

Aneurysmal Bone Cyst: Rapid Deterioration in a Long-Standing Tumor

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Aneurysmal bone cyst, first described by Jaffe and Lichtenstein¹ in 1942 as a rare, localized tumor of the long bones and spinal vertebrae, characterized by the "blowout distention" of the skeletal contour on a radiograph.² This neoplasm, usually found in children and young adults, constitutes 1.4 percent of all primary bone tumors.³ The cyst is occasionally associated with other tumors of the bone, such as osteoblastoma, osteosarcoma, giant cell tumor, and fibrous dysplasia,⁴ and has occasionally been reported to occur at a site of previous trauma to the bone.⁵ There is a tendency toward rapid growth with local expansion, which can exert pressure on an adjacent structure.³

Histologically, aneurysmal bone cyst is a cystic, osteolytic vascular tumor, replete with giant cells and fibrous septa. The tumor has a solid variant with fibroblastic proliferation, giant cell areas, stromal hemorrhage, and new bone production.⁶

When an aneurysmal bone cyst appears in the spine, it is generally diagnosed within a few months after the onset of clinical symptoms, which include local pain and swelling accompanied by reactive local muscle stiffness and restriction of movement. Only in advanced and rare cases does the aneurysmal bone cyst exert pressure on the spinal cord, which can lead to neurologic deficits.^{6,7} We describe the diagnosis, management, and follow-up of an aneurysmal bone cyst in a 14-year-old girl.

Case Report

A 14-year-old girl complained of a 2-year history of recurrent episodes of nonradiating low-back pain, which lasted for several days and was relieved with nonsteroidal anti-inflammatory drugs. Two weeks before she was admitted to the hospi-

tal, the girl had begun to experience worsening of the occurrences.

During the previous 2-year period, she had been examined several times by a pediatrician and orthopedist, but no history or physical finding could elucidate her pain. During these examinations a routine complete blood count, blood chemistry analysis, and urine analysis yielded normal values. Tests for antinuclear antibodies and rheumatoid factors, which can appear in juvenile rheumatoid arthritis and systemic lupus erythematosus, were negative. No abnormalities were found on an abdominal radiograph and sonogram, except for a small stone in the left kidney with mild widening of the calyces. Renal colic was suspected. Her blood sedimentation rate was 60 mm in the first hour, but no satisfactory explanation was provided.

When she was first examined in the emergency department, the girl appeared to be well developed and mildly obese, with no signs of distress. She complained of pain and tenderness in the lumbar spine region. Findings of a complete physical examination were unremarkable. The lumbar spine was found to be mildly tender without spasm or fullness. Muscle strength, sensation, and deep tendon reflexes of the lower extremities were normal. Rectal and genital examinations revealed no abnormalities. Complete blood count was normal. Laboratory results were normal, except for a high sedimentation rate. The pain was spontaneously relieved, and she was sent home.

One week later her condition was rapidly deteriorating. She complained of diurnal enuresis, and on examination there was increased weakness in both legs, especially in the proximal muscles, an increase in the patellar and Achilles tendon reflexes, positive bilateral Babinski reflexes, and footdrop in both legs.

An abdominal radiograph showed mild scoliosis in the lumbar region of the spine, but no other pathologic signs. Spinal computed tomography (CT) showed scoliosis to the left and a lytic lesion in the posterior elements of the vertebrae at the

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Figure 1. Computed tomographic scan showing a lytic lesion in the posterior elements of the vertebrae at the D10-D12 level, with expansion to the vertebral body from the left. This process with a thin periosteal border enters the spinal canal, pressing the cord forward and to the right.

level of D10-D12, with expansion to the vertebral body from the left. This process, which had a thin periosteal border, entered the spinal canal, pressing the cord forward and to the right (Figure 1). Magnetic resonance imaging (MRI) showed a space-occupying lesion at the level of D10-D12, extradurally pressing the spinal cord forward and to the right and destroying the pedicle and the lamina. On MRI with gadolinium injection, a nonhomogeneous multilobular process was seen (Figure 2). The tentative diagnosis was an aneurysmal bone cyst. During surgical exploration of the spine at the level of D10-D12, a gray-red mass 6 cm long was found invading the vertebrae through the left lamina to a depth of 2 cm from D10 and descending into the epidural space to D12. The tumor seemed to be epidural, attached to the dura, but easily separated from it. Cauterization and curettage of the cyst by way of a laminectomy were performed.

