Case Report

rare tumors

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Unicameral bone cyst in the pelvis: report of a case treated by placement of screws made from a composite of unsintered hydroxyapatite particles and poly-L-lactide

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Abstract

Unicameral bone cysts are fluid-filled benign lesions that occur mostly in the long bones. Unicameral bone cysts in the pelvis are extremely rare. Continuous decompression using titanium or hydroxyapatite screws has been reported as a treatment. Screws made from a composite of unsintered hydroxyapatite particles and poly-L-lactide can be used for the treatment. An adolescent male patient presented with a unicameral bone cyst in the ilium extending to a region adjacent to the hip joint. As initial treatment, the cortex was fenestrated when the patient was 13 years old and β-tricalcium phosphate implanted. The cyst first reoccurred when the patient was 15 years old and again when he was 17 years old. During the most recent treatment, unsintered hydroxyapatite particles and poly-L-lactide composite screws were placed, and no recurrence was observed during 2 years of follow-up. The slow biodegradability and absorbability of the screws may allow continuous drainage of unicameral bone cysts.

Keywords

Pelvis, unicameral bone cyst, solitary bone cyst, composite of unsintered hydroxyapatite particles and poly-L-lactide (u-HA/PLLA), screw

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Introduction

Unicameral bone cysts, or solitary bone cysts, are a unilocular cavity in the bone, lined by a fibrous membrane and filled with fluid, which often present in childhood and adolescence. These cysts most commonly occur in the metaphyseal regions of long bones, in particular the proximal femur and the proximal humerus. In addition to the long bones, the calcaneus is also a common site for unicameral bone cysts. Unicameral bone cysts of the pelvis are extremely rare.[1]

The etiology of unicameral bone cysts is unknown, but it is assumed that venous obstruction elevates intraosseous pressure, leading to cyst formation.[2] The pathogenesis of unicameral bone cysts in the pelvis in children could be similar to their pathogenesis in the long bones.[1] As a treatment for unicameral bone cysts in the long bones, minimally invasive methods to place cannulated screws or pins

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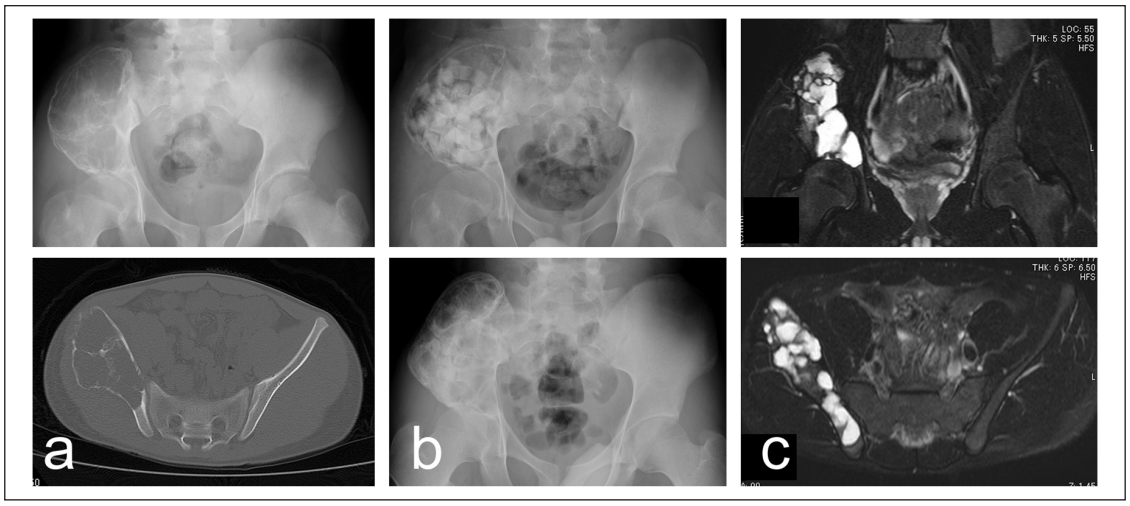

