Case Report

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Dentistry Section

Aneurysmal Bone Cyst with Ossifying Fibroma of the Mandible: A Case Report and Review of the Literature

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ABSTRACT

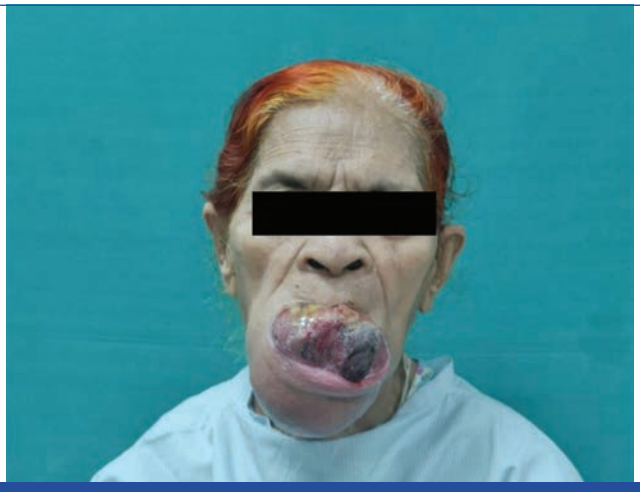
Aneurysmal Bone Cysts (ABCs) are uncommon benign bone lesions primarily affecting children and adolescents. They are distinguished by severe bone breakdown and expansive tissue growth, resulting in clinical symptoms and potential complications. ABCs typically occur in long bones, but reports have also documented their presence in the jaws, particularly the mandible. These lesions are commonly considered non-cancerous and are characterised by cystic or blood-filled chambers. There are two clinicopathological variations of ABC: primary ABC and secondary ABC. Primary ABC originates independently, while secondary ABC develops as a result of a pre-existing lesion such as a cyst, tumor, or Fibro-Osseous Lesions (FOL) like solitary bone cyst, ossifying fibroma, or giant cell granuloma. When ABC coexists with another bone lesion believed to be its precursor, it is referred to as an "ABC plus lesion". In this case report, a 75-year-old patient, presented with a painful growth in the lower anterior region of the jaw for the past 18 months. The diagnosis was established through clinical, radiological, and histopathological examinations. A complete surgical resection was performed, followed by uneventful reconstruction. Histopathological examination confirmed the presence of ABC with ossifying fibroma (ABC plus lesion). It is important to address ABC plus lesions as they can cause significant pain, deformity, and discomfort. Although non-cancerous, they can still disrupt normal bone structure and function. This case report emphasises the clinical, radiographic, and histopathological features of ABC plus lesions, aiding in disease identification.

Keywords: Benign, Bone breakdown, Odontogenic, Resection, Reconstruction

CASE REPORT

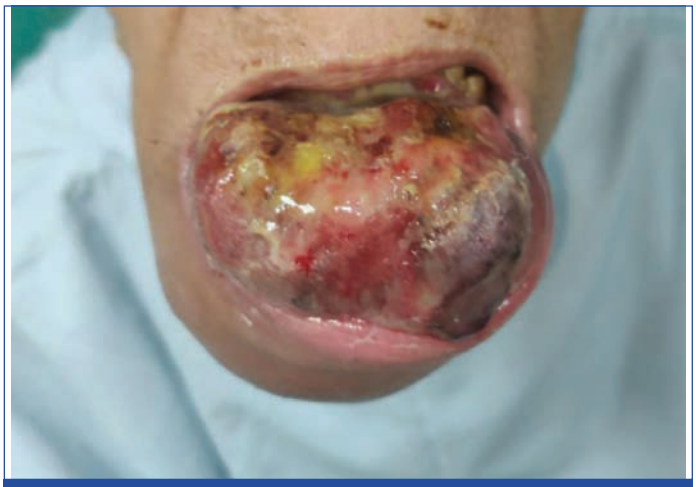
A 75-year-old female patient reported to the outpatient department with a growth over the lower front region of the jaw that had been present for approximately 18 months. Initially small, the growth had rapidly increased in size. The patient tested positive for Hepatitis B surface Antigen (HBsAg) and had a history of hypertension.

