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Masticatory muscle tendon-aponeurosis hyperplasia diagnosed as temporomandibular joint disorder: A case report and review of literature

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ABSTRACT

INTRODUCTION: Masticatory muscle tendon-aponeurosis hyperplasia (MMTAH) is a new clinical entity that presents mainly with trismus due to hyperplasia of the masseter aponeurosis and temporalis muscle tendon. However, the etiological factors of this disease are unknown; it is often mistreated as temporomandibular joint disorder (TMD).

PRESENTATION OF CASE: We report a 32-year-old female patient complaining of bilateral pain in her jaw and difficulty opening her mouth. She was first diagnosed as TMD and treated with a splint; however, her symptoms did not improve. Clinical examination revealed a square mandible, tenderness in the left and right temporalis muscles and masseter muscles, and tenderness along the anterior border of the masseter muscle. Her maximum mouth-opening was 30 mm. Short TI inversion recovery magnetic resonance imaging showed areas of low intensity at the anterior border of the masseter muscle and around the coronoid process where the temporalis muscle tendon attaches. Consequently, the diagnosis made based on the clinical and radiographic findings was MMTAH. Bilateral coronoidectomy was performed, followed by a rehabilitation program for six months. The maximum opening was maintained at 48 mm two years after the operation.

DISCUSSION: MMTAH was treated as type 1 TMD until it was recognized as a new disease at the conference for the Japanese Society for Oral and Maxillofacial Surgeons. Since then, many clinicians have become aware of this particular condition, and different treatment modalities have been proposed.

CONCLUSION: Clinicians should consider MMTAH as a differential diagnosis when the patient's chief complaint is gradually decreasing mouth-opening.

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1. Introduction

Temporomandibular joint (TMJ) hypomobility can occur for many reasons, including ankylosis, disc derangement, systemic disease, or a tumor [1,2]. The most common presentation of joint hypomobility is trismus, and it represents a diagnostic challenge because the clinician has to rule out the other possible causes

to reach a final diagnosis. In the past two decades, a new clinical entity was added as a TMJ hypomobility disorder, namely masticatory muscle tendon-aponeurosis hyperplasia (MMTAH). MMTAH is a novel clinical condition involving limited mouth-opening accompanied by hyperplasia of the masticatory muscle tendon and aponeurosis; this condition was first described by two Japanese groups [3,4]. MMTAH is a condition of unknown etiology, yet some authors have suggested that parafunctional habits like grinding and bruxism could be risk factors, as they might lead to hyperplasia of the tendon and aponeurosis of the masticatory muscles, thus causing muscle extension disorder [3]. The

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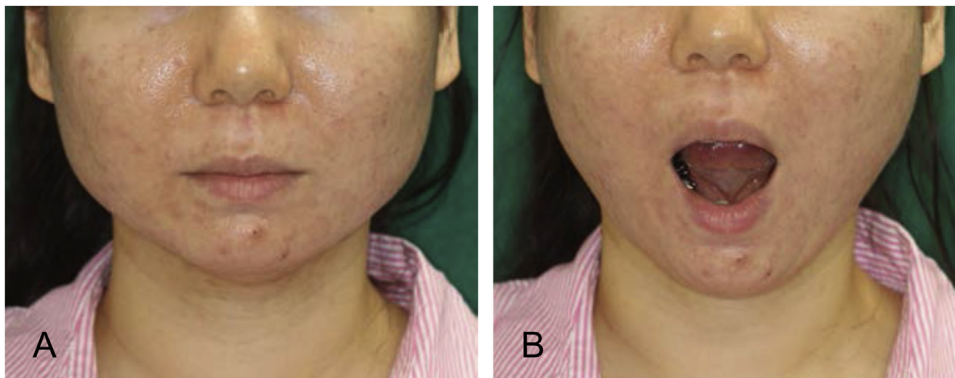


Fig. 1. Extraoral photograph at first visit.



