

Heterotopic Bone Formation in the Temporomandibular Joint: Report of a Case

Mohammad Hassan Samandari¹, Amirhossein Moaddabi², Parisa Soltani³, Sahar Goli Boushehri^{4*} and Atena Shiva⁵

¹Assistant Professor, Dental Implants Research Center, Department of Oral and Maxillofacial Surgery, School of Dentistry, Isfahan University of Medical Sciences, Isfahan, Iran

²Postgraduate Student, Department of Oral and Maxillofacial Radiology, School of Dentistry, Isfahan University of Medical Sciences, Isfahan, Iran

³Postgraduate Student, Department of Oral and Maxillofacial Radiology, School of Dentistry, Isfahan University of Medical Sciences, Isfahan, Iran.

⁴Dentist, Dental Students Research Center, School of Dentistry, Isfahan University of Medical Sciences, Isfahan, Iran

⁵Assistant Professor, Department of Oral and Maxillofacial Pathology, School of Dentistry, Mazandaran University of Medical Sciences, Sari, Iran

***Corresponding Author:** Sahar Goli Boushehri, Dentist, Dental Students Research Center, School of Dentistry, Isfahan University of Medical Sciences, Isfahan, Iran.

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Abstract

Heterotopic bone formation or heterotopic ossification is a pathological condition in which bone forms in nonskeletal tissues. It may occur mainly due to trauma or previous surgical procedures. This case report presents two cases with heterotopic bone formation in temporomandibular joint due to traumatic injury.

Keywords: Temporomandibular Joint; Heterotopic Ossification; Computed Tomography

Introduction

Heterotopic bone formation or heterotopic ossification is a pathological condition in which bone forms in non-skeletal tissues [1]. There are three recognized etiologies of heterotopic ossification: traumatic, neurogenic, and genetic. Traumatic HO typically occurs following fractures, dislocations, operative procedures, and severe burns [2]. The most common areas involved are upper and lower extremities [3]. Formation of this ectopic bone in soft tissues is a result of presence of several factors; precursor cells with the potential to differentiate into bone and cartilage, a conductive tissue environment, and an initiator or inducing event such as trauma. Most commonly, the heterotopic bone is normal in quality formed through endochondral or intramembranous osteogenesis. The mechanism by which this heterotopic bone is formed is parallel with the events that occur during normal embryonic bone and skeletal formation, as well as those occurring in bone regeneration during fracture healing. The pathophysiology of heterotopic ossification is caused by dysregulation of cell fate determination and in appropriate induction of the bone formation program [4].

Bone formation outside the skeleton is a common finding on radiographs from patients who have undergone arthroplasty of the hip, knee, shoulder, or elbow [5]. It also can be a consequence of trauma and arthroplasty of the temporomandibular joint [6]. In general, heterotopic bone is rarely encountered in the maxillofacial region. Examples include ossification of the stylohyoid ligament, periosteal hemorrhage or infection, and myositis ossifications of the masseter muscle [7]. This case report presents a case with heterotopic bone formation in temporomandibular joint due to traumatic injury.

Case Presentation

A 28-year-old male was referred to Maxillofacial Surgery Clinic of Alzahra Medical Center in Isfahan, Iran with a chief complaint of facial asymmetry and pain and limitation in mouth opening. He stated that he has experienced these problems for 3 months. The patient gave a history of trauma in childhood. No additional finding was noted in the patient’s history.

Extraoral examinations demonstrated deviation into the left side in the mandible (Figure 1a). No tenderness or swelling was observed in palpation. Maximum interincisal distance was approximately 7 mm. Examination showed significant deviation during mouth opening that caused more significant asymmetry.

Intraoral examination was incomplete due to severe difficulty in mouth opening. However unilateral cross-bite on the left side was evident (Figure 1b).