Figure 1. A unicameral bone cyst of the ilium. (a: top) Plain radiographs show an expansive osteolytic lesion in the ilium when the patient was 13 years old. (a: bottom) CT shows an expansive thinned cortex and osseous septa. (b: top) β-TCP was implanted after cortical fenestration. (b: bottom) When the patient was 15 years old, 23 months after the first operation, a recurrent osteolytic area was obvious. (c: top, coronal; bottom, axial) The recurrent lesion contained fluid with high signal intensity in T2-weighted images, with septa characterized by low signal intensity.

made of titanium or hydroxyapatite to provide continuous decompression of the cyst have been applied.[3,4] A cannulated hydroxyapatite, rather than titanium, screw or pin is recommended because it has a higher treatment success rate and its removal is unnecessary.[4] However, hydroxyapatite pins are not commonly used except for unicameral bone cyst treatment. Therefore, their commercial availability is apparently not widespread.

A composite of unsintered hydroxyapatite particles and poly-L-lactide (u-HA/PLLA) is available in a screw format (Super Fixsorb MX30; Johnson & Johnson, and Takiron), and this material is commonly used for maxillofacial surgeries.[5] u-HA/PLLA is biodegradable, absorbable, and osteoconductive, leading to its replacement by bone.[6] In the current case, u-HA/PLLA composite screws were used to treat a patient with a recurrent pelvic unicameral bone cyst.

Case report

A 13-year-old boy noticed pain in his right hip when he walked. He had no history of related trauma. His hip joint had a full range of motion and was the same bilaterally, but passive movements caused the hip pain. Plain radiographs showed an osteolytic lesion with an expansive and thinned cortex in the right ilium. The osteolytic lesion extended to a region adjacent to the hip joint (Figure 1(a)). Computed tomography (CT) confirmed that the cortex of the ilium was expanded and thinned, and revealed septa inside the lesion. The content of the lesion was homogeneous with a low CT image intensity, suggesting that the content was fluid, but not blood (Figure 1(a)). A unicameral bone cyst, rather than aneurysmal bone cyst characterized by blood content, was diagnosed based on the CT findings. Upon the diagnosis of the unicameral bone cyst, the cortex at the iliac crest was fenestrated, and the size of the fenestration was about 1 cm × 4 cm. The septa were removed as much as possible to provide a continuum between each sectional cavity. Then β-tricalcium phosphate (TCP) was implanted into the cavity (Figure 1(b)). Intraoperative findings showed the lesion was cystic, and its content was serous fluid, but not blood. The specimen from inside the lesion showed walls of a cystic lesion composed of fibrous tissue or granulation tissue with deposits of hyalinized eosinophilic or calcified material, and foci of new bone formation and hemorrhage. These features are compatible with those of a unicameral bone cyst. Postoperatively, partial weight-bearing was allowed for 3 months with a crutch, and then full weight-bearing was allowed without a crutch. The implanted β-TCP was gradually absorbed without bone incorporation. When the patient was 15 years old, 2 years after the initial surgery, the osteolysis had expanded (Figure 1(b)). Magnetic resonance imaging (MRI) demonstrated the lesion with high signal intensity in T2-weighted and with low signal intensity in T1-weighted images. Septa were characterized by low signal intensity on both T2- and T1-weighted images (Figure 1(c)). Because recurrence was diagnosed, a second operation was performed to fenestrate the same portion of the iliac crest as previously fenestrated, and β-TCP was implanted into the recurrent cavity (Figures 2(a)). Because there was no pain, full weight-bearing was allowed after this second operation. After 6–12 months of the second operation, bone consolidation was apparently obtained. However, when the patient was 17 years old, about 24 months after the second operation, an osteolytic region in the acetabular area was obvious on a plain radiograph and MRI (Figures 2(a) and (b)). A similar third operation was performed to fenestrate the same