Fig. 2. Panoramic X-ray at first visit.

characteristic features of patients who have MMTAH are as follows: a square mandible, cord-like masseter muscle aponeurosis, coronoid process hyperplasia, a small gonial angle, and limited mouth-opening [3,4]. Histopathological analysis of the cut tendon and muscle showed calcified nodules of silicon, calcium, and phosphorus [5,6] that explain the sound heard when the tendon is cut surgically. In this report, we describe the treatment of a case of MMTAH that was especially significant in the temporalis muscle tendon. This patient was diagnosed with temporomandibular joint disorder (TMD) and was treated for TMD for one year.

2. Case presentation

A 32-year-old female patient visited the general dental department in our hospital in August 2018 complaining of pain related to the bilateral temporal region and limited mouth-opening. The patient was previously diagnosed as TMD and treated with an oral splint for bruxism in a general dental clinic for one year before her first visit. Although she was treated with occlusal splint therapy continuously, her pain intensity did not show any improvement. She was referred to our department for further examination and treatment in October 2018. She has no past medical, surgical, family, psychosocial nor pharmacologic history. On inspection, the patient had a square configuration of the mandible (Fig. 1). Extraoral palpation revealed tenderness of both the temporalis and masseter muscles, though that of the temporalis muscles was more severe. Her maximal mouth-opening was 30 mm. A panoramic X-ray view was obtained and showed a square angle of the mandible and small gonial angles (Fig. 2). To ascertain that the trismus was

not related to an undiscovered TMD, computed tomography was performed, and it showed no obvious condylar changes, but both mandibular angles showed hyperplasia. In particular, the right mandibular angle was warped outward by the masseter muscle. Magnetic resonance imaging (MRI) was requested to determine the condition of the articular discs, muscles, and tendons. The findings showed that the articular discs exhibited intact physiologic disc shapes and positions. However, axial T1-weighted MRI showed enlarged masseter muscles (Fig. 3A, arrowheads), the axial short TI inversion recovery (STIR) view showed bilateral masseter muscle aponeurosis (Fig. 3B, arrowheads), and the axial STIR view showed bilateral thickened temporalis muscle tendons (Fig. 3C, arrowheads). The patient was diagnosed with MMTAH after the examination.

In December 2018, the patient underwent bilateral coronoidectomy under general anesthesia. After the anterior margin of the mandibular ramus was clearly indicated, the masseter aponeurosis hyperplasia on the periosteum was resected, but no firm fascia characteristic of MMTAH was found. However, the temporalis tendon and aponeurosis were firmly attached to the coronoid process (Fig. 4A (after removal) and B arrows), and a bilateral coronoidectomy was performed (Fig. 4C and D arrows). The masseter muscles were hypertrophic, and the right mandibular angle morphology was especially bent on the buccal side. To improve symmetry, a larger angle reduction was performed on the right than on the left. The mouth-opening was increased intra-operatively to 52 mm after the bilateral coronoidectomy. Fig. 4E shows the bilateral resected coronoid processes. The holes drilled in the coronoid processes were created so that the processes could be held with wires to

