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Title: Peritrochantric Unicameral Bone Cyst in a Nine-year-old Girl; A Case Report and Review of Literature

Running Title: A Report of a Peritrochantric Unicameral Bone Cyst Case

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Abstract

Introduction: The unicameral bone cyst (UBC) is a benign osteolytic lesion mostly found in the metaphyseal part of long bones in children. It is important as it can cause growth plate involvement, pathological fractures, and deformities of the affected limb. Our goal to report this case was to emphasize that hip area pathologies can be represented with knee pain and discomfort.

Case Presentation: The patient was a nine-year-old girl with a bone cyst in the right proximal femur, with functional knee pain and limping. The diagnosis was made after two years of pain in the knee area. As the cyst was symptomatic and the signs of impending pathological fracture were seen, the lesion was managed by a surgical method of curettage and fibular strut allograft and proximal humerus locking plate.

Conclusions: The patient has been examined for knee joint problems for a long time, and her femur bone cyst was diagnosed after two years of pain; while it could become a pathologic fracture or involve the growth plate and cause growth arrest of the limb, which could affect patient's quality of life for a lifetime. Therefore, in children with chronic knee pain, careful assessment of the hip area is recommended.

Keywords: Child, One cysts, Pain, Growth plate, Curettage, Allografts

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Introduction

The unicameral bone cyst (UBC), also called a solitary or simple bone cyst, is a benign lesion that occurs mainly in children and adolescents between 4-14 years old (1). It usually presents as an osteolytic cystic lesion filled with serous fluid and covered with a thin fibrous membrane, mostly found in the metaphyseal part of growing long bones (2). It usually disappears by itself when bone maturation completes (3). Nevertheless, it can lead to pathological fractures through destroying bony structure from the medulla toward the cortex (4). The lesion's etiology remains unknown, but the most popular theories are blocked fluid drainage, intramedullary venous obstruction, nitric oxide, and some lytic processes (5, 6).

The proximal humerus is the most common site of involvement, following with the proximal femur (2). Although the cyst is benign, the risk of pathological fractures and less frequently, the growth plate involvement makes it important to manage (4, 7). Symptoms consist of pain after pathologic fracture and limping, while most of the patients are asymptomatic, and the cyst is usually found incidentally or after a pathologic fracture (1, 3, 5, 8).

Follow-up is recommended for asymptomatic patients; however, it has an increased risk of pathologic fracture (athlete or weight-bearing bone) for those who have signs of an impending fracture, surgical intervention can be the choice treatment (9).

Our goal to report this case was to emphasize that hip area pathologies can be represented with knee pain and discomfort. As the patients with this problem are children and any possible complications can affect their quality of life, social appearance, and mental health for a lifetime, we found it beneficial to present a brief review of the treatment options performed up to this day.

Case Presentation

The patient was a nine-year-old girl who came with a chief complaint of functional knee pain and limping. The pain lasted for two years and had got worse in the last twelve months as the patient started doing sports, while her parents noticed limping in the patient's gait pattern.

Knee pain was more severe on the right knee's lateral side, usually presented after walking and running, and relieved by rest. Nighttime pains were reported only after daytime overactivity. The pain had no radiations to other parts, and there was no history of trauma or radicular pain. The patient had no associated symptoms such as fever, chills, nausea, vomiting, headache, dizziness, or lethargy. No past medical history such as trauma, surgery, or any particular disease existed. No family history of such orthopedic problems was reported.

During the physical examinations, no local tenderness or limited range of motion was detected in the suffering knee joint. Knee x-rays were also normal. With a normal knee and about two years of slowly progressing pain, suspicion of hip and pelvic problems arose. Following that, an X-ray, MRI, and bone scan was requested on both hip joints and pelvis. The lesion was found in the right femoral peritrochantric area (Figure 1A and 1B).

The roentgenograms and MRI showed a multi-locular lytic lesion in the neck and trochanteric part of the right femur, with a sharp margin and cystic structure. Thinning of the bone cortex of the affected area was also seen.

