Successful Treatment of Aggressive Aneurysmal Bone Cyst of the Pelvis With Serial Embolization

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Intralesional surgery is most commonly used for aneurysmal bone cysts. Rarely is en bloc resection used for active, aggressive, recurrent lesions and those located in expendable bones. However, persistence or recurrence of aneurysmal bone cysts is common. The clinical behavior of aneurysmal bone cysts is more aggressive in younger patients. Selective embolization is used as the primary treatment for aneurysmal bone cysts in surgically difficult anatomical locations and as an adjuvant to surgical treatment to reduce intraoperative blood loss and facilitate curettage.

This article describes a 3-year-old boy with an aggressive aneurysmal bone cyst of the pelvis involving the right ischiopubic rami that achieved curative treatment with 3 embolizations with N-2-butyl-cyanoacrylate. Biopsy was diagnostic; however, the clinical course was misleading. Twenty days after the first embolization, despite complete occlusion of the feeding vessels, the patient experienced severe pain, increased size of the lesion, and lateral subluxation of the right hip. Based on the imaging and histological diagnosis, intralesional hemorrhage was assumed, and repeat embolization was performed. After the second embolization, the patient experienced perineal skin necrosis from normal vessel embolization; it was treated with wound dressing changes and healed uneventfully. A third embolization was performed because of a persistent lesion. Six years after treatment, the patient was symptom free, and imaging showed complete ossification of the cyst.

Selective catheterization and occlusion of the feeding arteries with the appropriate embolic agent provide tumor devascularization, size reduction, pain relief, and induction of new bone formation. Multiple procedures are often necessary, and complications may occur.
Aneurysmal bone cysts are osteolytic hemorrhagic lesions of unknown origin causing bone expansion and destruction.\textsuperscript{1,3} Eighty percent occur in the first and second decades of life; in patients younger than 8 years, aneurysmal bone cysts are rare.\textsuperscript{3,4} Approximately 12\% occur in the pelvis.\textsuperscript{3,5} They are usually primary lesions; however, secondary aneurysmal bone cysts occurring in primary chondroblastoma, osteoblastoma, giant cell tumor, angiosarcoma, and osteosarcoma have been reported.\textsuperscript{1,3}

Rarely, indolent aneurysmal bone cysts heal spontaneously; however, the majority need to be addressed surgically.\textsuperscript{6} Patients with spontaneous healing are relatively older than typical patients with aneurysmal bone cysts.\textsuperscript{7,8} However, in cases of large, progressive, or secondary aneurysmal bone cysts, uncertain diagnosis, and risk of fracture, a treatment approach is required.\textsuperscript{5,7}

Given the benign biological behavior of aneurysmal bone cysts, intralesional procedures, such as curettage with or without bone marrow injection, and bone grafting are the most commonly used treatment options.\textsuperscript{1,3,7,9} Currently, less invasive treatments including cryotherapy, intralesional injection of calcitonin, methylprednisolone, or sclerosing agents,\textsuperscript{2,3,7,10-17} and selective arterial embolization as preoperative adjuvant to reduce intraoperative blood loss or as primary treatment have been reported.\textsuperscript{7,10,14-16,18} However, persistence, continuous growth, or recurrence of the cysts is not uncommon.\textsuperscript{1,4}

This article describes a 3-year-old boy with an aggressive, persistent aneurysmal bone cyst of the pelvis treated successfully with serial selective transcatheter arterial embolization. The parents gave written informed consent for patient’s data to be included in this study. This study was approved by the authors’ Institutional Review Board.

**CASE REPORT**

A 3-year-old boy was admitted to the authors’ institution with right hip pain, discomfort with hip motion, and pelvic pain with the inability to rest or sleep in the supine position. Radiographs of the pelvis and right hip showed an expansile osteolytic lesion involving the right ischiopubic rami (Figure 1). Computed tomography (CT) and magnetic resonance imaging (MRI) of the pelvis showed an osteolytic lesion with cavitary areas and fluid levels. Computed tomography–guided biopsy and histology showed an aneurysmal bone cyst. Because of the young age of the patient and the location of the lesion, selective arterial embolization was performed.

