Unicameral Bone Cyst of the Pelvis in a 13-year-old Boy Treated with Cannulated Screw Decompression After Open Curettage and Grafting: A Case Report

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Abstract

Although studied for more than a century, the cause of unicameral bone cyst (UBC) remains uncertain. UBC of the pelvis in younger patients has been particularly problematic because its rarity often results in misdiagnosis and improper treatment. Surgical treatment has typically involved percutaneous or open curettage and bone grafting; however, some studies have described the use of a continuous decompression device to address high recurrence rates of the lesion. We present a 13-year-old male patient in whom, after two unsuccessful operations, open curettage and grafting and insertion of a cannulated screw led to resolution of pain caused by a large, recurrent UBC in the left ilium. Results of clinical examinations 1.5 years after the third operation indicated successful treatment. The use of a cannulated screw after open curettage and grafting may provide a clinically effective option for treating UBC.

Introduction

Originally described by Virchow[1] in 1876, a simple or unicameral bone cyst (UBC) is a benign, fluid-filled lesion that affects tubular and flat bones. Eighty percent are found in young patients aged 3 to 14 years, with a higher occurrence in males than females (2.5:1).[2,3] Although possibly related to obstruction of venous or lymphatic outflow in bone,[4] the actual etiology of UBC remains unknown. The cysts are commonly found in the metaphyseal region of long bones (especially the femur and humerus) and tend to be asymptomatic and incidentally discovered on radiographs or after pathological fractures.[5-7]

Because of their rarity, UBCs of the pelvis are problematic in diagnosis and treatment.[8] Pelvic lesions compose only 2% of all UBCs and, unlike cysts of the femur and humerus, have been mostly reported in adults and older pediatric populations.[8-11] Pelvic UBCs are often misdiagnosed as aneurysmal bone cysts or fibrous dysplasia, which can result in improper treatment.[8] We describe one patient in whom open curettage and grafting for a large UBC in the left ilium led to worsened pain, whereas open curettage and grafting and insertion of a cannulated screw led to excellent reduction in pain and a return to competitive sports.

Case Report

In January 2011, a 13-year-old male cross-country runner presented to our clinic with pain in the left hip for the past year. The pain began spontaneously, without any history of trauma, and usually occurred while running. No pain was reported at rest or at night. The patient had noticed an increase in the size of his left hemipelvis relative to his right.

On initial physical examination, the patient was tender to palpation posteriorly in the region of the left sacroiliac joint and anteriorly inferior to the anterior superior iliac spine. He also had pain with hip flexion greater than 70°, which worsened with internal and external rotation. Results of sensory, motor, and vascular examinations in the left lower extremity were unremarkable, but testing in flexion, abduction, and external rotation of the left hip recreated the patient's posterior pain. A radiograph of the pelvis revealed a large expansile lytic lesion in the left ilium (Figure 1). An image obtained by magnetic resonance imaging (MRI) of the pelvis with contrast material revealed a multiloculated fluid-filled lesion in the left ilium; however, neither evidence of a solid component to the lesion nor fluid-fluid levels were detected. The MRI image did show signs of mild cortical thinning and expansion. After evaluating the image, we determined that the most appropriate diagnosis was UBC, with aneurysmal bone cyst being a less likely possibility.

In February 2011, the patient underwent open curettage and bone grafting with a posterior approach to the left ilium. Subperiosteal dissection was performed until the expanded cortex overlay the lesion, after which a cortical window was developed and clear yellow fluid was encountered in the lesion. Aggressive curettage allowed the cavity to be

irrigated and packed with 120 mL of cancellous allograft chips. Results of a pathological study of the lesion were consistent with UBC. Postoperatively, the patient noted gradual lessening of activity-related pain in his left hip; between 6 and 10 months after the initial procedure, however, the pain worsened to preoperative baseline. A radiograph (Figure 2A) and a computed tomography (CT) image (Figure 2B) obtained at 10 months showed reappearance of the lesion in the left ilium.