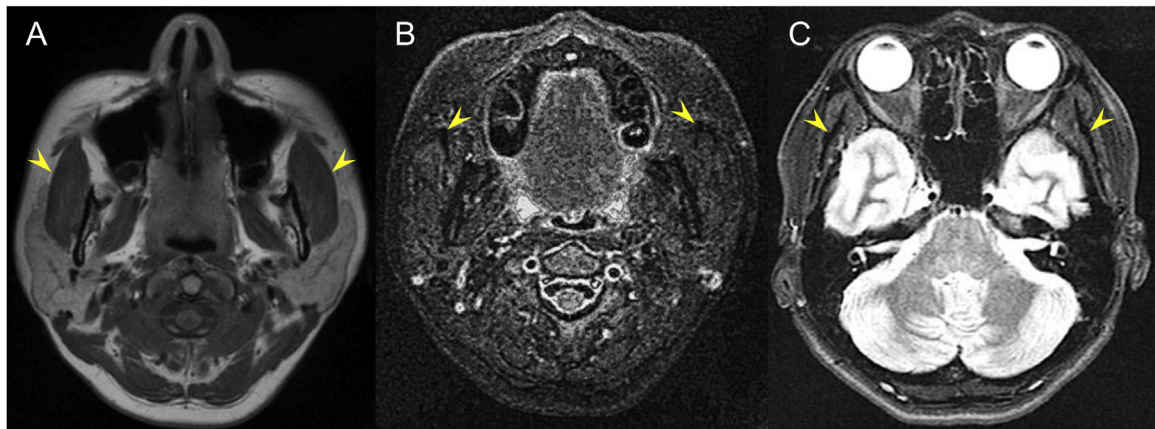


Fig. 3. Pre-operative magnetic resonance imaging (MRI). (A) Axial T1-weighted image (T1WI). Arrowheads: enlarged masseter muscles. (B and C) Axial short TI inversion recovery (STIR) view. Arrowheads: bilateral masseter muscle aponeurosis (B) and bilateral thickened temporalis muscle tendon (C).

