

Brief Report**Two mastication tests used in children with down syndrome: A feasibility study****W. J. A. in't Veld,¹ I. de Pijper,² M. van Gerven³ & L. van den Engel-Hoek³**¹ Prinsentichting, Purmerend, The Netherlands² Cordaan, Amsterdam, The Netherlands³ Donders Centre for Neuroscience, Department of Rehabilitation, Radboud University Medical Center, Nijmegen, The Netherlands**Abstract**

Background Children with Down syndrome can have problems with chewing and swallowing. However, no objective tests are available that give information about the efficiency and endurance of mastication. In this study, the feasibility of two mastication tests is studied by answering two research questions: are the Test of Mastication and Swallowing of Solids and the 6-min mastication test feasible to use in children with Down syndrome? Do these tests give information about the mastication efficiency and endurance in these children?

Method A total of 24 (4–18 years) children with Down syndrome participated. The collected variables in the Test of Mastication and Swallowing of Solids were masticatory time, discrete bites, masticatory cycles and number of swallows per cracker. The inter-rater reliability of these variables was tested. Collected variables in the 6-min mastication test were total masticatory cycles and percentage difference between minutes 1 and 6. Qualitative ratings of masticatory movements were made. In both tests, a comparison with a typically developing group was made.

Results In both tests, a total of 87.5% of the children completed the test. Both tests are feasible for children with Down syndrome with language comprehension skills of 3 years and over.

Conclusions The tests give information about the function and endurance during mastication in children with Down syndrome. The results showed that children with Down syndrome do not have endurance problems during mastication, but there is a lack of efficiency in mastication.

Keywords 6-min mastication test, Children, Down syndrome, Mastication, Test of mastication and swallowing of solids

Introduction

Children with Down syndrome often present chewing, drinking and swallowing problems because of impaired oral-motor function (O'Neill *et al.* 2005). The tongue and lips are hypotonic with the tongue having inefficient lingual lateralisation and a low and anterior resting position in the mouth (Hennequin *et al.* 1999). Mastication consists of a lateral movement of the tongue and an up-and-down movement of the mandible: the masticatory cycle (Le Reverend *et al.* 2014). Because of

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impaired oral-motor function, it may be difficult for children with Down syndrome to achieve a mature mastication efficiency (Frazier and Friedman 1996; Spender *et al.* 1996). In several studies in adults with Down syndrome, the masticatory time was found to be increased (Frazier and Friedman 1996; Spender *et al.* 1996; Allison *et al.* 2004; Faulks *et al.* 2008). Young adults with Down syndrome have a significantly lower mean chewing frequency, a significantly higher number of masticatory cycles with open mouth compared with controls (Allison *et al.* 2004; Hennequin *et al.* 2005a; Hennequin *et al.* 2005b). In summary, the masticatory efficiency of people with Down syndrome seems limited.

Recently, two tests were developed for the quantitative assessment of efficiency and endurance of mastication: (1) the Test of Mastication and Swallowing of Solids (TOMASS) (Huckabee *et al.* 2018) and (2) 6-min mastication test (6MMT) (van den Engel-Hoek *et al.* 2017). The TOMASS gives information about the masticatory efficiency. Total time needed for eating the whole cracker in seconds, discrete bites, number of masticatory cycles and swallows per cracker are all components of the masticatory efficiency and scored during this test. In two international studies, normative data for adults and children were collected (Frank *et al.* 2018; Huckabee *et al.* 2018).

The 6MMT gives information about the endurance of mastication, in terms of total number of masticatory cycles and the percentage difference between minute 1 (M_1) and minute 6 (M_6).

Because mastication problems are known in children with Down syndrome, it would be of interest to investigate if these two easy-to-perform tests are feasible in this group. Therefore, the aims of this study were to explore whether these tests are feasible for children with Down syndrome and to investigate what the nature is of the masticatory problems of this group.

