able to do the test

^aIn seconds

^bIn kilopascal

individuals than in children with DS ($p = 0.02$). However, the anterior and posterior endurance of the tongue did not differ significantly between the two groups (p -values were 0.20 and 0.13, respectively, Table 1).

Gender-Based Results

Gender Results Between Groups

Among the boys, anterior and posterior tongue strength and lip strength were significantly lower in children with DS ($p = 0.02$, 0.01, and 0.03, respectively) compared to the typical group. However, posterior and anterior tongue and lip endurance were not significantly different between boys with DS and typical boys (p -values were 0.15, 0.11 and 0.15, respectively). Among girls, similar to boys, the anterior and posterior tongue strength was significantly lower in children with DS ($p = 0.03$, and < 0.001 , respectively). However, there were no significant differences in the anterior and posterior tongue endurance and lip endurance and strength between the two groups ($p = 0.53$, 0.34, 0.07, and 0.06, respectively, Table 2).

Gender-Based Results in the Typical Group

In the typical group, the anterior tongue strength was significantly higher in boys ($p = 0.00$), but the posterior strength of the tongue in this typical group did not differ significantly between the sexes ($p = 0.11$). No significant difference was found between the lip endurance, anterior endurance of the

Table 2 Comparison of variables between children with Down syndrome and typical in each of the gender groups

Variable	Gender	Group	<i>n</i>	Mean ± SD	<i>p</i> value
LS ^b	M	DS	0		0.03
		T	10	26.82 ± 7.66	
	F	DS	4	19.08 ± 2.21	0.06
		T	17	25.67 ± 9.10	
LE ^a	M	DS	1	2	0.15
		T	10	32.08 ± 20.94	
	F	DS	4	10.75 ± 7.13	0.07
		T	17	35.94 ± 22.60	
ATE ^b	M	DS	1	2	0.11
		T	10	11.32 ± 6.29	
	F	DS	4	7.75 ± 5.12	0.53
		T	17	35.11 ± 7.05	
PTE ^a	M	DS	1	2	0.15
		T	10	9.00 ± 7.42	
	F	DS	4	4.75 ± 2.06	0.34
		T	17	7.64 ± 5.27	
ATS ^b	M	DS	2	9.10 ± 0.70	0.02
		T	10	42.75 ± 17.25	
	F	DS	2	14.69 ± 8.27	0.03
		T	10	29.51 ± 11.79	
PTS ^b	M	DS	2	8.30 ± 0.42	0.01
		T	10	42.02 ± 15.14	
	F	DS	2	15.21 ± 7.99	0.00
		T	10	31.56 ± 17.37	

LS lip strength, LE lip endurance, ATE anterior tongue endurance, PTE posterior tongue endurance, ATS anterior tongue strength, PTS posterior tongue strength, T typical, DS Down syndrome, *n* number of patients who were able to do the test

^aIn seconds

^bIn kilopascal

tongue, posterior tongue endurance variables and gender in this study in the typical group ($p=0.68$ and 0.75 , and 0.78).

Gender-Based Results in the Group with DS

In the group with DS, the anterior and posterior tongue strength was not significantly different between sexes ($p=0.16$ and 0.08 , respectively, Fig. 1).

In addition, only one boy with DS was able to perform posterior and anterior tongue and lip endurance testes, which was 2 s for all the three. In the group with DS, there were no significant differences in anterior and posterior tongue endurance ($p=0.15$ and 0.26), and lip endurance ($p=0.15$) between sexes.

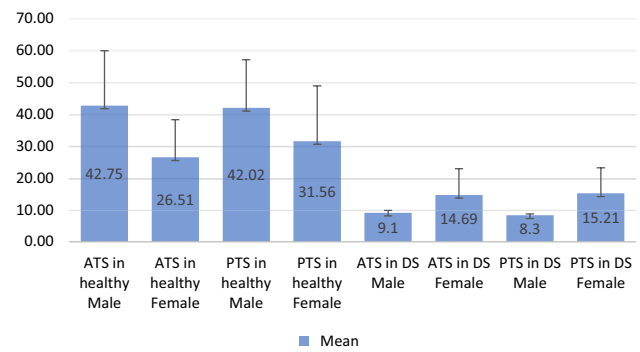


Fig. 1 Comparison of the tongue strength based on gender in each group. ATS anterior tongue strength, PTS posterior tongue strength, DS Down syndrome. ^aAll numbers are in kPa

Age-Based Results

Age Results Between Groups

Results showed that in 10–11 age group, the mean posterior tongue strength was significantly lower in children with DS ($p=0.05$) in comparison with typical individuals. However, there was no significant difference in posterior tongue strength in other age groups. Also, there were no significant differences in anterior tongue strength and lip strength in any age groups. The mean lip endurance was significantly higher in typical children than children with DS in age group of 12–13 ($p=0.05$). However, there were no significant differences in lip endurance between typical individuals and children with DS in any other age groups. Also, there were no significant differences between any of the age groups in terms of anterior and posterior tongue endurance (Table 3).

