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Case Report

Treatment of a calcaneal unicameral bone cyst by percutaneous CT-guided cement injection using a double-needle technique: A case report ^{☆,☆☆}

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ABSTRACT

Simple bone cysts (SBCs) are benign cavitory lesions that most commonly affect adolescent males in the first 2 decades of life. They are mainly asymptomatic but can manifest with pain or pathological fractures. Despite numerous proposed methods for managing calcaneal SBCs, the optimal approach toward these lesions remains controversial. Herein, we report a case of a 16-year-old girl with a calcaneal SBC. On local examination, tenderness was the only noteworthy sign. In an outpatient setting, under conscious sedation, 2 interosseous needles were simultaneously inserted into the cyst under the guidance of CT fluoroscopy. Without aspiration, a radiopaque bone cement mixture was injected into the cyst from 1 needle until serosanguineous fluid efflux from the second needle ceased. Over a 2-year follow-up period, the patient recovered without any complications. This novel technique has the potential to be used as a feasible and minimally invasive approach in the management of symptomatic unicameral calcaneal bone cysts.

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Abbreviations: SBCs, Simple bone cysts; UBCs, Unicameral bone cysts; CT scan, computed tomography scan; MRI, magnetic resonance imaging; DLP, dose length product.

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Introduction

Simple bone cysts (SBCs) are benign cavitary lesions within the metaphyseal region of bones [1]. They can be unicameral (single chamber) or multiloculated with partial septations [2]. Unicameral bone cysts (UBCs) are well-defined cystic cavities filled with serous or serosanguineous fluid lined by a fibrovascular connective tissue membrane [2,3]. Although the precise etiology of UBCs remains elusive, some researchers have suggested that venous drainage obstruction may be responsible for intramedullary interstitial fluid accumulation and cystic formation [4,5]. UBCs most commonly affect children and adolescent males (with a 2-3:1 male-to-female ratio) in the first 2 decades of life. They primarily involve metaphyseal regions of the proximal humerus and femur (accounting for up to 80% of cases) and are relatively uncommon in the calcaneus bone [5–7]. Calcaneal UBCs mostly arise from the anterolateral aspect of the bone, far from the area where most body weight is transmitted [8]. Although these lesions are mainly asymptomatic, they can present with pain or pathological fractures due to structural weakness [3]. The patient's age has the most significant impact on disease presentation [9]. Plain radiography yields high diagnostic accuracy and is considered the modality of choice for imaging [10]. Even though computed tomography scan (CT scan) and magnetic resonance imaging (MRI) add little to the diagnosis, they can help exclude other entities that can mimic a simple bone cyst [5,11]. Moreover, MRI can be combined with plain radiography to confirm the presence of fluid within a cyst and demonstrate its aggressive features [5].

Heretofore, various methods have been proposed to manage UBCs, including observational management, intralesional steroid injection, percutaneous/open curettage with or without bone grafting, percutaneous injection of allogenic demineralized bone matrix, and cannulated pin/screw insertion [12–19]. However, minimally invasive techniques are far more desirable due to the higher healing rate, lack of recurrence, shorter recovery time, and preservation of the periosteum, muscles, and blood supply.

The present study describes the successful management of a calcaneal UBC with a new minimally invasive technique utilizing 2 interosseous needles for cement injection into the cyst without aspiration.

Case report

A 16-year-old girl presenting with an 8-day history of pain and swelling over the right hind foot and calcaneal region and difficulty weight bearing was referred to our tertiary center. She had suffered from intermittent mild pain in the right calcaneus since age 3, aggravated by running and relieved by rest and cold compression. Her pain had become constant 3 weeks before coming to the hospital. The pain had increased over an 8-day period during which the patient had been playing volleyball most of the day. There was no history of trauma to the calcaneal region or family history of other pathological conditions.

On physical examination, the patient appeared relatively healthy and well-nourished. She did not suffer from any other medical conditions and took no medications. The local examination was unremarkable except for tenderness over the right calcaneal region. There was no ankle edema or muscle atrophy, and the limbs appeared symmetric with a fully active and passive range of motion. An MRI of the right ankle and foot demonstrated a homogeneous low T1 signal and a hyperintense T2 signal, suggestive of a simple bone cyst, which measured approximately $18.2 \times 21.5 \times 14$ mm (L×W×H), with an estimated volume of 2.86 ml in the anterior process of the calcaneus (Fig. 1).

