Expansive unicameral bone cyst occupying the distal tibia: A case report

by Andrew Robitaille, DPM; Lawrence M. Fallat, DPM, FACFAS

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Unicameral bone cysts (UBC) of the distal tibia are usually incidental findings. We present a case of a 50-year-old female who initially presented with chronic bilateral heel pain. Initial radiographs revealed plantar heel spurs, but also a large intraosseous cyst in the distal right tibia. Computed tomography was obtained which showed a large, multiseptated, lucent, expansile bone lesion in the central medullary canal the distal metaphyseal-diaphyseal junction of the distal tibia. To prevent fracture of the thin cortex and stop expansion of the cyst, surgical intervention was chosen. This case report serves to show how standard x-ray revealed a large UBC that could result in fracture of the distal tibia.

Keywords: bone tumor, cyst, unicameral, tibia, benign

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Unicameral bone cysts (UBC) are relatively uncommon benign bone tumors found mostly in the metaphysis of long bones, such as the humerus or femur with a male to female ratio of 3:1 [1]. UBC represents about 3% of primary tumors seen within the first two decades of life. In a meta-analysis by Kadhim, et al., in 2014, the distal tibia was only affected by UBC in 0.07% of reported cases [1]. Most of these lesions go unnoticed as they are usually asymptomatic in the absence of pathologic fracture [2]. In 1876, Virchow first described these lesions as cystic structures caused by abnormalities in local circulation [3].

Treatment goals for UBC include reestablishing bone strength, cortical thickness and elimination of the cyst [1,2]. There are various treatment modalities for UBC, which include conservative and surgical treatment [4]. Before surgical intervention, a thorough examination and clinical history must be obtained. Plain film radiography is usually sufficient for visualization and diagnosis of bone tumor [1]. Computed tomography (CT) or magnetic resonance imaging (MRI) should be obtained to evaluate the extent, size, and character of the tumor for treatment planning [2].

In this report, we present an unusual case of an adult female patient who was diagnosed on standard radiographs with an incidental finding of a large cystic lesion occupying the metaphyseal-diaphyseal region of the distal tibia. With the aid of CT, the lesion was further evaluated and then surgically treated with curettage and filled with allogenic bone chips and demineralized bone matrix (DBM).

Case Report

A 50-year-old female presented to the clinic with the chief complaint of bilateral plantar heel pain. Plain films were obtained and a large cystic lesion was noted to the patient's right distal tibia (Figure 1). The patient denied any inciting event. Upon physical exam, there was mild dull pain at the end range of dorsiflexion to the right ankle, but was otherwise unremarkable. Computed tomography (CT) of her right ankle was obtained to further evaluate the cystic lesion.
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CT results showed a large, multiseptated, lucent, expansile bone lesion with thin medial cortex in the central medullary canal in the metaphyseal-diaphyseal junction of the distal tibia. The lesion measured 3.9 x 4.1 x 6.8 cm (Figure 2). The patient was booked for surgical excision and curettage of the right distal tibia bone cyst with insertion of allogenic bone graft, DBM, and included a biopsy.

The patient was brought into the operating room and placed on the operating room table in the supine position. General anesthesia was administered. The right lower extremity was prepped, marked and draped in the usual aseptic manner and a pneumatic thigh tourniquet was then inflated to 325 mmHg.

An anterior incision 8 cm in length, just medial to the tibialis anterior tendon, was made overlying the anterior ankle which allowed direct visualization of the distal tibia. Four drill holes were made in the anterior tibia outlining the planned cortical window which measured 4 cm x 2 cm. The window was cut and removed in one piece revealing the contents of the cyst which contained multiple osseous septa with both hard and soft bone (Figure 3). The entire area of the bone cyst was curetted and excised. Complete cyst excision was confirmed with fluoroscopy and direct visualization. It was noted that there was a large posterior portion of the cyst tunneling 2 cm proximally and inferiorly from the cortical window. Following intramedullary debridement, the cortex remained intact on all sides with no evidence of fracture. All the material was removed from the cyst and was sent to pathology. The cavity of the cyst was
irrigated and 89% phenol was applied to the entire bone cyst area.

Figure 5 Histological slides displaying fragments of sclerotic trabecular bone, consistent with unicameral bone cyst (original magnification 10x and 40x, hematoxylin and eosin).

Due to the large defect post-curettage, the cavity was filled with a combination of crushed allogenic bone chips and DBM. The cystic cavity was completely packed and the bone window was replaced and tamped into position. To prevent displacement of the cortical window, a 7-hole 1/3 tubular plate was placed anteriorly with a distal bend to fit the contour of the tibia. Alignment of the plate was confirmed both visually and with fluoroscopy. Following this, two 3.5 non-locking cortical screws were placed proximal, and one 3.5 non-locking cortical screw was placed distally (Figure 4). The surgical site was flushed with copious amounts of antibiotic solution and closure was completed. Following the procedure, the patient was placed in a well-padded, bivalved, below-the-knee cast and instructed to remain non-weightbearing with the use of crutches. She was prescribed hydrocodone for pain and aspirin 325 mg twice daily to be taken for deep vein thrombosis prophylaxis. The pathology specimen revealed fragments of sclerotic trabecular bone with spindle cells, most consistent with unicameral bone cyst (Figure 5).