Age-Based Results in Typical Children

In typical individuals, there was no significant relationship between the anterior and posterior tongue strength and endurance and any of the age groups ($p=0.85$, 0.58 , 0.30 , 0.33 , respectively). Also, there were no significant differences between any of the age groups in terms of lip strength and endurance in the typical group ($p=0.30$ and 0.22 , respectively).

Age-Based Results in Children with DS

In the group with DS, there was no significant difference between any of the age groups in terms of anterior or posterior tongue strength ($p=0.72$ and 0.95 , respectively). Also, there was no significant relationship between the age groups and anterior endurance ($p=0.33$) of the tongue and lip strength ($p=0.09$) or lip endurance ($p=0.15$). However,

Table 3 Comparison of age variables among studied children

Variable	Age	Group	<i>n</i>	Min	Max	Mean ± SD	<i>p</i> value
PTS ^a	8–9	DS	3	7.00	21.66	12.42 ± 8.04	0.21
		T	7	5.66	51.66	28.9 ± 18.9	
	10–11	DS	3	3.66	20.66	13.55 ± 8.83	0.05
		T	13	2.33	57.66	35.22 ± 16.88	
	12–13	DS	2	8.00	22.00	15.00 ± 9.89	0.12
		T	13	11.33	60.66	37.38 ± 17.24	
ATS ^a	8–9	DS	3	3.60	18.33	10.17 ± 7.49	0.13
		T	7	7.30	50.66	29.08 ± 19.10	
	10–11	DS	3	4.60	21.33	15.31 ± 9.29	0.06
		T	13	6.60	57.33	30.98 ± 14.44	
	12–13	DS	2	9.60	20.33	14.96 ± 7.58	0.08
		T	13	13.33	65.66	33.14 ± 15.30	
PTE ^b	8–9	DS	1	7.00	7.00	7.00	0.20
		T	6	2.00	7.00	4.16 ± 1.72	
	10–11	DS	2	2.00	5.00	3.50 ± 2.12	0.09
		T	12	3.00	25.00	9.83 ± 6.65	
	12–13	DS	2	2.00	5.00	3.50 ± 2.12	0.18
		T	10	3.00	21.00	8.20 ± 6.06	
ATE ^b	8–9	DS	1	15.00	15.00	15.00	0.20
		T	6	3.00	15.00	7.16 ± 4.53	
	10–11	DS	2	3.00	7.00	5.00 ± 2.82	0.09
		T	12	5.00	20.00	11.55 ± 5.32	
	12–13	DS	2	2.00	6.00	4.00 ± 2.82	0.26
		T	12	2.00	32.00	13.63 ± 10.10	
LE ^b	8–9	DS	1	10.00	10.00	10.00	0.61
		T	6	2.00	60.00	33.77 ± 26.21	
	10–11	DS	2	7.00	21.00	14.00 ± 9.89	0.11
		T	12	7.00	60.00	42.12 ± 19.10	
	12–13	DS	2	2.00	5.00	3.50 ± 2.12	0.05
		T	10	5.00	60.00	24.86 ± 18.93	
LS ^a	8–9	DS	3	2.00	16.00	7.33 ± 7.57	0.13
		T	7	2.66	28.66	17.99 ± 9.83	
	10–11	DS	3	4.00	20.33	14.44 ± 9.06	0.09
		T	13	3.33	36.00	25.97 ± 9.75	
	12–13	DS	2	9.00	21.00	15.00 ± 8.48	0.17
		T	13	2.33	41.00	25.65 ± 10.51	

LS lip strength, *LE* lip endurance, *ATE* anterior tongue endurance, *PTE* posterior tongue endurance, *ATS* anterior tongue strength, *PTS* posterior tongue strength, *T* typical, *DS* Down syndrome, *n* number of patients who were able to do the test

^aIn kilopascal

^bIn seconds

the posterior endurance of the tongue in the group with DS increased significantly with age ($p = 0.05$).

Discussion

DS is the most common chromosomal disorder in humans [13]. Each year, about 6000 babies are born with DS, which is about 1 in every 700 [14]. Muscular hypotonia is a feature of trisomy. Commonly, it affects the tongue and lips, which

is noticeable on physical examinations [3]. The IOPI is an instrument typically used to measure tongue strength. This is the first study to use the IOPI with children with DS. In this study, we examined and compared the lip and tongue strength and endurance of children with DS and typical children. The results supported the hypothesis that the tongue strength was significantly lower in children with DS. However, this was not the case for tongue endurance. In addition, lips strength and endurance were both significantly lower in children with DS.

