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Traumatic Bone Cyst Possibly Following Removal of Third Molar: Review and A Case Report

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Abstract

Aim: The traumatic bone cyst (TBC) still remains a controversial entity. The aim of the paper was to review nomenclature, aetiology, pathogenesis, clinical, radiographic findings and management of traumatic bone cyst. This paper also presents a well-documented case of traumatic bone cyst involving angle/ramus of the mandible possibly an iatrogenic origin following removal of a wisdom tooth.

Methods: To review literature in past and document relevant findings regarding traumatic bone cyst of the jaw. To assess documented cases reported so far in angle of mandible possibly caused by removal of an impacted third molar.

Results: Review on nomenclature, aetiology, pathogenesis and classification still remains overlapping. Historically, this cyst has been known by several different pseudonyms, including solitary bone cyst (SBC), haemorrhagic cyst, extravasation cyst, and progressive bone cavity and still remains the same.

Conclusion: Clinicians should be aware of this lesion and that unexplained radiolucent lesions of the jaws should be documented and addressed accordingly.

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Traumatic bone cyst, Radiolucent lesions, Solitary bone cyst, Intramedullary haemorrhage

Clinical Relevance

Scientific Rationale of study

Traumatic bone cyst (TBC) of the jaw is an uncommon lesion usually detected as an incidental finding on dental radiographic examination. Considering its aetiology, clinical, radiological and histological presentation the term commonly used of traumatic bone "cyst" is inappropriate and should more accurately be called a traumatic bone "cavity".

Principal findings

Historically, this cyst has been known by several different names within the literature reflecting its uncertain nature.

The definitive diagnosis of TBC is invariably achieved at surgery.

Practical implications

TBC is an uncommon lesion of the jaw but clinicians should be aware of its clinical and radiographic characteristics and management.

Introduction

TBCs have been reported in the literature under a variety of names since first described by Lucas [1] in 1929: solitary bone cyst(SBC) [2-4], haemorrhagic bone cyst [5-7] extravasation cyst [8,9] progressive bone cavity [10,11] and unicameral bone cyst [12-14]. This varied terminology reflects the unexplained cause of the SBC as there is no unanimous agreement on the exact nature and possible cause of such cysts.

The SBC as this lesion will be called of the jaw is classified as a non-epithelial lined cyst according to World Health Organization, histological typing of odontogenic tumours, 2017 [2]. The lining, if present, is of connective tissue and contents, if any, are blood or its derivatives. It is usually asymptomatic and detected as an incidental finding on radiographic examination. Frequently it has a characteristic scalloped radiographic outline, and surgical exploration usually results in establishing the diagnosis and promoting the subsequent resolution. Since Lucas [1] first description, the lesion has attracted a great deal of interest in the dental literature, but its pathogenesis is still not clearly understood.

The criteria set out by Rushton [3] for the diagnosis of SBCs are as follows:

1. The cyst should be single and have no epithelial lining;
2. It should show no signs of acute or chronic infection;
3. It may contain red fluid but not soft tissue;
4. The walls of the cyst should be bony but of variable thickness.

Examination of the so-called red fluid, described by Rushton [3], by protein electrophoresis revealed a normal serum pattern. Donkor and Punnia-Moorthy [4] suggested a possible sub classification of SBCs based on their contents, i.e., empty cysts to be called idiopathic; those with solid content would be designated according to the histologic appearance of the bulk of the solid e.g. fibrous or granulation tissue and cysts containing fluid with the a biochemical profile similar to serum could be called extravasation cysts.

The SBC is mainly seen in young patients most frequently during the second decade of life. The sex distribution is reported to have a slightly male predilection. The majority of SBCs are primarily seen in the body of the mandible between the canine and last molar; the second most common site is the mandibular symphysis. There are reported cases involving the angle of the mandible, the ramus of the mandible and the maxilla.

