

Myositis ossificans traumatica of the temporalis muscle: a case report and diagnostic considerations

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Abstract

Background The present paper reported the case of a trauma-related myositis ossificans, with focus on considerations for a differential diagnosis process.

Case report A 50-year-old male with a severe painful limitation (12 mm) of jaw opening referred a trauma to the right temporomandibular joint (TMJ) area occurring about 40 days before. Posttraumatic TMJ ankylosis was ruled out on the basis of negative magnetic resonance and cone-beam computerized tomography findings, and the patient underwent treatment with arthrocentesis, botulinum toxin injections, and physiotherapy on the basis of two diagnostic hypotheses, viz., an anchored disk phenomenon or a myofibrotic contracture of the right masseter muscle due to prolonged myospasm. After 4 months, jaw opening was slightly increased to 23 mm, but limitation and pain persisted. A new CT was performed to investigate for the emerging clinical picture compatible with traumatic myositis ossificans of the right temporalis muscle. Once the diagnosis was confirmed, the patient underwent surgery for coronoidectomy. At the 6-month follow-up, mouth opening was increased to up to 35 mm and pain was absent.

Discussion It is fundamental that patients suspected of having uncommon clinical pictures leading to mouth opening restriction are promptly referred to specialized centers, where the differential diagnosis process should be

based on a comprehensive assessment taking into account for the potential etiologic factors described in the literature.

Keywords Myositis ossificans · Differential diagnosis · Temporalis muscle · Temporomandibular disorders · Coronoidectomy

Background

Myositis ossificans (MO) is a relatively rare disease characterized by bone neoformation in extraskelatal sites [1]. The disease can be either of primary or traumatic origin. Primary MO, known as progressive myositis ossificans or Munchmeyer's disease, is an hereditary condition with an autosomal dominant transmission occurring early during infancy and involves several muscles. Skeletal abnormalities, sexual development disorders, and deafness are usually associated with the disease, resulting in progressive functional limitations and a severe impairment [2, 3].

Myositis ossificans traumatica (MOT) is a more localized form involving muscles subjected to violent and/or repeated trauma. A palpable tumor-like calcified mass is often found within the injured muscle [4]. Common examples are horse-riders' bone, calvarymen's osseous plate on the outer thigh, and infantrymen's drill bone on the deltoid. Reports of this pathology occurring in the region of head and neck are rare, and there are very few cases described in the muscles of mastication [5]. In those cases, jaw range of motion limitation usually occurs, and the differential diagnosis has to exclude other potential causes of restricted mouth opening.

Considering these premises, the present paper reported the case of a trauma-related myositis ossificans, with focus on considerations for a differential diagnosis process.

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Case report

A 50-year-old male (LS) was referred from another hospital to the Department of Maxillofacial Surgery, University of Padova, Italy with a severe painful limitation of jaw opening. The patient, a professional carpenter, referred that symptoms and mouth opening restriction has occurred after a trauma injury (i.e., a piece of furniture sliding from its supports and hitting him hardly to the right temporalis area) dating back to about 40 days before. Pain developed immediately after the trauma, reached a peak (7 points on a 10-mm visual analog scale (VAS) with 0 being absence of pain and 10 being the worst pain ever experienced) about 2 weeks later, and then progressively decreased for the next couple of weeks without ceasing. After the trauma, jaw range of motion progressively decreased, and the patient asked for professional advice as soon as he was not able to eat comfortably and the pain increased again (approximately 1 month after the trauma). In the first hospital, the patient underwent both magnetic resonance (MR) and cone-beam computerized tomography (CBCT) of the temporomandibular joints (TMJ) to test for the diagnostic hypothesis of traumatic ankylosis of the right TMJ (Fig. 1). According to the first aid care givers, both imaging techniques were negative; attempts to force mouth opening during sedation were unsuccessful and symptoms did not disappear after a 1-week treatment with corticosteroids, so the patient was referred to our department.