Our study showed that both anterior and posterior tongue strength were lower in children with DS than in typical children. This is similar to the Hashimoto et al.'s study [6], in which they used pressure sensors to assess the tongue strength and pressure in 20 to 35 years old adults with DS. The duration and maximum tongue pressure in nine participants with DS were significantly shorter and lower than the control group. In addition, regarding the tongue strength, a study carried out by Gisel et al. examined oral dysfunction in children with DS, showing that they did not have the tongue strength to complete the oral phase of swallowing [15].

In this study, the anterior strength of the tongue was lower in the group with DS than in their typical peers. The mean anterior strength of the tongue was estimated to be 13.29 kPa in children with DS and 31.43 kPa in typical children. Also, in the study of Potter et al. using IOPI on the typical children between 3 and 16 years old, the mean anterior tongue strength was 43.32 kPa [16]. The difference in the tongue strength among typical children between our study and Potter et al.'s may be due to the different age range of the studied group.

Unlike the Hashimoto et al.'s study, in which the time of tongue pressure on the hard palate's midline was shorter in adults with DS, there was no significant difference between the posterior or anterior tongue endurance between children with DS and typical individuals in our study [6].

In the present study, the mean lip strength and endurance was significantly lower in individuals with DS. A study on swallowing problems in children under five years old with DS by videofluoroscopic swallow studies (VFSS) showed that almost 20% of them had oral swallowing disorder, of whom around 28% had weak lip closure [17]. In addition, the study of Smith et al. on adults with DS suggested that impaired lip closure, tongue movements, and mastication, either in isolation or combination were seen in most of them [18]. According to their study, impaired motor coordination, insufficient lip closure, and reduced tongue mobility lead to impaired chewing and swallowing in patients with DS.

In our study on children between 8 and 13 years, there was no significant association between the age and tongue strength and endurance, either in the group with DS or in the typical group. Besides, In a study by McKay et al. on 6 to 11-year-old children, tongue strength increased significantly

from age 6 to 8 years, but did not increase after that until age 11 [19]. The results support the theory that tongue strength does not increase significantly after the age of 8 until after age of 11. It should be considered that in our study, only one child with DS was able to perform anterior and posterior tongue endurance and lip endurance in the 8–9 years old group. In this age group, the mean endurance are higher in children with DS than in typical children. This can be due to a lack of data for other children with DS who were unable to perform the test correctly.

In the present study, the effect of gender on the strength and endurance of tongue and lips was also examined. Only the anterior tongue strength in typical children was significantly higher in boys. Potter et al.'s study on the children aged 3–16 years old, reported that the boys' tongue strength was only higher in ages 14 and 16, and no significant differences in the tongue strength were observed between the sexes before that age [16]. Also, Smith et al. have shown that girls have more tongue strength by age 6, and then there is no difference between the sexes [20]. In the study of Crow et al. on adults the tongue strength was higher in men, but the tongue endurance did not differ significantly between sexes [21]. However, In McKay et al.'s study, there was no significant difference in the tongue strength between the sexes [19]. Differences in the studied population and racial differences may be the factors contributing to these differences. As it is suggested in a study on healthy Belgians adults, that mean maximum tongue pressures in European population seem to be lower than Americans and are closer to Asian results [22].

One of the limitations of this study was the small number of the studied population. Additionally, even though each measurement was performed three times to achieve reliable results, some children were unable to follow the instructions correctly at some stages. This may be due to muscle fatigue, which can indicate muscle weakness. It is worth noting that some children with DS were unable to perform endurance tests; therefore the results of endurance tests mentioned in tables should be interpreted with caution. Due to lack of similar studies using IOPI in both children with DS and typical children, it is necessary to conduct similar studies with a larger sample size and other age groups. Also, it is crucial to focus on contributing factors related to the strength and endurance of the tongue and lips such as exercises and its effects on muscle function in swallowing and speech. Furthermore, in the present study, the posterior and anterior endurance of the tongue did not differ significantly between the two groups. There is a need to examine race, eating habits, and other factors on this matter.

Conclusion

In this study, the tongue, and lips strength and endurance in both children with DS and normal children were measured using IOPI for the first time. The anterior and posterior tongue strength was significantly lower in children with DS compared to their typical peers. This was not the case for the tongue endurance. In addition, lips strength and endurance were both notably lower in children with DS. The results of the present study provided quantitative data on the strength and endurance of the tongue and lips, and it can serve future studies for suitable interventions in this targeted group.

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Declarations

Conflict of interest The authors have no conflict of interest to declare.

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