Methods

Participants

Children with Down syndrome were approached by their own speech language therapist (SLT) to participate in the study. The participants were all aged between 4 and 18 years ($N = 24$) and had a

minimum language comprehension of 3.0 years of age. They were receiving speech and language therapy for problems in oral-motor functioning. Children with a feeding tube or gluten allergy, at risk of choking and/or eating only thick liquid pureed food were excluded. In both tests, the participants were compared with a typically developing Dutch group (van den Engel-Hoek *et al.* 2017, Frank *et al.* 2018).

Procedures

A short structured questionnaire for parents was used to collect data about gender, age, health and oral motor behaviour (see Appendix A). In both tests, the own SLT gave instructions to the participant following a standardised protocol, as described in the papers of Frank *et al.* (2018) and van den Engel-Hoek *et al.* (2017). The participant was asked to eat the cracker as quickly and comfortably as possible and to say his or her own name when he had finished. In the 6MMT, the participant was asked to chew for 6 min on a chewing tube, on one side of the mouth. If necessary, the SLT sat behind the child and was holding the chewing tube during 6 min. If needed, a time timer (countdown timer with visual support) was used. After the 6MMT, the participants from group 6MMT-2 filled in a face scale (Fig. 1) for pain and fatigue [converted to a visual analogue scale (VAS) score].

Materials

For the TOMASS, a 5 × 5-cm cracker of the brand Albert Heijn (as described in TOMASS study) was used. The chewing tube, used in the 6MMT, was 12–15 cm long and 1 cm thick and had a resistance of level 3 (Theratube[®]).

Data collection

Data collected from the TOMASS included the total time needed to eat a cracker, discrete bites, number of masticatory cycles and the number of swallows per cracker.

The variables collected from the 6MMT included the amount of masticatory cycles per minute, the total amount in 6 min and the percentage difference between M_1 and M_6 . In addition, the qualitative

