Int J Clin Exp Med 2016;9(12):23713-23720
www.ijcem.com /ISSN:1940-5901/IJCEM0030026

Case Report

Application of digital medicine techniques in the surgical treatment of myositis ossificans traumatica: a case report and review of the literature

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Received April 4, 2016; Accepted September 7, 2016; Epub December 15, 2016; Published December 30, 2016

Abstract: Myositis ossificans traumatica (MOT) is a rare heterotopic ossification disease which rarely occurs in masticatory muscles and thus, only 28 cases have been reported in the English literature since 2001. We present an unusual long-term case of MOT in temporalis muscle. The treatment protocol involved a radical lesion resection and bilateral coronoidectomy. According to the literature review, this is the first report of a case which utilized digital medicine techniques to aid in MOT surgical treatment. A conclusion of pathogenesis hypotheses and different treatment methods is also presented.

Keywords: Myositis ossificans traumatica, digital medicine, temporalis muscle

Introduction

Myositis ossificans (MO) is a rare heterotopic ossification disease involving muscles or soft tissues [1]. Myositis ossificans has been traditionally classified into 2 groups: myositis ossifican progressiva (MOP) and myositis ossificans traumatica (MOT), also known as post-traumatic MO (PTMO) [2] or myositis ossificans circumscripta (MOC) [3]. MOP is an autosomal dominant disease which causes symptoms from early infancy and involves several muscles. The consequent functional limitations are progressive and handicapping. MOT is a more circumscribed form, which involves single muscle, or muscle groups, subjected to violent or repeated trauma [4].

MOT is likely occurring in the femoral region, or brachium, but rarely in masticatory muscles. Thus, only 28 cases involving masticatory muscles have been reported in English literature since 2001 [5-13]. Among them, there are approximately 42.9% (12 cases) involving the temporalis muscle. Therefore, the temporalis muscle is one of the most vulnerable muscles of MOT in maxillofacial regions. The most accredited treatment is a radical surgical excision. However, because of the complex maxillofacial structure and the unpredictable vascular variation, the surgery can be very challenging. In this case, we successfully implemented computer-assisted surgery (CAS) and computer-aided design/computer-aided manufacture (CAD/CAM) techniques to aid in MOT excision and intraoperative maxillofacial reconstruction. To the best of our knowledge, this is the first case utilizing digital medicine techniques in MOT surgery.

In this article, we present an unusual MOT case, with the longest duration (45 years) and the most severe symptom (maximal mouth opening=0 mm) among all the cases reported in English literature since 2001. Furthermore, we discuss the treatment protocol, including the use of digital medicine techniques to improve the surgical outcome.

Case report

A 49-year-old Chinese woman was referred to the Department of Oral and Maxillofacial Surgery, Ninth People's Hospital (Shanghai, China) with a complaint of a reduced mouth opening for 45 years. The patient experienced a