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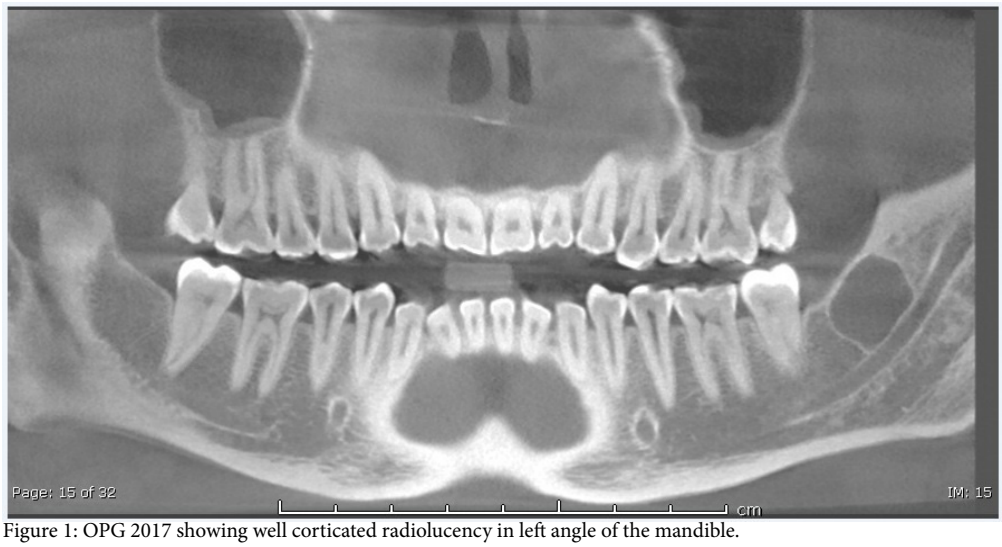

Figure 1: OPG 2017 showing well corticated radiolucency in left angle of the mandible.

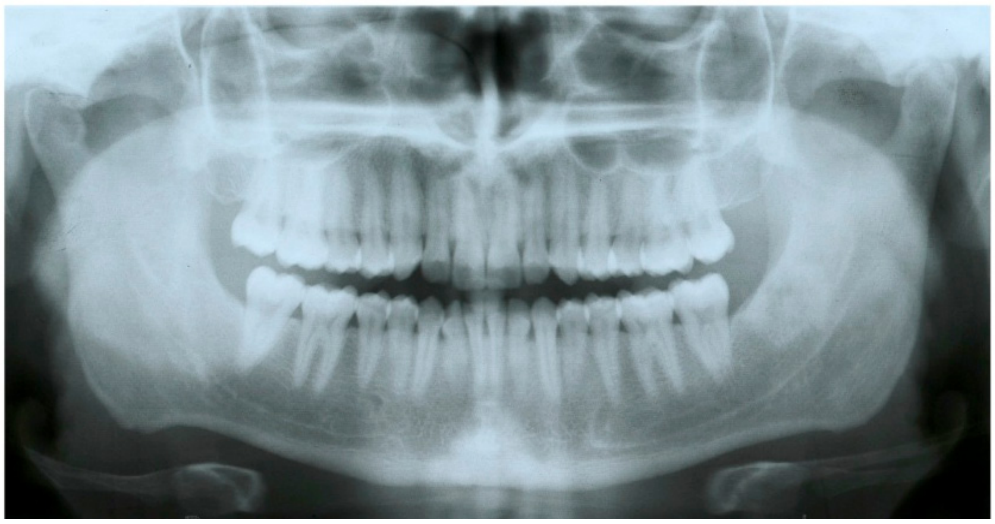

Figure 2: OPG 2010 showing early signs of radiolucency in left angle of the mandible.

