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Research Article

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DEVELOPMENT OF LIP CLOSING FUNCTION DURING TAKING FOOD INTO THE MOUTH IN CHILDREN WITH DOWN SYNDROME

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ABSTRACT

Children with Down Syndrome (DS) show developmental retardation of gross motor function including acquisition of oral movements related to eating and swallowing. To characterize the process of development/acquisition of eating/swallowing function of children with DS, interlabial pressure (IP) during taking food into the mouth was assessed. This study included 99 children with DS (birth to 4 year-old), and 112 age-matched control children showing typical development. IP during taking food into the mouth was measured as an objective index of lip closing function. The system for measuring IP during taking food into the mouth consisted of a strain gauge-pressure sensor connected to a strain-measuring device, which sent data to a personal computer installed with electromagnetic oscillograph software to display pressure waveforms. The DS and typically developing children were grouped into each age group and the data were compared between matched-age groups. IP during taking food into the mouth, pressure-time (PT), variation coefficient (VC) of IP during taking food into the mouth and VC of PT were analyzed using the unpaired t-test. Analyses showed a significantly higher IP during taking food into the mouth in the DS population than in the typically developing children in the 2 and 3 year old (P = .042 and .049, respectively) groups. No significant difference was observed between the DS and typically developing groups with respect to PT, VC of IP during taking food into the mouth or VC of PT for any age group. Children with DS showed a process of acquisition of lip closing function during taking food into the mouth similar to the process found in typically developing children, even though children with DS experience developmental retardation in gross motor and cognitive functions.

KEYWORDS: Down Syndrome, children, interlabial pressure, deglutition, eating, feeding behavior

INTRODUCTION

Following development of eating functions in infants, their suckling behavior is improved to ablactation around five-six months after birth. They acquire chewing movements before one year-old. Independent eating function is established around three years old. Then they start to eat in a manner similar to eating as adults eat (Morris & Klein, 2000). On the other hand the increased ability to close the lips during taking food into the mouth is well known to enforce eating function in typically developing children (Morris & Klein, 2000).

Development of labial functions has been well investigated with typically developing children using a relatively larger and thicker clothing button made of plastic connected with a digital tension gauge (Fukami, Saitoh, Inada, Oku, Iwase, Takemoto, Yamasaki, 2010). Many previous studies measuring lip closing force examined the maximum pressure during lip closing using specific equipment *i.e.* Myometer 160^R, or Lip De cum^R (Lambrechts, De Baets, Fieuws, Willems, 2010; Thüer & Ingervall, 1986; Ueki, Moroi, Sotobori, Ishihara, Marukawa, Iguchi, Higuchi, 2014). Therefore, it was determined under the maximum physical strength of the labial muscles which would not be feasible for infants lacking cognitive functions to understand the action to use such strength. Chigira, Omoto, Mukai, and Kaneko (1994) measured vertical lip closing force, which is naturally occurring during oral functioning by taking food into the mouth from a spoon, and demonstrated that interlabial pressure (IP) during taking food into the mouth increased with age. Their method eliminated any bias caused by producing the maximum physical strength which is a complexly subjective decision. Hence, it is considered to be applicable to very young children, as well as children with cleft lip, cleft palate (Chigira Watanabe, Nakane, Mukai, Kaneko, 1995; Kato, Funatsu, Sugiyama, Tomita, Saitou, Mukai, Inoue, 2011) or cerebral palsy (Chigira *et al.*, 1994), and elderly people in need of care (Tamura, Fukui, Kikutani, Machida, Yoshida, Yoneyama, Hamura, 2009)

In contrast, children with Down Syndrome (DS) demonstrated developmental retardation of gross motor function, particularly systemic hypotonia of muscles is one of the specific symptoms (Eid, Aly, Huneif, Ismail, 2017; Palisano, Walter, Russell, Rosenbaum, Ge´mus, Galuppi, Cunningham, 2001). These children are also slow in development of oral movements related to eating, swallowing, especially lip closure (Cooper-Brown, Copeland, Dailey, Downey, Peterson, Stimson, Van Dyke, 2008; Field, Garland, Williams, 2003). Children with DS demonstrate incomplete lip closure, tongue protrusion, swallow without chewing and spilling out food (Faulks, Collado, Mazille, Veyrune, Hennequin, 2008). However IP during taking food into the mouth has not yet been examined in the children with DS.

Lip closure plays an important role throughout the taking of food into the mouth for the swallowing processes. The functions ensure easy chewing and swallowing (Morris & Klein, 2000). Therefore it was hypothesized that the lip closing force of children with DS would be lower than that of typically developing children. Thus, this study aimed to complete the process of development/acquisition of the eating/swallowing function of DS children by measuring the IP during taking food into the mouth by using the method of Chigira *et. al*. (1994).

METHODS

Subjects

This study included ninety nine birth to 4 year old children with DS (52 boys, age range 9 to 59 months old, mean age 31.7±15.1 months old; 47 girls, age range 7 to 59 months old, mean age 31.7±14.9 months old) and one hundred and twelve birth to 4 year old children, who were free of remarkable systemic or stomatognathic abnormalities (64 boys, age range 6 to 57 months old, mean age 26.9±15.1 months old; 48 girls, age range 6 to 58 months old, mean age 35.4±16.4 months old), serving as controls. All subjects with DS were referred to our clinic for eating training, and were routinely consuming the entire quantity of food orally with no previous history of aspiration pneumonia. None of the children with DS had significant oral dryness. Data were collected prior to the eating training intervention (before the start of training). The typically developing children comprising the control group were pupils at local nursery schools with no eating problems.