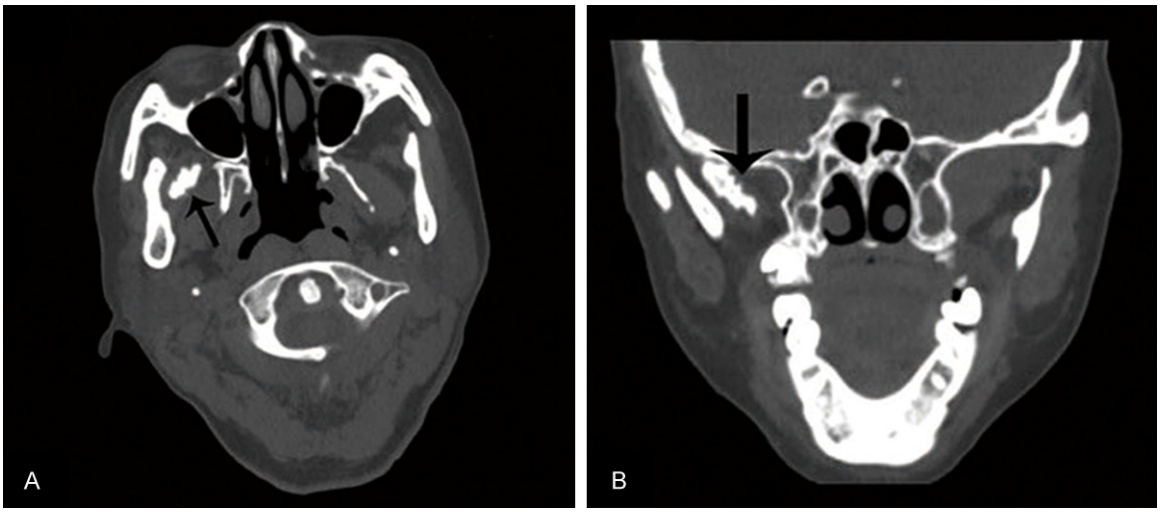


Figure 1. Preoperative CT imaging demonstrated ossification in the lower part of the right temporalis muscle (black arrow). A. Horizontal view. B. Coronal view.

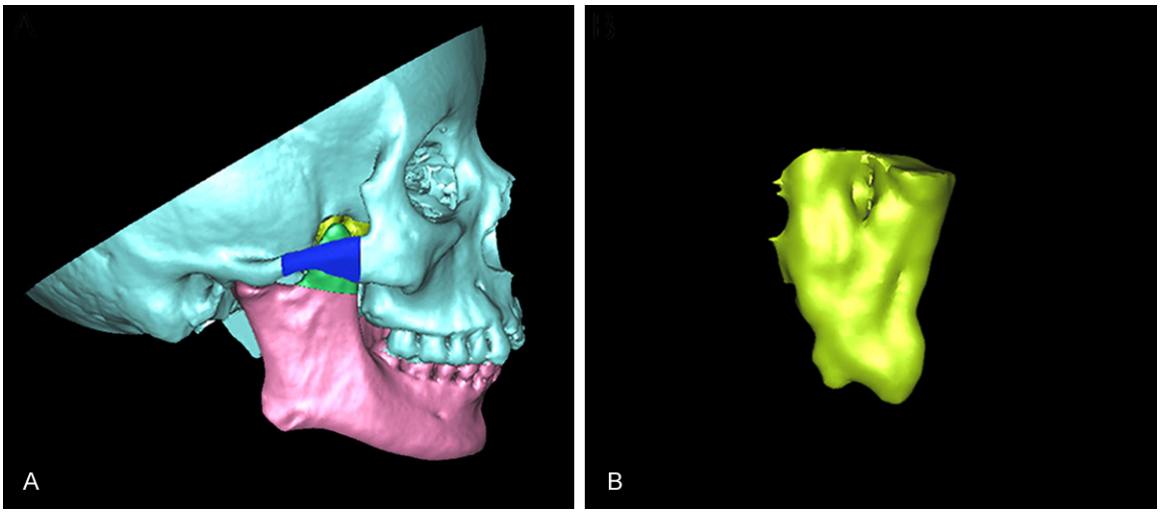


Figure 2. Three-dimensional reconstruction of the lesion. A. Space relationship of zygomatic arch, coronoid process and the ossified mass (yellow). B. The ossified mass (anterior view).

fall accident when she was 4 years old, and thereby developed a gradually reduced mouth opening. The patient had received no treatments before being referred to our hospital, and she had a diet of only soft food in small pieces.

Clinically, the patient was moderately nourished and demonstrated no evidence of developmental abnormalities. No facial asymmetry was obvious and the maximal incisal opening (MIO) was 0 mm, without temporomandibular joint (TMJ) clicking. Despite the limited range of motion, the patient reported no associated pain in the TMJ region upon palpation. The patient's dental hygiene was poor due to the reduced mouth opening.

