



# Intracranial Extradural Sphenoid Wing Aneurysmal Bone Cyst Extending into Orbit: A Unique Case

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## Abstract

Aneurysmal bone cysts (ABC) are benign, expansile, osteolytic and fibro-osseous tumour like lesions commonly seen in children with 80% occurring in patients less than 20 years of age. The origin of ABC is still debated. We describe a case of 8-year-old female with ABC involving right greater wing of sphenoid with intra-orbital extension leading to proptosis. Imaging revealed an expansile multiloculated cystic lesion with fluid-fluid level involving orbital surface of greater wing of sphenoid. The patient underwent right pterional craniotomy and excision. The lesion was multiseptate with dark chocolate coloured fluid and without any dural invasion. Early intervention preserved child's visual functions. Exact diagnosis by imaging studies and early surgical resection with complete resection of mass prevents recurrence.

**Keywords:** Aneurysmal Bone Cyst; Greater Wing of Sphenoid Orbit; Craniotomy

## Abbreviations

ABC: Aneurysmal Bone Cysts; CT: Computed Tomography; MRI: Magnetic Resonance Imaging.

## Introduction

Aneurysmal bone cysts (ABC) are benign, expansile, osteolytic and fibro-osseous tumour like lesions commonly seen in children with 80% occurring in the patients less than 20 years of age [1]. The pathogenesis of this entity is still not clear. These can be of 2 types of lesions, one secondary to an identifiable precursor and second, primary ABC without a pre-existing lesion. The metaphysis of a long bones (tibia and femur) is most common site and presents painful bony swelling and associated pathological fractures [1]. It is an uncommon tumour seen in Calvaria. Only 2-6 % of ABC's are

found in the skull with the involvement of temporal, occipital and frontal bones [2]. These tumors, although benign, are locally aggressive, causing mass effect on the surrounding structures. Depending upon the site, the presentation can vary. It can be painful, palpable swelling or associated with other symptoms like headache, proptosis, hearing loss, focal neurologic deficits, cranial nerve palsies and signs of raised intracranial pressure.

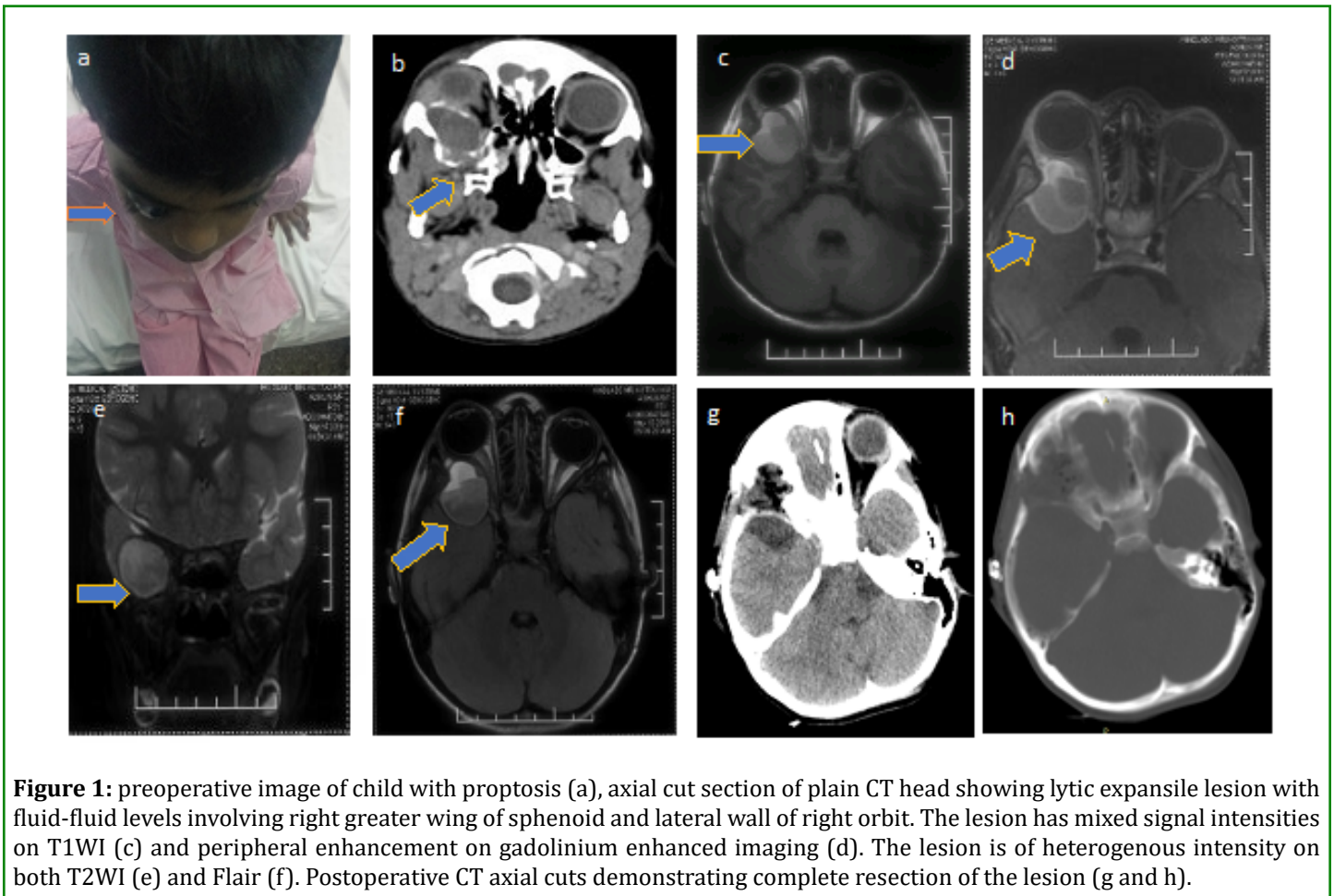
We report a case of 8-year female with aneurysmal bone cyst involving the right greater wing of sphenoid with intra-orbital extension leading to proptosis.

