International Surgery Journal
Anehosur V et al. Int Surg J. 2022 Oct;9(10):1755-1759
http://www.ijsurgery.com
pISSN 2349-3305 | eISSN 2349-2902

DOI: https://dx.doi.org/10.18203/2349-2902.isj20222602

Case Report

Surgical management of gigantiform osteoma of angle causing facial deformity

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Received: 09 August 2022
Revised: 07 September 2022
Accepted: 16 September 2022

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ABSTRACT

Osteoma is a rare, benign lesion wherein deposition of newly formed bone results in a tumor mass. Variants of osteomas can develop either as a peripheral mass arising from the periosteum or a central mass arising from the endosteum. Peripheral ones are usually asymptomatic and larger lesions primarily cause facial asymmetry. Central osteoma, in most cases results in cortical expansion and root displacement in some. Central osteomas pose a diagnostic dilemma and need to be discriminated from other similar lesions such as condensing osteitis, odontoma, ossifying fibroma, osteoblastoma. Isolated single osteomas are usually non-syndromic whereas, multiple osteomas are hallmark features of Gardner syndrome. Craniofacial bones such as paranasal sinuses and jawbones are frequently affected, although rare cases in other bones and soft tissues have been reported. This is a presentation of osteoma in the rare angle of mandible region in a young patient which was managed by intraoral approach.

Keywords: Bony swelling, Facial deformity, Surgical excision

INTRODUCTION

Osteoma is a benign overgrowth of bone, usually affecting the craniofacial region. It is characterized by abnormal proliferation of either compact or cancellous bone. It is a rare lesion and affects about 0.4-4% of the population.[1] They are solitary masses attached to the cortical plate or arise as a central lesion from the endosteal bone surface. Osteomas involve the craniofacial bone, commonly the paranasal sinuses and jawbone. Larrea et al in their study, noted that 49% of the osteomas were of the peripheral type followed by central osteoma constituting 29%. Also, in their series, 21% of the osteomas were located in the paranasal sinuses, of which the frontal sinus was most frequently affected followed by maxillary and ethmoidal sinuses.[2] Osteomas are usually slow-growing and asymptomatic mass. However, symptoms such as facial asymmetry, malocclusion, difficulty in swallowing and mastication, trismus and respiratory distress have been reported.[1] These symptoms are dependent on the size, location, and direction of growth of the lesion. The etiology of osteoma remains controversial, and several hypotheses have been suggested for the same. The 3 main theories proposed for the etiopathogenesis of osteoma are developmental defect, reactive lesion triggered by infection or trauma, neoplasm. Osteomas are unlikely to be developmental defects as they affect adults more than children and adolescents. The slow-growing nature of the lesion goes against the neoplastic nature theory. Osteomas are usually

found adjacent to areas with a previous history of trauma. This validates the theory of traumatic origin. The purpose of this case report is to present the clinical, radiographic, histologic, and surgical management of a peripheral osteoma in the angle of mandible.

CASE REPORT

A-24-year-old male reported Craniofacial center with a history of painless swelling over the right angle of mandible region for 6 years. There was no previous history of trauma. There was no history of difficulty in mastication, mouth opening was adequate, and no paresthesia was elicited. No significant personal or medical history was noted, and his blood investigation were within normal limits.

On extraoral examination, there was gross facial asymmetry with swelling noted over the right side of the face (Figure 1-3). A well-circumscribed mass was noted in the right mandibular angle region. The swelling extended from the parasymphysis region up to the postauricular region. The swelling did not cross the midline and no displacement of structures to the contralateral side was seen. The swelling was well-defined, hard in consistency and non-tender. The overlying skin appeared normal with no associated signs of inflammation. There was no local rise in temperature and no pulsation or thrill was felt. No evidence of lymphadenopathy or intraoral cortical expansion was present. There were no other similar lesions was noted in other regions. The orthopantamogram (OPG) radiograph revealed a well-circumscribed, radiopaque mass attached to the buccal surface of the angle and ramus region of the right mandible extending up to the inferior border (Figure 4). Routine blood investigations were normal including serum-calcium, phosphate, alkaline phosphatase, Thyroid stimulating hormone, Vitamin D, Parathyroid hormone to rule out fibro-osseous lesions and oncogenic osteomalacia. The planned procedure was surgical excision by an intraoral approach, exposure of entire bony mass and resection of followed by recontouring the mandible and primary closure.

The lesion was exposed through an intraoral incision under general anesthesia. The mass was spherical in shape, the surface was convoluted. The cleavage was identified which separated the mass from the adjacent healthy normal bone. Using a 703 straight fissure bur, grooves were made along the cleavage. A reciprocating saw was used to deepen the grooves. With the help of an osteotome, the mass was then completely separated from the underlying bone (Figure 5). Shaping and recontouring of the angle and the inferior border was done using a rosette bur (Figure 6 and 8). The postoperative course was uneventful. The specimen (Figure 7) was then sent for histopathological examination which revealed, dense mature bone trabeculae with numerous osteocytes within lacunae suggestive of an osteoma.

