A Large Mandibular Anterior Swelling—A Diagnostic Dilemma: A Case Report

Prasanna R. Sonar[1] Aarati S. Panchbhai[2] Suwarna B. Dangore[2]

[1] Sharad Pawar Dental College & Hospital, Datta Meghe Institute of Higher Education & Research (Deemed to be University), Sawangi-Meghe, Wardha, Maharashtra, India
[2] Department of Oral Medicine and Radiology, Sharad Pawar Dental College & Hospital, Datta Meghe Institute of Higher Education & Research (Deemed to be University), Sawangi-Meghe, Wardha, Maharashtra, India

Address for correspondence Prasanna R. Sonar, MDS, Sharad Pawar Dental College & Hospital, Datta Meghe Institute of Higher Education & Research (Deemed to be university), Sawangi-Meghe, Wardha 442001, Maharashtra, India (e-mail: jaysonar1234@gmail.com).

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Abstract

Hybrid lesions are lesions that combine characteristics of many pathologies. The literature has shown a rare relationship between aneurysmal bone cysts, fibro-osseous lesions, and a central giant cell lesion. In this case, we describe an unusual hybrid lesion in the mandibular anterior area. As a result, the goal of this article is to highlight the significance of comprehensive clinical, radiographic, and histological investigation in addition to presenting a rare instance of hybrid lesion. This will lessen the likelihood of misdiagnosis in these situations and aid in prompt treatment planning of the lesion.

Keywords

- hybrid lesions
- aneurysmal bone cysts
- ossifying fibroma
- central giant cell granuloma

Introduction

Hybrid lesions are incredibly uncommon compositions that have components of numerous pathologies, each of which has a specific tumor category.[1,2] One or more recognized odontogenic cysts and/or tumors that have mixed histological features and are present in the same primary location are referred to as hybrid odontogenic lesions.[3] Dental practitioners have a problem with this category of lesions since their histogenesis is debatable, their clinical behavior is still relatively unknown, and they exhibit a variety of clinical-radiographic symptoms.[3] Less cases have been reported recently of hybrid lesions that combine an ossifying fibroma (OF), an aneurysmal bone cyst (ABC), and a central giant cell lesion (CGCL).

According to the 2017 definition provided by the World Health Organization, CGCL is a benign osteolytic proliferation that can occasionally be locally aggressive and is made up of large, multinucleated osteoclast-type cells in a fibrous tissue stroma that also contains hemosiderin and hemorrhagic deposits.[4] The name "aneurysmal bone cyst" is misleading because this lesion is not a cyst and does not have an epithelial coating. It does have various-sized bloody spaces.[5] The ABC could start as a localized vascular disruption in bone.[6] ABC may be divided into three categories. The conventional or vascular kind (95%) appears as a damaging lesion that grows quickly, expands, and invades soft tissue. The solid kind (5%) may initially be seen as a radiolucency on a normal radiograph or as little swelling.[7,8] The characteristics of both the vascular and solid varieties can be seen in a third type or mixed variety. Because of reports of abrupt activation or fast expansion of stable lesions, it might represent a transient phase of the lesion.[8] OF, also known as cement-ossifying-fibroma, is a benign bone tumor that affects the facial skeleton. It is further divided into juvenile and conventional forms, each of which has trabecular and psammomatoid subtypes.[9]

CGCL associated with fibrous lesions may be the result of a secondary reaction in response to changes in the initial stroma of the lesion, where theoretically there is an activation of the osteoclasts and their subsequent transformation

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into multinucleated giant cells, all of this mediated by paracrine mechanisms. This is suggested as one potential pathogenesis for hybrid lesions involving giant cells.[10] There is a need to publish such instances in the literature because there are so few of them, making diagnosis and treatment more accurate.

