A Case Report of Aneurysmal Bone Cyst of the Mandible

Asha Raghunandan Iyengar[1], Seema Patil[2], Mansimranjit Kaur Uppal[3,*], Alisha Jason[4], Ganga Guddannanavar Karibasappa[5]

[1]Professor & Head, [2]Reader, [3,4,5]Post Graduate Student
Department of Oral Medicine and Radiology,
D.A Pandu Memorial R.V Dental College & Hospital, CA-37, 24th Main
1 Phase, J.P Nagar, Bangalore – 560078

***Corresponding Author**
E-mail: uppalsimran64@gmail.com

Abstract

Aneurysmal bone cyst (ABC) is a rare benign lesion of bone, which represents less than 1% of all the bone cysts biopsied. It is generally noted in long bones and spine and is less common in the craniofacial skeleton including maxilla and mandible. These rare jaw lesions are encountered mostly in the body and ramus of the mandible in the second or third decades of life. This case report describes a rare case of ABC in a 48 year old male patient involving the left body and ramus of the mandible. Clinically, it presented as an extra-oral swelling of 3 months duration with occasional mild pain and paraesthesia of the lip which are rare features of the lesion. Both conventional and CT imaging were suggestive of a multilocular (soap-bubble) lesion. An incisional biopsy was done and the histopathological report was suggestive of an Aneurysmal Bone Cyst. The patient was planned for hemimandibulectomy with free fibula reconstruction graft.

Key Words- Aneurysmal bone cyst, Mandible, Multilocular, KOT, Soap bubble.

Access this article online	
Quick Response Code:	**Website:** www.innovativepublication.com
	DOI: 10.5958/2395-6194.2016.00008.4

Introduction

Aneurysmal bone cyst (ABC) is a rare pseudocyst occurring in the long bones and vertebrae.[1] ABC is extremely rare in the maxilla and mandible constituting 0.5 % of all the jaw cysts.[2] The term ABC is a misnomer, as the lesion is neither an aneurysm nor a cyst. It is composed of numerous blood filled channels. ABCs generally occur in the first two decades of life and are infrequent beyond 20 years of age.[3] ABCs commonly present as asymptomatic bony swellings with a fairly rapid growth rate. One such rare case of ABC is presented here with unusual features such as occurrence in a relatively older subject and paraesthesia of the lower lip.

Case Report

A 48 year old male patient visited the department of Oral Medicine and Radiology with a chief complaint of swelling in the lower jaw on the left side since 3 months. The swelling was of a gradual onset; it was initially small and progressed to the present size. The swelling was associated with occasional mild pain and paraesthesia of left side of the lower lip and chin. The patient had not noticed any secondary changes such as ulceration or discharge. Patient gave a history of extraction of 38 twenty years ago which was uneventful. Patient did not give any previous history of trauma. The patient had consulted a private dental practitioner approximately 1 month prior for the same complaint wherein an antibiotic-analgesic course was prescribed and root canal therapy of 36 was initiated. There was no reduction in the size of swelling; hence he visited another dental practitioner wherein a CT scan of the mandible was advised.

The CT scan report was suggestive of Ameloblastoma / Keratocystic Odontogenic Tumor (KOT)/Osteomyelitis. An incisional biopsy intra-orally had been performed and the histopathological report suggested non-specific inflammation which was inconclusive. Hence, the patient visited this institution for further evaluation and treatment.

On extra-oral examination, mild facial asymmetry was seen due to a diffuse swelling in the left lower third of the face. Superoinferiorly, the swelling extended from a line joining the left corner of the mouth and tragus of the ear upto the inferior border of mandible. Anteroposteriorly, it extended 4cms posteriorly from the left corner of the mouth. Skin over the swelling appeared normal and there was no evident ulceration, suppuration /discharge. No visible pulsations were noted. On palpation, there was no local rise in temperature, the swelling was non-tender, smooth surfaced, firm to bony hard in consistency, non-compressible and non-reducible. Pulsations were not palpable.

On hard tissue examination, in 36, an endodontic access cavity preparation was evident and the tooth was non-tender on vertical and horizontal percussion. There was no abnormality in 37 and 38 was missing.

On soft tissue examination, a swelling was seen on the buccal aspect of mandibular teeth extending from 33 to 36. The swelling involved the marginal and attached gingiva, alveolar mucosa and vestibule. The mucosa over the swelling was pale pink and there was no evidence of ulceration, bleeding or suppuration. Sutures were seen with respect to 34 and 35 regions in the buccal vestibule (fig.1). On palpation, the swelling was non-tender except in the region of sutures and it was firm to bony hard in consistency.