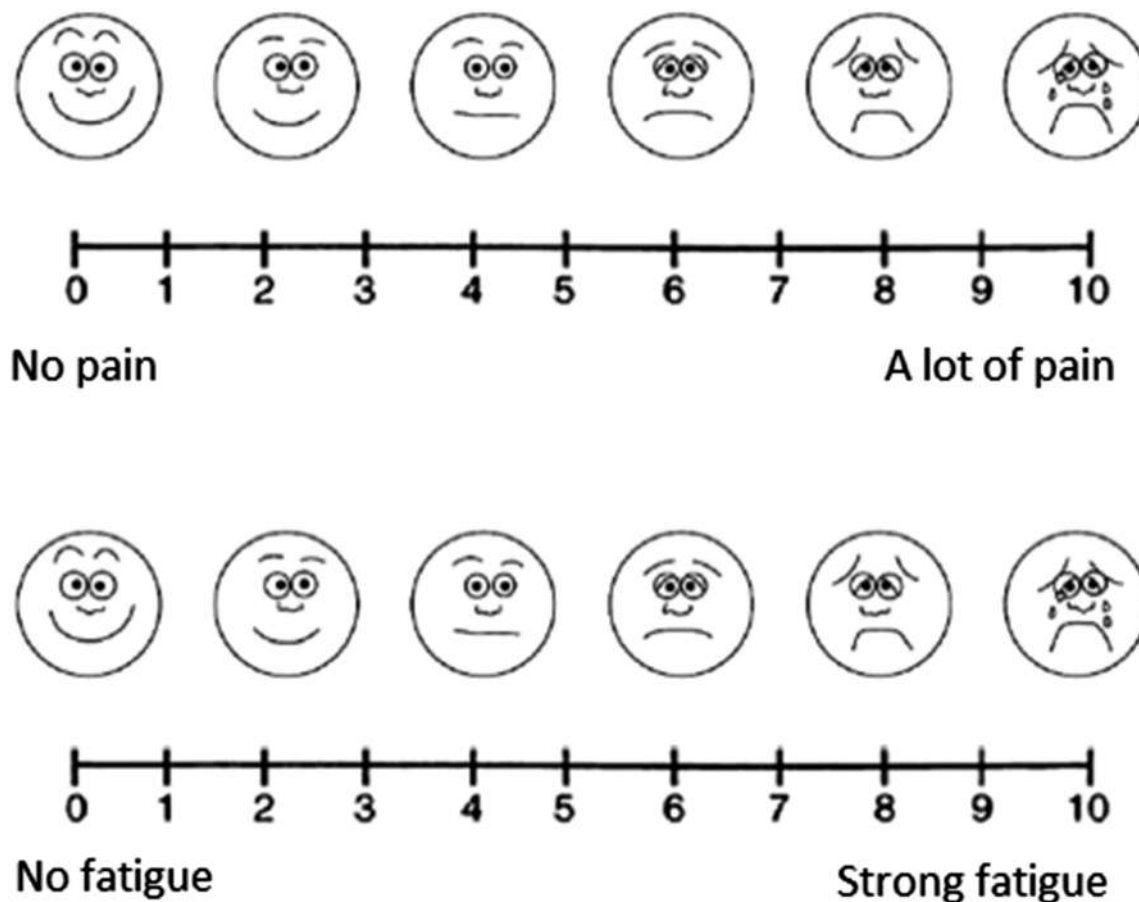


Figure 1. Face scale converted into a visual analogue scale.

rating of the masticatory movements in rhythm and size was made. Moreover, VAS scores for pain and fatigue were collected in the 6MMT-2 group. Both tests were video recorded in high definition by a Canon Eos 1100D digital SLR camera by the first author.

Reliability

Reliability for scoring the TOMASS and the 6MMT was reported in the papers of Frank *et al.* (2018) and van den Engel-Hoek *et al.* (2017). For the current study, the first and last authors scored the first video to obtain agreement on the scores. The first author scored the other 23 videotapes. In the TOMASS, the second author scored four cases (16.7%) to test the inter-rater reliability in this study.

Statistical analysis

The intraclass correlation coefficient (ICC) of the scores of the first and second authors were computed. The percentage of completed tests (both the TOMASS and the 6MMT) was calculated. For the TOMASS, the participants were divided into the same age groups as the typically developing Dutch group: 4:0–5:11 years old (group TOMASS-1) and 6:0–18:0 years old (group TOMASS-2) (Gerven *et al.* 2018).

For the 6MMT, the participants were divided into the same age groups as the typically developing group: 4:0–8:11 years old (group 6MMT-1) and 9:0–17:11 years old (group 6MMT-2) (van den Engel-Hoek *et al.* 2017). For the TOMASS and the 6MMT, mean and standard deviation of the z -scores were computed.

Descriptive statistics were used to document the qualitative rating and numeric VAS scores.

Ethical issues

This study was approved by the local Committee on Research Involving Human Subjects. Written informed consent was obtained from the parents of the participants. When the child found the task too difficult or did not want to cooperate, the SLT appropriately stopped.

Results

Participants

The study group consisted of 24 children with Down syndrome, age range (years : months) 4:7–17:11 (mean age 9:7), 13 were female and 11 were male (characteristics in Table 1). All participants had language comprehension skills of 3 years and over.

Feasibility

In the TOMASS, a total of three participants (12.5%) (all in TOMASS-2) could not completely eat the cracker. Three participants did not complete the 6MMT: two in group 6MMT-1 and one in group

6MMT-2 (in total 12.5%). One of them completed the test until minute 5. Two others did not start.

Results of the test of mastication and swallowing of solids

Reliability of the scoring was good: masticatory cycles per cracker (ICC = 0.99), swallows per cracker (ICC = 0.83) and discrete bites per cracker (ICC = 0.93). Total time was scored exactly the same by the two reviewers.

Total time, masticatory cycles per cracker and swallows per cracker, and discrete bites showed all z -scores $> +2$, except for discrete bites in the TOMASS-2 group (Table 2).

Results of the 6-min mastication test

The z -score of the total amount of masticatory cycles was within normal range (mean z -score group 6MMT-1 = -1.69 ; group 6MMT-2 = -1.32) (Table 3). The mean percentage difference between M_1 and M_6 showed also data within normal ranges. Results of the qualitative ratings are depicted in Table 4 and of the VAS scores in Table 5.

