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Treatment of pelvic aneurysmal bone cysts in two children: selective arterial embolization as an adjunct to curettage and bone grafting

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ABSTRACT
Treatment options of aneurysmal bone cysts include complete resection, curettage, curettage with bone grafting, selective arterial embolization (as a primary treatment or preoperative adjuvant therapy), and percutaneous injection of fibrosing agent. Treatment in pelvic locations is difficult because of the relative inaccessibility of the lesions, the proximity of the lesions to neurovascular structures, and the vulnerability of the acetabulum. Herein, we present 2 pediatric cases of pelvic aneurysmal bone cysts successfully treated with curettage with bone grafting following preoperative selective arterial embolization.

Key words: • *bone cysts* • *aneurysmal neoplasms* • *bone embolization* • *curettage* • *bone grafting*

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Aneurysmal bone cysts (ABCs) are benign, non-neoplastic, expansile, vascular, locally destructive lesions. They represent 1% of all primary benign bone tumors; 3% arise from the sacrum and 8%–12% occur in the pelvis (1–3). The definitive etiology of aneurysmal bone cysts is still controversial. ABCs can exist either as primary bone lesions (70% of cases) or as secondary lesions arising in other osseous conditions (30% of cases). Pelvic ABCs are usually large and highly vascular (4, 5).

Although curettage and en bloc excision are the treatments of choice for accessible lesions, other treatment modalities, such as radiation, cryotherapy, percutaneous intralesional injection, and embolization, have been used for less accessible or recurrent lesions. Embolization has been used as a surgical adjunct to reduce blood loss (6–10). We present in this report 2 pediatric cases of pelvic aneurysmal bone cysts successfully treated with curettage with bone grafting following preoperative selective arterial embolization.

Case reports

Patient 1

A 13-year-old male patient was admitted to our institution with left hip and low back pain, which began 4 months earlier following a sports-related accident. The physical examination was normal, except for sensitivity to palpation and a positive Lasegue test at 70° on the left hip. The laboratory findings were all normal. Lumbosacral magnetic resonance imaging (MRI) demonstrated a 7 × 5 × 5 cm, expansile, lobulated, lytic, multi-septated, cystic mass on the S2 and S3 vertebrae, which extended to the left neural foramina, sacroiliac joint, and paravertebral muscles (Fig. 1). The cyst had fluid-fluid levels. The signal intensity of the levels was hypo- and hyperintense on T1-weighted MR images and hyperintense on T2-weighted MR images. Additionally, there was bone marrow edema on both iliac bones. The septa of the lesion enhanced after intravenous (IV) administration of gadolinium. Radiological diagnosis was aneurysmal bone cyst of the sacrum. We performed an aspiration biopsy after radiological evaluation, and cytological findings confirmed the radiological diagnosis.

Because of the location of the lesion and in order to prevent massive intraoperative bleeding, we decided to perform preoperative selective arterial embolization. The right main femoral artery was cannulated, following the administration of local anesthesia, using a 5F introducer sheath. Diagnostic angiography of the pelvis was performed with an angiographic catheter (pigtail, Terumo, Japan). Diagnostic pelvic angiography demonstrated a marked tumoral blush on the sacrum. After pelvic angiography, left lateral sacral artery was selectively catheterized with a micro catheter (Tracker®-18 Infusion Catheter, Target Therapeutics,