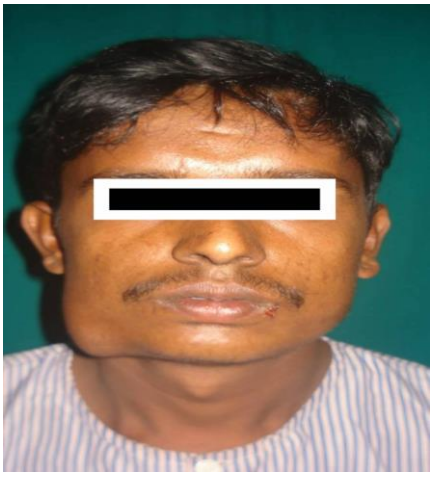

Figure 1: Frontal profile showing facial asymmetry on right side.

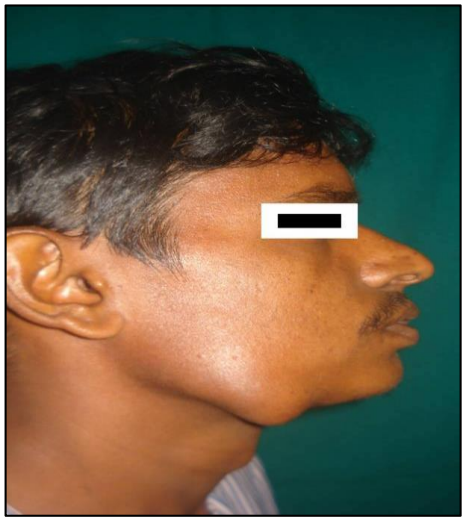

Figure 2: Lateral profile extent of swelling over the right angle.

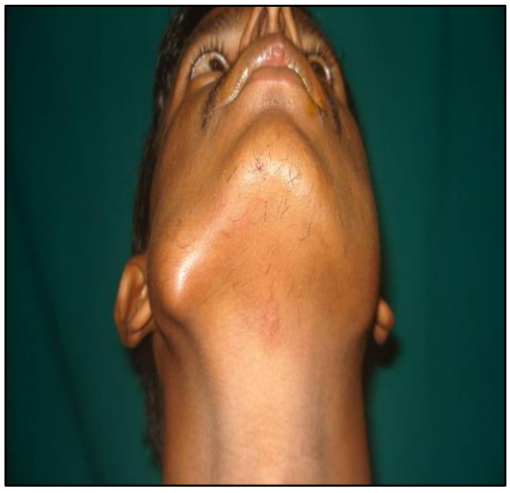

Figure 3: Worms view showing neck soft tissue expansion due to swelling.

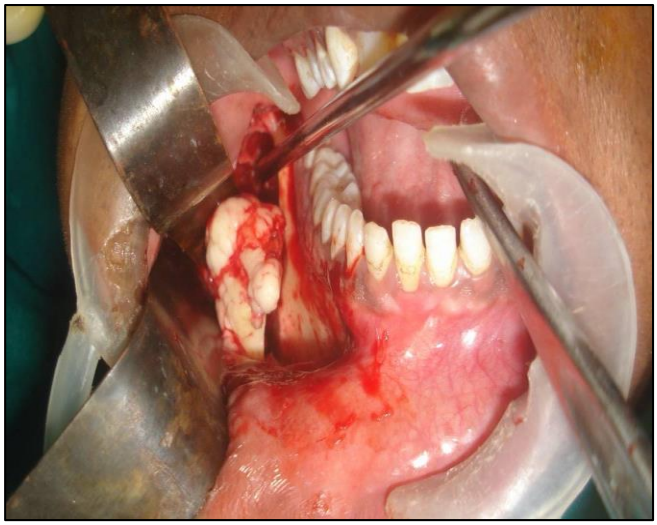

Figure 4: Intraoral obliteration of buccal sulcus.

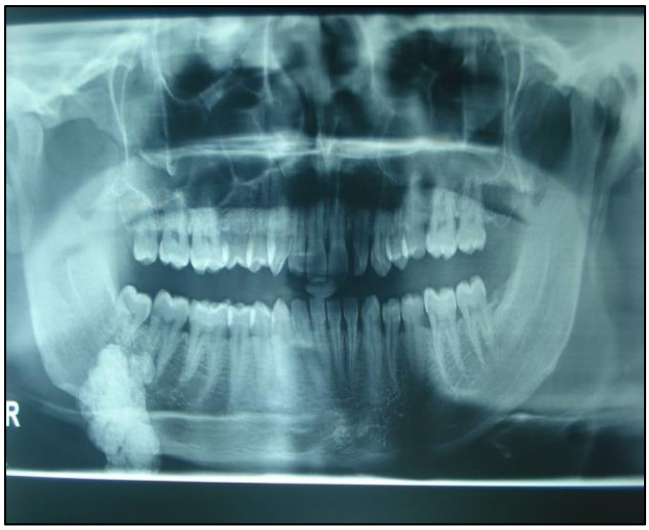

Figure 5: Preoperative panoramic radiograph.

