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

Reza Sharifi<sup>1</sup>, lotfollah kamali hakim<sup>1</sup>, Saeed Hasani Mehraban<sup>2</sup>, and Amirali Asadi<sup>1</sup>

February 26, 2021

## Abstract

Myositis ossification (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. In the head and neck region, masseter is most commonly involved muscle.

## Title page

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

- 1-Reza Sharifi : Assistant Professor, Department of Oral and Maxillofacial Surgery, Cranio maxillofacial research center. School of Dentistry, Tehran University of Medical Sciences, Tehran, Iran
- 2- Lotfollah Kamali Hakim: Resident of Oral and Maxillofacial Surgery, School of Dentistry, Tehran University of Medical Sciences, Tehran, Iran.
- 3-Saeed Hasani Mehraban: Resident of Oral and Maxillofacial Surgery, School of Dentistry, Tehran University of Medical Sciences, Tehran, Iran.
- 4-Amirali Asadi: Resident of Oral and Maxillofacial Surgery, School of Dentistry, Tehran University of Medical Sciences, Tehran, Iran. \*

\*Corresponding author: Amirali Asadi – Email: Amirali a 2003@yahoo.com

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

Myositis - Ossificans - masticatory - muscle -

<sup>&</sup>lt;sup>1</sup>Affiliation not available

<sup>&</sup>lt;sup>2</sup>Tehran University of Medical Sciences

## Key Clinical Message:

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. Surgical excision is the main treatment.

#### 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 familes. 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 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 1 A-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 the first Differential Diagnosis for the lesion (Figure 2 A\_D) CT Angiography was done but no Vascular lesions was 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 3 A, B).

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

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

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.

Author Contribution:

Majid Beshkar: Carried out the surgery

Lotfollah Kamali Hakim: Reviewed and revised the article Saeed Hasani Mehraban: Reviewed and revised the article

Amirali Asadi: Wrote the manuscript in consultation with other authors

Conflicts of interest:

The authors have no conflict of interest to declare.

#### References:

- 1. Aoki T, Naito H, Ota Y, Shiiki K. Myositis ossificans traumatica of the masticatory muscles: review of the literature and report of a case. Journal of oral and maxillofacial surgery: official journal of the American Association of Oral and Maxillofacial Surgeons. 2002;60(9):1083-8.
- 2. Boffano P, Zavattero E, Bosco G, Berrone S. Myositis ossificans of the left medial pterygoid muscle: case report and review of the literature of myositis ossificans of masticatory muscles. Craniomaxillofacial trauma & reconstruction. 2014;7(1):43-50.
- 3. Jayade B, Adirajaiah S, Vadera H, Kundalaswamy G, Sattur AP, Kalkur C. Myositis ossificans in medial, lateral pterygoid, and contralateral temporalis muscles: a rare case report. Oral surgery, oral medicine, oral pathology and oral radiology. 2013;116(4):e261-6.
- 4. Folpe AL, Inwards CY. Bone and Soft Tissue Pathology E-Book: A Volume in the Foundations in Diagnostic Pathology Series: Elsevier Health Sciences; 2009.
- 5. Nishio J, Nabeshima K, Iwasaki H, Naito M. Non-traumatic myositis ossificans mimicking a malignant neoplasm in an 83-year-old woman: a case report. Journal of medical case reports. 2010;4:270.
- 6. Aneiros-Fernandez J, Caba-Molina M, Arias-Santiago S, Ovalle F, Hernandez-Cortes P, Aneiros-Cachaza J. Myositis ossificans circumscripta without history of trauma. Journal of clinical medicine research. 2010;2(3):142-4.
- 7. Onen MR, Varol E, Tosun Mİ, Naderi S. Nontraumatic Myositis Ossificans as an Uncommon Cause of Scoliosis: Case Report and Review of the Literature. World Neurosurgery. 2019;123:208-11.
- 8. Kim SW, Choi JH. Myositis ossificans in psoas muscle after lumbar spine fracture. Spine. 2009;34(10):E367-70.
- 9. Merchant R, Sainani NI, Lawande MA, Pungavkar SA, Patkar DP, Walawalkar A. Pre- and post-therapy MR imaging in fibrodysplasia ossificans progressiva. Pediatric radiology. 2006;36(10):1108-11.
- 10. Zoccali C, Chichierchia G, Covello R. An unusual case of lumbar paravertebral miositis ossificans mimicking muscular skeletal tumor. Musculoskeletal surgery. 2013;97(3):251-3.
- 11. Mann SS, Som PM, Gumprecht JP. The difficulties of diagnosing myositis ossificans circumscripta in the paraspinal muscles of a human immunodeficiency virus-positive man: magnetic resonance imaging and temporal computed tomographic findings. Archives of otolaryngology—head & neck surgery. 2000;126(6):785-8.
- 12. Wiggins RL, Thurber D, Abramovitch K, Bouquot J, Vigneswaran N. Myositis ossificans circumscripta of the buccinator muscle: first report of a rare complication of mandibular third molar extraction. Journal of oral and maxillofacial surgery: official journal of the American Association of Oral and Maxillofacial Surgeons. 2008;66(9):1959-63.
- 13. Karaali S, Emekli U. Myositis Ossificans Traumatica of the Medial Pterygoid Muscle After Third Molar Tooth Extraction: A Case Report and Review of Literature. Journal of oral and maxillofacial surgery: official journal of the American Association of Oral and Maxillofacial Surgeons. 2018;76(11):2284.e1-.e5.
- 14. Jiang Q, Chen MJ, Yang C, Qiu YT, Tian Z, Zhang ZY, et al. Post-infectious myositis ossificans in medial, lateral pterygoid muscles: A case report and review of the literature. Oncology letters. 2015;9(2):920-6.
- 15. Torres AM, Nardis AC, da Silva RA, Savioli C. Myositis ossificans traumatica of the medial pterygoid muscle following a third molar extraction. International journal of oral and maxillofacial surgery. 2015;44(4):488-90.