In the process of subject enrollment 15 children with DS and 11 typically developing children were excluded from analysis because they refused to eat, or bit on a spoon, which is essential to measure IP with pressure waveforms during taking food into the mouth. In addition, seven of the 6 to 9 month old children with DS were excluded from analysis because they were unable to close their lips due to residual primitive feeding reflexes including: suckling and interfering oral movements; pushing out the spoon; or pushing the spoon up with the tongue. As suckling function remains longer in children with DS, more frequency is expected. Similarly, two of the 4 year old typically developing children were also excluded because of their apparently unusual eating styles (Figure 1).

The minimum required sample size for this study was calculated following Chigira *et al.* (1994). Assuming a significance level (α) of 5%, power of 80%, effect size of 3 kPa, outcome SD of 2.5 kPa and the same sample size of the two groups (1-year age groups) to be compared, the sample size for each age group was found to be 12. In the process of IP development during taking food into the mouth of typically developing children has been characterized (Chigira *et al.*, 1994), however no previous study examined IP during taking food into the mouth in children with DS. Given that children with DS show slower development of eating function although greater individual differences was found in comparison with typically developing children. The sample size for each age group was set at around 20.

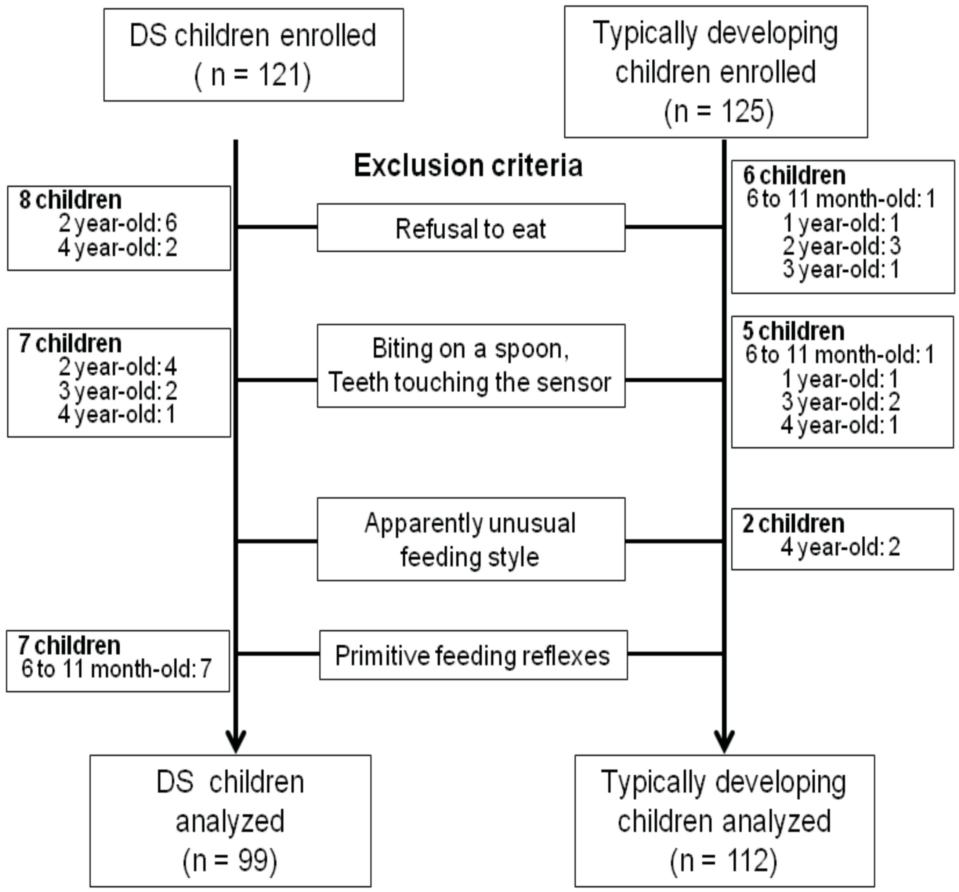

Figure 1: Flow chart of subject enrollment

Evaluation items

The baseline variables included age, sex, body height and weight, gross motor function (GMFM) (Russell, Palisano, Walter, Rosenbaum, Gemus, Gowland, Galuppi, Lane, 1998; Russell, Rosenbaum, Avery, Lane, 2002) and cognitive development (Ohta's staging) (Hashino, 1997). Body height and weight were compared between the DS and typically developing children using the growth standard based on the means and standard deviations (SD) of typically developing children in Japan (Isojima, Kato, Ito, Kanzaki, Murata, 2016). Moreover the subjects' oral cavities were also examined for the presence or absence of molar occlusion: when the occlusion was supported only by the first deciduous molar, it was classified as "F occlusal support, present;" and when supported by the first and second deciduous molars, as "S occlusal support, present". Children with unerupted or erupting first/second deciduous molars were classified as "no deciduous molar occlusal support".

