Abstract. Myositis ossificans (MO) is a disease where heterotropic bone forms within a muscle or other type of soft tissue. MO is classified into two groups, MO progressiva and post-traumatic MO. It rarely occurs in the masticatory muscles and thus, only 20 cases involving the masticatory muscles have been reported since 2001. The majority of the reported cases occurred due to trauma, repeated injury or surgical manipulation. However, in a small number of cases, no specific traumatic event was identified as the cause of MO. To the best of our knowledge, this is the first case of post‑infectious MO to be reported in the medial and lateral pterygoid muscles.

Introduction

Myositis ossificans (MO) is a disease where the formation of heterotropic bone occurs within a muscle or other type of soft tissue (1). MO is classified into two groups, MO progressiva (MOP) and post‑traumatic MO (PTMO) (2). MOP is an autosomal dominant disease observed within families in which multiple heterotopic ossifications develop systematically in various muscles, fascia, tendons and ligaments of the body (2,3). PTMO is characterized by heterotopic bone formation within muscle tissue as a result of a single or repetitive injury (2,4). PTMO is frequently reported in the orthopedic literature and is prevalent in the quadriceps femoris and brachialis anticus, where there is high risk for injury. However, MO is rare in the masticatory muscles. Only 20 cases of MO in the masticatory muscles were identified during a review of the literature (since 2001), which was conducted in the present study; 16 cases were associated with an evident traumatic cause and were diagnosed definitively as PTMO.

In the present study, a rare case of MO in the medial and lateral pterygoid muscles that was caused by odontogenic infection is presented, which was diagnosed as post-infectious MO (PIMO). To the best of our knowledge, this is the first case of PIMO in multiple masticatory muscles to be reported in the English literature. Written informed consent was obtained from the patient.

Case report

In January 2010, a 42-year-old female was referred to the Department of Oral and Maxillofacial Surgery, Ninth People's Hospital (Shanghai, China) with a complaint of the progressive, painless limitation of mouth opening for three years. The patient had no history of evident trauma, however, had experienced pain in the right upper jaw for approximately three years, for which the patient had not received any endodontic or periodontal treatment. In addition, the patient had experienced weakness when biting and chewing, which had endured for more than two years. The patient was administered non-steroidal anti-inflammatory agents by The First Hospital of Jiaxing (Jiaxing, China), however, no clinical improvement was observed.

Physical examination revealed that the patient was well-nourished and demonstrated no evidence of developmental abnormalities. No facial asymmetry was apparent and the limited range of motion, the patient reported no associated pain or changes in the occlusion. On palpation the masseter and temporalis muscles were normal. The lymph nodes (submandibular and deep cervical) were nonpalpable and nontender. Intraorally, the right maxillary third molar residual root and missing mandibular central incisors were examined. The patient's dental hygiene was poor, however, the oral mucosa and tongue appeared to be healthy.

The patient's general medical status was normal and the laboratory tests, including serum calcium (2.23 mmol/l; normal range, 2.08-2.65 mmol/l) and phosphorus levels (1.46 mmol/l; normal range, 0.78-1.65 mmol/l), were within the normal limits. A panoramic radiograph (Fig. 1) showed
chronic periapical lesions of the right maxillary third molar residual root (Fig. 1A). Computed tomography (CT) scans (Fig. 2) revealed heterotopic bone formation in the right lateral pterygoid and medial pterygoid muscles (Fig. 2A and B). Normal anatomic structures between the articular disk and the condyle were observed on magnetic resonance imaging (MRI; Fig. 3A and B). Accounting for the medical history and clinicoradiological results, the patient was diagnosed with MO in the right lateral and medial pterygoid muscles.

