Review Article

Traumatic Bone Cyst of Mandible: A Case Report of Rare Entity and Review of Literature

Abstract

The traumatic bone cyst was first described by Lucas in 1929 and later by Rushton. A solitary bone cyst (SBC) is a nonneoplastic osseous lesion affecting ≥95% of the metaphyses of long bones such as the proximal humerus and femur. The incidence of cyst affecting the jaws is 1% of all the cyst. About 89% of the lesion occurs in the mandible and 11% in the maxilla. Majority of the lesions occur in the posterior mandible, especially in the premolar–molar region. In this case report, we are presenting one such case of multiple lesions of SBC in mandible with systematic review of the literature.

Keywords: *Mandible, multiple lesions, traumatic bone cyst*

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Introduction

The traumatic bone cyst was first described by Lucas in 1929 and later by Rushton.[1] The term "traumatic bone cyst" is more widely used in the literature, but the International Histological Classification adopted by the WHO for odontogenic tumors uses the term "solitary bone cyst."[2]

A solitary bone cyst (SBC) is a nonneoplastic osseous lesion affecting ≥95% of the metaphyses of long bones such as the proximal humerus and femur.[3] The incidence of cyst affecting the jaws is 1% of all the cyst. Nearly 89% of the lesion occurs in the mandible and 11% in the maxilla. Majority of the lesions occur in the posterior mandible, especially in the premolar–molar region, while in the maxilla, they are more common in the anterior aspect.[4]

Til date, many cases of SBC have been reported in the orofacial region 34 out of them are cases comprising multiple lesions in the mandibular region. In this case report, we are presenting one such case of multiple lesions of SBCs in mandible with systematic review of the literature.

Case Report

A 25-years-old female patient reported to our unit with pain in the right lower back tooth region for 1 month. Pain was sudden in onset, throbbing in nature, and continuous in character. There was no history of fever, swelling, or paraesthesia associated with the pain.

On intraoral examination, pain seemed to be because of irreversible pulpitis with respect to 46. There was no sinus opening or tooth mobility present with respect to the same tooth. Electric pulp vitality test was negative. Medical history was noncontributory. IOPA was obtained and periapical radiolucency of 4 cm × 4 cm in size was seen with respect to the same tooth. Thereafter, an orthopantamogram (OPG) was recommended. The OPG revealed well-defined multilocular lesions [Figure 1], around 4 cm × 4 cm in size associated with respect to the right mandibular first and third molars and a well-defined unilocular radioluceny of about 12 cm × 5 cm size, extending from the mandibular canine to the third molar on the left side of the mandible. Another well-defined unilocular lesion of 10 cm × 10 cm in size was present with respect to the mandibular anterior region. There was no associated resorption of roots or pathological displacement of tooth.

A cone-beam computed tomography image was acquired to establish the position of the inferior alveolar neurovascular bundle and to determine the status of bone [Figure 2]. The findings revealed no buccal and lingual expansion of bone. The inferior alveolar

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Figure 1: Orthopantomogram showing multiple radiolucent lesions in the mandible

canal was severely displaced downward. On the basis of radiological and clinical findings, our differential diagnosis of the individual lesions consisted of SBC (traumatic bone cyst [TBC]), keratocystic odontogenic tumor (KCOT), and central giant-cell granuloma. Multiple KCOTs are seen in basal cell nevus syndrome. However, on clinical examination, no evidence was found for the same. We subsequently proceeded with the diagnosis of multiple TBC.

Aspiration was negative and an incisional biopsy specimen was obtained from the periapical region of the right second molar under local anesthesia. No tissue lining was obtained, so a second incisional biopsy was performed on the opposite side with respect to the left mandibular molars. Very scant tissue was obtained, which was sent for histopathological examination, which showed a thin band of vascular fibrous connective tissue with scant liquid content without any evidence of epithelial lining. These features were suggestive of a TBC.

Under general anesthesia, enucleation and curettage of the lesion were done, followed by bone graft placement. The lesion was approached by buccal crevicular incision. Full-thickness mucoperiosteal flap was raised. Surgical exploration revealed empty cavities with scarce amounts of tissue [Figures 3a and b]. A diagnosis of multiple SBC was surgically confirmed. Thorough curettage of the internal walls of the cavities was carried out and bleeding was induced. The cavity was packed with chips of Perioglas allogenic bone graft and closure was done using 3–0 vicryl sutures [Figure 4].