A provisional diagnosis of radicular cyst with respect to 36 was made. The differential diagnoses included Keratocystic Odontogenic tumor (KOT) and Ameloblastoma.

The investigations advised were Intra-oral periapical radiograph, mandibular cross-sectional occlusal radiograph, and panoramic radiograph. The Computed Tomography including 3D reconstructions was also reviewed.

Panoramic radiograph (fig.2) showed a well-defined multilocular radiolucency with a scalloped outline seen in the left body and ramus of the mandible. The radiolucency extended anteroposteriorly from 31 to approximately 2cms distal to 37 into the ramus. Superoinferiorly, it extended from the middle thirds of the roots of 31 to 37 up to the inferior border of mandible. In ramus, the lesion was seen extending from the sigmoid notch upto the inferior border sparing the angle of the mandible. A few faint wispy septae were seen in the ramus region. Blunted resorption of apices of 35 and distal root of 36 was seen. Uneven thinning of inferior cortical border of mandible was seen.

On CT scan (fig.3), a marginally expansile osteolytic lesion involving the mandible on left side with thinned out cortex at the superior aspect, also showing cortical breach posteriorly at the retromolar trigone, 3rd molar region was seen. There was evidence of central air locule within the lesion showing possible air fluid levels. The lesion apparently involved the alveolar process of mandible extending to ramus superiorly and body inferiorly.

The following radiographic differential diagnoses were considered; KOT, Odontogenic myxoma, ABC, Traumatic bone cyst, Central giant cell granuloma (CGCG) and Ameloblastoma.

As the previous biopsy report was inconclusive, another incisional biopsy was performed and the histopathological report (fig.4) revealed fibrovascular connective tissue and sinusoidal spaces. In some areas, the connective tissue comprised of mature fibrovascular connective tissue. In other areas, it was annular and contained multinuclear giant cells with few mitotic figures. Formation of osteoid and immature bone was observed. The histopathological report was suggestive of an Aneurysmal Bone Cyst. Hence a final diagnosis of ABC was made.

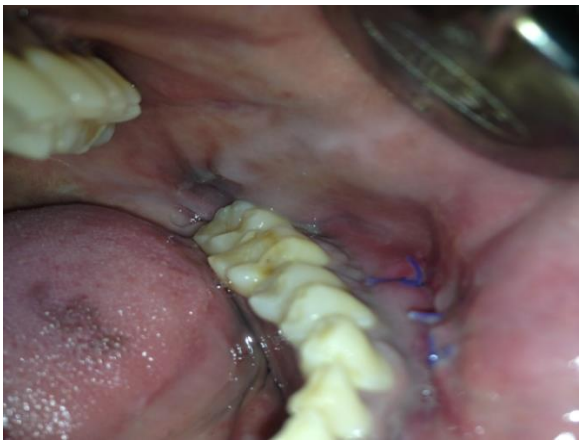


Fig. 1: Intra-oral photograph of the patient showing sutures on the buccal vestibule w.r.t 34 and 35

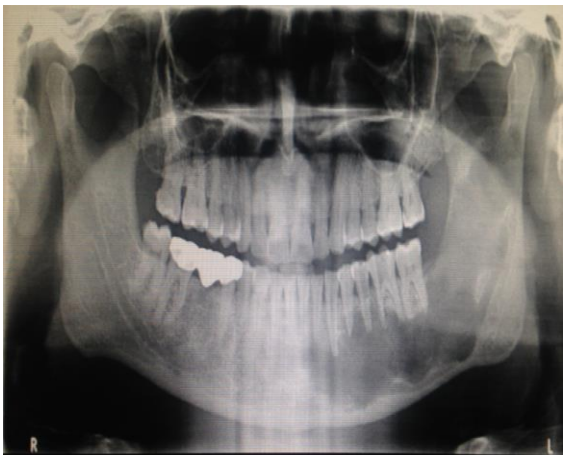


Fig. 2: Panoramic radiograph showing extensive multilocular radiolucent lesion involving the periapical region of 31 upto ramus and sigmoid notch region on the left side

