Reviewer’s report

**Title:** Solid Variant of Aneurysmal Bone Cyst of the Thoracic Spine: Case Report

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**Reviewer:** alejandro reyes-sanchez

Which of the following following best describes what type of case report this is?: Unexpected or unusual presentations of a disease

**Has the case been reported coherently?:** Yes

**Is the case report authentic?:** Yes

**Is the case report ethical?:** Yes

**Is there any missing information that you think must be added before publication?:** No

**Is this case worth reporting?:** Yes

**Is the case report persuasive?:** Yes

**Does the case report have explanatory value?:** Yes

**Does the case report have diagnostic value?:** Yes

**Will the case report make a difference to clinical practice?:** Yes

**Is the anonymity of the patient protected?:** Yes

**Comments to authors:**

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Aneurysmal bone cyst of the spine. Case report
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Introduction
Aneurysmal bone cyst is a fast-growing, expansile and occasionally aggressive benign tumor of undefined neoplastic nature. It comprises 1.4% of all bony tumors and 14 to 20% of all tumors that involve the vertebral column. Treatment of choice is complete surgical resection of the cyst, although with hypervascularized tumors there is risk of excessive bleeding. Occasionally, a problem is encountered in obtaining adequate stabilization of the spine. We present a case of aneurysmal bone cyst associated with severe neurological lesion, treated surgically.

Abstract
Aneurysmal bone cyst (ABC) is a fast-growing tumor of undefined neoplastic nature. It is occasionally an aggressive benign lesion whose treatment of choice is complete resection, even though the risk of profuse transoperative bleeding exists. We present a female patient with thoracic spine deformity with progressive paresthesias and muscle weakness of lower extremities that evolved to paralysis of both lower extremities and sphincter incontinence. Based on radiographic films, lytic lesions were identified at T7-T9 vertebrae as well as medullary space invasion. In electrophysiological tests, a complete somatosensorial pathway block was reported. Prior to resection of the neoplastic lesion and thoracolumbar stabilization, an incisional biopsy was performed. There was no postoperative medullary functional improvement. Morphological findings corresponded to an aneurysmal bone cyst at
T8. This lesion is mainly located in the long bones and less frequently of the spine, where instability and medullary compression may occur. It is possible to confuse this neoplasia with other lesions. Hence, definitive diagnosis with biopsy is necessary for determining an adequate therapeutic plan to eradicate risk of recurrence or associated neurologic sequelae, as well as to gain proper stability at the involved vertebral segments.

Key words: spine, aneurysmal bone cyst, spine instability.

Resumen

El quiste óseo aneurismático es un tumor de naturaleza neoplásica indefinida, de comportamiento benigno, crecimiento rápido y ocasionalmente de comportamiento agresivo, cuyo tratamiento de elección es la resección completa, aunque existe el riesgo de sangrado transquirúrgico excesivo. Se presenta el caso de una paciente con deformidad en columna torácica, con parestesias y debilidad muscular progresivas en extremidades inferiores, que evolucionó hasta la parálisis de dichas extremidades e incontinencia de ambos esfínteres. Mediante estudios de gabinete se localizaron lesiones líticas en cuerpos vetebrales T7 a T9 e invasión a conducto raquídeo. Los estudios electrofisiológicos identificaron bloqueo completo de la vía somatosensorial. Previo biopsia incisional, se realizó resección de la lesión y estabilización de la columna toracolumbar. La paciente evolucionó sin mejoría de la función medular. Los hallazgos morfológicos correspondieron a quiste óseo aneurismático en T8. Esta lesión se localiza principalmente en huesos largos y con mucho menor frecuencia en la columna vertebral, donde puede provocar inestabilidad y compresión de la médula espinal. Es posible confundirla con otras neoplasias, por lo que el diagnóstico definitivo mediante biopsia es imprescindible a fin de establecer el plan terapéutico adecuado, que elimine el riesgo de recurrencia o secuelas neurológicas asociadas, y lograr la estabilidad adecuada de los segmentos vertebrales afectados.
Clinical Case

We present the case of a 14-year-old female with an illness of 7 months duration prior to hospital admission. The patient first noticed a deformity in the posterior region of the thorax accompanied by slight pain; later parasthesias and muscular weakness of the extremities was noted, initially on the right side and later bilaterally, which caused frequent falls. A month before admittance, the patient presented paralysis of the extremities and incontinence of both sphincters.

At the time of hospitalization, the patient showed increase of thoracic kyphosis, pain upon palpation of the superior thoracic region in the spinous apophyses of T7-T9, and one sacral lesion. On neurological exploration, anesthesia of the distal dermatomes from T10 was found with paralysis of the extremities, absent patellar and Achilles reflexes, clonus and negative Babinski.

Radiographs showed increased thoracic kyphosis with collapse of the vertebral body of T8, slight scoliosis and retrolisthesis of T7. On computerized axial tomography, expansive lytic lesions affecting the vertebral bodies and the posterior arches of T7-T9, with tumor invasion to the rachidian conduit were appreciated (Figure 1).

In addition to the mentioned alterations, magnetic resonance showed a tumor that affected the posterior arches of T7-T9 and compression of the spinal marrow, to the level of the posteroinferior edge of T7 (Figure 2).

Evoked potentials indicated complete blockage of the somatosensory route. Incisional biopsy was performed that showed aneurysmal bone cyst and it was decided to perform surgical resection by means of double approach of the thoracic spine.

