Case Report

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Aneurysmal Bone Cyst at Dorsal Spine: Case Report and Review of Literature

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Abstract

An aneurysmal bone cyst (ABC) is a benign, locally proliferative vascular disorder of non-neoplastic osseous lesions in children and young adults, usually involving long bones. Spinal ABCs are rare. We are reporting a rare case of ABC involving dorsal spine presented with spastic paraplegia and discussing management along with review of literature.

Keywords: Spinal tumour; Extradural Tumour; Aneurysmal Bone Cyst.

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Introduction

An aneurysmal bone cyst (ABC) is a benign, locally proliferative vascular disorder of non-neoplastic osseous lesions in children and young adults. These are the large cystic blood-filled spaces. 75% of ABCs occur before the age of 20 years. They comprise 1.4% of all primary bone tumors, and commonly occur in the long bones.[1] Spinal ABCs are much rarer. Complete excision is the rule for symptomatic ABC but, asymptomatic ones (characterized by clinically insignificant destruction of the bone) are generally left alone with just close monitoring for any abnormal changes.

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We are reporting a rare case of ABC in the dorsal spine of 8 year old boy presenting with spastic paraparesis, which was successfully treated with complete excision.

Case Report

A 8 year old boy was presented to us with pain in upper back, progressive weakness in bilateral lower limb leading to spastic paraplegia with bowel bladder involvement (BBI). His neurological examination was showing power in B/L lower limb 2/5 with diminished sensations over B/L lower limbs. Knee and ankle jerks were exaggerated (3+) with significant increase in tone. His lab investigations were within normal range. Magnetic resonance imagimg (MRI) Dorsal spine with contrast was showing expansile lytic lesions in post elements (B/L laminae, right pedicle, spinous process) of D5 and D6 vertebra. Involved vertebrae reveals hyperintensity on both T2W images, T1W images and shows heterogeneous contrast enhancement. The expanded post elements are seen encroaching the posterior thecal sac leading to canal narrowing and cord compression. There are septae noted within the lesion showing heterogeneous enhancement

on Gadolinium contrast suggestive of Anueysmal bone cyst (Fig. 1). Patient underwent D5, D6 and partial D7 Laminectomy with involved right D5, D6, facet along with complete excision of tumor and dural decompression. The vertebral column was reconstructed with titanium transpedicular (B/L D4, D6,D7) screw and rod fixation. Post-operatively, his condition was improving. Histopathological examination microphotograph showed large cysts lined by fibroblasts, myofibroblasts and histiocytes spaces filled with blood and thin fibrous septa (Fig. 2A, H&E, 100X). Clusters of osteoclast-like multinucleated giant cells with loose spindly cellular stroma are seen along with hemosiderin deposits, charecteristics of ABC (Fig. 2). Pain was relieved gradually over few days and complete resolution of sensory motor symptoms was achieved over 10 weeks.

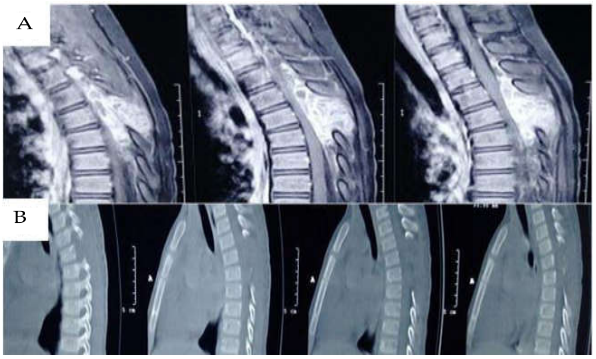

Fig. 1: Magnetic resonance imagimg (MRI) Dorsal spine with contrast (A) was showing expansile lytic lesions in post elements (B/L laminae, right pedicle, spinous process) of D5 and D6 vertebra. Involved vertebrae reveals heterogeneous contrast enhancement. Computed Tomography Scan of Dorsal Spine (B) shows expansile extra-axial lytic lesion causing destruction and thinning of posterior elements.

