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CASE REPORT

Mastication Forces and Orthodontic Treatment in Marfan Syndrome – A Case Report

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Abstract

The Marfan syndrome (MFS) based on an autosomal dominant genetic disorder. Beside to the main symptoms at the heart, the eves and the limbs the patients show a constricted maxillary bone with crowded teeth and reduced mastication forces. Main disorder in the reported 13.8-yearold patient case with MFS is the frontal and lateral open bite with crowded teeth, maxillary compression. Only three molars were in occlusion at the right side and the vertical occlusal distance increased from 3 mm on the right to 5 mm at the left side. Right and left upper first molars have been removed before treatment. Treatment was initiated with a Quad helix for expansion, followed by a fixed appliance for closing the bite and alignment of the teeth. We decided to remove the lateral left incisor in the mandible after expansion of the maxilla, thus gaining the space for all frontal teeth. Unilateral open bite was closed in the patient after 9 months of orthodontic treatment.

Second aim of the study was the ability for improvement of mastication function after closing the open bite and alignment of teeth. Bite force measurement with Bitefork System was done two times. T1 after occlusal contact of both sides and at T2 after debonding of fixed appliance nine months after T1. Bite force measurement shows normalization after interdigitation of all molars and bicuspids. Mastication exercises lead to strengthening of orofacial muscles.

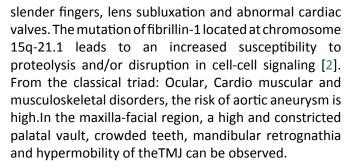
Introduction

Marfan Syndrome (MFS) is a genetic disorder of connective tissue. The inheritance is autosomal dominant with a prevalence of 1 to 10,000 up to 5,000. The age-standardized overall prevalence of MFS was 2.27 persons per 100,000 persons in South Korea [1].

The main clinical symptoms are long limbs, long



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In cases with an open bite and minimal teeth contacts from orthognathic surgery it is known that by post surgically increasing occlusal contacts, higher masticatory forces and strengthening of involved muscles were obtained. For patients with Marfan syndrome Maron [3] and Chaddha, et al. [4] recommends low intensity exercises with isometric loading of Oro-facial muscles. In orthognathic patients, Harzer [5] investigated post surgically changed fiber types in masseter muscle with increased tooth contacts from Myosin Heavy Chain (MHC) type I into type IIa. Increased content of type IIa fibers means the development of higher masticatory forces.

The aim of the case presented here are to demonstrate orthodontic treatment and improvement in muscle function in a patient with Marfan syndrome.

Diagnosis an Etiology

Clinical findings and diagnostic assessment

A 13.8-year-old patient with confirmed Marfan syndrome asked for orthodontic treatment. His main

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complaint was the open bite and crowding of teeth in the mandible. His medical history included high blood pressure treated with 47.5 mg of metoprolol daily for reduction and prevention of aortic aneurysm. Permanent mouth breathing was noted in the history.

The extraoral clinical examination revealed a concave profile and long face with higher lower part

(Figure 1). The oral examination showed an open bite of 5.0 millimeters with a slope of the maxillary occlusal plane to the left. Only four teeth were in occlusion on the right side. Maxilla and mandible were constricted with severe crowding in the frontal region. First molars at both sides in the maxilla had been extracted years before due to caries (Figure 2). The OPT showed all third molar germs and normal root lengths (Figure 2).



Figure 1: Long face syndrome of the Marfan patient, frontal (left), profile (right).

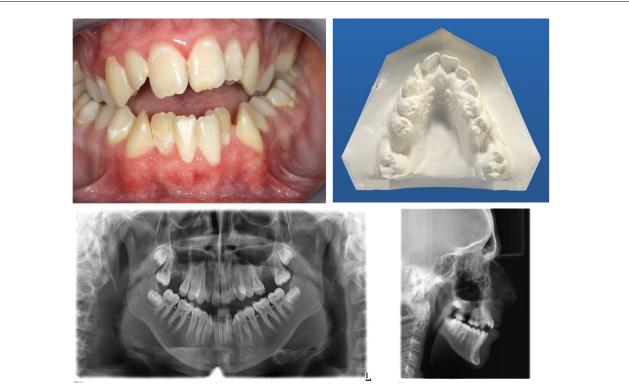


Figure 2: Open bite, three teeth in contact only (above left), narrow maxilla with missing both first molars (above right), OPT and cephalogram (below) frontal open bite of five millimeters in a Marfan patient.

The cephalogram reflected the skeletal open bite with a high ML/NL angle of 38.9 degrees. The ANB angle of 6.6 degrees signalized a Class II relationship. The proportion between the middle and the lower face underlined the dolichocephaly structure with 55.0 to 82.4 millimeters (Figure 3). Oral hygiene was poor.

