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"Imitating The Aggressive": A Case Report Of Cone-Shaped Simple Bone Cyst With Diagnostic And Therapeutic Insight"

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Abstract:
A simple bone cyst (SBC), also referred to as an idiopathic bone cavity, is a non-neoplastic, intraosseous pseudocyst characterized by the absence of an epithelial lining. While it predominantly affects long bones, its presence in the jaw especially the mandible is relatively uncommon. SBCs are most frequently observed in adolescents – second decade of life and are typically discovered incidentally on routine radiographic imaging due to their asymptomatic nature. Radiographically, they present as well-defined, unilocular radiolucent areas with scalloped borders, often extending between the roots of adjacent teeth without causing root resorption or displacement. The exact cause of SBC remains unclear, although trauma is often suggested as a contributing factor. Histological examination usually reveals an empty cavity or one containing a small amount of serosanguinous fluid, lined by a thin fibrous connective tissue. Management typically involves surgical exploration and curettage, is the gold standard treatment as it allows both diagnosis and treatment which promotes spontaneous bone healing. The prognosis is generally favorable, with a low rate of recurrence. Awareness of the clinical and radiographic characteristics of SBC is crucial for accurate diagnosis and effective treatment planning.

Key Word: *Traumatic bone cyst, Pseudocyst of Jaw, Cone shaped radiolucency, multilocular radiolucency, Giant cell lesion, Idiopathic bone cavity.*

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I. Introduction

Simple bone cysts (SBCs) are benign, nonneoplastic intraosseous cavities devoid of an epithelial lining, often surrounded by intact bone and either empty or filled with fluid or connective tissue. It was first described as a distinct pathological entity in 1929 by Lucas and Blum(1), SBCs are considered as pseudocysts due to the absence of an epithelial component, which differentiates them from true cysts. The WHO classified this as a part of Giant cell lesion which includes central / peripheral giant cell granuloma, cherubism and Aneurysmal bone cyst(2).

In the literature, SBCs have been referred to by various names, including traumatic bone cysts, solitary bone cysts, idiopathic bone cavities, hemorrhagic bone cysts, unicameral bone cysts, and extravasation cysts. They most commonly affect individuals in their second decade of life, with no clear sex predilection some female predilection has been reported in some literatures.(3). In the maxillofacial region, the mandibular body and symphysis are the most frequently affected areas, while maxillary and zygomatic involvement is relatively rare. These lesions are often asymptomatic and typically discovered incidentally during routine radiographic examinations(4).

The pathogenesis of SBCs remains unclear. Several theories have been proposed, and it remains inconclusive. Neither of theories proves to be evident as it has multitude of names which was forementioned. It attests to lack of understanding the true etiology and pathogenesis behind it(5,6).

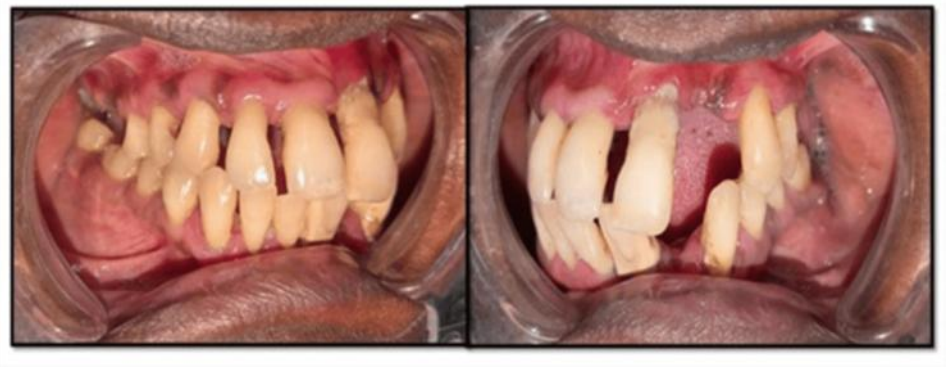
Radiographically, SBCs typically appear as well-demarcated unilocular radiolucent lession. A characteristic scalloping pattern between the roots of adjacent teeth may be observed when the lesion extends interdentally(7). The differential diagnosis includes Radicular cyst, odontogenic keratocysts, Ameloblastomas, central giant cell granulomas, odontogenic myxomas, Cherubism, Focal osteoporotic bone marrow defect, and other intraosseous vascular malformations and neoplasms(8). Surgical curettage remains the diagnostic and therapeutic gold standard. It induces bleeding within the cavity, promoting bone regeneration, which typically occurs over a 6 to 12-month period. The recurrence rate is extremely low, and the prognosis is generally favorable.

