Serial endovascular embolization as stand-alone treatment of a sacral aneurysmal bone cyst

Case report

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Aneurysmal bone cysts (ABCs) are destructive cystic lesions of the bone and are common in children. They are expansile in nature and, therefore, may become symptomatic. These have traditionally been treated surgically; but recently, endovascular embolization has shown promise as a stand-alone therapy. The authors describe a case of an ABC highlighting the effectiveness and efficiency of endovascular treatment. A 16-year-old boy was referred for a 4-month history of radiating back pain and urinary hesitancy. Findings from his neurological examination were normal, but he had problems ambulating because of pain. Magnetic resonance imaging and CT scanning showed a cystic mass in the sacrum; a biopsy was performed and diagnosis of ABC was confirmed. Treatment options were then discussed with the family.

The patient underwent 2 endovascular embolizations in approximately 1 month: Onyx 18 was involved in the first session, and N-butyl cyanoacrylate glue was used in the second session. After the first treatment, the patient experienced a dramatic decrease in pain and concomitant improvement in function. The patient went from being mildly symptomatic after the first treatment to completely asymptomatic after the second treatment. Clinical and radiographic follow-up obtained at 2, 6, and 18 months after initial treatment revealed the patient to be asymptomatic with progressive ossification.

Endovascular treatment can be effective in treating symptomatic cases of ABC in which surgery would carry significant risk. Selective arterial embolization can promote sclerosis and result in an immediate and significant decrease in pain.

(https://thejns.org/doi/abs/10.3171/2013.11.SPINE13412)

Key Words • aneurysmal bone cyst • embolization • sacrum • oncology

Aneurysmal bone cysts (ABCs) are destructive cystic lesions of the bone that are common in children in the 2nd decade of life. Approximately 11% of ABCs may be vertebral, and, as a whole, account for 1%–6% of all primary bone tumors. They are considered benign and are described as, “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.” Their expansile nature can cause pain through pathologic fracture or by neurological compromise. They may occur de novo, but as many as 25% may be secondary to another bone tumor. Although some ABCs can be managed conservatively, many will need treatment. Surgical treatment is the standard but can carry significant risk in cases in which the lesion is difficult to access. Most spine surgeons advocate a gross-total, or en bloc, resection. Adjuvant therapy with phenol or liquid nitrogen may be used to avoid recurrence. Selective arterial embolization (SAE) has been used more recently, but little is known of its efficacy because of the relatively few number of patients treated.

Case Report

This 16-year-old Caucasian boy was referred to our clinic in November 2011 for sacral and leg pain. Over 4 months, he described worsening pain in the sacral region, radiating into the back of his left leg, which prevented him from sitting for prolonged periods or from walking more than 100 ft at a time, making it impossible to attend school. He also experienced occasional urinary hesitancy but no sexual dysfunction. His neurological examination was nonfocal. The initial visual analog scale (VAS) score
was 1 for back pain and 7 for leg pain. His Oswestry Disability Index (ODI) was measured to be severely disabling (54%). A lumbosacral MRI study with and without contrast showed a cystic lesion containing fluid levels in the left sacrum. There was no enhancement. Computed tomography scanning confirmed the presence of the cystic lesion, destructive in nature, with involvement of the S-3 and S-4 segments (Fig. 1). There were thought to be some atypical features to the lesion, so needle biopsy was undertaken to rule out malignancy and confirmed the diagnosis of ABC. Treatment options were then discussed with the family.

In this case, surgical treatment carried considerable morbidity due to the lesion's location in the sacrum. A review of the literature revealed a recent case series from Italy that showed significant benefit and curative treatment with endovascular embolization. In December the family and patient agreed to this option and the patient was scheduled for embolization in January 2012. Angiography revealed 3 prominent feeders to the tumor. The first was a branch of the left iliolumbar artery and the second was a distal branch of the left internal iliac artery. Both were successfully embolized with Onyx 18 (Covidien), paying special attention to avoid embolization of vasculature to the skin. The third feeder was left alone with the intent to reevaluate and treat it in another session (Fig. 2A–C). The patient reported significant improvement in pain within 2 days of the first embolization. He returned to clinic a few weeks later and reported significant improvement in pain and urinary hesitancy, although he still did not have much stamina in standing for prolonged periods. The decision was made to pursue second-stage embolization 1 month later. In February 2012, angiography showed that the third feeder to the rostral part of the cyst was unchanged from the prior study. It was a dorsal branch from the internal iliac artery proximal to the prior two feeders. N-butyl cyanoacrylate (NBCA) was used in this session (Fig. 2D and E). The patient returned to the clinic in March; he was asymptomatic and was asking to advance to more rigorous activity. The VAS score and ODI were both 0. Additional CT scanning 2 months after the initial treatment revealed sclerotic bone formation along the margins, which at 6 months continued to show impressive progressive sclerosis and ossification of the mass (Fig. 3). The ODI and VAS scores remained 0, and the patient had begun a part-time job as an emergency medical technician. Follow-up CT scanning at 18 months demonstrated even further progression of sclerosis and ossification within the sacral mass (Fig. 4). Clinically, the patient had resumed athletic activities within 3 months after treatment and continued to function without limitation or pain.

