Simple bone cyst of mandible mimicking periapical cyst

Charan Babu HS,[1] Bhagawan Das Rai,[1] Manju A. Nair,[1] Madhusudan S. Astekar[2]

[1]Department of Oral and Maxillofacial Surgery; [2]Department of Oral and Maxillofacial Pathology, Pacific Dental College and Hospital Debari, Udaipur (Rajasthan), India

Abstract

Simple bone cysts (SBC) are pseudocysts occurring less commonly in the maxillofacial region. The uncertain and unclear etiopathogenesis led to numerous synonyms to refer this particular cyst. These cysts are devoid of an epithelial lining and are usually empty or contain blood or straw-colored fluid. In jaws initially it mimics a periapical cyst and later can lead to cortical bone expansion warranting for radical approach, which is seldom required. SBC is predominantly diagnosed in first two decades of life. Here we report a case of solitary bone cyst mimicking a periapical cyst of a mandibular molar in a 37-year-old patient.

Introduction

Simple or solitary bone cyst (SBC) is a less common pseudocyst of jaws, representing approximately 1% of all jaw cysts.[1] Lucas and Blum in 1929 for the first time described SBC as a separate disease entity.[2] According to WHO this cyst is defined as an unusual, benign, asymptomatic, slow growing, non-expansile, intra-osseous, empty or fluid filled cavity having a tenuous lining of connective tissue with no epithelium.[3] Most of times the diagnosis of SBC prior to surgery is difficult to a general dental practitioner as the lesion can have different presentation in various regions of the jaws. For these reasons, Rushton[4] adopted the following criteria for establishing diagnosis: i) a single lesion; ii) no epithelial lining; iii) no infection; iv) no perforation of the bony walls and no contents or fluid or connective tissue in the lesion. Afterwards, Hansen[5] added another criterion *i.e.* upon surgery, the lesion is essentially empty and occasionally the cavity may contain some fluid and/or small amount of tissues. The numerous synonyms of this cyst like traumatic bone cyst, hemorrhagic bone cyst, idiopathic bone cyst, osteodystrophic cyst, essential bone cyst, simple or solitary bone cyst, unicameral bone cyst, extravasation cyst, progressive bone cyst explain its unclear etiopathogenesis and presentation.[1,6]

SBC is mainly seen in young individuals, frequently during the first and second decade of life, and equally affecting both genders with a slight predominance to males.[1] The majority of SBCs are seen in long bones (90%) and far less frequently in jaw bones (10%). However body of the mandible between the canine and the third molar is the most common site (75%) in head and neck region followed by mandibular symphysis.[1] Very few cases are reported in the maxilla.[7,8]

Most of times this lesion is asymptomatic and discovered on routine radiographic examination only. However, sometimes the symptoms range from osseous expansion, pain, paraesthesia of the affected sides, failure of eruption of permanent teeth, displacement of the inferior dental canal, pathological fracture, and multiplicity of the lesion.[7-9] In addition, the lesion can be associated with other pathological conditions such as necrotic pulp, facial cellulitis, impacted third molar and odontogenic keratocyst.[8] Radiographically, it appears as an unilocular radiolucent area with an irregular but well defined outline, with or without sclerotic lining around the periphery of the lesion.[1,8] Characteristic of SBC is *scalloping* effect when it extends between the roots of the teeth.[9] The scalloped outline, however, is often found in edentulous areas too.[9] The definite diagnosis of SBC is usually made during surgical intervention by its gross appearance and contents.[1,9] The diagnosis will be confirmed after histopathological examination. A case of SBC in a rather older patient with localized buccal bone expansion mimicking periapical cyst is reported here.