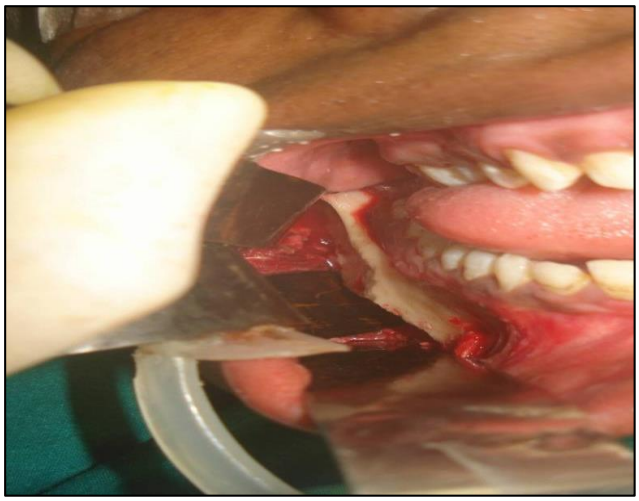

Figure 6: Intra operative osteoma separated from the underlying bone.

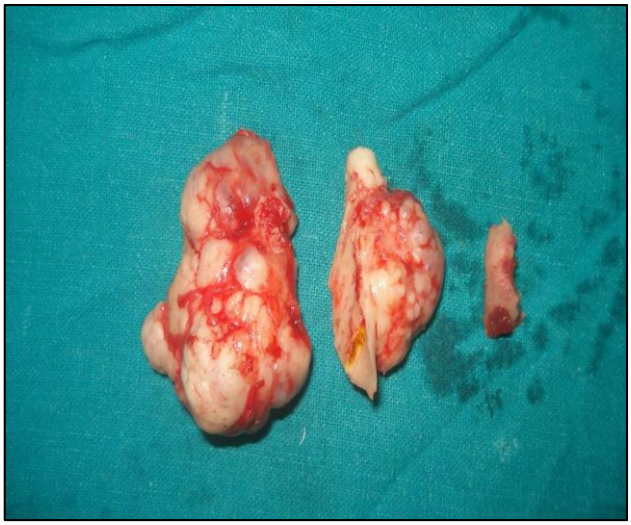

Figure 7: Excised specimen.

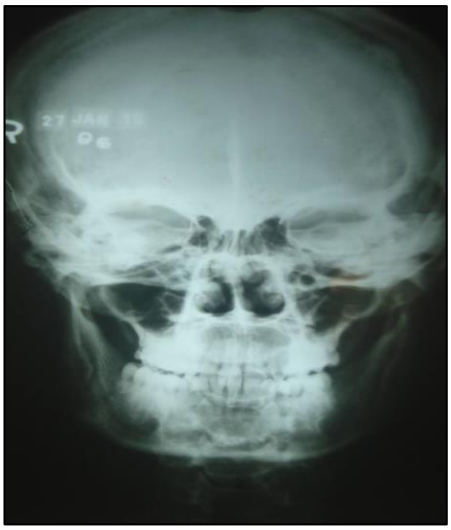

Figure 8: Postop X-ray showing symmetrical excision of pathology.

DISCUSSION

Osteoma is a slow-growing osteogenic lesion characterized by the proliferation of mature bone tissue-compact or cancellous. They originate from the membranous bones of the craniofacial skeleton and rarely affect the other bones. Frontal, ethmoidal, and maxillary sinuses are common sites for peripheral osteomas.[3] The diagnosis of paranasal sinus osteomas is delayed as the tumors are usually asymptomatic in the initial stages. As the lesion enlarges, it impinges the adjacent structures such as sinus ostium and orbital and intracranial structures. Frontal sinus osteoma is commonly associated with a frontal sinus infection. Forehead contour deformity is noted in lesions with anterior extension. Meningitis, seizure, or hemiparesis are noted in posterior intracranial extension.[4] Ethmoidal osteomas present with symptoms earlier than those in frontal sinuses.[5] The symptoms

usually include pain, facial deformity, nasal obstruction, persistent sinusitis, diplopia and epiphora. Other sites include orbit, temporal bone, zygoma and rarely jaws. Primary orbital bone osteomas are rare. Secondary orbital extension from the paranasal sinuses is reported with an incidence of 0.9-5.1%.[6] Orbital involvement results in symptoms like pain, diplopia, epiphora, globe displacement, dacryocystorhinitis and vision loss. Osteomas are commonly located in the frontal bone (28.57%), a mandible (22.85%) and maxilla (14.28%).[7]