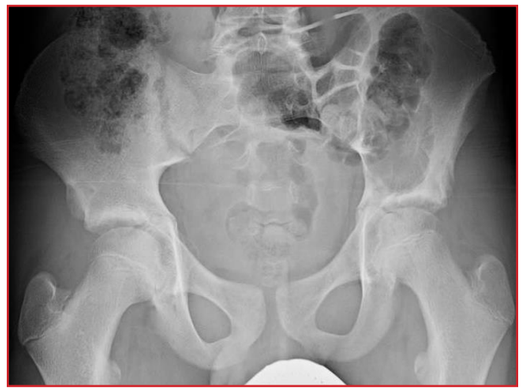

Figure 1. Radiograph of the pelvis obtained before the first procedure shows an expansile, large lytic lesion in the left ilium.

About 12 months after the first operation, the patient underwent another open curettage and bone grafting of the lesion in the left ilium. The skin was incised along the previously cut surgical scar using a similar posterior approach. Curettage was performed, the cavity was irrigated, and the opening was packed with 30 mL of cancellous allograft chips. Results of a pathological study were consistent with recurrent UBC, and a cyst wall was observed. Postoperatively, the patient's pain resumed and gradually worsened. By September 2012, he ambulated with a persistent limp. CT scans revealed persistence of a supra-acetabular defect that was not accessed by the posterior approach.

After evaluating a pelvic angiogram, we performed an alternative method to surgery by arterial embolization in the posterior division of the left internal iliac artery supplying the cystic lesion. Two successive CT scans revealed a recurrent, large lytic lesion in the left ilium (Figure 3). A surgical procedure was recommended because of the risk of pathologic fracture in the left acetabulum.

Twenty-two months after the index procedure, the patient underwent a third open curettage and bone grafting, involving aggressive curettage, irrigation with hydrogen peroxide, and grafting 240 mL of allograft cancellous chips (harvested from the right iliac crest) by an ilioinguinal approach. A cannulated screw was inserted into the cystic lesion. By 6 months postoperatively, the patient had complete resolution of pelvic pain and returned to competitive sports. A follow-up CT scan showed continued incorporation of the allograft, with a small persistent cystic component in the posterior aspect of the ilium (Figure 4). At 16 months after the third procedure, the patient remained asymptomatic (Figure 5) and results of a hip examination indicated pain-free range of motion with no tenderness.

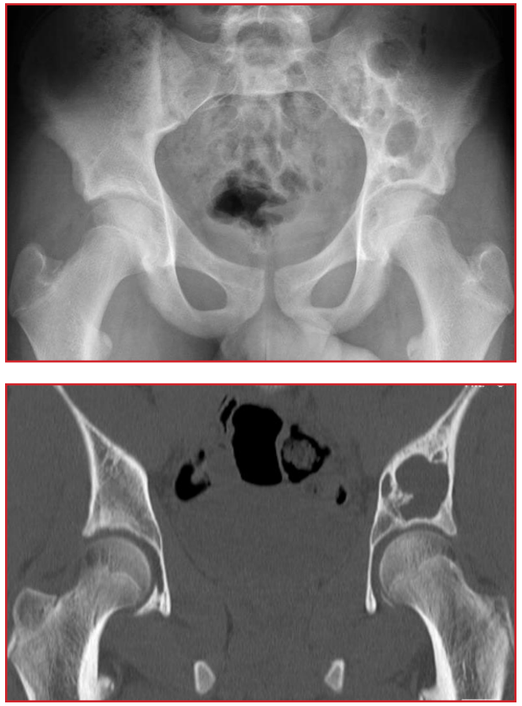

Figure 2. Scans of the pelvis obtained at 10 months after the first procedure reveal reappearance of the lesion in the left ilium. (A) Radiograph. (B) Computed tomography image.

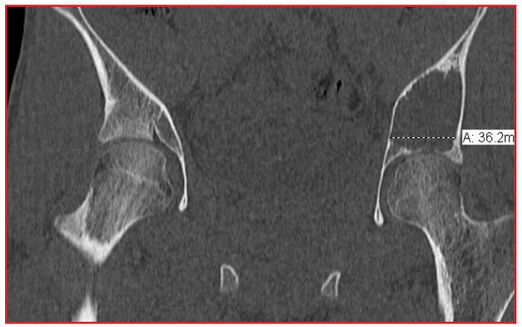

Figure 3. Successive computed tomography images of the ilium obtained after the second procedure and pelvic angiography show a recurrent, large lytic lesion in the left ilium.