Case Report

Having experienced painless swelling in her mouth, a 75-year-old woman visited a dental hospital. The swelling was present in the lower anterior region of the lower jaw for the last 18 months, which had grown gradually to its current size. The patient was hepatitis B surface antigen (HBsAg) positive with a history of hypertension that was present for 40 years and she was on medication for the same. The patient was not treated before for the same. No history of trauma was present. Habit history and family history were not significant.

Extraoral examination revealed a diffused swelling was present over the anterior region of the lower jaw extending anterior-posteriorly from the corner of the mouth on the left side to corner of the mouth on the right side, and superior-inferiorly from the vermillion border of the lip to 3 cm lower to the inferior border of the mandible. On inspection, the size was 5×8 cm approximately, the shape was roughly oval, the margins were well defined, the color was pinkish red, the surface was tense and edematous, and mentolabial fold was obliterated. On palpation, tenderness was absent, consistency was firm to hard and temperature was raised (►**Fig. 1**).

Intraoral examination revealed a diffuse swelling with labio-lingual expansion extending anterior-posteriorly from the 36 regions to 47 regions posteriorly causing labial vestibular obliteration and superior-inferiorly from the gingival margin into lingual and gingival labial sulcus. The overlying mucosa had lobulated swelling that was ulcerated. The size was 1.5×1 cm approximately, shape was roughly oval, borders were ill-defined, the surface was ulcerated and edematous, reddish pink in color, and the margins were well defined. On palpation, tenderness was absent, induration was absent, consistency was soft to firm, and fixity to the internal structure was absent. Grade II mobility was present with 31 32 41 42 (►**Fig. 1**).

The provisional diagnosis was given as aggressive central giant cell granuloma. Differential diagnosis was given as osteogenic sarcoma, ABC, and intermediate stage of fibro-osseous lesion.

Orthopantomogram and lateral skull radiograph revealed that osteolytic lesion extending from the symphysis region bilaterally involving the premolar and molar regions. Thinning of the inferior cortex bilateral up to molar region with irregular margins was seen with the remnant of internal septae seen in radiolucency (►**Fig. 2**). Computed tomographic scan, which was taken after 14 days after the first visit, revealed that there was a single large expansile, mixed lesion present in the body of the mandible with radiopaque-radiolucent foci, showing multiple radiolucent loculi with soap bubble appearance. The lesion showed corticated periphery (►**Fig. 3**).

Incisional biopsy revealed large multinucleated giant cell with 10 to 15 nuclei suggesting giant cell granuloma (►**Fig. 4**).

After the biopsy in 14 days, there was a rapid increase in intraoral swelling (►**Fig. 5**).

The patient underwent an elective surgical procedure under general anesthesia and nasotracheal intubation. As the patient was HBsAg positive, universal precaution was taken.

Affected teeth were extracted along with the removal of a portion of the mandible without disruption of the continuity of the mandible was planned. Segmental mandibulectomy from the angle of the mandible of the right side to the 38 regions of the left side of the mandible was done and reconstruction with pectoralis major myocutaneous flap was done (►**Fig. 6**).

