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Case Report

Aneurysmal Bone Cyst of the Mandible Associated with an Impacted Tooth; a Case Report

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Abstract

Introduction: Aneurysmal bone cyst is a non-neoplastic lesion affecting mainly the long bones and spine. Only 1.8% of aneurysmal bone cysts involve the head and neck area with mandible being more commonly affected. A wide range of pathologies are included in differential diagnoses of ABC.
Case Presentation: We report a case of mandibular aneurysmal bone cyst in a 15-year-old female that could be mistaken with a dentigerous cyst due to its association with an impacted tooth, although unlike dentigerous cyst it was accompanied by pain and swelling. Radiographically, an unilocular ill-defined radiolucency surrounding the third molar tooth germ was detected. performing fine needle aspiration, a low viscosity bloody fluid was aspirated. Incisional biopsy followed by microscopic evaluation revealed blood-filled spaces surrounded by hypercellular inflamed fibrotic connective tissue, suggesting an ABC. Due to the risk of association with true neoplastic lesions, the whole lesion was excised via extraoral approach and histopathologic evaluation confirmed the diagnosis of ABC. 7 years of follow-up showed no complications or signs of recurrence.
Conclusion: Association with unerupted or impacted tooth is not a common finding for aneurysmal bone cysts. It means that it can mimic radiographical features of dentigerous or other odontogenic cysts. Although aspiration may be helpful, but incisional biopsy and microscopic evaluation are mandatory for precise diagnosis.
Key Words: Aneurysmal bone cyst, Mandible, Unerupted tooth, Dentigerous cyst, Curettage

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Introduction

Aneurysmal bone cyst (ABC) is a descriptive term in which the word “aneurysm” refers to a marked expansion and the word “cyst” refers to a fluid-filled cavity. However, traditionally the aneurysmal bone cyst has been considered a reactive lesion and now cytogenetic evidence suggests that primary ABC (i.e., arising de novo) may be neoplastic in nature (1).

ABC accounts for 1% of all bone tumors with a reported incidence of 0.14 per 100,000 individuals and a prevalence of 0.32 cases per 100,000 individuals (2).

It is defined as an expansive osteolytic lesion consisting of various size blood-filled spaces of channels separated by connective tissue septas containing multinucleated giant cells and osteoid tissue by the world health organization (3).

Only about 1.8% of ABCs occur in the jaw bones with mandible being more commonly affected than maxilla and the molar and ramus region are more often involved than the anterior regions (4).

In most cases, ABC of jaws represents itself as a rapid-growing, painful or painless swelling with or without bony expansion but it can also be seen as an

asymptomatic, slow-growing lesion (5).

ABCs can mimic a wide range of pathologies including ameloblastoma, ossifying fibroma, fibrous dysplasia, giant-cell granulomas and odontogenic cycts (e.g. dentigerous cyst) and cause a diagnostic challenge requiring a comprehensive evaluation. Yet, according to authors' knowledge only few cases of ABC association with an impacted tooth are reported. We report a rare case of aneurysmal bone cyst associated with an impacted third molar tooth, mimicking a dentigerous cyst, in a 15-year-old girl, indicating the importance of step-by-step diagnosis and Correlation of clinical, histopathological and radiographic findings in jaw lesions.

Case Presentation

Patient information

A 15-year-old girl was referred to oral and maxillofacial surgery department of Tehran University of Medical Sciences (TUMS) with a chief complaint of painful swelling in her left lower face for the past 3 months. The patient had no medical history and reported no history of trauma to her jaw prior to developing this lesion.

Clinical findings

In the extraoral examination there was a mandibular swelling extending from left lip commissure to the periauricular region anteroposteriorly and extending superiorly from the inferior border of mandible along the ramus. The swelling was painful but with firm slightly tender consistency (Figure 1). There was no evidence of lymphadenopathy, trismus, or sensory changes.

