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**Authors** Jović Sasa1, Brajkovic Denis2, Borilovic Milena2, Marjanovic Uros2, Brkic Marko2, Kozomara Ruzica1,3, Stosic Srboljub1,3, Vojnosanitetski pregled (2019); Online First April, 2019.

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RECURRING MYOSITIS OSSIFICANS TRAUMATICA OF TEMPORAL MUSCLE: A CASE REPORT

Jović Sasa1, Brajkovic Denis2, Borilovic Milena2, Marjanovic Uros2, Brkic Marko2, Kozomara Ruzica1,3, Stosic Srboljub1,3

1Clinic for Maxillofacial surgery, Military Medical Academy, Belgrade, Serbia
2Resident at Clinic for Maxillofacial surgery, Military Medical Academy, Belgrade, Serbia
3Medical Faculty of the Military Medical Academy, University of Defence, Belgrade, Serbia

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Corresponding author:

Brajkovic Denis, DDS, PhD
Resident at the Clinic for Maxillofacial surgery, Military Medical Academy, Belgrade
Crnotravska 17, 11000 Belgrade, Serbia

E-mail: denis.brajkovic@gmail.com
Abstract

**Background.** Myositis ossificans traumatica (MOT) refers to a benign, localized ectopic bone formation within skeletal muscle bundles related to traumatic injury. MOT rarely affects masticatory muscles and it represents a major diagnostic and therapeutic problem for clinicians. Currently, the treatment of choice is complete excision of calcified mass after bone maturation and resection of affected bone.

**Case report.** A 47-year-old male presented with a month long severe restriction of mouth opening, following extraction of the right lower third molar tooth under local anesthesia. A CT scan revealed ectopic bone formation in the right temporal muscle extending to the right coronoid process. Surgical excision of the calcified mass was performed. Six years following the surgery, patient reported same symptoms. CT scan revealed a calcified mass of the right temporal muscle extending to the medial pterygoid muscle. The patient was reoperated and postoperative physical treatment is in progress.

**Discussion** MOT represent a major diagnostic and therapeutic challenge for surgeons. Recurrences after surgical treatment are often found. Further research is needed to clarify the mechanisms of ossification in MOT in order to develop conservative treatment approaches.

**Key words:** myositis ossificans, trauma, masticatory muscles, temporal, treatment

Apstrakt

**Uvod.** Traumatski osificirajući miozitis se odnosi na benigno, lokalizovano ektopično formiranje koštanog tkiva unutar skeletnih mišića nakon trauma mišića. Traumatski osificirajući miozitis retko zahvata mastikatorne mišiće i tada predstavlja ozbiljan dijagnostički i terapeutski problem za hirurge. Trenutno je tretman izbora potpuna ekscizija kalcificifikovane mase i resekcija zahvaćene kosti.

**Prikaz slučaja.** Pacijent se javio na pregled maksilofacijalnog hirurga zbog otežanog otvaranj usta unazad mesec dana, nakon ekstrakcije desnog donjeg umnjaka pod lokalnom anestezijom. Kompjuterizovana tomografija glave pokazala je stvaranje ektopične kosti u
Myositis ossificans is a rare disease in which ectopic benign bone formation occurs in muscle tissue. It has been divided into myositis ossificans progressiva (MOP) and myositis ossificans traumatica (MOT). MOP is an autosomal dominant disease characterized by systemic ossification of muscles and soft tissues with poor prognosis. MOT refers to a benign, localized ectopic bone formation and ossification of fibrous connective tissue within skeletal muscle bundles related to traumatic injury. The pathological mechanism of MOT is not completely understood, and proposed pathogenesis theories suggest inflammatory response in muscle tissue related to trauma followed by displacement of osteoprogenitor cells and overexpression of bone morphogenetic proteins, leading to ectopic bone formation.

MOT is most commonly seen in young patients due to the bone metabolism, renewing periosteum and richness in mesenchymal cells. MOT rarely affects masticatory muscles and so far a limited number of reports have been published. MOT of masticatory muscles represents a major clinical problem since there is no unified algorithm for diagnosis and treatment. The main clinical sign of MOT of masticatory muscles is progressive trismus following trauma to the head and neck. The only treatment modality widely accepted is complete excision of calcified mass after bone maturation and resection of affected bone.