Laboratory tests consisting of complete blood count, erythrocyte sedimentation rate, levels of calcium, phosphate, serum albumin, renal function tests (blood urea nitrogen creatinine), urinalysis, coagulation tests (partial thromboplastin time, prothrombin time, international normalized ratio) were analyzed, and the results were all within normal limits.

Based on the clinical and radiological findings, a simple bone cyst was diagnosed, and radiological intervention was planned for the patient. Written informed consent was obtained from the patient and her family before the procedure. After conscious sedation with 1 mg midazolam, 1 mg/kg ketamine, and 1mg/kg propofol and local anesthesia with 5 CC of lidocaine 2%, a VerteStable cannula was inserted percutaneously into the cyst under the guidance of CT fluoroscopy to inject cement. An Ostycut needle (16G) × 100 mm (Angiomed/Bard, Karlsruhe, Germany) was also inserted percutaneously to allow the efflux of serosanguineous fluid from the cyst (Fig. 2B).

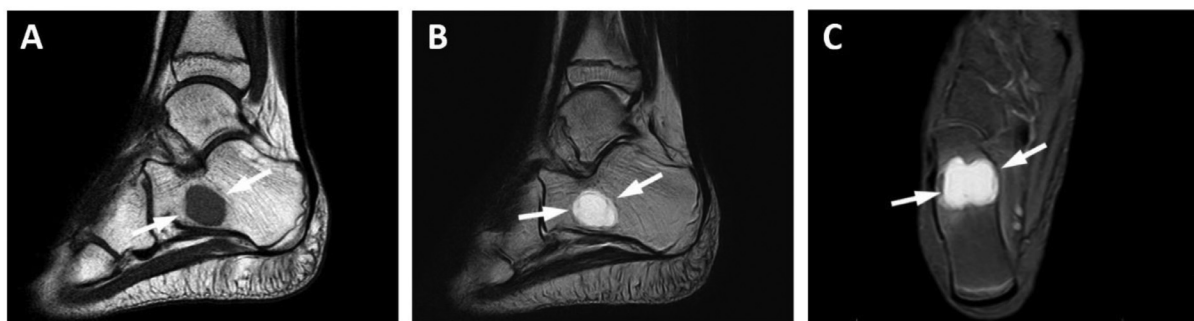


Fig. 1(A–C) – MRI indicated a homogeneous intraosseous sharply margined lesion in the calcaneus (arrow), (A) hypo-signal in the sagittal T1-weighted image, (B) intensity hyper-signal in the sagittal T2-weighted image, (C) axial fat-suppressed T2-weighted image, indicating exclusively fluid content.

