CASE REPORT

Microscopic Changes In a Case of Bilateral Myositis Ossificans of Musculature of the Jaws - Relevance of Clinical & Microscopic Perspectives in the Disease Progression

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ABSTRACT: *Myositis Ossificans is a benign, solitary, self-limiting, ossifying soft-tissue mass typically occurring within skeletal muscle.Myositis Ossificans can occur almost anywhere in the body including the orofacial region. Despite the characteristic clinical, radiological and microscopic features the diagnosis of Myositis Ossificans still remains unclear. The most that we can do at the present time is to make sure that we recognize the signs of this malady and take advantage of every opportunity to study it. This article aims to bring forth together the more important clinical and pathological data concerning this disease, to analyze the histological spectrum seen in the Myositis Ossificans and to hypothesize its possible histogenesis through interpretation of the mineralized components to differentiate the early and late changes in Myositis Ossificans which were not clearly visualized under light microscopy and further analyzed by using polarizing microscopy.*

Keywords: Myositis ossificans, polarizing microscopy, histogenesis, muscle degeneration, ossification

Introduction

Myositis ossificans (MO) is an unusual benign disease which is infrequently seen in the maxillofacial region.[1] It is a benign heterotropic ossification within a muscle chiefly affecting active adolescents and young adults, with a slight male predominance.[2,3] Ackerman(1958) described MO as an "extra-osseous, localized, non-neoplastic formation of bone and cartilage". This is somewhat inaccurate because often the lesion may not even contain or be associated with bone. Moreover, the term "Myositis" is unsatisfactory as there is no good evidence of inflammation.[4,5]

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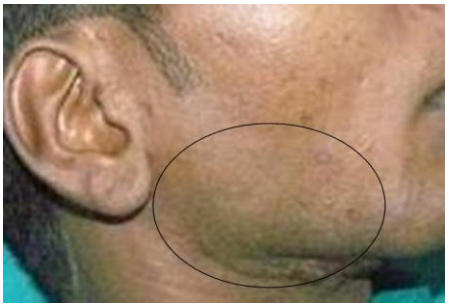
MO diseases of the muscle may be seen in either of two different forms: the myositis ossificans progressiva (MOP) and the myositis ossificans circumscripta, called by others traumatic myositis ossificans (TMO), localized myositis ossificans, or fibrodysplasia ossificans circumscripta.[6] Noble ((1924) classify the MO in to three types: Myositis ossificans progressiva, is believed to be a metabolic disorder occurring in children with widespread metamorphosis of muscle into bone with all of the skeletal muscles becoming involved progressively. It is ultimately fatal. Traumatic myositis ossificans circumscripta, follows a local trauma which may be either acute (a single injury), or chronic (repeated slight injuries or an occupational injury, such as strain of the adductor longus tendon in jockeys), and the last is Myositis Ossificans Circumscripta; without history of trauma.[5,6] This is usually found in paraplegia, chronic infections, burns and poliomyelitis, but may occur independently of these conditions.[2] 75% of cases of MO are associated with trauma.[4,5] The lesions are predominantly localized at high-risk sites of injury, such as the thigh, buttocks, elbow, and less often, the shoulder and calf. However, MO can occur

almost anywhere in the body, even in the sternocleidomastoid muscle, masseter muscle, and the muscle of the chest wall. Posttraumatic myositis results in a soft tissue mass which ossifies with a lacy pattern by 4-8 weeks.[2] Review of literature does not show many cases reported in the maxillofacial region bilaterally, including the masticatory muscles. But some cases are seen affecting the muscles of mastication with the highest incidence in the masseter unilaterally.[6]

Even with the characteristic clinical, radiological and microscopic features the diagnosis of MO still remains obscure.[2] The medical profession till date has not been able to offer the specific treatment modality to the patients of MO. The most that we can do at the present time is to make sure that we recognize the signs of this malady and take advantage of every opportunity to study it. This article aims to bring forth together the more important clinical and pathological data concerning this disease, analyze the histological spectrum seen in the MO and to hypothesize its possible histogenesis through interpretation of the mineralized components in MO with the use of polarizing microscopy.

Case report

A 22 year old male patient reported to the Out Patient Department, with the chief complaint of difficulty in mouth opening from past 6 years. The patient gave the history of a small extra-oral swelling 6 years back on the right side of the lower jaw with no associated pain. The swelling gradually increased in size which subsequently started discharging extra-orally by itself within a few days.(Figure 1).



Following this episode he noticed mild pain on the same side with gradual decrease in the mouth opening. The swelling gradually increased in size and was asymptomatic. Patient gave the history of tobacco chewing which was stopped as soon as the reduced mouth opening was observed. Patient also noticed restricted movements on left & right side but more extensively on right side.