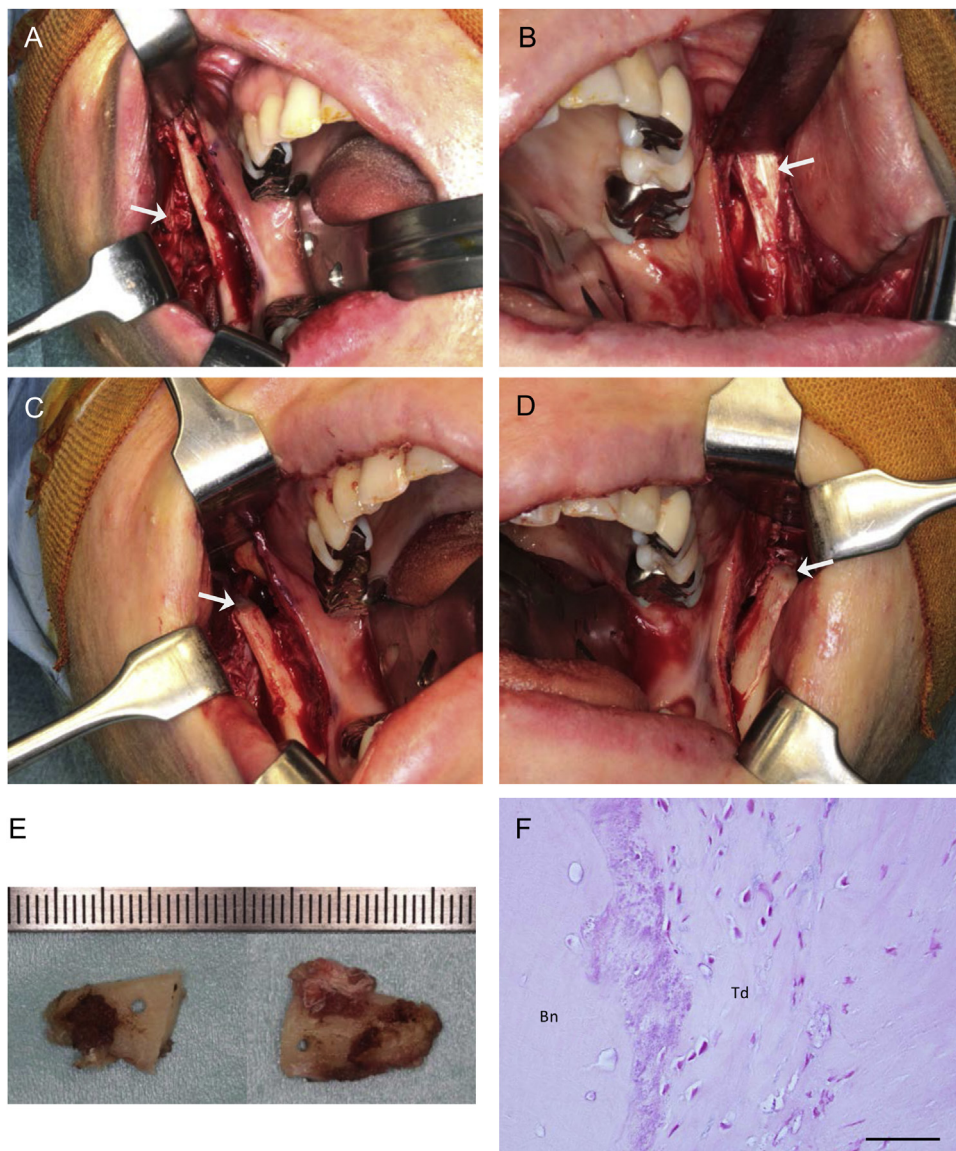


Fig. 4. Perioperative photo and histology of the coronoid process. Anterior border of the right (A) and left (B) ramus. Arrowheads: temporalis tendon. After the resection of the right (C) and left (D) coronoid processes. Arrowheads: resected edge of the coronoid process. E) Resected coronoid processes. F) Histology of the coronoid process H&E staining, bar 50 μ m. Bn: Bone, Td: Tendon.



Fig. 5. Post-operative panoramic X-ray.

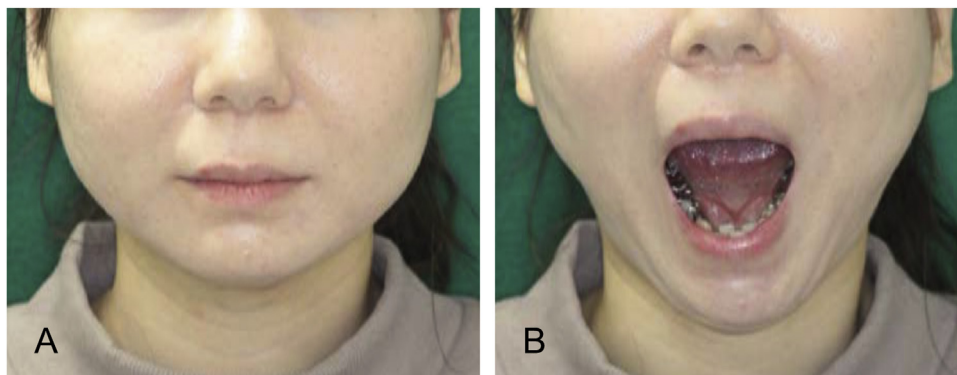


Fig. 6. Extraoral photograph one year after operation. Closed mouth (A) maximum opening (B).

prevent them from being pulled upward by the temporal muscle during the coronoidectomy. Pathological examination of the resected coronoid processes revealed that the temporal muscle tendon and aponeurosis tissues showed normal structures in H & E staining (Fig. 4F).

A postoperative panoramic X-ray view showed the resected coronoid processes and right mandibular angle compared with presurgery (Fig. 5). The rehabilitation program started four days after the surgery and included mouth-opening with the aid of a mouth opener (product number: 14674 (8-9286-01), YMD Co. Tokyo, Japan) four times per day, three times before each meal and one time before sleep. Each time, the patient had to hold her mouth open 40 mm for at least 30 s for at least half a year. Initially, the maximum mouth-opening after the surgery was 25 mm, but it gradually increased to 48 mm one week after surgery. Follow-up was performed in our hospital every few weeks for the first six months after discharge, thereafter every few months, and the amount of opening and the symptoms of the masseter and temporal muscles were assessed every time. The patient was instructed to continue using the night guard to prevent bruxism, and the maximum mouth-opening remained 48 mm at one year after surgery (Fig. 6A and B). Two years have passed now, but the maximum opening has been maintained at 48 mm, and the patient's progress is good.

