

CASE REPORT

Non-traumatic myositis ossificans of the Right masseter muscle: a case report and review of the literature

Reza Sharifi¹ | Lotfollah Kamali Hakim² | Saeed Hasani Mehraban² | Amirali Asadi² 

¹Department of Oral and Maxillofacial Surgery, Cranio maxillofacial research center, School of Dentistry, Tehran University of Medical Sciences, Tehran, Iran

²Resident of Oral and Maxillofacial Surgery, School of Dentistry, Tehran University of Medical Sciences, Tehran, Iran

Correspondence

Amirali Asadi, Resident of Oral and Maxillofacial Surgery, School of Dentistry, Tehran University of Medical Sciences, Tehran, Iran.

Email: Amirali_a_2003@yahoo.com

Abstract

Myositis ossificans (MO) is a rare disease in which ossification develops in the muscle or soft tissue. MO traumatica is recognized by ossification of the soft tissues after acute or repetitive trauma, burns, or surgical procedures. It usually involves the extremity muscles. In the head and neck region, the masseter is most commonly involved in MO traumatica. Surgical resection of the ossified tissue has been the most commonly used treatment for this disorder, with a high postoperative recurrence rate. We report a case of non-traumatic MO of the Masseter muscle with different size round calcifications and to review the literature reported data about MO traumatica involving the masseter muscle.

KEY WORDS

masticatory, muscle, Myositis, Ossificans

1 | INTRODUCTION

Myositis ossificans (MO) is a rare disease in which ossification develops in the muscle or soft tissue. MO is divided broadly into myositis ossificans progressiva (MOP) and myositis ossificans traumatica (MOT). MOP is an autosomal dominant disease in which multiple, heterotopic ossifications develop in the systemic muscles, fascias, tendons, and ligaments, sometimes within families. MOT, also called traumatic myositis ossificans, myositis ossificans circumscripta, localized myositis ossificans, or fibrodysplasia ossificans circumscripta, is a disease in which muscles are ossified after trauma or inflammation.¹

Unlike MOP, MOT is often remitted through surgical treatment, including excision of the ossification, some patients have repeated relapses and are refractory to treatment.²

In the head and neck region, the masseter is most commonly involved in MOT because it is on the lateral side of the mandible and is most likely to receive external forces directly.³

The aim of this article is to present and discuss a case of non-traumatic MO of the Masseter muscle with different size round calcifications.

2 | PRESENTATION OF CASE

A 24-year-old male was admitted to our outpatient clinic in Shariati Hospital Tehran, Iran. He complained of swelling in right masseter region from 1.5 year ago. During physical examination painful and movable separate masses were evident in the right ramus of mandible, the patient also mentioned pain on chewing and mouth opening. He had no limitation on mouth opening and the skin over the right cheek region and the intraoral soft tissue were normal. The right parotid duct exhibited normal salivary flow. He denied any blunt trauma to the area and his medical history and systemic review were unremarkable. A core needle biopsy was taken from the patient 8 months before and that report was "Fibromuscular tissue with a few blood vessels." There was no history of

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familial involvement and the patient denied any anesthesia or dysesthesia.

MRI was taken from the region and a mass with 55*32 mm diameter in right masseter muscle with scattered signal void foci related to calcifications with few enhancing after Gd injection. The lesion was high on T2 signal and iso on T1(Figure 1A-D).

The CT revealed a 65*29*51 mm soft tissue mass which was located in right masseter muscle with foci of round calcifications: Radiologist suggested hemangioma as the first differential diagnosis for the lesion (Figure 2A-D), CT angiography was done but no vascular lesions were found.

As we noticed calcification in masticatory muscles, the laboratory tests including calcium, alkaline phosphatase, phosphorus, and Vitamin D were ordered and they were all within normal ranges.

Eventually, the patient's history, and the clinical and radiographic findings, allowed us to diagnose myositis ossificans involving the right masseter muscle.

Intraoperatively, under local anesthesia, the right masseter muscle was subjected to blunt dissection and eleven calcified masses were identified and excised. Approximately, masses measured 3 mm to 1 cm in diameter (Figure 3A,B).

Histopathologic examination revealed relatively mature calcifications formed from irregular bone trabeculae, surrounded by masseter muscle fibers. (Figure 4A,B).