Computed tomography (CT) scan were performed. Based on coronal CT images (Figure 1c), a radiopaque mass with a margin of cortical bone can be seen superomedial to the right mandibular condyle. Treatment was planned for surgically removing this mass in the temporomandibular region. The surgery was performed under general anesthesia. Preauricular incision (Al-Kayat Bramley modification) allowed access to the condyle and its medial space. The mass was exposed and removed from the region (Figure 1d). Finally, the incision was closed in layers. The mass had a size of 1 × 2 cm. The result of the histopathological examination of the sample was normal cancellous and cortical bone suggesting heterotopic bone formation. 8-week postoperative follow-up of the patient revealed normal mouth opening and alleviation of the temporomandibular pain. Also, the asymmetry was resolved partially after the surgery (Figure 1e).

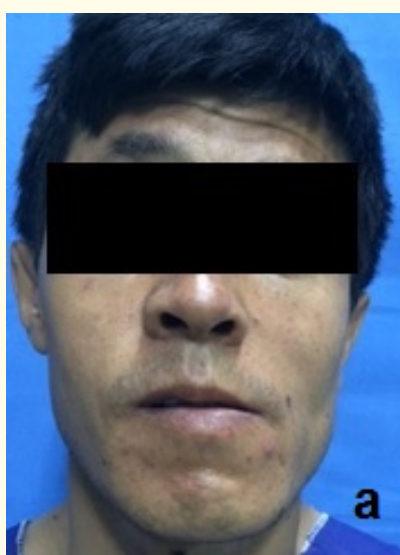


Figure 1: (a) patient photograph depicting significant facial asymmetry and chin deviation to the left side; (b) intraoral photograph showing unilateral cross-bite on the left side; (c) Coronal CT image demonstrating the radiopaque mass in the superomedial aspect of the right mandibular condyle; (d) intraoperative photograph showing the heterotopic bone mass; and (e) follow-up photograph demonstrating normal mouth opening.

Discussion

It is suggested that following trauma or operation in the joint region, pluripotential mesenchymal cells are stimulated to differentiate into osteoblastic and chondroblastic stem cells [8]. The mechanism of this stimulation is unknown, but a factor in the bone matrix is the most likely agent [9]. Other theories have also been reported [10] and the causes may be multifactorial. In summary, heterotopic bone formation is a result of dysfunction in the intricate, dynamic system of bone formation and remodeling of the body.

Any direct or indirect trauma to the joint may alter the existing balance of temporomandibular joint and cause lead to problems such as effusion, hemarthrosis, dislocation, internal de-arrangement, fibrous adhesion, ankylosis, fracture and limitation or deviation of jaw opening [11]. Bone formation is a rare reaction of the affected tissue after traumatic injury. Heterotrophic bone can form in any part of body. Hips, shoulders, elbows and knees are the joints most commonly affected by heterotopic ossification [12]. Temporomandibular can also be involved after arthroplastic surgery. In the study performed by Lindqvist, *et al.* [6] 52% of the temporomandibular joints developed heterotopic ossification after alloplastic temporomandibular joint reconstruction. However, formation of bone within the TMJ after head trauma is rare.

This report, presented a case of heterotopic bone formation secondary to trauma in childhood. It had caused significant deformity of the face and chin deviation. Also it had impaired the patient's mouth opening due to pain and limitation. Surgical resection of the heterotopic bone was indicated due to pain and loss of function. Recurrence of heterotopic bone formation within the joint is significant [13] and treatment must be performed meticulously in order to prevent it. Prophylactic measures available for prevention of recurrence of heterotopic ossification include postoperative radiotherapy and non-steroidal anti-inflammatory drugs, particularly Indomethacin. In the past, bisphosphonates were also used for prophylaxis, but were abandoned after reports of their recurrence after discontinuing the use of drug [2]. The decision to provide prophylactic treatment must balance a patient's risk of heterotopic bone formation against the potential risks of preventive treatment. In the case of the present patient, follow-up sessions were planned to monitor the patient for possible recurrence.

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