CT scanning (**Figure 1**) revealed an expanding, hyperdense mass in the lower part of the right temporalis muscle, indicating heterotopic bone formation. A three-dimensional reconstruction (**Figure 2**) was performed with the help of *Simplant Pro 11.04* software (Materialise Dental, Leuven, Belgium), which showed an ossified mass overlapped the right coronoid process and zygomatic arch.

Accounting for her trauma history and clinicoradiological results, the patient was diagnosed

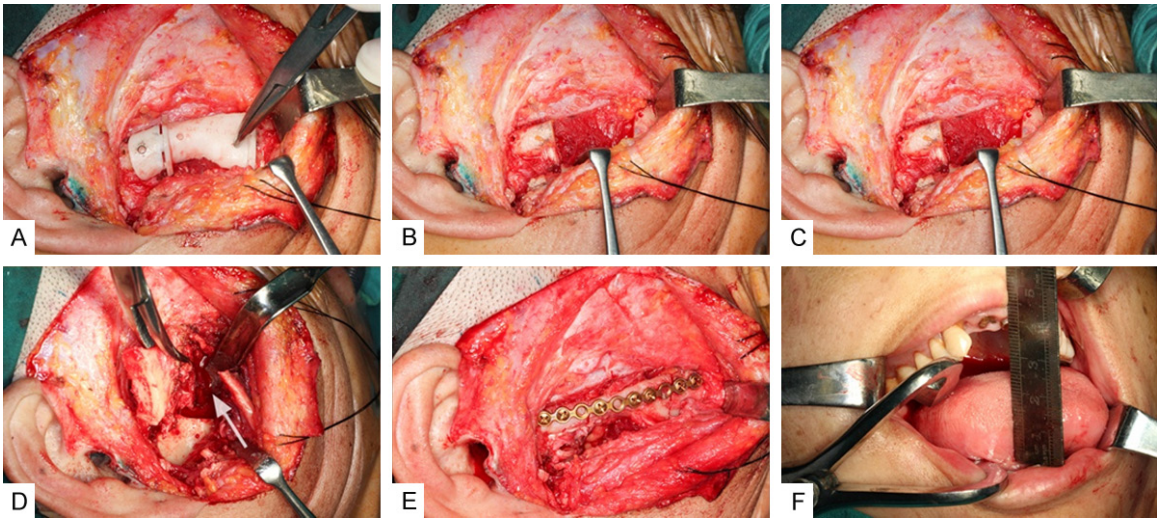


Figure 3. Digital templates were used to guide osteotomy and titanium plate implantation. A, B. Zygomatic arch template used to drill location holes and to do temporary zygomatic arch osteotomy. C, D. Coronoid process templates used to do bilateral coronoidectomy. The white arrow shows the ossified mass. E. Reconstruction of zygomatic arch with titanium plate according to location holes. F. Intraoperative forced maximal incisal opening was 43 mm.