Figure 1. preoperative photography showing the swelling of lower left face

The intraoral examination showed a large swelling extending from the buccal aspect of second molar tooth to middle area of ramus in retromolar area with the overlying mucosa being normal without any signs of bleeding, ulcer or pus discharge.

Diagnostic assessment

Panoramic radiograph showed a unilocular radiolucent lesion with ill-defined borders extending anteroposteriorly from mesial of unerupted third molar tooth to the left mandibular angle and superoinferiorly from inferior border to sigmoid notch of mandible surrounding the unerupted tooth. There was no evidence of cortical perforation (Figure 2).

Figure 2. Panoramic radiograph showing a unilocular radiolucency in left posterior area of mandible.

Based on radiographic and clinical findings central giant cell granuloma, ameloblastic fibroma, ossifying fibroma and dentigerous cyst were considered as differential diagnosis. Fine needle aspiration was performed and low viscosity bloody fluid was aspirated which was in favor of high vascular nature of the lesion.

Incisional biopsy was done for the lesion. The surgical impression during the incisional biopsy was the solid-cystic, spongy tissue with moderate amount of bleeding in favor of bloody nature of the lesion.

Microscopic evaluation of the sample revealed numerous various-sized blood-filled spaces surrounded by hypercellular inflamed fibrotic c onnective tissue (Figure 3). Scattered multinucleated giant cells and reactive bone formation were seen. These features were compatible with aneurysmal bone cyst. However, since ABC may be associated with some true neoplastic lesions such as fibro-osseous lesions or central giant cell granuloma,

we suggested for the surgeon to excise the whole lesion to reach a definitive diagnosis.

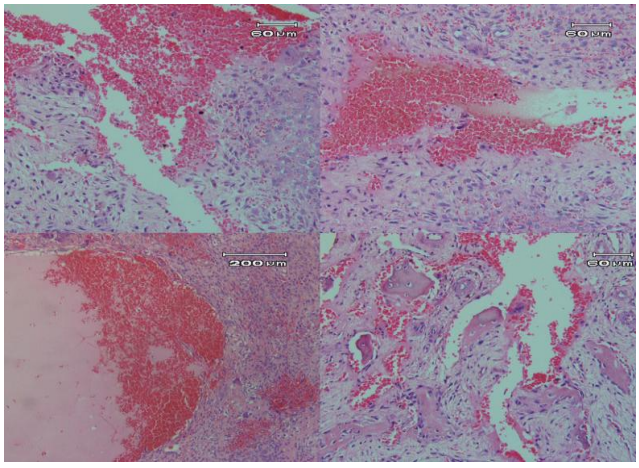

Figure3. histopathologic view showing various-sized blood-filled spaces surrounded by hypercellular inflamed fibrotic connective tissue

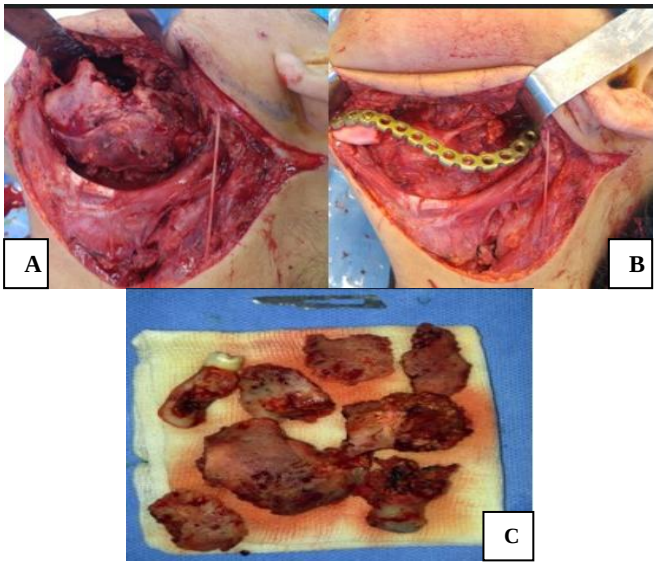

Figure 4. (A) surgical treatment via extroral approach. **(B)** reconstruction of mandible using a titanium plate. **(C)** excised lesion and the third molar germ.