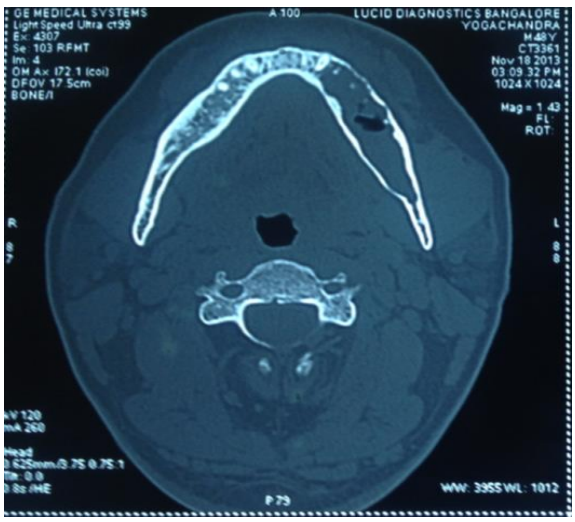


Fig. 3: (a) Axial sections of CT showing osteolytic lesion extending from anterior mandible to the ramus

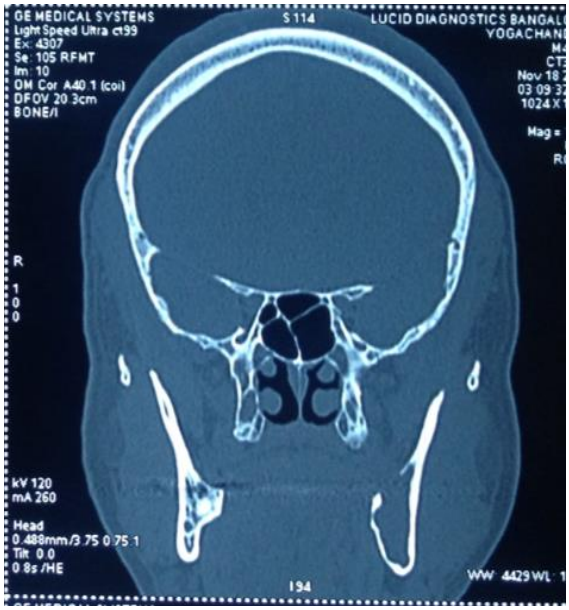


Fig. 3: (b) Coronal section of the CT scan showing an expansile lesion with a breach in the cortical plate in the retromolar trigone region

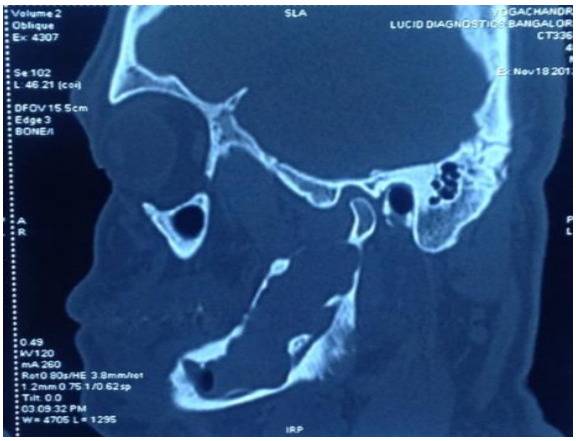
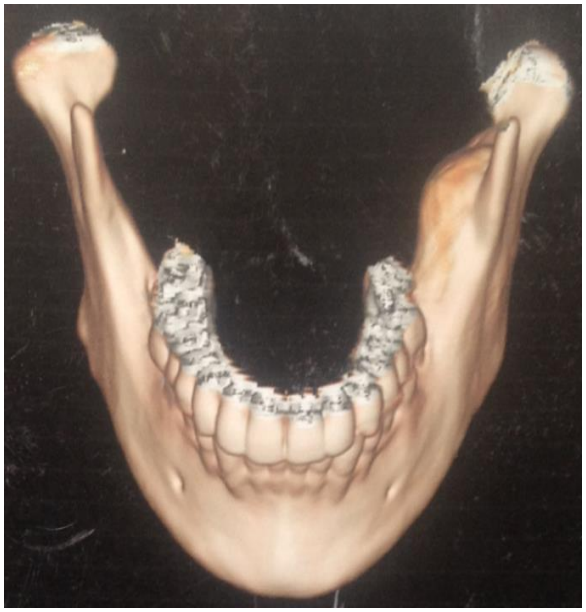


Fig. 3: (c) Sagittal section of the CT scan showing osteolytic lesion involving the body and entire ramus



