Aneurysmal Bone Cyst of the Thoracic Spine: Evolution after Particulate Embolization

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Summary: A case of aneurysmal bone cyst of the thoracic spine treated with serial particulate embolization responded with involution of soft-tissue and cystic components and diffuse ossification of the mass. Reappearance of foci of bony rarefaction or cystic change at 4-year follow-up indicates the need for continued surveillance.

Index terms: Spine, abnormalities and anomalies; Interventional neuroradiology

Aneurysmal bone cysts account for about 1.4% of all bone tumors, and approximately 14% to 20% of these involve the spine (1, 2). The treatment of choice for aneurysmal bone cysts has been complete surgical resection, but in selected cases the risks of unacceptable surgical morbidity and excessive hemorrhage in hypervascular tumors and the challenge of maintaining spinal stability are indications for adjunctive or alternative therapy. We describe a case of thoracic spinal aneurysmal bone cyst treated effectively with serial particulate embolization.

Case Report

A 15-year-old girl was first seen after several months of right upper back pain. Imaging studies showed a large paraspinal and posterior mediastinal mass. Subtotal resection via right thoracotomy was performed in Mexico. Pathology reports were consistent with aneurysmal bone cyst. The patient was seen at our institution 6 months later. Neurologic examination was remarkable for decreased light touch and pinprick sensation over the right upper back and axilla in the T-2 dermatome region. Motor strength was intact in all extremities, and bowel and bladder function were normal.

Chest radiography showed an 8-cm right apical paraspinal mass with rim calcification. There was destruction of the right second and third ribs and lytic changes in the T-2 and T-3 vertebrae. Computed tomography (CT) and magnetic resonance (MR) imaging of the spine (Fig 1) showed epidural extension of the mass and involvement of both T-2 and T-3 vertebral bodies, posterior elements, and the second and third ribs. CT-guided needle biopsy obtained only a bloody aspirate, and open biopsy obtained tissue confirming the diagnosis of aneurysmal bone cyst.

Treatment options were discussed among various consultants and presented to the patient and family. Embolization therapy with possible surgery to follow was elected. Spinal angiography (Fig 2) was performed and showed the hypervascular mass supplied by branches of the right thyrocervical trunk, right bronchial artery, and right intercostal-bronchial trunk. Four staged selective embolizations (Fig 2) with 250 to 350 μm of polyvinyl alcohol sponge particles were accomplished at 1-month intervals. For each vessel, embolization was performed nearly to the point of stasis. Delivery systems included 5F catheters alone or with a coaxial Tracker-18 Hi Flow (Target Therapeutics, San Jose, Calif). Coils were not used in order to permit repeat embolizations as needed. In total, thyrocervical trunk branches were embolized on all four occasions, the right bronchial artery three times, and the right intercostal-bronchial trunk twice.

CT 6 months after the initial embolization (3 months after the last session) showed extensive decrease in soft-tissue mass and development of ossification (Fig 3); CT findings 2 years later were unchanged. At 4 years, the patient remained symptom free and the neurologic examination was completely normal. Follow-up CT and MR showed sclerotic changes in the vertebral bodies reflected as decreased T2 signal on MR (Fig 4A) and the development of cystic areas within the ossified mass on CT (Fig 4B).

Discussion

Aneurysmal bone cyst is a nonneoplastic expansile primary bone lesion that is uncommon in the spine. The long bones and flat bones (particularly the pelvis) are involved more often (2). Vertebral lesions tend to begin in the pos-
terior elements and may spread through the pedicle into the vertebral body and epidural space (3). In the spine, involvement of two adjacent vertebrae (as in this case) can be an important clue to diagnosis (2).

Treatment options include surgical curettage, complete wide resection (with or without bone grafting), radiation therapy, hyperthermia, cryotherapy, or vascular occlusion. The treatment most likely to effect a cure is a total wide resection; however, morbidity and concerns for structural integrity have led to a search for less radical surgical techniques (4). Curettage has an approximately 19% recurrence rate, usually in the first 2 years (2). In the long bones, recurrence after curettage and bone grafting can be as high as 54% (4). Regression of aneurysmal bone cysts has been reported after open biopsy and diagnostic angiography (5). Successful treatment by radiation therapy alone has been reported (6), but it is usually used in combination with surgery or in cases of persistent postoperative recurrence. Radiotherapy to the spine can be complicated by myelopathy (2.9%) (3) and carries a small risk of postirradiation sarcoma (2).

Arterial embolization has been effective either as an adjunct to surgery or as sole therapy. Numerous successful cases have been reported, particularly in the pelvis and long bones (7–14). In the spine, arterial embolization was first used preoperatively to decrease vascularity and intraoperative hemorrhage (15, 16). Several cases of successful embolization of spinal aneurysmal bone cysts have been reported in the orthopedic literature (17–19). The response of aneurysmal bone cysts to embolization has been involution of the soft-tissue component, sclerosis, and ossification. This mineralization becomes apparent 2 or more months after embolization (7, 17). In some of these cases, the involution of mass and ossification was so profound that surgery was avoided altogether.

Arterial embolization in the treatment of aneurysmal bone cysts is indicated when surgical risk or morbidity is unacceptable, particularly when spinal stability or neural elements might be jeopardized. Technically, we chose to perform serial particulate embolizations and did not use occlusive coils. Four staged embolizations were performed because of a concern that embolization might be associated with tumor necrosis, bleeding, or swelling. In view of the large tumor mass adjacent to the spinal cord, any swelling might have been associated with cord compression. The choice of medium-sized (250 to 350 μm) polyvinyl alcohol sponge particles was based on the concern that smaller particles might behave more like liquid agents and could increase the risk of complications such as ischemia to normal tissue, especially nerves. One might argue that smaller particles would pene-
trate the tumor bed more deeply, which could have reduced the number of treatment sessions. Because hypervascularity seemed recurrent even in vessels thoroughly embolized, the absence of coils allowed continued access. One could argue that coils would offer a more lasting occlusion, and coils are probably appropriate if surgery is to be performed soon after the embolization. Previous authors (17) have suggested monitoring with somatosensory evoked potentials and selective sodium amytal injection to improve the margin of safety during spinal embolization.

Ideally, angiography should be done to establish the normal vascular supply to the anterior spinal axis before embolization in that region. Although we did not completely map the normal spinal cord circulation in this case, we frequently restudied each embolized pedicle angiographically as the embolization progressed, recognizing the potential for flow dynamics to change during embolization.

Finally, in our case, despite the development of diffuse ossification, the reappearance of foci...
of bony rarefaction or cystic areas (Fig 4B) after 4 years suggests that caution should be exercised in declaring the lesion cured, and continued surveillance (beyond the 2-year follow-up indicated in most case reports) is prudent.

References