Surgical excision was performed under general anesthesia and, following fiber optic assisted intubation, access to the mandibular was achieved via a preauricular incision, which extended to the temporal region and exposed the condylar process and sigmoid notch, as well as the coronoid process. To remove the calcification in the lateral pterygoid muscle and reduce the tension on the mandible caused by the temporalis, a right coronoidectomy was performed using a reciprocating saw. The majority of the calcified and fibrotic fibers of the upper head of the right lateral pterygoid muscle were removed. An additional intraoral approach to the medial aspect of the mandibular was adopted via a mucoperiosteal incision of the retromolar area to reveal and access the calcified medial pterygoid muscle. A pedicled buccal fat pad (BFP) flap was used to fill the dead space. A calcified mass was identified and subsequently excised. In addition, the masseter attachment was stripped from the ramus of the
Table I. Case reports of myositis ossificans in masticatory muscles (20 cases reported since 2001).

| Author (Ref.) | Patient (Age, years/ Gender) | Location | Chief complaint | History of trauma | Disease duration | MIO (preoperation) | Treatment | MIO (Intraoperation) | MIO (follow-up) | Outcome          |
|---------------|-------------------------------|----------|-----------------|-------------------|-----------------|--------------------|-----------|----------------------|----------------|------------------|
| Nemoto *et al* (5) | 39/M                          | Masseter; temporalis; lateral pterygoid; frontalis | Trismus mass | Repeatedly struck on the face with a plastic hammer | >1 year | 5 mm | Excision + muscle release + bilateral coronoidectomy | 55 mm | 37 mm (1 year) | No recurrence |
| Jayade *et al* (2) | 25/F                          | Medial pterygoid; lateral pterygoid; temporalis | Pain; swelling; trismus | None | >6 months | 2 mm | Osteotomy + excision + contralateral coronoidectomy | 45 mm | 39 mm (3 months) | No recurrence |
| Guarda-Nardini *et al* (4) | 50/M                          | Temporalis | Pain; trismus mass | Trauma injury | 40 days | 12 mm | Excision + coronoidectomy | Good | 35 mm (6 months) | No recurrence |
| Choudharya *et al* (6) | 31/M                          | Medial pterygoid | Trismus mass | Panfacial trauma | 3 years | 8 mm | Excision | 37 mm | 27 mm (30 months) | No recurrence |
| Thangavelu *et al* (11) | 36/F                          | Medial pterygoid | Trismus; pain | Anesthesia injection + tooth extraction | 3 months | 3 mm | Osteotomy + excision + abdominal fat graft | 32 mm | 28 mm (9 months) | No recurrence |
| Godhi *et al* (19) | 21/M                          | Medial pterygoid; lateral pterygoid; temporalis | Pain; swelling; trismus mass | None | 6 years | 5 mm | Osteotomy + excision + reconstruction plate with a condyle | 42 mm | Gradual decline (1 year) | Unknown |
| Trautmann *et al* (12) | 33/M                          | Medial pterygoid | Trismus; tenderness; swelling | Anesthetic injection + endodontic treatment | 2 months | 5 mm | Coronoidectomy + partial resection of calcified medial pterygoid | Unknown | Limitation (3 years after the second surgery) | Recurred twice |
| Ramieri *et al* (13) | 64/M                          | Medial pterygoid | Swelling; trismus | Anesthetic injection + tooth extraction | 3 years | 15 mm | Excision | 38 mm | Unknown | Unknown |
| Kruse *et al* (20) | 35/F                          | Masseter | Trismus; tenderness | None | 12 years | 10 mm | Conservation treatment | Unknown | Unchanged (10 mm) | Regular follow-up |
| Conner and Duffy (1) | 18/F                          | Medial pterygoid; temporalis masseter | Pain; trismus | Anesthetic injection + tooth extraction | 4 months | 4 mm | Excision + coronoidectomy | Unknown | 25 mm (>18 months after the third surgery) | No recurrence |
Table I. Continued.