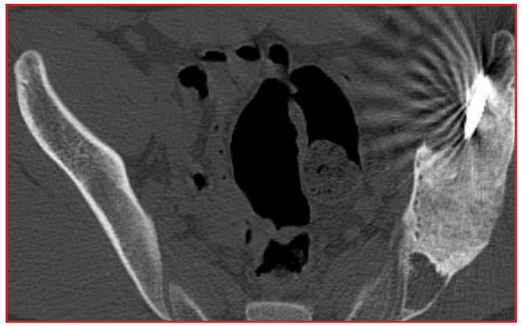

Figure 4. Computed tomography image obtained at 6 months after the third procedure shows the allograft cancellous chip and a small persistent cystic component in the posterior ilium.

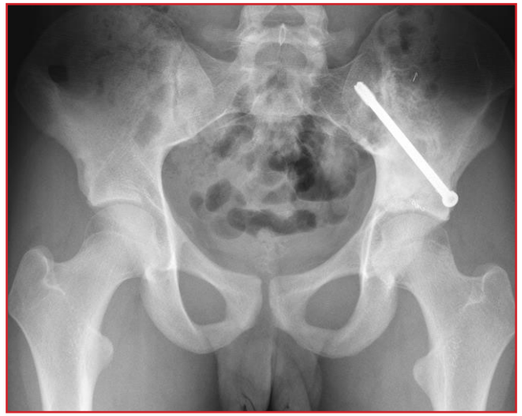

Figure 5. Radiograph obtained at 16 months after the third procedure shows the successful treatment of the unicameral bone cyst, with no recurrent lesion and a cannulated screw inserted in the left ilium.

Discussion

The findings of the current case are consistent with the diagnosis of UBC. Radiographs and CT scans of pelvic UBC generally show lytic lesions centered between the inner and outer table of the ilium, with the cyst fluid measured less than 20 Hounsfield units.[12,13] In our study, MRI images revealed the multiloculated fluid-filled nature of the lesion; however, the lesion did not show fluid-fluid levels or solid components, which would have been more indicative of aneurysmal bone cyst or fibrous dysplasia, respectively.[14] Despite imaging findings consistent with simple cyst, results of a pathological study were needed to confirm the diagnosis. Results of the two independent pathological studies of samples of the cyst lining indicated a fibrous membrane with occasional giant cells, which is consistent with signs of UBC.[3]

The goals in treating UBC are to prevent future pathologic fracture and lessen pain associated with the cyst by encouraging bone to fill in the cavity.[9,12] Surgical treatment, which ideally entails fewer procedures and complications, can be separated into percutaneous (including aspiration, autogenous bone marrow injections, steroid injections, and curettage and grafting) and open techniques (including curettage and grafting and subtotal resection).[9,15] Additionally, some authors advocated the use of cannulated screws, K-wires, or flexible intramedullary nails to provide continuous decompression of fluid within the cyst to stimulate healing.[16,17] Hou et al[15] reported a significantly shorter time to solid union of UBCs treated with curettage and grafting and decompression with a cannulated screw compared to curettage and grafting alone or percutaneous procedures.

The findings of the current case are also consistent with the high risk of recurrence after surgical treatment of UBC. Published recurrence rates have been high (up to 50%) after open curettage and grafting, with larger cysts most likely to recur.[18] Most authors recommended complete removal of cyst lining before grafting,[3,18] although some suggested placing continuous decompression devices such as cannulated screws to decrease recurrence rates. In a case series of 26 patients, Tsuchiya et al[17] found success rates of 80% and 100% when inserting either a titanium cannulated screw or cannulated hydroxyapatite pin, respectively, after curettage and grafting; interestingly, the only twice-recurrent lesion was UBC of the ilium. Recurrence in our case reflects the difficulty in accessing the entirety of a large, iliac UBC by using one approach.

Although the difficulties in diagnosis and treatment are well described, pelvis UBCs continue to present a challenging problem to surgeons and younger patients. Our study exemplifies the problems in diagnosing and treating a large UBC of the ilium. However, the successful outcome with use of a cannulated screw after open curettage and grafting may offer an effective method of treatment.

References

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