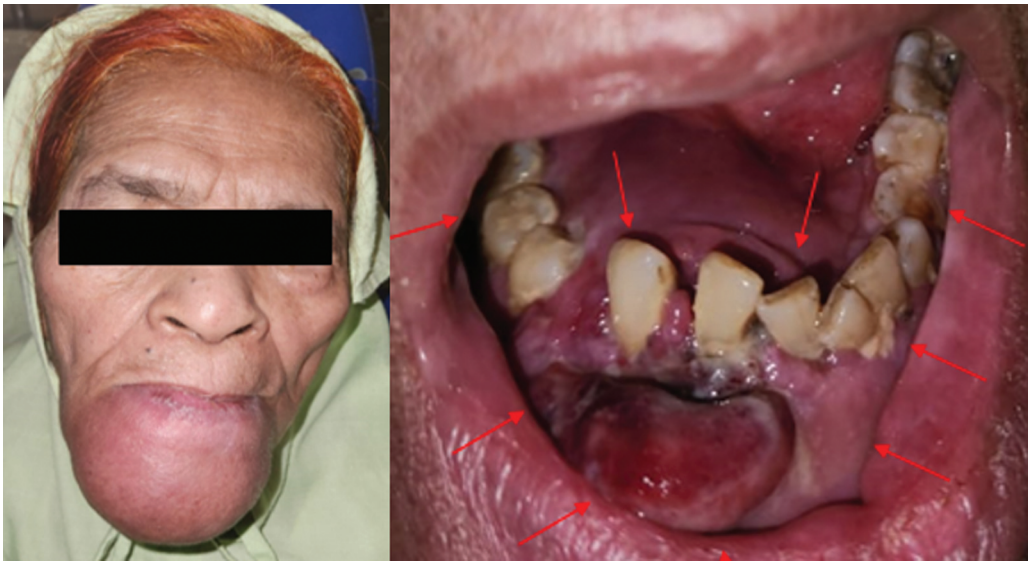

Fig. 1 Extraoral and intraoral photo on the first visit.

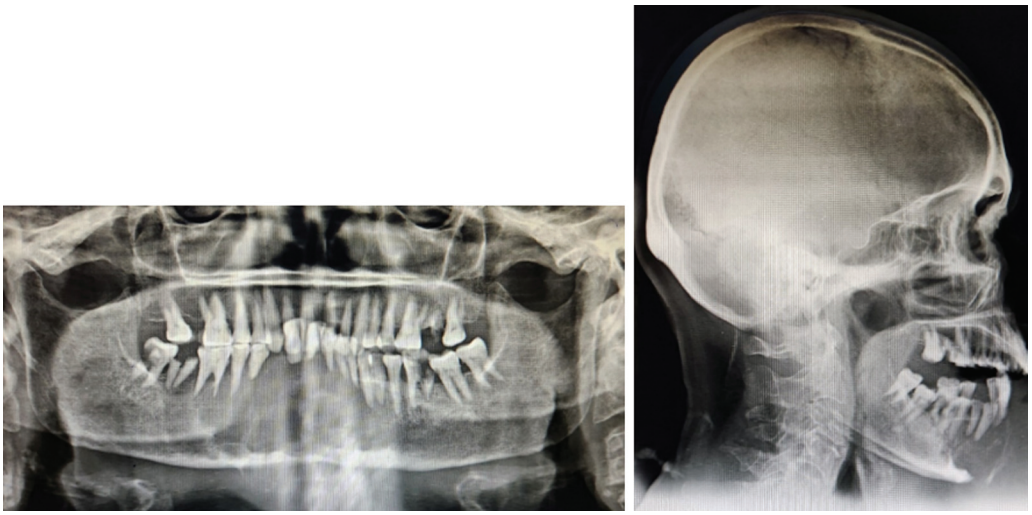

Fig. 2 Orthopantomogram and lateral skull radiograph.

