

TRAUMATIC BONE CYST OF THE MANDIBLE; DIAGNOSTIC CHALLENGE AND MANAGEMENT. A CASE REPORT

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ABSTRACT

The traumatic bone (TBC) cyst is an uncommon benign empty or fluid containing cavity within bone that is not lined by epithelium. The etiopathogenesis of TBC is still unknown. TBC is frequently encountered in young patients during the second and third decades of life. Sex predilection is equal but some studies in literature suggest clear female predominance. 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. The cysts are usually asymptomatic. Associated teeth are usually vital with no resorption or displacement. It expands the cortices and, seldom, intraoral or extra oral swelling may be seen. Most of the TBCs are diagnosed incidentally in orthopantomogram (OPG). On radiographic examination, a unilocular irregular but well defined lytic lesion is seen characteristically extending between the roots of the teeth. TBC is representing approximately 1% of all jaw cysts. A final diagnosis of a TBC is almost invariably made at the time of surgery, where in identification of an empty air-filled cavity serves as a valuable diagnostic tool. Surgical exploration was proved not only essential in making the right diagnosis but also curative from a treatment plan perspective. Recurrence of TBC is assumed to be extremely rare. However, a distinct proportion of recurrences may occur.

INTRODUCTION

TBC shows its non-neoplastic behavior and is defined by World Health Organization (WHO) as “an intraosseous cyst with fragile connective tissue lining without any epithelium.⁽¹⁾ TBCs are also referred to by various other names in the literature, including extravasation cyst, hemorrhagic bone

cyst, progressive bone cavity, simple bone cyst, solitary bone cyst, and unicameral bone cyst.⁽²⁾ The multitude of the names applied to this lesion attests to the lack of understanding of the true aetiology and pathogenesis.⁽³⁾ TBCs were described in 1929⁽⁴⁾ by Lucas and Blum as separate disease entities.⁽⁵⁾ Diagnostic criteria were established

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in 1946, comprising of a single bony cavity without epithelial lining, encompassed by bony walls, lacking contents or containing liquid and/or connective tissue.⁽⁶⁾ Afterwards, Hansen 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.⁽⁷⁾ WHO classified traumatic cyst as a non-neoplastic osseous lesion due to absence of epithelial lining, which demarcates it from other true cysts.⁽⁸⁾ 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.⁽⁹⁾

For explaining this lesion various theories have been put forth by many authors,⁽¹⁰⁾ none of them explains all the clinical and pathological characteristics of the lesion.⁽¹¹⁾ Regarding the presence of a history of trauma in most reported cases,⁽¹²⁾ the traumatic–hemorrhagic theory seems to be the most accepted.⁽¹³⁾ The explanation underlying the hypothesis of trauma⁽¹⁴⁾ according to this theory TBC arises from a focus of intramedullary hemorrhage that causes a hematoma after trauma.⁽¹⁵⁾ This hematoma subsequently liquefies and fails to organize and be replaced with tissue.⁽¹⁶⁾ The other theories assert failure of growth and development of osseous tissue, biochemical and mesenchymal differentiation, degeneration of benign neoplasms and low-grade infection.⁽¹⁷⁾ According to Wakasa et al. Florid cemento-osseous dysplasia is the initiative factor for TBC to develop due to lymphatic drainage obstruction.⁽¹⁸⁾

The TBC is an uncommon,⁽²⁾ The lesion is mostly diagnosed in patients below 30 years of age with an approximate mean age of 20 years.⁽¹⁹⁾ Reduced prevalence of TBCs in older age group has led to the speculation that this lesion may be self-repairing.⁽²⁰⁾ Sex predilection is equal.⁽²¹⁾ Although some studies state they are more frequently in men.⁽²²⁾ But some studies in literature suggest

clear female predominance (14:7).⁽²³⁾ The majority of cases are seen in long bones (90%).⁽¹⁾ TBCs are rare lesions, accounting for 1% of all jaw cysts.⁽²⁴⁾ Most commonly, the TBC involves the body or symphysis of the mandible,⁽²⁵⁾ with only a few cases reported in the condyle.⁽²⁶⁾ Maxillary lesions tend to be uncommon, although the reasons for this are unclear.⁽²⁷⁾

