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# Evaluation of a mechanical stretching device, the TheraBite®, in patients with restricted maximal mouth opening and neuromuscular disorders: a case series

**Bitte Ahlborg DDS[1], Birgitta Johansson Cahlin DDS[1], Åsa Mårtensson DDS[1] and Anna-Karin Kroksmark PT PhD[2]**

*[1]Mun-H-Centre, National Orofacial Resource Centre for Rare Disorders; [2]The Regional Pediatric Rehabilitation Centre, Queen Silvia Children's Hospital: Gothenburg, Sweden*

**Abstract**

**Aim:** Inability to open the mouth completely is a feature that develops in several of the neuromuscular disorders. The aim of this case series was to evaluate whether stretching with TheraBite® had any effect on maximal mouth opening in patients with Duchenne muscular dystrophy and spinal muscular atrophy.

**Methods:** Three patients with spinal muscular atrophy and six patients with Duchenne muscular dystrophy were treated during a two-year period. Maximal interincisor opening was measured to evaluate the results and the patients also kept diaries.

**Results:** The patients with spinal muscular atrophy preserved their maximal mouth opening or increased their results slightly. One patient with Duchenne muscular dystrophy increased the maximal mouth opening, while, in five patients mouth opening was slightly reduced during the treatment period.

**Conclusion:** This series suggests that stretching with the TheraBite® should be introduced the moment a progressive limitation is noticed, as it appears to be easier to preserve rather than increase mouth opening.

***Key words:*** *Maximal mouth opening, neuromuscular disorders, spinal muscular atrophy, Duchenne muscular dystrophy, TheraBite®, stretching programme*

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## Introduction

The inability to open the mouth completely is a feature that develops in several of the neuromuscular disorders, even if it is not frequently described (Botteron *et al*., 2009). Severely reduced maximal mouth opening (MMO) affects the ability to chew, swallow and maintain oral hygiene (Messina *et al*., 2008). Sufficient mouth opening is also important for dental care.

Neuromuscular disorders are a group of many different disorders caused by a disturbance in some part of the motor unit. One of the most common neuromuscular disorders is Duchenne muscular dystrophy (DMD) (Emery, 1991). The disorder leads to progressive muscle wasting with degeneration and necrosis of muscle fibres that are gradually replaced by adipose and connective tissue (Dubowitz, 1995a). Contractures are a common feature, partly caused by pathological changes in the muscle tissue but also as compensation due to muscle weakness (Eagle, 2002a). No cure is available, but, with better co-coordinated care and mechanical ventilation, life span has increased during the last decade (Eagle *et al*., 2002b). Today, many patients with DMD survive into adulthood. Several studies have shown that the oral cavity is also affected in DMD. A combination of severe muscle weakness, an enlarged tongue and the dominance of less affected perioral muscles leads to gradual development of malocclusion (Eckhardt and Harzer, 1996). In a cross-sectional study, Botteron and co-workers (Botteron *et al*., 2009) showed that patients with DMD also develop a restricted MMO compared with controls.

Spinal muscular atrophy (SMA) is another neuromuscular disorder. A degeneration of anterior horn cells in the spinal cord causes denervation of skeletal muscles, with muscle weakness of varying degrees (Dubowitz, 1995b). In the more severe cases, there may also be an involvement

of the cranial nerves, resulting in bulbar weakness (van den Engel-Hoek *et al*., 2009). SMA is clinically divided into three subgroups, based on age of onset and the maximum motor function achieved (Dubowitz, 1995b). Although the disorder is said not to be progressive, increasing weight and height may cause functional loss (Zerres *et al*., 1997). Despite the development of scoliosis and reduced pulmonary function, most of the patients with SMA have a normal life span. Contractures are sometimes a feature in SMA and develop as a consequence of inactivity or compensatory manoeuvres due to profound muscle weakness. Orofacial problems with affected growth and reduced MMO have previously been described in patients with SMA. In a study of patients with SMA, Granger and co-workers (Granger *et al*.,1999) showed that MMO was reduced to approximately 50% of reference values. They also showed that the masticatory muscles were less efficient and fatigued more easily than in the healthy control group. In a study of 409 patients with different neuromuscular disorders, a reduced MMO and difficulty eating and swallowing were reported in the patients with SMA (Willig *et al*., 1994). Messina and co-workers (Messina et al., 2008) described 122 patients with SMA, where 30% had limitations in the ability to open the mouth; these problems were increasingly more frequent with age.

The effect of treatment with the TheraBite® has previously been evaluated in patients with restricted MMO resulting from head and neck cancer (Dijkstra et al., 2004; Cohen *et al*., 2005). To our knowledge, the TheraBite® has not been evaluated in patients with neuromuscular disorders. The aim of this case series was to evaluate whether stretching with the TheraBite® had any effect on MMO in patients with DMD and SMA.

## Materials and methods

*Subjects*

Data relating to MMO from three patients with SMA (23, 27 and 62 years of age) and six patients with DMD (19-32 years of age) were evaluated retrospectively. The patients were referred by their physicians to a national orofacial centre due to an observed successive reduction in MMO.