Case Report

A female patient aged 37 years reported to the Department of Oral and Maxillofacial Surgery with a complaint of asymptomatic swelling in right lower back tooth region since eight months. The swelling was relatively insignificant extra orally and intraoral inspection revealed a sessile swelling localized to buccal vestibule of otherwise normal right mandibular first molar and measuring about 1×1 cm in size (Figure 1). The overlying mucosa was intact and normal in color. On palpation the swelling was hard and non-tender. There was no significant lymph nodes involvement. The orthopantomograph revealed periapical radiolucency in relation to right mandibular first molar (tooth #46), measuring about 1.5×2 cm. The radiolucency was extending into the inter-radicular region of right mandibular first molar without involving the interdental bone on distal and mesial sides of the tooth (Figure 2). The occlusal radiograph revealed a prominent buccal expansion and thinned out cortical bone around the affected tooth (Figure 3). The vitality test revealed nonvitality of right mandibular first molar. A diagnosis of periapical cyst was made. Endodontic treatment of the tooth and surgical enucleation of the cyst was planned. The surgical approach was achieved by crevicular incision with anterior release and reflection of mucoperiosteal flap. Exposed bone revealed a well-localized expansion in relation to right mandibular first molar with thinned out cortical plate (Figure 4). A bony window was made at that site, but no contents were found except for a thin tissue lining which lead to a diagnosis of SBC (Figure 5). The bony wall was curetted to stimulate bleeding and to remove thin connective tissue lining, irrigated well and primary closure was done. The small amount of tissue obtained was sent for histopathological examination. The microscopic examination using hematoxylin and eosin stain (H and E stain) showed loosely arranged fibrocellular connective tissue with interspersed bony trabecular pattern without any epithelial lining suggesting SBC (Figure 6). The patient is under regular follow-up.

Discussion

SBC is an uncommon radiolucent, pseudocystic lesion affecting jaw bones. The etiopathogenesis of SBC is still unclear. The most widely accepted theory for the development of this cyst is associated to an intramedullary hemorrhage as a result of trauma, which fails to organize and the subsequent degeneration of the clot, producing an empty

Correspondence: Madhusudan Astekar, Department of Oral and Maxillofacial Pathology, PAHER University, Pacific Dental College and Hospital, Debari, Udaipur (Rajasthan) - 313024, India. E-mail: madhu.tanu@gmail.com

Key words: bone cysts, solitary, aneurysmal, jaw cysts, pseudocyst, radicular cyst, periapical diseases.

Conflict of interests: the authors declare no potential conflict of interests.

Received for publication: 20 January 2012.
Revision received: 13 April 2012.
Accepted for publication: 16 April 2012.

Clinics and Practice 2012; 2:e59
doi:10.4081/cp.2012.e59

cavity within the bone.[1,3,6] Other theories for the pathogenesis include: i) infection of bone marrow; ii) loss of blood supply to a hemangioma or lymphoma; iii) cystic degeneration of existing bone tumor; iv) changes and reduction in the osteogenic activity; v) faulty calcium metabolism as a result of a systemic condition, such as parathyroid diseases; vi) ischemic necrosis of the fatty bone marrow; vii) low grade chronic infection; viii) imbalance between the osteoclastic and osteoblastic activity; ix) developmental defect; x) failure of mesenchymal tissue to form bone and cartilage, and instead becomes immature as multiple bursa-like synovial cavities.[1,3,6,8]

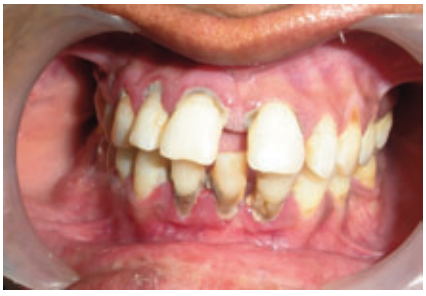

Figure 1. Localized swelling in buccal vestibule in relation to mandibular right first molar.

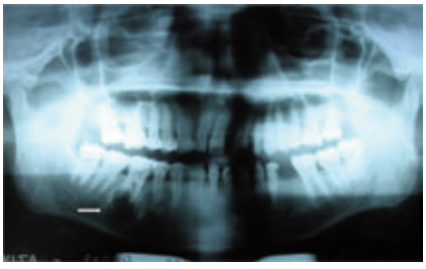

Figure 2. Orthopantomograph showing periapical radiolucency in relation to mandibular right first molar extending high into inter-radicular region.

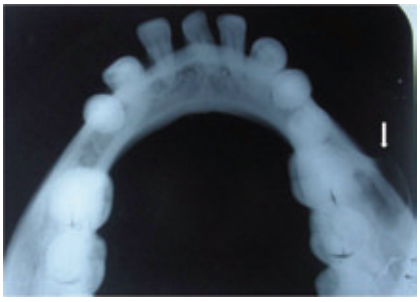

Figure 3. Occlusal radiograph showing localized prominent buccal cortical plate expansion in relation to mandibular right first molar.

