AN UNUSUAL PSEUDOCYST OF THE JAW: A CASE REPORT WITH A BRIEF REVIEW

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ABSTRACT
Solitary bone cyst (SBC), a relatively uncommon lesion of the jawbones. It is a type of the so-called “pseudocysts” of the jaws since the bone cavity is not lined with proper cystic membrane. Because of the numerous synonyms coined with this lesion make it obvious that there has been much disagreement on the pathogenesis of the lesion. It is an uncommon intraosseous non-neoplastic lesion of the jaws that almost affects patients in the second decade of life and have an excellent prognosis. It is an asymptomatic, slow growing, usually non-expansile lesion commonly diagnosed incidentally during routine radiographic examination of the jaw bones. The present literature reports a case of SBC in 24 year old male patient in posterior mandibular region along with a short review.

KEYWORDS: Solitary bone cyst, pseudocyst, etiopathogenesis.

INTRODUCTION
Solitary bone cyst (SBC) was first recognized by “Virchow” in 1876. Later it was described by “Lucas” in 1929. In dentistry, “Blum” reported first three cases in 1932. “Jaffe and Lichenstein” gave a discussion on this in 1942.1,2 It is known by numerous other names. These names include: traumatic bone cyst, traumatic bone cavity, simple bone cyst, idiopathic bone cyst, and, hemorrhagic bone cyst, single chambered tumor-like lesions.1,2

The international histological classification adopted by the World Health Organisation for odontogenic tumours uses the term “solitary bone cyst” however the term “traumatic bone cyst” (TBC) is more widely used in the literature. The WHO classification describes TBC as a non-neoplastic osseous lesion because it shows no epithelial lining, which differentiates this lesion from the true cysts.3 The multitude of names applied to this lesion attests to lack of understanding of true etiology and pathogenesis.

CASE REPORT
A 24 year old male was referred by his dentist to Oral and Maxillofacial surgery with a chief complaint of painless swelling since three months which was gradually increasing in size. On extra oral examination evidence of facial swelling was noted on left angle of the mandible and was approximately 4 x 2.5 cm in size, oval in shape, bony hard consistency, non-tender with normal overlying mucosa. (Figure no. 1) Intra oral examination revealed swelling extending from anterior border of master to posterior border of mandible. (Figure no. 2) No cortical expansion was noted. Correlating with the history and clinical examination a provisional diagnosis of odontogenic keratocyst was given.

Figure no. 1: Extra oral examination showing facial swelling on left side of the jaw.
Intra oral examination revealed swelling extending from anterior border of master to posterior border of mandible.

Panoramic and lateral oblique view radiograph revealed an oval, partly well-defined unilocular radiolucency extending from 36 to coronoid process. (Figure no. 3) FNAC of the lesion was performed which yielded negative aspiration. The patient was posted for surgical exploration under general anesthesia and the lesion was found to be an empty cavity, with no evidence of epithelial lining and containing very little amount of fluid mixed with blood.

OPG revealed an oval, partly well-defined unilocular radiolucency extending from 36 to coronoid process.

Curettage of the cavity was performed and the excised bone done to enter the bony cavity was sent for histopathological examination which yielded cystic lumen with no epithelial lining but surrounded by loose to more dense peripheral capsule. (Figure no. 4) The subadjacent regions of capsule is loose vascular fibrous tissue with areas of haemorrhage, extravasated RBCs, few inflammatory cells, scattered giant cells and plump fibroblasts. (Figure no. 5) The decalcified H & E stain section of bone shows loose vascular fibrous tissue with areas of osteoclastic cavity. (Figure no. 6) Thus, on the basis of clinical, radiological and histopathological findings a final diagnosis of Solitary bone cyst was made.
DISCUSSION

In the present case of SBC, the diagnosis is well documented radiographically and histopathologically. It is an interesting case of possible iatrogenic origin which is also located in a rather unusually site, the left ramus of the mandible.

SBC is not a common lesion. In a literature review, Hoffmeister and Harle in 1985 showed 0.6% of frequency. While Sheffield showed 0.5% of frequency as noted in literature by Jones and Franklin in 2006.6,7 Howe (1965) determined certain criteria for including SBC cases. It represents like cyst should be single, have no epithelial lining, no evidence of acute or prolonged infection, contain principally fluid and no soft tissue, walls should be of bone which is hard although possibly thin in parts.8 SBC in the jaws may affect patients between the ages of 2 and 75 years, but 56 to 70% of the cases present in the second decade of life and only 15% of the patients are more than 40 years old. Men are affected more than women (M: F 3:2), but one series reported by Hansen et al 1974 and Saito et al 1992 suggested that SBC were more common in women in the older age group.6,9,10

Majority of cases of SBC generally occurs in the mandible. However, Howe reported one case in maxilla.8 The majority of cases are diagnosed fortuitously and almost all of these were chance radiographic findings. Swelling is a symptom in 27%, pain in 10%, labial paraesthesia in 2%. Some cases show both pain and swelling. Over half of the patients gave a history of significant trauma to the area, the time lag between injury and diagnosis varied from 1 month to 20 years.6

Other unusual symptoms include tooth sensitivity, fistulas, delayed eruption of permanent teeth, displacement of the inferior dental canal and pathologic fracture of the mandible. Adjacent to the lesion related teeth are usually vital and there is no mobility associated with it.1

