CASE REPORT

A Case of "Camouflage": Aneurysmal Bone Cyst Simulating a Radicular Cyst

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ABSTRACT: Aneurysmal Bone Cyst (ABC) is an uncommon non-neoplastic pseudocyst. It usually affects the long bones, spine and occasionally involves the jaws. Radiographically, ABC may appear as an unilocular or multilocular radiolucency with expansion and thinning of the surrounding cortical bone. However, due to the lack of a pathognomonic feature of its own, the diagnosis still remains ambiguous. The following is a report of ABC located at the right body of the mandible in an Indian female patient. The lesion initially camouflaged as a simple radicular cyst. Later, ABC was confirmed only with histo-pathological investigation.

Key words: *bone cysts, aneurysmal/diagnosis, mandible/pathology, female, pseudocyst*

The Aneurysmal Bone Cyst (ABC) has been described as a pseudocyst due to the absence of an epithelial lining. ABC was first recognized by Jaffe and Lichtenstein in 1942 and they described it as an intraosseous, osteolytic lesion most commonly affecting the metaphyseal region of the long bones. Later on, Bernier and Bhaskar in 1958, described the first case of ABC in the jaws. Only 2% of ABC lesions are found in the head and neck region, two thirds of which are located in the body-ramus region of the mandible[1]. Despite a descriptive history of more than 60 years, it's untrue nature and character makes the optimal treatment of ABC still an obscure and debatable one.

CASE REPORT

A 14 year old girl presented with a mild, intermittent, painful tooth in the right posterior region of the lower jaw for more than a year's duration. She had visited a dentist for the complaint 6 months back, and the pain subsided on taking a course of antibiotics and analgesics. In the last one week, she noticed a painful swelling in the right submandibular region causing a facial asymmetry . It was diffuse, smooth surfaced, with no surface discoloration or secondary changes. It was not associated with paraesthesia or anaesthesia (Fig-1).

Generally, she was in good health with no remarkable medical history. On intra-oral examination, inferior buccal cortical expansion extending from 45 to 47 region was evident. Overlying mucosa was erythematous with no surface discoloration or sinus opening. On palpation, it was firm in consistency. No bruit was heard on auscultation. Hard tissue examination revealed a non-tender grossly decayed 46, retained grossly decayed 84 and retained 83 (Fig-2).

Considering the history and the clinical findings, a provisional diagnosis of Infected Radicular Cyst was made. The radiographic examination included intra-oral periapical radiographs (IOPA), occlusal and panoramic radiograph. The radiograph revealed a homogenous, osteolytic, unilocular radiolucency with no internal structure in the right body of the mandible in relation to 46. Corticated border was evident in the anterior aspect of the lesion. There was also a radiolucency in relation to coronal portion of 46 that extended into the dentin (Fig-3). Right lateral mandibular occlusal projection revealed an uninterrupted smooth bicortical expansion (Fig-4).

On the contrary, panoramic radiograph revealed a bilocular radiolucency in the right body of the mandible, with one faint internal septa margined by a corticated border. It had caused a spiked type of tooth resorption in 46 and mesial displacement of 45, 46, 47. The lesion appeared to push the inferior alveolar canal downwards (Fig-5). There was also a radiolucency in the coronal portion of 46, extending into dentin, involving the pulpal horns.

Received :17.09.12 Accepted : 31.12.12

Aspiration of the involved region yielded, a dark coloured, blood like fluid (Fig-6). Considering the above investigations, odontogenic myxoma, aneurysmal bone cyst, ameloblastoma and keratocystic odontogenic tumour were considered for differential diagnosis.

As part of the preliminary treatment, extraction of carious tooth and incisional biopsy was done under local anesthesia. The incised specimen was thereafter sent for histo-pathological examination which revealed a Solid type of Aneurysmal Bone Cyst (Fig-7). Following this, enucleation was carried out under general anesthesia (Fig-8). The patient was followed up for 6 months and reported with no recurrence.

DISCUSSION

World Health Organization defines ABC as a 'benzign, tumour like lesion with an expanding osteolytic lesion consisting of blood filled spaces of variable size separated by connective tissue septa containing trabeculae or osteoid tissue and osteoclast giant cells.[1] The term "aneurysmatic" refers to the blow-out effect or expansion of the affected bone[2].