The aim of this case report is to emphasize and document the unusual occurrence of this lesion and rare radiographical appearance which can lead to misdiagnosis and over treatment of the same.

II. Case Report

A female in the early sixty presented with complaint of missing teeth and mobility in relation to lower anterior region, persisting for past two years. Clinical examination revealed tooth mobility and periodontal opinion was sought for further assessment. A routine panoramic radiograph was performed to evaluate the extent of alveolar bone loss. Incidentally, the radiograph revealed a well-defined, irregular radiolucency with characteristic interdental scalloping, extending from the distal aspect of 48 to 44 and 36 to 34 region, (cone shaped margins with apex towards the anterior region) below the apices of the involved teeth with no root resorption and tooth mobility bilaterally. For further confirmation on its extent and to assess the lesion in 3dimension – CBCT was taken. Though the lesion was extended well beneath the canal, patient hasn't reported on paresthesia and the integrity of the lower border of the mandible was preserved. Upon further inquiry, the patient admitted to being aware of the lesion for approximately one year but had delayed seeking treatment due to the absence of pain. On comparing the OPG the lesion was extended in the distal aspect towards the ramus of mandible (Figure1,2, 3)

Figure 1: Intra oral picture which shows no significant swelling, or vestibule obliteration.



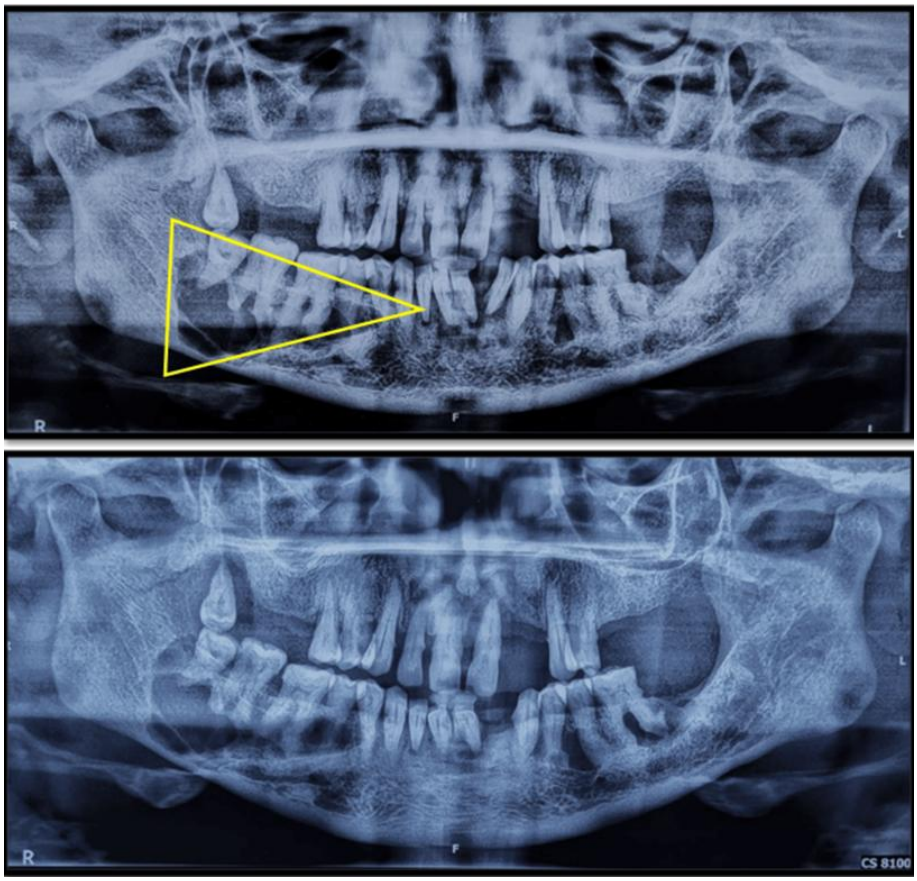


Figure 2: Sequential OPGs which reveals cone shaped radiolucency in the posterior mandible

