INTRODUCTION

Myositis ossificans is a rare disorder in which ossification develops within skeletal muscles or soft tissue. It is broadly classified into two clinical subtypes; myositis ossificans progressiva (MOP) and myositis ossificans traumatica (MOT). MOP is acquired as an autosomal dominant disease in which ossification develops within muscles, fascia, ligaments or tendons and sometimes multiple members within a family are affected.1 It often begins in early childhood causing progressive restriction in movement and ankylosis of joints eventually leading to death by the involvement of respiratory muscles.1

MOT (also known as myositis ossificans circumscripta or fibrodysplasia ossificans circumscripta); develops after trauma or inflammation within muscles. MOT is frequently reported in the orthopedic literature, and it is most common in the quadriceps femoris and branchialis anticus. There have been few reports of this lesion in the masticatory muscles. Of these, most previous cases involved the masseter muscle.2 Remission is achieved through surgical excision of the ossified mass but recurrence has also been extensively reported.3

CASE REPORT

A 19 years male patient came to the Oral and Maxillofacial Surgery outpatient department with chief complaint of progressively reducing mouth opening for the past four months. History, clinical and radiological examination strongly suggested it to be a rare case of myositis ossificans traumatica which was subsequently confirmed histopathologically. The patient was managed by extirpation of the ossified left lateral and medial pterygoid muscles with interpositioning of split thickness temporalis muscle and fascia flap and post-operative mouth opening exercises. Satisfactory mouth opening was achieved. However recurrence/relapse was noted soon after.

Key words: Muscles of mastication, Ossification, Temporalis muscle and fascia flap

ABSTRACT

A 19 years male patient came to the Oral and Maxillofacial Surgery outpatient department with chief complaint of progressively reducing mouth opening for the past four months. History, clinical and radiological examination strongly suggested it to be a rare case of myositis ossificans traumatica which was subsequently confirmed histopathologically. The patient was managed by extirpation of the ossified left lateral and medial pterygoid muscles with interpositioning of split thickness temporalis muscle and fascia flap and post-operative mouth opening exercises. Satisfactory mouth opening was achieved. However recurrence/relapse was noted soon after.

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which he reported to the dental OPD of another hospital where he was diagnosed as having odontogenic infection and was put on intravenous antibiotics, without success. He was then advised various laboratory investigations, imaging and was re-diagnosed as having fibrous adhesions at the left temporomandibular joint. The patient was operated under general anesthesia with blind nasal endotracheal intubation and jaw stretching was performed with dental mouth gags. The patient reported signs of improvement and was discharged on the fifth postoperative day. There was progressive limitation of mouth opening in subsequent weeks for which he was referred to oral & maxillofacial department, PIMS for further management. On examination spontaneous mouth opening was measured to be 5 mm (Figure 1). Arch bar was in place. Orthopantomogram (OPG) showed carious left first mandibular molar (Figure 2) while all laboratory investigations including complete blood chemistry (CBC), serum calcium and phosphate, alkaline phosphatase, parathyroid hormone, vitamin D3 and serum calcitonin were within normal limits. Conventional conservative methods to improve mouth opening were employed after admitting the patient but they yielded no favorable results. Computed tomography (CT) was done which demonstrated calcified plaques in the left medial and lateral pterygoid muscles extending from the lateral pterygoid plate to their insertions into the ramus neck of the mandible respectively (Figure 3). A differential diagnosis of heterotopic bone formation, osteosarcoma and myositis ossificans was formulated and the patient was scheduled for surgery under general anesthesia. After administrating general anesthesia by fiberoptic nasal endotracheal intubation, the medial surface of the mandibular ramus was approached using a submandibular Risdon incision (Figure 4). The ossifications in the medial and lateral pterygoid muscles at their insertions in the mandibular ramus and neck were then extirpated using bur and osteotome (Figure 5). Pterygoid process was approached from the oral cavity and the ossified mass was detached from the base of pterygoid process with a chisel. A split thickness temporalis muscle and fascia flap was taken using Al-Kayat Bramley incision which was invested on the medial surface of the mandibular ramus and secured to the masseter muscle at the inferior border of mandible to prevent heterotopic bone formation (Figure 6). Intraoperative mouth opening was recorded at 42 mm (Figure 7). The retrieved surgical specimen as well as a submandibular lymph node was submitted for histopathological analysis. The patient recovered uneventfully from anesthesia. Furthermore light jaw stretching was instituted on the 2nd postoperative day whereas rigorous exercises on the 7th day after the surgery. The result of histopathology confirmed the diagnosis of Myositis Ossificans Traumatica of Lateral and Medial Pterygoid.
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was discharged on 10th postoperative day with spontaneous mouth opening of 33 mm and recalled for weekly follow up visits. On his 2nd follow up visit the spontaneous mouth opening was 28 mm then after reducing to 21 mm on the 4th visit. The patient failed to report back to the department after that.