During the extraoral examination, a diffuse growth was observed over the anterior region of the mandible, extending anteroposteriorly from the left corner of the mouth to the right corner of the mouth, and superoinferiorly from the vermillion border of the upper lip to 3 cm above the inferior border of the mandible. The growth measured approximately 7.5×6 cm and had a firm consistency. It was non-tender [Table/Fig-1].



[Table/Fig-1]: Extraoral photograph of the patient.

Intraoral examination revealed insufficient mouth opening. A 7×5 cm lesion was observed, extending mesiodistally from the 45 to 35 region and superoinferiorly from the gingival border of the lower anterior teeth into the lingual and gingivolabial sulcus. The edges were smooth with well-defined borders, and the consistency ranged from soft to firm [Table/Fig-2].

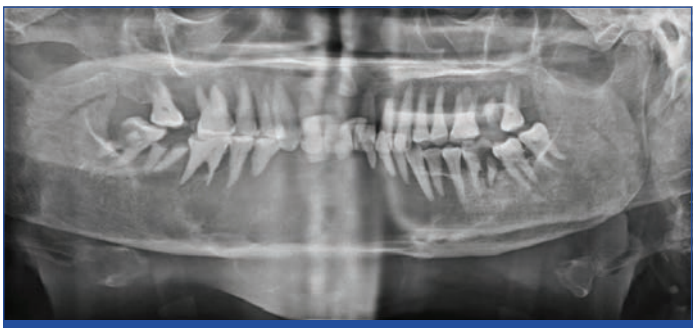


[Table/Fig-2]: Photograph showing lesion.

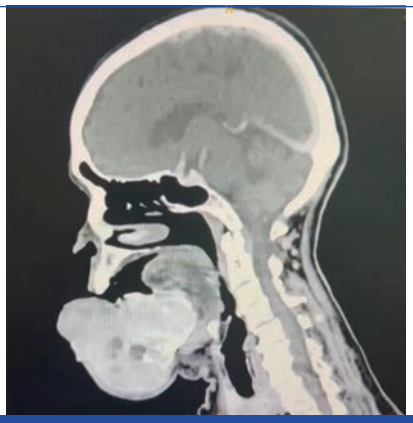
Radiographic examinations were performed. The Orthopantomogram (OPG) revealed a well-defined multilocular radiolucency in the mandibular symphysis region, extending from the 35 to 46 region [Table/Fig-3]. The Computed Tomography (CT) scan [Table/Fig-4] showed a 7.5×5.3×6.2 cm expansile large cystic lesion in the mandibular symphysis menti. A provisional diagnosis of Central Giant Cell Granuloma (CGCG) was made. The differential diagnosis at the time included ameloblastoma and keratocystic odontogenic tumour, also known as Odontogenic Keratocyst (OKC).

Under general anaesthesia, a segmental mandibulectomy was performed from the angle of the mandible (right) to the 38 region on the left side of the mandible. Reconstruction was carried out using a Pectoralis Major Myocutaneous Flap (PMMC) on the right side, and

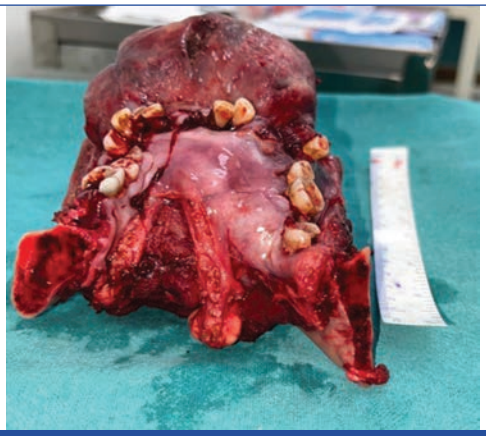
a tracheostomy was performed. The resected specimen was sent for histopathological examination [Table/Fig-5].



[Table/Fig-3]: Orthopantomogram (OPG) showing multilocular radiolucency in the lower anterior jaw.

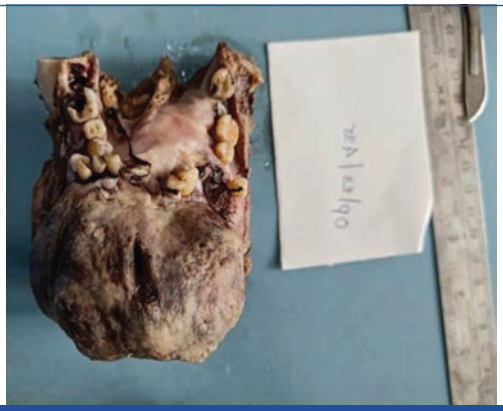


[Table/Fig-4]: Computed Tomography (CT) showing cystic lesion in mandibular symphysis menti.