Patient underwent 2 surgeries for the same at SafdarJung Hospital, New Delhi. In the first surgery, done in January 2007 bilateral exploration of the TMJ was done, to rule out fibrous ankylosis and to attempt condylectomy. TMJ was found normal. The problem persisted and hence the second surgery was attempted in December 2007; at the time of surgery ossification of the medial pterygoid muscle was identified. Mandible was osteotomized at the angle–ramus region; a pseudo-joint was created by interpostioning platysma into the osteotomized space. Patient was advised physiotherapy to increase the mouth opening. Subsequently after surgeries, he again noticed slight swelling on the left side which was also associated with slight pain.

On examination, the patient was moderately built and nourished with all the vital signs within normal limits. Extra-oral examination presented with facial asymmetry (Fig.1). Right lateral excursion was restricted to 2mm, left lateral function was also found restricted. There was deviation towards the right side on mouth opening. A hard palpable mass was felt with respect to the lower right masseteric region of face leading to a progressive diminution of the mouth opening. The neck examination was normal and no masses were noted. There was no tenderness or lymphadenopathy.

On radiographic examination, orthopantomograph (OPG) & the lateral view showed increased bone density leading to radio-opacity in the region of the right coronoid process as compared to the left side (Fig.2&3). CT scan examination revealed ossification of the right lateral pterygoid process (Fig 4,5,6).

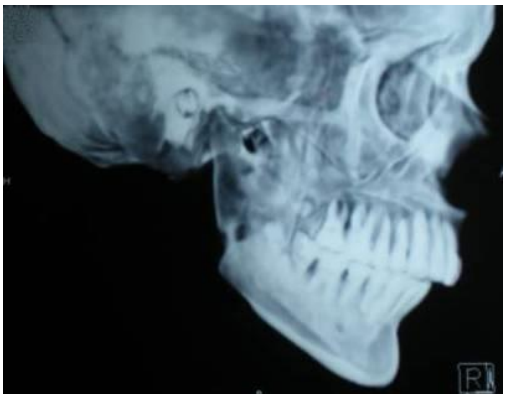
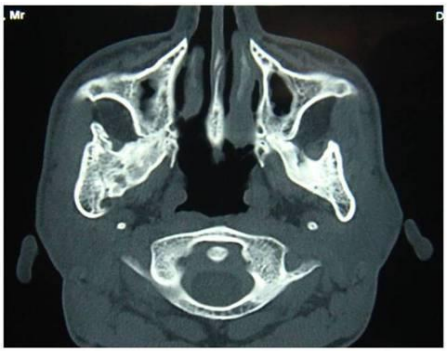
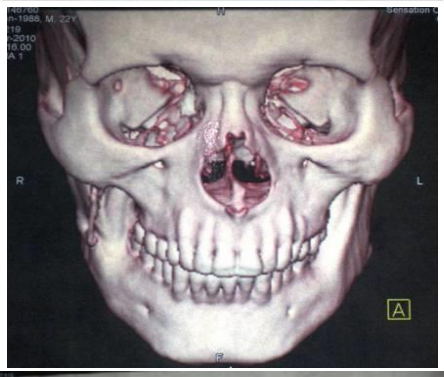


Figure-2 & 3 Lateral view: showing bilateral calcification involving muscles of mastication; more pronounced on the right side as compared to the left side.





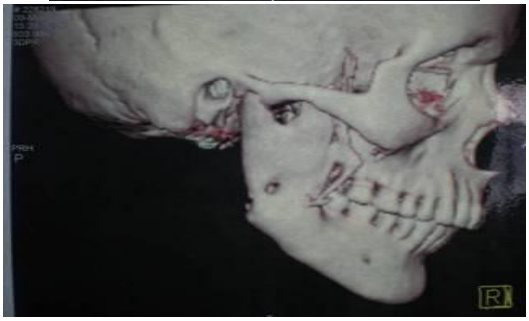
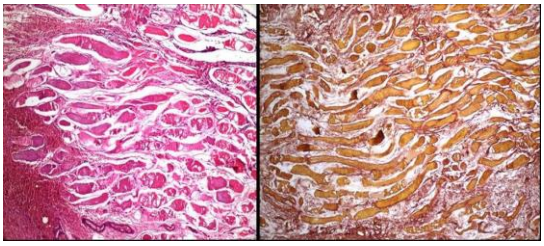


Figure 4,5,6 showing calcifications and osteoarthritic changes and ossifications in condylar region.

After the incisional biopsy multiple soft and hard tissue bits, from both left and right condylar areas were received. The lesion was grayish-white in color and the largest bit measuring 3.4×2.5 cm in dimension & was gritty in consistency. The hard tissue bits were underwent for decalcification and the following features were noted.