At the first appointment, the patient had a maximum mouth opening (MMO) of 12 mm, not increasing with forced assistance (Fig. 2). The right and left lateral excursions were 6 mm and 1 mm, respectively, and protrusion was 3 mm. Pain level was rated 8/10 in a VAS. Palpation of the jaw muscles provoked pain in the right temporal and masseter areas and revealed muscle contracture and hypertone localized in the same areas. On the basis of the negative MR and CT, which were not sent to our observation, two diagnostic hypotheses were considered, viz., an anchored disk phenomenon [6] or a myofibrotic contracture of the right masseter muscle due to prolonged myospasm [7].

The patient underwent a bilateral intra-articular injection of mepivacaine 2% (Carbocaine, Sanofi Winthrop, NY,

USA), and an arthrocentesis of the TMJ was performed according to a single-needle technique increasing the injection–ejection fluid pressure [8]. The single-needle technique provides the under pressure fluid injection with the patient in a mouth-open position, in order to expand the joint cavity; after the injection, the patient is asked to close the mouth and the fluid is taken off with the same injection needle. The fluid injection–ejection process must be performed for up to 10 repetitions (for a total amount of about 40 ml). The under pressure injection of fluid is mostly useful to break joint adhesences that are responsible for the reduced translatory movement of the condyle and are mainly called into cause to explain the phenomena of disk anchorage to the fossa and/or eminence, thus allowing an immediate improvement in mouth opening. This makes the single-needle technique indicated in the case of hypomobile joints with strong adhesences or joints with degenerative changes that make the insertion of the second needle difficult, thus seeming the most indicated strategy to help the patient restoring normal mouth opening. In the immediate postinjection phases, the patient was subjected to forcedly assisted physiotherapy with the aim to increase jaw range of motion, and a 16-mm mouth opening was achieved. After 1 week, at the first follow-up appointment, mouth opening was decreased to 10 mm, VAS score was 3/10 and muscle contracture was still detected with palpation.

Thus, bilateral botulinum toxin injections were then performed in the masseter and temporalis area with a total of about 150 U of drug (Dysport®, Ipsen, UK) injected per treated side. The application technique adopted in the investigation was described elsewhere [9] and already adopted in a previous investigation [10]. The patient was asked to clench the jaws in order to properly identify the muscle to be injected and then multiple injections were performed in the more prominent area of the muscles, with an injection covering on average a 2-cm skin surface over the target muscle tissue. A minimum of five injections with a reverse pyramid pattern were performed in the masseter muscles, and a chessboard pattern was used for the temporalis muscles. After 1 month, jaw range of motion was not improved significantly yet (MMO=12 mm; right, left, and protrusive movements=8, 3, 6 mm, respectively)

Fig. 1 Cone-beam computerized tomography performed in the first hospital focused only on the area around the temporomandibular joint condyle

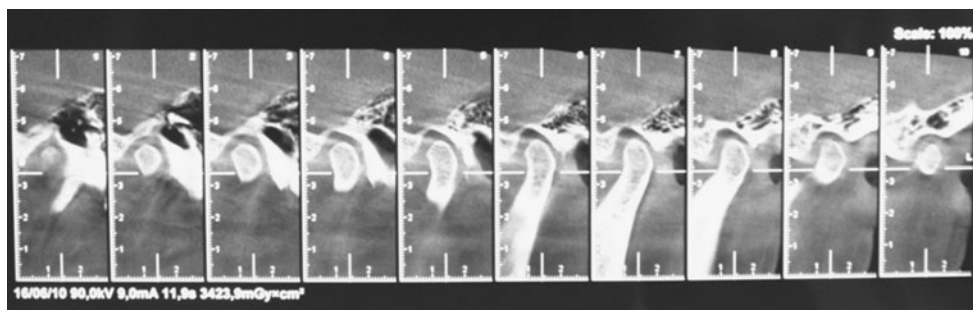




Fig. 2 Preoperative mouth opening

and the patient was encouraged to start a home-based protocol of passive physiotherapy with a commercially available device (TheraBite Jaw Motion Rehabilitation System®, Therabite, Philadelphia, PA, USA).

