

A Case of Bilateral Unicameral Bone Cysts

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Abstract

Unicameral bone cysts typically occur in the metaphyseal-diaphyseal regions of long bones within the first 10 years of life. These unicameral bone cysts are considered a benign self-limiting disease that spontaneously heals after skeletal maturity. They are relatively common in the humerus, femur, and tibia; yet rarely develop in the calcaneus. Incidence of bilateral cysts is extremely rare. The case presented is of a 38-year-old female who suffered from bilateral unicameral bone cysts of the calcaneus that required surgical intervention.

Keywords Calcaneal bone cyst; Calcaneal stress fracture; Simple bone cyst

Background

Simple bone cysts, also known as unicameral bone cysts are vague in etiology [1]; however intraosseous lipomas, myxomas, and vascular abnormalities have been proposed as predisposing factors [2]. Unicameral bone cysts typically occur in the metaphyseal-diaphyseal regions of long bones within the first 10 years of life [3]. These unicameral bone cysts are considered a benign self-limiting disease that spontaneously heals after skeletal maturity. These cysts are relatively common in the humerus, femur, and tibia; yet rarely develop in the calcaneus [4]. Of 429 unicameral bone cysts reported, only 18 [4%] were reported in the calcaneus [5-9]. Unicameral bone cysts in the calcaneus are less susceptible to pathological fractures that are typically associated with cysts occurring in the femur and humerus [10]. Since the cysts are usually located in the anterior aspect of the calcaneus, physiological stresses are lower [2,11,12]. Calcaneal cysts, like all unicameral bone cysts, are typically asymptomatic with incidental diagnosis as the result of another injury. Most calcaneal unicameral bone cysts can be treated conservatively with few needing surgical intervention [4]. In addition the incidence of multiple cysts within individuals is extremely rare. Of 36 reported cysts, only 3 were bilateral [4]. Of those 3 reported bilateral cysts, symptom resolution was achieved through conservative treatment. There are no data to support bilateral unicameral bone cysts with surgical resolution.

Case Study

A 38-year-old active female, avid runner [36 to 40 miles per week], presented with acute left, medial heel pain. Twenty-five months prior to this incidence of left, medial heel pain; patient was diagnosed with unicameral bone cyst of the right calcaneus. The initial unicameral bone cyst of the right calcaneus was diagnosed when the patient was 36 years old. Patient presented with pain in her right foot that spanned from lateral mid-foot to the rear foot and pain was exacerbated during running. Patient history revealed reoccurring stress fractures of the lateral mid-foot for a period of seven years. Prior to unicameral bone cyst diagnosis, the initial orthopedic examination during the patient's latest stint with pain revealed a cuboid stress fracture in her right foot.

The patient was immobilized in a Cam Walker [DJO Global, Inc., Vista, CA] for six weeks. Following six weeks of immobilization the patient attempted to run and noted the same severity of pain as reported prior to immobilization. Magnetic resonance imaging [MRI] revealed pathological marrow signal intensity in the mid portion of the calcaneus that extended laterally near the site of pain symptoms. A computed tomography [CT] scan was performed to compare with the MRI. The CT displayed a 1.8 cm smoothly marginated area of soft tissue radiodensity that had several coarsened trabeculae extending through the site of marrow edema in the mid portion of the calcaneus (Figure 1). Diagnosis revealed a unicameral bone cyst with a stress reaction in the mid-body of the right calcaneus. Due to the pain and activity level of the patient, surgical resolution of the cyst was recommended and elected.

