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Case report

Aneurysmal Bone Cyst of D2 in a Child complicated with paraplegia.

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Abstract

Aneurysmal bone cysts (ABCs) are benign osteolytic lesion representing 15% of all primary spine tumors. We report a case of a 9-year-old girl who had an ABCs localized in D2.

Symptoms involved back pain and paraplegia. Radiology investigations showed osteolysis of D2 and anterolisthesis of C7 and D1.

The patient had a posterior decompression and laminectomy of D2, D3 and D4 without neurological improvement. Surgical biopsy confirmed the diagnosis.

Computed tomography scan showed tumor remnants. An embolization of the tumor and an anterior liberation associated with bone graft were performed.

The result was a spectacular neurological improvement with disappearing of all neurological symptoms. Radiology investigations follow up showed only spine instability but no residual tumor.

Key words: Tumor; Cyst; Bone; Spine.

Introduction:

Aneurysmal bone cysts (ABCs) are benign and locally aggressive osteolytic lesions. They represent 1.4% of primary bone tumors; 9.1% of all bone tumors; and only 15% of primary spine tumors [1]. These lesions occur either in thoraco-lumbar or cervical spine. These locations are problematic due to the frequency of spine instability. The reconstructive surgery is always challenging [2].

Case presentation:

Our case is a 9-year-old girl with no previous medical history. She first presented with back pain and progressive paraplegia. There was no history of trauma or fever. Physical examination showed neck stiffness, flaccid paraplegia and abolition of tendon reflexes. There was no sensory neither sphincterian disorders.

The X-ray showed osteolysis of D2 and anterolisthesis of C7 and D1. The CT scan showed an osteolytic lesion in the posterior arch of D2 vertebrae (Figure 1). The lesion was compressing the spinal cord. The MRI showed hypersignal of the vertebrae D2 in T1 and T2 weighted sequences with vascular enhancement and medullary compression (Figure 2).

The patient first had a posterior decompression and laminectomy of D2, D3 and D4, stabilized by a halo cast. A surgical biopsy was also performed, and it was in favor of ABC (Figure 3).

There was no neurological improvement after the surgery. Spasticity of lower appeared one month later. A CT scan showed tumor remnants in the medullar cavity (Figure 4). An embolization of the tumor was performed (figure 5), associated with anterior liberation, bone graft and stabilization with halo cast for three months.

6 months later, the neurological defect has disappeared. The muscular testing was normal.

CT scan follow up showed that there is no recurrence of the tumor (Figure 6). At 9 years after the surgery, the patient is living a normal life and has no complaints. No recurrence of the tumor was observed.

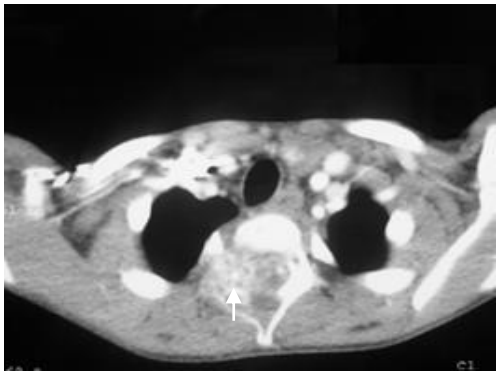
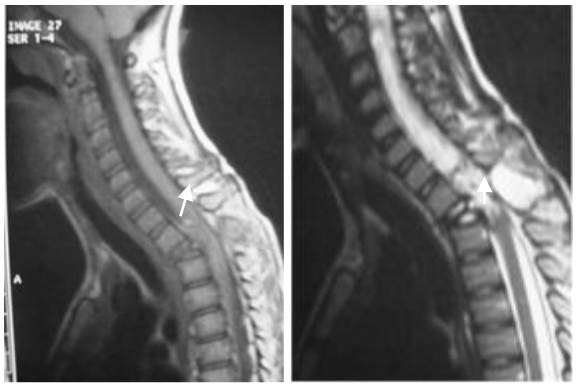
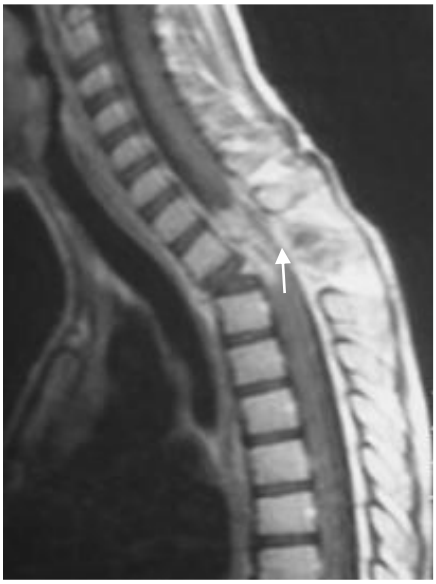