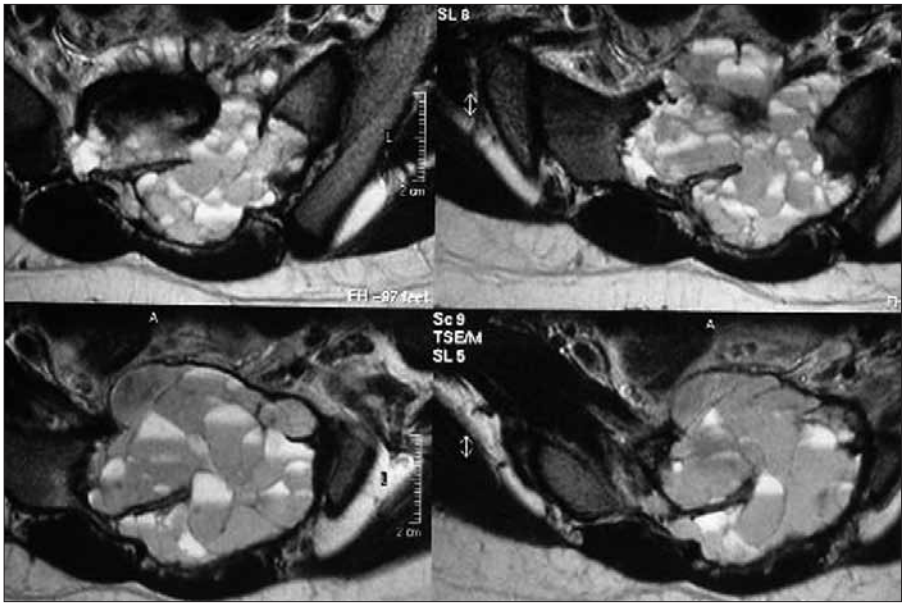

Figure 1. Serial axial T2-weighted MR images show an expansile, lobulated, lytic, multiseptated, cystic mass on the S2 and S3 vertebrae, which extends to the left neural foramina, sacroiliac joint, and paravertebral soft tissue.

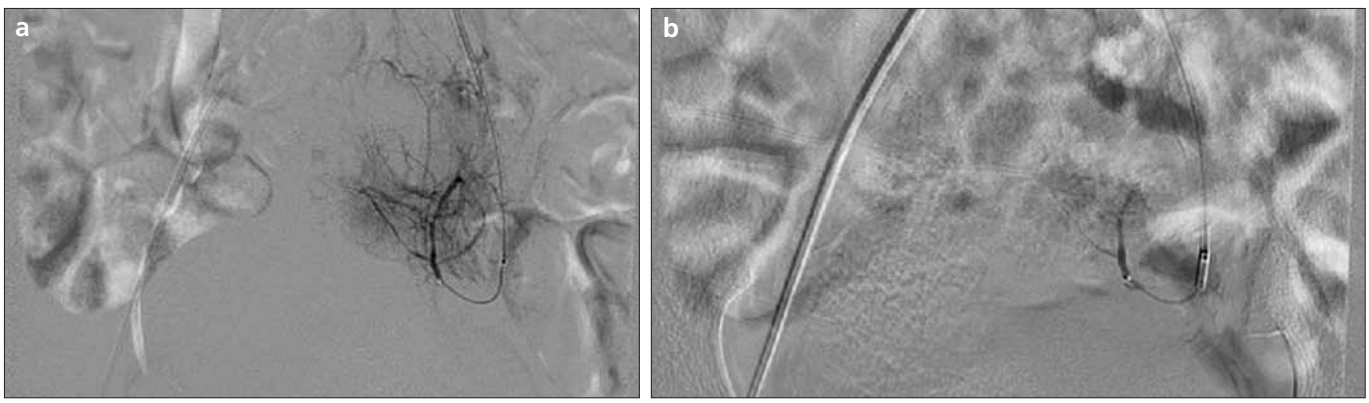

Figure 2. a, b. Left lateral sacral angiography **(a)** reveals a prominent tumoral blush on the sacrum. Follow-up angiography **(b)** after embolization with PVA shows no residual tumoral blush.

Boston Scientific, USA) and guide wire (0.016 microgold guide wire, Terumo, Japan), and arteriography revealed prominent perfusion of the lesion (Fig. 2a). Embolization was performed with 500-700 µm polyvinyl alcohol (PVA) particles (Contour®-PVA Embolization Particles, Target Therapeutics, Boston Scientific, USA) under constant fluoroscopic guidance until stasis within the target vessel was observed. There was no residual tumor perfusion after embolization (Fig. 2b). There was no enhancement of the lesion on selective angiography of the median sacral and right lateral sacral arteries. The day after embolization, curettage associated with bone grafting was performed through the posterior approach and there was no intraoperative bleeding. The patient was discharged on the seventh postoperative day. He is currently symptom-free and no residual or recurrent lesion has been seen during the 2-year follow-up.