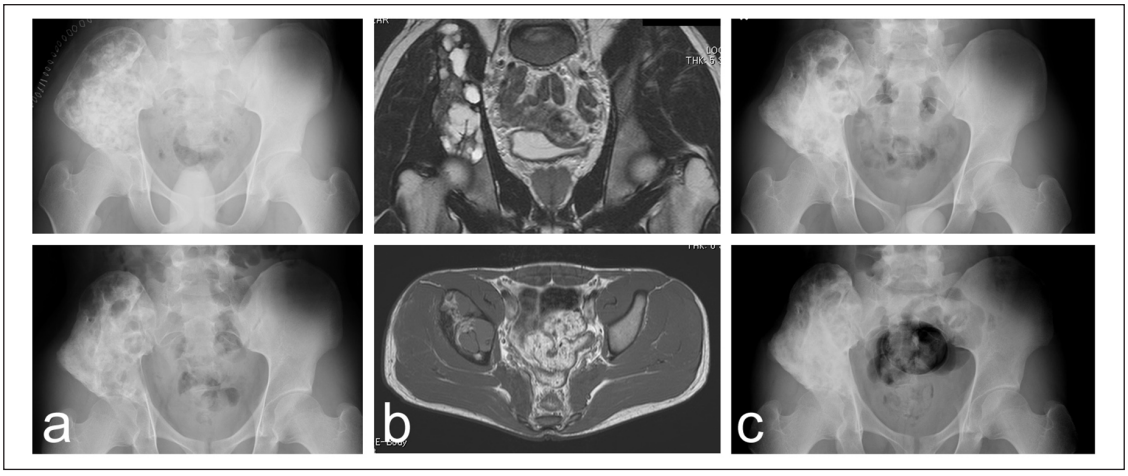

Figure 2. Reoperation for a recurrent unicameral bone cyst of the ilium. (a: top) When the patient was 15 years old, a second operation was performed to implant β-TCP after fenestration of the cortex. (a: bottom) When the patient was 17 years old, 24 months after the operation, an osteolytic region at the acetabular area was obvious. MRI shows fluid in the ilium as having (b: top) high signal intensity in T2-weighted and (c: bottom) low signal intensity in T1-weighted images. (c: top) To decompress the cyst, the cortex was fenestrated, β-TCP was implanted, and u-HA/PLLA composite screws were placed. (c: bottom) Bone consolidation is seen 24 months after the third operation, without recurrence of the osteolytic region.

portion of the iliac crest as previously fenestrated, and β-TCP was implanted into the recurrent cavity to treat the recurrent lesion. In addition, 3 u-HA/PLLA composite screws were placed (Figures 2(c)). At 24 months after the third operation, irregular bone consolidation was obtained and recurrence of osteolytic region was not seen, at least not in the acetabular area. His hip joint had a full range of motion and was the same bilaterally, without any hip pain.

Discussion

Unicameral bone cysts are characterized by an expanded cystic lesion with a thinned cortex. Long bones are the preferential site for unicameral bone cysts, especially in the proximal metaphysis of the humerus and femur. The differential diagnosis of unicameral bone cysts among cystic lesions is an aneurismal bone cyst, characterized by expansive cystic lesion containing blood. In the current case, the absence of fluid–fluid lines as a result of blood sedimentation in CT examination excluded the diagnosis of an aneurysmal bone cyst. The diagnosis of a unicameral bone cyst was confirmed based on the interoperative findings of a cystic lesion with serous content.

The pathogenesis of unicameral bone cysts in the long bones has been assumed to be blockage of circulation because of venous obstruction.[7] The pelvis is rare as a site for unicameral bone cysts, while the cysts are relatively common in the long bones.[8] Unicameral bone cysts in the long bones are found mostly in the metaphysis adjacent to the epiphyseal plate in children and adolescents.[8] Unicameral bone cysts tend to improve spontaneously in most patients, especially after skeletal maturity. Therefore, the development of the lesions may be related to skeletal growth. The pathogenesis of pelvic unicameral bone cysts in childhood may be similar to that seen in the long bones.[1] However, in a previous study of 16 cases of unicameral bone cysts in the pelvis, the cysts were diagnosed in both young and older patients.[1] The patients ranged in age from 9 to 69 years with the mean age of 30 years.[1] Cysts in the pelvis may develop later in life.[9] Unicameral bone cysts in the elderly have been assumed to be associated with sacropelvic joint degeneration because of findings that unicameral bone cysts occur in the posterior portion of the ilium, adjacent to the sacroiliac joint in the elderly.[1] The different pathogenesis or etiology of unicameral bone cysts based on site specificity has been reported in long bone and calcaneus. Accordingly, the pathogenesis could be different in the pelvis.