Microscopic examination showed numerous muscle fiber bundles with architectural changes. Evidence of degeneration with fragmentation of muscle fiber and separation into individual fibrils was seen. The muscle bundles were interspersed with areas of calcification. At few places, the muscle tissue was replaced by calcified masses. Many large engorged blood vessels along with extravasated R.B.Cs, areas of hemorrhage and adipose tissue were also noticed.(Fig 7&8)

Figure :7&8

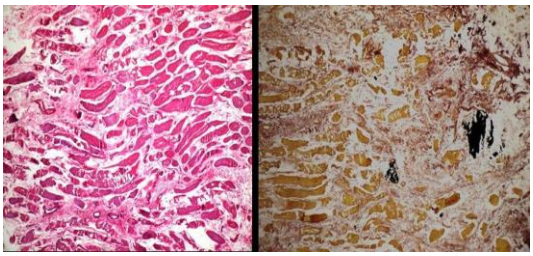


Photomicrograph showing Early changes-with numerous muscle bundle degeneration and architectural changes.

In the first surgery, the early changes in the MO were present in the form of muscle degeneration with slight calcifications. These early changes were followed by complete ossification of muscles which was observed during the second surgery.

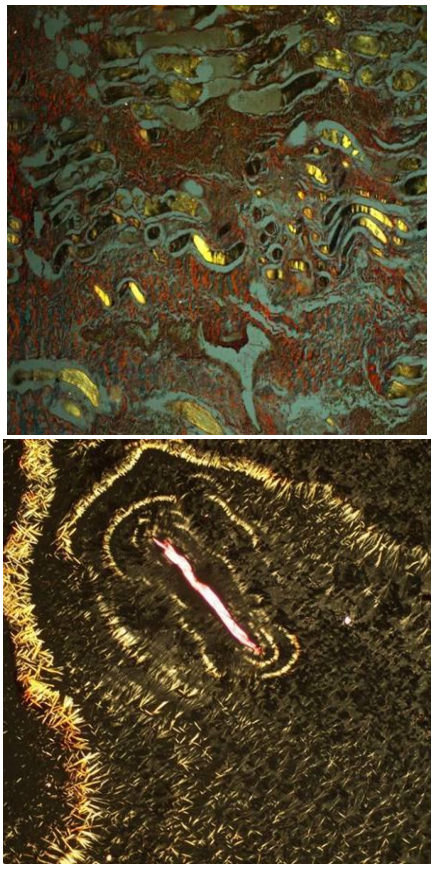
To further evaluate the pattern of muscle degeneration and ossification, including the early and late changes special stains like-Van Gieson and Vonkossa were used. Van Gieson to highlight the muscle degeneration (yellow) pattern. Vonkossa stain was used to demonstrate the calcified areas (black) (Fig.9& 10). The two staining procedures clearly defined the degeneration and the ossification.

Figure:9&10



Photomicrograph showing Late changes in the form of areas of calcification stained in van gieson. (black).

To further evaluate the mineralized masses in the late changes of MO, Van Gieson stained sections were subjected to polarizing microscopic analysis (Model BX41; Olympus System Microscope) and the ossification patterns of mature bone, with proper lamellar pattern of bone formation were analyzed. The early changes were in the form of muscle degeneration and slight calcification. (Fig 11,12)



Photomicrograph showing areas of muscle degeneration with calcification and areas of ossifications in lamellar pattern. (Polarizing Microscopy)

Discussion

MO was described initially by Thoma in 1958 as a condition generally caused by calcification and progressive ossification of an intramuscular hematoma after trauma. [6,7] MO is a benign, self-limiting and localized lesion characterized by ossification of fibrous connective tissue within and between skeletal muscle bundles after multiple traumatic episodes with muscle bleeding.[6]

MO usually occurs in early adult life, predominantly in healthy, young men.[2] According to Trautman et al, the pathogenesis of MO remains uncertain although many authors consider it as an aberrant physiological healing.[6] It is possible, but as yet unproved, that in cases presenting with an apparent history of traumatic injury, the process commences with muscle damage resulting in tissue necrosis or hemorrhage followed by exuberant reparative fibroblastic and vascular proliferation with eventual ossification.[2,7,11] Others consider that it is initiated by an excessive proliferation of fibroblasts while some speculate that the new bone arises by metaplasia of local fibrous tissue. Injury is undoubtedly an important factor but it is obviously not the only cause of condition. In a small number of cases, etiologies may include burns, infections or drug abuse.[2,8]