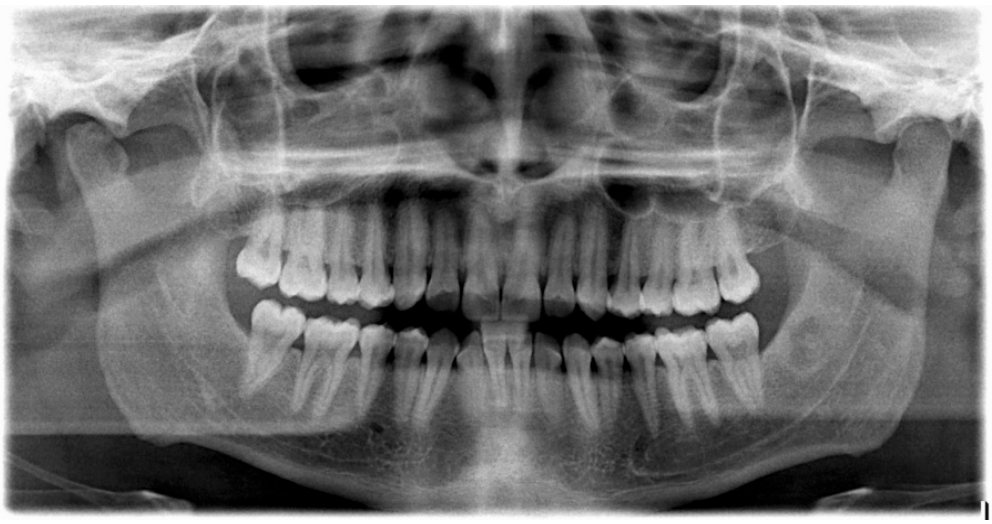

Figure 3: OPG 2012 showing increase in size of radiolucency in left angle of the mandible.

Radiologically, the lesion appears as a radiolucent area but with a less well-circumscribed boundary than the odontogenic cyst. A scalloped effect is often seen around the roots of the adjacent teeth, which are normally vital. The radiolucency is situated superior to the inferior alveolar nerve in contrast to the Stafne's idiopathic bone cavity that is located below the mandibular canal.

This paper presents a case of SBC of possible iatrogenic origin and discusses its pathogenesis.

Case Report

A 34 year old man was referred to the oral surgery clinic HCF Dental Centre, Sydney, Australia by his dentist for opinion regarding radiolucency involving the left angle of the mandible (Figure 1).

He was medically fit and well. At the age of 25, he had left lower wisdom teeth removed and described it as a traumatic experience. Unfortunately he did not keep his previous radiographs. There was no history of any previous trauma other than removal of the wisdom tooth. The patient was completely free of symptoms. There was no expansion of cortical bone, either buccally or lingually. There were no palpable lymph nodes present. Radiographically on OPG there was a unilocular radiolucency with well-defined scalloped borders just above inferior dental canal. OPGs from 2010 and 2012 at the age of 27 and 29 (Figure 2 & Figure 3) were available and it was evident that the radiolucency had developed over the past 7 years. CBCT confirmed a 16 X 12mm diameter radiolucent lesion on the left angle of mandible extending to the ramus (Figure 4). Differential diagnosis of odontogenic keratocyst, unicystic ameloblastoma, residual cyst and TBC was considered. The patient consented to exploration of the lesion to achieve definite diagnosis and management.

Exploration was carried out under general anaesthesia, a mucoperiosteal flap was reflected and a window was opened in to the lesion. The cavity was seen to be empty, except for a small amount of straw-colored fluid in the base; there was no soft tissue present. The surgical site was closed primarily. A definitive diagnosis of TBC was established. The Post-operative phase was uneventful and he was advised to have a review in six months time or earlier if any concern arose. The patient came for review nine months post op with no complaints and OPG showed complete bone healing (Figure 5).

Clinical Presentation

SBCs are usually asymptomatic and are detected as an incidental finding on radiographic examination. Characteristic radiographic features associated with SBCs are generally unilocular, unilateral and asymptomatic radiolucent area with scalloping. The classic scalloping appearance suggests that the resorption process follows the path of least resistance and may reflect the absence of hydrostatic pressure that is associated with the growth of more common cysts. Trabecular bone has low resistance to resorption as compared to cortical bone or root dentine; this may explain the apparent sparing of roots and lamina dura. Howe [6,7] reported in his series of review of 60 cases, 2 cases presenting with pain and paraesthesia, 2 presenting with pain and swelling and another 2 presenting with only pain. Other features include an inconsistent lamina dura and an occasional root resorption [23-26]. As with any intra-bony lesion, bone resorption occurs in SBCs.