Eating function of children with DS was also assessed by one dentist with more than 8 years of experience with eating/swallowing rehabilitation. The food textures eaten by the subjects and jaw movements were examined. Food textures were divided into six levels: 1) fluids, 2) pureed foods, 3) mashed foods, 4) soft pieces, 5) soft mechanicals, 6) hard mechanicals (Groher & Crary, 2016), with 1 to 4 defined as "soft food" and 5 and 6, as "solid food". Jaw movements were assessed using four grades: 1) minimal movement, 2) vertical movement, 3) diagonal rotary chew, 4) circular rotary chew (Morris & Klein, 2000). Moreover the degree of lip closure and tongue protrusion during resting time, taking food into the mouth, bolus formation or swallowing food were assessed (Omoto, Mukai, Shishikura, Ihara, Kaneko, 1986). The degree of lip closure was assessed in five grades: 1) the upper lip everted, 2) the upper lip totally immovable, 3) lips could not be closed, but moved toward closing, 4) lips could be closed occasionally, and 5) lips could be closed anytime (equivalent to the normal status). Of these, 1, 2, 3 and 4 were defined as "poor", and 5 as "good" lip closure. Similarly, the degree of tongue protrusion was assessed in four grades: 1) the tongue always protruded beyond the lips, 2) the tongue sometimes protruded beyond the lips, 3) the tongue located between the teeth and lips and 4) the tongue located behind the mandibular teeth. Of these, 1, 2 and 3 were defined as existing tongue protrusion, and 4 as not existing tongue protrusion. Typically developing children were not evaluated for assessment of eating function, since advancement process of eating function in typically developing children has been well-established (Groher & Crary, 2016; Morris & Klein, 2000). The purpose of this study focused on eating function for the children with DS.

Objective assessment of lip closure

An objective index of lip closure, IP during taking food into the mouth, was measured following the method of Chigira *et al.* (1994). This system is consisted of a strain gauge pressure sensor (PS-2KA; Kyowa Electronic Instruments, Co., Ltd., Tokyo, Japan) connected to a strain measuring device (EDX-10A and EDX-11A, Kyowa) by copper string which sent data to a personal computer installed with electromagnetic oscillograph software to display pressure waveforms. The pressure sensor to measure IP during taking food into the mouth was 0.6 mm in thickness, and was attached to a spoon such that the total thickness of the pressure measuring device was 1.5 mm. IP during taking food into the mouth is affected by the thickness of the measuring device, with thicker devices producing higher measurement values (Fukui, Kikutani, Tamura, Inaba, 2005). Therefore, to minimize the effect of the thickness of the measuring device, we embedded the pressure sensor in a flat-headed spoon, and coated it with quick-polymerizing dental resin for waterproofing. The flat head of the spoon allowed the subjects to consume the test food without experiencing discomfort. PS-2KA was calibrated following the method of Chigira *et al.*(1994), and then electric output from the sensor was recorded when 100g was loaded on PS-2KA under 23℃ and 50% humidity (Figure 2).

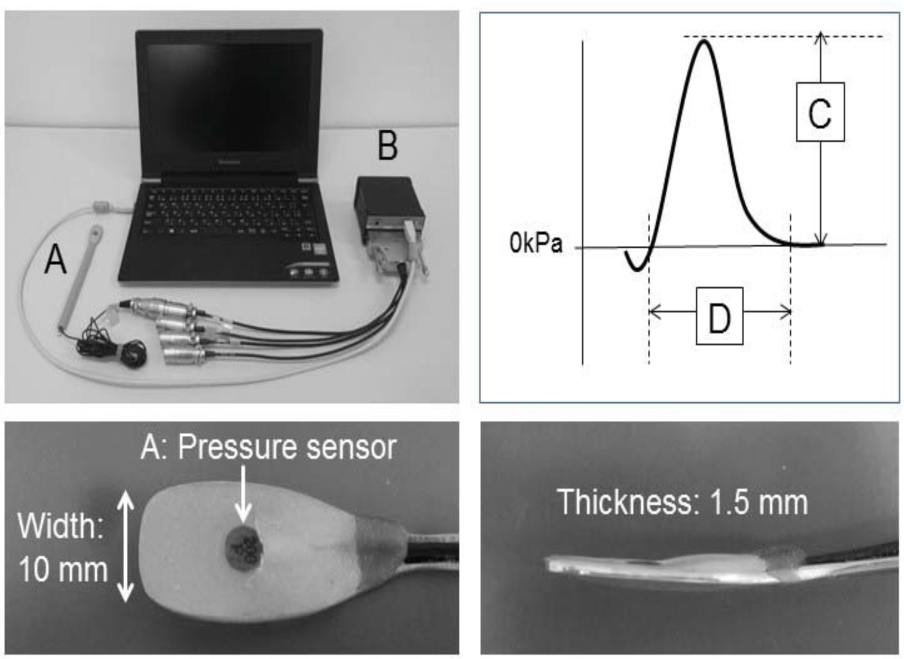

Figure 2 : **System for measuring IP during taking food into the mouth, and illustration of a pressure waveform displayed on the oscillograph**

The system consists of a strain gauge-pressure sensor (A) connected to a strain measuring device (B). The pressure sensor was embedded in a flat-headed spoon. In the waveform illustration, the vertical axis represents IP (C), and the horizontal axis represents PT (D).

Since visual and auditory stimuli had to be eliminated as they are known to affect subject's eating behaviors (Morris & Klein, 2000), all measurements were performed in a soundproof room, in which only desk and chair were, were provided, in the authors' medical institution. In addition, measurements were conducted in the presence of the subjects' parents to prevent the children becoming excessively anxious.