Under general anesthesia, digital subtraction angiography and selective arterial embolization were performed through the left common femoral artery. At the first embolization, 3 feeding arteries were identified originating from the right obturator artery (Figure 2A). The feeding vessels were selectively catheterized with a 4 French diagnostic catheter and a microcatheter (Cordis Corporation, East Bridgewater, New Jersey). Embolic agent N-2-butyl-cyanoacrylate in 33\% lipiodol ultramix was successfully injected, sandwiched with 5\% glucosate solution to prevent polymerization with blood. One flacon (1 mL) of N-2-butyl-cyanoacrylate was mixed with 2 mL of 33\% lipiodol. From the mixture, 1 mL was aspirated in a 1-mL insulin syringe, and 0.1 to 0.2 mL of the aspirate mixture was injected, sandwiched with 2\% glucose solution under fluoroscopic control. Then, angiography was performed to evaluate occlusion and remaining pathological vasculature (Figure 2B). The patient was discharged from hospital 2 days after the procedure pain free.

Twenty days after the first embolization, the patient experienced increased pain at the right hip without systemic symptoms of infection (fever) or increased white blood cell count and C-reactive protein. Radiographs and CT of the pelvis showed increased size and extension of the lesion to the posterior pelvis and thigh and lateral subluxation of the right hip (Figure 3). These were explained by intracystic hemorrhage, and a second embolization was performed.

At the second embolization, 4 feeding arteries were identified originating from the right profunda femoris artery (Figure 4A). Complete occlusion of the feeding arteries using the same procedure was performed (Figure 4B). The patient experienced pain relief 4 days after the second embolization; however, perineal skin necrosis occurred (Figure 5A) from embolization of branches of the external pudendal arteries (Figure 5B). Necrosis was treated with wound dressing changes, including wound cleaning with saline, sterilization with povidone iodine, and packing with a dry dressing pad, and healed uneventfully over the next 4 months.

Three months after the second embolization, the patient was pain free; however, clinical examination and imaging (Figure 6) showed a large posterior pelvic mass from persistent cyst, and a third embolization was performed. At the third embolization, 5 feeding arteries were identified originating from the right internal iliac, inferior gluteal, and obturator arteries (Figure 7A). All feeding arteries were successfully embolized using the same procedure with no complications (Figure 7B). The patient was discharged from hospital 2 days after the procedure pain free.
Routine follow-up evaluation, including standard radiographs of the pelvis and right hip, was performed every 3 months for the first 2 years, every 6 months for the next 3 years, and then annually. Radiographs showed progressive homogeneous trabecular bone formation and gradual reduction of the aneurysmal bone cyst. At 6 years, the patient was symptom free. Imaging studies showed complete ossification of the lesion (Figure 8).

**DISCUSSION**

Radiographs remain key in the diagnosis of aneurysmal bone cyst. The diagnosis is usually suggested by CT and MRI findings of blood cavities separated by septa. Fluid–fluid levels are suggestive of aneurysmal bone cysts, although they may also be observed with other lesions, such as telangiectatic osteosarcoma, Ewing’s sarcoma, and pathological fractures, in unicameral bone cysts. Biopsy and histology are paramount for safe diagnosis.

The current article described a 3-year-old boy with an aggressive aneurysmal bone cyst. Biopsy was diagnostic; however, the clinical course was misleading. The cyst persisted despite complete occlusion of the feeding vessels with N-2-butylcyanoacrylate embolization. Although the authors acknowledge that 1 embolization may not be efficient for complete tumor devascularization, in the current patient, 20 days after the embolization, the lesion continued to be painful and expanded, mimicking a sarcoma. Based on the imaging and histological diagnosis, the authors’ clinical experience, the young age of the patient, and the difficult location, the authors proceeded with repeat embolization that was finally successful.

Intralesional surgery is most commonly used for aneurysmal bone cysts. Rarely is en bloc resection used for active, aggressive, and recurrent lesions and those located in expendable bones. However, persistence or recurrence of aneurysmal bone cysts is common; the recurrence

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**Figure 2:** Angiography at the first embolization showing 3 feeding arteries originating from the right obturator artery (arrows) (A). Angiography after embolization showing occlusion of the feeding arteries (B).

**Figure 3:** Anteroposterior radiograph (A) and computed tomography scan (B) of the pelvis 20 days after the first embolization showing increased size and extension of the lesion to the posterior pelvis and thigh and lateral subluxation of the right hip (arrows).