Most of times this lesion is asymptomatic and discovered on routine radiographic examination only.⁽²⁸⁾ However, dull pain is associated in 10–30% of the patients,⁽⁷⁻¹⁵⁾ swelling, tooth sensitivity, and less commonly with fistula, root resorption, paresthesia, displacement of inferior alveolar canal,⁽²⁹⁾ delayed eruption of permanent teeth. Cortical plate expansion is seen in some cases, which occurs commonly on buccal side and pathological fracture of the mandible.⁽³⁰⁾ The adjacent to the lesion teeth are usually vital and there is no mobility.⁽³¹⁾

Radiographic features of TBCs are non-pathognomonic.⁽⁶⁾ However, on radiologic evaluation, the lesion exhibits unilocular or multilocular radiolucency with well-defined or ill-defined margins. When multiple teeth are affected the radiolucencies involving the roots shows a dome like projections that scallops between the roots⁽³²⁾ and often scallops between the roots of the teeth, almost always diagnostic.⁽³³⁾ The scalloped outline, however, is often found in edentulous areas also.⁽⁷⁾ Furthermore, scalloped margin is a sign of possible recurrence, although this should not be confused with the interdental scalloping associated with an intact lamina dura.⁽³⁴⁾ However, This radiographic pattern may vary, which includes multilocular presentation, association with impacted teeth, and multiple occurrences in the same patient.⁽²⁰⁾ The histopathological evaluation reveals a vascular connective tissue or a thick myxofibromatous proliferation intermixed with normal bone,⁽³⁵⁾ with a scant yellowish, even blood colored liquid,⁽³⁶⁾ devoid of epithelial lining in the edges of lesion.⁽³⁷⁾ 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.⁽³⁸⁾

A final diagnosis of a TBC is almost invariably made at the time of surgery, when identification of an empty air-filled cavity serves as a valuable diagnostic tool.⁽²⁴⁾ Diagnosis established with aspiration of lesion and opening of bone window and accumulation of bleeding and fibrin inside the cavity is usually efficient for resolution of bone.⁽⁷⁾ Because of a lack of unique clinical and radiographic features, it is important to establish the differential diagnosis between TBCs and other radiolucent lesions of the jaws.⁽⁴⁾ Differential diagnosis included central giant cell granuloma, aneurysmal bone cysts, haemangioma,⁽³⁹⁾ dentigerous cysts, keratocystic odontogenic tumors, ameloblastomas, odontogenic myxomas, calcifying epithelial odontogenic tumor, adenomatoid odontogenic tumor, focal osteoporotic bone marrow defect, intraosseous vascular malformations, early stage of cementoma (periapical cemental dysplasia), fibrous dysplasia and cherubism.⁽²¹⁾ Several treatment modalities have been reported, including resection, curettage, bone grafting, corticosteroid injection and more recently, injection of autologous medullary bone.⁽⁴⁰⁾ The widely recommended treatment for TBCs is surgical exploration followed by curettage of the bony walls.⁽²³⁾ It serves as both a diagnostic maneuver and a definitive therapy by producing bleeding in the cavity. Haemorrhage in the cavity forms a clot which is eventually replaced by bone,⁽²⁵⁾ offering a good prognosis.⁽²³⁾ It is believed that in some cases there may be a spontaneous resolution,⁽⁴¹⁾ that might allow more aggressive lesions keep growing, leading to additional complications such as pathological fracture, involvement of adjacent teeth and need for wider surgical margins.⁽¹⁸⁾

Recurrence of TBC is assumed to be extremely rare.⁽³⁰⁾ If there are multiple cysts recurrence rates

would be high⁽⁴²⁾ or those associated with florid cemento-osseous dysplasia.⁽²³⁾

Case Report

A 23 years old Libyan male was referred to the Oral and Maxillofacial Surgery Department, Faculty of Dentistry, University of Tripoli, for evaluation and management of an asymptomatic unilocular radiolucency in the posterior right mandible which was discovered during routine OPG screening for orthodontic treatment, with an insignificant medical history.

On extra oral and intra oral clinical examination no any pathological abnormality detected, the teeth related to lesion were vital and immobile as well as the facial symmetrical was observed.