- 16. Ramieri V, Bianca C, Arangio P, Cascone P. Myositis ossificans of the medial pterygoid muscle. The Journal of craniofacial surgery. 2010;21(4):1202-4.
- 17. Thangavelu A, Vaidhyanathan A, Narendar R. Myositis ossificans traumatica of the medial pterygoid. International journal of oral and maxillofacial surgery. 2011;40(5):545-9.
- 18. Trautmann F, Moura P, Fernandes TL, Gondak RO, Castilho JC, Filho EM. Myositis ossificans traumatica of the medial pterygoid muscle: a case report. Journal of oral science. 2010;52(3):485-9.

Figure legends:

Figure-1(A D): Preoperative MRI revealed a mass in right masseter muscle with round calcifications.

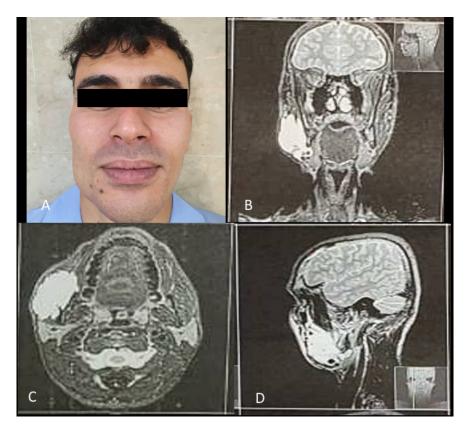
Figure-2 (A D): Preoperative CT scan revealed round calcifications at right masseter region.

Figure-3: The lesion surgically excised which shows Fragments of round different size calcifications.

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

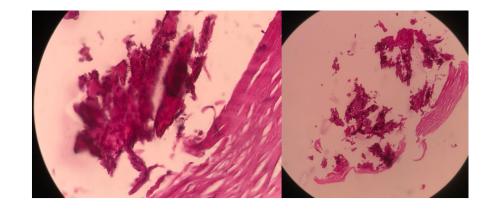
Figure-5: Post-operative radiographic images showed some remained round calcification after surgery located in regions with difficult intraoral access

Figure-6: patient's mouth opening at 6 months follow up













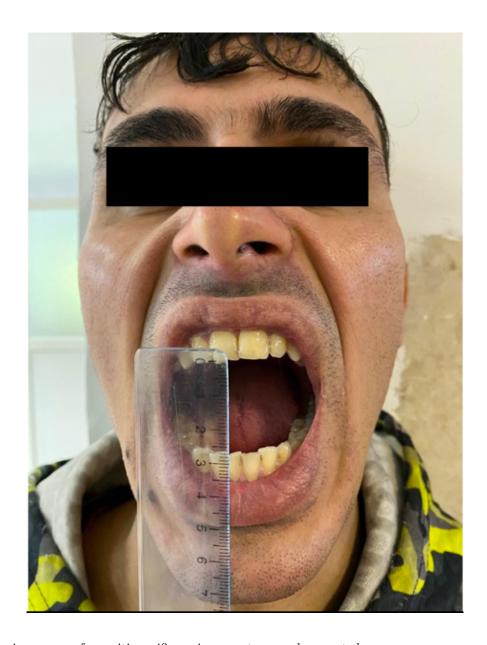


Table 1: Previous cases of myositis ossificans in masseter muscle reported.

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

Author& Year	Patient Gender& Age	Muscles involved	History of trauma or infection	Chief complaint	Intervention
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
Piombino 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

