Review Article

Masticatory muscle tendon-aponeurosis hyperplasia: A new clinical entity of limited mouth opening

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Summary Limited mouth opening is a common health problem that interferes with eating, makes examination of the oral cavity difficult, and may increase the mortality rate during emergency intubation. Here we introduce a disease designated as masticatory muscle tendon-aponeurosis hyperplasia, which is a new clinical condition of limited mouth opening. Most oral surgeons and dentists are still unaware of this disease condition, thus increasing the risk of incorrect diagnosis as some other disease, such as temporomandibular joint disorder. We will review the clinical features, epidemiology, pathophysiology, etiology, diagnosis, treatment, and prognosis of this disease and also appraise the literature available on the subject.

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1. Introduction

Limited mouth opening interferes with eating and medical procedures that require access to the oral cavity, thus increasing rates of mortality during emergency intubation [1,2]. It may be caused by various diseases and factors such as trauma, inflammation, temporomandibular joint disorders, neurological disorders, rheumatoid arthritis, side effects of drugs, tumors, and hyperplasia of the coronoid process [3–6]. In this review, we introduce a disease known as masticatory muscle tendon–aponeurosis hyperplasia (abbreviated MMTAH), a new clinical condition of limited mouth opening. We will review the clinical features, epidemiology, pathophysiology, etiology, diagnosis, treatment, and prognosis of this disease and also appraise the literature available on the subject.

This disease was first described by two Japanese groups, independently, in 2000. Inoue et al. reported a patient with hyperplasia of only the masseter muscle aponeurosis, accompanied by limited mouth opening and a square mandible [7]. Moreover, Murakami et al. [8] reported 10 chronic severe mandibular hypomobility patients exhibiting square mandible and hyperplasia of the coronoid process and the mandibular angle. In both the reports, the authors believed that the mandibular hypomobility was a result of contracture of the masticatory muscles and not coronoid process interference.

Thereafter, some case reports of this disease were presented in domestic conferences. 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 for the Japanese Society of Temporomandibular Joint in 2008 [9].

2. Clinical features

MMTAH is a condition in which the tendon and aponeurosis of the bilateral masticatory muscles exhibit hyperplasia, thus restricting muscle extension. One of the main symptoms of MMTAH is limited mouth opening. Because the condition progresses very slowly from adolescence, patients have few subjective symptoms and limited mouth opening may only be identified by dentists during visits to the dental clinics.

A characteristic feature of this disease is a square mandible with a small gonial angle and flattening of the occlusal planes (Fig. 1A–C). We have previously proposed that MMTAH patients can be classified into three categories, based on analysis of cephalometric radiographs [10], as follows: for Type 1, both occlusal plane and mandibular plane exhibits counter-clockwise rotation; for Type 2, only mandibular plane exhibits counter-clockwise rotation; for Type 3, both occlusal plane and mandibular plane exhibits no rotation.

Square mandibles which exhibit prominent mandibular angle may be due to a hyperplastic aponeurosis and tendon. Although the zygomatic bone thickens in the front, back, inside, and outside, the coronoid process does not interfere with it on mouth opening.

In addition, a bilateral overhang of the masseter muscle along the anterior border of the mandibular ramus is noted. Minowa et al. reported that aponeuroses of the masseter muscles are rarely found below the lower half of the anterior margin of the masseter muscles [11]. However, thickened aponeuroses are often observed to extend onto this lower half in patients with MMTAH. Kobayashi et al. reported that it is useful to visualize tendons and aponeuroses using magnetic resonance imaging (MRI) [12]. On maximal mouth opening, intraoral palpation along the anterior border of the masseter muscle reveals a hard cord-like structure, consistent with the findings on the MRI (Fig. 2).

3. Epidemiology

MMTAH is highly prevalent in women. Arika et al. reported that the ratio of men to women with MMTAH is 2:5 [13]. According to our preliminary investigation, approximately 1% of 1000 individuals in Japanese elementary and junior high schools suffered from MMTAH (unpublished data).

4. Pathophysiology

From a histopathological viewpoint, Chiba [14] observed that the tendons and aponeuroses in MMTAH appeared normal because of a lack of both inflammation and transformation, indicating that the excess tissue was a result of hyperplasia (Fig. 3A). However, the aberrant sound heard on cutting the tendon tissues with scissors during a surgery indicates hardness of these tissues, suggesting occurrence of anomalous changes [15]. We analyzed the microstructural characteristics of muscles and tendons in patients with MMTAH using electron microscopy and energy dispersive X-ray analysis to determine its elemental composition and compare it to patients with facial deformities (FD) and normal tendons [16]. We found that mineralized nodules were observed in the tendon tissues of patients with MMTAH but not in those with FD. These calcified regions contained irregular particles, 10–100 μm in diameter, as observed on scanning electron microscopy; they are probably responsible for the unusual sounds created when cutting the tendon with scissors during a surgery. On energy dispersive X-ray analysis, we observed silicon along with calcium and phosphorous in
the tendons of MMTAH patients (Fig. 3B). Notably, this silicon was detected in the uncalcified regions of the tendon, suggesting a possible role of silicon accumulation in the initial stages of heterotopic calcification. Although some investigators have reported that silicon depletion resulted in stunted growth and profound defects in the bone and connective tissues [17,18], a reliable experiment has suggested that silicon deficiency results in inhibition of growth plate closure and increase in longitudinal growth in vivo [19]. On the basis of our findings, we speculate that silicon accumulation is involved in heterotopic calcification, specifically occurring in the mandibular tendons or aponeuroses as a result of intramembranous ossification.