**Figure 4:** Angiography at the second embolization showing 4 feeding arteries originating from the right profunda femoris artery (arrows) (A). Angiography after embolization showing occlusion of the feeding arteries (B).
rates for aneurysmal bone cysts range from 10% to 59%.1,3,7,19,22 The clinical behavior of aneurysmal bone cysts is more aggressive in younger patients.1,4,7 Moreover, surgical treatments have been associated with complications, including hip instability, growth plate injury, skeletal deformities, infection, neurovascular injury, pulmonary embolism, and long rehabilitation, and with the need for reconstruction procedures.3,9,21

Given the benign biological behavior of aneurysmal bone cysts, intralesional injection of sclerosing agents,12 radiation therapy,1-3,10 cryotherapy,13 systemic calcitonin therapy, and embolization have been reported with variable success rates.2,3,7,10-17 Local recurrences are reported in up to 59% of cases, but may be higher in children.1-4,7,10,11,17,18 Radiation therapy is not recommended for aneurysmal bone cysts because of the risk of post-radiation complications.

Selective embolization is used as a primary treatment of aneurysmal bone cysts in surgically difficult anatomical locations and as an adjuvant to surgical treatment to reduce intraoperative blood loss and facilitate curettage.10,15-17,23-26 Aneurysmal bone cysts are usually supplied by ≥1 feeding arteries. The feeding arteries and collaterals, their relationship
with adjacent vessels, and the possible arterial venous fistula inside the tumor must be carefully evaluated in angiography to avoid embolizing adjacent normal tissue and embolic complications. Selective catheterization and occlusion of the feeding arteries with the most appropriate embolic agent provide for tumor devascularization, size reduction, pain relief, induction of new bone formation, and avoidance of complications. Multiple procedures are often necessary, as was seen in the current patient.

The main advantage of selective arterial embolization is the avoidance of surgery, postoperative complications, and complex reconstructive procedures. The success rate of embolization for aneurysmal bone cyst ranges from 75% to 94%. The recurrence rate after embolization for aneurysmal bone cyst ranges from 39% to 44%. Persistence or recurrence of aneurysmal bone cysts after embolization has been associated with cysts >5 cm. In cases of persistent or recurrent lesions, repeat embolization can be performed successfully. However, embolization may also be associated with complications, which may be significant, as in the current patient. The overall risk of embolization-related complications is as high as 6%; these complications consist of dissection of the femoral artery at the site of transarterial catheterization, pain due to ischemic necrosis of the tumor, accidental embolization into nontumor vessels, infection, and postembolization syndrome.

Embolization of adjacent normal vessels can result in a large zone of tissue loss and may be associated with risk of nerve palsy, skin breakdown and subcutaneous or muscle necrosis, and infection because of tissue ischemia. The risk of complications is higher in certain anatomic regions. In the pelvis and hip, the lateral circumflex femoral artery, the supply to the sciatic, femoral, and lateral cutaneous nerve of the thigh, and the pudendal arteries should be avoided to prevent ischemic complications at the perineal area, as in the current patient. In the humerus, the posterior circumflex humeral artery, and the spine, the Adamkiewicz artery that originates between the T5 and L2 vertebrae should be avoided. To avoid embolizing adjacent normal tissue, operator’s experience, careful evaluation of the relationship of the feeding arteries with the adjacent vessels, and selective catheterization and embolization of the pathological arteries with the most appropriate embolic agent is required.

Embolic agents include gelfoam, polyvinyl alcohol particles, liquid (absolute alcohol), coils, adhesives, ethanol, and microfibrillar collagen. Speed and reliability of delivery, duration of occlusive effect, and preservation of normal tissue are considerations for choosing an embolic occluding agent. N-2-butylcyaanoacrylate is a liquid embolic agent that spreads according to its polymerization time and the vascular flow and can pass through bent catheters, navigating tortuous blood vessels. A distinct advantage of the N-2-butyl-cyanoacrylate in lipiodol is its dense radiopacity that allows for its exact site of occlusion to be observed. The current authors use N-2-butylcyaanoacrylate for permanent occlusion of the target vessels and complete devascularization of the lesion. Bolus administration of 0.1 to 0.2 mL of N-2-butylcyanoacrylate in 33% lipiodol sandwiched with 5% glucosate solution is necessary for safety. However, complications occur from embolization of adjacent normal vessels, as in the current patient.

**REFERENCES**

Case Report