[Table/Fig-5]: Resected specimen.

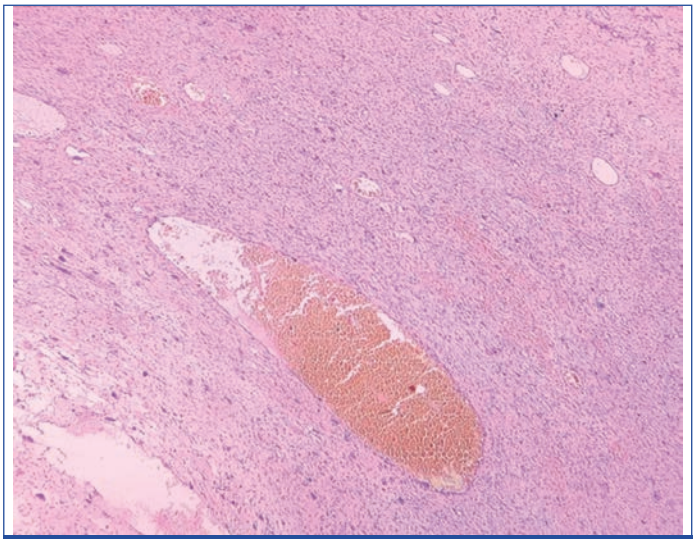
During gross examination, an exophytic greyish-black extensive lesion measuring 8×7×6 cm was observed on the lower anterior jaw [Table/Fig-6]. On the cut section, excessive bleeding was encountered, resembling a sponge soaked in blood with cavernous spaces [Table/Fig-7].



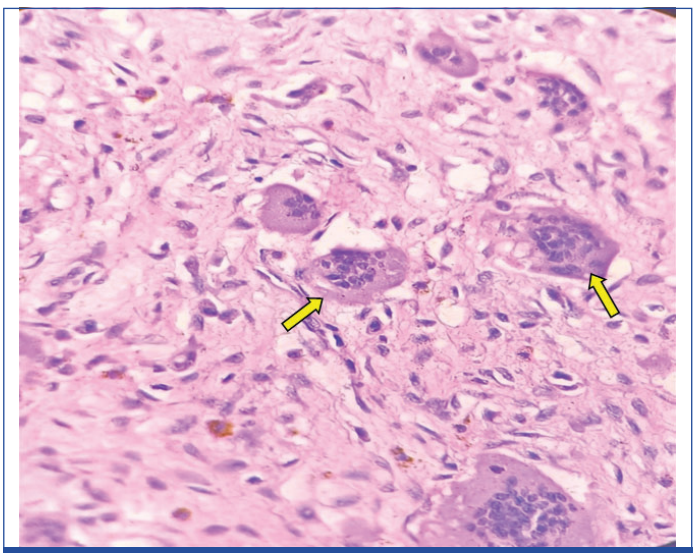
[Table/Fig-6]: Gross examination of the resected specimen.

[Table/Fig-7]: Cut section of the specimen showing large sinusoidal spaces filled with blood.

