



Peripheral osteoma associated with peripheral ossifying fibroma: A case report

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ARTICLE INFO

Article Type: Case Report

Received: 2 Jan. 2018

Revised: 19 Feb. 2018

Accepted: 10 Mar. 2018

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ABSTRACT

Osteoma is a benign osteogenic neoplasm composed of mature compact or cancellous bone and classified as central, peripheral or extra-skeletal. Peripheral type of osteoma is rare. The maxillary sinuses are the most common sites of involvement. It is rare in jaws. Peripheral ossifying fibroma is a reactive lesion of oral tissues, associated with local factors such as trauma or irritation factors. In this paper, we presented the first case of solitary peripheral osteoma associated with peripheral ossifying fibroma. The lesion was located in the lingual surface of the left posterior area of mandible in a 32-year-old woman. The lesion was removed surgically, and no recurrence has been observed after six months follow up.

Keywords: Peripheral osteoma, Peripheral ossifying fibroma, Mandible.

Introduction

Osteoma is a benign, slow growing asymptomatic osteogenic neoplasm composed of mature compact and/or cancellous bone. It is classified as central (endosteal), peripheral (parosteal, periosteal, or exophytic) or extra-skeletal (osseous choristoma osteoma) [1-3].

Although the pathogenesis of osteoma is controversial, it is thought that osteoma may be as true neoplasm, developmental anomaly or reactive lesion induced by trauma, muscle traction or infection [4,5]. Peripheral type of osteoma is rare. It is more common in craniofacial bones in

comparison with other bones [1,3,4]. The most common sites are paranasal sinuses followed by orbital wall, temporal bone, pterygoid process and external ear canal [3-6]. Although the prevalence of peripheral osteoma of jaws is quiet rare, the frequency of the lesion in mandible is more than maxilla, most often in the posterior body of mandible, condyle, angle, ascending ramus, coronoid process, anterior body, and sigmoid notch, respectively [4,6].

Although osteoma is seen in any ages, it mostly occurs in young adults and affects men and women equally [7]. It is usually seen as solitary lesion however multiple osteomas may be seen in association with Gardner syndrome [1].

Peripheral ossifying fibroma is a reactive lesion which is relatively uncommon [8]. It occurs as a result of trauma or irritation factors including micro organisms, plaque, calculus, poor restorations, and dental appliances [8,9]. The lesion is most common in young adults and has predilection to females [10]. The condition affects mainly the gingiva, especially the anterior region of the maxilla [9-11]. It appears as a solitary nodule with slow and progressive growth. Microscopically, peripheral ossifying fibroma is characterized by fibrocellular connective tissue, composed of plump ovoid or spindle-shaped fibroblasts with bony trabeculae and/or cementum-like material [11]. The purpose of this paper is to present the first case of large peripheral osteoma of the mandible associated with peripheral ossifying fibroma in a 37-year-old woman.

Case Report

A 32-year-old woman was referred to the oral and maxillofacial surgery department because of an exophytic lesion in lingual plate of the left side of mandible. She had been aware of the slow growing lesion that had increased in its size over the past four months. The lesion was not painful, and there was no record of bleeding. She had no previous systemic disease or allergic history. Intra oral examination revealed a large, well-defined, round lesion on lingual portion of left first mandibular molar measuring about 3cm in diameter and looks over growth of periodontium (Figure 1). The lesion was firm on palpation. The overlying oral mucosa was intact and normal in color. Cone beam computed tomography (CBCT) revealed a peripheral radio opaque lesion with well-defined border, attached to lingual portion of mandible in first molar location progressing longitudinal to first premolar in mesial aspect (Figure 2). Surgical excision of the lesion was done under local anesthesia (Figure 3). Microscopic examination showed a nodular mass composed of large pieces of bone trabeculae with extensive fibrofatty marrow in deep area which surrounded by dense fibrotic connective tissue (Figure 4a).

The connective tissue was highly cellular in some areas composed of plump mesenchymal cells with osteoid formation (Figure 4b). The lesion was covered by parakeratinized acanthotic stratified squamous epithelium. Various sized blood vessels, diffuse mild

chronic inflammatory cell infiltration were also seen in connective tissue. The patient presented for a post-operative visit 6 weeks after surgery, and the healing was progressing normally (Figure 5). There were no postoperative complications. There was no evidence of recurrence after six months follow up.



