

Multifocal Infratemporal Heterotopic Ossification: An Unusual Clinical Feature: A Case Report

Ali Hossein Mesgarzadeh* Ali Mortazavi*

* Department of Oral & Maxillofacial surgery, Tabriz University of Medical Sciences, Tabriz, Iran

Abstract: True ossification of the face spaces is a very rare clinical entity. We present a rare case of unilateral ossification of infratemporal space the in an 11-years old male with swelling in the right temporal and orbital area. His medical history was unremarkable. Radiographic evaluation with spiral CT scan revealed separated ossified particles inside of infratemporal space. After 2 separate procedure all bony particles were removed at operation room. Pathologic evaluation showed normal bone structure. Postoperative period was uneventful.

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Introduction

Heterotopic ossification (HO) is the presence of bone in soft tissue where bone normally does not exist. It is a pathological condition in which bone arises in tissues not in the osseous system and in connective tissues usually not manifesting osteogenic properties. It is a relatively rare, well-defined entity¹. Early description of heterotopic ossification was performed by Reidel in 1883 and Dejerne A and Ceillier A (1918). They reported ectopic ossification among soldiers that had experienced spinal cord trauma during World War⁷.

In some literatures it widely known as miositis ossificans. It is a reactive bone producing inside of muscles or other connective tissues. When occurring in submucosal or subcutaneous fat it is often referred to as paniculitis ossificans or fasciitis ossificans.

Miositis ossificans is divided broadly into miositis ossificans progressive (MOP) and myositis ossificans traumatic (MOT)^{1,4}.

A more serious and extensive form is myositis ossificans progressive or fibrous dysplasia progressive that involves skeletal muscles, tendons, fascia aponeuroses and ligaments and it has an autosomal dominant genetic pattern.

The progressive form is also associated with assorted congenital anomalies especially in toes and thumbs with ankylosis of digits and a history of joint pain and swelling. Multiple and massive heterotopic ossification and calcification may develop in several other conditions such as fibro-osseous pseudotumor, florid reactive periostitis and bizarre parosteal osteochondromatous proliferation that are probably variation of heterotopic ossification.

The formation of ectopic bone may be experimental induced in any soft tissues through implantation of demineralized bone or dentine contained bone morphogenetic protein⁵.

Heterotopic ossification may occur after acute or chronic trauma to a muscle. The musculatures of head and neck region are an uncommon site for this phenomenon but occasional cases have occurred in the masseter and other facial muscles.

Majority of authorities presume these lesions originate from an intramuscular hematoma with metaplastic transformation of pleuripotential stromal cells but traumatic implantation of periostum is another local explanation for selected cases.

Heterotopic ossification of the head and neck region typically occurs in the masseter muscle after single severe injury¹⁰. There is no gender predilection.

Here we report a case of multiple heterotopic ossifications inside of infratemporal space without any history of trauma and functional disabilities.

Case Report



Figure 1. Asymmetry of temporal region due to expansion of right zygomatic arch

An 11-year-old male patient referred to the oral & maxillofacial surgery department clinic for his swelling in the right temporal and orbital area. He had also complained about slow growing asymmetry of his face and occasional preauricular pain. Swelling located around his right zygomatic arch and lateral to orbital rim without any visual or auditory disturbance. (Figure 1)

Palpation of swelling area was non tender without any pitting edema. Overlying skin texture and color was normal. Dental examination showed normal deciduous and permanent teeth. Intraoral examination did not reveal any bulging and swelling around zygomatic buttress. Overlying mucosa was normal. Patient did not present any history of trauma, abscess

formation or any surgical intervention in affected area. His laboratory tests (CBC, biochemistry, ca, p, Alkaline Phosphatase) were in the normal range. No unusual sign & symptom were found in the other part of body and his medical condition was unremarkable. He did not present any restriction on his mouth opening. Inter incisal distance was more than 35mm. Fine needle biopsy was unremarkable. Radiographic evaluation with spiral CT scan revealed separated ossified particles inside of infratemporal space and medial to zygomatic arch without any union to adjacent bony structures and surrounded by well-defined radiolucent space (Figure 2). Patient was scheduled for incisional biopsy.