Moreover, we conducted basic research using proteomic analysis in the temporalis muscle tendon of MMTAH patients and compared it to FD. A pilot study conducted on one MMTAH patient and one FD patient, using two-dimensional fluorescence difference in gel electrophoresis system, revealed that four proteins were upregulated and three proteins were downregulated [20]. Furthermore, we also performed proteomic analysis of tendons from four test subjects and four control subjects using two-dimensional polyacrylamide gel electrophoresis with matrix-assisted laser desorption/ionization time-of-flight mass spectrometry system [21]. We observed that fibrinogen fragment D and β-crystallinA4 (CRYBA4) were upregulated whereas myosin light chain 4 (MYL4) was downregulated in MMTAH patients but not in FD patients. These studies demonstrated tendon sample analysis using a proteomic analysis for the first time in the oral-maxillofacial surgery field.

Inactivation of myosin II causes abnormalities in the collagen fibers of the tendon along with downregulation of MYL4, suggesting that MYL4 reduction is associated with pathological conditions in MMTAH. CRYBA4, which plays a vital role in protecting cells in non-lens tissues from damage caused by environmental and/or metabolic stress, is upregulated in tendons of MMTAH, thus indicating that it may have a role in the stress response or pathology associated with MMTAH development.

We also observed hyalinization, fatty infiltration, and microcalcification in MMTAH cases using electron microscopy [16]. These observations were also observed in tissues of
patients with tendinosis, where the tendons are characterized by ruptures and degenerative changes [22,23]. In tendinosis, fibrinogen exhibits a fine, granular, or linear pattern along the collagen fibers [24]. Accumulation of fibrinogen in the tendon tissue is typical in MMTAH, suggesting that it may resemble tendinosis.

Because we could not obtain the masseter muscle from patients with FD as it causes difficulties for the patients, we did not conduct the experiments in the masseter muscles of patients with MMTAH. Therefore, we could not clearly show the similarities and differences in pathology between the masseter and temporal muscles.

5. Etiology

The etiology of MMTAH still remains unclear, although parafunctional habits are often associated with it. Inoue et al. suggested that work hypertrophy played a role because patients were often observed to display clenching or grinding [7]. Because MMTAH progresses bilaterally and is a juvenile-onset disease, we hypothesized that disease progression involves both environmental and genetic factors.

6. Diagnosis

The following two findings help confirm MMTAH diagnosis:

(1) Limited mouth opening that progresses very slowly from adolescence and no limitation of lateral or anterior mandibular movement.

(2) There is no limitation of lateral or anterior mandibular movement.

(3) A hard cord-like structure found along the anterior border of the masseter muscle on intraoral palpation (Fig. 4).

Figure 2  Horizontal section in MRI. Overhang of masseter muscle along the anterior border of mandibular ramus (yellow arrow). (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

Figure 3  Histological analysis of MMTAH [14]. (A) Hematoxylin and eosin staining of tendon tissues. (B) Element mapping of tendon tissues. Silicon is indicated by red arrow. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)
Secondary diagnostic features include a square mandibular structure [15]. Although MRI can help visualize tendons and aponeuroses, the criteria for hyperplasia in these tissues have not yet been established. The main and key symptom of MMTAH is limited mouth opening. If the patient feels the limitation of mouth opening as compared to previous condition, he or she should consult with the doctor for care.

7. Treatment

Conservative treatment, including mouth opening training, has shown no significant effects. Surgical treatment options include aponeurectomy of the masseter muscle with coronoidectomy. Our previous paper was the first report of satisfactory outcome on surgical treatment of 10 MMTAH patients [15].

Although mandibular anglectomy or masseter muscle myotomy improves restricted mouth opening temporarily, good long-term results have not been obtained [7]. Murakami et al. [8] suggested that mandibular anglectomy was necessary to release the muscle sling of the medial pterygoid and masseter muscles. Aponeurectomy of the masseter muscle relieves tension in this muscle. Mandibular anglectomy is unnecessary, but can still be performed for esthetic reasons. Moroi et al. [25] and Yoshida et al. [26] also reported that they sometimes carry out mandibular anglectomy for a square mandible. Yoshida et al. [26] suggested that mandibular anglectomy induces disuse atrophy of the masseter and medial pterygoid muscles.