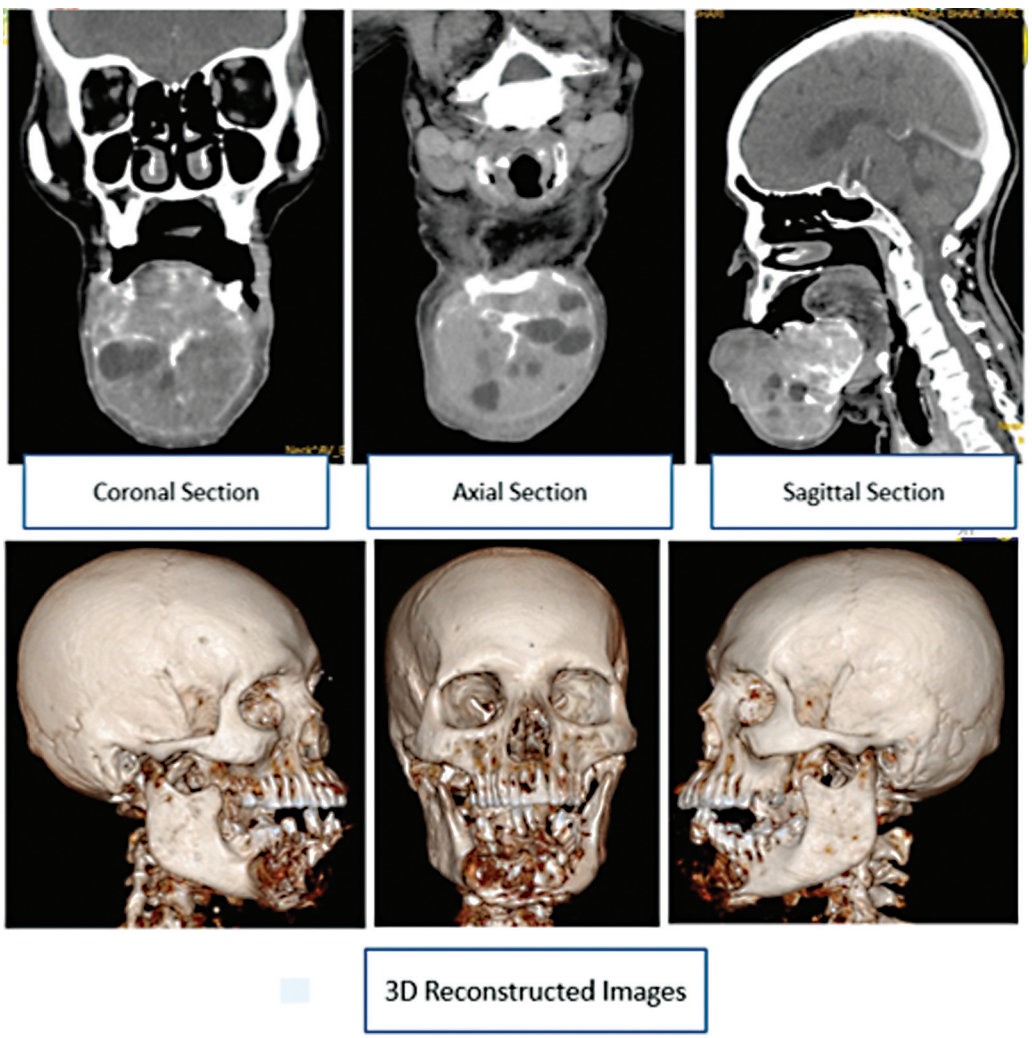

Fig. 3 Computed tomographic scan showing coronal, axial, and sagittal sections.

The resected specimen showed an exophytic grayish-black extensive lesion present over the lower anterior jaw (►**Fig. 7**). On the cut section, excessive bleeding was encountered resembling a blood-soaked sponge with large cavernous spaces.

When the resected specimen was sent for histopathology, hematoxylin and eosin stained lesional tissue section in scanner view showed fibrocellular with large cavernous or sinusoidal blood-filled spaces. In low power view, immature

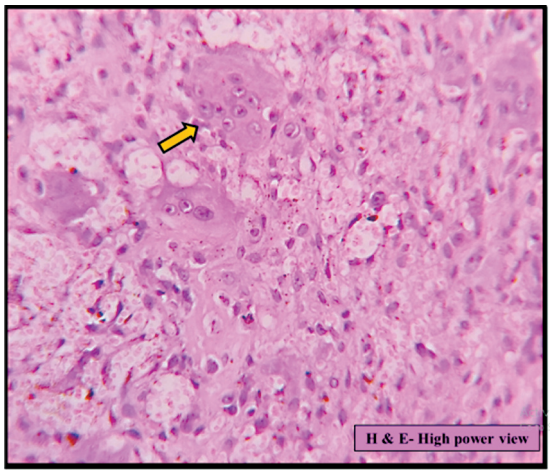

Fig. 4 Histopathology of incisional biopsy showing large multinucleated giant cell.