Radiographically it is manifested as a well-defined, radiolucent, unilocular area between 61 and 79%, which occasionally presents a typical festooned pattern around the apexes of the adjacent teeth5,6 An occlusal view shows the radiolucency extending along cancellous bone.2,6 However, 21% have radiopaque foci and 7% may show cloudiness. The border, although irregular, can vary from well-defined to a complete absence of cortical outline. Copete et al (1998) described a characteristic cone shaped radiographic morphology. Some case show one lateral margin of the lesion formed an angular interface with normal bone with two planes converging at a 45° angle to produce a sharp cone effect. Most often (66%) the cone pointed anteriorly towards the midline.11

The growth plate is normally not affected by these expansive cysts. The cortex of the bone have a tendency to get thin and weak. This thinned cortex is an area of structural weakness and is prone for a pathological fracture. The ‘fallen leave’ sign is indicative for SBC.4 Scalloping or interdigitation between the roots of teeth was a common feature in 44% to 68% of the cases. It can also be away from the teeth.2,6

Loss of lamina dura is predominant in patients over 30 years of age and there is minimal involvement in younger people. Displacement of teeth and root resorption are rare although 9 to 22% cases were reported in few literature.2,6 Occasionally bone expansion can be noted buccally and lingually. On review of cases 63% showed some degree of marginal condensation but not as sharp or opaque as with radicular cysts.6 Bone septa may be present and the lesions are sometimes interpreted as multilocular, which can lead to an erroneous diagnosis.6

The pathogenesis of SBC is not clear although number of theories have been proposed. Olech et al (1951) suggested the following possible pathogenesis, based upon traumatic aetiology and Howe proposed an essentially similar natural history. Olech et al introduced their hypothesis that following trauma to a bone, causes intramedullary haemorrhage, a failure of early organization of haematoma in some of marrow spaces and subsequent liquefaction of the clot can lead to formation of traumatic cyst. Cyst develops only after injury to those areas of bone where spongy bone containing haemopoietic marrow is enclosed in a heavy compact cortical layer. This explains the most frequent sites in metaphyses of long bones and in mandible and it develops in young individuals and also explains the presence of blood within the cavity at the time of surgical exploration.12,13

This opens the possibility that microtrauma of teeth and the alveolar ridge are involved in the pathogenesis of traumatic bone cysts.13 According to Olech et al, the primary haematoma will not be organized if it is not in contact with reactive and fibrous connective tissue and this will not be present if intramedullary haemorrhage has led to necrosis of bone marrow itself and related endosteum. The trabeculae of medullary bone are slowly resorbed by osteoclastic activity on their opposite surfaces and by the time viable connective tissue gains contact with haematomas, the latter has liquefied.12

Beasley (1976) observed areas of haemorrhage associated with necrosis and myxoid degenerative changes. Majority of SBCs contain air or some other gas, some may contain blood or serosanguineous fluid which...
tends to support concept of haematoma break down.\textsuperscript{14} This break down produces a local rise in osmotic pressure which leads to transudation into cyst fluid. In presence of intact cortical bone there is increase in intrasosseous pressure which leads to resorption of bone by osteoclastic activity and sometimes swelling by concurrent periosteal bone deposition. As transudation occurs, fluid is diluted so that intracystic pressure drops but further bleeding may be responsible for progression of lesion. Once the bleeding is stopped, gradual absorption of serous fluid in the cavity occurs which becomes empty.\textsuperscript{19}

When the cyst is operated SBCs are generally empty. Sometimes blood, serosanguineous or serous fluid may be present.\textsuperscript{6}

Kuhmichel & Bouloux have noted that such content in the bone cavity may represent different stages in the development of traumatic bone cysts.\textsuperscript{13}

SBCs consists of loose vascular fibrous tissue membrane of variable thickness with no epithelial lining, fragments of fibrin with enmeshed red cells may be seen. Haemorrhage and haemosiderin pigment are present and scattered small multinucleated cells are found. Long standing cases are densely fibrous. The adjacent bone shows osteoclastic resorption on its inner surface. Beasley (1976) described areas of haemorrhage associated with necrotic tissue or tissue showing myxoid degeneration which occurred in cavities adjacent to areas of bone resorption.\textsuperscript{14}

When cysts are multiple, empty cystic cavities appeared to be secondary to cystic degenerative changes in a pre-existing lesion.\textsuperscript{6}

Treatment is surgical. The surgical operation consists of evacuation of the content, cocheleation of the cavity in order to stimulate bleeding in the cavity. The wound is then sutured. This is followed by the formation/creation and organisation of a clot, and healing by the formation of new bone. Some TBC heal spontaneously without any intervention. This is probably the reason why they are rarely found in older age groups. Recurrences are rare after surgical treatment.

CONCLUSION

In everyday practice, during work with patients and when analysing radiograph images, it is possible to come across formations which radiographically and clinically have the appearance of cystic changes. In some cases it is not immediately possible to determine the cause. For example there is no non-vital tooth nearby, remaining root, impacted tooth, the case history shows no evidence of trauma in the affected area, and neither are there any significant subjective disorders in the patient. If the case history shows the existence of trauma in the affected area then the decision is probably somewhat easier. This is the point when we can differentially diagnostically suspect a traumatic cyst. In order to decide on the final diagnosis it is then necessary to carry out exploration of the area and cocheleation of the cavity, which at the same time is the main therapeutic procedure.

A histopathological diagnosis will confirm the existence of a TBC if during the operation some tissue is removed from the cavity. If no tissue is found in the cavity for histopathological examination a decision on diagnosis of a TBC will depend on individual experience. Knowledge of the symptomatology, correct interpretation of the radiograph and correct recording of case history will be helpful when deciding on the diagnosis and relevant treatment.

REFERENCES