Table 1 Patient characteristics

Participants Male, $N = 11$ Female, $N = 13$ Mean age (years : months) 9:07 (range 4:07–17:11)		No N (%)	Yes N (%)	N total
1.	Respiratory infections or pneumonia	13 (56.5%)	10 (43.5%)	23
2.	Swallowing problems in the past	10 (41.7%)	14 (58.3%)	24
3.	Swallowing problems in the present	17 (70.8%)	7 (29.2%)	24
4.	Showing aversion for mealtimes	21 (87.5%)	3 (12.5%)	24
5.	Difficulties with mastication	17 (70.8%)	7 (29.2%)	24
6.	Many choking (more than once a day)	20 (83.3%)	4 (16.7%)	24
7.	Coughing during a meal	22 (95.7%)	1 (4.3%)	23
8.	Saliva loss or food loss during a meal	17 (70.8%)	7 (29.2%)	24
9.	Cramming	11 (45.8%)	13 (54.2%)	24
10.	Eating enough	2 (8.3%)	22 (91.7%)	24
11.	Open mouth behavior	10 (41.7%)	14 (58.3%)	24
12.	Drooling	17 (73.9%)	6 (26.1%)	23
13.	Speech and language therapy for oral motor skills in past	0 (0%)	24 (100%)	24
14.	Speech and language therapy for oral motor skills in present	4 (16.7%)	20 (83.3%)	24
15.	Problems during tooth brushing	18 (81.8%)	4 (18.2%)	22

Table 2 Mean and SD of the *z*-scores per age group per parameter for the TOMASS

Parameter	Group TOMASS-1 4:0–5:11 years (N = 5)		Group TOMASS-2 6:0–17:11 years (N = 16)	
	Mean <i>z</i> -score (SD)		Mean <i>z</i> -score (SD)	
Total time	4.53	(2.06)	5.55	(4.87)
Discrete bites	2.53	(1.99)	1.39	(2.22)
Masticatory cycles per cracker	4.73	(2.82)	4.73	(6.6)
Swallows per cracker	11.30	(5.03)	8.40	(3.48)

z-scores (for the TOMASS and 6-min mastication test) are calculated using the equation: $\frac{\text{measured value} - \text{normal value}}{\text{standard deviation of normal value}}$
SD, standard deviation; TOMASS, Test of Mastication and Swallowing of Solids.

Table 3 Mean and SD of the *z*-scores per age group for total amount of masticatory cycles and the percentage difference between minute 1 (*M*₁) and minute 6 (*M*₆) in the 6MMT

Parameter	Group 6MMT-1 4:0–8:11 years, N = 8		Group 6MMT-2 9:0–17:11 years, N = 13	
	Mean <i>z</i> -score (SD)		Mean <i>z</i> -score (SD)	
Total masticatory cycles	−1.69 (0.94)		−1.32 (1.89)	
Percentage difference between minute 1 and minute 6	1.53 (5.34)		0.78 (3.22)	

6MMT, 6-min mastication test; SD, standard deviation.

Table 4 Qualitative ratings of the masticatory movements on the 6MMT

Qualitative rating	Group 6MMT-1 4:0–8:11 years N = 9		Group 6MMT-2 9:0–17:11 years N = 13	
Rhythm				
Rhythmic, N (%)	0 (0.0%)		1 (7.69%)	
Variable rhythmic, N (%)	0 (0.0%)		0 (0.0%)	
Not rhythmic, N (%)	9 (100%)		12 (92.23%)	
Movements				
Normal movements, N (%)	5 (55.56%)		4 (30.76%)	
Big movements, N (%)	0 (0.0%)		1 (7.69%)	
Small movements, N (%)	0 (0.0%)		2 (15.38%)	
Variable movements, N (%)	4 (44.44%)		6 (46.15%)	

6MMT, 6-min mastication test.