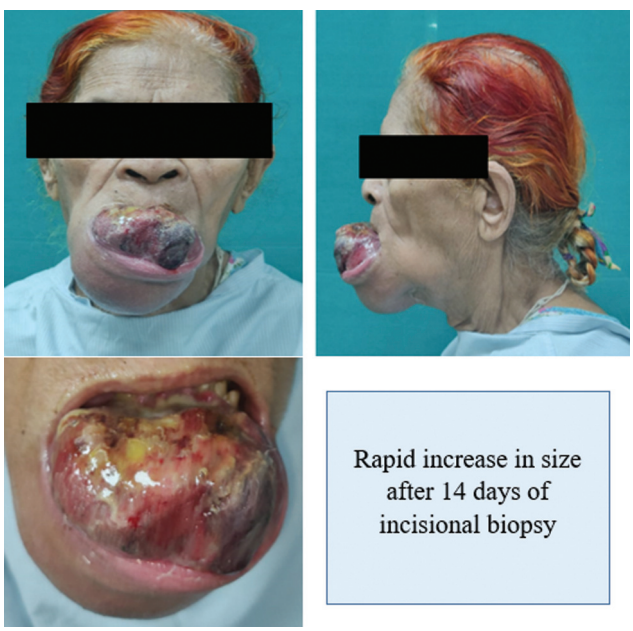

Fig. 5 Rapid increase in the size of the lesion after 14 days of biopsy (preoperative photos).

Fig. 7 Resected specimen.

bony trabeculae lined by osteoblastic rimming with numerous osteocytes are seen. Also, multinucleated giant cells and young fibroblasts are seen in the connective-tissue stroma seen in low power view. Under higher magnification, multinucleated giant cells with 10 to 15 nuclei and immature plump fibroblasts are seen in the connective tissue stroma. Histopathological diagnosis was given as an ABC with OF (►**Fig. 8**).

Discussion

The differential diagnosis for this lesion includes aggressive central giant cell granuloma, ABC, intermediate-stage of fibro-osseous lesion, and osteogenic sarcoma. It takes a combination of clinical, radiographic, and histological assessment to correctly diagnose these disorders.

A benign yet locally destroying lesion that mostly affects the jaws is aggressive central giant cell granuloma. Aggressive

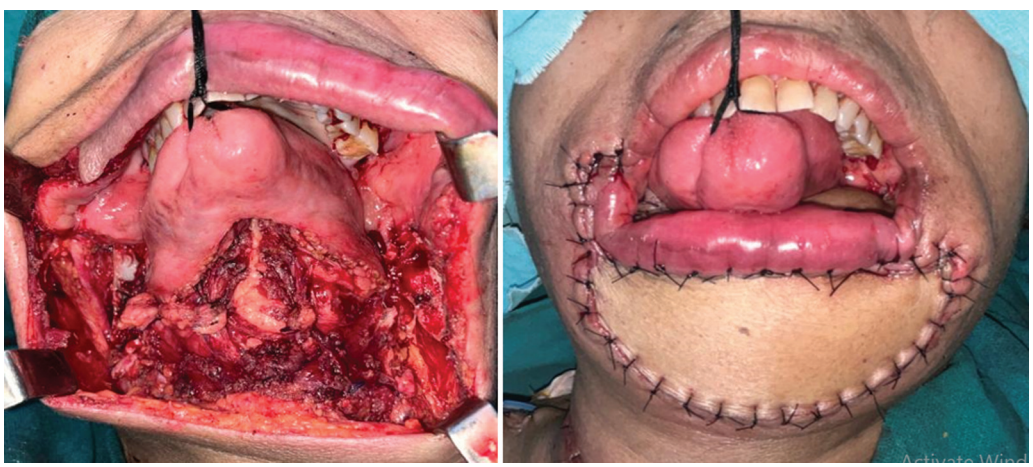

Fig. 6 Segmental mandibulectomy with reconstruction with pectoralis major myocutaneous flap.