Treatment objectives

The orthodontic treatment hierarchy involved the following steps:

- Palatal expansion, extraction of an incisor in the mandible
- Alignment of teeth
- Closing of open bite

- Improvement of occlusion
- Increase of bite force with a measurement tool

Treatment alternatives

The patient and his parents rejected the opportunity for rapid palatal expansion and orthognathic surgery after detailed medical information.

Treatment progress

We started with bonding and insertion of a Quad-Helix in the maxilla. Meanwhile the restorative therapy in the mandible was finished. We decided to remove the lateral left incisor in the mandible after expansion of the maxilla, thus gaining the space for all frontal teeth. For this we used a fixed appliance. The left part



Figure 3: Treatment result after closing the open bite with fixed appliance *in situ*, 18 months after start of treatment and occlusal contacts on both sides (T1 of bite force measurement). The second left incisor has been removed because of crowding in the mandible.

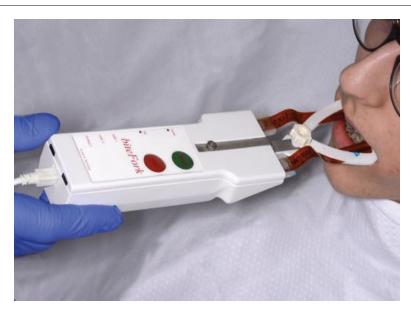


Figure 4: Bitefork System for bite force measurement. Maximum bilateral bite force at T1 ranged between 93.6N, left, and 104.8N right, in total, 198.4N.

of the arch wire in the maxilla was activated through a pronouncedcurve of Spee. Loop contraction TMA wire 0.016×0.022 provided orthodontic space closure between 17-15 and 27-25. For alignment and vertical movement up and down elastics were used.

Bite force measurement with the Bitefork System [6] was done twice, at T1 after occlusal contact on both sides and at nine months after debonding of the fixed appliance (T2) (Figure 4). The measurement was based on calibrated strain gauges. The position of the pressure sensors was fixed with a silicone impression material between the upper bicuspids on both sides (Figure 4). All measurements were repeated for 10 seconds with maximum bilateral and unilateral masticatory force.

Follow up and treatment outcomes

1.5 years after start of treatment all remaining 25 teeth were in occlusion (Figure 3). The appliance was removed six months later. Because of asymmetric extraction there was no alignment of the mid lines and no full interdigitation of all teeth. The patient confirmed improvement of masticatory function, mouth closure and permanent nose breathing. We recommended hard food and long-lasting mastication for strengthening of masticatory muscles (Figure 5).

The Cephalogram at the age of 16.5 years showed a slight reduction in the ML/NL angle of 3.3 degrees and the ANB angle was reduced from 6.6 degrees to 4.5 degrees (Table 1 and Figure 6).

At T1 the maximum bilateral bite force ranged between 93.6 N left and 104.8 N right, a total of 198.4 N (Figure 7). The maximum unilateral biteforce on the right, i.e. the contact side at T1 was 171.78N and on the left side 68.65N only. There was no tooth contact in this part at the start of treatment on this side. At T2, nine months after debonding, the maximum mastication force increased to 252.51 N on the right side and 157.53 N on the left side, a total of 410.04 N. The unilateral force on the right side was 180.54 N and on the left side 157.29 N (Figure 7 and Table 2). The occlusion after debonding and 12 months of retention with acryl splints remains generally stable.

Discussion

The patient shows all symptoms of Marfan

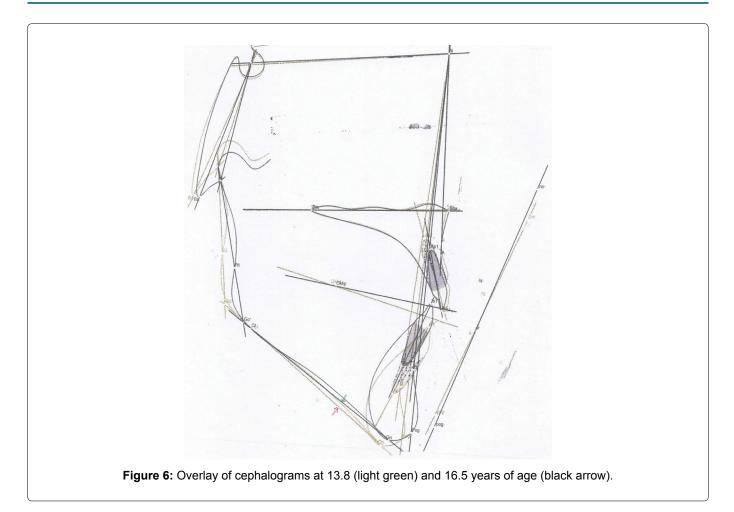
Table 1: Cephalometric	analysis at the	e start of treatment and	
after.			