The patient's postoperative follow-up course was uneventful, and she rapidly regained strength in

both legs. A histologic examination of the cyst showed it to be an osteolytic lesion replete with giant cells and fibrous septa, with a few solid areas of fibroblasts (Figure 3).

At a 1-year follow-up examination the girl had resumed full daily activities; was pain-free; had normal muscle strength, sensitivity, and reflexes; and had no urinary incontinence.

Discussion

Primary tumors of the vertebrae are rare in infancy and childhood. An aneurysmal bone cyst is a rare, expanding benign lesion that appears most commonly in children and young adults, but can occur later in life.³

It has been suggested that aneurysmal bone cyst is not a true neoplasm, but rather a vascular malformation. It probably begins with an arteriovenous fistula inside the bone, expands into the medullar space, and forms a cyst with blood-filled spaces surrounded by a thin shell of periosteum.^{2,5} When the cyst appears in the spine, the neural arch is the

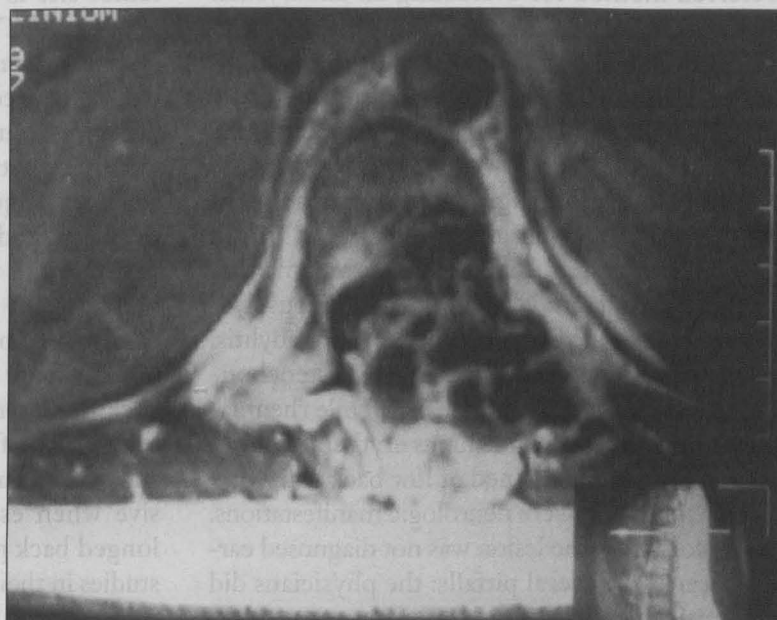


Figure 2. Magnetic resonance imaging after injection with gadolinium shows a nonhomogeneous multilobular lesion at D10-D12 level, extradurally pressing the spinal cord forward and to the right, destroying the pedicle and the lamina of the vertebra.

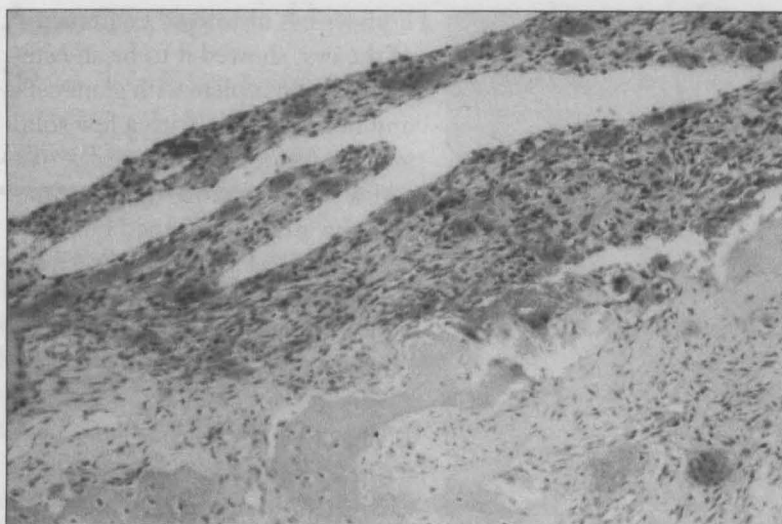


Figure 3. Histologic sample of a cystic osteolytic lesion replete with giant cells, fibrous septa, and a few solid areas of fibroblasts.

most frequently involved portion of the vertebrae, although other parts of the body can occasionally be affected. Clinically the patient usually has pain and stiffening in the affected portion of the spine with restricted motion. A palpable mass appears in only 6.0 percent of cases.¹ The lesion can enlarge rapidly within a few months and cause extensive bone destruction. On radiographic examination, the lesion usually appears as an expanding, lytic process, surrounded by a thin cortical shell.^{2,8} The preferred method for evaluating an aneurysmal bone cyst, after conventional radiography, is an MRI, which will provide as much or more information than a CT.⁵ In our case, findings from both CT and MRI were suggestive of aneurysmal bone cyst.