In this case report we presented a case of reoccurring MOT affecting right temporal muscle after extraction of right lower third molar under local anesthesia.
Case report

A 47-year-old male presented to the Department for Maxillofacial surgery in 2011, with a month long complaint of restriction of jaw motion. The patient previous medical history was uneventful. Patient reported that he underwent extraction of the right lower third molar due to the pericoronal infection, under local mandibular anesthesia in private dental clinic approximately 2 months before. There was no history of other trauma to the head and neck.

Head and neck examinations revealed severe trismus, with a maximum incisal opening of 2 mm.

There was no tenderness of the right masseter and temporalis muscles. Intraoral examination was incomplete due to the trismus. Computed tomography (CT) scan of the facial bones depicted radiopaque entity attached to and extending superiorly to the right coronoid process and the insertion of the right temporalis muscle. The lesion appeared as a central radiolucency surrounded by circumscribed ossified periphery. Calcification in the right temporal muscle was approximately 43x15 mm in size in when measured from coronoid processus to the upper part of the lesion, extended to the temporal fossa and well-defined from the surrounding structures. There were no signs of bone destruction and infiltration of other masticatory muscles (Figure 1.).

Figure 1. Preoperative computed tomography (CT) demonstrating heterotopic calcification in the right temporal muscle (red arrows). A) Sagittal view; B) Axial view; C) 3D reconstruction
At this point the differential diagnosis of extra skeletal bone formation included myositis ossificans, fibrodysplasia ossificans, osteochondroma, chondrosarcoma, osteosarcoma, osteoma, and vascular malformation with phleboliths.

We decided to perform a right mandibular coronoidectomy and extirpation of osseous tissue. By intraoral approach and elevation of mucoperiosteal flap, after striping the temporalis attachment from the coronoid process, a calcified mass extending along the temporalis muscle from the coronoid process, 4x3 cm in size, was visualized. Due to the size and localization of the lesion, extraoral temporal approach was performed to gain access to the mass in right temporal fossa which was excised along with the coronoid processus. The resected coronoid process and calcified mass were normal-appearing bone with no evidence of surrounding bony or soft tissue destruction or infiltration. After resection, there was a 2 cm gap between the mandibular ramus and the temporalis muscle. Immediate intraoperative maximal incisal opening, measured from the maxillary to the mandibular incisal edges was 40 mm, compared to 2 mm preoperatively.

Histological finding revealed zonal pattern of the lesion. The innermost zone consisted of immature vascularized fibroblastic zone with mild degree of pleomorphism, sparse inflammatory cells and rare multinucleated giant cells. Intermediate zone consisted of irregular bone trabecula and a peripheral zone revealed ossification and mature lamellar bone (Figure 2). The histological finding was consistent with myositis ossificans.

Figure 2. Photomicrograph of a excised extraosseous bony lesion. A) Section of specimen showing central loose connective tissue with immature bone containing osteocytes and mature bone at the periphery (H&E stain, ×100); B) Photomicrograph showing central connective tissue zone surrounded by immature bone containing osteocytes in the lacunae (H&E stain, ×200); C) PAS staining
Postoperatively, the patient was able to open mouth passively up to 25 mm without assistance.

Two years after the right mandibular coronoidectomy, partial resection of right temporal muscle tendon and extirpation of osseous tissue from temporal fossa, the patient presented to our clinic for a follow-up evaluation. He reported difficulties in mouth opening in the morning, but no restriction or pain during mouth opening during the day. Extraoral and intraoral examination were uneventful. The patient’s maximum incisal opening was 30 mm without assistance or pain. However, CT scan revealed an osseous lesion in the right temporal fossa, in the vicinity, but not attached to the resected coronoid processus, extending to the temporal muscle. The radiological features of the lesion were similar to previous findings (Figure 3).