Extraoral and intraoral clinical examinations revealed no abnormalities. There were no signs of swelling, buccal vestibule obliteration, bony crepitus or any carious lesions with no evidence paresthesia. The patient was asymptomatic and did not report any discomfort. Her medical history was non-contributory, with no reported history of trauma. Vitality testing confirmed that all involved teeth were vital. Aspiration of the lesion yielded a straw-colored serosanguinous fluid. Based on these findings, the patient was scheduled for a surgical biopsy to establish a definitive diagnosis. A crevicular incision was made, followed by elevation of a buccal mucoperiosteal flap. Surgical access to the lesion was achieved through a buccal corticotomy using a round bur. Upon entry, an empty cavity with internal septations was observed, with no epithelial lining present, confirming the diagnosis of a simple bone cyst (SBC) (Figure 4). Small piece of bone which was taken from septations was sent for Histopathological examination (Figure 5). The cavity was thoroughly curetted, and the flap was repositioned and sutured using 3-0 silk sutures. The postoperative recovery was uneventful.

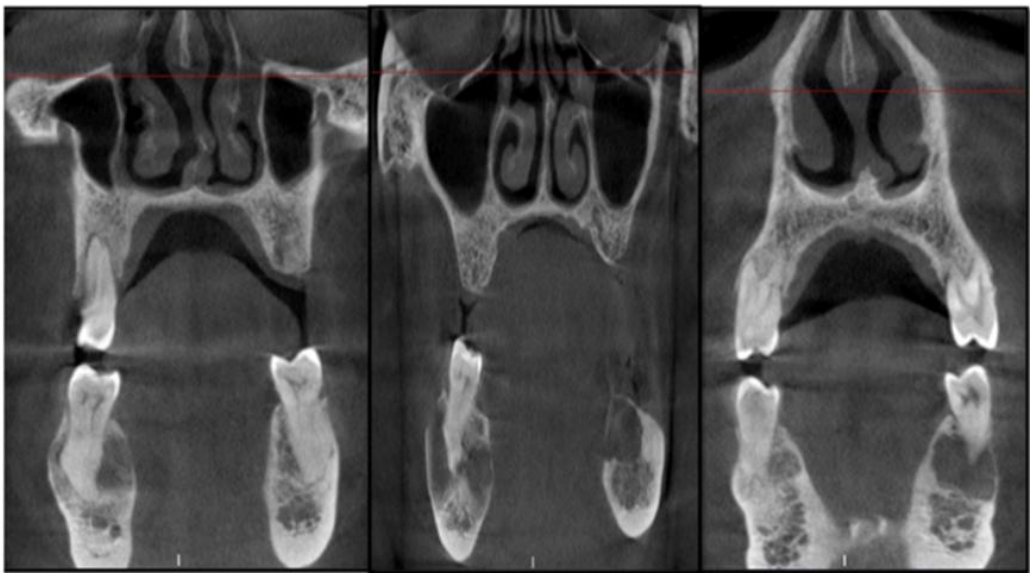


Figure 3: CBCT coronal section shows bilateral involvement with intact lower border of mandible and extension beneath the canal

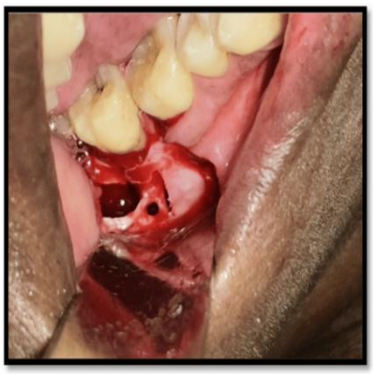


Figure 4: Intra-cystic septations with empty cavity and without epithelial lining

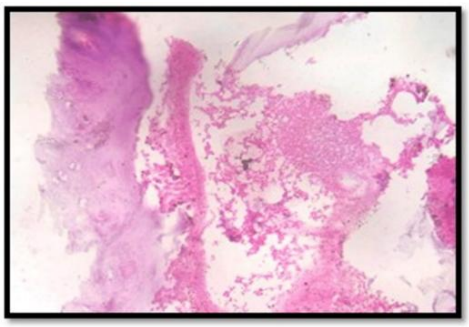


Figure 5: Histopathological section which reveals the normal architecture of the bone

III. Discussion

Simple bone cysts have been known for many years, nevertheless, still currently they are underrepresented in the available literature with respect to true incidence, and most likely due to their asymptomatic nature and the fact that spontaneous resolution occurs in some of these cases. Though consistent discrepancies exist in the literature, the lesion seems to be most commonly seen in the second decade of life with a tendency to occur in the posterior mandible. The pathogenesis of the lesion is not well understood and remains a subject of twin debate between various authors.