Measurements for taking food into the mouth were performed seven times for each child, while in a sitting position, with approximately one gram of yogurt on the sensor-embedded spoon.
In consideration of the effect of the spoon angle on IP during taking food into the mouth (Kayanaka-Sekine, Saiki, Tamura, Kikutani, Matsumoto, 2011), assistance for taking food into the mouth for all children was provided by a dentist with more than 8 years of experience in eating/swallowing rehabilitation, to ensure minimal differences in assisting behaviors. The data of the 3rd to 7th measurements, in which all children in this study were able to take the test food into the mouth, were referred for analysis. The obtained waveforms were analyzed using an analysis software program (DIAdem, National Instruments Japan Corp., Tokyo, Japan) to determine the maximum pressure as the IP during taking food into the mouth, and the time of lip contact with the pressure sensor as the pressure time (PT) (Figure 2). Also the variation coefficient (VC) of IP during taking food into the mouth, that the standard deviation (SD) of IPs is divided by the mean IP during taking food into the mouth, was attained. The VC of PT, that the SD of PTs is divided by the mean PT. The IP during taking food into the mouth represents the pressure caused by the lips upon coming into contact with the spoon when food is taken into the oral cavity, the PT represents the time of the lip contacting with the spoon. The VC of IP and PT during taking food into the mouth represent the dexterity of lip closing function during taking food into the mouth: lower values indicate higher dexterity.

Statistical analysis

Statistical analyses were performed using IBM SPSS ver. 25 for Windows. Each DSs or typically developing children were divided into five age groups, and data were compared between the DS and typically developing children of the same age groups. The Pearson's X^2 test for independence was applied for the analyses of SDs of body height and weight, gross motor function, cognitive development and molar occlusion. The unpaired t-test was used for IP during taking food into the mouth, PT, VC of IP or VC of PT during taking food into the mouth.

For the DS group, the Pearson's X^2 test for independence was utilized for the analyses of age-related changes in the incidence of incomplete lip closure or tongue protrusion. Age-related changes in IP during taking food into the mouth, PT, VC of IP or VC of PT during taking food into the mouth in the both DS and typically developing children were analyzed using the Bonferroni test (Armstrong, 2014).

ETHICAL CONSIDERATIONS

The children included in this study and their parents received explanations of the content of the study both verbally and in writing, and provided consent prior to study initiation. This study was conducted upon obtaining approval from the ethics committee at the Nippon Dental University School of Life Dentistry (Approval No. NDU-T2015-30). The study (IRB-FY2017053) was approved by the Human Subjects Research Committee at Nippon Dental University.

CONFLICT OF INTEREST

The authors have stated explicitly that there are no conflicts of interest in connection with this article. This study was supported in part by a Research Grant (16K11870) from of the Ministry of Education, Culture, Sports, Science and Technology, Japan.

RESULTS

Comparison between DS and typically developing children

Children with DS had significantly lower body heights than typically developing children (*P* = .01, =.004, < .001, = .006 and < .001, for 6 to 11 month-old, 1, 2, 3 and 4 year-old, respectively). Children with DS had significantly lower weights than typically developing children before 2 year old (*P* = .043, =.001, = .008, = .103 and = .054, for 6 to 11 month-old, 1, 2, 3 and 4 year-old, respectively). Children with DS showed developmental delay in gross motor function as compared to typically developing children (*P* = .113, < .001, = .003 and= .167, for 6 to 11 month old , 1, 2 and 3 year-old, respectively), also developmental delay in cognitive function as compared to typically developing children (*P* < .001, for 1 to 4 year-old, respectively). Moreover, a degree of deciduous molar occlusal support similar to that of typically developing children at the 3 year old or older was observed (*P* = .025, = .014, for 1 and 2 year-old, respectively) (Table 1).

Table 1-1 Comparison of characteristics between DS and typically developing children

Group	Children with DS (n = 99)					
Age	**6 to 11M**	**1Y**	**2Y**	**3Y**	**4Y**	**Total**
Number of children (%)	**9 (100)**	**26 (100)**	**25 (100)**	**19 (100)**	**20 (100)**	**99 (100)**
Sex						
Male	6 (66.7)	12 (46.2)	13 (52)	11 (57.9)	10 (50)	52 (52.5)
Female	3 (33.3)	14 (53.8)	12 (48)	8 (42.1)	10 (50)	47 (47.5)
Height (SD)						
−2 SD ≥	4 (44.4)	17 (65.4)	14 (56)	7 (36.8)	12 (60)	54 (54.5)
−1 SD ≥ and > −2SD	3 (33.3)	5 (19.2)	6 (24)	8 (42.1)	7 (35)	29 (29.3)
+1 SD > and > −1 SD	2 (22.2)	3 (11.5)	5 (20)	4 (21.1)	1 (5)	15 (15.2)
+2 SD > and ≥ +1 SD	0 (0)	0 (0)	0 (0)	0 (0)	0 (0)	0 (0)
≥ +2 SD	0 (0)	1 (3.8)	0 (0)	0 (0)	0 (0)	1 (1)
Weight (SD)						
−2 SD ≥	4 (44.4)	13 (50)	4 (16)	5 (26.3)	2 (10)	28 (28.3)
−1 SD ≥ and > −2SD	3 (33.3)	9 (34.6)	11 (44)	4 (21.1)	9 (45)	36 (36.4)
+1 SD > and > −1 SD	2 (22.2)	4 (15.4)	10 (40)	9 (47.4)	9 (45)	34 (34.3)
+2 SD > and ≥ +1 SD	0 (0)	0 (0)	0 (0)	1 (5.3)	0 (0)	1 (1)
≥ +2 SD	0 (0)	0 (0)	0 (0)	0 (0)	0 (0)	0 (0)
Gross motor function						
Lying & rolling over	7 (77.8)	3 (11.5)	1 (4)	0 (0)	0 (0)	11 (11.1)
Sitting	1 (11.1)	6 (23.1)	0 (0)	1 (5.3)	0 (0)	8 (8.1)
Crawling & kneeling	0 (0)	2 (7.7)	0 (0)	0 (0)	0 (0)	2 (2)
Standing	1 (11.1)	12 (46.2)	8 (32)	2 (10.5)	0 (0)	23 (23.2)
Walking	0 (0)	3 (11.5)	16 (64)	16 (84.2)	20 (100)	55 (55.6)
Cognitive development						
≤ I-2	9 (100)	15 (57.7)	7 (28)	1 (5.3)	0 (0)	32 (32.3)
I-3	0 (0)	11 (42.3)	18 (72)	12 (63.2)	13 (65)	54 (54.5)
II	0 (0)	0 (0)	0 (0)	4 (21.1)	5 (25)	9 (9.1)
III-1	0 (0)	0 (0)	0 (0)	1 (5.3)	0 (0)	1 (1)
≥ III-2	0 (0)	0 (0)	0 (0)	1 (5.3)	2 (10)	3 (3)
Presence/absence of deciduous molar occlusal support						
No deciduous molar occlusal support	9 (100)	20 (76.9)	0 (0)	0 (0)	0 (0)	29 (29.3)
F occlusal support, present	0 (0)	6 (23.1)	16 (64)	0 (0)	0 (0)	22 (22.2)
S occlusal support, present	0 (0)	0 (0)	9 (36)	19 (100)	20 (100)	48 (48.5)