| Author (Ref.) | Patient (Age, years/Gender) | Location | Chief complaint | History of trauma | Disease duration | MIO (preoperation) | Treatment | MIO (Intraoperation) | MIO (follow-up) | Outcome |
|---------------|-----------------------------|----------|-----------------|-------------------|-----------------|-------------------|-----------|-------------------|----------------|---------|
| Bansal et al (16) | 20/F | Buccinators; medial pterygoid | Trismus | Dento-alveolar trauma | 2 years | 1 mm | Excision + bilateral coronoidectomy + ipsilateral palatal pedicle flap | 35 mm | 30 mm (1 year) | No recurrence |
| Rattan et al (18) | 45/M | Medial pterygoid | Trismus | Absolute alcohol injection | 8 months | 7 mm | Excision + pedicled buccal fat pad flap | 30 mm | 45 mm (2 years) | No recurrence |
| Mazano et al (21) | 51/M | Temporalis | Trismus; mass trauma | Severe trauma | 25 years | 13 mm | Excision | Unknown | 38 mm (1 year) | No recurrence |
| Yano et al (8) | 34/M | Masseter | Trismus | Criminal violence | Half year | 5 mm | Excision + coronoidectomy | Unknown | 40 mm (10 months) | No recurrence |
| Uematsu et al (22) | 38/F | Temporalis | Masseter; mass trismus | None | 2 weeks | Unknown | Excision + coronoidectomy + excision + a penrose drain placed | Unknown | 40 mm (3.5 years) | No recurrence |
| St-Hilaire et al (14) | 68/M | Temporalis | Masseter; temporalis | Anesthesia injection + tooth treatment | 2 weeks | 5 mm | Coronoidectomy + excision + tooth treatment | Unknown | Unknown | Unknown |
| Saka et al (9) | 33/M | Temporalis | Trismus; pain; swelling | Blunt trauma | 3 weeks | Limited mouth opening | Excision | Unknown | Unlimited mouth opening (4 years) | No recurrence |
| Aoki et al (10) | 44/M | Masseter; lateral pterygoid; Trismus; pain; swelling; tenderness | Blunt trauma to the face | 1 year | 7 mm | Excision | Unknown | 32 mm | 10 mm (10th day after surgery) | Recurrence |
| Kim et al (15) | 30/F | Lateral pterygoid; Trismus | Anesthesia injection + tooth treatment | 3 years | 8 mm | Coronoidectomy + excision + interpositional abdominal fat graft placed | 27 mm (in the final surgery) | 12 mm passively (after the final surgery) | Multiple recurrences |
| Mevio et al (17) | 55/F | Temporalis | Trismus | Dental surgery | 18 months | 6 mm | Coronoidectomy + excision | Unknown | Correct mouth opening | No recurrence |

MIO, maximal incisal opening.
mandible to weaken the contractile force of the masseter muscle and mouth opening of 40 mm was achieved intraoperatively. The right maxillary third molar residual root was removed. The wound was closed in layers following the achievement of complete hemostasis. The healing period was uneventful and a postoperative panoramic radiograph and a CT scan were performed four days following surgery, which revealed that the ossification had been excised (Figs. 1B and 2C). Histopathology of the excised tissue specimens (Fig. 4) identified the novel formation of bone and osteoid within the muscle fibers. Physical therapy was initiated in the immediate postoperative period using suitable analgesics (200 mg celecoxib, twice a week, for one week) and was continued following discharge from the hospital. Maximum spontaneous mouth opening of 30 mm was achieved seven days following surgery.

The patient was followed up for a total of 36 months postoperatively. At present, the patient exhibits a stable interincisal opening of 25 mm.

Table II. Clinical features of myositis ossificans in the masticatory muscles (20 cases reported from 2001) for 12 males and eight females (mean age, 36.75 years).

| Parameter       | Patients, n |
|-----------------|-------------|
| Location        |             |
| Masseter        | 6           |
| Lateral pterygoid| 6           |
| Medial pterygoid | 11          |
| Temporal        | 10          |
| Chief complaint |             |
| Trismus         | 20          |
| Pain            | 8           |
| Mass            | 5           |
| Swelling        | 6           |
| Tenderness      | 3           |
| Recurrence      |             |
| No              | 13          |
| Yes             | 4           |
| Unknown         | 4           |

Table III. Precipitating factors of myositis ossificans in the masticatory muscles (20 cases reported from 2001).

| Precipitating factor                  | Patients |
|---------------------------------------|----------|
|                                       | n  | %  |
| Facial trauma                         | 8  | 40 |
| Local infiltration of anesthetics     | 6  | 30 |
| Dental surgery                        | 1  | 5  |
| Local infiltration of absolute alcohol| 1  | 5  |
| Unknown                               | 4  | 20 |