SBCs are said to occur mainly in the body and symphysis regions of the mandible in the facial skeleton. Occasionally these lesions may extend to the ramus. However, a few cases have been reported with a location, which is entirely in the ascending ramus [21,22]. Hoffmeister

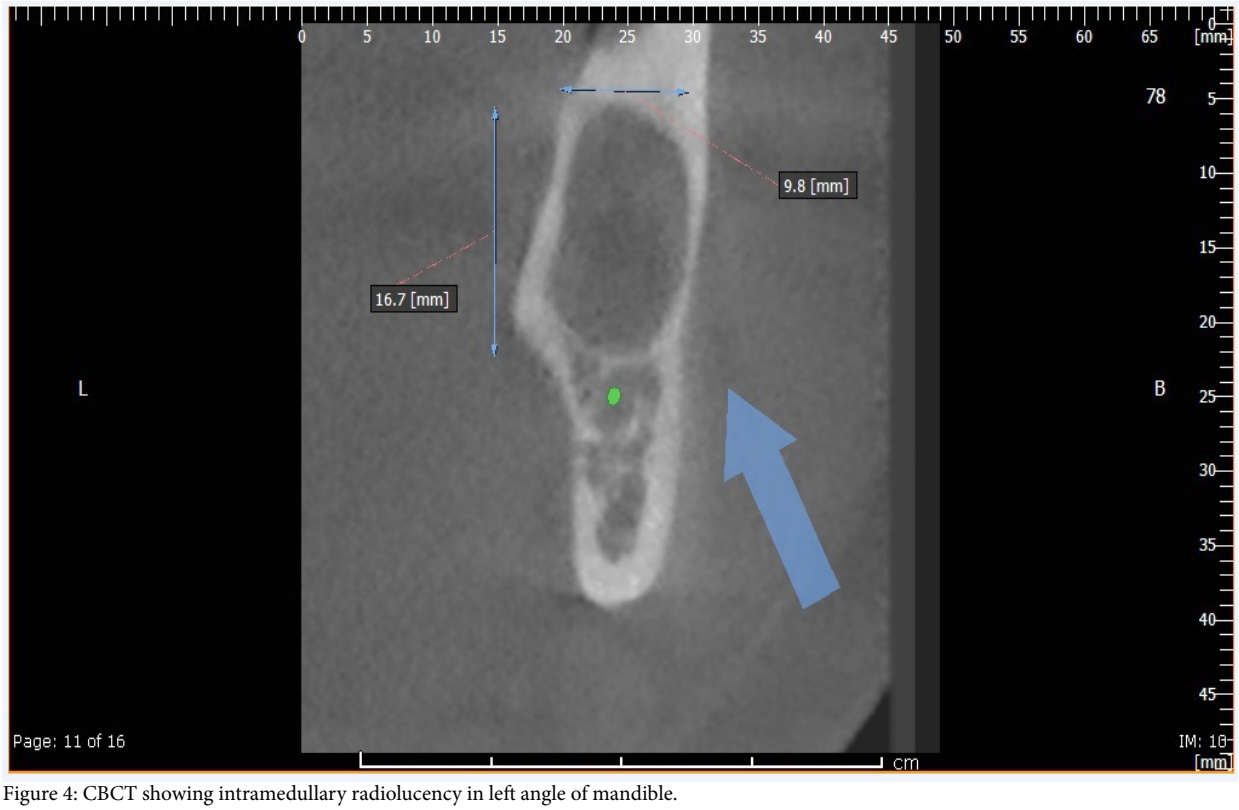

Figure 4: CBCT showing intramedullary radiolucency in left angle of mandible.

and Harle [23] reported a frequency of 0.6% in their review of 3,353 jaw cysts. The frequency seems to be greater in males and they tend to present within the second decade of life. The majority of cases reported occurred in the mandible and rarely in the maxilla.

Pathogenesis

In order to explain the unusual behaviour of SBC, the following theories have been put forward (Table 1).