Pelvic lesions may remain asymptomatic longer when they occur in non-weight-bearing regions.[1] Another reason for pelvic unicameral bone cysts being found in adult age may be that long-standing asymptomatic pelvic lesions are being found coincidentally. There is no consensus on best treatment, but treatment is generally unnecessary in asymptomatic individuals. However, unicameral bone cysts in the long bones seldom heal, even after fracture.[10] For this reason, surgery is recommended, especially for lesions with impending fracture or prefracture.

As a treatment for unicameral bone cysts, many minimally invasive methods have been proposed, such as cortical decompression, percutaneous calcium sulfate grafting, and autogenous bone marrow injection.[11] Curettage of unicameral bone cysts in the long bones with bone grafting is followed by a high rate of recurrence.[12] Corticosteroid injection may also require multiple procedures. Bone grafting and curettage tend to be applied for larger bone cysts.

The choice of treatment is controversial, but many factors, such as size, bone strength, age of the patient, and activity level, need to be considered for appropriate treatment.[13,14]

In a previous report, nine patients with pelvic unicameral bone cysts underwent curettage of their lesions, and the lesion was filled with a bone graft in one case. Eight of the patients were treated with one curettage, and one patient had a second curettage 21 years after the first procedure.[1] In the treatment of pelvis, asymptomatic unicameral bone cysts of the pelvis in non-weight-bearing regions do not usually require surgical intervention. In the current case, surgery was necessary because the lesion extended to a region adjacent to the hip joint, and the patient had pain, suggesting impending fracture. Screw placement was chosen in the current case. A cannulated screw can be inserted through a small fenestration to provide continuous decompression as a minimally invasive treatment,[3,4] and this method provides superior results to curettage and bone grafting.[3] The use of titanium or hydroxyapatite cannulated screws or pins has been reported. However, a cannulated pin made of hydroxyapatite, rather than titanium, is recommended because they have a higher treatment success rate and their removal is unnecessary.[4]

In the present case of a recurrent pelvic unicameral bone cyst, u-HA/PLLA composite screws were used for treatment. u-HA/PLLA composite screws are not cannulated. Taking into consideration the assumed etiology of unicameral bone cysts in which venous obstruction elevates intraosseous pressure leading to cyst formation,[2] a cannulated form could be expected. However, a previous review reported treatment with intramedullary nails appears to provide more favorite clinical results than that with cannulated screws.[15] This reminded us the drainage possibly occurred around the nails. In our experience, cannulation in cannulated screws is easily obstructed by soft tissue or ossification at the hollow end of the screw. The current u-HA/PLLA composite screws appear to provide continuous drainage based on the successful clinical result. The u-HA/PLLA composite composed of u-HA and PLLA shows degradative and bioresorptive behavior. Experimentally, the PLLA matrix degrades after about 4–5 years, and almost all of the osteoconductive u-HA particles are replaced by bone tissue after about 5.5 years.[16]

In the current pelvic lesion, no recurrence was observed within a 2-year follow-up after surgery for u-HA/PLLA composite screw placement. The effectiveness of treatment with u-HA/PLLA composite screws is not conclusive in the current case, and fenestration alone might have been sufficient, although a comparative control is lacking. However, considering the relatively young age of the patient, possible recurrence was considered. Theoretically, screw placement has a positive effect in terms of reducing any possible recurrence. The slowly biodegradable and absorbable character of u-HA/PLLA makes this a method for continuous drainage that warrants consideration in the treatment of unicameral bone cysts. Moreover, like the use of hydroxyapatite pins, the use of u-HA/PLLA screws avoids a second operation because their removal is unnecessary.

In summary, unicameral bone cysts of the pelvis are rare. u-HA/PLLA degenerates and can be absorbed. To provide continuous drainage, u-HA/PLLA composite screws were placed in a fenestrated area of the cortex surrounding the cyst. We recommend u-HA/PLLA composite screws be considered for the treatment of unicameral bone cyst in the long bones and pelvic cysts. Further clinical assessment of the use u-HA/PLLA composite screws to treat unicameral bone cysts is warranted.

Author contributions

A.S. drafted the manuscript. T.O. and S.M. designed the study. All authors approved the final version of the manuscript.

Conflict of interest

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Ethical approval

For this retrospective case report, permission from a local ethics committee was not required.

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Informed consent

The patient and guardians represented in this study were informed that the data from their case would be deidentified and used in a journal publication.

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