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SINE QUA NON RADIOLOGY-PATHOLOGY

Myositis Ossificans of the Temporalis Muscle

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Abstract Traumatic myositis ossificans (TMO) is a rare ossifying disease that occurs in the muscle or soft tissues. A case of TMO isolated in the temporalis muscle is reported. In the case described, calcification in the temporalis muscle was confirmed after computed tomography. Surgery, physiotherapy, and histopathological analysis were performed. One year after treatment, further ossification was present but without interference in function. The most accepted treatment for TMO in the maxillofacial region is excision followed by physiotherapy. The high rate of non-recurrence may be concealed due to the short follow-up period. TMO is a lesion that may frequently recur and long-term follow-up must be provided.

Keywords Myositis ossificans · Masticatory muscles · Heterotopic calcification · Closed head injuries · Stomatognathic system

Introduction

Myositis ossificans (MO) is a rare ossifying disease that occurs in the muscle or soft tissues. It is divided into progressive ossifying myositis (POM), also called fibrodysplasia ossificans progressive [1] or Muncher-Meyer disease [2], a dominant autosomal hereditary disease with systemic manifestations; and traumatic myositis ossificans (TMO) in which a palpable mass appears more locally [2–8]. Other names for TMO are: circumscribed myositis ossificans (when a specific history of trauma can't be identified), localized myositis ossificans, ossifying hematoma, and heterotopic non-neoplastic bone formation [1].

The first report found about this condition was reported by Ivy and Eby [5, 9] in 1924. TMO is caused by a major trauma episode or multiple less traumatic events [5, 8, 10].

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TMO is widely reported in the orthopedic literature [1, 7, 10, 11]. The quadriceps femoris and brachialis anticus are the most affected muscles, particularly in young athletes [2, 5, 8, 10, 12–15]. In the facial region, it frequently occurs in the third decade of life and the most affected muscle is the masseter [2–8, 11, 14]. The temporalis muscle seems to be less affected than the masseter and medial pterygoid muscles. Only six cases have been described in the literature involving the temporalis muscle in isolation without association of other facial muscles; the seventh case is reported in the present article. Other muscles affected involving the mandible are the medial and lateral pterygoid, mentalis, buccinator, genioglossus, platysma and others of the neck area [1, 10]. In all of the cases of TMO, there are traumatic antecedents [5, 14], with rare exceptions.

To date, the nature and cause of TMO has not been fully elucidated [8, 9, 11], suggesting that metaplasia of connective tissues, hematoma ossifications, and penetration of bone fragment into the muscle with osteogenic cells are all possible etiologies [4, 5, 7, 8, 10, 15, 16]. However, they all seem to occur as an aberrant form of the physiological healing process. Carey [17] proposes four main theories for the development of TMO, as follows: displacement of bone fragments in the soft tissue with subsequent proliferation; detachment of periosteal fragments into the surrounding tissue with proliferation of osteoprogenitor cells; migration of subperiosteal osteoprogenitor cells around the soft tissue through periosteal perforation induced by the trauma; and metaplasia of extraosseous cells exposed to bone morphogenetic proteins (BMPs) derived from lysis of the bone fragments displaced within the soft tissue during traumatic injury.

The first detection of TMO is commonly 3–4 weeks after trauma, but it can take up to 20 years before the first symptoms appear [13, 18].

TMO is frequently treated by surgical procedures, including ossification excision, but some patients have repeated recurrence and they become treatment resistant [3, 7].

The objective of this study is to report a case of TMO of temporalis not associated with others masticatory muscles.

Case Report

A 17-year-old boy, victim of a traffic accident, suffered laceration in the infra-orbital region and zygomatic-orbital complex fracture on the right side. Surgical reduction and fixation of fractures was performed without intercurrences. During the postoperative period, the patient did not present improvement in mouth opening although there was no zygomatic arch interference. Even after intensive physiotherapy, mouth opening could not be recovered.