Many authors suggest that a history of trauma to the jaws plays a role in its initiation. The most commonly accepted theory proposed by Pommer, is that it is caused by intramedullary haemorrhage following any sort of trauma, including tooth removal. Both Hansen et al. [10] and Beasley [12] reported trauma to the jaws implicated in 26.9% and 81% of cases, respectively. It has been suggested that this precipitating trauma causes intramedullary haemorrhage within the bone. Failure of haematoma organisation and liquefaction of the clot, coupled with possible factors such as venous insufficiency and high intra-cystic hydrostatic pressure, results in bone cavity formation (Figure 6). In the literature, however, cases of SBC have been reported where there has been no apparent relationship with trauma. The role of trauma in inducement of this bleeding is controversial and coincidental. Degering [14] advocates periodic radiographic checks after injuries to the face in anticipation of such lesions.

The haemorrhagic theory is unsatisfactory in a number of ways. It fails to explain why, as in cases reported by Biewald [15], resolution occurs in SBCs of the jaws by simple injection of autogenous blood. Nor does it account for their predilection for sites in the rest of the skeleton not necessarily prone to trauma (Whinery [6]). Matsumura et al. [18] examined 53 SBCs to correlate between histologic and radiographic findings and to discuss its aetiology. The study showed bone expansion and radio-opacity were closely related to their histological characters and recurrence was more likely to be observed in patients with partially thickened wall with dysplastic bone formation.

Some authors consider the SBC is developmental in origin [17]. Hosseini [18] suggested a developmental origin involving failure of differentiation of osteogenic cells from mesenchymal cells. Osteogenic cells undergo metaplastic change and form synovial cells, which coalesce to form a fluid filled cavity. Mirraet al. [19] reported synovial cells in the wall of a simple bone cyst of femur. The theory of failure of osteogenesis and formation of synovial hamartoma, derives some support from Johnson & Kindred [20]. These authors studied the walls of 200 cases of extra-facial bone cyst. The lining they encountered varied from a few micra to a centimetre in thickness. When thin, the lining resembled a juxtra-articular ganglion; when thick, a bursa. Other possible theories include intraosseous vascular abnormalities, degeneration of bone tumours and low-grade infection. However, despite numerous other hypotheses, the defect, which leads to the formation of these lesions, remains unknown.

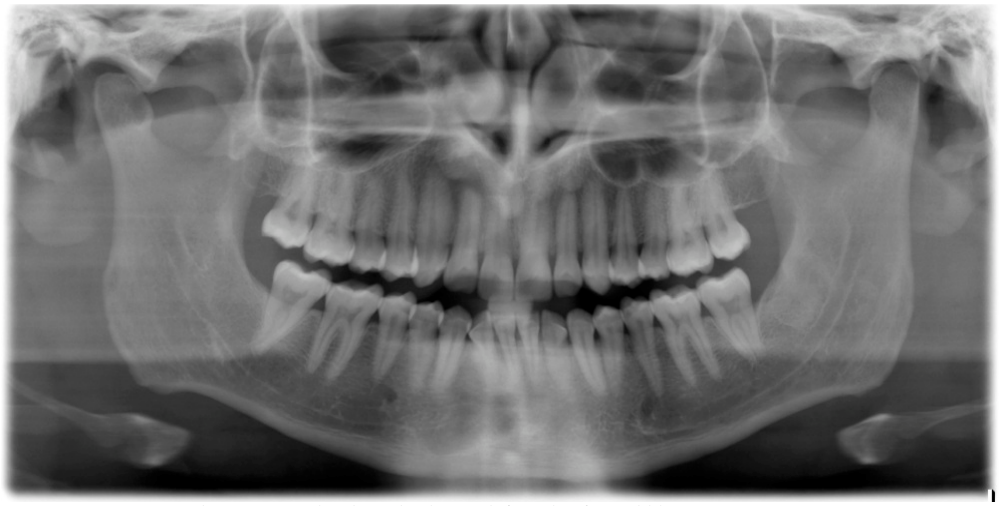

Figure 5: Postop OPG showing complete bone healing in left angle of mandible.