The mandible is the most affected anatomic region of the jaw, specifically the angle and inferior border, followed by the alveolus and the maxilla. Multiple lesions are often associated with Gardner syndrome. It is an autosomal dominant genetic disorder characterized by a triad of multiple intestinal polyps, skeletal abnormalities and impacted/supernumerary teeth. Other features of this syndrome include epidermal cysts and cutaneous fibromas. The risk of malignant transformation of the intestinal polyps increases as they approach the age of 40. This syndrome was ruled out in our patient with negative clinical history.

Osteoma though benign, infiltration of interdental bone and abnormal histologic bone structure suggests the neoplastic nature of the lesion. Osteomas can be of three variants: central, peripheral, or extra skeletal. Peripheral osteoma appears on the bone surfaces whereas the extra skeletal is immersed in the soft tissues. Osteomas are usually long-standing and are asymptomatic in the initial stages and may be detected accidentally in routine radiographs. However, in later stages, bone deformation and/or compression of adjacent structures are seen.[2] Central osteomas are even slower compared to peripheral osteomas to present clinical manifestations, as considerable growth must occur before the evident expansion of the cortical plate occurs. The exact pathogenesis of osteoma is uncertain. Endocrine disturbances have been considered as a possible etiologic factor.[8] The formation of the osteoma could be a result of either trauma or reaction to inflammation, infection or hamartomatous events.[9] The combination of trauma along with muscle traction, best explains the pathogenesis of osteoma. The inner surface of the bone is lined by endosteum, comprising osteoprogenitor cells. These cells along with periosteum aid in nutrition, bone growth and repair. Trauma causes subperiosteal bleed and oedema. The muscle traction further elevates the periosteum. The osteoprogenitor cells in endosteum, begin to differentiate into osteoblast-trauma trauma. This initiates an osteogenic reaction which is further enhanced by constant muscle traction.[3] Osteomas appear at any age group but most commonly affect the younger age group as seen in this report without any gender predilection.[10] However, Sayan et al reported a male-to-female ratio of 1.9:1.

Peripheral osteomas manifest as unilateral, well-circumscribed masses, sessile with varying diameters ranging from several millimetres to centimetres. However, the true size of the lesion may be difficult to determine when part of the lesion is within the bone. There is no age or gender predilection. The patient is usually asymptomatic although he might complain of facial asymmetry or interferences in dental occlusion in later stages. Radiographically, osteoma appears as a radiopaque, circumscribed mass with a thin radiolucent zone surrounding the radiopaque mass. It may have a central trabecular or a sclerotic pattern.

The differential diagnosis includes periosteal ossifying fibroma, chondroma, exostoses and osteoblastoma, fibrous dysplasia.[9] Exostoses such as tori are true hamartomas, and they usually remain dormant after puberty. They are reactive or developmental in origin and not considered as true neoplasm. Osteomas, on the other hand, are considered true neoplasm and they continue to grow even after the completion of the skeletal growth.[11] Osteoblastoma is fast-growing in nature and is usually associated with pain. They arise from the medullary bone and cause cortical expansion. Microscopically, they show abundant osteoid trabeculae anastomosing in a loose fibrovascular connective stroma. Osteomas resemble normal compact or cancellous bone with fibrofatty bone marrow.[10]

Surgical intervention is recommended in only symptomatic patients. If the osteomas are asymptomatic and relatively smaller in size, the patient can be managed non-invasively with periodic follow-up. However, if the osteoma results in severe facial disharmony, exuberant growth rate or functional disorders, surgery is recommended.[9] Surgery involves complete excision of the lesion from the base. However, partial excision of the lesion is recommended when preservation of bone tissue is essential for prosthetic rehabilitation.[12]

Diagnostic workup can be aided with use of panoramic radiographs/computed tomography. Tomography is considered a superior modality as it provides better resolution on size, location and anatomical relation with adjacent structures. The definitive diagnosis however requires an appropriate correlation between clinical, radiographic and histologic findings as numerous osteoblastic lesions have overlapping features. Recurrence after resection is rare and malignant transformation has not been reported in literature.[12]

CONCLUSION

Osteoma although are commonly occurring in the facial bone, the one reported here adds to the similarly reported, but the basic steps of understanding the pathology like clinical and radiographic presentation, surgical approach and histopathogical examination remain the cornerstone for the successful management.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Anehosur V, Kotian D, Kumar N. Surgical management of giganti-form osteoma of angle causing facial deformity. Int Surg J 2022;9:1755-9.