## Case Report

A 5-year-old female patient was admitted to our institute complaining of painless non-pulsatile proptosis of right eye

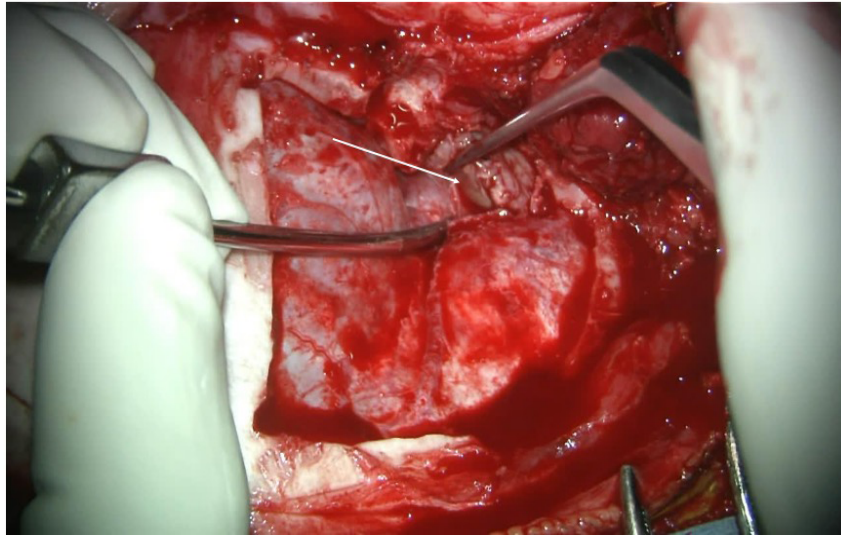
for 3 months of duration. No history of headache, vomiting, blurring of vision or seizures, trauma. Her perinatal history was normal without any history of developmental delay. On examination, higher mental functions were within normal limits. Extra-ocular eye movements and pupils were unremarkable, however, there was proptosis of right eye. Her visual acuity was 6/6 in both eyes. There was no temporal swelling or localized tenderness, no bruit, conjunctival congestion, or epiphora. Routine blood investigations were within normal limits. Visual field analysis did not show any field cut. There was no prolongation of P100 latencies on visual evoked potential. On X-ray skull, a lytic lesion on the supero-lateral orbital margin was noted. CT head showed

well defined expansile lesion involving right greater wing of sphenoid and lateral wall of right orbit with medial displacement of right optic nerve. There were fluid levels within with hyperdense contents in the dependent part. MRI brain revealed an expansile multiloculated cystic lesion with fluid-fluid level involving orbital surface of greater wing of sphenoid compressing right anterior temporal lobe posteriorly and right optic nerve medially. It measured 2.8x2.4x2.3 cm and had mixed signals intensities on T1WI, T2WI and FLAIR sequences. On contrast imaging, there was peripheral enhancement of lesion and right middle cranial fossa dura Figure 1.

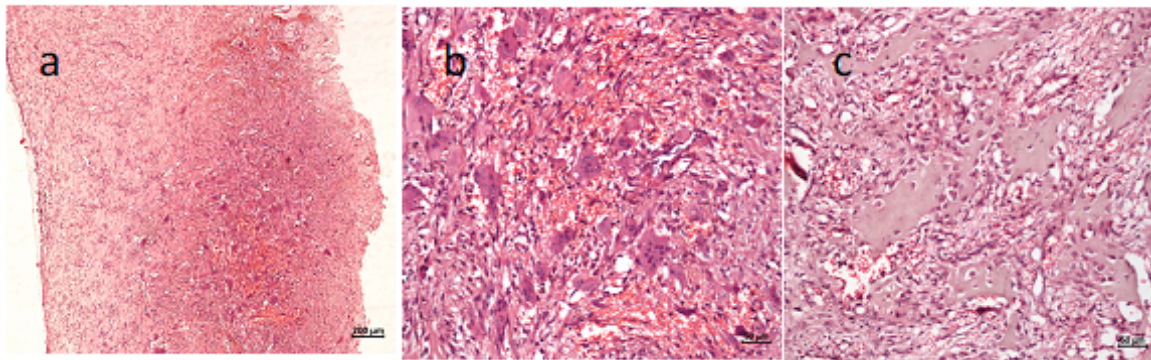


The patient underwent right pterional craniotomy and excision. During craniotomy, the lesion with its septations were uncovered near the sphenoid wing with drainage of dark chocolate coloured fluid. The thin septations were opened and the soft cystic walls were excised meticulously without injuring temporal pole dura and periorbital tissue relieving the compression. Figure 2 There was no dural invasion. Frontotemporal bone flap was replaced and fixed