On radiographic examination, OPG revealed well defined unilocular radiolucent lesion measuring approximately 3cm.x1.5cm. that extended from the distal root of the right lower first molar to the distal aspect of right lower third molar (fig.1), with scalloped superior border between the roots (fig.2). Aspiration with 19 gauge disposable needle showed negative result. On the basis of these findings the differential diagnosis was keratocystic odontogenic tumor, ameloblastoma and traumatic bone cyst.

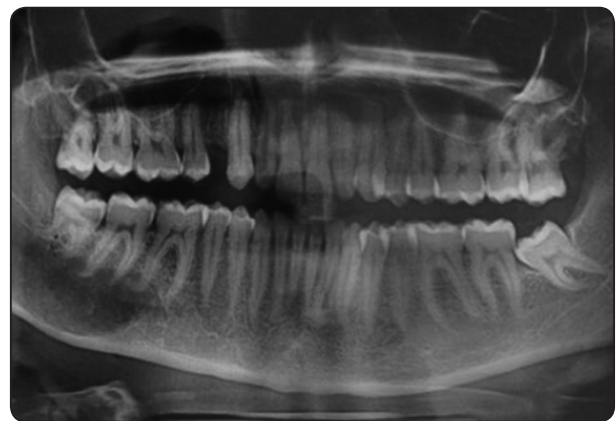


Fig. (1) Preoperative Orthopantomograph showing well defined unilocular radiolucent lesion that extended from the distal root of the right lower first molar to the distal aspect of right lower third molar.

Surgical exploration procedure was done under general anesthesia, intra-oral incision was done extended from the right lower first premolar to the retromolar area, a mucoperiosteal flap was reflected and bone window was created buccally. Empty bone cavity with small amount of serosanguineous fluid was found without any epithelial lining. The entire bony wall was curetted carefully to stimulate bleeding and to remove thin connective tissue lining, irrigated well and closure of the flap was done by using 4/0 absorbable suture. A small amount of tissue fragments of viable bone and scant soft tissues curetted was sent for histopathological examination. The histopathologic result revealed fibrous connective tissue, with interspersed bony

trabecular pattern without any epithelial lining, few areas of hemorrhage and intense inflammatory cell infiltrate (fig.3). Correlation between clinical, radiographic and histopathological result along with the findings of surgical exploration were confirmed the final diagnosis of the lesion was TBC. Furthermore, the final diagnosis was confirmed after regression of the lesion as well as evidence of bone regeneration with follow-up without recurrence after one year (fig.4 and 5).



Fig. (2) Preoperative Orthopantomograph showing scalloped superior border between the roots.

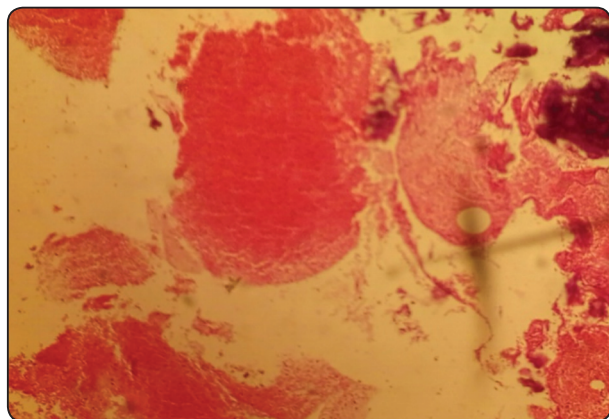


Fig. (3) A photomicrograph showing fibrous connective tissue, few areas of hemorrhage and intense inflammatory cell infiltrate (H&E stain, x40).



Fig. (4) Six months follow-up orthopantomograph showing new bone formation.

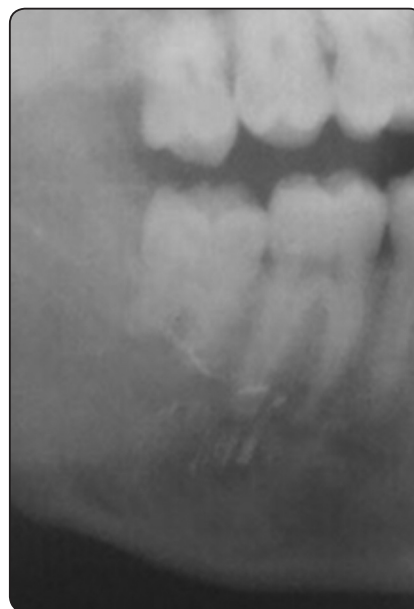


Fig. (5) One year follow-up orthopantomograph showing regression of the lesion, progressive osseous regeneration with no evidence of recurrence.