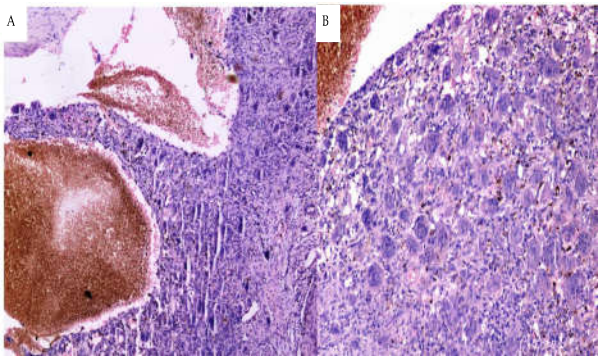

Fig. 2: Photomicrographs show large cysts lined by fibroblasts, myofibroblasts and histiocytes spaces filled with blood and thin fibrous septa (Fig. 2A, H&E, 100X). Clusters of osteoclast-like multinucleated giant cells with loose spindly cellular stroma are seen along with hemosiderin deposits (Fig. 2B, H&E, 200X).

Discussion

Jaffe and Lichtenstein first described ABC in 1942, when they noted "a peculiar blood containing cyst of large size". ABCs can be primary and secondary ABC. If ABC is having association with entities such as fibrous dysplasia, giant cell tumors and/or osteoblastomas are defined as secondary ABC and this association was found approximately in 20-30% of cases.[2] In 69% of primary ABCs there is characteristic association of clonal t (16;17) genetic translocation which can lead to an upregulation of the TRE17/USP6 oncogene. These lesions were staged according to Capanna et. al.'s classification.[3] Inactive cysts have a complete periosteal shell with defined sclerotic bone limits. Active cysts have an incomplete periosteal shell and defined bone limits. Aggressive cysts have an indefinite margin and show uniform osteolysis. The active or aggressive character in certain localisations of ABCs in children requires either curettage with a considerable recurrence rate or a radical segmental excision raising complex reconstructive challenges.[4] Cyst maturation with subsequent ossification may be observed either spontaneously or after incisional biopsy.[5]

ABCs involves metaphyses of long bone usually. In 10% of cases of all ABCs, Spinal involvements are reported; mostly in the thoracic and lumbar regions. It starts with the involvement of pedicle and then lesion extends into the vertebral body anteriorly and posterior elements. Spinal ABCs are commonly seen in 10-20 years with slight female preponderance similar to long bone ABC.[6,7]

Pain is found to be most common symptom. patient may also present with sensory disturbance, motor weakness, paravertebral mass, or kyphoscoliosis. They can also present with acute symptoms of root/cord compression. Differential diagnosis of ABCs includes giant cell tumor, hemangioma, fibrous dysplasia, osteosarcoma, osteoblastoma and metastatic lesions.[2]

Complete resection of the lesion is considered to be the treatment of choice with excellent outcome. Total excision must include the entire cyst wall, all abnormal tissues that feel spongy and bone surfaces that are lined with fragile and hypervascular membranes. For extensive lesions and those having instability in spine require stabilization.[7]

It is found that ABC is thought to arise from a pre-existing lesion due to a superimposed secondary vascular phenomenon which start a periosteal or intraosseous arteriovenous malformation resulting in the erosion of the osseous trabeculae into a cystic cavity. In patients is not having any neurological involvement and with pain as the only symptom can be treated with selective arterial embolization alone or by intralesional calcitonin and methylprednisolone injection or vertebroplasty. The results of embolization can be seen in the form of involution of the soft-tissue component, sclerosis, and ossification. The intralesional injection of calcitonin can induce the formation of cancellous bone and inhibit the osteoclastic activity. Methylprednisolone can reduce the fibroblastic action and the angiogenesis.[8,9]

Radiation alone is not having any beneficial effect and may be associated with additional risk of malignant transformation. Adjuvant radiation may be given to patients with inoperable lesions, hypervascular lesions, aggressive recurrent disease, or high risk surgical candidates.[10]

Conclusion

Aneurysmal bone cyst is a rare extradural tumour of the spine presented with charectrestic radiological faeatures. Complete excision is the rule for cure and stabilization may be required in the presence of instability. Age, location, size and number of mitotic figures have been suggested for recurrence. Histological examination should be done to prevent overlooked of an underlying more aggressive neoplasm.

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