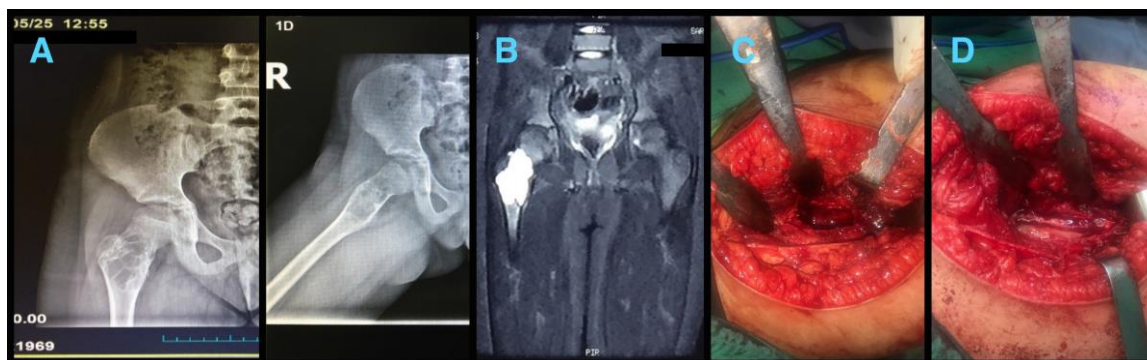


Figure 1. A. AP and lateral view radiographs of patient's right pelvis and hip joint at the time of diagnosis, B. T2 MRI of patient's pelvis and hip joints showing the defect as a hyper-signal lesion in the neck and peritrochantric area of right femur, C. Photography of the patient during surgery showing an open window into the lesion, D. Photography during surgery showing placed fibular allograft in the defect.

The patient was referred to our center to manage the lesion. As the condition was almost serious, due to the great risk of pathologic fracture, the surgery was done with a probable diagnosis of a unicameral bone cyst of the neck of the right femur.

Under general anesthesia, the surgery was done in supine position and Watson Jones approach, under C-Arm radiographic control. We opened a window into the lesion, similar to the lesion in the anterior of the right femur (Figure 1C). Curettage was done with curette and burr; two fresh frozen fibular allograft segments were fitted in the lesion as strut graft (Figure 1D). Then the locking plate (ARTA Orthoped Co.; Iran) was placed on the lateral side and fixed with locking and cortical screws. The result was checked under C-Arm radiography (Figure 2A).

The patient was discharged on day two after surgery and maintained non-weight bearing for six weeks. She is doing well right now, and her last follow up visit six months from surgery revealed complete healing of the lesion and good general condition and movement (Figure 2B).



Figure 2. Post-surgery radiographies. A. Immediate post-op AP and lateral radiography of right hip; B. Radiographies of the last follow-up.

Discussion and Review of Literature

We reported a unicameral bone cyst of the proximal femur in a nine-year-old girl, treated with the surgical technique. The unicameral bone cyst is a benign lytic lesion representing almost 3% of primary bone tumors in pediatrics. It tends to grow toward the cortex and predispose a bone to fracture (1-3). Patients with UBC are usually asymptomatic until a pathologic fracture is present (3, 4, 8). The pain of the affected area, thigh pain and limping is also seen as a complaint (10). To review the literature, we have searched different qualitative databases with different terms (unicameral bone cyst, simple bone cyst, children, pediatrics, allograft, fibular allograft, surgery); the found related cases are shown in Table 1.