MO may have autosomal dominant inheritance with complete penetrance but variable expressivity, and most cases results from a sporadic mutation. Genes for bone morphogenic proteins, in particular BMP-4, are thought to be a plausible candidate genes[5] which resulting in stem cells differentiating into osteoblasts and finally leading to ossification.[3] MO rarely affects the head and neck muscles. Not more than 30 cases have been reported in the maxillofacial region. None of the cases enlisted have substantiated the diagnosis of MO with respect to the pattern of ossification. The highest incidence of MO involving the masticatory

muscles was in the masseter, but case reports have described occurrence in temporal, medial and lateral pterygoid muscles.[7, 8] A possible explanation of MO occurrence within the masseter muscle is that, anatomically, this muscle is most likely to receive the full impact of any direct trauma, for example, as a result of inter-personal violence. Seehra J, R. Lloyd. suggested the implication of pericoronitis and local anaesthesia infiltration of the mandibular foramen as contributing factors to the development of MO within the medial pterygoid.[9] Narang, Dixon and Parkash *et al.* have reported cases of MO of the medial pterygoid muscle following the extraction of a lower wisdom tooth.[9]When affecting the masticatory muscles, MO can be asymptomatic and often produces severe trismus. It has also been reported that MO can affect other muscles of the head and neck region, including the soft tissues associated with the chin and buccinator, genioglossus, platysma and sternocleidomastoid muscles.[10,11]

It has been found out that symptoms can manifest 2–5 weeks post injury and then ossification sites can be detected radiographically.[4] Plain radiographs highlight an area of abnormal calcification seen at the periphery and has a ring-like configuration. CT is the best imaging modality for diagnosing MO: as it demonstrates soft tissue involvement and is a useful aid for surgical planning. MRI is a sensitive technique for identifying small, early lesions but is non-specific. Extensive muscle edema may be seen. Bone scintigraphy is very sensitive in the early detection of MO, demonstrating increased uptake in damaged muscle.[4,5,10]

Biopsy remains the most accurate form of diagnosis. Histologically, a distinctive appearance of zonal ossification is seen with muscle degeneration and calcification dispersed in a fibrocellular stroma.[9] The histopathology can guide us to the early changes which can prevent the diagnostic dilemmas as well as prevent patient from undesired trauma. The use of special stain as Van-Gieson and Vonkossa led to clear visualization of the muscle degeneration as well as the ossified bone that led us to define early and late changes in MO, which were confirmed by using polarized microscopy.[10]

The distinction between these mineralized masses is quite arbitrary in routine H & E sections which can be accurately identified in polarizing microscopy. Polarizing microscopy selectively visualizes anisotropic structures, which appear bright and shining on dark field. The study of MO with polarizing microscope gave an added advantage as it clearly helped us in delineating the early and late changes in collagen fibers arrangement based on the changing direction of the fibers, based upon the principle that if the new direction of these fibers were perpendicular to the surface, they would not be birefringent since collagen is isotropic when viewed.. Young collagen, fibrils, which are more hydrated and less perfectly aligned than those of mature collagen, is also less or not at all anisotropic. Cross linking between fibers determine the amount of birefringence.[8] The muscle pattern ranging from degeneration of muscles to formation of calcified bodies was also analyzed. A possible explanation for this phenomenon could be that in muscles compound birefringence occurs and both the intrinsic and the form birefringence contribute to the positivity which is weaker than that of the collagen. Atrophy or loss of functional activity in muscle markedly affects the intensity of its birefringence. Fischer found reduced birefringence in degeneration and atrophy of muscle .[8,9,10]

In the present case, we could also identify the calcified patterns of bone which was found to be mature concentric lamellar patterns which led to the conclusions that the patient suffered from MO in the early stage: even when it was incorrectly diagnosed as ankylosis of TMJ. Secondly, it depicted the late changes in MO due to the repeated surgical explorations. The possible explanation for the birefringence of bone could be that in bone both the organic matrix and the inorganic crystals are anisotropic hence birefringence can be studied in both. In

decalcified bone the main anisotropic compound is collagen.[8,10]

Studies carried out with the help of polarizing microscope to decipher the presence of the kind of mineralized tissue has led to the clear differentiation between immature and mature bone, and also had highlighted the early and late changes in the form of ossification of the degenerated muscle bundles, which were not clearly evident in light microscopic analysis.

Conclusion

The presence of the tissue architecture and the type of cell also further emphasizes that the origin of MO as a *de novo*. This would lead to an analysis that could suggest the origin of these lesions to be in deeper structures. Further studies at the tissue/ cellular levels need to be emphasized for this predominantly clinically discussed disease. Also, molecular/ subcellular findings could be the direction in which the enigma of the disease entity called Myositis Ossificans needs further research.[11,12]

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