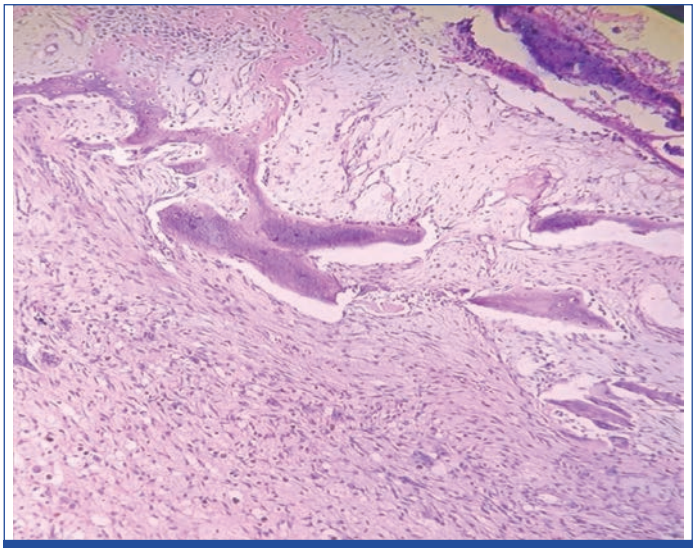
Histopathological examination of the Haematoxylin and Eosin (H&E) stained lesional tissue section showed a fibrocellular connective tissue stroma with large, cavernous or sinusoidal spaces filled with blood [Table/Fig-8]. The connective tissue stroma exhibited multi-nucleated giant cells with 10-15 nuclei and immature plump fibroblasts [Table/Fig-9]. Other sections revealed immature bony trabeculae lined by osteoblastic rimming and numerous osteocytes [Table/Fig-10]. The connective tissue stroma exhibited delicate interlacing collagen fibrils interspersed with a large number of actively proliferating fibroblasts [Table/Fig-11]. A final diagnosis of aneurysmal bone cyst with ossifying fibroma was made. No special stains were used in this case.



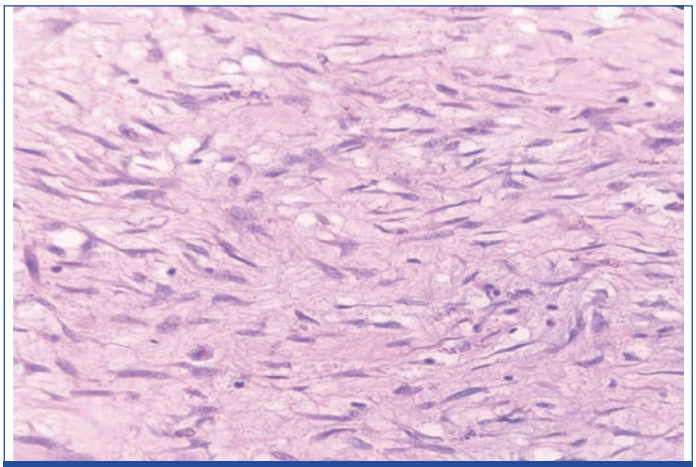
[Table/Fig-8]: Large cavernous spaces are seen in fibro cellular connective tissue stroma (H&E- 4x).



[Table/Fig-9]: Multi-nucleated giant cells with 10-15 nuclei (H&E- 40x).

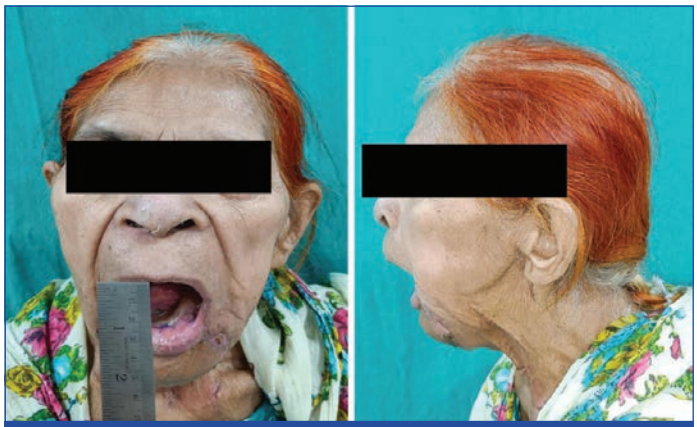


[Table/Fig-10]: Immature bony trabeculae lined by osteoblastic rimming (H&E-10x).



[Table/Fig-11]: Plump immature fibroblasts with interlacing collagen fibrils (H&E- 40x).

The patient experienced full recovery and had no pain or other symptoms after six months of follow-up. There was no evidence of recurrence [Table/Fig-12,13].



[Table/Fig-12]: Postoperative photograph-front view of the patient.
[Table/Fig-13]: Post-operative photograph-lateral view of the patient. (Images from left to right)

DISCUSSION

ABC is a rare non-cancerous osteolytic tumour of bone tissue characterised by multiple sponge-like, blood-containing areas of varying sizes. These areas often lack endothelial lining and contain bone elements and osteoclast-like giant cells [1,2]. ABCs are commonly seen in areas with greater marrow and venous content. Due to the low venous pressure in skull bones, ABCs can be unpredictable lesions [3]. They are usually found in the metaphysis of long bones, such as the tibia and femur (more than 50%), and in the spine (12-30%) [4]. ABCs are less common in the craniofacial skeleton (2-12%) [4-6]. However, the mandible is a common location for ABCs in the head-neck region [6].