with plates and screws. Post operatively patient developed right eye chemosis that was managed conservatively. Post-operative visual functions were intact, and proptosis reduced significantly. Histopathological examination was suggestive of cyst wall composed of bland fibroblasts with osteoclast-type multinucleate giant cells and reactive woven bone rimmed by osteoblasts Figure 3.



**Figure 2:** Intraoperative image of the lesion seen after front-to-temporal craniotomy (white arrow) at sphenoid region.



**Figure 3:** Histopathological features show cyst wall composed of bland fibroblasts (a) with osteoclast-type multinucleate giant cells (b) and reactive woven bone rimmed by osteoblasts (c) [Haematoxylin and Eosin, Original magnification: 50X (a) and 200X (b, c) Scale bars: 200µm (a) and 50µm (b, c)].

## Discussion

Aneurysmal bone cyst is a benign, multi-cystic, vascular lesion of younger age group involving metaphysis and spine. Calvarial involvement is seen rarely with a known incidence of 2-6%. In this case, patients have presented symptoms of painless proptosis of very short duration [2]. Other symptoms in ABC's of skull are diplopia, ptosis, headache, visual deterioration, and nasal congestion. The duration of symptoms to be brought into notice is about 2 months and sudden proptosis secondary to rapid expansion of the lesion due to haemorrhage is common [2]. Erosion of the affected bone has been reported due to this entity [3].

There are 4 radiological phases described – osteolysis, rapid growth, stabilisation and healing with progressive ossification by Dabska and Burdue to toewski. Short history suggests a phase of rapid growth [4]. Majority of cases seen are extracranial with mass effect as seen in our case causing proptosis.

Temporal lobes were also seen to be compressed extracranially by this lesion. Jaffe [5] and Lichtenstein [6] described first aneurysmal bone cysts and are well known to affect long bones, spine and pelvis. It comprises of a macroscopic appearance of cyst, blood-filled cavity within an expanded bony wall thereby giving rise to terminology aneurysmal bone cyst.

There are several proposed theories of aneurysmal bone cysts. Several cases have been reported as post traumatic events. Another causative factor has been defined as reaction secondary to another bony lesion such as giant cell tumors [7]. Another pathogenesis as suggested by Lichtenstein [6] is that ABC is due to local circulatory disturbance due to sudden occlusion of venous drainage from that segment of bone or development of an arteriovenous shunt which in turn causes cystic blood-filled spaces leading to bony expansion with atrophy [8,9]. As an etiological factor trauma has a controversial role [6]. Costantini FE, et al. [10] postulated the proliferation of the internal periosteum through the dural erosion for the intracranial origin dural elements were found on histology. Amidst variable clinical presentation the diagnosis of an ABC can be challenging with varied locations of this entity [10].

In the present case, the patient presented with proptosis due to compression of orbital contents from laterally over optic nerve. In our case, craniotomy offered a complete excision of cyst as well as cyst wall, the orbit and optic nerve with relief of proptosis seen evidently on post-operative period. Imaging modalities such as CT and MRI are essential to aid both diagnosis and for surgical planning. CT in majority of lesions have expansile, lucent bone lesions with osseous remodelling and cortical thinning [10].

MRI features are fluid-fluid levels, multiple internal septations, soap bubble appearance due to the presence of small multiple small cysts which are projected from larger cysts as seen in our case. The radiological differential diagnosis with fluid-fluid levels is chondroblastoma, osteosarcoma, giant cell tumor, fibrous dysplasia, osteoblastoma, and tumoral calcinosis [11]. Diverticula is also a feature in this case which is the presence of small cysts projected from larger cysts. Due to these features a bubbly appearance is also seen in these cases [12]. Histology is the definite answer of this entity in establishing the diagnosis which demonstrates irregular, blood-filled chambers with islands of bone and fibrous tissue [13].

Surgical excision has been suggested for these aggressive tumours although cryosurgery was previously suggested. Option of radiotherapy with a dose of 30 to 36 Gy has also been considered for large size tumour to reduce vascularity preoperatively to facilitate the excision and has been cited in cases of recurrent cases of temporal bone ABCs [6,14,15].

Role of pre-operative embolization to decrease the vascular burden of the tumor and facilitate surgical removal is also demonstrated [16]. Other modalities reported in the literature treated with repeated intralesional calcitonin injections using an Ommaya reservoir [16].

## Conclusion

To conclude, it requires adequate imaging at an earlier period with MRI and CT scans which will give an idea about the primary pathology so early intervention can be planned to improve the outcomes.

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