Figure 1. Intraoral view showing a well-defined, pedunculated lesion covered by normal oral mucosa. Note the attachment of the lesion to the lingual plate of the left posterior area of mandible.

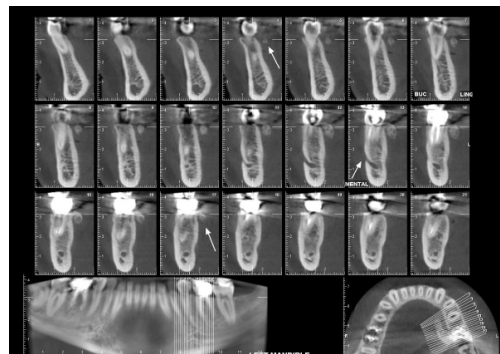


Figure 2. CBCT view of the lesion shows a round well defined radio opaque lesion. The only area of attachment is in lingual aspect of the first molar tooth.



Figure 3. Macroscopic view of the lesion shows nodular mass with bony hard consistency.

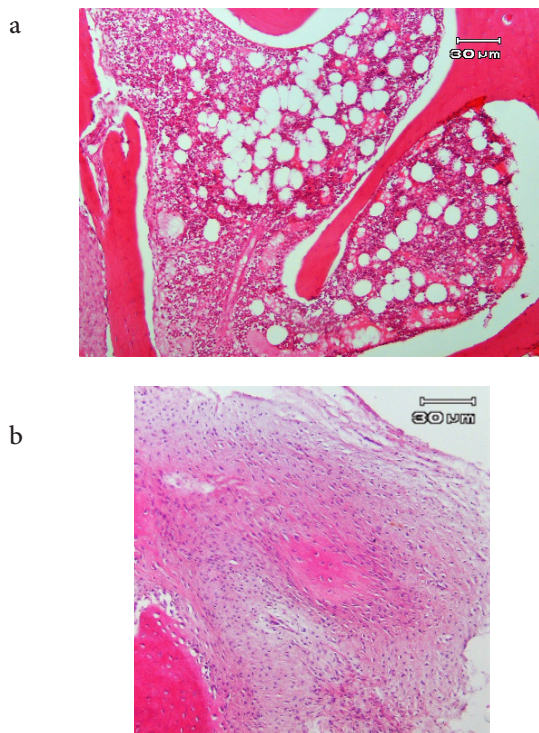


Figure 4. a) Microscopic features show lamellar compact bone with extensive fibrofatty marrow. b) Plump mesenchymal cells with osteoid formation.



Figure 5. Normal healing is evident in post-surgical follow up 6 weeks after treatment.

Discussion

The etiology of osteomas and its real prevalence are still unknown. Some investigators consider it as a true neoplasm; while others accept it as a reactive lesion or developmental anomaly. Woldenberg et al suggested that some peripheral osteomas may be reactive rather than neoplasms, probably associated with trauma [12]. However, in the case has been described in this paper, there was no information as the possible cause, no history of previous trauma or infection. Peripheral ossifying fibroma arises from periodontal ligament cells, and local factors such as trauma, dental biofilm, calculus, and microorganisms have been associated with this condition [11].

Osteomas are often asymptomatic and detected incidentally in routine clinical and radiographic examination. As in our case, the lesion had not any evidence of pain or significant symptom. However, based on the location of the tumor, secondary problems with occlusion and functional may occur [1]. Panoramic radiography or computed tomography is used for imaging [2]. However, as demonstrated in this case, the CT is the best imaging modality for determining the location and real extension of the lesion [5]. An oval, radiopaque, well-defined mass attached by a broad base to the cortical bone is a classic radiographic hallmark of peripheral osteomas [2]. Pedunculated peripheral osteoma has a narrow contact area between the lesion and the compact bone [1]. In our case, the lesion consisted of dense, uniformly opaque compact bone with a narrow pedicle demonstrated by CT.

Some lesions are considered in the differential diagnosis of peripheral ossifying fibroma including exostoses, erupted odontoma and peripheral ossifying fibroma. Exostoses are bony outgrowth that arise from bone surface. Unlike osteomas, their growth usually stops after puberty. An erupted odontoma appears as a well-defined radiopaque lesion. The density of odontoma is similar or greater than density of tooth structure. Also narrow radiolucent rim is surrounded the lesion unlike osteoma [2]. We described an unusual presentation of peripheral osteoma due to coincidence with peripheral ossifying fibroma, unusual because of its correlation with a reactive lesion that has not been reported until now. Due to persistence of one solitary lesion in this described case, Gardner's syndrome was not considered.