Children with typically developing (n = 112)

Table 1-2 Differences between children with DS and typically developing children

Age	6 to 11M	1Y	2Y	3Y	4Y	Total
Number of children (%)	**20 (100)**	**22 (100)**	**26 (100)**	**21 (100)**	**23 (100)**	**112 (100)**
Sex						
Male	14 (70)	15 (68.2)	16 (61.5)	11 (52.4)	8 (34.8)	64 (57.1)
Female	6 (30)	7 (31.8)	10 (38.5)	10 (47.6)	15 (65.2)	48 (42.9)
Height (SD)						
−2 SD ≥	1 (5)	4 (18.2)	1 (3.8)	2 (9.5)	2 (8.7)	10 (8.9)
−1 SD ≥ and > −2SD	2 (10)	8 (36.4)	7 (26.9)	3 (14.3)	6 (26.1)	26 (23.2)
+1 SD > and > −1 SD	16 (80)	10 (45.5)	18 (69.2)	14 (66.7)	14 (60.9)	72 (64.3)
+2 SD > and ≥ +1 SD	0 (0)	0 (0)	0 (0)	2 (9.5)	1 (4.3)	3 (2.7)
≥ +2 SD	1 (5)	0 (0)	0 (0)	0 (0)	0 (0)	1 (0.9)
Weight (SD)						
−2 SD ≥	1 (5)	1 (4.5)	2 (7.7)	1 (4.8)	1 (4.3)	6 (5.4)
−1 SD ≥ and > −2SD	6 (30)	7 (31.8)	2 (7.7)	2 (9.5)	3 (13)	20 (17.9)
+1 SD > and > −1 SD	10 (50)	11 (50)	21 (80.8)	17 (81)	17 (73.9)	76 (67.9)
+2 SD > and ≥ +1 SD	3 (15)	3 (13.6)	1 (3.8)	0 (0)	2 (8.7)	9 (8)
≥ +2 SD	0 (0)	0 (0)	0 (0)	1 (4.8)	0 (0)	1 (0.9)
Gross motor function						
Lying & rolling over	6 (30)	0 (0)	0 (0)	0 (0)	0 (0)	6 (5.4)
Sitting	9 (45)	0 (0)	0 (0)	0 (0)	0 (0)	9 (8)
Crawling & kneeling	1 (5)	0 (0)	0 (0)	0 (0)	0 (0)	1 (0.9)
Standing	4 (20)	2 (9.1)	0 (0)	0 (0)	0 (0)	6 (5.4)
Walking	0 (0)	20 (90.9)	26 (100)	21 (100)	23 (100)	90 (80.4)
Cognitive development						
≤ I-2	20 (100)	0 (0)	0 (0)	0 (0)	0 (0)	20 (17.9)
I-3	0 (0)	12 (54.5)	0 (0)	0 (0)	0 (0)	12 (10.7)
II	0 (0)	10 (45.5)	16 (61.5)	0 (0)	0 (0)	26 (23.2)
III-1	0 (0)	0 (0)	10 (38.5)	1 (4.8)	0 (0)	11 (9.8)
≥ III-2	0 (0)	0 (0)	0 (0)	20 (95.2)	23 (100)	43 (38.4)
Presence/absence of deciduous molar occlusal support						
No deciduous molar occlusal support	20 (100)	10 (45.5)	0 (0)	0 (0)	0 (0)	30 (26.8)
F occlusal support, present	0 (0)	12 (54.5)	24 (92.3)	0 (0)	0 (0)	36 (32.1)
S occlusal support, present	0 (0)	0 (0)	2 (7.7)	21 (100)	23 (100)	46 (41.1)