The CT showed the presence of cortical thickening and solid periosteal reaction of the coronoid process of the mandible associated with increase in volume of the surrounding soft tissues and obliteration of the retromaxillar fat suggesting osteomyelitis or a non-infectious inflammatory process 30 days after fracture reduction (Fig. 1). The patient showed no laboratory alterations in his levels of calcium, phosphorous, alkaline phosphatase, vitamin D, calcitonin, parathyroid hormone, or osteocalcin.

Coronoidectomy with excision of the mass was performed intraorally and the release movement of mouth opening was immediately observed. Specimens were submitted to histopathological analysis. Microscopically, osseous metaplasia in the striated muscle tissue was observed with ossification of the interfascicular connective tissue within the striated muscle tissue. Immature bone was present in the periphery of the muscle tissue, in continuity with mature lamellar bone, showing numerous lacunae filled with osteocytes (Figs. 2, 3).

In the postoperative period after excision of the lesion, the patient underwent physiotherapy consisting of mouth opening exercises until normal mouth opening was achieved. During one-year of control/follow-up, the patient reported discomfort in the right retromolar region and a new CT showed further ossification in the temporalis muscle, but without interfering in mouth opening (Fig. 4). By the time of 30 months of follow-up the patient had no complaints or limitations of mandibular movements.

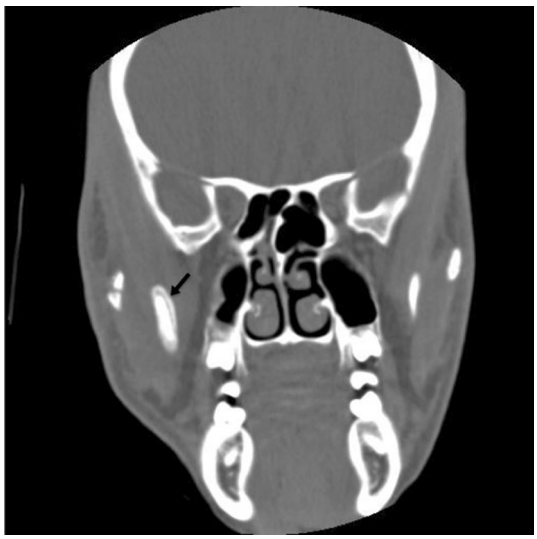

Fig. 1 CT 30 days after fracture reduction with cortical thickening and solid periosteal reaction of the coronoid process (*arrow*) of the mandible associated with increase in volume of the surrounding soft tissues

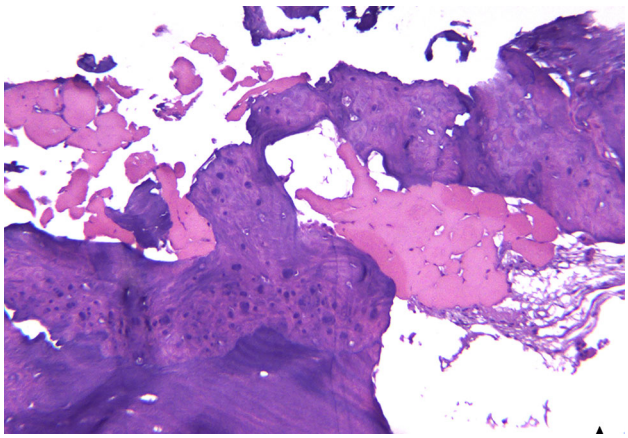

Fig. 2 Hematoxylin and eosin (HE) staining. Osseous metaplasia occurring in the striated muscle tissue (100X)

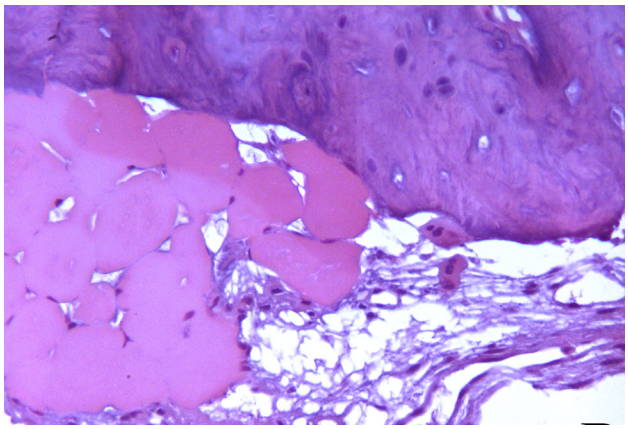

Fig. 3 High magnification of Fig. 2 (400X)