DISCUSSION

Myositis ossificans traumatica is elaborately reported in orthopedic literature. Even though the maxillofacial region is a common site of trauma, reports of MOT are extremely rare. A review of the literature showed not more than thirty previously reported cases of MOT in the maxillofacial region.\(^4\) Approximately two-thirds of these cases involved the masseter muscle, which does not seem unreasonable, because the muscle is broad and covers the lateral side of the mandibular ramus and is susceptible to damage as a result of such incidents as fights or work-related accidents.\(^4\) MOT in more than one muscle, as in our case is even rarer (5 previously reported).
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There is male predilection of 3:1 due to more male involvement in outdoor activities and contact sports. 5 Some authors believe myositis ossificans traumatica to be an inadequately descriptive term because in as many as 25% of cases, no history of trauma can be obtained. 6 Apart from myositis ossificans progressiva, which has a genetic origin, European authors have described two other variants of myositis ossificans as traumatic myositis ossificans circumscripta and atraumatic myositis ossificans circumscripta. 7 These authors have suggested an infective origin to atraumatic myositis ossificans. 8 In the present case report history of trauma during mastication was established.

The exact mechanism for the development of MOT remains unknown. Carey proposed four main theories of its development: 1) displacement of bony fragments into the soft tissue with subsequent proliferation, 2) detachment of periosteal fragments into the surrounding tissue with proliferation of osteoprogenitor cells, 3) migration of subperiosteal osteoprogenitor cells into surrounding soft tissue, through periosteal perforation induced by trauma, and 4) metaplasia of extraosseous cells exposed to bone morphogenic proteins derived from the lysis of bone fragments displaced within the soft tissue during traumatic injury. 9 Among above mentioned theories, the latter theory seems to be accepted most widely. It has been proposed more recently that MOT results from initial intramuscular hemorrhage with exuberant proliferation of vascular granulation tissue that undergoes subsequent metaplasia to cartilage or bone. 10 Molloy and McGuirk also listed four theories for the pathogenesis of MO: infection, ossification of a hematoma, tearing of the periosteum with subsequent callus, and metaplasia of muscle and fibrous connective tissue to cartilage and bone. 11

Despite the recent advances in radiographic imaging, diagnosis of MOT still remains difficult. It is extremely difficult if not impossible to diagnose MOT with conventional plain radiography but computed tomography (CT) at different planes can facilitate tremendously in diagnosis. CT can define the extraskeletal location, extent of the lesion and the degree of invasion into the surrounding soft tissue. The clinical picture of MOT is often confused with osteosarcoma 12, and biopsy is arguably the most accurate method of confirming the diagnosis. However, a surgical procedure done too early can apparently lead to a greater recurrence rate. 13 Thus biopsy should be performed only on lesions when the diagnosis is in doubt after a precise work-up. We finally performed excisional biopsy to secure a definitive diagnosis as well as aid the restricted oral opening and function.

The only treatment modality accepted universally is complete excision of the ossified mass as early as possible followed by aggressive postoperative physiotherapy. 14 Various pharmacologic agents have been used as adjunctive treatment modality. Preoperative and postoperative bisphosphonates, warfarin, low-dose radiation therapy, corticosteroids, and nonsteroidal anti-inflammatory drugs have been used empirically but have shown limited additional benefit or no benefit at all. 15 Of the previously reported cases, almost all were treated by surgical excision followed by physiotherapy. The surgical procedure in these cases consisted of excision of the calcified mass along with removal of a portion of the muscle. Such therapy typically yielded early success as measured by an increase in opening of the jaws. However, follow-up revealed decreased opening of the jaws after less than 3 months. 16 Calcification in the muscles was seen in failed cases, and it was presumed that surgical hemorrhage occurring during the procedure to remove the original calcified mass was contributory. It was also suggested that inadequate physical therapy may have contributed to relapse of diminished jaw opening. 17

Interpositional materials have also been suggested to minimize the chances of recurrence. Lesions in lateral pterygoid muscle, with its limited accessibility, are more prone to recurrence. Spinazzze et al used silicone rubber sheets to cover the medial aspect of the angle; however recurrence was noted after a time lapse and the sheets had to be removed. 18 Narang and Dixon interposed silastic sheets for a case of recurring MOT successfully. 19 The use of abdominal and buccal pad of fat in case of MOT has been reported. 6,19 The use of temporalis muscle and fascia flap to prevent heterotopic bone formation in temporomandibular joint surgery has been reported frequently 20 therefore we used partial thickness temporalis muscle and fascia flap as an interpositional resource on the medial side of mandibular ramus, securing it at the inferior border of masseter to prevent recurrence.
CONCLUSION

Little was found about MOT in current literature especially of lateral and medial pterygoids and there is no universally accepted protocol for the management of such cases. The recommendations are based purely on personal experiences and no long term follow-up support is available in literature to validate any particular treatment. Although conservative measures may not prove to be adequate in treating MOT; but it is suggested that surgery should also always be planned with caution and a clear understanding of the potential morbidity and recurrence.

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REFERENCES