Age	*P* value*				
	6 to 11M	1Y	2Y	3Y	4Y
Hight, SD	.01	.004	<.001	.006	<.001
Weight, SD	.043	.001	.008	.103	.054
Gross motor function	.113	<.001	.003	.167	—
Cognitive development	—	<.001	<.001	<.001	<.001
Presence/absence of deciduous molar occlusal support	—	.025	.014	—	—

Differences between DS and typically developing children (by Pearson's X^2 test for independence).
—: Statistics were not calculated because the frequency of appearance is constant.
SD: standard deviation.M: month old and Y: year-old

Assessment of eating function in children with DS

Jaw movement

Diagonal rotary chewing or more complicated jaw movements were acquired by 22.2% and 76.9% of the 6 to 11 month old and 1 year old children with DS, respectively. Circular rotary chewing movement was acquired by 16% and 78.9% of the 2 and 3 year old children with DS, respectively (Tables 2).

Food texture

Solid foods (soft mechanicals and hard mechanicals) were eaten by 0%, 7.7%, 52% and 89.5% of the 6 to 11 month old, 1, 2 and 3 year old children with DS, respectively (Tables 2).

Table 2 Eating function in children with DS - Food textures and jaw movement assessment by external observation -

Group	Children with DS (n = 99)					
Age	6 to 11M	1Y	2Y	3Y	4Y	Total
Number of children (%)	9 (100)	26 (100)	25 (100)	19 (100)	20 (100)	99 (100)
Eating function						
Food texture						
Fluids and pureed foods	4 (44.4)	0 (0)	3 (12)	0 (0)	0 (0)	7 (7.1)
Mashed foods	4 (44.4)	10 (38.5)	3 (12)	2 (10.5)	0 (0)	19 (19.2)
Soft pieces	1 (11.1)	14 (53.8)	6 (24)	0 (0)	0 (0)	21 (21.2)
Soft mechanicals	0 (0)	2 (7.7)	8 (32)	5 (26.3)	5 (25)	20 (20.2)
Hard mechanicals	0 (0)	0 (0)	5 (20)	12 (63.2)	15 (75)	32 (32.3)
Jaw movement						
Minimal movement	1 (11.1)	1 (3.8)	0 (0)	0 (0)	0 (0)	2 (2)
Vertical movement	6 (66.7)	5 (19.2)	4 (16)	0 (0)	0 (0)	15 (15.2)
Diagonal rotary chew	2 (22.2)	19 (73.1)	17 (68)	4 (21.1)	2 (10)	44 (44.4)
Circular rotary chew	0 (0)	1 (3.8)	4 (16)	15 (78.9)	18 (90)	38 (38.4)

M: month old and Y: year-old

Incomplete lip closure

More than 85% of children with DS in all age groups showed incomplete lip closure at rest. During eating, 77.8% of the 6 to 11 month old children with DS showed incomplete lip closure during taking food into the mouth, as compared to only 10% of the 1 to 4 year old children with DS. Incomplete lip closure during bolus formation was observed in 100% of the 6 to 11 month old children with DS, as compared to

83.3% of the 1 to 4 year old children with DS. Incomplete lip closure during swallowing was observed in 55.6% of the 6 to 11 month old DS children, as compared to 22.2% of the 1 to 4 year old children with DS. Significant differences among age groups of children with DS in the incidence of incomplete lip closure were observed only during taking food into the mouth ($P < .001$) (Tables 3).

Tongue protrusion

Tongue protrusion was found in 77.8% of the 6 to11 month old children with DS at rest, as compared to 24.4% of the 1 to 4 year old children with DS. During taking food into the mouth, tongue protrusion was found in 66.7% of the 6-11 month old children with DS, but not in the 3 year old children with DS or older. Tongue protrusion during bolus formation was found in 77.8% of the 6 to 11 month old children with DS. The percentage decreased with age, only 15% of the 4 year old children with DS demonstrated tongue protrusion during bolus formation. Tongue protrusion during swallowing was found in 44.4% of the 6 to 11 month old children with DS, whereas only 16.7% of the 1 to 4 year old children with DS demonstrated it. Significant differences among age groups of children with DS were observed in the incidence of tongue protrusion at rest ($P = .008$), during taking food into the mouth ($P < .001$) and during bolus formation ($P < .001$) (Table 3).

Table 3 Eating function in children with DS- Assessment of lip closure and tongue protrusion by external observation-

Group		Children with DS (n = 99)					
Age		6 to 11M	1Y	2Y	3Y	4Y	P value[a]
Lip closure							
Rest	Good	0(0)	3(11.5)	3(12)	1(5.3)	3(15)	0.701
	Poor	9(100)	23(88.5)	22(88)	18(94.7)	17(85)	
Taking food into the mouth	Good	2(22.2)	22(84.6)	21(84)	19(100)	19(95)	<0.001
	Poor	7(77.8)	4(15.4)	4(16)	0(0)	1(5)	
Bolus formation	Good	0(0)	8(30.8)	3(12)	1(5.3)	3(15)	0.085
	Poor	9(100)	18(69.2)	22(88)	18(94.7)	17(85)	
Food swallowing	Good	4(44.4)	18(69.2)	19(76)	16(84.2)	17(85)	0.145
	Poor	5(55.6)	8(30.8)	6(24)	3(15.8)	3(15)	
Tongue protrusion							
Rest	Not exist	2(22.2)	18(69.2)	18(72)	17(89.5)	15(75)	0.008
	Exist	7(77.8)	8(30.8)	7(28)	2(10.5)	5(25)	
Taking food into the mouth	Not exist	3(33.3)	25(96.2)	24(96)	19(100)	20(100)	<0.001
	Exist	6(66.7)	1(3.9)	1(4)	0(0)	0(0)	
Bolus formation	Not exist	2(22.2)	16(61.5)	20(80)	18(94.7)	17(85)	<0.001
	Exist	7(77.8)	10(38.5)	5(20)	1(5.3)	3(15)	
Food swallowing	Not exist	5(55.6)	21(80.8)	19(76)	18(94.7)	17(85)	0.154
	Exist	4(44.4)	5(19.2)	6(24)	1(5.3)	3(15)	