Table 1. Reviewed literature of published cases with a unicameral bone cyst

Authors	Year of Publishing	Study Design	Cases Information	Manifestation	Treatment
Rosario et al. (5)	2017	Case Report	A 26-year-old kickboxing coach (Male)	A painful right shoulder which on radiographs and magnetic resonance (MR) imaging showed a proximal humeral lesion with signs of ossification	Diagnostic aspiration of the lesion was firstly done, revealing straw-colored fluid. The patient then underwent intralesional curettage with alpha-tricalcium phosphate cement reconstruction of the lytic defect.
Shapira et al. (9)	2012	Case Report	An 11-year-old girl	A 6-week history of left leg pain; The only significant finding upon physical examination is point tenderness over the anterior aspect of the proximal left tibia. There is no swelling, warmth, or erythema of the left leg or knee.	Not Accessible
Gregory et al. (11)	2010	Case Report	a 13-year-old boy	Bilateral distal femoral unicameral bone cysts (UBCs) associated with acquired generalized lipodystrophy.	The patient underwent bilateral cyst aspiration and cystogram, followed by incisional biopsy and intraoperative frozen section.
Aiba et al. (12)	2018	Case Series	37 patients (24 males and 13 females) Mean age 14.7 years	Simple bone cysts	Endoscopic curettage was performed with the support of an arthroscope via 7-8mm holes penetrated by cannulated drills with a small incision.
Roukhsi et al. (13)	2013	Case Report	An 11-year-old boy	Painful limping. Clinic, radiographic and pathologic findings and cyst aspiration	Repeated intralesional steroid injection

				evocated solitary bone cyst	
Langston et al. (14)	2016	Case Report	A 26-year-old woman	Secondary hip arthritis from developmental dysplasia of the hip and a large pertrochanteric bone cyst.	Staged total hip arthroplasty
Yilmaz et al. (15)	2010	Case Report	A 7-year-old boy	Recurring pain in the left knee. Physical examination of the patient, laboratory tests, and radiological examination of the knee joint were normal	Roentgenograms performed for a presumed hip joint problem revealed a focal cortical defect on the left femoral head and an increased effusion in the left hip joint space compared with the right counterpart.
Cho et al. (16)	2012	Case Series	25 patients (21 males and 4 females) Mean age 11.1 years	A unicameral bone cyst	Intramedullary decompression followed by grafting of demineralized bone matrix
Abdel Mota'al et al. (17)	2008	Retrospective study	14 cases	UBC of proximal femur	Treated surgically using intra-lesional excision
Subramanyam et al. (18)	2018	A Report of Two Cases	14-year-old and 13-year-old girls	14 (a history of pain in the right hip for 4 years); 13 (inability to weight bear on the left lower limb)	Both cases were operated in the fracture table under general anesthesia. Hip joint was accessed through Watson Jones approach.

Our patient had complained of knee pain and limping, which has not been mentioned before in the literature (Table 1). Knee pain can be a symptom of many important diseases in children, e.g., septic arthritis, rheumatoid disease, malignancies, and hip joint problems. It is fair to emphasize those hip problems such as developmental hip dysplasia can be present with no hip symptoms but in the knee joint (15). We found it valuable to share as a diagnostic tip in children presenting with chronic knee joint discomfort.

Different types of treatments have been suggested for UBC. Steroid injection was popular in the 1970s as a less invasive and more effective treatment than surgery (19). However, several studies since then demonstrated less effectiveness of this method and unpredictability of the outcome, even with multiple repeated injections (20). Intra-medullary decompression and bone grafting is another popular treatment that has been done with variant graft materials, e.g., medical-grade calcium sulfate, demineralized bone matrix, cancellous chips, and hydroxyapatite (HA) (3, 16, 17, 21).

Gokce et al. has done a minimally invasive surgery technique, including percutaneous decompression and using medical-grade calcium sulfate pellets as a graft. They worked on 55 patients, including seven patients with proximal femur cysts. Eighty percent of patients healed after one operation needed no further treatment. Eleven patients have undergone reoperation. The healing rate after the second surgery was 94%. Two patients developed pathologic fractures between the first and second surgery (18). Dormans et al. used the same technique on 24 patients with 91.7% complete healing, two patients underwent a second surgery, and one remained persistent even after the second surgery (22). This method slightly increases the risk of pathologic fractures and reoperation through the curettage of the cyst wall and applying no bone fixation. Also, a recurrence rate of 37% has been reported in a trial of 35 patients with the method mentioned above (23).