ABC is divided into three categories. The traditional or vascular type presents as an expansile, rapidly expanding destructive lesion that causes cortical perforation and soft tissue invasion. The solid variant may appear as a small, painless growth initially detected as a radiolucent lesion on routine radiographs, or as a clinically significant tumour [7,8]. The mixed type exhibits characteristics of both the vascular (classic) and solid varieties. Rapid expansion and activation of stable lesions have been documented, suggesting that it may represent a transient phase of the lesion [8].

Clinicopathologically, ABC has two forms: primary (congenital or acquired) and secondary, which arises from a pre-existing lesion. Congenital ABC is determined by factors such as arteriovenous malformation, tooth growth, and tooth maturation during infancy. Acquired ABC is often associated with trauma. Secondary ABC can be linked to the progression of pre-existing lesions, such as a cyst, tumour, or fibro-osseous lesions like solitary bone cysts, ossifying fibromas, or giant cell granulomas [9].

Ossifying Fibroma (OF), a benign bone tumour usually referred to as a form of FOL, can affect both the mandible and maxilla, but it is more frequently observed in the mandible, accounting for 70-90% of cases [10]. Clinically, this tumour manifests as a slowly expanding intrabony tumour that often lacks symptoms and rarely grows to a size that causes facial asymmetry [11]. Only a few cases in the literature describe the close association between ossifying fibroma and ABCs, as in the present case [12]. In the facial bones, the occurrence of ABCs subsequent to ossifying fibroma is still a rare finding [13].

A literature review of thirty-two ABC-plus-lesions revealed that males are more commonly affected, and there is a higher propensity for mandibular involvement. Sixty-eight percent of the thirty-two ABC-plus-lesions were associated with FOLs, while giant cell lesions accounted for 32% of the cases. ABC presents with a wide range of clinical characteristics, ranging from a painless lesion identified through radiographic examination to a symptomatic (painful), expanding, and destructive pattern [14]. However, the majority of cases were painless, as was the situation in this case. In contrast to ABCs in long bones, ABC-plus-lesions typically cause discomfort and exhibit a tendency for rapid growth, whereas ABCs in other locations are often associated with malignant tumours such as osteosarcomas and chondrosarcomas [15].

The radiological appearance of jaw ABCs is highly variable. The lesion may exhibit bony growth, a cyst-like appearance resembling a soap bubble or honeycomb, or it may have an unconventional inflated appearance. The cortex may be perforated or destroyed, and a periosteal response may occur [16]. The lesion can appear as radiopaque, radiolucent, or mixed. In this case, a multiocular radiolucency was observed, causing expansion of the cortical plates and thinning of the inferior mandibular border. Root resorption in the affected teeth was also noted. However, the diagnosis based solely on radiographic examination is uncertain, as other lesions, such as ameloblastoma, odontogenic cysts or tumours, myxoma, or central haemangiomas, can have similar radiographic appearances [17].

ABC exhibits numerous sinusoidal spaces filled with blood in a fibrocellular connective tissue stroma, along with bone/osteoid material and multiple multinucleated giant cells. The presence of haemosiderin pigments is also variably observed, which are pathognomonic features of the vascular variant of ABC [18]. On the other hand, the solid variant shows foci of haemorrhage with numerous fibroblasts and fibrohistiocytes. Additionally, osteoclast-like giant cells, areas of osteoblastic differentiation with bone elements, and calcifying fibromyxoid tissue are present. The mixed variant demonstrates both solid and vascular characteristics. Aneurysmal bone cyst plus-lesions exhibit a combination of classic

(vascular) and solid forms, along with related lesions, featuring multiple vascular spaces in the fibrocellular stroma, multinucleated giant cells, and bone tissue production [19]. The histological findings in this case were consistent with the aforementioned criteria, indicating an ABC with OF.