Cause	References
Degeneration of bone tumor	Blum, 1955 [36] Huebner GR, Turlington EG, 1974 [17]
Faulty calcium metabolism	Bloodgood JC, 1942 [39]
Low grade infection	Bloodgood JC, 1942 [39]
Local disturbance of bone growth	Jaffe HL, 1942 [11]
Venous obstruction	Cohen J, 1970 [38]
Excessive osteolysis	Ogden JA, Griswold DM, 1970 [40]
Intramedullary haemorrhage	Pommer G, 1951 [5] PogrelMA, 1987 [4]
Combination of the above	Bernier JL, Johnson LC, Whinery JG, 1955 [41]

Table 1: Etiology of Solitary/Traumatic Bone Cyst.

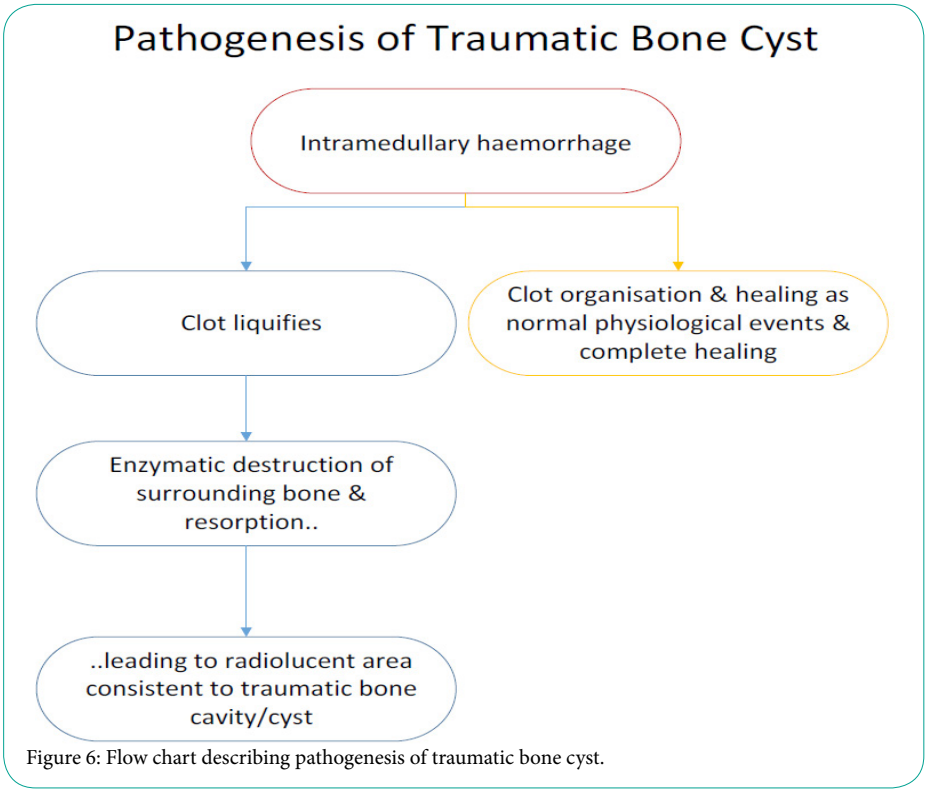

Figure 6: Flow chart describing pathogenesis of traumatic bone cyst.