DISCUSSION

The TBC is an uncommon nonepithelial lined cavity of the jaws,⁽⁴³⁾ characterized by an empty bone cavity⁽¹¹⁾ or contain blood or straw-colored fluid.⁽¹⁾ In the presented case, empty bone cavity with small amount of serosanguineous fluid was found without any epithelial lining, this finding was found in most previous studies particularly in the mandibular lesions. It usually occurs in the metaphyseal region of long bones and it is unusual in the maxillofacial region, with a prevalence of 0.5–1.2% of all jaw cysts.⁽¹³⁾ However, TBC is uncommon and coincide with many previous researches. TBCs are considered almost exclusively mandibular lesions, with a preference for the posterior areas (body and ramus),⁽⁴⁴⁾ 95% TBCs occur in the body of mandible⁽⁸⁾ or symphysis area with only a few cases reported in the condyle.⁽²⁴⁾ However, in our case the lesion was located in the mandibular molars region in agreement with many previous literatures. TBCs are generally detected in patients in the second and third decades of life, although in 15% it was found above 40 years of age.⁽⁵⁾ In the presented case the lesion was diagnosed at the age of 23 years old which fall within age range in which TBCs are most commonly reported. The sex distribution is reported to be quite even⁽³¹⁾ or men are affected somewhat more frequently.⁽⁴⁵⁾ our patient in this presented case was male is inconsistent with Cortell-Ballester et al 2009 who reported female predominant.⁽²³⁾ The pathogenesis of traumatic bone cyst remains a matter of controversy.⁽⁴⁶⁾ However, there are many etiological theories postulated that has been suggested to elaborate this lesion. Among them the mostly accepted theory is the trauma hemorrhagic theory which explains its clinical & histological features.⁽³¹⁾ Nevertheless, the traumatic etiology hypothesis is challenged by the fact that more than 50% of cases have no traumatic history.⁽⁷⁾ Only 23.8% of the patients reported antecedents of orofacial traumatism in the retrospective study of Ballester et al.⁽²³⁾ Our patient did not give any evident history of trauma.

Furthermore, the trauma-hemorrhage theory may be an explanation because the mandible, which has more cortical bone, repairs itself more slowly compared to the maxilla. Clinically, the lesion is asymptomatic in the majority of cases and⁽⁴⁷⁾ may be incidentally diagnosed on routine dental treatment.⁽⁵⁾ our case was absolutely asymptomatic and coincide with Paulo Ricardo et al study(2012)⁽⁴⁾ and most previous studies. Teeth vitality was notified in most cases.⁽⁴⁸⁾ In our case report, the teeth involved were vital and did not show root resorption, this finding is in agreement with Lago CA et al 2006⁽⁴⁹⁾ and Neuschl M. et al. 2014.⁽⁴⁰⁾ Radiographic examination usually reveals a unilocular homogeneous radiolucency⁽⁵⁰⁾ with a well-circumscribed margin.⁽⁵¹⁾ our findings were similar and coincide with, Naveillie, Shaffer, Lucas C, and Hansen. Characteristic for the TBC is the “scalloping effect” when extending between the roots of the teeth⁽⁵²⁾ as we found in our presented case.

Most of the histopathologic findings reveal fibrous connective tissue and normal bone. There is no any evidence of an epithelial lining. The lesion may exhibit areas of vascularity, fibrin, erythrocytes and occasional giant cells adjacent to the bone surface,⁽²⁵⁾ most of these features were evident in our presented case and is in accordance with observation of MacDonald-Jankowski D⁽³¹⁾, Kaugars⁽⁵³⁾ and Hansen⁽⁷⁾. Aspiration result in cases of TBC, a positive aspirate of straw colored fluid is observed.⁽³⁵⁾ The amount of fluid diminishes with the age of the lesion, and the lesion eventually becomes empty.⁽¹⁹⁾ In our case aspiration biopsy from the lesion showed no cystic fluid and air bubbles with some blood, this findings were coincided with L. K. Surej Kumar et al. 2015.⁽⁵⁾ and Kumar Pushpanshu et al. 2013.⁽²⁴⁾