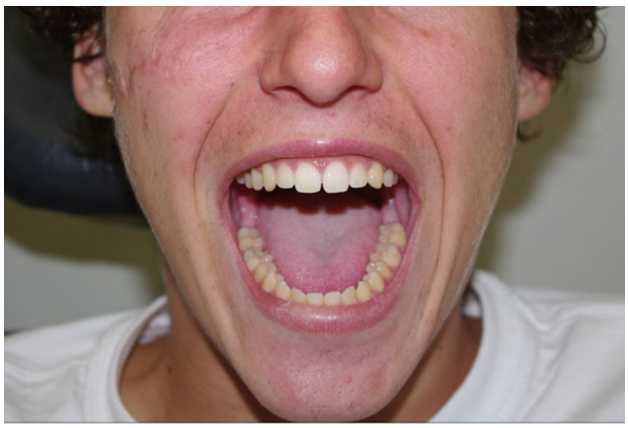

Fig. 4 Normal mouth opening 12 months after surgery

Discussion

TMO is frequently related to a single major traumatic event. On the other hand, in the orthopedic literature, injuries caused by repetitive low intense trauma are frequent, as those that occur in the deltoid muscle of soldiers or the adductors of horse riders [5, 8, 11, 12, 15].

TMO most commonly occurs in men during the third decade of life. In the masticatory muscles the ratio found of 2.23:1 of men in comparison with woman is similar to that found by Aoki et al. [3] (16:7 = 2.28:1).

The masseter muscle is the most frequently affected masticatory muscle due to its external position, which makes it more susceptible to trauma [3, 4]. Seventeen cases of TMO involving the temporalis muscle are described in the literature, seven isolated lesions (including the one reported in this article) and ten involving other masticatory muscles. After the masseter muscle, the medial pterygoid seems to be the most affected, followed by the temporalis and lateral pterygoid muscle in this order. In the case presented the patient had involvement only of the temporalis muscle without other muscles associated.

In most cases, the signs and symptoms of the TMO start immediately after the injury or up to 6 months later, but they may appear 20 years after trauma, as in some cases. These signs and symptoms may include trismus, firm/solid persistent edema, and calcifications seen on radiographs [3, 5, 13–15, 18]. Trismus is one of the main complaints of patients followed by increase in volume, edema and pain. Increase in local temperature, difficulty in swallowing, complaints in ear area, asymmetry or facial deformity and increase of dental mobility are rare complaints [3–18]. In the case reported, the patient showed signs a few days after surgery, which were persistent trismus, persistent edema, and calcification present on CT exam. During this period, some of the signs and symptoms of the disease, such as edema, pain, or even trismus, were mistaken with those of the postoperative period after fracture reduction and fixation, making diagnosis difficult. In this case, the patient didn't present trismus previous to the trauma, but started to present right after the accident, probably due to the dislocation of the zygomatic complex. The persistent trismus even after the reduction of the fracture and the post-treatment physiotherapy and the presence of calcification observed on the CT exam allowed the hypothesis of TMO. The calcification observed on CT could indicate also a pre-existing coronoid hyperplasia, but in this case, once the trismus is present after the treatment/post-surgery period the trismus would need to be present previous to trauma, or it would not be present previous to trauma after the treatment/post-surgery period.

Factors related to direct traumatic impact such as trauma and fractures, violence, traffic and work-related accidents, gunshots and falls are the main causes of TMO. Dental extractions, local infiltration of anesthetics, previous surgeries, abscesses and infections, poorly performed orthodontic treatment, occlusal trauma, and orthopedic treatment after cervical trauma are other causes of TMO [4].

