

# A two-year follow-up of surgical and non-surgical treatments in patients with masticatory muscle tendon-aponeurosis hyperplasia

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**Abstract.** This study re-examined the usefulness of surgery for the management of masticatory muscle tendon-aponeurosis hyperplasia (MMTAH) through a comparison of the outcomes between patients who underwent surgery and those who did not. The duration of follow-up was 2 years. Twenty-eight patients who attended the study hospital and were given a diagnosis of MMTAH were included. Nineteen patients underwent surgery (surgical group) and nine patients were instructed to open their mouths wide once a day and did not undergo surgery (non-surgical group). Maximum mouth opening, impairment of daily activities, satisfaction, and the status of mouth opening training were evaluated after surgery. The mean increase in mouth opening after 2 years was 20.2 mm in the surgical group and 2.4 mm in the non-surgical group. Adequate mouth opening training led to satisfactory results 2 years postoperative, and sustained mouth opening training for 6 months after surgery was a key factor for obtaining good outcomes. The general condition and personality of individual patients should be evaluated carefully before surgery to estimate whether or not they can endure the pain associated with postoperative mouth opening training. The results of this study suggest that the surgical procedure is useful for the management of MMTAH.

**Key words:** masticatory muscle; tendon-aponeurosis; hyperplasia; limited mouth opening; non-surgical treatment; surgical treatment.

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Masticatory muscle tendon-aponeurosis hyperplasia (MMTAH) is a condition characterized by bilateral hyperplasia of the tendons and aponeuroses of the masticatory muscles, which restricts muscle

extension. Patients with MMTAH thus present with limited mouth opening.

MMTAH was designated and approved as a new disease entity by the Japanese Society for the Temporomandibular Joint

in 2008<sup>1</sup>, and is becoming a frequently reported disease in Japan. Unfortunately, it is not recognized as a disease entity by most clinicians throughout the world, potentially leading to misdiagnoses such as

temporomandibular disorders. However, in 2014, Worni et al. reported a case in Switzerland<sup>2</sup>. In 2015, Lehman et al. reported four female patients who presented with limited mouth opening who had lateral and protrusive movements within normal limits. These patients underwent bilateral coronoidectomy, which resulted in a clinically significant improvement in their limited mouth opening<sup>3</sup>. The present authors consider it likely that these patients had MMTAH<sup>4</sup>.

The limited mouth opening in MMTAH progresses very slowly from adolescence. A characteristic feature of MMTAH is a square mandible associated with a small gonial angle and flattening of both the mandibular plane and occlusal plane<sup>5</sup>. Although the coronoid process thickens longitudinally and laterally, it does not interfere with the zygomatic bone, in contrast to mandibular coronoid process hyperplasia. On maximum mouth opening, intraoral palpation along the anterior border of the masseter muscle can confirm a hard, cord-like structure resulting from hyperplasia of the aponeuroses<sup>1,6</sup>; this is the most typical clinical condition used to diagnose MMTAH.

Histopathologically, the tendons and aponeuroses in MMTAH appear normal because of a lack of both inflammation and transformation; the excess tissue is thus a result of hyperplasia<sup>7</sup>. However, electron microscopy has shown the presence of calcified nodules containing irregular particles of 10–100  $\mu\text{m}$  in diameter in tendon tissues affected by MMTAH, and in addition to calcium and phosphorus, silicon has also been detected on energy dispersive X-ray analysis of the tendons of patients with MMTAH<sup>8</sup>. Furthermore, proteomic analysis has demonstrated up-regulation of fibrinogen fragment D and  $\beta$ -crystallin A4 (CRYBA4) levels and down-regulation of myosin light chain 4 (MYL4) levels<sup>9,10</sup>.

The aetiology of MMTAH remains unclear. Parafunctional habits are often associated with MMTAH. Inoue et al. suggested that work hypertrophy is involved because patients often display clenching or grinding<sup>11</sup>. As MMTAH is a juvenile-onset disease that develops bilaterally, it is hypothesized that disease progression involves both environmental and genetic factors<sup>6</sup>.