Chigira et al. have introduced the multiple drill-holes methods believing venous obstruction as the etiology of cysts. They tried it on seven patients, from which four has completely healed, 2 had partial recurrence after healing, and one had no response to the treatment. They believe the Kirschner Wires (KW) used for drilling can help drain the cyst through the holes, and as a foreign body stimulates the immune system in the area, leading to more blood flow and reconstructing factors to help the healing of the cyst (6).

Roposch et al. worked on the intramedullary nailing method on 12 patients with a defect on the proximal femur. This method prevents further iatrogenic trauma to the bone, as it does not need curetting the cyst wall. It has no blood loss as it is not open surgery and also prevents pathologic fractures. Though a complication rate of 25%, including nail perforating the bone's lateral cortex and shortening of the nail following the patient's growth, makes it an unreliable treatment in children (7).

Li et al. have compared treating the patient with two methods of autogenous bone marrow (ABM) injection and titanium elastic intramedullary nailing (TEN) on 46 patients, including 16 on the femur. The results showed 60.9% of complete healing in the ABM method and 69.6% in TEN, with a recurrence rate of 13% in both. Four pathologic fractures happened in the group treated with ABM, while one in the other group. Autogenous bone marrow injection needs multiple injections to gain acceptable results and does not prevent further pathological fractures. In contrast, TEN has the same disadvantages as other intramedullary nails mentioned above (24).

In a novel study done by Zhang et al., intramedullary nailing was combined with curettage and bone grafting and was compared with other groups; it had undergone curettage and bone grafting alone. The results showed a 90% healing rate in the first group and 68.8% in the second group, suggesting that intramedullary nailing could be added to the curettage and bone grafting as a safe and easy way to increase the rate of treatment effectiveness (25, 26).

Abdel-Mota'al et al. treated 13 patients with UBCs on proximal femur surgically using the intralesional excision technique. They filled the lesion with autogenous grafts in 3 cases, hydroxyapatite matrix in 8 cases, and a combination of two in 2 cases. They also used internal fixation with DHS in 5 cases, KW in one case, and external fixation in another case. Recurrence appeared in one case nine months after surgery. Malunion and growth arrest occurred in another patient that has presented with a pathological inter-trochanteric fracture. They found the HA matrix a useful and reliable substitute in the case of limited autogenous bone grafts in children (17). In another study, Subramanyam et al. has reported one case of pathologic fracture non-union due to UBC that had been previously treated with DHS. They treated a patient with open reduction, filling the lesion with cancellous and fibular strut graft and fixed with valgus osteotomy fixation, locking plate, and screws. They gained great results with normal ROM and healing of the cyst with a full union of the fracture (18). These two studies suggest that the DHS is not an appropriate fixation for the lesion than locking plate fixation (17, 18).

Jamshidi et al. worked on 14 cases using fibular strut graft and locking plate as fixation. Six patients had presented with pathologic fracture, 5 with thigh pain, and three patients have been found incidentally. Of 14 patients treated with this method, ten have healed completely, and four parts. No recurrence was reported at the time of follow up. No major complications were found; only one mild coxa vara and one heterotopic ossification. No patient discomfort or growth arrest

was reported during the follow-up. Casting and further interventions were not needed in any cases. They also noted a higher incidence of healing after a pathologic fracture, while all of the six patients presented with pathologic fracture versus half of the others (without fracture) healed completely (1). In addition to Subramanyan et al., these promising results encouraged us to use this technique in our patients (1, 18).

Conclusion

The patient has been examined for knee joint problems for a long time, and her femur bone cyst was diagnosed after two years of pain; while it could become a pathologic fracture or involve the growth plate and cause growth arrest of the limb, which could affect patient's quality of life for a lifetime. Therefore, in children with chronic knee pain, careful assessment of the hip area is recommended.

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Compliance with ethical guidelines: All ethical principles are considered in this article. The participants were informed about the purpose of the research and its implementation stages; they were also assured about the confidentiality of their information; moreover, they were free to leave the study whenever they wished, and if desired, the research results would be available to them

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