SBC occurs most commonly during first two decades of life and males are affected more than females,[1,5,10] but Cortell-Ballester and coworkers report a female prediliction.[11] In the present report the patient was in her fourth decade of life, the age which is quite unusual for this cyst. In the maxillofacial region, 95% SBCs occur in the mandible.[7,8] The most affected area, including the present case, is the posterior premolar-molar area of the mandible, although cases involving the symphysis, the mandibular condyle and rarely the maxilla have been reported.[3,12] One case of SBC diagnosed after orthodontic treatment has been reported.[13] Hosseini[14] suggested that according to existing theories of pathogenesis, the SBC should appear with the same frequency in the maxilla as in the mandible. Hansen[5] and Kaffe *et al*.[15] assumed that the low incidence of maxillary SBC is due to the superimposition of various anatomical radiolucencies in the maxilla which obscures the presence of SBC and might go undiagnosed if asymptomatic.

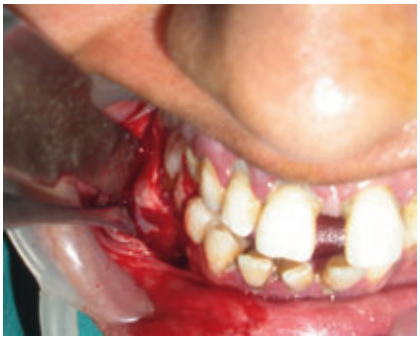

Figure 4. Thinned out buccal cortical plate with bluish hue.

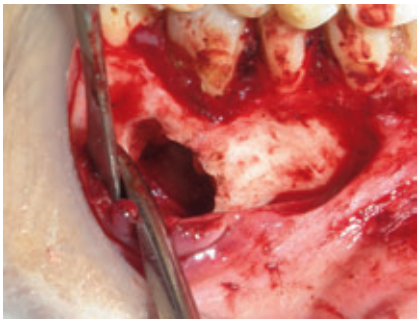

Figure 5. Cystic cavity without any significant lining and contents.

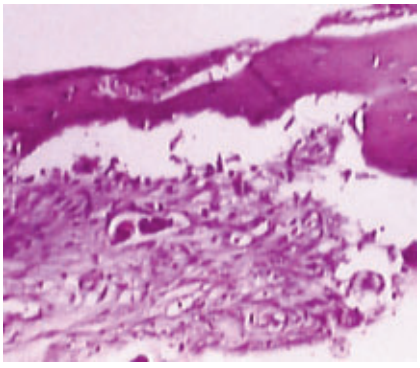

Figure 6. Histopathological view using hematoxylin and eosin stain (10x).

Root resorption because of this cyst is rare, but can cause disappearance of the lamina dura.[3] The results of a study on experimental model of mandible indicated that the operative finding of air in the cavity of SBC might have been an error at least in few cases.[16] Contrast enhanced magnetic resonance imaging (MRI) can be confirmatory for distinguishing these cysts from other cysts and tumors.[12]

Macroscopically cystic wall is usually composed of a thin connective tissue membrane, gray-yellowish in color, very friable, hemorrhagic, and difficult to remove for microscopic examination.[1,6] The cystic contents seem to change according to the SBC's evolution and location; the cyst can be filled with blood or serohematic or serous fluids and also empty in most of the cases as well as in the present case, especially in mandibular lesions. The protein composition in the cystic fluid is similar to that of serum. The walls of SBC have the property of a semi-permeable membrane.[6] Microscopic examination typically shows the cystic wall as a connective tissue membrane with numerous collagen fibers, with no epithelial lining. Numerous fibroblasts and giant cell like osteoclasts are sometimes visible, with some newly formed trabecular bone surrounded by numerous osteoblasts. Numerous congested capillaries and cholesterol crystals related to the osseous necrosis also may be present.[1,6,17] The present case fulfilled these criteria at the time of surgical intervention leading to a diagnosis of SBC which was later confirmed by histopathology. Various treatment modalities are suggested for SBC:[5,8] i) Keeping the case under observation and waiting for spontaneous regression, if it is asymptomatic; ii) Aspiration of the contents; iii) Surgical exploration and curettage to stimulate bleeding, healing, and initially to confirm the diagnosis; iv) Packing with gel foam saturated with thrombin and penicillin; v) Endodontic intervention alone; vi) Injection of methyl prednisolone acetate solution for treatment of long bone cases; vii) Injection of autogenous blood with bone graft or hydroxyapatite to stimulate the osteogenic activity; and viii) Bone grafting. Widely accepted and recommended treatment option for this cyst is surgical exploration and curettage of bony walls.[9,11] Careful curettage of the lesion helps progressive bone regeneration, offering a good prognosis and reduces relapse.