**Discussion**

This case illustrates a new approach for our team in the treatment of ABCs. Although the etiology of these lesions is uncertain, given their vascularity, it is logical that SAE might have a beneficial effect as a primary treatment. In the past, it has been used as a preoperative treatment. With the advances of endovascular technique and improved imaging, patients with ABCs have begun to be treated exclusively by these means (Table 1), most prominently in Italy. In 1992 De Cristofaro et al. published a case series of 24 patients, 19 with ABCs and 5 with angiomas, 17 of whom had resolution of their symptoms with SAE. Recurrence developed in 2 patients; 1 of these recurrences resolved with re-treatment. The authors also went on to state that this treatment may be preferential in the spine, sacrum, and pelvis, given the operative risks.

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**Fig. 1.** Initial CT images (A–C) reveal an expansile sacral mass with cortical destruction and absent calcified tumor matrix. Magnetic resonance images (D and E) reveal a lobulated mass with cystic spaces containing fluid-fluid levels representing blood products. There is involvement of the sacral spinal canal and extension both ventral and dorsal to the sacrum.
These authors’ latest series is the largest case study to date, with 36 patients with ABCs; 6 of the lesions were located in the spine and 5 were specifically located in the sacrum. They had a 94% success rate and 3 complications of skin necrosis and transient paresis.

Endovascular treatment for ABCs has been performed in the US before. Mohit et al. reported a case involving the craniocervical junction that, again, was high risk given its location, but was successfully treated with endovascular therapy. As with our case, surgical treatment was deemed to carry a significant risk of morbidity given the location and, therefore, embolization was pursued. The lesion was successfully treated with NBCA, and the authors were able to eliminate pain and reform bone.

Some authors have proposed certain exclusion criteria that must be met before pursuing embolization as a primary treatment; these include structural instability,
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pathological or impending fracture, neurological symptoms, and technical feasibility. In our case, there was considerable bone involvement, and a case for impending fracture could be made. Neurological involvement, radiculopathy, and urinary symptoms were all relieved. In the series of De Cristofaro et al., 11 patients had almost complete ossification at 22 months. In our patient, CT scans were obtained approximately 4 months apart and demonstrated how quickly a young patient can recover. Perhaps most importantly, this case demonstrates that arterial embolization does not deter new bone growth.

Intralesional curettage, widely considered the mainstay of surgical treatment, carries significant risk and may even require aborting the procedure because of blood loss. Attempts at removing tumor and sparing nerve roots might have been possible but are technically challenging. In this instance, the risk of CSF leakage and subsequent fistula cannot be ignored. Recurrence rates with intralesional curettage range from 10% to 25%. An en bloc resection would have entailed a total or hemisacrectomy with root sacrifice from S-1 and below. This might have led to compromised bowel and bladder function, even with unilateral root sacrifice. Given the size of this large sacral lesion, sacropelvic instability is another concern with resection. Sacropelvic fixation would have been required, perhaps leading to additional operative morbidity.

Our case also exemplifies the cost-to-benefit ratio of this procedure. This patient was diagnosed and treated very efficiently. He was hospitalized for 2 days total and was able to maintain his daily activities and school work. We therefore feel that arterial embolization of spinal column aneurysmal bone cysts is a safe and effective treatment strategy for lesions in which resection carries significant morbidity. This case documents long-term bone remodeling, coinciding with reduction in the patient’s axial and radicular pain. The early radiographic response suggests local control, although long-term follow-up is necessary to establish absence of recurrence.

Acknowledgment

We wish to thank Andrew J. Gienapp for his editorial and publication assistance.

Disclosure

No sources of support were received for this report. Dr. Arthur has served as a consultant for Terumo/MicroVention, Johnson & Johnson, Covidien/eV3, and Stryker. Drs. Doss, Weaver, and Didier have no financial conflicts of interest to disclose.

Author contributions to the study and manuscript preparation include the following. Conception and design: Arthur, Weaver. Acquisition of data: Doss. Analysis and interpretation of data: all authors. Drafting the article: Arthur, Doss. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Arthur. Study supervision: Arthur.

References


TABLE 1: Studies of endovascular treatment for ABCs

<table>
<thead>
<tr>
<th>Authors &amp; Year</th>
<th>No. of Patients</th>
<th>Location</th>
<th>Outcome</th>
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<tbody>
<tr>
<td>Rossi et al., 2010</td>
<td>36</td>
<td>mixed (5 sacral)</td>
<td>94% success at 5 yrs w/ NBCA</td>
</tr>
<tr>
<td>Rossi et al., 2012</td>
<td>1</td>
<td>pelvis</td>
<td>3 treatments w/ NBCA; complete ossification &amp; symptom free at 6 yrs</td>
</tr>
<tr>
<td>Boriani et al., 2001</td>
<td>41 (4 w/ SAE)</td>
<td>mobile spine</td>
<td>3/4 treated w/ SAE resolved; 1 recurrence in SAE &amp; curettage; higher incidence of deformity w/ curettage</td>
</tr>
<tr>
<td>Mohit et al., 2004</td>
<td>1</td>
<td>craniovertebral junction</td>
<td>3 treatments w/ polyvinyl alcohol; complete ossification</td>
</tr>
<tr>
<td>De Cristofaro et al., 1992</td>
<td>19 ABCs, 5 angiomas</td>
<td>mixed</td>
<td>2 recurrences (median 22-mo follow-up); 11 had complete ossification treated w/ NBCA; cyst resolution &amp; ossification at 3 yrs</td>
</tr>
<tr>
<td>Marushima et al., 2009</td>
<td>1</td>
<td>thoracic spine</td>
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