Year	Author	Cases reported/as	Treatment/others
1929	Lucas et al [1]	1 case as HBC	Surgical intervention
1946	Rushton M [3]	3 cases as SBC	Discussed diagnostic criteria
1955	Thoma KH [36]	1 case as HBC	Used Gelfoam to treat the case
1955	Whinery JG [9]	3 cases as SBC	Surgical intervention
1955	Blum T [35]	1 case as HBC	Resolved by aspiration
1955	Jacobs MH [7] ref from Howe GL 1965	4 cases as TBC	Curettage
1956	Boyne PJ [31]	2 cases as TBC	Treated with freeze-dried homogeneous bone
1963	Howe GL [6,7]	3 cases HBC	Surgical intervention
1964	Fordyce GL[55]	1 case as HBC	Surgical intervention
1965	Howe GL [8]	60 cases as HBC from previous studies	Review of treatment
1987	Pogrel et al [4]	2 cases as SBC	Surgical intervention
1998	Matsumura et al [18]	53 cases as SBC	Histological study
2006	XanthinakiAA et al [30]	1 case as TBC	Surgical intervention
2008	Harnet JC et al [56]	SBC review	Etiopathologic review
2009	Cortell-Ballester I et al[47]	21 cases as TBC	Surgical intervention
2009	Seehra J et al [29]	1 case as SBC	Surgical intervention
2012	Dincer O et al [49]	1 case as TBC	Surgical intervention
2012	Martin-Filho PR et al [50]	26 cases as TBC	Surgical intervention
2012	Manor E et al [57]	Review of cystic lesions 322 cases	16 were TBC
2013	Shah KM et al[48]	1 case as TBC/SBC	Surgical intervention
2013	Salem AS et al [46]	1 case as TBC	Surgical intervention
2014	Kumar S et al [52]	2 cases as TBC	Surgical intervention
2014	Madiraju G et al [54]	1 case as SBC	Surgical intervention
2015	Sathyan P et al[44]	2 cases as TBC	Surgical intervention
2016	Reddy GS et al [43]	1 cases as HBC	Surgical intervention
2016	Falci SG et al [53]	1 case bilateral mandible as SBC	Surgical intervention
2017	Nagaraj T et al [45]	1 case as TBC	Surgical intervention
2018	Zemmouri Y et al [51]	1 case as SBC	Surgical intervention

Table 2: Review of data published as TBC/SBC.

Management

Although SBC may heal spontaneously, exploration is generally considered advisable to confirm the diagnosis, and this in itself constitutes definitive treatment. Simple exploration of the cyst is usually curative. Curettage of the bony walls is generally recommended and bone regenerates within a few months. Treatment options suggested have included enucleation and primary closure, injection of autogenous blood and primary closure, insertion of gelfoam and primary closure, insertion of bone grafts, and exploration and packing to obtain secondary healing. All methods of treatment appear to be equally successful and recurrence has not been reported. Some authors advise the avoidance of curettage of the roof of the cavity when present in dentulous area of the jaws, in an attempt to preserve the vitality of the adjacent teeth. It has been documented that the contents of this cyst can range from an empty cavity to one containing either blood or serous fluid. Histological examination of SBCs reveals a loose vascular fibrous tissue membrane of variable thickness with no epithelial lining [28-31]. It is unlikely that these cysts will predispose to fracture of the mandible. However, the general consensus is that a surgical intervention is required, not only to confirm the diagnosis, but also to stimulate healing.

Discussion

SBC lesions are usually asymptomatic on clinical presentation [30-32,35]. The definitive diagnosis of the SBC is invariably achieved at surgery following exploration [33,34,36].A fibro-osseous lesion and a SBC may have similar radiolucent appearance during the initial formation and early presentation of the lesion. It is only during maturation that a distinction can be made, in that; the TBC retains its original radiolucent appearance while the fibro osseous lesion develops calcifications leading to opacities. Although not diagnostic, this distinction may help to differentiate the SBC from more common fibro osseous lesions. Considering its aetiology, clinical, radiological and histological presentation the term commonly used of traumatic bone "cyst" is inappropriate and should more accurately be called as traumatic bone "cavity" [37]. The literature review revealed overlap of terminologies. Most commonly used were solitary, haemorrhagic and traumatic bone cyst and the most of the authors advise surgical exploration as management option (Table 2).

The case reported had possibly an iatrogenic origin and was well documented clinically and radiologically to be appropriately diagnosed as traumatic bone cavity. Since trauma and intramedullary haemorrhage, with subsequent abnormal healing appears to be the most plausible explanation for the development of TBC in this case, it seems quite reasonable that this might have occurred following surgical removal of the third molar [38]. There are a few case reports in the literature-suggesting incidence of TBC following removal of third molar teeth including Pogrel [37], Xanthinakiet al. [29]. This case can also be attributed to the history of a previous traumatic procedure, as there was no history of other trauma.

Summary

TBC remains a controversial entity. Simple surgical intervention helps in diagnosing and treating these lesions. Recurrence remains unlikely. Our case suggests that previous traumatic removal of a wisdom tooth seems to be the most likely aetiological factor.

Competing Interests

The author declare that there is no competing interests regarding the publication of this article.

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