Post-operative radiographic images showed some remained round calcification after surgery located in regions with difficult intraoral access. (Figure 5A,B).

Postoperative course was uneventful and no abnormality of masticatory or sensory function was noted. After surgery, the patient had some pain in the right cheek region, with restriction of jaw movement. Therefore, A period of physiotherapy was carried out and the patient was instructed for active and passive mouth opening exercises.

After 6 months follow-up, the patient returned to our clinic and he was satisfied with resolution of swelling and increased MMO (Figure 6). The patient is now under supervision for further progression of the lesion and if condition worsens, revision surgery will be performed.

3 | DISCUSSION

Myositis ossificans is a heterotopic ossification of muscular tissue which is benign and is characterized by well differentiated bone formation.⁴

Trauma is the most frequent etiological factor seen in almost 60–75% of the cases.⁵ It is most frequently encountered in the arm, shoulder, thigh, and hand, in order of frequency.⁶ Fortunately, MOT is rare in the maxillofacial region. The masseter muscle is commonly involved because of its position on the facial skeleton, which makes it more prone to

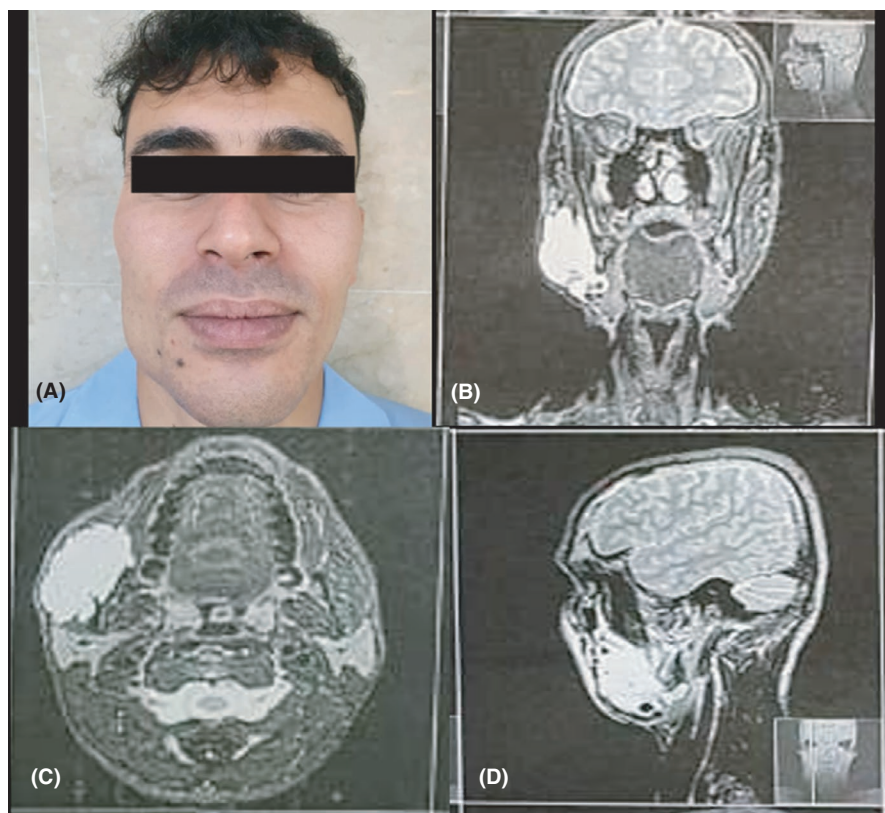


FIGURE 1 A-D, Preoperative MRI revealed a mass in right masseter muscle with round calcifications

FIGURE 2 A-D, Preoperative CT scan revealed round calcifications at right masseter region