In two patients (cases 8 and 9), MMO had been investigated previously at 14 years of age by one of the authors (BA). According to medical records, case 8 had more than 45 mm and case 9 had 53 mm mouth opening at that time. At 19 years of age, the MMO had decreased to 25 mm and 37 mm respectively and the patients were referred to the national orofacial centre due to their restricted mouth opening.

*Intervention*

The TheraBite® (Atos Medical AB, Hörby, Sweden) was used for jaw mobilisation (*Figure 1*). The device is constructed with a mandibular mouthpiece that moves downwards in an anatomically correct track when the handle is squeezed. Broad mouthpieces with foam cushions spread the forces to protect the teeth. A precision-adjustment screw enables slow opening and permits precise setting. A C-shaped 'hand aid' assists the patient or the helper to maintain constant opening during the stretching procedure.

The stretching programme was constructed to include ten consecutive repetitions daily with a short interval of rest in between – where each repetition should have a duration of 30 seconds. Helpers were instructed to squeeze the handle, when the patients were too weak to perform the task.

*Method*

Three dentists were involved in the project and were calibrated in the examination technique before the data were collected. The dentist examined and measured MMO at the national orofacial centre every third month during a two year period. The MMO is defined as the interincisal measurement of tooth 21/31 plus or minus vertical overbite (*Figure 2*). The reference value for jaw-opening capacity in children below the age of 10 years is more than 35 mm. In adolescents more than 13 years of age, the jaw-opening distance is more than 40 mm (Agerberg, 1974a; Agerberg 1974b; Garnett *et al*., 2008), and is used as a reference value for adult population as well.

Between the appointments at the national orofacial centre, the patients used diaries every day to record whether the stretching programme had been performed. The diaries were collected and registered at every visit.

## Results

Data were obtained before treatment started and at six occasions during the treatment period. The patients with SMA preserved their MMO or increased their results slightly through the treatment period, (*Table 1*, *Figure 3 A* and *B*). One patient with DMD increased the MMO, while, in five patients with DMD, MMO was slightly reduced during the treatment period.

Several of the patients found it easier to eat and to brush the teeth after stretching with the TheraBite®. No patient experienced pain and most of the patients found the treatment comfortable.

*Figure 1. TheraBite® – a jaw stretching device for adults and children*

*Figure 2. Measuring interincisal distance 21/31, 21 mm*

## Discussion

This case series showed that patients with SMA were able to preserve and even increase the MMO during the treatment period. In the patients with DMD, the MMO increased in one patient but decreased slightly in five.

DMD is a rapidly progressive disorder with a generalized deterioration in muscle strength and the development of severe contractures around the joints. The result in this case series, with preservation and only a slight reduction in MMO, indicates that the intervention with the TheraBite® may have delayed the rate of the development of the restriction.

To our knowledge, the natural course of MMO in DMD is not well investigated or described. There are, however, two cross-sectional studies of MMO in patients with DMD with different results. In a group of 16 boys between 6-20 years of age, the result showed a significant difference compared with controls (Botteron *et al*., 2009), whilst, in another study, no significant difference was found in a group of 24 young men with DMD, 17-30 years of age, compared with controls (Ueki *et al*., 2007). It might be expected that the significant difference would have been observed in the older age group, but this was not the case. Neither of these studies were, however, longitudinal and the relation between age and mouth opening has not been investigated.

In the group of patients in this case series, two young men with DMD were investigated at the age of 14 by one of the authors (BA). At this age, no limitation was recorded. In one patient (case 8), MMO decreased by more than 20 mm during the following five-year period. In the other patient (case 9), MMO had just begun to deteriorate to under a normal level when the patient was referred to the clinic. The findings of this case series and the findings from the two cross-sectional studies (Ueki *et al*., 2007; Botteron *et al*., 2009) indicate that there is a variation regarding the rate of development of restrictions of MMO and perhaps only some of boys/men with DMD will develop this restriction. A great deal is still unknown about the factors influencing the condition and at present there is not sufficient knowledge to predict who will preserve sufficient mouth opening and who will not.

The findings of this case series indicate further that it appears to be difficult to increase MMO in DMD when the limitation has already occurred. One reason for this might be the increase in fat and connective tissue replacing necrotic muscle fibres as part of the disease progression. This degenerative process, muscle weakness and disuse result in stiff, unstretchable structures around the joints. For this reason, the findings of this case series suggest that stretching with the TheraBite® should be introduced the moment a progressive limitation is noticed, in order to attempt to preserve sufficient mouth opening.

To obtain information about how often and when the stretching programme was performed, all the patients were provided with diaries. All the patients were well motivated, but during the treatment period the compliance varied, depending on health conditions, or if the patient had been busy with other priorities. Periods with excellent compliance have obviously influenced the results in a beneficial way. In one patient with SMA, MMO continuously increased during the period – in this patient, compliance had been good for most of the treatment period. The patient had been encouraged by the results that have been achieved and had also experienced well-being in connection with the stretching exercise. Every millimetre gained had been important and had improved the ability to brush the teeth, eat and speak.