Pharmacotherapy, occlusal splint therapy, and physical therapy are ineffective. Although it has previously been reported that surgical treatment including aponeuroctomy of the masseter muscle with coronoidectomy is effective<sup>6</sup>, there have been no reports directly comparing surgical

treatment with non-surgical treatment. Furthermore, sufficient mouth opening cannot be obtained for all patients after surgery. The present authors believe that mouth opening training might be related to the prognosis of MMTAH.

In this study, the outcomes at 2 years after surgical treatment were compared with those obtained with non-surgical treatment. The relationship between post-operative mouth opening training and treatment outcomes was also examined.

## Patients and methods

### Patients

This was a retrospective study involving 28 consecutive patients who attended the Department of Oral and Maxillofacial Surgery, Saitama Medical University Hospital, with a chief complaint of limited mouth opening between January 2004 and March 2014, and given a diagnosis of MMTAH. MMTAH was diagnosed on the basis of the following two findings: (1) limited mouth opening progressing very slowly from adolescence, and (2) the presence of a hard, cord-like structure on intraoral palpation along the anterior border of the bilateral masseter muscles. The presence of a 'square mandible' configuration was an auxiliary factor in the diagnosis<sup>6</sup>.

Magnetic resonance imaging (MRI) showed that there were no abnormalities in the position or shape of the articular disc in all patients. Although patient 22 had undergone bilateral arthroscopic surgery of the temporomandibular joint (TMJ) at another hospital 6 years ago, the position of the articular disc was normal. Additionally, it was confirmed that there was no clicking of the TMJ and no limitation of lateral or anterior mandibular movement by palpation in all patients, with the exception of patient 22. With regard to patient 22, fibrous adhesions in the upper joint space were suspected on arthroscopy.

The surgical treatment was explained to all patients, and the patients themselves decided whether or not to undergo this surgery. Those who chose not to undergo surgery were instructed to open their mouths wide once a day and were asked to return to the hospital every 4 months for an assessment of their mouth opening.

### Surgical procedures

The following surgical procedures were performed for all patients who elected to undergo surgical treatment<sup>6,12</sup>: With the patient under general anaesthesia, an inci-

sion was made in the oral mucosa overlying the external oblique line. The anterior part of the masseter muscle was bluntly stripped to expose the silvery white aponeurosis (Fig. 1). A section of the anterior part of the aponeurosis of approximately 20 × 30 mm in size was then resected bilaterally. The temporal tendon attached to the coronoid process was exposed and subsequently removed from the coronoid process. Because the coronoid process does not interfere with the zygomatic bone, it was possible to identify the apex of the coronoid process. To completely release the tendon, especially the parts arising from the posterior and superior aspects of the coronoid process, a coronoidectomy was performed bilaterally. The excision of the coronoid process was evaluated by panoramic radiography.

### Postoperative mouth opening training procedures

To prevent an open bite, oral intake and intentional clenching training are required immediately after surgery. Mouth opening training was commenced on the fifth day postoperative. The mouth was opened wide to the point of endurable pain, for 30 seconds, using a mouth gag (Fig. 2). This procedure was repeated at least three times per day. Discharge was considered when the patient was able to open their mouth more than 40 mm using the mouth gag on their own. The same training was continued by the patient at home for 2 months after discharge and performed daily until 6 months after surgery. At 6 months after surgery, the patients were instructed to check their mouth opening once per day and to perform the training if they were unable to open their mouth wide.

### Evaluations

#### Maximum mouth opening

For the patients who underwent surgery (surgical group), the maximum unassisted mouth opening was measured at the following times: first examination and 1 month, 6 months, 1 year, and 2 years postoperative. The maximum assisted mouth opening with a mouth gag was measured immediately after surgery in the operating room. For the patients who did not undergo surgery (non-surgical group), the maximum unassisted mouth opening was measured at the following times: first examination, 6 months later, 1 year later, and 2 years later.

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