![Figure 3. Postoperative computed tomography (CT) 2 years after surgical therapy demonstrating recurrence of heterotopic calcification in the right temporal muscle (red arrows). Coronoid processus is resected. A) Coronal view; B) Sagittal view](image)

Because there were no functional problems, the patient was scheduled for a 6 months follow up. CT scan was performed again after two years (2015) revealing unchanged condition, however the ossification extended towards the mandibular attachment of the right medial pterygoid muscle (Figure 4).
Figure 4. Postoperative computed tomography (CT) 4 years after surgical therapy demonstrating recurrence of heterotopic calcification in the right temporal muscle (red arrows) extending towards the right medial pterygoid muscle (yellow arrow). Note demarcated ectopic bone formation in right temporal fossa. A) Axial view; B) Sagittal view; C) Coronal view

The patient was referred again to our clinic in April, 2017 because a two weeks long inability to open mouth. CT scan revealed enlargement of the ossified mass in the right temporal muscle extending from temporal fossa to muscular space affecting medial pterygoid muscle (Figure 5).
Figure 5. Postoperative computed tomography (CT) 6 years after surgical therapy demonstrating recurrence of heterotopic calcification in the right temporal muscle (arrow). Note fusion of calcification in temporal muscle to resected coronoid processus. A) Axial view; B) 3D reconstruction; C) Coronal view; C) Sagittal view; D) Preoperative photograph of the patient showing minimal mouth opening.

The decision on surgical treatment was made. An intraoral incision was made along the external oblique ridge of the mandible and a calcified mass, extending along the resected mandibular coronoid to the temporal muscle was visualized and partially extirpated. Because of poor visibility and intraoperative bleeding, an extraoral submandibular incision was performed. With preservation of major vascular and nerve structures, the right temporal fossa was approached and identified an osseous lesion in the temporal muscle extending towards the medial pterygoid muscle tendon. The osseous mass was extirpated.
Postoperatively, the maximal incisal opening was 40 mm. The tissue macroscopically resembled the previously extirpated tumor and the histological finding was similar (Figure 6).

Figure 6. Photomicrograph of a excised extraosseous bony lesion. A) Section of specimen showing central loose connective tissue with immature bone containing osteocytes and mature bone at the periphery (H&E stain, ×100); B) and C) Photomicrograph showing central connective tissue zone surrounded by immature bone containing osteocytes in the lacunae (H&E stain, ×200)

At the release from the department, the patient was able to open mouth passively up to 10 mm without assistance and he was instructed for physical treatment consisting of aggressive mandibular range-of-motion exercises. At the regular check-ups, the mouth opening was still reduced to 10mm, and the patient is currently performing proposed physical therapy. On the CT scan follow-up 6 month after the surgery the ectopic calcified mass affecting right temporal and medial pterygoid muscles was still present (Figure 7).
Figure 7. Postoperative computed tomography (CT) 3D reconstruction 6 months after second surgical treatment showing resected coronoid processus and ectopic calcified mass in temporal and medial pterygoid muscle