	25/03/2019	14/12/2021	
SNA (°)	84.3	85.0	
SNB (°)	77.7	80.5	
ANB (°)	6.6	4.5	
NL-NSL (°)	2.9	2.7	
ML-NSL (°)	41.3	38.4	
ML-NL (°)	38.9	35.7	
U1/NA (°)	19.8	13.8	
L1/NB (°)	21.4	19.5	
L1/ML (°)	98.0	99.4	
U1/L1 (°)	132.1	142.1	
U1/NA (mm)	7.3	4.0	
Pg-NB (mm)	3.4	1.9	
NSBa (°)	119.2	118.5	
Wits Appr. (mm)	1.1	2.0	

SNA-Sella-Nasion-A-point angle, Sella-nasion-B-point angle, A-point-nasion-B-point angle, nasal-line to nasion-sella-line, mandibular-line to nasion-sella-line angle, mandibular-nasalline angle, upper central incisor-line to nasal line angle, lower central incisor line to mandibular line angle, upper central incisor to lower central incisor line angle, upper central incisor to lower central incisor line angle, upper central incisor to nasal A-point line, pogonion to nasal-B-point line, nasion-sella line to sella-basion angle, Wits appraisal.



Figure 5: Frontal view after debonding (above left), maxilla expansion and space closing after loss first molars (above right), right and left occlusion are not ideal because of extraction of one lower incisor (below).



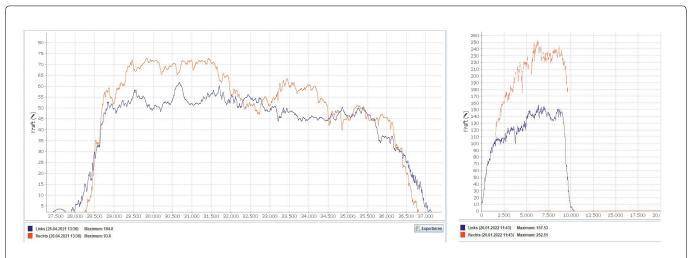


Figure 7: Maximum bilateral bite force at T1 ranged between 93.6N, left, and 104.8N right, in total, 198.4N. At T2, nine months after debonding, the maximum mastication force increased to 252.51 N on the right side and 157.53 N left side in total, 410.04 N.

T1 Measurement	Left maxim.	Force left	Right maxim.	Force right	Duration
1	24/25	93.6	14/15	104.8	9 sec.
2	24/25	68.8		103.6	10 sec.
3			14/15	171.76	10 sec.
4	24/25	89.31			10 sec.
T2 Measurement	Left maxim.	Force left	Right maxim.	Force right	Duration
unilateral 1	24/25	99.14	14/15	147.97	12 sec
bilateral 2	24/25	157.53	14/15	252.51	14 sec

Table 2: Repeated bite force measurements at T1 and T2.

syndrome. Body height increased to 2.04 meters and all cephalometric values changed minimally. Bilodeau [7] described retreatment in a patient after surgery and orthodontic intervention which were unsuccessful. The patient exhibited an open bite, crowding, severe root resorption and TMJ disorders. The second treatment included surgery and extraction and resulted in a balanced and harmoniousfacial relationship, as we achieved without surgery. We did not see root resorptionin our patient or periodontal disease as Utreja and Evans [8] observed in two patients. Lida, et al. [9], in a systematic review, found a wide variety of research in children and adults with Marfan syndrome.

The increase in masticatory force between T1 and T2 is probably a result of a higher number of teeth in occlusion. Ingervall [10] determined that bite force was correlated with the number of occlusal contacts. Masticatory force from both sides of 410.04 N corresponds with 367.46 N in healthy probands [6]. However, a difference remains between right and left sides. The reason for that could be the long absence of occlusal contacts connected with unilateral muscle atrophy on the left side. Mastication of food is linked to the right occlusion side. Kiliaridis [11] showed that in growing patients with unilateral crossbite the thickness of the masseter muscle is different. Percheron, et al. [12] investigated the muscle strength and body composition in women and found a decreasein lean leg mass and a reduction in strength.

Dean [13] emphasized the effectiveness and limitation of sport activities in Marfan patients. Peripheral blood vessel resistance and diastolic blood pressure increases during static exercises and decreases with dynamic exercises. He concluded that Marfan patients should avoid exercises with high intensity but should participate in moderate dynamic exercises.

Patient Perspective

- Unilateral open bite was closed in a Marfan patient after 9 months of orthodontic treatment.
- Bite force measurement shows normalization after interdigitation of all molars and bicuspids.
- Mastication exercises led to stability, balance and strengthening of orofacial muscles.

Conflicts of Interest

There are no conflicts of interest.

Authors Contributions

Dr. Harzer designed and wrote the manuscript. He measured all mastication force values with exception of the direction of the handling of the tool; Dr. Weisskircher developed the measurement tool and the main author rented the tool from him. He explained the measurement procedure and helped for the interpretation of the measurement results.

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