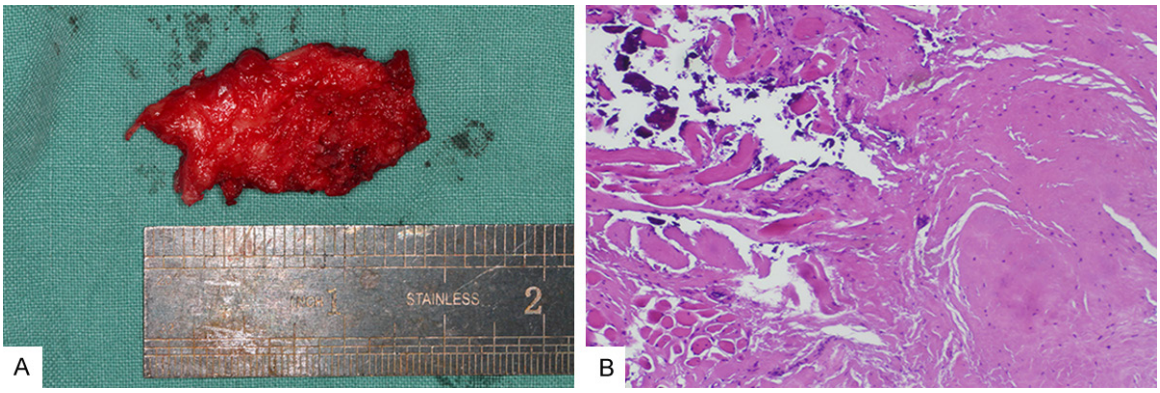


Figure 4. Excised tissue specimen and pathological microphotograph. A. Extracorporeal specimen showed the long axis of ossification mass was 14 mm. B. Microphotograph identified the hyperplasia of bone and osteoid within muscle fibers (Haematoxylin-eosin stain, magnification: 100×).

with MOT in the lower part of the right temporalis muscle.

Under general anesthesia, we performed the surgical excision through a modified preauricular approach which extended from earlobe to the temporal region to expose the zygomatic arch. To remove the ossification located on the inside of the coronoid process and reduce the tension on the mandible, bilateral coronoidectomy and bilateral masseter attachments remission were performed. Furthermore, we cut off the zygomatic arch temporarily to secure that the ossifying mass can be removed en bloc. In addition, digital templates, which were manufactured with CAD/CAM techniques preoperatively, were applied in both the ossification resection and zygomatic arch osteotomy, making the surgery more accurate as well as less time-consuming (**Figure 3A-E**). The long axis of the ossification mass was 14 mm and the forced MIO achieved intraoperatively was 43 mm (**Figures 3F** and **4A**). The healing period was uneventful and a postoperative mouth-opening physiotherapy was advised for at least 1 month. The patient claimed no facial numbness after surgery.

Histopathology of the excised tissue specimen (**Figure 4B**) identified the hyperplasia of the bone and osteoid, surrounded by muscle tissue.

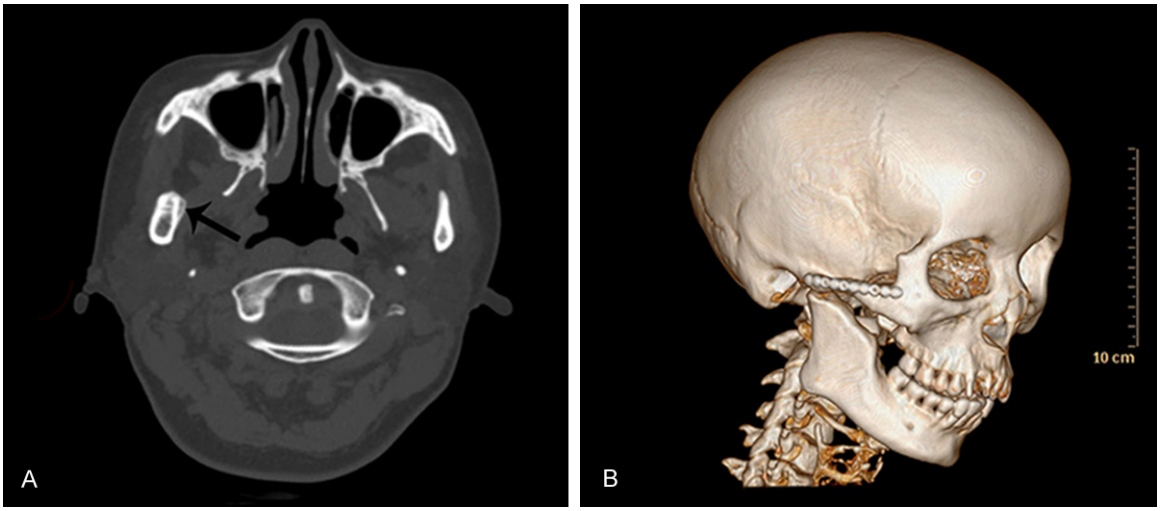
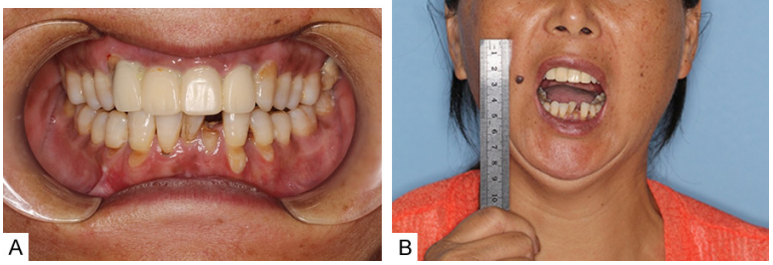