At the 4-month follow-up, mouth opening was improved to 20 mm, but no improvement was recorded as for right, left, and protrusive movements, and VAS score was still 4/10. Because of persistent limitation, another TMJ arthrocentesis was programmed now followed by hyaluronic acid injection (Hyalgan®, Fidia, Abano Terme, Italy). One week later, MO was increased up to 23 mm, but VAS was still 3/10. Importantly, the feeling of an hard end-feel maneuver never disappeared, thus suggesting that obstacles to a full-mouth opening could not be only related to the early hypotheses of muscle spasm and/or anchored TMJ disk phenomenon.

The emerging clinical picture was compatible with traumatic myositis ossificans, and a new CT focusing not only on the TMJ area was prescribed to investigate for the above hypothesis. A 3D CT confirmed that a tumor-like osseous neoformation was present in the area of the insertion of the right temporalis muscle on the coronoid process (Fig. 3), and the patient was planned for a surgical intervention of coronoidectomy.

The classical intraoral approach to the coronoid process area was adopted by a mucoperiosteal incision of the retromolar area. Soft tissues were lifted to achieve an optimal exposition of the anterior margin of the mandibular ramus and the coronoid process (Fig. 4). A couple of subperiosteal channeled retractors were placed near the sigmoid incisures to protect soft tissues on both lingual and buccal sides. A Lindemann bur was used to perform an horizontal osteotomy of the coronoid process at the level of the sigmoid incisures basis, and the calcified attachment of the temporalis muscles on the process was detached with concurrent removal of the coronoid process. During

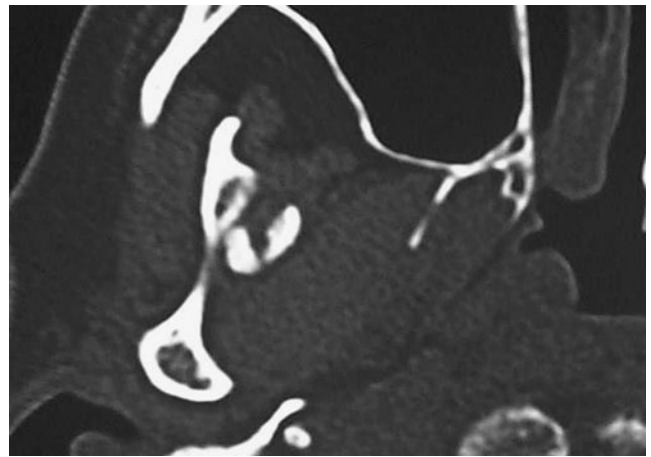


Fig. 3 Computerized tomography showing the newborn bone tissue attached to the right coronoid process

surgery, forced mouth opening was performed to assess the jaw range of motion, and tissues were then sutured. The postoperative course was uneventful, and the patient was advised to keep on performing physiotherapy during rehabilitation. At 1 month postsurgery, mouth opening was improved and pain was absent. At the 6-month follow-up, mouth opening was increased up to 35 mm and pain was still absent (Figs. 5 and 6).

Discussion

The present paper reported the case of a patient developing myositis ossificans after a violent trauma in the temporalis area. The literature on such disease occurring in jaw muscles is scarce, with only 23 cases reported in the 1980s and 1990s [11]. Over the years, several theories were proposed to hypothesize the pathogenesis of MOT, but little is known about the mechanisms involved at the tissue and cellular levels leading to ossifications of extra-osseous sites



Fig. 4 Surgical intervention for coronoidectomy. Intraoral access to the mandibular ramus and dissection of the coronoid process