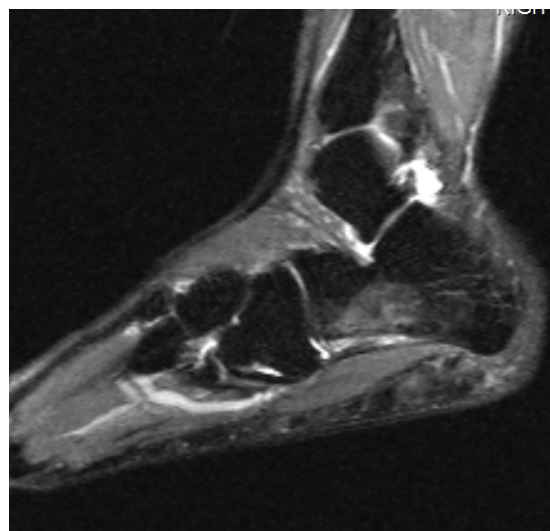


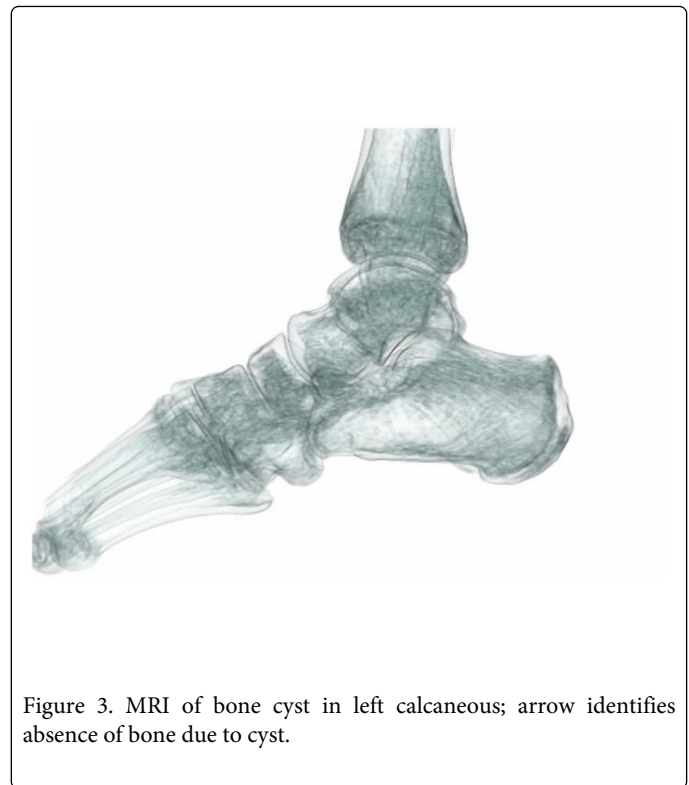
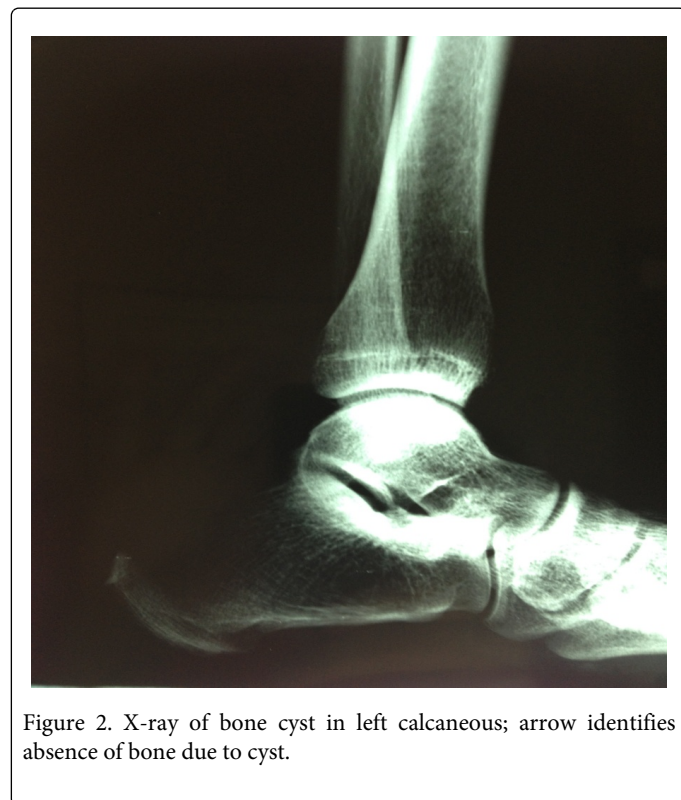
Figure 1. CT of lateral calcaneal marrow intensity. The arrow indicates the area of increased bone marrow intensity.

Surgery was performed to evacuate the bone cyst with curettage and then pack with osteocele allographic bone. Surgical procedure

consisted of an 8 cm lazy S linear incision along the level of pathway of the peroneal tendons on the lateral aspect of the right calcaneus proximal to the calcaneal cuboid joint. Once the cystic structure was identified, a window was made on the lateral aspect of the calcaneus to facilitate curettage and evacuation of the contents of the cyst. The window was freed from the restraining bone and removed from the surgical site. The cystic structure was noted to be approximately 3 cm in depth and 1.5 cm in diameter with several small branches off the cystic structure protruding into other areas of the calcaneus. Two milliliters of osteocele allograft material was prepared, irrigated, and then compressed into the cystic structure to fill the defect created by curettage. After the cyst was packed, the window was reapplied and press fitted into the previously made window site on the lateral calcaneus.