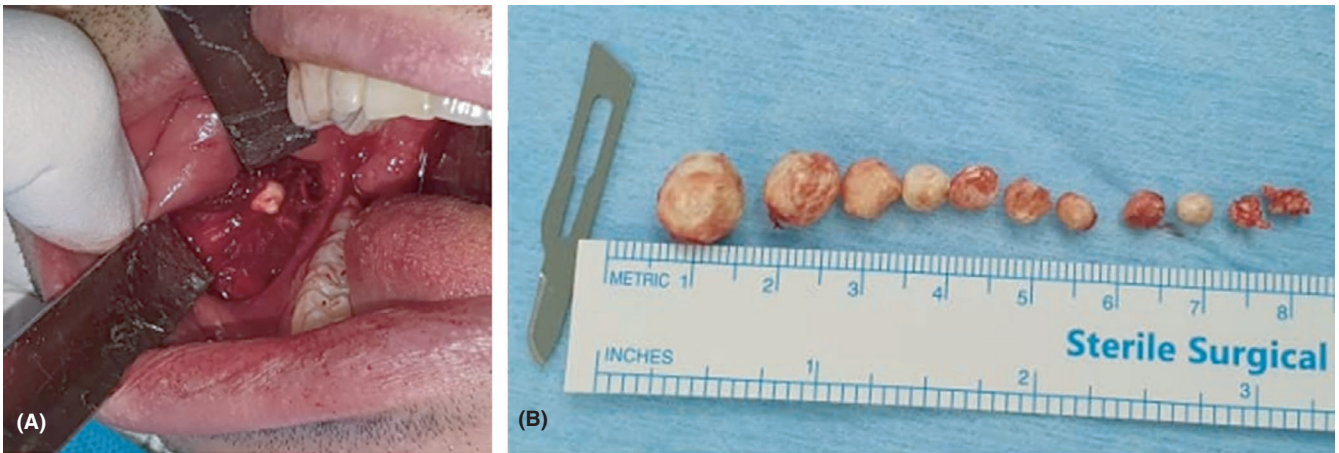
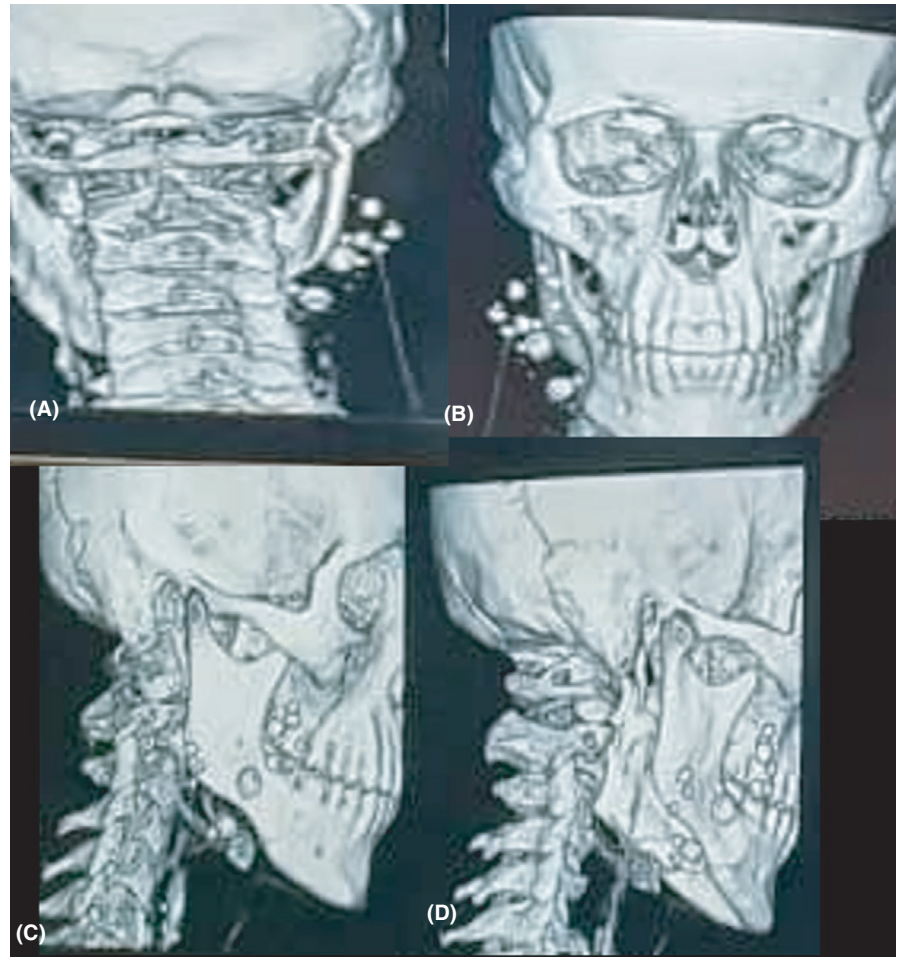