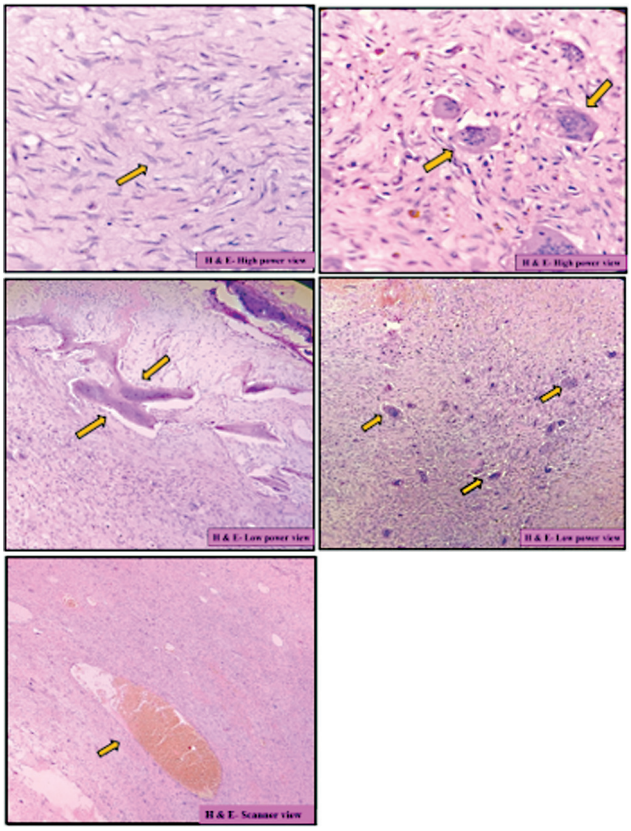

Fig. 8 Histopathology of the resected specimen.

growth, cortical bone damage, and a high recurrence rate are its defining features.[11] OFs and fibro-osseous lesions are two distinct medical conditions with unique histological characteristics. A series of benign bone illnesses known as fibro-osseous lesions are distinguished by the replacement of healthy bone with a fibroblastic stroma and various degrees of mineralization.[12,13]

The process of ossification results in the development of mineralized bone and fibrous tissue. Other bones, such as the long bones and craniofacial skeleton, can also develop OFs. They appear as well-circumscribed, expansile lesions on radiographs that are both radiolucent and radiopaque in appearance. OFs exhibit trabeculae of woven bone together with fibrous tissue histologically.[14,15]

The most frequent primary malignant bone tumor, osteogenic sarcoma, primarily affects children and young adults. The formation of osteoid, or young bone, distinguishes it as it develops from primitive bone-forming cells. The metaphysis of long bones, especially those at the knee, is a common site of involvement. Osteogenic sarcoma presents radiographically as a destructive lesion that is aggressive and mixed in lytic and sclerotic patterns. Histologically, it shows the production of osteoid and pleomorphic malignant cells.[14,15]

A rare hemorrhagic bone condition called ABC only sometimes affects the jaws. The lesion, which was caused by a localized vascular disruption, is widely accepted.[6] The "blow-out" effect or growth of the damaged bone that manifests in these sorts of lesions is referred to as "aneurysmatic."[16] The jaw's ABC lacks epithelial lining (pseudocyst).[11] It makes up 5% of all cranial and maxillofacial bone lesions and is most prevalent where the marrow and vascular contents of the bone are both reasonably high. This explains why ABC in the skull bones, which have less venous pressure, is uncommon.[17,18]

The concept of a hybrid lesion involving ABCs was first proposed by Kransdorf and Sweet in 1992. They described cases in which the histological features of ABCs were present along with additional features of other bone tumors, such as giant cell tumors, osteoblastoma, or fibrous dysplasia. This hybrid nature of the lesion makes it challenging to diagnose and treat accurately. The diagnosis of ABC hybrid lesions relies on a combination of clinical, radiological, and histopathological findings. Radiographically, these lesions may show a mixed pattern with characteristics of both ABC and the associated lesion. Magnetic resonance imaging can provide further information on the extent of the lesion and its relationship with adjacent structures.[19]