Following surgery, the patient was non-weight bearing and placed on crutches with a soft splint for three weeks. Three weeks post-operative, the patient was weight bearing in the Cam Walker [DJO Global, Inc., Vista, CA] without the assistance of crutches. Six weeks post-operative, the patient was released to walk and resume physical activity without restriction. Within four weeks [10 weeks post-operative] of physician's release the patient completed a 5K run at pre-injury performance level with no pain.

Patient returned to her running regimen with no complications. Twenty-five months later, patient presented with acute pain at the medial aspect of her left heel. Patient had no history of left lower leg, heel or foot pain or injury. Patient self diagnosed as possible unicameral bone cyst and implemented 4 weeks of conservative treatment. Once conservative treatment did not relieve symptoms, X rays (Figure 2) and MRI (Figure 3) were performed. MRI reported an impression of a pseudo-cyst in calcaneus and not a pathological condition. X-ray revealed a unicameral cyst.



The patient's activity level, current pain status, and the surgical success of her contralateral unicameral bone cyst, led her to elect for surgical evacuation of the unicameral bone cyst. An 8 cm lazy-S linear incision was made on the lateral aspect of the left calcaneus, immediately over the area identified as the area of cystic structure in the bone at the mid calcaneus. A window approximately 1.2 cm² was placed into the lateral aspect of the calcaneus at the point of the presence of the apparent bone cyst. The window was excised and put into sterile saline. The cystic structure appeared to go from the lateral calcaneal wall all the way to the medial calcaneal wall. The area was debrided and irrigated extensively. After debridement, 4.5 mL of Ostocel bone chips by NuVasive were pressed into the defect created by the cyst. After packing the window was put back into place and sutured in place.

Following surgical evacuation with debridement, the patient was placed in a soft splint, non-weight-bearing using crutches for three weeks. At week three, the soft splint was replaced with a Cam Walker [DJO Global, Inc., Vista, CA]. At three weeks the patient was allowed to perform partial weight bearing with crutches. At six weeks the patient was able to ambulate in the Cam Walker without the assistance of crutches. Patient was in Cam Walker for an additional 7 weeks. At 13 weeks the patient began walking and jogging. Eighteen weeks post-operative the patient competed in a 5K run with no pain.

Discussion

The presented patient sustained a rare unicameral bone cyst in her right calcaneus that was finally diagnosed after episodes of repeated stress reactions spanning more than seven years. Surgical evacuation with debridement of the cyst was performed and the patient was able to return to running 10 weeks post-operative. Then 25 months after the surgical procedure for the right unicameral bone cyst, the patient

experienced symptoms of a pseudo fracture of her left calcaneus. Surgical evacuation with debridement was performed to evacuate the cyst and after 13 weeks of assisted mobilization she began running again.

Despite the continual research into unicameral bone cysts, the etiology is illusive [13]. Unicameral bone cysts typically occur in the metaphyseal-diaphyseal regions of long bones, within the first 10 years of life [3], while this patient was in her later 30s. Though unicameral bone cysts are more common in the femur and humerus of youth, retrospectively it has been reported that calcaneal unicameral bone cysts are not as rare as once believed in adults [4]. Average age of diagnosis has been 15 years of age [14,15] with diagnosis after 17 years the location in the calcaneus is more common [16] with the incidence of bilateral occurrence being rare [14,17]. Typically unicameral bone cysts are asymptomatic until pathological stress fracture occurs [13] and are more prevalent in males than females [18]. In addition, of the reviewed calcaneal cases [4], most have been asymptomatic with no surgical treatment. In the current case the cyst in the right calcaneus was revealed after repeated episodes of stress reactions within the foot over a period of seven years, while the left unicameral cyst became symptomatic when an acute pseudo fracture occurred. Only 14% of unicameral bone cysts reported have been in the calcaneus with rare bilateral occurrence [4,14,17]. This is the first case to present not only a case of bilateral unicameral bone cysts in the calcaneus, but it also the requirement of surgical intervention bilaterally.

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