Figure 1: CT axial images show an expansive and lytic lesion in the vertebral body, right pedicle, transverse and spinous process of D2 which enhance after contrast injection with moderate canal compromise



(a)



(b)

Figure 2: a) T1, T2 tumoral appearances b) T2 with injection of gadolinium contrast: hypersignal of the vertebrae D2 in T1 and T2 weighted sequences with vascular enhancement and medullary compression

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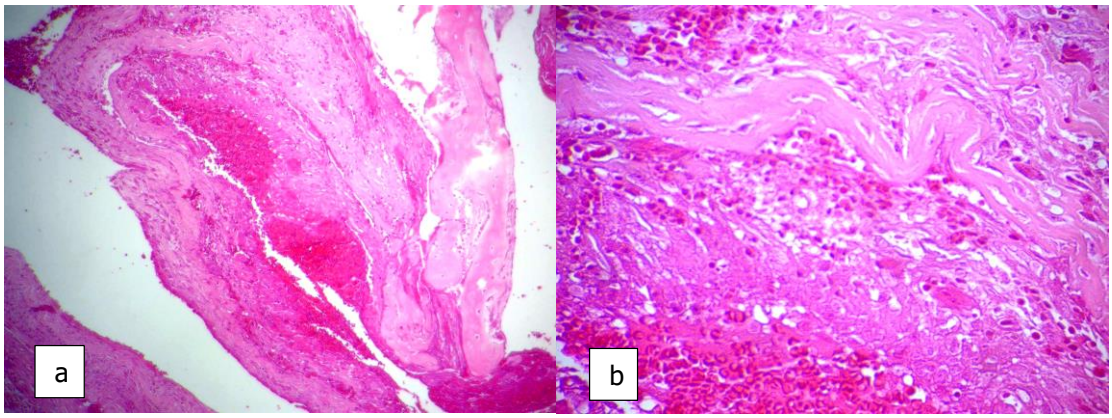

Figure 3: Histological study a: cavities separated by septa of various thickness. These cavities were filled with red blood cells. b: Thin osteoid hyaline bands close of the borders of the cavities