The pathophysiology of ABCs is still a subject of debate. It could be caused by reactive vascular malformation, post-traumatic stress disorder, or hereditary susceptibility [20].

In most cases, total resection of the lesion is the preferred treatment modality for ABC plus-lesions. Surgical resection and curettage are considered the gold standard treatment. Diagnostic and therapeutic embolisation, curettage, block resection, radiation reconstruction, and systemic therapy with calcitonin are also utilised. Some studies have reported self-healing over a long follow-up period. For patients with aesthetic deformity, mandible discontinuity, or a high risk of fracture, early restoration of the defect using autogenous grafts has been recommended [5,14, 21-23].

In this case, surgical resection was performed as the treatment approach, and regular monitoring was conducted. No evidence of residual lesion was observed after six months of follow-up. The case reports of ABC plus-lesions in the head and neck region have been compiled in [Table/Fig-14] [3,24-41].

S. No.	Year	Author(s)	Age/ Sex	Site	Diagnosis (ABC plus)
1.	1964	Yarington CT et al., [24]	48/F	Maxilla	Giant cell (reparative granuloma)
2.	1970	Costas JB and Pietropinto J [25]	22/F	Mandible	Giant cell granuloma
3.	1971	Buraczewski J and Dabska M [26]	26/F	Mandible	Fibrous dysplasia
4.	1972	Ellis DJ and Walter PJ [27]	17/M	Maxilla	Cementifying fibroma
5.	1973	Oliver LP [28]	20/F	Mandible	Fibrous dysplasia
6.	1978	Bertrand G et al., [29]	28/M	Mandible	Fibrous dysplasia; Ossifying fibroma
7.	1980	El Deeb M at al., [30]	19/M	Mandible	Fibrous dysplasia
8.	1980	Goldmann ME and Sisson GA [31]	10/M	Maxilla	Fibrous dysplasia
9.	1984	Pankey ER et al., [32]	20/M	Mandible	Central giant cell granuloma
10.	1985	Robinson PD [33]	13/M	Mandible	Cementifying fibroma
			11/M	Maxilla	Cemento-ossifying fibroma
			41/M	Mandible	Cemento-ossifying fibroma
			11/M	Mandible	Cemento-ossifying fibroma
			14/F	Mandible	Cemento-ossifying fibroma
			18/F	Mandible	Benign osteoblastoma
			47/F	Mandible	Cemento-ossifying fibroma
11.	2010	Sun ZJ et al., [34]	12/F	Maxilla	Cemento-ossifying fibroma
			17/M	Mandible	Ossifying fibroma
			27/F	Mandible	Ossifying fibroma
			30/F	Mandible	Ossifying fibroma
			9/M	Mandible	Central giant cell granuloma
			14/M	Mandible	Cemento-ossifying fibroma
			7/M	Mandible	Cemento-ossifying fibroma
12.	2011	Sankaranarayanan S et al., [35]	6/F	Maxilla	Juvenile ossifying fibroma
13.	2011	Westbury SK et al., [36]	17/F	Maxilla	Central giant cell granuloma
14.	2011	Tabrizi R et al., [37]	26/M	Mandible	Non-ossifying fibroma
15.	2012	Henriques AC et al., [38]	21/M	Mandible	Ossifying fibroma
			18/M	Mandible	Giant cell lesion
16.	2014	Arora SS et al., [39]	61/M	Mandible	Giant cell granuloma
17.	2014	Moghe S et al., [3]	8/F	Maxilla	Ossifying fibroma
18.	2018	Sarode SC et al., [40]	10/M	Maxilla	Juvenile ossifying fibroma
19.	2022	Sonone A et al., [41]	17/M	Mandible	Cemento-ossifying fibroma

[Table/Fig-14]: Review of ABC plus case reports in the head and neck region [3,24-41].

CONCLUSION(S)

The ABC is often associated with fibro-osseous jaw lesions, which are referred to as aneurysmal bone cyst plus-lesions. The clinical, radiological, and histological data were all considered in this case of a secondary ABC-plus-lesion. Due to the diverse pathophysiology of ABC-plus lesions, diagnosing and identifying them, as well as determining the appropriate therapy, poses a challenge for surgeons. A definitive diagnosis can only be obtained through an incisional biopsy. Biopsies should be performed after ruling out vascular lesions.

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