Figure 5. CT imaging and three-dimensional reconstruction (1 year after surgery). A. CT imaging revealed no residual ossification mass and formation of bony callus (black arrow); B. Three-dimensional reconstruction of craniofacial region revealed reconstructed zygomatic arch and absence of coronoid process.

Figure 6. Occlusion and facial appearance (1 year after surgery). A. Neutroclusion (Class I); B. Maximal incisal opening (MIO) was 17 mm, no apparent facial asymmetry was noticed.



Postoperative CT scanning (**Figure 5A**) and three-dimensional reconstruction (**Figure 5B**) showed the complete resection of the ossification mass and the accurate reconstruction of the right zygomatic arch. Neither facial numbness nor pain was apparent 1 year after the surgery. The occlusion was neutral and MIO was 17 mm (**Figure 6**).

Discussion

MOT involving the temporalis muscle is a rare disease with only 8 cases reported in the English literature from 2001 to 2015 (**Table 1**). All of them shared a definite history of facial trauma and a common complaint of restricted mouth opening. Although the duration of MOT symptoms varied from 40 days to 25 years, no recurrences have been reported after excision. In general, the mean age of MOT patient was 33.8 years (range, 12-68 years), and a male to female predominance of 4:3 was observed (**Table 2**). According to the literature review, the duration of our case (45 years) is the longest among all the MOT cases involving masticatory muscles in the English literature.

As its name suggests, MOT is mainly caused by trauma, which can be both iatrogenic and non-iatrogenic. According to the previous research, several causes of MOT have been revealed: tooth extraction [1, 14], local anesthetic injection [14, 15], dental surgery [4], alcohol injection [16], genioplasty, badly performed orthodontic treatment [10], facial skeleton fractures [17] and facial trauma. Although all the factors above have been considered to be the triggers of MOT, the exact mechanism for the pathogenesis remains unclear. Several theories have been proposed. The most accredited hypothesis was proposed by Carey EJ [18]. It has been mentioned in many scientific articles [2, 10, 11, 14], suggesting 4 main theories for the development of MOT: (1) displacement of bony fragments into the soft tissue with subsequent osteoprogenitor cells proliferation; (2)

Table 1. Case reports of MOT involving temporalis muscles (8 cases reported since 2001)

Author	Patient (Age/Gender)	Chief complaint	History of trauma	Duration	Treatment	Outcome
Reddy *et al* [9]	21/M	Trismus	Hit by a heavy vehicle jack rod	2 months	Excision + ipsilateral coronoidectomy	No recurrence
Nemoto *et al* [24]	39/M	Trismus; mass	Repeatedly struck on the face with a plastic hammer	>1 year	Excision + bilateral coronoidectomy	No recurrence
Guarda-Nardini *et al* [25]	50/M	Pain; trismus	Trauma injury	40 days	Injection + coronoidectomy	No recurrence
Conner and Duffy [14]	18/F	Pain; trismus	Anesthetic injection + teeth extraction	4 months	Extended excision + ipsilateral coronoidectomy + disarticulation	No recurrence (after the third surgery)
Mazano *et al* [26]	51/M	Trismus; mass	Severe trauma	25 years	Excision	No recurrence
St-Hilaire *et al* [15]	68/M	Trismus	Anesthesia injection + tooth treatment	2 weeks	Excision + ipsilateral coronoidectomy	No recurrence
Saka *et al* [27]	33/M	Trismus; pain; swelling	Blunt trauma	3 weeks	Excision	No recurrence
Mevio *et al* [4]	55/F	Trismus	Dental surgery	18 months	Excision + ipsilateral coronoidectomy	No recurrence

F: Female; M: Male.