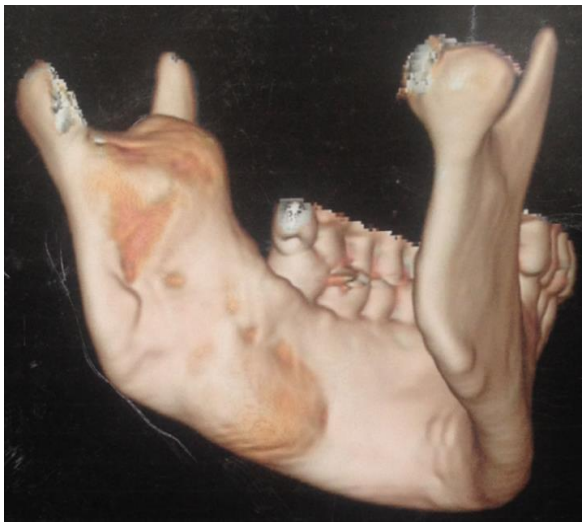


Fig. 3: (d) 3D reconstruction of the CT-scans

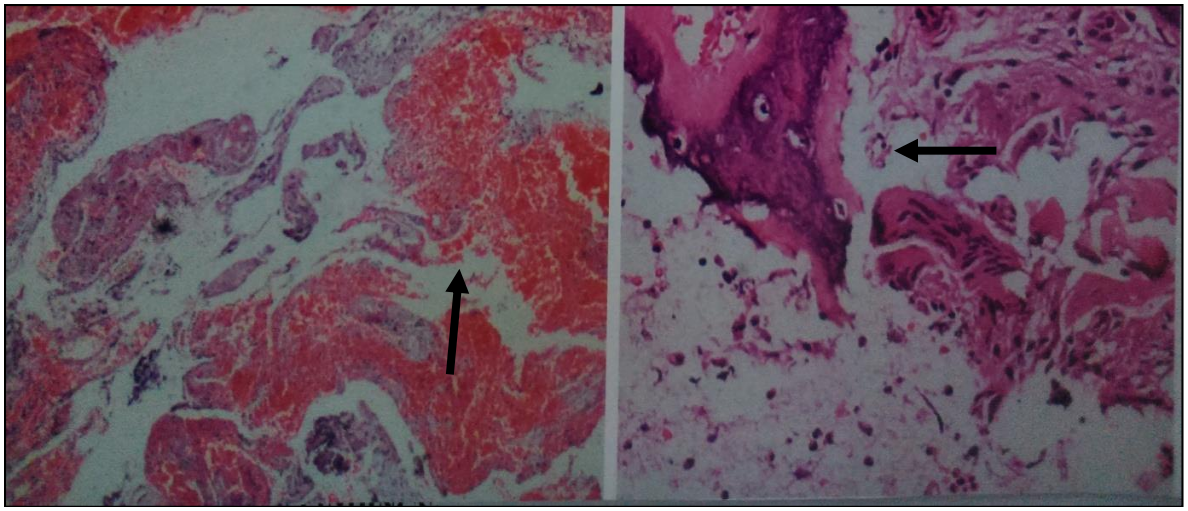


Fig. 4: (a) Sinusoidal vascular spaces, (b) Bone formation and multinucleated giant cells

Discussion

Aneurysmal bone cysts were first described by Jaffe and Lichenstein in 1942.[1] The cause and pathogenesis of aneurysmal bone cysts are poorly understood. Several investigators have proposed that it arises from a traumatic event, vascular malformation, or neoplasm that disrupts the normal osseous hemodynamics and leads to an enlarging, hemorrhagic extravasation. In support of this theory, it has been suggested that aneurysmal bone cyst and giant cell granuloma are closely related. An aneurysmal bone cyst may form when an area of haemorrhage maintains connection with the disrupted feeding vessels, giant cell granuloma-like areas can develop after loss of connection with the original vascular source. Another school of thought suggests that it may occur either as a primary lesion or as a result of disrupted vascular dynamics in a pre-existing intra-bony lesion.[5]

Cytogenetic analysis has demonstrated the presence of various chromosomal abnormalities in some cases, particularly those involving 17p11-13 and 16q22.However, the significance of these chromosomal abnormalities in the molecular pathogenesis of the aneurysmal bone cyst remains unclear.[5]

Aneurysmal bone cysts are located most commonly in the shaft of a long bone or in the vertebral column. It is generally found in patients younger than the age of 30, although a wide age-range is noted. Gnathic aneurysmal bone cysts are uncommon, with approximately 2% reported from the jaws.A mandibular predominance is noted, and the vast majority arises in the posterior segments of the jaws. No significant gender predilection is noted. ABC usually presents as a rapidly growing swelling. Paraesthesia is a very rare feature. Crepitus and perforation is also uncommon. On occasion, malocclusion, mobility, migration of involved teeth may be present.[4] Maxillary lesions often bulge into the adjacent sinus; nasal obstruction, nasal bleeding, proptosis, and diplopia are noted uncommonly.[5]

In the present case, ABC was found in a 48 year old male involving the left mandibular body and ramus. The age of the patient was found to be relatively higher than what has been reported in the literature. Certain rare features such as occasional mild pain and paraesthesia with respect to the left lower lip were also noted.