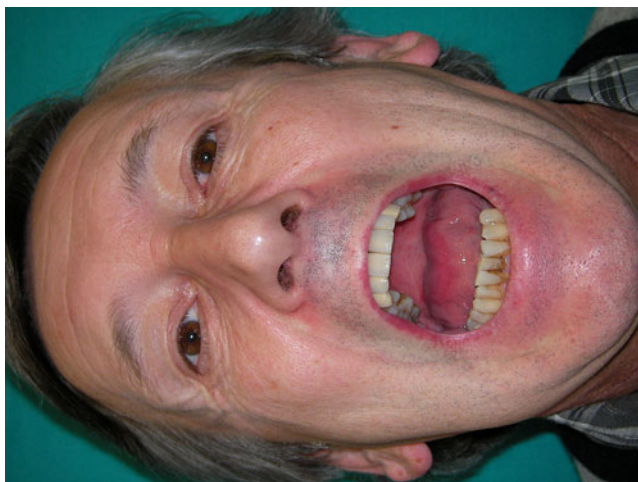


Fig. 5 Postoperative mouth opening

after a trauma. It is likely that bone tissue growth within muscles is due to metaplasia of connective tissues cells after bleeding from a trauma and myonecrosis, but it was also hypothesized that the penetration of periosteum fragments with osteogenic cells into the muscle is a potential factor for the onset of myositis ossificans.

Notwithstanding that, and since little is known on its actual pathogenesis, from a clinical viewpoint, the most interesting issue for discussion is represented by the difficulties in differential diagnosis. In the present case, time was spent before the correct diagnosis was made, and the negative MR and CBCT scans performed in the first hospital, which were mistakenly performed with focus on the temporomandibular joints only, were surely a key factor to address clinicians toward a combination of an intra-articular (e.g., anchored disk phenomenon) and an extra-articular (e.g., muscle spasm) diagnostic hypotheses. Temporomandibular joint ankylosis, enlargement of the coronoid process, and tumor were other three potential diagnoses to be considered when restricted mouth opening



Fig. 6 Orthopantomography at 6 months postsurgery showing the dissection of the right coronoid process

is observed, but they were excluded in this case because of the sudden onset occurrence of jaw motion limitation, which is not typical of any of the above diseases.

The fact that TMJ arthrocentesis and botulinum toxin injections aiming to relapse joint adhesions and muscle contracture as well as to achieve pain relief allowed achieving small improvement in jaw range of motion and that the patient was strongly motivated to perform a home regime of physiotherapy represented other confounding factors for the diagnosis with respect to other literature cases, which usually did not report any preoperative improvement independently by the therapeutic regime adopted. So, this is an example of the diagnostic challenges encountered when rare disease occur in body district or areas where they are not frequently observed. The temporomandibular disorders, orofacial pain, and maxillofacial surgery literature is plenty of studies describing the complex diagnostic pathway leading to the detection of the cause for jaw motion limitation [12–14]. In all cases, the choice of the right imaging technique prescribed by expert practitioners on the basis of a plausible diagnostic hypothesis is the fundamental step for achieving the correct diagnosis [15].

In the case under description, some interesting aspects are worthy to be also discussed. First, no fractures apparently occurred in the coronoid process area, thus bone formation in the temporalis tendon was due to the coagulation of posttraumatic fluids; second, no other areas of ectopic bone formation were referred, and laboratory exams did not show any abnormalities as for bone metabolism; third, there was also the possibility of an iatrogenic origin of the temporalis muscle ossification, related with the trauma of the needles adopted for botulinum toxin injections, but this rare occurrence was not compatible with the clinical picture and patient's history, which showed pain symptoms and mouth opening limitation already before the injections.

Myositis ossificans traumatica is a benign disease and surgical coronoidectomy is the treatment of choice when it is localized at the insertion of the temporalis muscle on the coronoid process. The uneventful postoperative course and the quick establishing improvement after surgery were common with similar interventions due to coronoid hyperplasia, but in case of long-lasting preoperative MOT-related pain, one cannot exclude that central sensitization phenomena, viz., pain is still present after its peripheral cause is removed [16], occurs if surgery is not performed in a reasonable time after trauma and symptoms onset.

In consideration of the above, it is fundamental that patients suspected of having uncommon clinical pictures leading to mouth opening restriction are promptly referred to specialized centers where the differential diagnosis process should be based on a comprehensive assessment taking into account for the potential etiologic factors described in the literature.

Conflicts of interest The authors declare no conflicts of interest.

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