FIGURE 3 The lesion surgically excised which shows fragments of round different size calcifications

traumatic injuries. Incidence of the pterygoid and temporalis muscle involvement is rarely reported in literature.³

MO may develop at any age, but is most often seen in adolescents and young adults.⁷

Theories have been discussed about etiology, such as myositis ossificans being a step in an organizing hematoma's development. It has also been suggested that

osteoblasts escape from periosteum and migrate to soft tissue and create this condition. Another theory is mechanical trauma that can cause osteoblasts to be pushed into muscle and therefore result in ectopic calcification in a muscle.⁸ Burns, infections, and drug abuse are other rare factors which may cause MO.⁹ Nontraumatic MO is very rare in the literature.¹⁰ Repetitive microtrauma, tissue

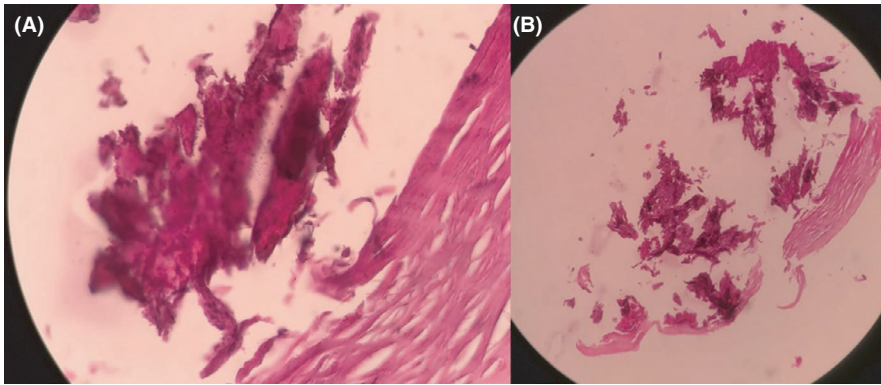


FIGURE 4 Histopathologic examination revealed relatively mature calcifications formed from irregular bone trabeculae, surrounded by masseter muscle fibers

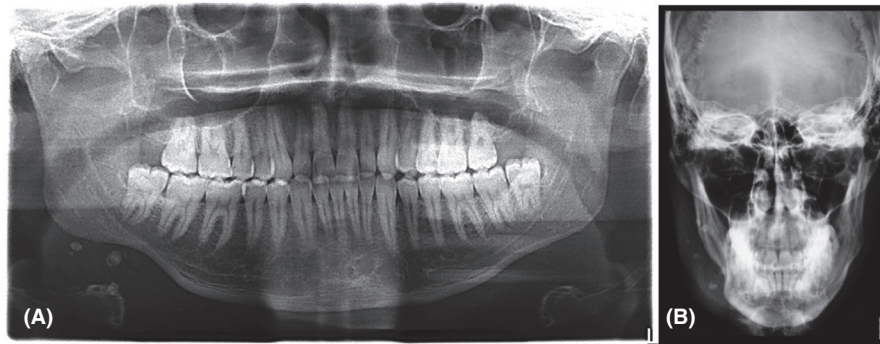


FIGURE 5 Post-operative radiographic images showed some remained round calcification after surgery located in regions with difficult intraoral access



FIGURE 6 Patient's mouth opening at 6 months follow-up

ischemia, and inflammation were addressed as the causative mechanisms of the non-traumatic MO.¹¹ The most widely accepted theory states that trauma to skeletal muscles induces the expression of bone morphogenic protein at the site of injury, which in turn stimulates the primitive stem cells to differentiate into osteoblasts, resulting in heterotopic ossification.¹² Table 1 reviews 6 cases of myositis

ossificans in masseter muscle in the literature previously published.^{2,13-18}

Hanisch et al.¹⁹ in 2018 reviewed 63 cases of the masticatory muscles Myositis ossificans, they found that wisdom-tooth infection, dental extraction, mandibular nerve block, and periodontitis therapy were reported as MOT cases. Medial pterygoid muscle was the most affected muscle. Females were more affected than men. They also reported most reported clinical symptom of MOT was trismus, followed by swelling and pain. These findings are not similar to our case which the patient denied any history of trauma or dental treatments. In contrast, clinical symptoms such as pain and swelling were expressed by our patient.