Patient 2

A 14-year-old female patient was admitted to our institution with left hip and leg pain, which began 6 months earlier after she had fallen down. Physical examination revealed a shortness of the left lower extremity; all other findings were normal. Plain radiograph of the pelvis showed destruction of the left acetabulum. MRI demonstrated a 9 × 8 × 6 cm, expansile, lobulated, multiseptated, cystic mass on the left acetabulum, which extended to the ischium (Fig. 3). There was minimal edema on the pubic bone. The left hip joint was normal, except for minimal effusion. Radiological diagnosis was ABC. Due to the location of lesion and in order to prevent hazardous intraoperative bleeding, we decided to perform selective arterial embolization. The right main femoral artery was cannulated under local anesthesia using a 5F introducer sheath, and diagnostic angiography of the pelvis was performed with the use of an angiographic catheter (pigtail, Terumo, Japan). Pelvic angiography demonstrated a tumoral blush on the left acetabulum. The feeding arteries of the lesion originated from both the left internal iliac artery and left inferior epigastric artery. Following pelvic angiography, the left internal iliac artery was selectively catheterized with an angiographic catheter (cobra, Terumo, Japan). The lesion showed minimal enhancement on angiography and the feeding branches, which were shown to originate from the left inferior epigastric artery (Fig. 4a), were selectively catheterized with a micro catheter (Tracker®-18 Infusion Catheter, Target Therapeutics, Boston Scientific, USA)

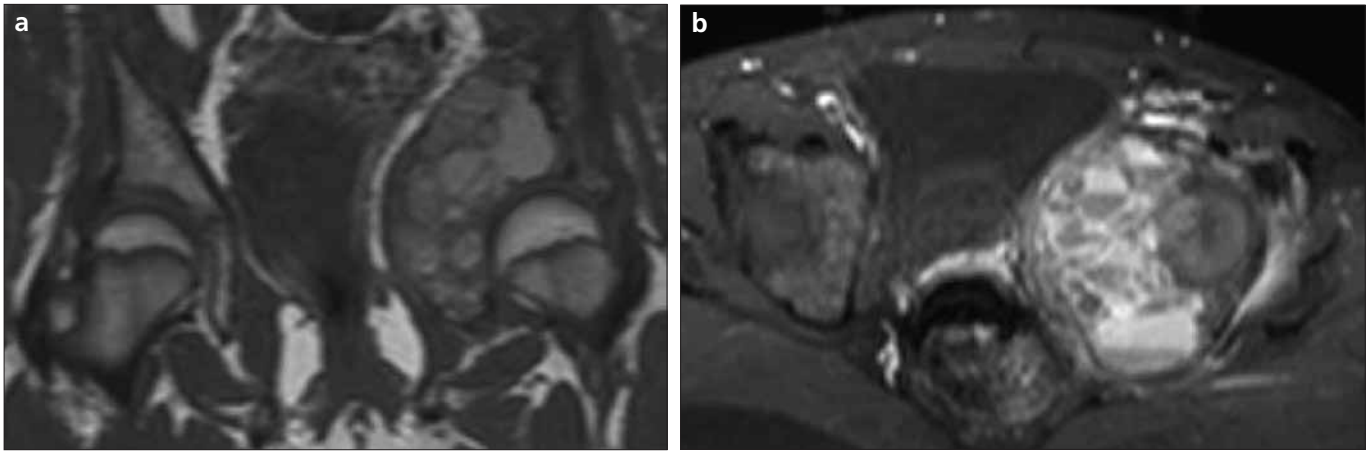

Figure 3. a, b. Coronal T1-weighted MR image **(a)** shows an expansile, lobulated, multiseptated, cystic mass on the left acetabulum. The mass has fluid-fluid levels hyper- and isointense with regard to muscles. Post-contrast axial fat saturated T1-weighted MR image **(b)** shows prominent enhancement of the lesion.