The pathogenesis of ABC is controversial, and a number of theories have been advanced. Although trauma has been postulated, there is little evidence to support this. Lichtenstein in 1950 postulated that, ABC is a manifestation of altered hemodynamics[3]. According to Mankin JH et al, presence of circulatory disturbance leads to congestion, and pressure secondary to vascular dilatation and engorgement, resulting in bone resorption, with deposition of fibrous connective tissue, osteoid and

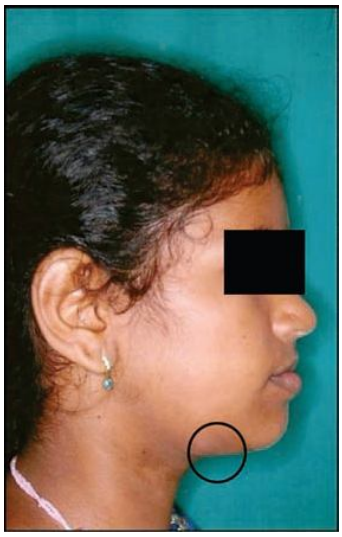

Fig 1: Clinical extra-oral photograph showing a swelling in the right submandibular region .

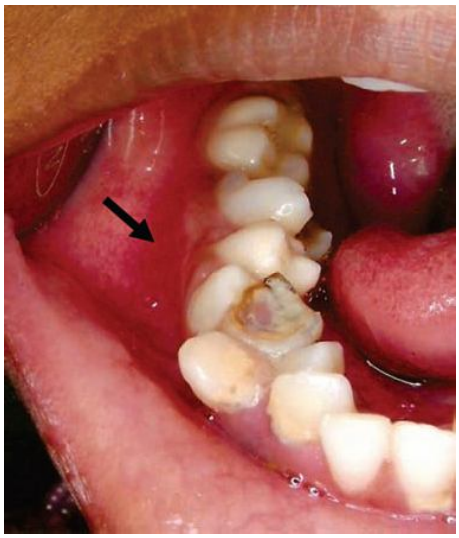

Fig 2: Clinical intra-oral photograph showing an erythematous area of the vestibule pertaining to 45, 46, and 47 and a grossly decayed tooth in 46, grossly decayed retained 84 and malposed retained 83

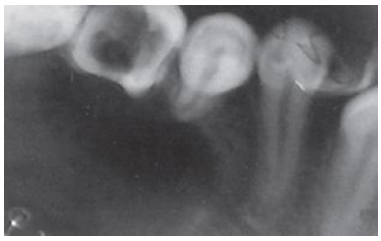

Fig 3: IOPA showing an osteolytic lesion of the right posterior mandible.

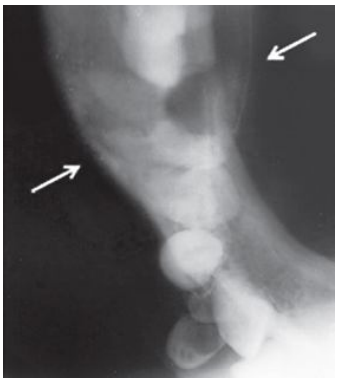

Fig 4: Right lateral mandibular occlusal projection showing bi-cortical expansion.

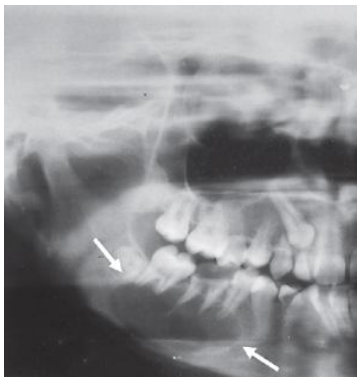

Fig 5: The cropped panoramic radiograph showing bilocular radiolucency in the right body of the mandible.

new bone[4].

Later, Bernier and Bhaskar described that, ABC resembles the central giant cell reparative granuloma of the jaw [5] and the only difference being, the presence of blood containing spaces in the former[3].

A third hypothesis was given by Beisecker et al , who proposed that, a primary bone lesion initiates an osseous, arteriovenous malformation and thereby creates through its hemodynamic forces, a secondary reactive lesion of the bone, leading to the production of an abnormal vascular component[3] .

The lesion usually occurs in females below 20 years of age having a predilection to mandibular body-ramus area[3] . This was consistent with the present report. She also had a bony hard swelling associated with pain and expansion, which are the other features of the entity.

Radiographically, ABC may appear as an unilocular or multilocular radiolucency. If multilocular, it may either have a honey comb or soap bubble appearance[3]. However, the radiological features for ABC are non-specific. In this case, a bilocular feature with one faint internal septa was observed.

The natural history of aneurysmal bone cyst has been described as evolving through four radiologic stages:

1. In the initial phase, the lesion is characterised by a well-defined area of osteolysis with discrete elevation of the periosteum.

2. This is followed by a growth phase, in which the lesion grows rapidly with progressive destruction of bone and development of the characteristic blown-out radiologic appearance.

3. Then, the period of stabilization sets in, where the characteristic soap bubble appearance develops as a result of maturation of the bony shell.