The confirmatory diagnosis of these lesions is usually achieved by histopathological examination. Peripheral ossifying fibroma is highly cellular mass of connective tissue which shows large number of plump fibroblasts in the fine fibrillar stroma. Mineralized material consisting of mature, lamellar or woven osteoid, cementum-like material and also lamellar or dystrophic calcifications can be seen in peripheral ossifying fibroma.

It seems that removal of an asymptomatic peripheral osteoma is not generally necessary. Surgical intervention is indicated only in large lesions [12]. However, Horikawa et al claimed that the treatment choice for osteoma is surgery [5]. However, we agree with him based on our unusual demonstrated case considering accompaniment peripheral osteoma and a reactive lesion; peripheral ossifying fibroma. This accompa-

niment has been achieved by surgical excision of the lesion.

Although the recurrence of peripheral osteoma is rare but the recurrence rate of peripheral ossifying fibroma has been reported approximately 20% [11]. So it is clear that to minimize treatment failure in this lesion, it must be removed completely with removal of other source of irritants. As we mentioned, there was no evidence of recurrence in our case after six months follow up. There is not any report of peripheral osteoma undergoing malignant transformation.

Conclusion

Based on our knowledge, this is the first report of peripheral osteoma associated with peripheral ossifying fibroma on the lingual surface of the mandibular body. Although recurrence of peripheral osteoma after surgical excision is extremely rare, however; it is better to follow up the lesion after surgical excision.

Conflict of Interest

There is no conflict of interest to declare.

References

- [1] Sasidharan A, Gohil A, Koshy S, Kumar Gupta A. Bony tumour in an unusual location on the mandible. *Indian J Plast Surg.* 2015 May-Aug; 48(2): 200–203.
- [2] Kshirsagar K, Bhate K, Pawar V, Kumar S, Kheur S, Dusane Sh. Solitary Peripheral Osteoma of the Angle of the Mandible. *Case Reports in Dentistry* 2015, 430619, 4 pages.
- [3] Singhal P, Singhal A, Ram R, Gupta R. Peripheral osteoma in a young patient: A marker for precancerous condition?. *J Indian Soc Pedod Prev Dent* 2012; 30:74-7.
- [4] Bulut E, Acikgoz A, Ozan B, Gunhan O. Large peripheral osteoma of the mandible: A case report. *Int J Dent* 2010; 2010:834761.
- [5] Horikawa FK, de Freitas RR, Maciel FA, Gonçalves AJ. Peripheral osteoma of the maxillofacial region: a study of 10 cases. *Braz J Otorhinolaryngol.* 2012; 78(5):38-43.
- [6] Sayit AT, Kutlar G, Idilman IS, Gunbey PH, Celik A. Peripheral osteoma of the mandible with radiologic and histopathologic findings. *J Oral Maxillofac Radiol* 2014; 2:35-7.

- [7] Hemant Shakya. Peripheral Osteoma of the Mandible. *J Clin Imaging Sci.* 2011; 1: 56.
- [8] Sairam V, Padmaja K, Praveen Kumar B, Naresh G, Vikas Reddy G. Peripheral Ossifying Fibroma: A Case Report. *Advances in Dentistry and Oral Health.* 2016;1(3).
- [9] Bhasin M, Bhasin V, Bhasin A. Peripheral Ossifying Fibroma. *Case Reports in Dentistry* 2013, 497234, 3 pages.
- [10] Mohiuddin Kh, Priya N.S, Ravindra Sh, Muthy S. Peripheral Ossifying Fibroma. *J Indian Soc Peridontol.* 2013; 17(4):507-9.
- [11] El Achkar VNR, Medeiros RDS, Longue FG, Anbinder AL, Kaminagakura E. The role of Osterix protein in the pathogenesis of peripheral ossifying fibroma. *Braz Oral Res.* 2017 Jul 3;31:e53.
- [12] Woldenberg Y, Nash M, Bodner L. Peripheral osteoma of the maxillofacial region. Diagnosis and management: a study of 14 cases. *Med Oral Patol Oral Cir Bucal.* 2005 Jul 1; 10Suppl 2:E139-42.

Please cite this paper as:

Safarian M, Derakhshan S, Moradzadeh Khiavi M, Baghi A; Peripheral osteoma associated with peripheral ossifying fibroma: A case report. *J Craniomaxillofac Res* 2018; 5(2): 91-94