Aneurysmal bone cyst of the thoracic spine

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Introduction

An aneurysmal bone cyst (ABC) is a non-neoplastic osseous lesion with a benign, locally proliferative vascular disease in children and young adults. Seventy-five percent of ABCs are formed before the age of 20 and occur most frequently in the long bones. Spinal ABCs are substantially relatively uncommon. We discuss a rare occasion of ABC affecting the thoracic spine that was successfully treated surgically.

Case Report

A 12-year-old girl presented with two weeks history of progressive paraparesis without pain. On examination, she had weakness of grade 3/5 both sides with sensory impairment below the knee level. Her bladder and bowel habits were normal. There was no previous history of trauma.

Computed tomography (CT) scan of cervico-thoracic spine with reconstruction showed an expansile osteolytic lesion with several thin internal septations and sclerotic margin, involving the T1 vertebral body and its bilateral pedicles and transverse processes, bilateral par interarticularis and spinous process. No definite cortical destruction is noted [Figure 1].

Magnetic resonance imaging (MRI) showed a bone mass at T1 level involves body, lamina and spinous process with fluid level inside the mass [Figure 2]. There was extension of the lesion into the spinal canal and pressure on the both sided nerve roots.

She underwent angiography with tumor embolization, then laminectomy T1 with tumor removal with posterior fixation C6-7 and T2-3, followed by anterior approach with T1 corpectomy with fusion with iliac bone graft. The intraoperative findings showed tumor encasing bilateral laminar, pedicle and facet with pathologic fracture, egg shell (fragile) consistency at T1 level. The tumor was completely excised. Histopathology of the lesion showed Histopathology of the lesion showed uniformly distributed osteoclast type giant cells having multiple nuclei [Figure 3]. There were few cysts containing hemorrhage and were lined by histiocytes and osteoclast giant cells. These features were suggestive of secondary aneurysmal bone cyst. Post operatively, she had complete improvement in weakness. Post operative CT showed the implant in position [Figure 4].

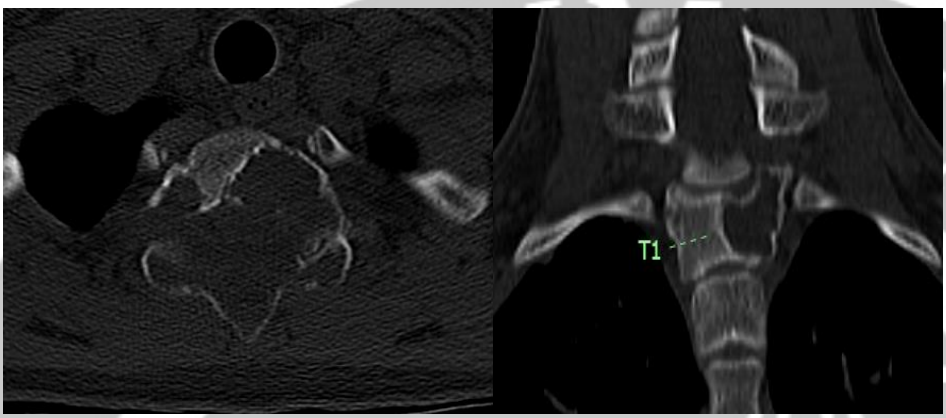

Figure 1 Thoracic spine CT scan showing well-circumscribed expansile lytic with periosteal calcification lesion (eggshell appearance) involving the T1 vertebral body