Diagnosis of Traumatic bone cyst prior to surgical intervention is greatly difficult in most of the instances.⁽⁵⁾ Because of a lack of unique clinical and radiographic features, it is important to establish the differential diagnosis between TBCs and other radiolucent lesions of the jaws.⁽²⁴⁾ Therefore, TBCs can make differential diagnosis with aneurysmal

bone cysts but (form very expansile soap-bubble radiolucencies and usually in the mandibular ramus and angle. Active and aggressive cysts tend to recur), dentigerous cysts but (contain the crown of an unerupted tooth which is usually displaced). Keratocystic odontogenic tumors but (most lesions, are unilocular, with as many as 40% noted adjacent to the crown of an unerupted tooth and 50% of mandibular lesions produce buccal expansion, with a propensity for root divergence). Calcifying cystic odontogenic tumor but (Because calcifications are usually small, lesions tend to occur as a diffuse radiolucency with faint flecks calcified structures, lesions have indistinct lines of demarcation with the surrounding bone and usually occurs over unerupted teeth it may be a radiolucent or mixed unilocular lesion). Ameloblastomas but (It appears radiographically as a multilocular radiolucency and usually presents between ages 30 and 50 years old). Odontogenic myxoma but (a radiolucent area with a soap-bubble or honeycomb appearance and show gross expansion of the mandible). Focal osteoporotic bone marrow defect but (The radiographic appearance is variable, ranging from a rounded, barely noticeable mixed radiolucent-opaque lesion to a diffuse, irregularly shaped radiolucent area with faint wisps of a trabecular pattern). Intraosseous vascular malformations but (usually diagnosed with imaging studies such as Doppler angiography or contrast time-lapse angiography). Central giant cell lesions but (on occlusal radiographs, often exhibit complete cortical bone loss, movement of associated teeth and resorption of tooth roots is commonly observed) and early stage of periapical cemental dysplasia but (mostly black females are affected and lesions are multiple and is considerably more common in the anterior mandible).

The definitive diagnosis is mainly based on⁽⁴⁹⁾ the analysis of clinical features, radiological features, histological features and surgical exploration findings.⁽³⁵⁾ However, the clinical, radiographic, histopathological findings and surgical exploration of the presented case that fulfill the diagnostic

criteria, were enough to confirm the final diagnosis of TBC in the right mandible.

The widely recommended treatment for TBCs is surgical exploration followed by curettage of the bony walls.⁽⁵⁴⁾ Surgical exploration was proved not only essential in making the right diagnosis,⁽⁵⁵⁾ but also favors progressive bone regeneration after formation of a stable blood clot, offering a good prognosis.⁽²³⁾ In our case, surgical exploration and curettage of bone walls was performed under general anaesthesia.

Recurrence of TBC is assumed to be extremely rare,⁽²⁶⁾ a recent study has reported the recurrence rate of TBC in facial bone to be about 20%.⁽⁵⁶⁾ On recurrence, a marked increase in the size with multilocular appearance.⁽⁵⁷⁾ If present, recurrence usually occur within three months after surgery.⁽²⁰⁾ Our sixth month postoperative radiograph did not show any recurrence and revealed excellent wound healing and in our presented case study simple curettage of the walls of the bone cavity and surgical exploration produced favorable results characterized by regression of the lesion and progressive osseous regeneration within 6 months duration with no evidence of recurrence even one year after the surgical treatment and our patient is still under observation and follow up :

CONCLUSION

Diagnosis of TBC prior to surgical intervention is a challenge in most of the instances, as the lesion is asymptomatic and usually discovered on routine radiographic examination for dental treatment tend to be misdiagnosed with many tumors and cystic lesions of the jaws. Surgical exploration followed by careful curettage of the bony walls has been the most widely recommended treatment for TBCs. It serves as a diagnostic tool and as a definitive treatment by inducing fresh bleeding which undergo organization and followed by formation of new bone. Clinical and radiological follow up is obligatory to ensure complete osseous healing after 6-12 months and avoid possible recurrences.

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