*: Differences between age groups (by Pearson's X^2 test for independence).
M: month old and Y: year-old

Measurement results

IP, PT, VC of IP and VC of PT during taking food into the mouth were initially analyzed separately for boys and girls in the groups consisting of DS and typically developing children, but revealed no significant differences based on sex. Thus, all of the following analyses were performed on pooled data from boys and girls (Figure 3).

IP (interlabial pressure)

A significant difference in IP during taking food into the mouth was shown between 6 to 11 month old and 3 year old children in DS group (*P* < .001) and between 6 to 11 month old and 4 year old children (*P* = .001). In the typically developing children, a significant difference in IP during taking food into the mouth was found between 6 to 11 month old and 3 year old (*P* = .008), 6 to 11 month old and 4 year old (*P* = .009), 2 year old and 3 year old (*P* = .023), and 2 year old and 4 year old (*P* = .028) groups. Subsequent analysis by age group showed a significant difference in IP during taking food into the mouth between DS and typically developing children in the 2 year old (*P* = .042) and 3 year old (*P* = .049) groups (Figure 3).

PT (pressure time)

A significant difference in PT was demonstrated between 6 to 11 month old and all of the other (1 to 4 years) year old children in DS groups (*P* < .001 for all). Similarly, in the typically developing children, a significant difference was also found among the 6 to 11 month old and all of the other (1 to 4 years) age groups (*P* < .001 for all). Subsequent analysis by age group showed no significant difference in PT between DS and typically developing children in any of the age groups (Figure 3).

VC of IP (variation coefficient of interlabial pressure)

A significant difference in VC of IP was indicated between 6 to 11 month old and 2 year old (*P* = .008), 6 to 11 month old and 3 year old (*P* = .002) and 6 to 11 month old and 4 year old (*P* = .014) children in DS group. In the typically developing children, no significant difference was determined between any of the age groups. Subsequent analysis by age group showed no significant difference in VC of IP during taking food into the mouth between DS and typically developing children in any of the age groups (Figure 3).

VC of PT (variation coefficient of pressure time)

In the DS group, there were no significant differences in VC of PT among all age groups. Similarly, in the typically developing children, there were no significant differences among all age groups. Subsequent analysis by age group showed no significant difference in VC of PT between DS and typically developing children in all age groups (Figure 3).

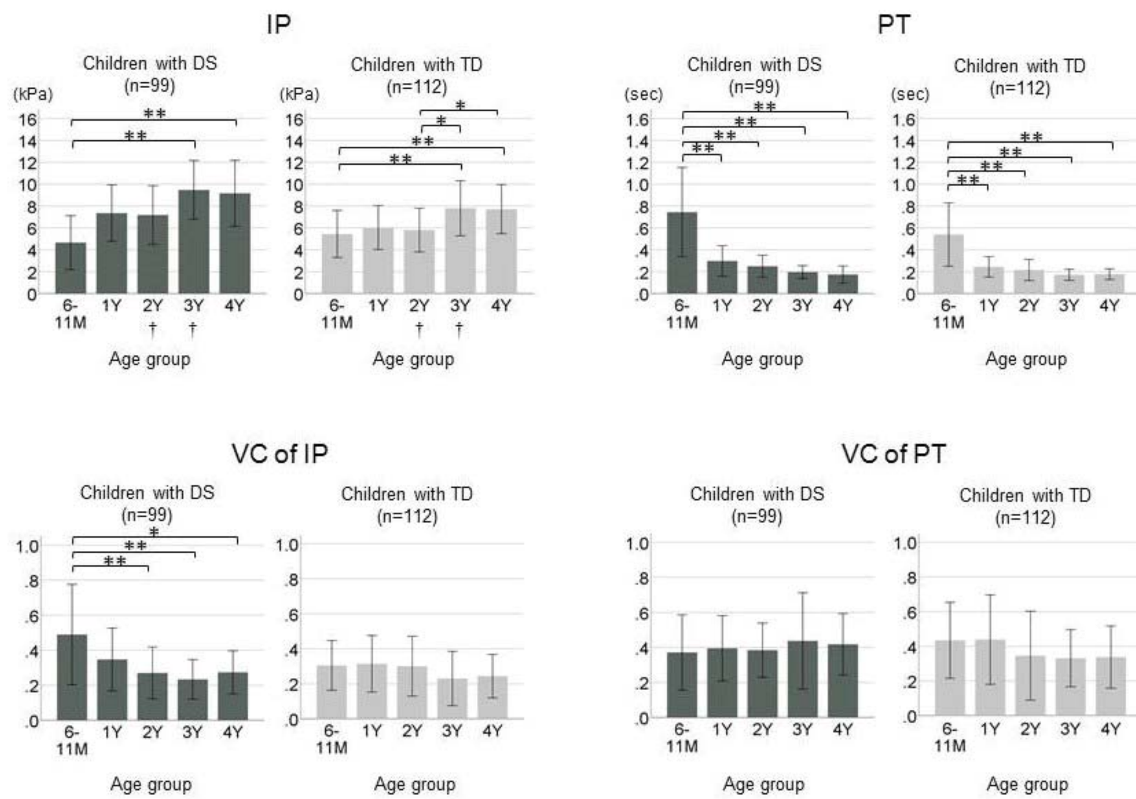

Figure 3 IP, PT, VC of IP and VC of PT during taking food into the mouth in children with DS and typically developing (TD) children

IP of DS children increased till 3 year-old, PT was decreased since 6 month-old till 1 year-old. VC of IP in children with DS decreased with ageing, because lip closing function during taking food into the mouth was developed.