Figure 6 Anteroposterior and lateral foot view of patient at twelve months postoperatively highlighting complete consolidation of cortical window.

The patient continued to present to the clinic on a regular basis for postoperative evaluation and serial radiographs. On the first two postoperative appointments (week 1 and 3), the patient’s visual analog pain score (VAS) was 2 out of 10. Radiographs showed incorporation of bone graft material and the patient was allowed to partial weight bear as tolerated. By the patient's third postoperative appointment (8 weeks), the patient’s VAS score was 0 out of 10. Radiographs were taken, revealing consolidation of the cortical window in the right tibia with no recurrence of bone cyst. At this time, the patient was transitioned into normal shoe gear and sent to physical therapy with goals of decreasing edema, increasing range of motion, and increasing strength. At the patient’s one-year follow-up, radiographs were taken revealing no recurrence of bone cyst and the patient remained asymptomatic and had no limitations on full activity (Figure 6).

Discussion

Multiple theories have been postulated for the pathogenesis of unicameral bone cysts. Blockage in the venous drainage is the most favored mechanism which occurs in rapidly growing and remodeling cancellous bone [2, 3, 4]. This increased pressure may lead to the resorption of bone. Others have postulated that there could be a disturbance in bone growth, intramedullary hemorrhages secondary to trauma that do not completely resolve, degenerative phase of benign tumor, and osteomyelitis [2,10]. Cyst fluid analysis has shown increased levels of prostaglandin e2, II.1 beta, and proteolytic enzymes
which could lead to bone resorption and cyst formation [5].

Treatment for UBCs is either observation or surgical intervention [2]. Reported surgical treatment includes medullary decompression with cannulated screws or intramedullary nails, steroid injections, or curettage with autograft or allograft [1, 2, 5-8]. Scaglietti, et al., were first to describe percutaneous injection of methylprednisolone acetate for UBC treatment in 1974 with only 24% healing rate after one injection [8]. Several authors have found satisfactory results with healing rates between 50-90% with steroid injections, but most reported that several procedures were necessary for cyst consolidation [1, 6]. It has been reported that steroid injection may prevent the pro-inflammatory cytokine activity that leads to cyst formation as well as to relieve cyst pressure due to trepanation [1, 8].

Other injectable materials including bone graft and demineralized bone matrix (DBM) have been evaluated. Lokicic, et al., in 1996 was the first to report the use of autologous bone marrow injections for UBC treatment in children with 100% success rate [9]. Other studies have used bone marrow graft in combination with DBM [7, 11]. These studies attribute the high success rate due to bone marrow's osteoprogenitor cells in combination with the osteoinduction and osteoconduction properties of DBM [7, 11]. Multiple authors have evaluated the effectiveness of DMB alone as an injection with high success rates [6, 7]. Cho, et al., in 2012 evaluated twenty-five patients with a unicameral bone cyst who were treated with intramedullary decompression followed by grafting of demineralized bone matrix [2]. They used a small incision to create a cortical window to allow for curettage and decompression of the cyst and subsequently injection a mixture of allograft bone and DMB. Their success rate was 100% with a mean healing time of 6.6 months. Two patients required a second procedure, which they determined the initial amount of bone void filler was not enough to fill the entire space. The authors concluded that mixture of bone graft material, DBM, and completely filling the cyst was successful with satisfactory results [2].

Due to the size of our patient's cyst, we used a longer incision and created a cortical window over the anterior distal tibia. This approach made it possible for visualization and use of curettes to completely remove all cyst material and to obtain a biopsy. This approach may appear more aggressive than other reported procedures, but because of the large size of the cyst and age of the patient, it was necessary for visualization and complete curettage, biopsy, and filling of the cyst with allograft and DBM. Similar techniques have been reported from multiple authors with healing rates greater than 90% [1, 6, 7]. The surgical curettage is necessary to resolve the cyst, but also allows for biopsy, which is necessary to rule out malignancy such as Ewing's sarcoma and osteosarcoma because they also present as cystic lesions radiographically [5].

Unicameral bone cysts are usually incidental finding with many factors that could contribute to their formation. Although there is no standardized treatment for UBC, the goal of treatment is to prevent pathological fracture and, in children and adolescents, to prevent skeletal deformities during growth. Surgical procedures such as curettage with allogenic bone graft have been shown to be successful treatment with low rates of recurrence [1, 6, 7]. The surgical curettage is necessary to obtain tissue for biopsy to determine pathology including Ewing's sarcoma and osteosarcoma [5]. In this report, we present a patient who was treated successfully with surgical curettage and allogenic bone graft for a distal tibia UBC with no cyst recurrence after one year. Long-term clinical follow-up is necessary for observation of potential cyst recurrence.

References

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