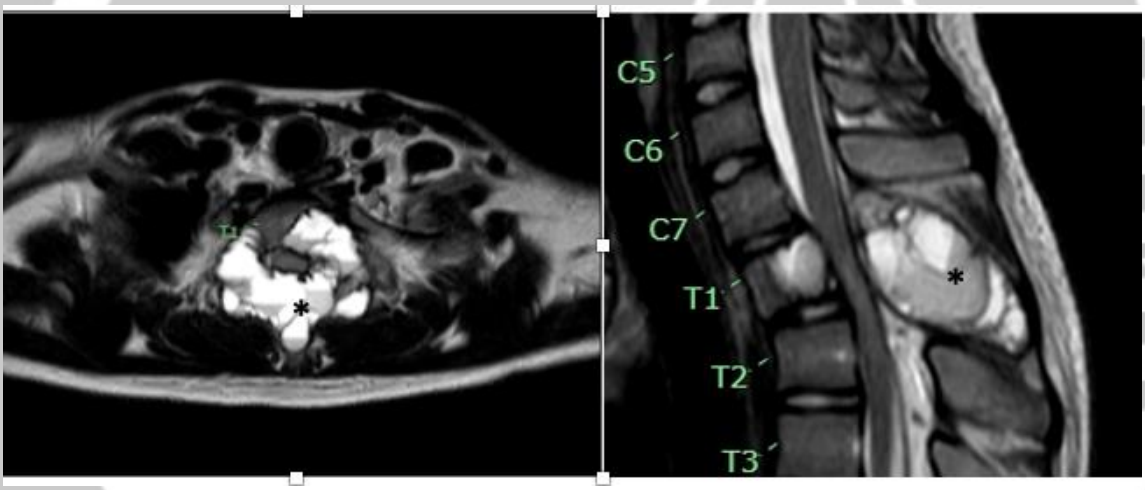

Figure 2 MRI scan of the thoracic spine showing multiple fluid-fluid levels involving T1 vertebral body, bilateral pedicles, lamina and spinous process –"Soap bubble" (Asterisk) with spinal cord compression

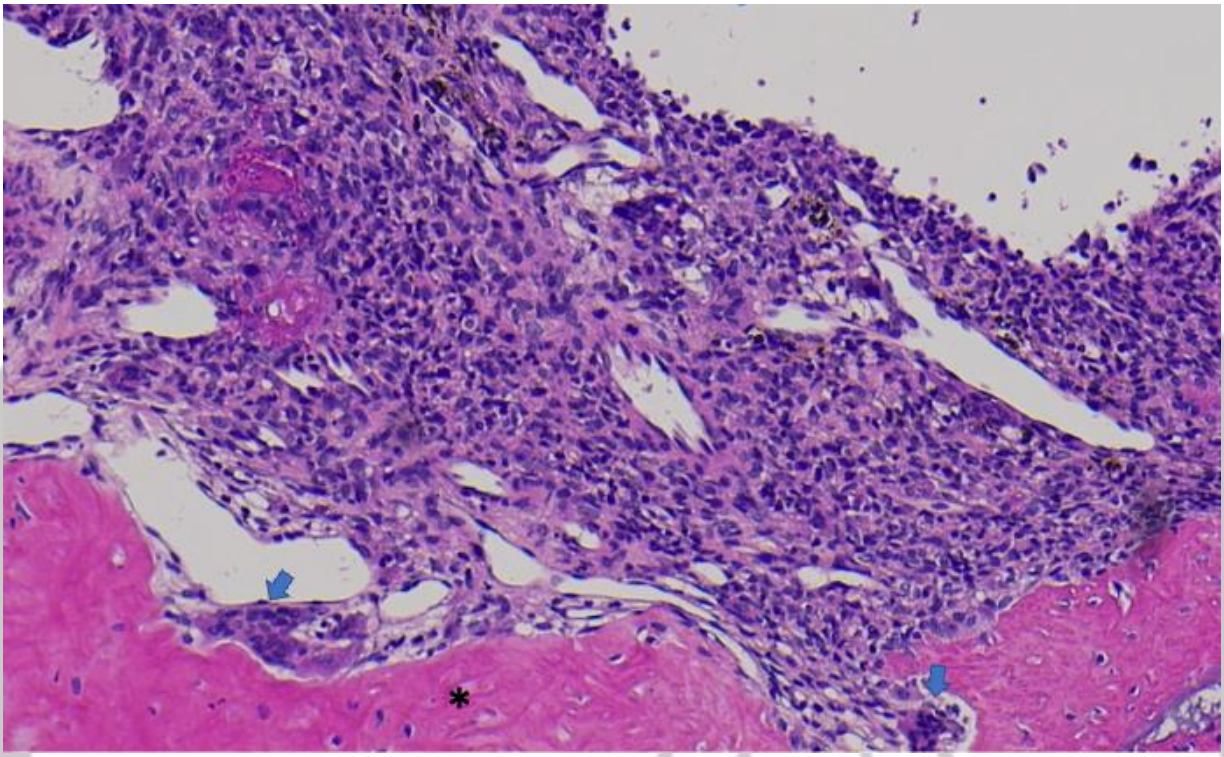

Figure 3 H and E stains showing osteoclast type giant cells having multiple nuclei (Arrow head). Cyst (Asterisk) containing hemorrhage and lined by histiocytes and osteoclast giant cells can be noted.