Generally, the radiographic features of ABC are not diagnostic. These lesions are usually unilocular, but long standing lesions may show a "soap-bubble" appearance and may become progressively calcified.[6] Destruction of the bone cortex and periosteal reaction is rare.[7]

In the present case, conventional radiographs revealed extensive bony involvement extending from the left symphysis to ramus of the mandible with a few faint wispy septae depicting a multilocular soap bubble appearance. CT scan is far superior to plain radiography and often reveals a thin cortex of bone, expansile nature of lesion and fluid-fluid levels within the lesion. CT-scans also accurately identify tissue septae. MRI findings of fluid-fluid levels inside the lesion are highly specific of aneurysmal bone cysts.[3] Air-fluid levels are due to layering of solid blood components within the cyst.[8]

The differential diagnosis of a swelling in the mandibular posterior region includes Infected Radicular cyst, KOT, Ameloblastoma, Central giant cell granuloma, Odontogenic myxoma, Traumatic bone cyst and Aneurysmal bone cyst. In a case of radicular cyst, a history of trauma or a source of infection like a deep dental caries may be present. A similar swelling may be present but expansion of the cortex is rare.

KOT presents as a slow growing swelling in the mandibular posterior region with a buccal cortical expansion.

Ameloblastomas are painless slow growing swellings with mobility of the teeth in the involved region and root resorption may be a feature along with paraesthesia.

CGCG also presents as a painless swelling affecting mostly females under the age of 30 commonly affecting the anterior mandible. It rarely perforates the cortex.

Odontogenic myxoma is a slowly enlarging painless expansion of the jaw with possible spreading, loosening and migration of teeth. There is a slight female preference and the age range affected is 10 to 50 years.

Traumatic bone cyst usually will present with a history of trauma with the lesions being mostly asymptomatic and detected on routine radiographs.[4]

In the present case, CT scans revealed perforation of the cortex in the retromolar trigone in the 3rd molar region which is an unusual feature of ABC.

On macroscopic examination ABCs have sponge-like appearance, consisting of blood-filled cavities separated by thin, fibrous septa.[7]

Microscopically, the aneurysmal bone cyst is characterized by spaces of varying size, filled with blood. It is surrounded by cellular fibroblastic tissue containing multinucleated giant cells and trabeculae of osteoid and woven bone. On occasions, the wall contains an unusual lacelike pattern of calcification that is uncommon in other intraosseous lesions. The blood-filled spaces are not lined by endothelium. In approximately 20% of the cases, aneurysmal bone cyst is associated with another pathosis, most commonly a fibro-osseous lesion or giant cell granuloma.

At the time of surgery, intact periosteum and a thin shell of bone are typically found covering the lesion. Cortical perforation may occur, but spread into the adjacent soft tissue has not been documented. When the periosteum and bony shell are removed, dark venous blood frequently wells up and venous like bleeding may

be encountered. The appearance at surgery has been likened to that of a "blood-soaked sponge."[5]

With regard to the treatment, conservative surgical resection of the lesion is the treatment of choice. This must be limited to careful enucleation or curettage of the mass as it is a benign process. Segmentary resection must be done only in case of multiple recurrences or extension to overlying tissues. Other options are the surgical excision and curettage of the cavity. The use of radiotherapy is not recommended because of the probability of radio-induced tumours.[7] The present case was planned to be treated with hemimandibulectomy and free fibula reconstruction graft as the lesion was quite an extensive one with perforation of the cortex.

Recurrence rates range from 20% to 30% according to different series and seems to occur most frequently within the first year after surgery. This is usually attributed to incomplete removal of the lesion especially in soft tissue invasive cases. Several authors recommend immediate reconstruction of the defect with autogenous grafts in cases of aesthetic deformity and in cases with high risk of fractures and loss of mandibular continuity.[3]

Conclusion

Aneurysmal bone cyst still remains an enigma, not only regarding the cause, but also regarding clinical diagnosis, imaging, and optimal treatment. Recently some markers have been identified which are specific for aneurysmal bone cyst. There is still no reliable system which would establish diagnosis, support and define different treatment modalities so as to eliminate the problems encountered both by the patient and the surgeon.

Conflict of Interested: None
Source of Support: Nil

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