Theories for pathogenesis postulated in the literatures:

Some theorize they may be the result of resolution of a traumatic hematoma or development of venous obstruction while others suggest a developmental defect; neither of which has been conclusively proven.

- Trauma- Hemorrhagic theory – trauma induced intraosseous hematoma that fails to organize and does not undergo remodeling, but the liquefaction predominates resulting in bony defect.
- Traumatic Theory suggests, Post trauma clot breaks down and leaves an empty defect.
- Developmental anomalies involving the incorporation of synovial tissue, and compromised venous drainage leading to interstitial fluid accumulation and subsequent bone resorption.
- Other less commonly proposed theories include ischemic necrosis, cystic degeneration of benign bone tumors, low-grade infections, calcium metabolism disorders, Defects in local bone development, Osteoclastic resorption due to decreased tissue pH occurred due to venous stasis.

Radiographically, simple bone cysts may present in a variety of ways from smooth unilocular radiolucency to irregular, scalloped lesions with multi locular appearance which may mimic an odontogenic cyst or tumor. As illustrated in this case report, the lesion takes on a cone or Flask-like shape, as it is typically described in Shafer's textbook and has no literature documentation (9). However, atypical presentations can mimic aggressive jaw lesions, leading to misdiagnosis and over-treatment. In our case, the presence of internal septations created a multilocular appearance, closely resembling odontogenic keratocyst, ameloblastoma, or aneurysmal bone cyst which was misleading our provisional diagnosis. It is very imperative to take a note on Extension of the lesion in the presented case. It was positioned well beneath the canal which was note-worthy, since the all reported cases of SBCs were positioned above the canal.

Diagnostic challenge:

Clinical correlations of this peculiar cone / flask -shaped radiographic appearance with bilateral presentation haven't been documented in published case reports and subsequently often leads to misdiagnosis, especially with a vague or no history of trauma and with minimal to no clinical symptoms that contributes to the observed variability in radiographic aspects, which often presents a diagnostic dilemma. This variation in radiographic features contributes to frequent misdiagnosis. SBCs may be mistakenly identified as odontogenic keratocysts, unicystic ameloblastomas, or central giant cell granulomas — all of which often prompt more radical and extensive treatment approaches, including resection or enucleation under general anesthesia.

There is considerable diagnostic uncertainty from a clinician's point of view, which reinforces the need for clinical, radiology and surgical merge. This case helps to highlight the need to report these lesions, not just in being able to support classical textbook descriptions with clinical cases, but also in being able to help educate and warn clinicians not to overtreat. The way we had a representation of a classic case also supports the need to document it, as there are fewer classic cases that have been published in literature, and documenting cases like this adds credibility when the lesion was well seen by the imaging, surgical findings, and follow-up, which occurred in this report.

This case reinforces that while SBCs are benign and often self-limiting, their radiographic presentations are not always "simple." A cautious interpretation of radiological features, especially when atypical, is essential to avoid unnecessary aggressive treatment.

Table 1, summarizes the documented SBCs in the literature. In which we could able to infer it has a female sex predilection with unilocular radiolucency and occurrence in 2nd and 3rd decades of their life which was supported by various literatures. This was again contradicting with our case report with regard to age, appearance and site of occurrence.

Table1:

AUTHOR/YEAR	AGE	M:F RATIO	SITE OF OCCURENCE	RADIOGRAPHIC APPEARANCE	ETIOPATHOGENESIS
Marques et al/ 2021[10]	14	M	Body of mandible	Regular/ well defined/ unilocular/ corticated border	No history/ Asymptomatic

Ishimoto et al/2016[4]	13	F	Anterior mandible	well defined/ multilocular/ scalloped border	No relevant history
Paolo et al/ 2024[11]	23	F	Body of the mandible	Regular/ well defined/ unilocular/ corticated border	Asymptomatic / intra oral swelling
Charan et al/ 2012[12]	30	F	Body of the mandible	Regular/ well defined/ unilocular/ corticated border	Asymptomatic/ intra oral swelling
Varun et al/[13]	21	F	Anterior mandible	Regular/ well defined/ unilocular/ corticated border	No history/ Asymptomatic

IV. Conclusion

1. SBCs are not necessarily associated with trauma – history. It may present symptomatic / asymptomatic
2. SBCs are filled with serosanguinous fluid, blood or empty with devoid of epithelium.
3. Shouldn't be taken as "simple" since it has transformation potential into sarcomas which has been documented in literatures (10).

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