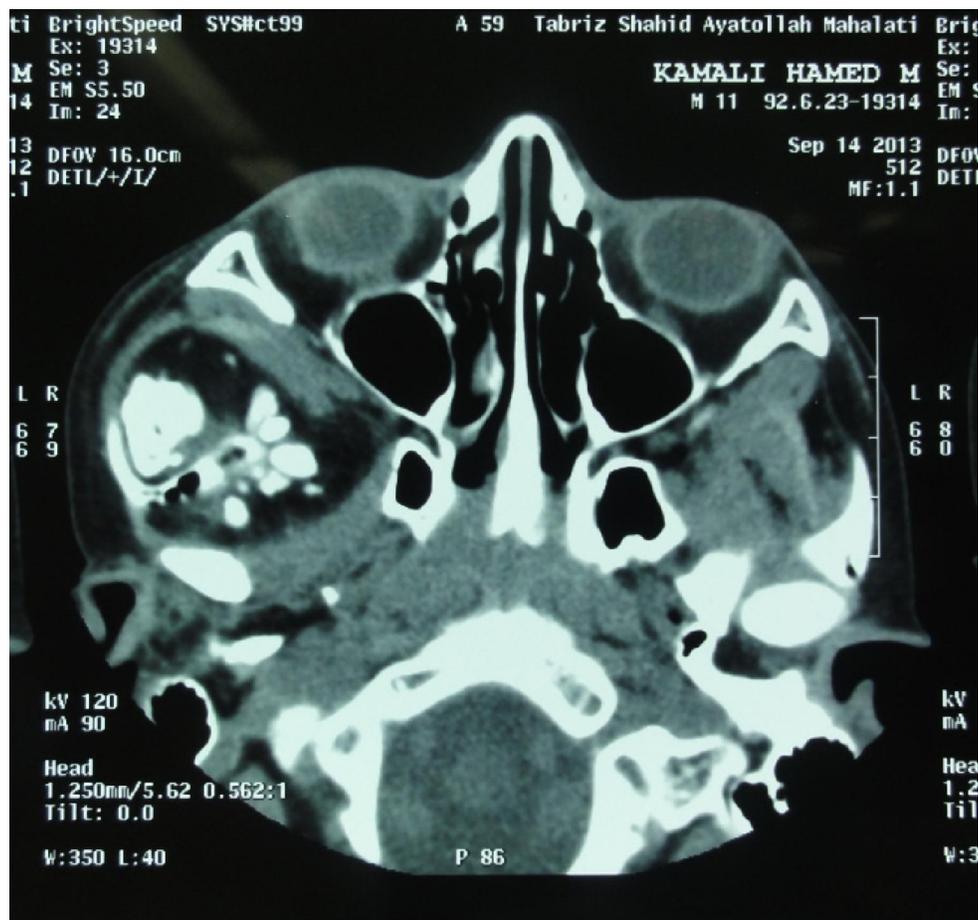


Figure.2. Axial view of midface shows multifocal ossified particles inside of infratemporal space

By lateral eyebrow approach surgical team tried to have an access to the ossified particles and two separate portions were obtained from medial aspect of zygomatic arch. Bony particles were surrounded by lipomatous tissues without any fusion to zygomatic arch or temporal bone. Histopathologic examination of the mass revealed presence of normal mature lamellar bone and some lobulated adipose tissues around

(Figure 3 & 4). After first procedure another CBCT (Figure 5) was performed to localize remaining particles and make the best surgical access. At the second surgical intervention we used preauricular approach to access remaining particles. Four bony particles were removed (Figure 6). Postoperative period was uneventful.