Previously patients were advised to use a simple wooden device for stretching exercises. The device functioned like a clothes peg, with a rubber band providing the resistance. The device was placed between the incisors and the wooden blades are squeezed together in order to stretch the mouth opening. It was inexpensive and simple, but there was a risk that the upper incisors would be moved and proclined. In comparison, the TheraBite® has broad mouthpieces following the entire row of teeth and works by pressing the mouthpieces apart along the natural ana-

*Table 1. Diagnosis, age and maximal mouth opening when treatment started and after 21 months of treatment. Maximal mouth opening is given as a distance in millimeter plus/minus vertical overbite.*

| Patient | Diagnosis | Age at start of treatment (years) | Mouth opening at start of treatment | Mouth opening after 21 months of treatment |
|---|---|---|---|---|
| Case 1 | SMA | 62 | 21-1 mm | 27+0 mm |
| Case 2 | SMA | 27 | 16+2 mm | 18+2 mm |
| Case 3 | SMA | 23 | 18+4 mm | 20+2 mm |
| Case 4 | DMD | 34 | 14-3 mm | 11-3 mm |
| Case 5 | DMD | 32 | 20+0 mm | 19-1 mm |
| Case 6 | DMD | 24 | 20+1 mm | 22+2 mm |
| Case 7 | DMD | 25 | 17+1 mm | 15+0 mm |
| Case 8 | DMD | 19 | 23+2 mm | 21+2 mm |
| Case 9 | DMD | 19 | 35+2 mm | 31+2 mm |

SMA= spinal muscular atrophy
DMD= Duchenne muscular dystrophy

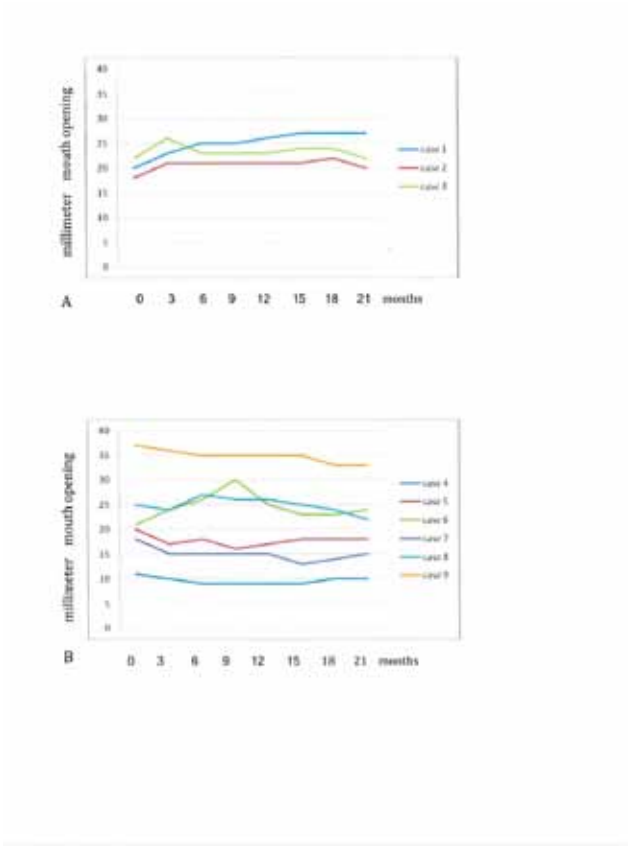


*Figure 3A and B. Maximum mouth opening distance in millimetres in three patients with SMA and in six patients with Duchenne muscular dystrophy before and during 21 months of treatment with TheraBite®*

tomical path of the jaw, thereby decreasing the risk that the teeth would procline.

Häggman-Henriksson *et al*. (2006) showed in a study that head immobilization can impair jaw function. This was confirmed by clinical observation of the patients with SMA with profound muscle weakness in neck flexors and extensors. To stabilise the head in a safe position to avoid the head dropping backwards, patients clench their teeth and retract the mandible. This habitual position of the head in combination with constant activity in the masticatory muscles may contribute to the development of restriction in MMO in this group of patients. This compensatory head posture has also been described (van den Engel-Hoek *et al.*, 2009).

## Conclusions

Stretching with the TheraBite® preserved or slightly increased MMO in some of the treated patients with SMA and DMD. As it appears to be easier to preserve than to increase MMO, the findings of the case series would suggest that stretching with the TheraBite® should be introduced the moment a progressive restriction is noticed. Life span has increased in several of these disorders and as severely restricted MMO affects health and quality of life, more attention should be paid to these symptoms and treatment. To our knowledge, this is the first study on the treatment with TheraBite® in neuromuscular disorders. Further research is, however, needed to develop an optimal method and timing of this intervention.

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***Address for correspondence:***

Anna-Karin Kroksmark
Physiotherapist, PhD
Regional Pediatric Rehabilitation Centre
Box 21062, 418 04 Göteborg
Sweden
Email: anna-karin.kroksmark@vgregion.se