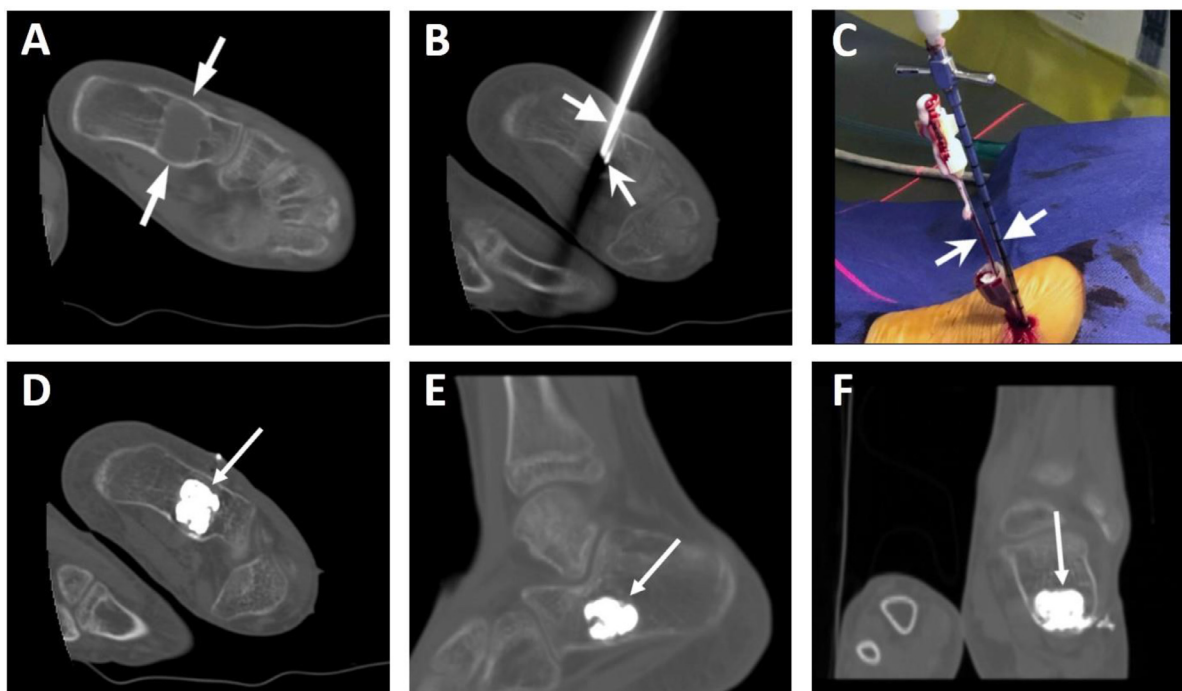


Fig. 2(A) – Axial noncontrast CT image of the right hind foot shows the intraosseous sharply margined calcaneal lesion (arrow). **(B, C)** Under CT fluoroscopy guidance, a VerteStable cannula (thick arrow) and an Ostycut needle (wide arrow) were inserted percutaneously into the cyst for the injection of a radiopaque bone cement mixture. The radiopaque bone cement mixture was injected through the VerteStable cannula until there was no more serosanguineous fluid efflux from the Ostycut needle, and only cement mixture efflux was observed. **(D–F)** The actual noncontrast CT image and reconstructed sagittal and coronal images show a complete replacement of cyst content with radiopaque cement material (thin arrow).

Unlike previous techniques, aspiration was only performed to reveal serosanguineous fluid content. Subsequently, through a VerteStable cannula, the radiopaque bone cement mixture (BonOs Inject 1×24, OSARTIS GmbH, Munster, Germany) was injected into the cyst. The injection from 1 needle was immediately associated with the efflux of serosanguineous fluid from the Ostycut needle. Cement injection was continued until there was no more serosanguineous fluid efflux from the second needle, and only cement efflux was observed (Fig. 2C). The procedure was performed under fluoroscopy control to warrant a complete replacement of cyst content with cement material (Figs. 2D–F). The system was kept closed to prevent air embolism. After filling the cyst completely, the needles were removed, and a dressing was placed over the injection sites. The duration of the procedure was estimated to be 15 minutes, and the total dose length product (DLP) and smart view DLP were 577.9 mGy*cm and 13 mGy*cm, respectively. After the procedure, the patient's hemodynamic status was stable, and she was discharged the next day. No weight-bearing was recommended for 1 month postoperatively, and full weight-bearing without support was initiated 1 month after the procedure. The response to intervention was assessed by the relief of symptoms. The patient experienced itching and warmth at the procedure site immediately after the intervention. She returned to her country and didn't come for her scheduled follow-ups, therefore there is no follow-up imaging. On a phone call 3 weeks after the procedure, the patient reported pain with weight-

bearing activities and limping after prolonged standing. Phone call follow-ups were conducted approximately every 6 months for 2 years after the procedure. There were no reports of itching or warmth at the procedure site or complaints of limping. The patient had experienced a few scattered episodes of mild pain after prolonged standing, relieved with rest.

Discussion

UBCs are non-neoplastic fluid-filled cavities that account for up to 3% of primary bone lesions and solely arise in children and adolescents [2,6,7,10]. Even though the exact cause of UBCs' formation has not been elucidated, many theories have been proposed, such as local disruption in bone formation, small nests of synovial cells trapped in bones, and venous drainage blockage [4,5,20]. The most widely accepted theory is venous drainage blockage due to observations showing a slight increase in the involved bone's internal pressure compared to the bone marrow's normal pressure and oxygen's lower partial pressure in the cyst fluid compared to arterial or venous blood [20]. UBCs are mainly diagnosed by clinical presentations and plain radiography [6]. Although CT scan and MRI add little to the diagnosis, they can help eliminate other entities that mimic a simple bone cyst [1,11]. UBCs of the calcaneus primarily affect the anterolateral aspect of the bone,

which is far from the area where most body weight is transmitted [8].

Various strategies range from nonoperative management, intralesional steroid injection, percutaneous or open curettage with/without bone grafting, percutaneous injection of allogenic demineralized bone matrix, more invasive procedures such as cannulated pin/screw insertion, and surgical procedures have demonstrated acceptable efficacy. However, the optimal management of these lesions remains an issue of debate. Nonoperative management with close follow-up is recommended in incidentally diagnosed patients with no risk of pathological bone fractures. Spontaneous resolution of heel pain has been reported in 1%-2% of patients managed nonoperatively [4,19].