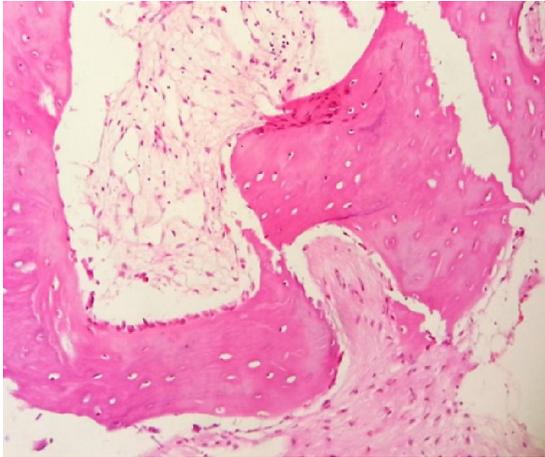


Figure.3.Normal bone structure showing

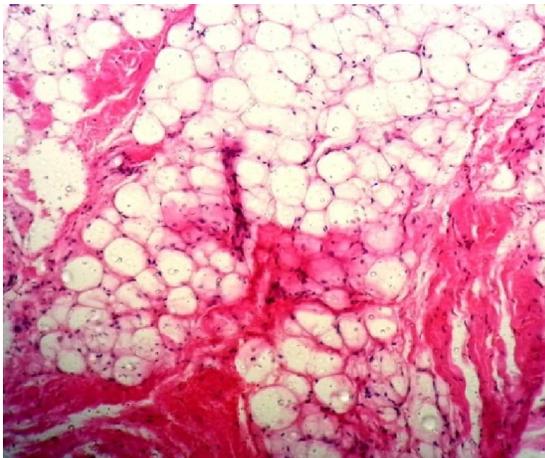


Figure.4.Structure of normal adipose tissue Haversian canals and osteocytes



Figure.5. CBCT shows bony particles at Medial side of temporal portion of zygomatic arch



Figure 6. Four bony particles were removed after second surgical intervention.

Discussion

Heterotopic ossification (HO) is the formation of mature lamellar bone in soft tissue. It forms outside of the joint capsule and periosteum. It is usually associated with symptoms of pain, hypomobility, tenderness or swelling but in some situation can remain asymptomatic^{1,7}. Symptoms depend on the size and proximity to certain anatomical site.

The most common cause of HO is trauma¹, although, atraumatic HO has also been reported. Trauma could be in the form of musculoskeletal injury, surgical trauma, or warfare injuries. Other causes of HO are hereditary⁴, burn¹⁰ and neurogenic injury¹. HO can occur in muscle, adipose or connective tissue.

Very few cases of HO have been previously reported in the maxillofacial region. Miositis ossificans has been reported in muscles of mastication and a case of atraumatic HO in scalp following transposition of temporalis muscle in the cheek for the facial paralysis^{11,12,8} and paniculitis ossificans in submental region⁹, have been reported.

Successful treatment of HO relies on understanding of the risk factors, the pathophysiology and anatomy of the region. Different modalities have been proposed for treatment or prevention of HO such as surgical intervention³, Radiotherapy², NSAID therapy⁶ and Bisphosphonate therapy⁹.

Our case is unique as he presents HO at a very young age, without history of trauma, in a very unusual site, and in connective tissue. There is no unusual finding or growth disturbance at the mandibular bone and its function.

Based on the clinical and radiological findings a differential diagnosis should include myositis ossificans circumscripta, myositis ossificans progressive, osteoma, nodular fasciitis, osteosarcoma and chondrosarcoma, Slowly calcifying lesions synovial sarcoma, rhabdomyosarcoma, and malignant fibrous histiocytoma should also be included¹². The most important pathology to be excluded is osteosarcoma. Histopathologically presence of mature lamellar bone confirms the diagnosis of HO.

Myositis ossificans traumatica (MOT) was described initially by Thoma in 1958 as a condition generally caused by calcification and progressive ossification of an intramuscular hematoma after trauma^{1,5,12}. A review of literature showed MO is a very rare clinical entity in maxilla facial region. To the best of our knowledge about 30 cases have been reported in maxillofacial field.

Surgical trauma, chronic infection like pericoronitis, tooth extraction and acute trauma have been proposed as an etiologic factor for this pathologic condition.

The highest incidence of MO involving maxillofacial muscles was in masseter but very few case reports have described in temporal, medial and lateral pterygoid muscles. Heterotopic ossification without history of trauma is extremely rare and it can originate by non traumatic myositis ossificans. According to Rattan the pathogenesis of MOT remains uncertain although many authors consider it as an aberrant physiological healing⁵. Intramuscular hemorrhage, which is followed by the exuberant formation of vascular granulation tissue^{4,5,8}, maturation of granulation tissue results fibroblastic proliferation with progression to the synthesis of osteoid and chondroid.

The unique histopathologic feature of our case is the presence of multiple and separated ossification foci inside of lipomatous tissues without any union to each other and adjacent bones like zygomatic arch and coronoid process. Another unique entity is lack of hypomobility in his jaw movement. Enlargement of ossified particles may lead to swelling of face and zygomatic arch expansion in very young age. Well defined enlargement of sigmoid notch due to the presence of heterotopic ossification and surrounding of bony segments by lipomatous tissue make it interesting, on the other hand it can explain non union of bony particles to the adjacent skeletal tissues. We propose that ectopic extension of buccal fat pad into the infratemporal and sigmoid notch and subsequent

mandibular function and its mobility could be a reason for micro hemorrhage islands inside of adipose tissue and finally multiple bone formation from collected hematoma.

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