4. Final healing results in calcification and ossification with the lesion transformed into a dense bony mass .[6]

The Solid type presents as a solid mass without a cystic compound of irregular density. It contains multiple hemorrhagic foci and is a fibroblastic, osteoblastic and osteoclastic compound[7].

Considering the clinical features, the lesion was provisionally diagnosed as infected radicular cyst. However, keratocystic odontogenic tumour, odontogenic

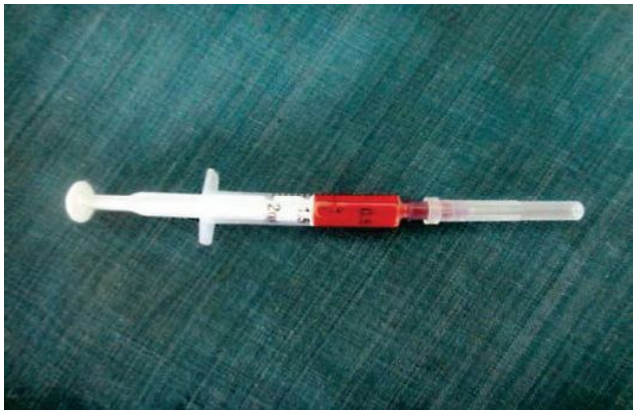

Fig 6: Aspirate from the involved region yielded blood like fluid.

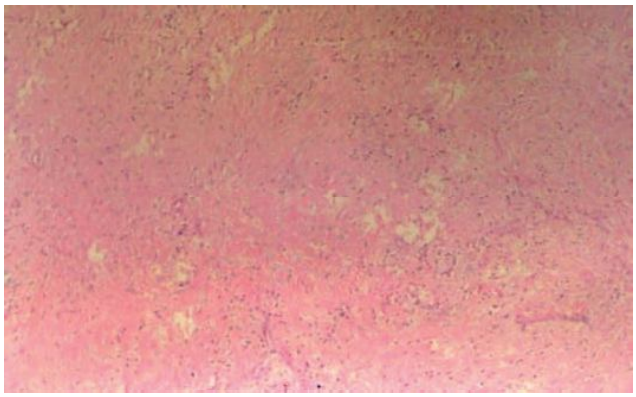

Fig 7: The photomicrograph (x100) showing fibrous connective tissue stroma with abundant mononuclear spindle cells and extensive extravasation of RBC's in the intervening stroma.

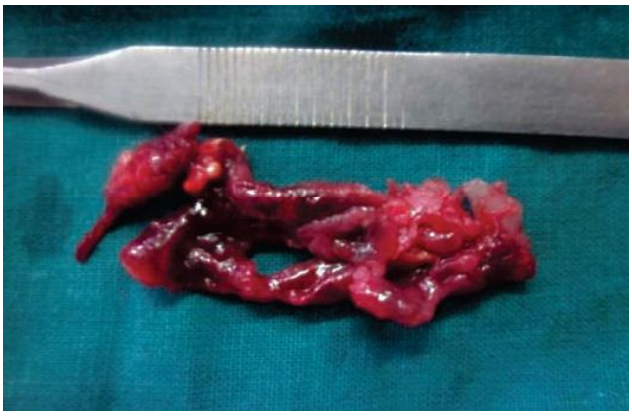

Fig 8: Showing the enucleated specimen from the right mandibular posterior region

myxoma, aneurysmal bone cyst and ameloblastoma were considered in the differential diagnosis.

Ameloblastoma was excluded from consideration as it commonly presents in the middle aged people. Odontogenic myxoma typically has a tennis racket radiographic appearance, which was absent in our case. The occurrence of keratocystic odontogenic tumour in a young patient with absence of unerupted tooth is rare.

In this case, presence of a pulpally involved tooth led to a provisional diagnosis of radicular cyst. However, the findings of the orthopantamogram and the content of aspirate indicated ABC. Histopathological examination confirmed solid type of ABC.

The range of investigations for ABC includes, the basic intra-oral and extra-oral conventional radiographs, along with CT, MRI, angiogram and biopsy. Management of ABC is not definite. Although, curettage is the most commonly adapted treatment, cryotherapy has also been suggested as an alternative treatment modality. In case of extensive lesions, segmental resection is indicated. Radiation is not recommended as sarcomatous change has been reported in these lesions after irradiation [8].

CONCLUSION

Though ABC has varied patterns of presentation, its clinical diagnosis is still possible. With cautious aspiration, biopsy and histo-pathological analysis, the camouflage can certainly be unveiled.

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How to cite this article:

Aneetha Raman G, Ravi David Austin, Kumar C Srivastava, Maria Rajathi. A case of "camouflage": Aneurysmal Bone cyst simulating a radicular cyst. Journal of Scientific Dentistry 2012;2(2):47-50.

Source of Support: Nil, **Conflict of Interest:** None declared