Thirty-two ABC plus lesions were found after a study of the literature; it was noted that there is a preponderance of men and that the mandible is more likely to be afflicted. The posterior area of the jaw is where more than 90% of ABC plus lesions are found. Sixty-eight percent of the 32 ABC plus lesions had fibro-osseous lesions in common. Giant cell lesions were linked to 32% of the lesions. From a lesion having no symptoms to a painful, expanding, and destructive pattern, ABC exhibits a wide spectrum of clinical characteristics.[20] The majority of the lesions, however, were asymptomatic swellings, which also existed in our case. Female patient having a lesion in the mandibular anterior region is rare that was present in our case.

In contrast to ABCs that develop in other long bones, ABC hybrid lesions usually exhibit discomfort along with a propensity for rapid development, whereas ABCs that develop in other regions of the body are frequently linked to malignant tumors including chondrosarcomas and osteosarcomas.[18] ABCs have never encountered malignant or metastatic bone lesions. Additionally, they are very infrequently linked to malignant ameloblastomas and aggressive neoplasms like osteoblastoma.[21]

Jaw ABC's radiological appearance is somewhat unpredictable. The lesion may seem bony, cystic with a honeycomb or soap bubble appearance, or eccentrically inflated. The cortex may be perforated or destroyed, and a periosteal response may appear.[21] A mixed, radiopaque, or radiolucent lesion might exist. We found an expansile, mixed lesion with radiopaque–radiolucent foci in the current instance. Additionally, the lower anterior teeth appeared to be mobile. Since other lesions with a similar appearance on radiographs, such as odontogenic cysts, ameloblastomas, central giant cell granulomas, myxomas, or central hemangiomas of the bone, exist, it is impossible to diagnose the lesion based solely on its radiographic appearance.[22]

According to histopathology, ABC is made up of many blood-filled sinusoidal gaps, osteoid material, and multinucleated giant cells in a fibrocellular connective tissue stroma. Variable quantities of hemosiderin pigments are also detected. This description is diagnostic of ABC in its classic

or vascular form.[16] In contrast, hemorrhagic foci with many fibroblastic and fibrohistiocytic components, gigantic cells that resemble osteoclasts, osteoblastic differentiation zones with osteoid, and calcifying fibromyxoid tissue are seen in the solid form. In the mixed form, one can see both vascular and solid characteristics. The ABC + lesions have a combination of vascular and solid form, as well as related lesions; they exhibit multiple blood-filled holes in the fibrocellular stroma, as well as multinucleated giant cells and the production of osteoid tissue.[23] The histological characteristics of the lesion in this instance matched those indicated above, suggesting an ABC with an OF.

The total eradication of the lesion is often the goal of the ABC plus lesion treatment strategy. The lesions might be divided by many bone septae and are often multilocular. This makes surgical removal challenging. The methods of treatment are systemic calcitonin therapy, curettage, block resection, diagnostic and therapeutic embolization, and block removal. Several self-healing cases have been documented in some studies after extensive follow-up. For the treatment of cases of aesthetic deformity, loss of continuity of the mandible, and situations with a high risk of fractures, several writers have advocated for prompt restoration of the defect using autogenous grafts.[20] In the present case, segmental mandibulectomy with reconstruction was done.

Conclusion

Only a very small number of cases of these associated hybrid lesions have been reported in the literature. Due to their rarity and vague initial symptoms, these patients confront doctors with diagnostic difficulty. Dentists have a critical role in the diagnosis and treatment of hybrid lesions, and each lesion must be correctly classified. Such cases should be reported in the literature so that a more accurate diagnosis and course for treatment may be implemented.

Conflict of Interest
None declared.

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