Coronoidectomy or coronoidotomy should be performed to remove the tendon arising from the posterior and superior aspects of the coronoid process. There are few reports of reformation or rebuilding of the coronoid process after coronoidectomy and/or coronoidotomy for hyperplasia. There are also few reports of reformation or rebuilding of the mandibular angle after mandibular anglectomy for a square mandible. These recurrent cases often cause limitations of mouth opening. Thus, most of these cases required another surgical operation [27–29]. Yoshida et al. [26] also reported a case of reformed coronoid process and mandibular angle after coronoidectomy and anglectomy for masticatory muscle tendon-aponeurosis hyperplasia. Furthermore, they suggested that postoperative mouth opening training is an important prognostic factor for preventing the limitation of mouth opening in masticatory muscle tendon aponeurosis hyperplasia. Other reports described mouth opening training as a rehabilitation exercise in detail [30,31].

Previously, we have reported a case presentation with videos [32], where we introduce the surgical procedures for MMTAH patients in detail. Surgical treatment is performed under general anesthesia. Muscular relaxation does not improve restricted mouth opening. Conforming to the...
incision line for sagittal split ramus osteotomy, an incision is made in the oral mucosa and the anterior margin of the mandibular ramus is exposed. The fascia of the masseter muscle on the outer aspect of the mandibular ramus will be visible, and on separating this fascia, the anterior margin of the masseter muscle can be clearly identified. The aponeurosis is revealed and exhibits a silvery-white color on exfoliating upward and outward (Fig. 5A). The aponeurosis is detached from the muscle tissue and excised as large as possible using scissors or electrosurgical knives. If the masseter muscle appears hard on palpation, the aponeurosis may be located within the muscle. Aponeurectomy of the masseter muscle should be performed bilaterally. The periosteum on the anterior margin of the mandibular ramus can be exfoliated upward. An abnormal sound can be heard on exfoliating this periosteum with a raspatory. The temporal tendon attached to the coronoid process is then exposed and subsequently removed from the coronoid process (Fig. 5B). Because the coronoid process does not interfere with the zygomatic bone, its apex can easily be identified. Although the posterior tendon should be cut as much as possible, it is often difficult to excise it completely. By clamping the coronoid process tightly with the help of a Pean or Kocher, we can excise it using cutting instruments such as the Lindemann drill. After confirming that mouth has opened >45 mm, the surgical incision can be closed using sutures. Continuous intraoral drainage is effective in reducing postoperative swelling and conducting mouth opening training. Immediate oral intake and training in intentional clenching are required postoperatively to prevent open bite. Mouth opening training should be started 5 days after surgical treatment. The training involves widely opening of the mouth for 30 s with the help of a mouth opener, which is repeated more than 3 times, using analgesics to control the pain. The patient is discharged once they can open the mouth >40 mm without using the mouth opener. This training should be continued for at least 6 months.

8. Prognosis

Long-term satisfactory results can be obtained by continuous mouth opening training. Our preliminary study revealed that, although immediate postoperative occlusal force was less than half the standard force, occlusal force returned to the normal range within 6 months (unpublished data). After 1 year, the temporal muscle was observed to reattach to the resected stump of the bone (Fig. 6A and B) [33]. A reduction in the cross-sectional area of the masseter muscle was observed, despite resection of only the tendon without the muscle (Fig. 6C and D).

9. Related papers

Unfortunately, MMTAH is not well recognized in countries other than Japan. Beckers [34] reported that bilateral
resection of the masseter muscles did not improve limited mouth opening in patients diagnosed with bilateral masseter muscle hypertrophy. MM-at-H should be considered in this case. Recently, however, the first case of MM-at-H outside Japan was reported in Switzerland [35]. In contrast, Lehman et al. reported that four female patients presenting with limited mouth opening and lateral and protrusive movements within the normal range had been treated by bilateral coronoidectomy and showed a significant improvement in mouth opening [36]. Although they did not mention our papers, we consider that these cases must be MM-at-H [37]. Anesthesiologists also reported cases of MM-at-H that resulted in difficult laryngoscopy after initiation of anesthesia [38]. In case of emergency intubation, limited mouth opening may increase the mortality rates.

10. Conclusion

At present, most clinicians are unaware of this condition, resulting in an increased risk of incorrect diagnosis as other diseases, such as masseter muscle hypertrophy and temporomandibular joint disorders. Some patients may have undergone irreversible surgeries. When oral surgeons, dentists, and general physicians notice limited mouth opening on examination of the oral cavity, they should have sufficient knowledge of the diseases such as MM-at-H that may have caused it.

Ethical statement

Informed consent was obtained from an individual participant included in the study.

Conflict of interests

None declared.

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