Giovanini et al.²² in 2019 reported a Traumatic myositis chondro-ossificans of masseter muscle after direct masseter muscle injury and correlate it to bone and cartilage biomarkers upregulation. Nine days after trauma the patient had severe mouth opening limitation. After CBCT and ultrasonography study, there was incomplete fracture between the posterior board of inferior jaw and coronoid process as well as calcification within masseter muscle. The treatment was excisional biopsy of calcification, coronoid process removal to enhance mouth opening as well as incomplete condyle fracture monitoring. After histological analysis, immunohistochemistry was conducted and it was found that chondro-ossification biomarkers such as TGF- β 1, Indian hedgehog (IHH), BMP2, osteopontin (OP), and osteocalcin (OC) were upregulated.

TABLE 1 Previous cases of myositis ossificans in masseter muscle reported

| Author & Year | Patient Gender & Age | Muscles involved | History of trauma or infection | Chief complaint | Intervention |
|----------------------|----------------------|----------------------------|---|---|---|
| Kayal et al. 2018 | Male-36 | Left masseter muscle | History of falling | Restricted mouth opening - swelling | Excision |
| Yang et al. 2021 | Male-21 | Bilateral Masseter muscles | After orthognathic surgery | limitation of mouth opening | Excision of the ectopic bone |
| Kruse et al. 2009 | Female-35 | Bilateral Masseter muscles | chemoradiotherapy and critical illness neuropathy | progressive decrease in the range of motion | No treatment |
| Demirkol et al. 2015 | Female-64 | Left Masseter muscle | Blunt trauma to the left cheek | Pain on chewing and mouth opening | Surgical excision of the calcified masses |
| Plombino et al. 2018 | Female-62 | Right masseter muscle | Colon resection | reduction of the mouth opening | Surgical excision by trans-oral approach |
| Trepat et al. 2016 | Female-49 | left masseter muscle | repetitive infection of her left wisdom tooth | pain, swelling, difficulty in mouth opening | Surgical removal of the tumor |

Joshi et al.²³ in 2018 described a case report of Myositis ossificans in left masseter muscle. The patient's chief complaint was restricted mouth opening for past 3 months. The patient had a history of trauma which resulted in swelling on the left side of his face. The swelling gradually hardened over a period. The swelling was palpable and when the patient clenched his teeth and he had restricted mouth opening to 2mm. CT scan revealed evidence of hypertrophy of left masseter muscle and intramuscular mass with a rim of ossifications. The mass was excised and the histopathologic examination revealed mature lamellar bone correlating with Myositis Ossificans.

Differential diagnosis is important in these lesions, First of all ossifying fibromas are important, because their clinical behavior is similar, they arise in the third and fourth decade, mainly in the posterior of mandible, they grow slowly, are encapsulated, and they can cause expansion and displacement of adjacent structures.²⁰ Another differential diagnosis to explain is sialolithiasis, as in this case, the patient had round calcifications in parotid area also mimicking salivary gland stones. Both of the lesions can appear as round radiopaque masses on panoramic radiographs and they can appear superimposed on the mandible, but sialolithiasis exhibits a higher incidence in males aged between 30 and 60 years.²¹

As mentioned in the table, female is more frequently involved than males. In all of the cases, trauma to the masticatory system or infection is noticed. As our patient did not give any obvious history of trauma, we feel our case is unique of being nontraumatic and having no simultaneous predisposing factors, we can name it as idiopathic round calcification of masticatory muscles, which is rare.

4 | CONCLUSION

Myositis ossificans is a disease with the main feature of formation of heterotropic bone involving muscle or any other soft tissue. Its main clinical features in Oral and Maxillofacial region are trismus, pain, and swelling in some cases. It is more common in females. Surgical excision is the main treatment for MO and it is essential that clinicians visit these patients on regular intervals to monitor for signs of inflammation, decreased range of motion, or radiographic evidence of recurrent calcified mass formation.

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CONFLICTS OF INTEREST

The authors have no conflict of interest to declare.

AUTHOR CONTRIBUTIONS

Reza Sharifi carried out the surgery. Lotfollah Kamali Hakim reviewed and revised the article. Saeed Hasani Mehraban gathered the information. Amirali Asadi wrote the manuscript in consultation with other authors.

ETHICAL APPROVAL

Written informed consent was obtained from the patient for their anonymized information to be published in this article.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ORCID

Amirali Asadi  <https://orcid.org/0000-0003-0206-4676>

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