Discussion

In this study, the TOMASS and 6MMT were tested for the first time in children with Down syndrome. In both tests, 87.5% could complete the TOMASS and the 6MMT. It is concluded that both tests are feasible to use in children with Down syndrome (% complete the test >80%).

The secondary objective of this study was to determine the nature of mastication problems in children with Down syndrome in terms of efficiency and endurance. The TOMASS showed that these children need more time and have more mastication cycles per cracker and swallowing acts per cracker than typically developing children of the same age.

Table 5 Mean VAS scores for pain and fatigue per group 6MMT-1 and 6MMT-2, directly after the 6MMT

VAS scores	Group 6MMT-1 4:0–8:11 years	Group 6MMT-2 9:0–17:11 years
VAS score pain (SD; range)	N/A	N = 8: 0.3 (0.7; 0–2)
VAS score fatigue (SD; range)	N/A	N = 7: 0.9 (1.6; 0–4)

6MMT, 6-min mastication test; N/A, not applicable; SD, standard deviation; VAS, visual analogue scale.

These results are in accordance with the literature (Allison *et al.* 2004).

Results of the 6MMT suggest that the total masticatory cycles during 6 min in both age groups are within the standard range in comparison with typically developing children. The difference in percentages between M_1 and M_6 in both age groups is also within the normal range. In the 6MMT-1, the number of mastication cycles in M_1 was generally low and increasing during the test. It is the author's hypothesis that young children with Down syndrome need more time to get used to the chewing tube. The qualitative ratings of the 6MMT showed a difference in comparison with the typically developing group. In Down syndrome children, jaw control was found to be limited (Frazier and Friedman 1996). Jaw control is necessary for rhythmic and constant chewing (Le Reverend and Hartmann 2013). This could be an explanation for the differences found between the Down syndrome and typically developing group.

Self-reports of children's pain intensity must be considered with caution (Von Baeyer 2006). Tsze *et al.* (2013) found that convergent validity of the VAS for describing pain was questionable in typically developing children <7 years old (Tsze *et al.* 2013). In our study, only participants older than 9 years old completed the VAS directly after the 6MMT. Because of their developmental delay, the interpretation must be considered with care.

Although the feasibility was established in this study, some remarks have to be made. To complete both tests in children with Down syndrome, the SLT and the child must have a good rapport. During the TOMASS, we found that some children benefited from support to their motivation through suggesting they compete to eat a cracker as fast as possible, external commentary from the SLT to continue and sometimes by adding a competition element. During the 6MMT, some children needed

'inner speech': they need the SLT to articulate their thoughts to continue mastication.

Conclusion and recommendations

The TOMASS and the 6MMT are with some adjustments feasible for children with Down syndrome with 3-year-level comprehension skills and above. This study shows that these children, although known with hypotonia, do not have endurance problems during mastication, but their efficiency in mastication is reduced. Therefore, the TOMASS rather than de 6MMT seems to be meaningful for the assessment of chewing problems in children with Down syndrome and could give direction to tailor made masticatory exercises. In addition, the TOMASS could be a useful outcome measure to see if chewing exercises were effective.

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Conflict of Interest

No conflicts of interest have been declared.

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Appendix

Questionnaire about problems in mastication and swallowing

Does the child experience respiratory infections	Yes – No
Have there been swallowing problems (past)	Yes – No
Does the child experience swallowing problems (present)	Yes – No
Is the child dreading the mealtimes	Yes – No
Does the child experience mastication difficulties	Yes – No
Does the child choke? ('yes' when choking more than once a day)	Yes – No
Is there coughing during mealtimes	Yes – No
Saliva loss or food loss during a meal	Yes – No
Does cramming occur	Yes – No
Is the daily intake sufficient	Yes – No
Drooling	Yes – No
Speech and language therapy for oral motor skills in past	Yes – No
Speech and language therapy for oral motor skills in present	Yes – No
Problems during tooth brushing	Yes – No

According to Franssen and van den Engel-Hoek, 2015 (14)

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