Recurrence of the lesion is not commonly encountered, however, it is being postulated

that existence of another cyst within the bone cavity, which may not have been enucleated, is a cause for recurrence.[14] Some SBC heal spontaneously without any intervention. This is probably the reason why they are rarely found in older age groups.[6,11] Recurrences are rare after surgical treatment.[6] Some authors recommend operative interference, which consists in exposing the involved area, cutting a large enough window to give access to the entire cavity and carefully curetting the entire contents down to cortical bone. The cavity should then be swabbed out with an escharotic, such as carbolic acid, alcohol, or zinc chloride saturated solution, followed by irrigation with normal saline solution.[18] Radiation therapy may be employed, but the results are more uncertain and less satisfactory than those obtainable by surgery.[18] The possibility of damaging the growth centers by over radiation should also be considered.

Conclusions

This simple cyst with potential complications is quite difficult to diagnose clinically. If left untreated can regress itself or can lead to all possible complications ranging from simple pain to dysesthesia, mere facial asymmetry to complicated fractures, simple root resorption to secondary infection of facial structures. One can suspect this cyst on radiological examination and confirm by surgical intervention or by contrast MRI. Early treatment and regular follow-up are recommended till it regresses.

References

1. Harnet JC, Lombardi T, Klewansky P, et al. Solitary bone cyst of the jaws: a review of the etiopathogenic hypotheses. J Oral Maxillofac Surg 2008;66:2345-8.
2. Lucas C, Blum T. Do all cysts of the jaws originate from the dental system. J Am Dent Assoc 1929;16:659-61.
3. Barnes L, Eveson JW, Reichart P, Sidransky D. Editors. World Health Organization classification of tumours. Pathology and genetics of Head and Neck Tumours. Lyon: IARC Press; 2005.
4. Rushton MA. Solitary bone cysts in the mandible. Br Dent J 1946;81:37-49.
5. Hansen LS, Sapone J, Sproat RC. Traumatic bone cysts of jaws: report of sixty-six cases. Oral Sur Oral Med Oral Pathol 1974;37:899-910.
6. Sandev S, Sokler K, Grgurevic J. Traumatic bone cysts. Acta Stomatol Croat 2001;35: 417-20.
7. Oral B, Oral F, Piskin B, Ozkan BT. Traumatic bone cyst of the anterior maxilla. Int J Dent Case Reports 2011;1:41-6.
8. Ahmed KA, Al-Ashgar F. Maxillary solitary cyst: review of literature and case report. Saudi Dent J 1991;3:109-13.
9. Xanthinaki AA, Choupis KI, Tosios K, et al. Traumatic bone cyst of the mandible of possible iatrogenic origin: a case report and brief review of the literature. Head Face Med 2006;12:40-4.
10. Forssell K, Forssell H, Happonen RP, Neva M. Simple bone cyst: review of the literature and analysis of 23 cases. Int J Oral Maxillofac Surg 1988;17:21-4.
11. Cortell-Ballester I, Figueiredo R, Berini-Aytes L, Gay-Escoda C. Traumatic bone cyst: A retrospective study of 21 cases. Med Oral Pathol Oral Cir Bucal 2009;14:e239-43.
12. Yokokawa M, Tanaka S, Ono M, et al. Traumatic bone cyst of the mandible diagnosed using contrast-enhanced magnetic resonance imaging. Int J Oral Med Sci 2011;10:31-6.
13. Motta AFJ, Torres SR, Coutinho ACA. Traumatic bone cyst–report of a case diagnosed after orthodontic treatment. Revista Odonto Ciência 2007;22:377-81.
14. Hosseini M. Two atypical solitary bone cysts. Br J Oral Surg 1978;16:262-9.
15. Kaffe I, Littner MM, Begleiter A, et al. Traumatic bone cyst of the maxilla - a rarity? Clin Prev Dent 1983;5:11-2.
16. Suei Y, Tanimoto K, Wada T. Simple bone cyst. Evaluation of contents with conventional radiography and computed tomography. Oral Surg Oral Med Oral Pathol 1994; 77:296-301.
17. Jaffe HL, Lichtenstein L. Solitary unicameral bone cyst: with emphasis on the roentgen picture, the pathologic appearance and the pathogenesis. Arch Surg 1942;44:1004-25.
18. Coley BL, Higinbotham NL. Solitary bone cyst: the localized form of osteitis fibrosa cystica. Ann Surg 1934;99:432-48.