Therapeutic intervention

The surgical treatment was performed via extraoral approach under general anesthesia. The lesion and the tooth germ were excised, followed by a thorough curettage of bone. The mandible was then reconstructed using a titanium plate to provide the continuity of the lower jaw (Figure 4). The histopathologic examination of the whole excised lesion confirmed the diagnosis of ABC. In addition, no tumoral proliferation or cystic lining was seen in sub serial sectioning of the whole excited lesion.

Follow-up

The patient was followed for 7 years and there was no sign of recurrence or complication.

Consent form

Written informed consent was obtained from the patient for publication of this case report and enclosed images. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity. A copy of the written consent is available.

Discussion

ABCs are benign osteolytic lesions that commonly appear in the first 2 decades of life predominantly involving the mandible if in craniofacial region (6).

Etiology and classification

The etiology of ABCs is not completely known; Traditionally they are considered reactive lesions. A traumatic event, vascular malformation or neoplasm may disrupt normal osseous hemodynamics, resulting in an area of hemorrhage and osteolysis. In contrast, cytogenic evidence suggests that primary ABCs may be neoplastic in nature (1). ABCs can also be categorized according to their clinical and radiological behavior as 1st stage – inactive, with well-defined periosteal and sclerotic borders, 2nd stage – active, with well-defined radiographic appearance and interrupted periosteal borders, 3rd stage – aggressive, with rapid growth and diffuse ill-defined radiographic appearance (6, 7). Based on clinical and radiographic findings, it seems that our case was in 3rd stage.

ABCs show painful swelling in most reported cases. Although Triantafillidou reported a case of ABC in a 7-year-old child which showed painless swelling (5). The most common type of ABC is “classic or vascular form” that is composed of blood filled cavities, sinusoidal spaces with hemosiderin-laden macrophages, and multinucleated giant cell separated by cellular fibrous connective tissue with more aggressive and rapid growth. The other less common form is the “solid” type (5%), which is a

noncystic variant of ABC, composed mainly of soft tissue with slow growth and discrete clinical appearance. Hemorrhage and fibroblastic spindle cells proliferation is often seen. Areas of osteoblastic differentiation with osteoid calcification can be seen. A third form also known as "mixed type" contains solid and cystic areas (6, 7). According to surgical impression, our case showed vascular form of the lesion.

Differential diagnosis

Wide range of pathologies with osteolytic radiographic appearance is included in the differential diagnosis of ABC, i.e. odontogenic cysts, non-odontogenic jawbone cysts and tumoral and nontumoral lesions like ameloblastoma, ossifying fibroma, fibrous dysplasia, giant-cell granulomas, osteosarcoma, etc.

In our case, the cystic formation could be mistaken with an odontogenic cyst (dentigerous cysts), due to its association with the mandibular left third molar germ. Although, there were significant swelling and pain which are not common findings in dentigerous cysts. Mihaylova reported a case of ABC in a 14-year-old male which mimicked a dentigerous cyst similar to our case (7). Table 1 shows previously reported unusual ABCs.

Histopathology

Microscopically, ABC may be mistaken with some other pathologic lesions composed of some nonneoplastic lesions such as central giant cell granuloma and also some neoplastic lesions such as osteosarcoma, giant cell rich variant. Central giant cell granuloma shows abundant number of giant cells proliferation in background of plump mesenchymal cells and lack of any sinusoidal spaces. Giant cell rich variant of osteosarcoma shows more atypia in spindle cells and osteoid formation beside the malignant cells. Presence of lace-like osteoid formation can also be helpful in diagnosis of osteosarcoma.