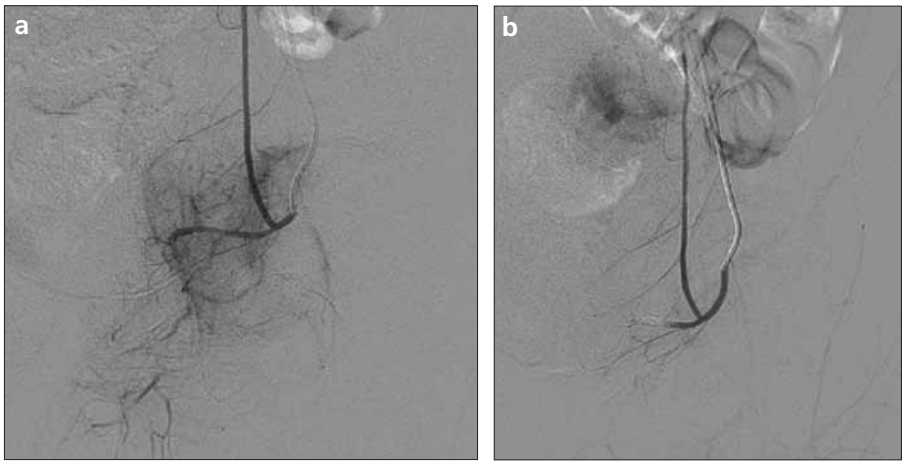

Figure 4. a, b. Left inferior epigastric angiography **(a)** demonstrates a prominent tumoral blush on the acetabulum. Follow-up angiography **(b)** after embolization with PVA and micro coil; no residual tumoral blush is seen.

and guide wire (0.016 microgold guide wire, Terumo, Japan). Embolization was performed with 500-700 µm PVA particles (Contour®-PVA Embolization Particles, Target Therapeutics, Boston Scientific, USA). The lesion's main feeding artery originated from the left inferior epigastric artery, as seen on the left external iliac angiography (Fig. 4a). There was no tumoral enhancement on the follow-up angiography after the embolization (Fig. 4b). The day after the embolization, curettage associated with bone grafting was performed through lateral incision. There was no intraoperative bleeding. The patient was discharged symptom-free and no residual or recurrent lesion was seen during the 6-month follow-up.

Discussion

Treatment of pelvic ABCs is difficult because of the relative inaccessibility of the lesions, associated intraoperative bleeding, proximity of the lesions to neurovascular structures, and the vulnerability of the acetabulum or sacroiliac joint. Treatment options include complete resection, curettage, curettage associated with bone grafting, selective arterial embolization as a primary treatment or preoperative adjuvant therapy, and percutaneous injection of fibrosing agent (3).

Recently, percutaneous injection of a fibrosing agent (Ethibloc, Ethicon) was employed with promising results (11–13). There are 2 published cases of pelvic ABC treatment by Ethibloc injection with good results (11, 12).

The method of treatment of ABCs of the pelvis and sacrum must be individualized based on location, and the extent and aggressiveness of the lesion. Lesions <5 cm that exhibit minimal destruction or expansion of the cortical bone, which do not threaten the integrity of the acetabulum or sacroiliac joint, are best treated with intralesional curettage, with or without bone grafting. Lesions >5 cm require a more aggressive approach with the excision-curettage technique (3). Rates of local recurrence after primary treatment, except en bloc excision, have been reported to range from 12% (14) to 31.5% (15), and even as high as 59% in a series of 44 patients treated by curettage alone, with or without bone grafting (16); however, complete resection is often impossible in this location.

A delayed diagnosis contributes to the difficulty of surgical extirpation (17–19); therefore, selective arterial embolization has been advocated as an adjuvant or definitive treatment of aneurysmal bone cysts in cases that are not accessible for primary surgical treatment.

Selective embolization of arterial feeders has been previously used as a primary modality of treatment in long bones and the thoracolumbar spine (8). The main advantage of selective arterial embolization is the avoidance of radical surgery. Given similar local recurrence rates for selective arterial embolization and intralesional excision, the use of selective arterial embolization as a primary modality of treatment seems reasonable. The disadvantages of selective arterial embolization in the treatment of ABCs include the risk of embolizing arterial supply to adjacent normal tissue and embolic complications (20).

Intraoperative bleeding is most common with curettage of sacral or acetabular lesions. Preoperative selective

arterial embolization is currently performed to minimize intraoperative hemorrhage (3). We chose to use preoperative selective arterial embolization as a means to decrease the vascularity of the cysts and thereby make subsequent surgery easier. Indeed, a significant reduction in arterial perfusion is essential to make the present lesion accessible to surgical treatment, thus contributing to its successful management. We performed selective arterial embolization before surgery in 2 patients, with PVA only in the sacral lesion, and PVA and micro coil in the acetabular lesion, both without recurrence.

In conclusion, we think that preoperative embolization of pelvic ABCs with PVA particles prevents massive intraoperative bleeding and facilitates surgery. Preoperative selective arterial embolization and curettage associated with bone grafting is an effective treatment method for pelvic ABCs.

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