Discussion

Although MOT of masticatory muscles presents a benign reparative ectopic bone formation in muscle, accompanying trismus is a major functional problem to the patients. Initial trauma causes inflammatory response in the muscle and periosteum with subsequent displacement of bone fragments and osteoprogenitor cells into muscle bundles which induce ectopic bone formation. However in about 25% of cases the history of trauma is not found. Previous reports found that most commonly affected masticatory muscles were the masseter and medial pterygoid muscle, but in several reports more than one muscle was affected. Reports of affection of more than one muscle indicate that the inflammatory response following trauma is not localized on one individual muscle. Most MOT lesions were caused by direct trauma to the masseter muscle or trauma to the medial pterygoid muscle after local anesthetic injection. Temporal muscle is not commonly affected with MOT. In the present case MOT could not be linked to any apparent trauma to the head except the mandibular anesthesia and extraction of the third molar which occurred two months before the initial symptoms began. However, this type of surgical trauma would not cause the direct trauma to the temporal muscle. There are indices that chronic subclinical infection, which often accompanies third molars due to the pericoronitis, could lead to the inflammation and periosteal reaction and subsequently cause bone formation. In present case the trigger of ectopic ossification is still questionable. The most interesting clinical feature for discussion were difficulties in diagnosis and differential diagnosis. The diagnosis of MOT was based on clinical picture, radiological examination and histological findings. Bone formation was confined to the temporal muscle tendon and mandibular coronoid processus with no other areas of ectopic bone formation. Laboratory tests revealed no specific abnormalities of bone metabolism. However, even after mandibular coronoidectomy and resection of temporal muscle tendon the recurrence of the disease was observed after two years. Furthermore, seven years following the surgical treatment the patient had limited mouth opening because of the enlargement of ectopic bone and affection of the nearby medial pterygoid muscle by the ossification process. The following
surgical treatment included extirpation of osseous lesion in the temporal muscle and the medial pterygoid muscle tendon. The uneventful postoperative course and relapse of the disease could be linked to ongoing inflammatory process and activity of osteoprogenitor cells leading to continuous process of ectopic bone formation as proposed in other trials. MOT represents a significant diagnostic challenge for the clinicians. The main criteria for diagnosing MOT include a history of local injury, clinical and radiological evidence of ossification within two months following injury and localization of the ossification in the muscle tissue. Differential diagnosis for MOT include other benign bone-forming lesions: fibroosseous dysplasia progressive, calcified fibromatosis, phleboliths, osteoma, osteoblastoma as well as malignancies such as osteosarcoma, chondrosarcoma, rhabdomyosarcoma.

Radiographic appearance of MOT depends on the maturity of the lesion. Radiologically, Shirkoda et al. described 4 phases of MOT. Initial phase is characterized with inflammation and mesenchymal stem cell proliferation, without calcification. Initial bone formation is seen after 1-2 weeks after trauma. Intermediate phase with peripheral ossification is seen after 4 weeks. Mature phase is seen after 6 weeks and lesion appears as a central radiolucency surrounded by peripheral mature bone. During this phase, the lesion is well delineated from surrounding tissue and surgical treatment could be performed with minimal adverse events. CT scan is sensitive for identifying ossification. The radiological appearance of MOT is consistent with the zonal histological pattern of the lesion with well circumscribed ossified periphery and a low attenuating central portion. Early lesions appear as amorphous calcifications within soft tissue, while mature lesions are well separated from surrounding bone by a thin radiolucent area, however older lesions can appear attached to the adjacent bone.

Histologically, the hallmark of MOT is the zonal pattern. Central or cellular zone represents the innermost region of lesion, showing mitotic activity, undifferentiated cells, necrotic muscular tissue, giant multinucleated cells and loose fibrovascular tissue. The middle or intermediate zone contains active osteoblasts and immature osteoid. Peripheral or outer zone of the lesion shows mature bone with active osteoclasts and collagenous fibrous stroma. The microscopic and radiographic zone pattern is strongly suggestive of a reactive lesion and helps to rule out a diagnosis of sarcoma.
Standard treatment of MOT is surgical excision of the ossification along with osteotomy. In the mature phase the ectopic bone is well demarcated from surrounding tissue and is easiest to excise. Several authors proposed interposition of soft tissue graft between resected bone and muscle to prevent bone formation and prevent formation of hematoma. Although wide surgical excision of the lesion is performed, relapses are often reported. When several muscles are affected, surgical treatment may resolve with significant functional impairment. Several trials reported use of anti-inflammatory drugs, radiotherapy and drugs affecting bone metabolism as a mean to control postoperative inflammation and ossifying potential of the tissue. Other treatment modalities include physical therapy, acetic acid iontophoresis, magnesium therapy and bisphosphonate therapy. However these report are confined to single case studies, and development of new treatment methods is needed.

**Conclusion**

MOT represents a major diagnostic and therapeutic challenge for surgeons. It is fundamental that patients with unspecific clinical history leading to trismus are referred to specialized centers to diagnose. When MOT of masticatory muscles is suspected, CT scan could be the both, a diagnostic and a prognostic radiological tool. Surgical treatment remains the treatment of choice, and should involve excision of osseous lesion and osteotomy of muscle attachment region of the bone. In cases when several muscles are affected surgical treatment may resolve with a major disfunction of masticatory function. Thus, further research is needed to clarify the mechanisms of ossification in order to develop conservative treatment approaches.

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