Histopathological examination of the excised specimen confirmed the diagnosis.

Postoperatively, the patient had mild diffuse swelling for 3–4 days, which subsided eventually. The patient has been followed up for 9 months and is asymptomatic with no positive findings of pain, swelling, and paraesthesia. Follow-up panoramic radiograph showed satisfactory bone healing without any evidence of enlargement of lesion and recurrence.

Materials and Methods

We systematically searched the Medline, Pubmed, and Google Scholar database using the keywords traumatic bone cyst, solitary bone cyst, idiopathic bone cyst, empty bone cavity, maxilla, mandible, atypical, unusual, oral cavity, and palate. However, our search was not just limited to these terms only. We checked the references of these articles for other cases and thoroughly studied them. In total, we found 34 cases of multiple SBC of jaws. The clinical and radiographic findings of these cases are shown in Table 1.[1,5-32]

Figure 2: Cone-beam computed tomography Image showing multiple bone cavities in the mandible

Discussion

The TBC is an uncommon, nonepithelial lined, intraosseous bone cavity of the jaws. They have been reported in the literature under a variety of names: SBC, hemorrhagic bone cyst, extravasation cyst, and simple bone cyst.[33] This is suggestive of the lack of understanding of the true etiopathogenesis of this lesion. However, the term "TBC" is most widely used at present.[34]

In the classification suggested by the World Health Organization (WHO), TBCs are included in the group of bone-related lesions, together with the aneurysmal bone cyst, ossifying fibroma, fibrous dysplasia, osseous dysplasia, central giant-cell granuloma, and cherubism.[33] The presence of multiple cysts is rare. So far, 34 cases have been reported in literature with only 5 cases being detected after the third decade of life.

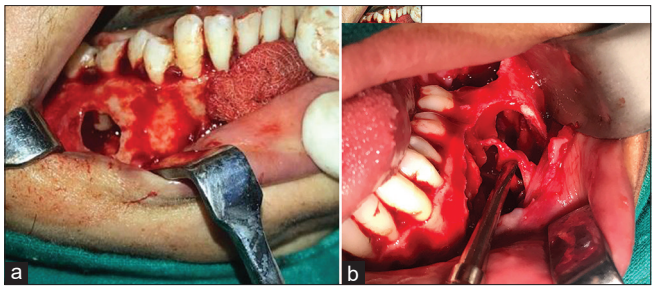

Figure 3: (a and b) Intraoperative photographs showing bone explorations

Suei *et al*. reported that TBC of the jaws have an equal prevalence in both genders, with most lesions occurring among patients in the second decade of life. However, the lesion is also been found in older age brackets.[35] The mandibular body is the frequent site of TBC; however, cases involving the mandibular symphysis, ramus, condyle, and even anterior maxilla have been reported.[36] Cases have also been reported where TBC was present in the zygomatic bone.[37]

The pathogenesis of the TBC is not yet completely understood; several theories have been suggested. Trauma has been frequently suggested as an etiological factor. However, the incidence of a positive history of trauma has been rather variable in the reported series of cases from 17% to 70%.[33] In the present case report, no history of trauma was elicited.

Trauma results in the formation of intraosseous hematoma. Subsequently, the blood clot liquefies, and surrounding bone is destroyed by enzymatic activity. Blum and Thoma suggested that a previous history of trauma to the jaws contributes to the formation of most of TBCs.[36] Thoma believed that trauma results in a subperiosteal hematoma that reduces the blood supply to the region, causing bone resorption by osteoclasts.

There might be an underlying systemic component in patients with multiple SBCs, such as predisposition to vascular anomalies. This might also explain why women are more predisposed, who tend to have more conditions with vascular coagulopathies.[38] Another proposed mechanism considers these lesions to be synovial cysts arising from a developmental juxta epiphyseal error with the intraosseous incorporation of synovial tissue. However, this is not consistent with our case of multiple cysts where no synovial lining was found.