Figure 3. Anteroposterior radiographs (left) and lateral (right) of thoracolumbar column (postoperative) showing adequate
alignment of the thoracic column after the instrumentation with a universal system.

Figure 2. Magnetic resonance of thoracic column in T1 (left) and T2 (right). In addition to the kyphotic deformity, complete destruction of the vertebral body of T7 can be observed, involvement of the posterior arches and the rachidian conduit, as well as severe compression of the spinal marrow.

Figure 1. Computerized axial tomography of T6 showing involvement of the posterior arch with tumor invasion of the rachidian conduit in both cuts.

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For the first time, anterior approach was performed for marrow decompression, tumor resection, placement of intersomatic bone graft, and screw and rod fixation of T7-T9. Ten days later, during a second surgical event, posterior approach for tumor resection was performed, freeing marrow and fixation with universal type instrumentation of T2-L1 (Figure 3). The patient evolved during the postoperative period with left pleural effusion that responded to medical treatment with diuretics and respiratory therapy. Postoperatively, there were no signs of recovery of marrow function.

Surgical biopsy of the eighth thoracic vertebra consisted of multiple fragments. Macroscopically, it was irregular, light brown tissue of medium consistency that conformed 2 cm³ of rough surface, with firm solid areas. Pathological results determined aneurysmal bone cyst. There was a second piece of 10 cm³ of surgically resected tissue. Macroscopically it consisted of multiple fragments of reddish-brown color with a fleshy appearance and cystic areas of hematic content limited by fibrous tissue septa and bony trabecular tissue.

Morphological findings in both histopathological studies corresponded to benign cystic lesion, constituted by cystic spaces filled with blood with fibrous tissue walls, delimited by flat
cells (Figure 4) and stroma composed of fusiform cells of fibroblastic collagen, small blood vessels, giant multinucleated osteoclast-type cells, and hemosiderin (Figure 5). Some areas predominated with vascular spaces without endothelial coating and zones in which reactive bone tissue was identified. The established diagnosis corresponded to aneurysmal bone cyst of T8.

Discussion
Aneurysmal bone cyst is a benign tumor of undefined neoplastic nature1-4 that presents a pattern of rapid, expansive growth, sometimes erosive. It generally affects persons during the second decade of life.5,6 There is no gender bias, but some studies report a greater frequency in females (2:1).7 Sites of distribution of this lesion are generally the femur, humerus, tibia, sacrum, pubis and vertebral column.8-12 In the metaphyseal region of the long bones, it presents more frequently in the distal region of the femur and proximal region of the tibia. It is less common in the vertebral column, affecting primarily the pedicles and spinous apophysis, frequently producing a paravertebral or mediastinal mass effect,13,14 according to its localization.14,15 It constitutes ~14% of the total of bone tumors and 14 to 20% of the tumors that involve the vertebral column,16 appearing more commonly in thoracic vertebrae and sometimes in cervical vertebrae.17

Almost 30% of aneurysmal bone cysts of the column present in pediatric patients causing instability of the vertebral column and compression of the spine marrow.5,8,18,19 Recently, familial type incidence has been established and in cytogenetic studies alteration of the short arm of chromosome 17 has been identified, with translocation in the long arm of chromosome 16, suggesting a hereditary component in the etiology of this entity.20

Sometimes cystic growth is rapid, which provokes the appearance of a paravertebral mass that can cause irreversible damage to the spine marrow,13 as happened in the present case. Currently, study of aneurysmal bone cyst is facilitated with
diagnostic studies (computerized axial tomography and magnetic resonance),21 but given that it can be confused with other types of lesions such as osteosarcoma or giant cell tumors, biopsy of the lesion is necessary to obtain definitive diagnosis.

Figure 4. Cystic injury constituted by fibrous tissue walls that delimit hematic content spaces.

Figure 5. Fibrous stroma constituted by fibroblasts, small blood vessels, giant multinucleated osteoclast-type cells and hemosiderophages.

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Macroscopically, the aneurysmal bone cyst is a lesion of variable size, delicately delimited by a fine bony tissue layer, although sometimes it can show a pattern infiltrative to the soft tissues, similar to a honeycomb or great blood-filled cavernous vascular spaces.

Microscopically, the lesion has a variable morphology in which vascular spaces can be observed that lack a smooth muscle wall and endothelial coating. Differential diagnosis is necessary to rule out other primary lesions or aneurysmal bone cyst secondary to another neoplasia, for example, telangiectactic osteosarcoma. These two entities can be clinically and radiologically confused. However, microscopically, morphologic diagnosis establishes the difference with a basis in the presence of sarcomatous, anaplastic cells with high nuclear pleomorphism in the osteosarcoma.1

Treatment by means of embolization and curettage has the risk of recurrence, and although curettage and radiotherapy have demonstrated effectiveness, they can cause vertebral deformity. Before neurological compromise, treatment of choice is decompression, complete resection of the lesion, and placement of bone graft for reconstruction of the vertebral column18,22,23 as well as instrumentation of the affected segment to obtain suitable alignment and stability of column.24 Radiotherapy should be considered when resection of the injury is not posible.25

Conclusions
It is important to establish the differential diagnosis in benign aggressive lesions with others of a malignant nature, favoring suitable and opportune treatment. In the present case, the patient received medical attention at a late stage of the disease and although surgical treatment corrected the deformity and eliminated pain, postoperatively there was no neurological recovery due to marrow damage. For this reason, it is important to consider that in patients with neurological symptomatology surgical treatment must be performed in a timely manner before permanent neurological damage is established.

References
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