A Rare Case of an Aneurysmal Bone Cyst of Temporal Bone

Islam KMT1, Alom S2, Haque M3, Chowdhury D2, Hossain M4

1Dr. K. M. Tarikul Islam, Associate Professor, Paediatric Neurosurgery, Department of Neurosurgery, Bangabandhu Sheikh Mujib Medical University, Dhaka, Bangladesh
2Dr. Shamsul Alom, Associate Professor, Vascular Neurosurgery, Department of Neurosurgery, Bangabandhu Sheikh Mujib Medical University, Dhaka, Bangladesh
3Dr. Shaan Muhammad Iran, Department of Neurosurgery, Bangabandhu Sheikh Mujib Medical University, Dhaka, Bangladesh
4Prof. Moududul Haque, Department of Neurosurgery, Bangabandhu Sheikh Mujib Medical University, Dhaka, Bangladesh
5Prof. Dhiman Chowdhury, Skull Base Neurosurgery, Department of Neurosurgery, Bangabandhu Sheikh Mujib Medical University, Dhaka, Bangladesh
6Prof. Mohammad Hossain, Spinal Neurosurgery, Department of Neurosurgery, Bangabandhu Sheikh Mujib Medical University, Dhaka, Bangladesh

Abstract: Aneurysmal bone cyst are uncommon lesions of long tubular bone. It is benign and nonneoplastic. Aneurysmal bone cyst is unusual in temporal bone that may extend intracranially. We report here a patient of 6 years old boy presented with swelling over left temporal region for 2 months. CT and MRI findings and histopathology revealed aneurysmal bone cyst. Total surgical resection was ensured though lesion was extended intracranially. Prognosis is excellent after total removal as total surgical excision is curative. Good surgical result of this uncommon lesion prompted our report.

Keywords: tubular bone, bone cyst, Prognosis, swelling.

INTRODUCTION

Aneurysmal Bone Cysts (ABCs) are rare benign bone lesion which mainly occurs in the metaphyseal ends of the long bones [1, 2]. ABC is uncommon in skull with reported incidence of only 1% [3]. Most of the cases of ABC occur within 3rd decade of life. Although rare, occurrence of ABC after trauma has been documented. Aneurysmal bone cysts occupy the grey area between distinctly non-neoplastic conditions of the bone and true neoplasms [4]. An uncommon case of ABC in left temporal region of young child is reported here.

CASE REPORT

A 6 years old boy presented with swelling on the left temporal region for 2 months following trauma to head sustained during sports. Patient's parents also complained intermittent pain around the swelling for same duration. There was no history of ear discharge, vertigo, nausea, vomiting or seizure. There was no symptoms of raised ICP; neither any visual or ocular symptoms.

Clinical examination showed fullness of left temporal region. A non tender mass was evident in left temporal region. A non tender mass was evident in left temporal region for 2 months following trauma to head sustained during sports. Patient's parents also complained intermittent pain around the swelling for same duration. There was no history of ear discharge, vertigo, nausea, vomiting or seizure. There was no symptoms of raised ICP; neither any visual or ocular symptoms.

X-Ray skull both view reveals normal findings. CT scan of left temporal bone showed an expansile lytic lesion involving petrous, squamous and mastoid part of left temporal bone with thinning out of squamous part. MRI of Brain revealed expansile complex cystic lesion with minimal internal hemorrhage on left temporal lobe with evident compression over the left lateral ventricle resulting in midline shift. Multiple fluid level were present. The lesion was exposed by left temporal approach. Incision was extended below zygomatic arch to gain access to infratemporal part of the tumor. After reflecting the temporalis muscle, lesion was found, outer table of temporal bone was thinned out and perforated through which dark-colored blood was coming out. Tumor was...
thoroughly curetted in all directions until the normal tissue was visible. Medially, tumor was removed until dura was reached. Total surgical excision was ensured. Cranioplasty was done with bone cement. Postoperative period was uneventful and patient was followed up to 3 months and was symptom free.

Histopathology report showed cystic spaces without endothelial lining. The cystic spaces are separated by fibrocollagenous septa containing woven bone, multinucleated giant cells & infiltration of chronic inflammatory cells. The areas of haemorrhage are also seen.

Fig. A: The patient showing swelling in the left temporal region
Fig. B: CT scan showing fluid level and erosion of temporal bone
Fig. C: MRI showing heterogenous lesion with fluid levels
Fig. D: Intraoperative image showing well defined capsulated lesion, soft in consistency involving adjacent petrous bone
Fig. E: Peroperative image showing cranioplasty
Fig. F: Microphotograph of ABC shows cystic spaces containing RBCs separated by septa containing spindle shaped cells and scattered multinucleated giant cells
DISCUSSION

ABC was first described by Jaffe & Lichtenstein in 1942, when it was shown as distinct pathological identity of bone [5]. 80% of ABCs occur within the first two decades of life with a mean age at presentation of 16.1 years [5]. ABCs has a slight female preponderance with a male female ratio of 0.84:1 [6]. Most of ABCs occurred in the temporal and occipital bone observed by Sheikh et al. The cause behind the pathogenesis is still not to be fully understood. Local trauma has been suggested as a cause of ABC [7-10].

ABC has association with other disease identities like Fibrous dysplasia, Chondrosarcoma, Osteoclastoma, Non-ossifying fibroma, Giant cell tumor & Osteoblastoma. It can occur both in developing and mature bone. ABC can occur anywhere in the skeleton but predilection to long bones and vertebrae is evident. Their occurrence in skull is rare with an incidence of 3-6% [11]. Histologically anastomosing vascular channels of varying sizes are seen. These spaces don't have wall like those seen in blood vessels.

CT scan of affected part is superior to plain radiograph to well delineate the lesion. Multiple fluid levels are important characteristics of ABC on CT scan which signifies sedimentation of RBC in the blood filled cavities [12].

Different treatment modalities include simple curettage, complete surgical excision, radiotherapy, cryosurgery and endovascular embolisation. Among all treatment of choice is total excision as it is curative [13, 14]. Simple curettage is related with high recurrence rates from 21 to 50%. Radiotherapy is not recommended though it is written in the literature, advocated for deep lesions at the base of the skull with dural involvement where total excision is not possible [15]. Radiotherapy is contraindicated in the treatment of ABC with fibrous dysplasia as there is chance of malignant transformation [16]. In the treatment of ABC chemotherapy has no role. To reduce bleeding during surgery endovascular embolisation has marked contribution for preoperative devascularization. Endovascular embolisation may be applied where surgical resection difficult [17].

Declaration of Patient Consent

We have obtained all appropriate patient consent forms. Patient’s attendant has given consent for clinical informations and images. His identity will be concealed but anonymity cannot be guaranted.

Financial Support and Sponsorship: Nil.

REFERENCES


Conflict of Interest: No conflict of interest.