3. Discussion

Patients who suffered from slowly progressing trismus and a cord-like aponeurosis were usually misdiagnosed as intractable type I TMD, hyperplasia of the mandibular coronoid process, or

masseter hypertrophy and were treated accordingly for a long time. H. Lehman et al. reported a case of limited mouth-opening who had been treated for a long time as having a disorder of muscular origin without success [7]. In 2005, MMTAH was recognized as a new disease at the conference for the Japanese Society for Oral and Maxillofacial Surgeons. Subsequently, the disease was defined and approved at the conference of the Japanese Society for Temporomandibular Joint in 2008 [8]. Since then, many clinicians have become aware of this particular condition, and different treatment modalities have been proposed. MMTAH can be treated conservatively with mouth-opening exercises, but they have not shown significant results. On the other hand, MMTAH can be treated surgically by aponeurectomy to relieve the muscle tension. However, aponeurectomy should be combined with coronoidectomy as reported by Yoda et al. [6]. Coronoidectomy or coronoidotomy was reported to remove the temporalis muscle tendon that is attached to the posterior and superior aspects of the coronoid process, thus improving the range of mouth-opening. In this case, we expected a long-term stable result without reattaching the temporalis muscle tendon to the coronoid process, so a more reliable coronoidectomy was performed. Mandibular anglectomy is also one of the surgical treatments used to release the pterygomasseteric sling, yet it has only achieved temporary results and is used mainly for esthetic purposes [4]. In our present case, we performed bilateral coronoidectomy combined with anglectomy in consideration of symmetry, owing to the fact that the temporalis muscle tendon was thickened. We consider this case to be an uncommon variant of classic MMTAH because it clearly presented with temporalis muscle tendon hyperplasia with or without affecting the masseter muscle, unlike in the usual presentation. Mouth-opening