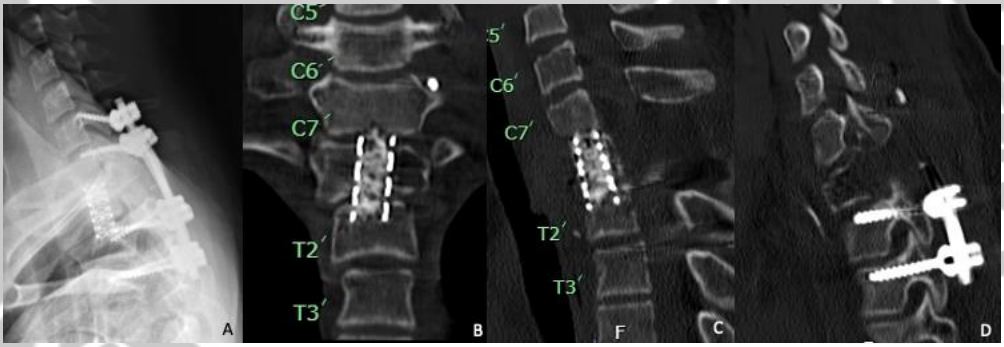

Figure 4: (A) Radiograph showing implantation from C6-T3. (B-D) CT of the cervicothoracic spine showing T1 anterior reconstruction with C6-C7 lateral mass screws and T2-T3 pedicle screws

Discussion

Aneurysmal Blood Cysts (ABC) has been defined by the World Health Organization (WHO) as a benign tumor like lesion, "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." (Schajowicz F. Berlin, Germany: Springer-Verlag; 1992)

ABC has been first characterized in the year of 1942 where it has been identified as "a peculiar blood-containing cyst of large size."(Jaffe HL, Lichtenstein L. 1942) ABC is not at all common, comprising only around 1.4% of all bone tumors. Metaphysis of long bones including the humerus, femur, tibia, and the flat bones of the pelvis are most frequently affected. 10% of cases have spinal involvement, with the thoracic and lumbar regions being the most common affected areas. With a slight female preponderance, ABC is frequently seen in children and teenagers with the age range of 10 to 20 years old. Typically, the lesion begins at the posterior elements, then to the pedicles and progresses to the vertebral body. [3,4] [5,6] ABC is believed to develop from a pre-existing lesion as a result of a superimposed secondary vascular phenomena and causes an intraosseous or periosteal arteriovenous malformation which the results in osseous trabeculae eroding into a cystic cavity. Histologically speaking, a primary ABC is described as anatomizing fibrous-walled channels with an endothelial cell lining, either fully or partially, but lacking the elastic lamina or muscle layers of true blood vessels. The microscopic features of a secondary ABC are identical to the primary ABC, but often with evidence of a concurrent, benign or malignant lesion.

For lesions difficult to access surgically, either intralesional injections or selective embolization may be used to decrease tumor size (Bush et al., 2010; Rossi et al., 2010; Rossi et al., 2017). Multiple types of substances have been used in intralesional injections, including but not limited to, polidocanol, calcitonin and prednisolone, each with varying degrees of effectiveness (Chang et al., 2017; Deventer et al., 2021). However, surgery is considered first-line treatment for ABCs (Shih et al., 1998, Ibrahim et al., 2012).

Surgical procedure depends on ABC staging. Stage 1 ABCs, which are located centrally in the bone, can be treated via intralesional curettage, in which only the tumor is removed while the roof of the spine is left intact (Chowdry et al., 2010). Stage 2 ABCs, which are located centrally and invade the bone's entire diameter, require intralesional excision. This means that a bone window has to be opened before the surgeon can remove the ABC's contents. En-bloc or wide excision is

typically reserved for stage 3 ABCs, which are eccentrically located and not amenable to intralesional excision. These types of ABCs usually require reconstruction via structural allografting or prosthetics post-tumor removal (Park et al., 2016).

From our radiologic findings, it is evident that our patient has a stage 3 ABC. Her aneurysmal bone cyst is located eccentrically at the T1 level. Due to the nature of the lesion, surgery will include either en-bloc or wide excision followed by reconstruction. Before the series of operations, our patient first underwent preoperative embolization of the left ascending cervical artery, to reduce intraoperative bleeding. Three days later, her bone tumor was removed via a T1 laminectomy. and her spine was stabilized via posterior fixation at C6-7 and T2-3. Four days after her tumor removal, she underwent a T1 lumbar corpectomy and fusion with an iliac bone graft.

Conclusion

Aneurysmal bone cysts (ABCs) are benign but locally aggressive bone tumors that are most commonly found in the femur, tibia and spinal vertebrae. They mainly occur in pediatric patients, and as such, growth plate involvement and limb-length deformities usually are the primary concerns (Sasaki et al., 2017; Klein et al., 2019; Zhao et al., 2020). Surgery is only required if patients present with signs of root or cord compression. In such cases, total excision is required to achieve total remission. For extensive lesions or patients with spinal instability, additional spinal stabilization is required (Park et al., 2016). Radiation alone may have no benefit due to additional risk of malignant transformation. However, adjuvant radiation may be given to patients with inoperable lesions, hypervascular lesions, aggressive recurrent disease, or high risk surgical candidates.

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