Table 2. Clinical features of MO involving the masticatory muscles (28 cases reported since 2001) for 16 males and 12 females

Parameter	Patients, n	Percentage, %
Location		
Masseter	9	32.1
Lateral pterygoid	9	32.1
Medial pterygoid	14	50.0
Temporalis	12	42.9
Chief complaint		
Trismus	26	92.9
Pain	10	35.7
Mass	6	21.4
Swelling	7	25.0
Tenderness	3	10.7
Epilepsy	1	0.04
Recurrence		
No	16	57.1
Yes	6	21.4
Unknown	6	21.4

detachment of periosteal fragments into the soft tissue with subsequent osteoprogenitor cells proliferation; (3) migration of subperiosteal osteoprogenitor cells into the surrounding soft tissue, through a periosteal perforation induced by trauma; (4) metaplasy of extraosseous cells exposed to bone morphogenetic proteins (BMP). Another widely accepted hypothesis, cited by many articles [10, 19-21], suggests that MOT results from an initial intramuscular hemorrhage and the proliferation of vascular granulation tissue.

CT, magnetic resonance imaging (MRI) and panoramic radiograph are efficacious tools in locating, as well as shaping the ossification mass. The pathognomonic feature of MO is a well-circumscribed, high-attenuating periphery with a low-attenuating central portion [11], which is compatible with our case (**Figure 1**). According to Arima R *et al* [22], the interval between the trauma and the first detection of ossification mass ranged from 3 weeks to more than 20 years.

Although surgeons are well-equipped in detecting the ossification mass, it is still challenging to make the definitive diagnosis between MOT and other diseases which can result in a restricted mouth opening. Spinzia A *et al* [10] suggests that both intra-articular (ankylosis, the anchored disc phenomenon, and bilateral anterior disc displacement without reduction) and extra-articular (infection, foreign body reaction, musculoskeletal injury or disorder, enlargement of the coronoid process and neoplasm) causes must be considered in the diagnostic hypotheses. Therefore, a definite history of trauma will contribute to the diagnosis of MOT and the planning of surgical treatment.

Different treatment strategies of MOT have been proposed in literature. Usually, a radical excision of the ossified mass is adopted by most surgeons, however, the surgical timing is controversial. Some authors [22, 23] hold the view that early surgical intervention(within 6 weeks after symptoms emerged) will lead to an ideal prognosis, whereas others [10, 14] insist the surgical excision should not be performed until the ossification is matured, usually at 6 to 12 months after the onset, to avoid recurrence. Other therapies includes: physiotherapy, medical therapy (nonsteroidal anti-inflammatory drugs, warfarin, etc.) and low-dose radiation therapy [20]. To date, the effectiveness of nonsurgical therapy for MOT remains unclear, and requires further research.

Although the radical surgical excision is a world-accredited treatment for MOT, it remains challenging due to the complexities of the craniomaxillofacial anatomy. Excessive bleeding, facial nerve injuries, and incomplete resection of lesions are the main complications. Nevertheless, the advent of CAS and CAD/CAM techniques makes the change. Digital medicine, as an uprising branch of modern medicine, possesses significant advantages such as accurate, customizable, minimally invasive, etc. With the application of CAS and CAD/CAM techniques, we reconstructed a three-dimensional craniomaxillofacial model according to the CT data, manufactured customized digital templates and performed surgery simulation. During the surgery, with the help of the digital templates and the three-dimensional model, surgeons can easily determine the location and depth of osteotomies. Consequently, not only minimal invasion but also fewer complications were achieved during and after the surgery.