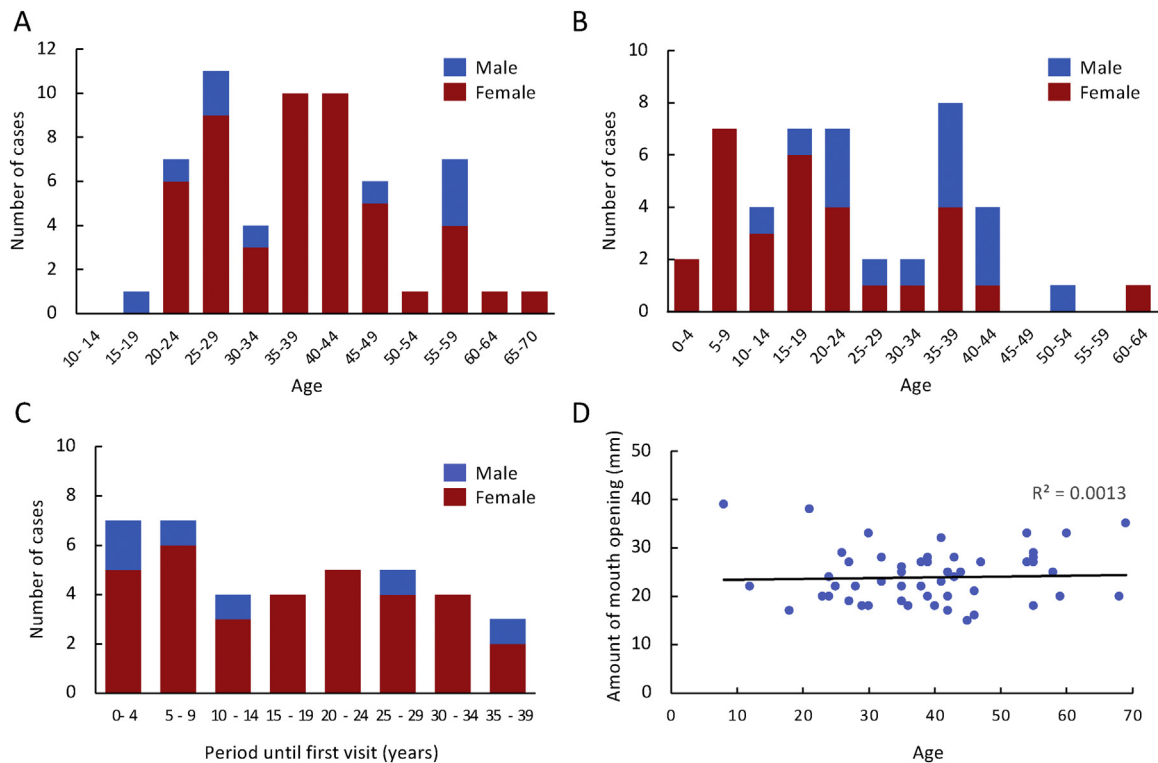


Fig. 7. Data collection from included studies grouped in graphs. A) The age distribution at the first visit. B) The age at which trismus was first identified. C) The time taken until the first visit. D) Scatter diagram analysis between the age at the first visit and the mouth-opening distance. The data were reviewed in references [4], [6], [7] and [9–11].

exercises after the surgery are crucial to prevent any limitation of mouth-opening, so they were started within the first week post-operatively, and the rehabilitation program was continued for six months. As previously mentioned, MMTAH is a silent condition that is identified clinically by a dentist or subjectively when the patient experiences limited mouth-opening. Upon a review of the literature in English, we found few case reports or clinical studies. K. Murakami et al. [4], T. Yoda [6], H. Lehman et al. [7], K. Nakaoka et al. [9], T. Sato et al. [10] and N. Hayashi et al. [11] reported jaw hypomobility due to hypertrophy of the masseter muscle tendon and aponeurosis. The disease was found to be more prevalent in females in a wide age range (Fig. 7A). A column graph shows that the incidence of the patient first visiting the dentist due to limited mouth-opening was highest in the age range of 25–29 years old, followed by 35–44 years (Fig. 7B). Data collected from the aforementioned articles showed that the age range in which the highest number of patients first identified their limited mouth-opening was 35–39 years, followed by 5–9 years and 15–24 years (Fig. 7C). Thus, there are big differences between the ages of patients when they identify their jaw hypomobility. By correlating this finding to the aforementioned one, we can anticipate that patients started to seek treatment when they encountered difficulty in opening their mouth during dental treatment. Additionally, we found that the time taken by patients to seek advice ranged between 0–39 years. The highest number of patients took from 0 to 9 years between their first identifying their difficulty in mouth-opening until their first visit seeking treatment. However, among these time intervals, the longest interval of 35–39 years occurred in the smallest number of patients. There was no correlation between the age of the patient and the amount of mouth-opening that they were capable of ($r^2 = 0.0013$) (Fig. 7D). A small number of patients visited the dentist at an advanced age because they experienced reasonable functional mouth-opening during their daily lives for many years. This report has been reported in line with the SCARE 2018 criteria [12].

4. Conclusion

Careful attention should be paid to this condition when a patient’s chief complaint is gradually decreasing mouth-opening with normal protrusive and lateral movements accompanied by thick masticatory muscle tendon-aponeurosis that is evident both clinically and on MRI. It is important that this condition not be misdiagnosed as TMD. In addition, it is important to evaluate the patient’s compliance with mouth-opening training before surgery, including the patient’s general condition and social background, as it can determine the outcome of the case.

Declaration of Competing Interest

None to declare.

Funding

None to declare.

Ethical approval

Ethical approval was exempted by our institution.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request. Identifiable patient information has been removed.

Author contribution

Nagwan Elsayed: Writing of original draft

Tsuyoshi Shimo: Main surgeon involved in care of patient and conceptualising and writing of the paper
 Fumiya Harada: Editing manuscript
 Shigehiro Takeda: Surgeon involved in care of patient
 Daichi Hiraki: Collection of data and analysis
 Yoshihiro Abiko: Pathological examination
 Eiji Nakayama: Analysis of the MRI data
 Hiroki Nagayasu: Contributed in critical reading

Registration of research studies

Not Applicable.

Guarantor

Tsuyoshi Shimo.

Provenance and peer review

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