Probably the calcification of the temporalis muscle was not present before the trauma (traffic accident or surgery) because there were no symptoms indicating its presence, but we cannot affirm this because the calcification couldn't be identified in the pre- and post-surgery X-ray exams because of the superimposition of structures. The CT exams aren't solicited by routine for every type of fracture because of their high cost and exposure to a high dose of radiation. The first CT was performed after 30 days when we identified a mouth opening limitation and only at this time the calcification of the temporalis muscle was identified (Fig. 1).

It is believed that TMO results from intramuscular hemorrhage with vascular proliferation and exuberant granulation tissue that undergoes subsequent metaplasia to cartilage or bone, but the details are unknown [3, 8, 13, 15, 16]. After muscle necrosis, macrophages invade the injured site and release osteogenic growth factors [3].

Some authors attempted to induce TMO experimentally in the laboratory by intramuscular injection of alkaline phosphatase, but with no results, and they concluded that the elevated alkaline phosphatase is a result of the evolution of TMO and not its cause [4, 5]. Intramuscular injection of blood also failed [4, 5]. The transplantation of decalcified bone fragments into normal connective tissue succeeded in producing ectopic osteogenesis, but when the intercellular substance of the non-decalcified bones is transplanted, the induction effect rarely occurs [19]. The autolysis of scattered bone fragments releases BMPs (bone morphogenetic proteins) and induces differentiation of perivascular mesenchymal cells in the muscle tissue [11].

The accepted treatment is complete excision and osteotomy involving the region of muscle insertion [3, 4, 7, 13]. The timing of surgical intervention may vary, depending on the risks and benefits of each of these options [15]. Early surgical procedure apparently seems to lead to a high rate of recurrence [11]. Thorndike [20] recommends that surgical removal should not be performed until the bone is mature and bone activity has decreased (detectable by bone scintigraphy), usually between 6 and 12 months. It is believed that the recurrence of lesions will occur in 35 % of the cases [14]. However, Conner and Duffy [5] advocate earlier intervention when the TMO is located in the head and neck due to components such as pain and trismus and they add that other authors do not advocate surgery if these components are not present, for up to 35 % of cases of TMO may spontaneously regress after several months.

Complete excision is the most common treatment for TMO and it is frequently associated with postoperative physiotherapy. Complementary treatments are rare, but they involve partial biopsy, local dissection, and others such as drugs, local injection of steroids, insertion of grafts and biomaterials and in more aggressive cases, cervical resection. In the case reported the patient presented pain and trismus with significant functional impairment, so early intervention (30 days) was instituted, followed by physiotherapy.

After surgical treatment, compression of the surgical area is recommended to prevent recurrence, followed by intense physiotherapy [7, 11, 13]. Acetic acid iontophoresis, injection therapy with magnesium sulfate and magnesium lactate and Etidronate disodium administered orally (EHDP [Didoronel; Procter & Gamble, Cincinnati, OH, USA] a bisphosphonate used for prophylaxis and treatment of Paget's Disease), low doses of radiation to inhibit osteogenic action, anti-inflammatory drugs, corticosteroids, indomethacin, warfarin, and retinoids can be used as adjuvant treatment [5, 11, 14]. Up to the present, there is no consensus with regard to non-surgical treatment and there is a lack of clinical and scientific evidence for the above-mentioned treatments [5].

Most cases of recurrence recorded in the literature occurred within a 6-month period after surgery. However, the high number of cases without recurrence may be concealed due to the short postoperative follow-up period of the patients (usually less than 12 months). In the case described, significant recurrence of the bone mass occurred within 12 months, but signs and symptoms such as pain and trismus have not been present during the 30 months of follow-up. Since there are no signs and symptoms on the follow-up period, we have to consider that the calcification observed on the 1 year post-surgery CT could also be bony remnants of the coronoid and not a true recurrence of TMO.

TMO is an uncommon lesion, but it must be considered as differential diagnosis when the patient has trismus and persistent edema associated with calcifications in the soft tissues present in the imaging exam. The most accepted treatment for cases of TMO is complete excision followed by physiotherapy. TMO is a lesion that may frequently recur and long-term follow-up must be provided.

Compliance with Ethical Standards

Conflict of interest None declared.

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