MO presenting in the masticatory muscles is rare; a review of the literature, which was conducted in the present study identified only 20 cases reported since 2001. The results of the literature review are presented in Tables I-III. The mean patient age was 36.75 years (range, 18-68 years). Nineteen of the 20 patients initially attended the hospital presenting with restricted mouth opening. Of the 20 patients, 16 were diagnosed with PTMO as a result of facial trauma (4-10), local infiltration of anesthetics (1,11-15), dental surgery (16,17) or absolute alcohol injection (18). However, the remaining four patients had no evident history of trauma, tooth extraction or infection (2,19,20,22). Among the patients diagnosed with PTMO, males predominated (ratio of males to females, 11:5), which may be attributed to the fact that males are more likely to be subject to trauma in daily life (10). The region where PTMO most frequently occurred was the medial pterygoid muscles, which was often caused by a local anesthesia injection, followed by the application of external force directly to the temporalis and masseter muscles. The present review of the literature revealed that out of the 20 cases observed, the surgical management of one case (14) was performed at the early stage of PTMO in the temporalis without the observation of calcification, however, in the other cases, surgery was conducted when trismus occurred and calcification was identified via CT. In addition, of the 17 patients that were followed up, 10 patients were continued with the follow-up for more than one year and four patients exhibited recurrence subsequent to the first surgical treatment.

The exact mechanism for the pathogenesis of MO remains unclear, however, trauma is considered to be the inciting event. According to the literature, a signal, such as a bone morphogenetic protein (BMP) signal from the site of injury, may induce mesenchymal cells to differentiate into osteoblasts or chondroblasts, given the appropriate environment (23,24). In the field of stomatology, odontogenic infection is a common condition when accompanied by trauma. In the present case,
Ossification, the symptom of MO, may be observed by diagnostic imaging tests a minimum of 2-5 weeks subsequent to injury (25-27). After eliminating temporomandibular joint disease using MRI, CT and three-dimensional CT scans are considered to be particularly efficacious investigative tools in the oral and maxillofacial region. These imaging techniques aid with identifying the exact location and shape of the ossification, as well as establishing the association between the lesion and surrounding tissues, which is important for surgical treatment. Although a panoramic radiograph may not be effective for determining the exact extent of the lesion, due to the superimposition of the cranial bones, it may aid with the identification of odontogenic infection foci. Bone scans and ultrasound may also be used, however, are rarely applied for the craniofacial region (28).

Treatment of PTMO and PIMO usually includes surgical excision of the calcification and the surrounding muscles. Patients with MO of the temporalis or masseter area often undergo a coronoidectomy and the excision of the involved calcified muscles; whereas MO of the pterygoid muscle is more debilitating and, thus, the management of these types of patient is more complicated than that of the patients exhibiting MO of other masticatory muscles. According to the experiences of the present study, the following approaches should be considered: i) Transoral and extraoral approaches, which are often used to provide access to the medial aspect of the mandibular ramus to allow complete excision of the ossified muscle; ii) protection of the internal maxillary artery and inferior alveolar nerves (this is considered to be critical); and iii) using a BFP flap to fill the dead space for preventing hematoma formation and heterotopic bone reformation (29,30). Two types of free fat, abdominal fat and the BFP, have been reported that may serve as interpositional material. The BFP has been identified as a particularly effective autogenous tissue, which has been demonstrated in a multitude of surgical procedures in the maxillofacial region (31,32). The BFP lies in close proximity to the site of surgery and may be used as a pedicled or random-pattern flap along with its own blood supply, so there are fewer instances of resorption when compared with an abdominal fat transfer.

In conclusion, a case of PIMO in the medial and lateral pterygoid muscles is presented and chronic low-grade infection was identified to be an important consideration in addition to other possible precipitating factors in the occurrence of MO. Panoramic radiography revealed the source of infection and CT scans effectively delineated the calcified mass. A positive outcome was achieved for the patient by the surgical excision of the calcification and ossified muscles, and via the use of a BFP flap to fill the dead space. This study indicates that symptomatic wisdom teeth must be removed as soon as possible, to prevent infection. In addition, it is important to consider infected as a factor which may lead to myositis ossificans.

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