By following the patient for 1 year, we found that her MIO decreased to 17 mm as well as the occlusion was neutral and stable. Due to

the long-term trismus, masticatory muscles strength declined, which in turn led to a restricted mouth opening. The further recovery of opening requires time and exercise.

It must be pointed out that not all MOT patients should adopt surgical treatment. The surgical excision is recommended only if the patient has persistent pain or severe restricted motion. Given the long duration of the disease (45 years) and severe reduced mouth opening (MIO=0 mm), a radical excision of the ossified mass was performed.

Conclusion

MOT involving the temporalis muscle is unusual, with confusing terms of diagnosis. However, its pathognomonic radiographic features and history of trauma will contribute to a definitive diagnosis. In this report, we present a rare case with a disease duration of 45 years. A positive outcome was obtained by performing the surgical excision with CAS and CAD/CAM techniques. Nevertheless, further research of pathological mechanisms of MOT is highly needed.

Acknowledgements

This work was supported by the National Natural Science Foundation of China (81371168, 81671010), Technology Commission of Shanghai Municipality Sci-ence Research Project (14DZ2294300), the Medical-Engineering Cross Fund of Shanghai Jiao Tong University (YG2013MS63), Shanghai Municipal Education Commission-Gao feng Clinical Medicine Grant Support (20152226), the Innovation Fund of Translational Medicine, Shanghai Jiao Tong University School of Medicine (15ZH2007), Science and technology commission of Shanghai biological medicine support project in 2016 (16441908800), Key project of Shanghai municipal health and family planning commission in 2016 (201640001), Ministry of science and technology research and development of the national key projects in 2016 (2016YFC1100600) and the tenth Students' Innovative Training Project of Shanghai Jiao Tong University School of Medicine (2016059).

Disclosure of conflict of interest

None.

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References

[1] Torres AM, Nardis AC, da Silva RA and Savioli C. Myositis ossificans traumatica of the medial pterygoid muscle following a third molar extraction. Int J Oral Maxillofac Surg 2015; 44: 488-490.

[2] Jayade B, Adirajaiah S, Vadera H, Kundalaswamy G, Sattur AP and Kalkur C. Myositis ossificans in medial, lateral pterygoid, and contralateral temporalis muscles: a rare case report. Oral Surg Oral Med Oral Pathol Oral Radiol 2013; 116: e261-266.

[3] Aoki T, Naito H, Ota Y and Shiiki K. Myositis ossificans traumatica of the masticatory muscles: Review of the literature and report of a case. J Oral Maxillofac Surg 2002; 60: 1083-1088.

[4] Mevio E, Rizzi L and Bernasconi G. Myositis ossificans traumatica of the temporal muscle: a case report. Auris Nasus Larynx 2001; 28: 345-347.

[5] Jiang Q, Chen MJ, Yang C, Qiu YT, Tian Z, Zhang ZY and Qiu WL. Post-infectious myositis ossificans in medial, lateral pterygoid muscles: A case report and review of the literature. Oncol Lett 2015; 9: 920-926.

[6] Demirkol M, Aras MH and Tutar E. Myositis Ossificans Circumscripta in the Masseter Muscle Mimicking Phleboliths. J Craniofac Surg 2015; 26: 2020-2021.

[7] Mashiko T, Akizuki T, Watanabe Y, Sasaki R, Yokoyama M, Yoshimura K and Mineda K. Clinicopathologic Assessment of Myositis Ossificans Circumscripta of the Masseter Muscles. J Craniofac Surg 2015; 26: 2025-2026.

[8] Kumar N, Austin RD, Mathew P, Sakthivel S and Vijayalakshmi L. Traumatic myositis ossificans of the masseter muscle: a case report with conventional and advanced imaging features. Gen Dent 2014; 62: 75-77.

[9] Reddy SP, Prakash AP, Keerthi M and Rao BJ. Myositis ossificans traumatica of temporalis and medial pterygoid muscle. J Oral Maxillofac Pathol 2014; 18: 271-5.