The differential diagnosis of bony causes of back pain in children are other tumors of the bone, such as osteblastoma, osteosarcoma, osteoid osteoma, giant cell tumor, and fibrous dysplasia. Nonneoplastic causes comprise spondylitis, osteomyelitis, Scheuermann disease (osteochondritis of the vertebral epiphyses), juvenile rheumatoid arthritis, and systemic lupus erythematosus.

Our patient complained of low back pain for 2 years before there were neurologic manifestations. In all probability the lesion was not diagnosed earlier because of several pitfalls: the physicians did not suspect a skeletal neoplasm; there were no external findings on physical examination; the girl was at an age at which low back pain, because of rapid growth, is common; and the pain was not progressive, but intermittent and relieved by mild

analgesia. Moreover, the abdominal radiograph did not indicate any pathologic finding because it was of an area below the level of the lesion, and the left nephrolithiasis found on a sonogram was misleading as a source of the pain. In retrospect, the physicians should have had a higher index of suspicion for bony lesion, and earlier thorough examinations, including CT or MRI, should have been performed.

In our case, prompt curettage of the bone cyst was successful, and there was no need for bone grafting. When possible, this method of treatment is preferred. Even either incomplete curettage or open

biopsy can result in obliteration of the cyst. Radiotherapy has also been used effectively in patients who are at high risk for surgery or who are resistant to surgical treatment, although the possibility of grave complications, such as myelopathy, sarcomatous changes, and deformation of the vertebrae, make this mode of treatment undesirable. In large tumors that have a high risk of bleeding and in places where curettage would be difficult, such as the pelvis or spine, selective embolization of the tumor site is possible, followed by curettage, if necessary.⁹ The prognosis of aneurysmal bone cyst in children is most promising, although there is a high percentage of recurrence, mainly in the first year after the operation and in patients who undergo partial resection of the tumor. These children require more careful follow-up and might need additional treatment.⁷

Conclusion

Aneurysmal bone cyst is a rare skeletal neoplasm, but it is only one of the various causes for low-back pain that is a common complaint in the adolescent. The family physician and the pediatrician should be aware of these entities, be more aggressive when establishing their diagnosis of prolonged back pain, and include advanced imaging studies in their workup.

References

1. Fry VG, Van Dellen JR. Aneurysmal bone cyst of the spine. *S Afr Med J* 1980;258:211-3.
2. McArthur RA, Fisher RG. Aneurysmal bone cyst in-

- volving the vertebral column. A case report. *J Neurosurg* 1966;24:772-6.
3. McLurin RL. Extramedullary spinal tumors. In: *Pediatric neurosurgery: surgery of the developing nervous system*. American Association of Neurological Surgeons, Section of Pediatric Neurosurgery. New York: Grune & Stratton, 1982:541-9.
 4. Mintz MC, Dalinka MK, Schmidt R. Aneurysmal bone cyst arising in fibrous dysplasia during pregnancy. *Radiology* 1987;165:549-50.
 5. Miron D, Mogilner G, Horowitz J, Moreli D, Siplovitz L. [Aneurysmal bone cyst of a lumbar vertebra in a child.] *Harefuah* 1993;124:82-5.
 6. Oda Y, Tsuneyoshi M, Shinohara N. "Solid" variant of aneurysmal bone cyst (extragnathic giant cell reparative granuloma) in the axial skeleton and long bones. A study of its morphologic spectrum and distinction from allied giant cell lesions. *Cancer* 1992;70:2642-9.
 7. MacDonald P, Letts M, Sutherland G, Unruh H. Aneurysmal bone cyst of the upper thoracic spine. An operative approach through a manubrial sternotomy. *Clin Orthop* 1990;June:127-32.
 8. Goodman ML. Aneurysmal bone cyst in early childhood. *Orthop Rev* 1986;15:457-60.
 9. DeRosa GP, Graziano GP, Scott J. Arterial embolization of aneurysmal bone cyst of the lumbar spine. A report of two cases. *J Bone Joint Surg Am* 1990;71-A:777-80.