Data are shown as a mean ± standard deviation.

Differences between the age of children showed *P* value by Bonferroni test ($^{*}P < .05$ $^{**}P < .01$).

Differences between children with DS and TD showed IP during taking food into the mouth in the 2 and 3 year old (†*P* = .042 and .049, respectively) groups by unpaired test.

M: month old and Y: year-old

DISCUSSION

The present study was conducted to characterize eating function of children with DS during taking food into the mouth. Most of the children with DS who were less than 1 year old were not eating solid food (soft mechanicals and hard mechanicals), while 2 year-olds or older were eating solid food. This study demonstrated that children with DS show retardation of oral or mandible functions related to eating or swallowing compared with typically developing children who acquire the functions at 9 to 11 month old after birth (Morris & Klein, 2000).

There are many reports related to incomplete lip closure (Cooper-Brown *et al.*, 2008; Faulks D, *et al.*, 2008; Price, Orvidas, Weaver, Farmer, 2004). Specifically, incomplete lip closure was found during resting time of the mouth for DS patients (Oliveira, Paiva, Campos, Czeresnia, 2008). The present study assessed lip closing function not only for resting time, but for taking during food into the mouth, bolus formation and swallowing of food for children with DS. The present study found that lip closure during taking food into the mouth and swallowing tended to be more frequent. In addition, it clearly demonstrated that lip closing function during food taking into the mouth was acquired at earlier stages. However, the function, that is required during resting time and during bolus formation or swallowing, was difficult to acquire.

The incidence of tongue protrusion during bolus formation was found to be lower with ageing, although incomplete lip closure was frequently found. To close the lip during chewing, the masticatory muscles, such as the masseter muscle, must be deactivated whereas the lower lip also must be activated (Hanawa, Tsuboi, Watanabe, Sasaki, 2008; Takada, Miyawaki, Tatsuta, 1994; Tomiyama, Ichida, Yamaguchi, 2004). The space for the tongue in DS children is too small to accommodate the tongue in the oral cavity proper because of morphological characteristics of DS, *i.e.* maxillary hypoplasia, high palate and narrow palate (Alió, Lorenzo, Iglesias, Manso, Ramírez, 2011; Fink, Madaus, Walker, 1975; Klingel, Hohoff, Kwiecien, Wiechmann, Stamm, 2017; Skrinjarić, Glavina, Jukić, 2004). That is, for producing enough room to move the tongue, the jaw may be required to open farther, therefore closing lips would be difficult.

IP during taking food into the mouth in children with DS increased with age until three years of age, and the PT was also rapidly shortened since the age group of 6 to 11 months until 1 year, as well as in typically developing children. These results support the results reported by Chigira *et al*. (1994). Moreover, an age-related change in the VC of IP during taking food into the mouth was found to decrease with ageing in children with DS. In contrast, no such age-related change in the VC of IP during taking food into the mouth was found in typically developing children, suggesting that these children become able to dexterously close the lips soon after weaning diets. IP during taking food into the mouth was showed higher in children with DS than in typically developing children in the 2 year-old and 3 year-old groups. The result suggests that children with DS might have poor ability of adjust force although they have enough force to close the lips.

In the typical developmental process, gross motor functions, including walking, advance between birth to 1 year old (Morris & Klein, 2000). In children with DS included in the present research, gross motor function including walking and oral functions for chewing developed between birth to 3 years-old. Despite children with DS having smaller physical constitution and retardation of cognitive function compared to typically developing children, children with DS showed similar development to that of typically developing children in lip closing function during taking food into the mouth. Although children with DS demonstrate lower tonicity of the muscles (Eid *et al.*, 2017; Palisano *et al.*, 2001), children with DS in this study demonstrated adequate ability of taking food in to the mouth. This may partially be explained by the habitual eating behavior: the time of introduction of weaning diets in children with DS was considered to be almost the same as that for typically developing children. Lip closing function during taking food into the mouth was developed through repeated food eating after weaning diets. Accordingly, a way to assist eating for children with DS soon after weaning diets may affect their development of the skill of lip closing function during taking food into the mouth.

In this study, we found some limitations of the research design: IP was measured by a pressure sensor attached to a spoon. Because of these conditions, some children with difficulties in eating might have affected the measurement. Moreover some children refused to eat the test food. Psychological effects of the presence of the examiner and the use of a pressure sensor, which may have had an effect on the lip function could not be completely ruled out.

CONCLUSION

The present study revealed that children with DS show a process of acquisition of lip closing function during taking food into the mouth similar to that in typically developing children, even though they experience developmental retardation of gross motor and cognitive functions. Not training lip force but rather training lip dexterity may improve incomplete lip closure for children with DS.

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