Treatment approaches

The treatment plan must be in accordance with the location of the ABC, clinical presentation and radiographic findings. Standard surgical management is curettage with or without bone grafting. Available alternative therapies include (a) selective arterial embolization as a preoperative adjuvant modality to minimize intraoperative blood loss; (b) polymethylmethacrylate injection in benign bone lesions in children, although long-term effects may be deleterious (no growth potential, stress shielding increasing the risk of future pathologic fractures, possible nidus for infection); (c) percutaneous alcoholic solution of zein injection, with potential complications including local transitory inflammatory reaction, aseptic bone necrosis, and pulmonary embolism; (d) radiation therapy, which is associated with excellent control of ABCs but with chronic effects such as impaired function or secondary malignancies; (e) intralesional injections of steroids or doxycycline; and (f) bisphosphonate or denosumab treatments (8).

Recurrence rates range from 20 to 30% in different groups and it usually occurs within the first year after surgery probably due to insufficient excision or curettage of the lesion, especially in soft tissue invasive cases. Studies recommend immediate reconstruction of the defect with autogenous grafts in cases of esthetic deformity as well as those with a high risk of fractures and loss of mandibular continuity (9). A previous study by Motamedi et al stated that initial resection is not necessary and did not observe any recurrences following surgical curettage of mandibular lesions(10). In this case, a thorough curettage was performed after the removal of lesion and the involved tooth germ. We also did the reconstruction of the area immediately after excision of the lesion. 7-year follow-up showed no recurrence and no functional complications in the early and late postoperative period.

Conclusion

ABC in head and neck area is not a common finding and ABC associated with an impacted tooth is even less common and can cause a diagnostic challenge. It emphasizes the importance of comprehensive evaluation, including radiographic imaging and histopathological examination. The radiologic and clinical presentation of ABCs is extremely variable and a great emphasis is placed on histopathologic examination for the diagnosis. Due to the hemorrhagic nature of the lesion, adequate surgical removal by an oral and maxillofacial surgeon is recommended to achieve a good long-term prognosis.

Table 1. Reported cases of unusual ABC. Abbreviation: F: female, M: male, Mu: multilocular, U: unilocular, y: year(s), L: left, R: right, Man: mandible, Max: maxilla, NR: not reported

	Author	Multi/ Unilocular	Year	Age (y)	Gender (M/F)	Location	Symptoms	Effect on teeth	Why is reported?	Work up	Follow-up
1	Khurshida Banu	Mu	2012	19	M	L body of Man	Painful swelling	Associated with an impacted molar	Giant ABC	Resection	1 y
2	Katherine Triantafillidou	NR	2012	7	F	R Max & maxillary sinus	Painless swelling	Displacement and impaction of teeth	ABC in association with an ossifying fibroma	1.curettage 2.partial maxillectomy (after recurrence)	17 y (2 recurrences in 4 years)
3	Roshan Kumar Verma	Mu	2013	8	M	R Max	Swelling and dull pain	Ectopic molar	ABC of maxilla with an ectopic molar	Endoscopic excision	3 months
4	Sandhya Tamgadge	Mu	2014	7	M	L Man body & ramus & condyle & coronoid	Swelling	Displacement of D, E, 36 & 37	Juvenile psammomatoid ossifying fibroma with an ABC	Enucleation abd curettage/ chemical cauterization	2 months
5	Aadithya B.urs	Mu	2014	13	F	L posterior Man	Painful swelling	Impacted third molar	NR	NR	1.5-2 y
6	Ana Belen Marin Fernandez	Mu	2016	10	F	L mandibular ramus & condyle	Swelling and TMJ dysfunction	NR	ABC in mandibular condyle	Embolization/ curettage	7 y
7	Zornista Mihaylova	U	2019	14	M	L mandibular angle	NR	Impacted third molar	Mimicking a follicular cyst	Curettage	Long term(?)
8	Our case	U	2024	15	F	L Man	Swelling	Impacted third molar	Association with an impacted molar	Enucleation and curettage	7 y

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