Intralesional steroid injection was first introduced as a successful management strategy for simple bone cysts. Despite the reported outcomes, recent studies on calcaneal UBCs have demonstrated a high recurrence rate even after multiple steroid injections [13,21]. Scaglietti et al. reported only 24% cyst healing after a single injection and incomplete recovery even after multiple injections. Glaser et al. [22] performed nine steroid injections on 6 patients, and curettage combined with bone grafting on 9 patients. All of their patients were asymptomatic. The study reported among cysts treated with steroid injections, there was zero percent healing and the persistence of the 2 cysts. Conversely, complete healing without recurrence occurred in cases who underwent curettage with bone grafting after a minimum follow-up period of 12 months. Moreover, a few studies have suggested that steroids have minimal effect on resolving the cyst, and the mechanical effect of punctures, which helps normalize local circulation, is the key factor. Percutaneous or open curettage, which can be performed with or without bone grafting, is the traditional method for managing symptomatic calcaneal UBCs. Although open curettage with bone grafting has demonstrated significant improvement in heel pain, it has been associated with a high recurrence rate in cases where the graft had been incompletely packed [16]. Innami et al. [12] performed endoscopic curettage with percutaneous injection of calcium phosphate to treat symptomatic calcaneal UBCs. Their findings showed the complete resolution of pain and radiological healing in all patients after a 3-year follow-up. Aiba et al. [14] performed endoscopic curettage on 6 calcaneal UBCs and reported complete recovery without cyst residue after a median of 4-month follow-up.

Abdel-Wanis et al. [18] used minimal curettage, multiple drilling, and continuous decompression as an alternative to autografting in children. Due to the patients' young age, harvesting a large volume of autografts can be difficult. On the other hand, the risk of disease transmission by autografting should not be overlooked, no matter how small is. This study reported complete radiographic healing in all patients and total pain relief without recurrence in 59% of cases. Other methods, such as a cannulated screw placed for continuous decompression, have demonstrated lower recurrence rates, higher healing rates, and a significant reduction in heel pain following decompression. Saraph et al. [23] treated nine calcaneal UBCs with continuous decompression using cannulated screws, among whom 8 cases showed complete healing and one showed a residual region on imaging with complete

pain relief after a minimum follow-up period of 24 months. The only complication reported in the study was irritation at the screw insertion site observed in 1 patient, which led to the early removal of the screw. Shirai et al. [24] managed 23 simple bone cysts in the calcaneus by using cannulated hydroxyapatite pins, which were used to achieve continuous decompression. After a mean follow-up period of 5 months, their results showed complete healing in all patients. In another study, Park et al. [15] compared open chip allogenic bone grafting with percutaneous injection of bone powder. Complete healing was reported in 70% of open chip allogenic bone grafts and 50% of cases percutaneously injected with bone powder after a mean follow-up of 49.4 months. Persistent cysts were reported in 15% of patients treated by open chip allogenic bone grafts, and 20% of those managed by percutaneous injection of bone powder.

In this study, we treated a calcaneal unicameral bone cyst using a double-needle technique by percutaneous cement injection until the cyst was completely filled. Utilizing 2 needles enabled us to fully drain the cysts with a radiopaque bone cement mixture in a relatively short time (15 minutes) without any complications.

This novel technique has the potential to be used as a feasible minimally invasive approach to manage symptomatic simple bone cysts in the calcaneus. Nevertheless, further studies are required to validate its long-term safety and efficacy with regard to the management of symptomatic calcaneal simple bone cysts.

Availability of data and material

Research data is available upon request. To request the data, contact the corresponding author of the article. The authors declare that they had full access to all of the data in this study and the authors take complete responsibility for the integrity of the data and the accuracy of the data analysis.

Author's contributions

AA, AH, and HG designed the concept of the study. AA and HG defined intellectual content. AA and HG were major contributors to clinical studies. AA, HG, and GA edited and reviewed the final manuscript. AH conducted the literature search. AH analyzed and interpreted data. AA, AH, and GA contributed to data acquisition. HG is the guarantor of the paper. All authors read and approved the final manuscript.

Patient consent

Written informed consent was obtained from the patient and her family prior to the procedure.

Ethics approval

Tehran University of Medical Sciences Hospital's ethical board committee has approved the present study, and it was performed in compliance with the 1996 Health Insurance Portability and Accountability Act (HIPAA).

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