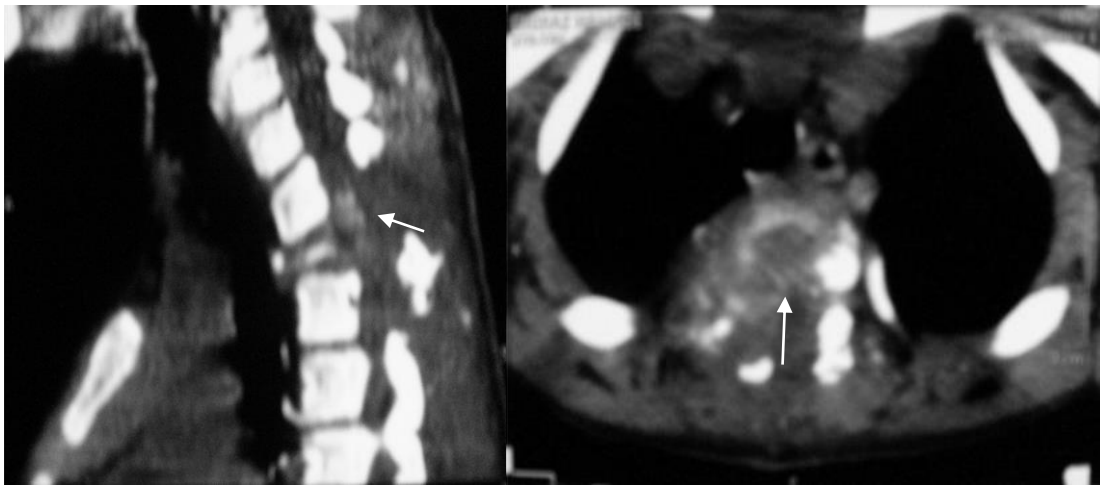

Figure 4: Tumor remnants in the medullar cavity

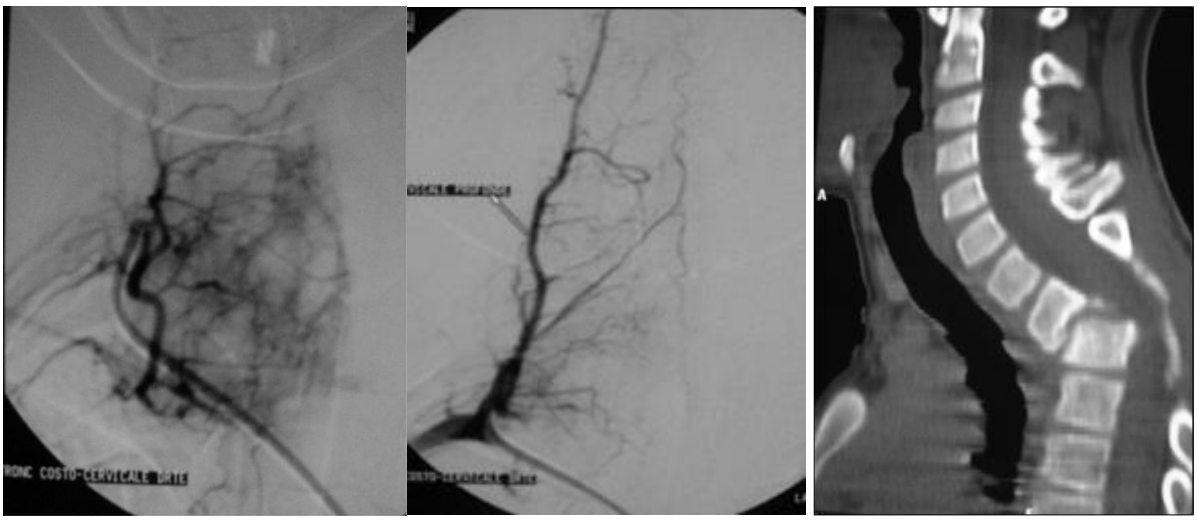

Figure 5: Pre-operative embolization of the tumor

Figure 6: follow up CT scan at 6 months

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Discussion:

ABC was first considered as a variety of giant cell tumors, then as an isolated tumor-like bone dystrophy. But its real nature is still unknown. ABC was first described in 1942 by Jaffe and Lichtenstein [3]. This terminology was, since then, world widely used, even ABCs are neither cysts, nor aneurysms. It is a tumor-like lesion and can be individualized in two forms: primitive ABC, which is an independent entity (70% of the cases), and secondary ABC (30% of the cases), which is a reactional and developed on a preexisting lesion [4].

Pathogenic mechanisms of ABC are still discussable. Recent researches, particularly genetic and immuno-histochemical, are tending to prove that ABC is more a tumor than a tumor-like lesion. However, pathogenesis of ABC is probably multifactorial. In an epidemiological multicentric study about 411 children with primitive ABC, femur (22%), tibia (17%), spine (15%), humerus (10%), pelvis (9%) and fibula (9%) were the most frequent localizations [5]. Lumbar region is the most frequently affected in the spine. ABC is first found in the posterior arc (40% of unique lesions), then fills the vertebral body in the front side via the pedicle, adjacent vertebrae in the up and downside via the articular processes, and the ribs laterally. Isolated localization in the body of the vertebrae is very rare [6]. ABC can present in a form of stiff and painful scoliosis, with an important functional disability. A neurological syndrome is found in 50% of the cases. Neurological signs, like radicular compression, are either progressive due to the growth of the size of the tumor, or brutal due to the damage caused in the vertebral body. X-rays and CT scan findings depend on the stage of ABC. During the osteolytic stage, radiological images are usually zones of eccentric bone depletion. The active growth stage shows a sub-periosteal eruption. Healing stage is characterized by progressive calcification and ossification of the cyst [7].

MRI can localize the lesion and its extension, confirm its sub-periosteal situation and analyze the surrounding vessels and noble structures. Some images are very revealing, like a well-limited expansive bone lesion, a decrease of signal in T1 associated with increase of the signal in T2 (liquid compound), a peripheral border of low signal enhanced by the injection of gadolinium, multiple small cavities confined by septa and the presence of liquid-liquid levels.

Association of X-ray and MRI is helpful for the diagnosis of ABC, but biopsy is mandatory before the treatment for histological confirmation [8].

Selective arterial embolization, used as the only treatment, or during the pre-operative phase (an uncontrollable bleeding in this region can be fatal) is admitted by all the authors. It is widely used when the ABC affects the spine and the pelvis where we can't use a pneumatic tourniquet. Complications as ischemia of neurological structures or other organs are possible [9].

If surgery is indicated, it must fulfill three obligations: the complete excision of the tumor, decompression of the spinal cord and reconstruction and stabilization of the spine. It is essential, especially in this localization, to treat the lesion in only one surgical procedure. further surgeries are challenging and always complicated [10].

Surgical curettage is the most appropriated treatment for ABC of the spine. It consists in accessing the cyst via a window, performing a careful curettage of its cavity and excising its lining. We can combine this technique with bone graft. Most of the recurrences occur during the first months after the treatment (3 to 6 months). There are usually less chances of recurrence in the vertebral localizations [11,12].

Conclusion:

ABCs are benign and rare tumors of the child. A stiff and painful back is the most frequent warning sign. This tumor can be severe when it is localized in the spine because of its neurological risks. Surgical treatment is essential when neurological symptoms are present.

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Conflict of interest: none

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