[10] Spinzia A, Moscato G, Broccardo E, Castelletti L, Maglitto F, Orabona GD and Piombino P. A rare isolated unilateral myositis ossificans traumatica of the lateral pterygoid muscle: a case report. J Med Case Rep 2014; 8: 230.

[11] Boffano P, Zavattero E, Bosco G and Berrone S. Myositis ossificans of the left medial pterygoid muscle: case report and review of the literature of myositis ossificans of masticatory muscles. Craniomaxillofac Trauma Reconstr 2014; 7: 43-50.
[12] Kamalapur MG, Patil PB, Joshi S and Shastri D. Pseudomalignant myositis ossificans involving multiple masticatory muscles: Imaging evaluation. Indian J Radiol Imaging 2014; 24: 75-79.
[13] Almeida LE, Doetzer A, Camejo F and Bosio J. Operative management of idiophatic myositis ossificans of lateral pterygoid muscle. Int J Surg Case Rep 2014; 5: 796-799.
[14] Conner GA and Duffy M. Myositis ossificans: a case report of multiple recurrences following third molar extractions and review of the literature. J Oral Maxillofac Surg 2009; 67: 920-926.
[15] St-Hilaire H, Weber WD, Ramer M and Lumerman H. Clinicopathologic conference: Trismus following dental treatment. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2004; 98: 261-266.
[16] Rattan V, Rai S and Vaiphei K. Use of buccal pad of fat to prevent heterotopic bone formation after excision of myositis ossificans of medial pterygoid muscle. J Oral Maxillofac Surg 2008; 66: 1518-1522.
[17] Ungari C, Filiaci F, Riccardi E, Rinna C and Iannetti G. Etiology and incidence of zygomatic fracture: a retrospective study related to a series of 642 patients. Eur Rev Med Pharmacol Sci 2012; 16: 1559-1562.
[18] Carey EJ. Multiple bilateral bone and callus femurs and left traumatic parosteal formations of the innominate bone: report of case. Archives of Surgery 1924; 8: 592-603.
[19] Abdin H and Prabhu S. Traumatic myositis ossificans of lateral pterygoid muscle. J Oral Med 1983; 39: 54-56.
[20] Thangavelu A, Vaidhyanathan A and Narendar R. Myositis ossificans traumatica of the medial pterygoid. Int J Oral Maxillofac Surg 2011; 40: 545-549.
[21] Ungari C, Quarato D, Gennaro P, Riccardi E, Agrillo A, Mitro V, Cascino F, Reale G, Rinna C and Filiaci F. A retrospective analysis of the headache associated with temporomandibular joint disorder. Eur Rev Med Pharmacol Sci 2012; 16: 1878-1881.
[22] Arima R, Shiba R and Hayashi T. Traumatic myositis ossificans in the masseter muscle. J Oral Maxillofac Surg 1984; 42: 521-526.
[23] Parnes EI, Hinds EC. Traumatic myositis ossificans of the masseter muscle: report of case. J Oral Surg 1965; 23: 245-250.
[24] Nemoto H, Sumiya N, Ito Y, Kimura N, Akizuki A and Maruyama N. Myositis Ossificans Traumatica of the Masticatory Muscles. J Craniofac Surg 2012; 23: e514-e516.
[25] Guarda-Nardini L, Piccotti F, Ferronato G and Manfredini D. Myositis ossificans traumatica of the temporalis muscle: a case report and diagnostic considerations. Oral Maxillofac Surg 2012; 16: 221-225.
[26] Manzano D, Silván Á, Saez J and Moreno JC. Myositis Ossificans of the temporalis muscle: Case report. Med Oral Patol Oral Cir Bucal 2007; 12: e277-280.
[27] Saka B, Stropahl G and Gundlach KK. Traumatic myositis ossificans (ossifying pseudotumor) of temporal muscle. Int J Oral Maxillofac Surg 2002; 31: 110-111.