Traumatic bone cysts are mostly detected accidentally as they are usually asymptomatic.[36] Cases were reported in literature where the lesion was asymptomatic and was detected during routine radiological examination. In most cases, intraorally, the soft tissues were found to be unaffected. Moreover, there was no increased mobility of the teeth or changes in their color. In our review, we found only three cases where nonvital teeth were associated with the lesion. The presence of symptoms is quiet variable. While

Figure 4: Suturing done

some patients may be asymptomatic, others may develop symptoms such as altered sensations, numbness, swelling, and pain.[33] There may also be cortical plate expansion.[35] In the present case report, the patient had throbbing pain due to irreversible pulpitis but no paresthesia.

The majority of TBC range from a lesion of 1 cm in diameter to those which involve the entire body and ramus of the mandible. In contrast to other cysts of the jaw, the hydrostatic pressure of TBC is relatively low, while the osmotic pressure in relation to blood is slightly higher.[33] The protein composition in the cystic liquid is similar to that in serum. It contains slightly more bilirubin.[39] The growth of some TBC can cause pathological fracture of the mandible. On the other hand, some TBC can have spontaneous regression.

The structure of the inferior alveolar canal may in some cases be preserved but not always. There are chances of contact with remaining roots and impacted wisdom teeth. There is no resorption of the teeth roots.[33] The following differential diagnoses can be considered:[40] periapical cyst of radicular cyst, KCOT, lateral periodontal cyst, odontogenic tumors-like ameloblastoma, fibrous dysplasia, cherubism, central giant-cell granuloma, aneurysmal bone cyst, arteriovenous malformations, and hemangiomas.

The treatment is surgical. The procedure comprises the evacuation of the cystic contents followed by curettage of the cavity to stimulate bleeding in the cavity.[39] The wound is then sutured. This is followed by the formation/creation and organization of a clot and healing by the formation of new bone.[41] Cases have been reported where TBC heal spontaneously without any intervention.[33] This is probably the reason why they are rarely found in older age groups and mostly restricted to younger age groups.[40] Recurrences are rare after surgical treatment. A histopathological diagnosis will confirm the existence of a TBC using some of the tissues obtained from the bone cavity.[42] Histological examination of slides shows fragments of fibrovascular

Table 1: Review of literature

Reference	Age/sex	Presentation	History of trauma	Tooth vitality	Radiological features	Recurrence	Histopathology
Hankey 1947	15/male	Routine radiographic examination	No	NS	Scalloping	Not up to 6 months follow-up	NS
Hutchinson 1954	23/NS	Routine radiographic examination	No	NS	Scalloping, loss of lamina dura	NS	NS
Thoma 1963	15/female	NS	NS	NS	NA	NS	SBC
Szerlip 1966	15/female	Routine radiographic examination	NS	Vital	Scalloping	4 years 5 months	SBC
Grasso *et al*. 1969	35/male	Routine radiographic examination	No	Vital	Scalloping, multilocular expansion	7 months	Sclerotic bone
Morris *et al*. 1970	21/female	Routine radiographic examination	NS	Vital	NA	NS	NS
Hubner 1971	11/female	Routine dental examination	No	Vital	NS	6-month postoperation no recurrence	NS
Stewart *et al*. 1973	17/male	Routine radiographic examination	No	NS	Scalloping	6 months	Lamellar bone and loose erythrocytes
Schofield 1974	20/female	Routine radiographic examination	Yes	Vital	Scalloping, multilocular buccal erosion	4-5 months	Trabeculae bone, connective tissue
Ruprechet and Reid 1975	17/male	Pain	NS	Vital	NS	7 months	NS
Heimdahl 1978	16/male	Swelling	NS	Vital	Scalloping and expansion	3 months	NS
Pogrel 1978	18/female	Routine radiographic examination	NS	Vital	Scalloping and expansion	1 year	Bone fibrous tissue
Markus 1979	13/female	Routine radiographic examination	NS	NS	Multilocular tooth displacement, loss of lamina dura, root resorption, expansion	6 months	Lamellar bone, fibrous tissue
Raibley *et al*. 1979	13/female	Routine radiographic examination	No	NS	Scalloping multilocular expansion		Granulation tissue, foreign body giant cells
Kuroi 1980	36/female	Routine radiographic examination	No	Vital	NA	Recurrence 3 months, second year 9 months,	Traumatic bone cyst
Patrikou *et al*. 1981	15/NS	Pain	NS	Vital	Scalloping, expansion	16 months	Traumatic bone cyst
Foress *et al*. 1988	NS	NS	NS		Scalloping	Yes/NS	Cancellous bone, lamellar compact bone
Brannon and Houston 1991	21/female	Routine radiographic examination	No	NS	NS	NS	Reactive bone, fibrous, and granulation tissue
Saito *et al*. 1992	<30, NS	Routine radiographic examination	NS	Vital	NA	Yes	NS
Saito *et al*. 1992	>30, NS	Routine radiographic examination	NS	Vital	NA	Yes	NS
Saito *et al*. 1992	>30, NS	Routine radiographic examination	NS	Vital	NA	Yes	NS
Prakash *et al*. 1992	32/male	Pain/swelling	NS	Vital	Scalloping, expansion	16 months	Bone fragment, inflammatory granulation tissue
Feilding 1992	15/female	Routine radiographic examination	No	Vital	Scalloping, expansion	1.5 years	Dense cortical bone
Jones and baughman 1993	23/female	Pain and swelling	No	Vital	Expansion	Not up to 2 years	Bone trabeculae, fibrous connective tissue

Contd...

Table 1: Contd...

Reference	Age/sex	Presentation	History of trauma	Tooth vitality	Radiological features	Recurrence	Histopathology
Oda *et al*. 2002	12/male	Swelling	Yes	Vital	Multilocular, tooth displacement, loss of lamina dura, root resorption, expansion	8 months	Granulation tissue
Kraut and Robin 2003	43/female	NS	NS	NS	Scalloping and tooth displacement	1st recurrence, 18 months, 2nd 14 months	Fragment of bone, granulation tissue
Mupparappu *et al*. 2008	39/female	Pain, swelling	NS	Vital	Multilocular, loss of lamina dura	1st recurrence - 2nd year, 2nd recurrence-NS	Bone trabeculae, fibrous connective tissue, rims of osteoid, acute and chronic inflammatory cells
Kuhmichael and boulox 2010	32/female	Routine radiographic examination	No	Vital	Scalloping, expansion	12 months	NS
De Oliviera *et al*. 2012	22/male	Routine radiographic examination	No	Vital	Scalloping	12 months	NS
Martins filho *et al*. 2012	10/male	NS	No	NS	Scalloping, multilocular expansion	NS	NS
Mathew *et al*. 2012	15/female	Swelling	No	NS	Scalloping, multilocular expansion	4 months	NS
Seo -Yungan *et al*. 2012	11/female	Routine radiographic examination	No	Vital	Loss of lamina dura, expansion	2 years	NS
	17/male	Routine radiographic examination	No	Vital	Scalloping, loss of lamina dura, root resorption	NS	NS
	18/male	Routine radiographic examination	No	NS	Scalloping, loss of lamina dura, root resorption	3 months	Viable bone, dense fibrous connective tissue

SBC: Solitary bone cyst; NS: Not stated; NA: Not available

connective tissue, cholesterol clefts, and foreign body giant cells.

In our case report, there was no sign of a fibro-osseous or any other lesion and numerous extravasated red blood cells were present. The specimen was noted for the absence of epithelium prompting the diagnosis of a traumatic bone cyst. However, if no tissue is found in the cavity for histopathological examination, a decision on diagnosis of a TBC will be according to the clinical experience of the operating surgeons.

There maybe the possibility of damage to the inferior alveolar nerve in cases where the mandible is operated. In cases where substantial bone resorption occurs, there is also the possibility of a fracture. In our case, however, no nerve damage was seen and at 7 months follow-up, there was no paresthesia. Radiological examination showed that satisfactory bone formation was present, with no evidence of recurrence.

Conclusion

Multiple traumatic bone cysts are a very rare finding. Although successful treatment outcomes are well reported, the precise nature and etiopathogenesis of the lesion is not completely understood. The treatment modality in most cases is the same. A thorough clinical examination, patient history, and careful radiological evaluation will be helpful when deciding on the final diagnosis and treatment plan. Surgical exploration followed by histopathological examination can establish the final diagnosis. Recurrence after surgery is extremely rare.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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