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A Unique Technique for Precise Targeting in Treatment of Rare Bifocal Intraosseous Ganglion Cysts of the Talus: A Case Report and Review of the Literature

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Data Interpretation D
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Conflict of interest: None declared

Patient: Male, 51-year-old
Final Diagnosis: Intraosseous ganglion cyst
Symptoms: Discomfort • pain
Medication: —
Clinical Procedure: Arthroscopy • bone graft
Specialty: Orthopedics and Traumatology

Objective: Rare disease

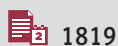
Background: This article presents a rare case of 2 separate intraosseous ganglion cysts of the talus in a 51-year-old man, treated with a unique technique of precise lesion targeting to avoid extensive bone loss and minimize articular chondral injury of the talus.

Case Report: Two separate intraosseous ganglion cysts of the talus were diagnosed in a 51-year-old man with chronic ankle pain. A single straight-line incision with an entry point through the talonavicular joint was created to spare the precarious blood supply of the talus network. The 2 distinct subchondral lesions were approached under fluoroscopic control for curettage and autologous bone grafting using the anterior cruciate ligament tibial guide in a pair-of-compasses fashion. In almost 5 years of follow-up the patient has been asymptomatic. Magnetic resonance imaging has revealed no signs of degenerative changes in the ankle or the talonavicular joint, and the intraosseous edema has almost disappeared.

Conclusions: To the best of our knowledge, this case is the first report of 2 distinct intraosseous ganglion cysts of the talus. We recommend the precise targeting technique used in our case for treating intraosseous talar lesions with intact articular cartilage.

Keywords: Adult • Bone Cysts • Ganglion Cysts • Histological Techniques • Teaching • Talus • Teaching

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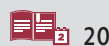
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Background

Intraosseous ganglia are benign cystic lesions, first described by Fisk in 1949 [1]. Many names have been used for this type of lesion, such as subchondral, intraosseous mucoid, or synovial bone cyst, mainly due to the differences in the interpretation of their etiology [2]. The peak incidence of intraosseous ganglion is in the fourth and fifth decades of life, and it is rare in children [3]. Intraosseous ganglia contain a gelatinous material with a histopathologic appearance similar to that of a soft-tissue ganglion, and they are rarely reported lesions. They have a tendency to occur in the metaphysis of long bones of the lower extremity, such as distal tibia; however, the wrist, femoral head, medial malleolus, and carpal bones are other well-recognized sites [4,5]. Patients with osseous ganglia experience intermittent pain that frequently increases with activity. Patients can also present with neurologic dysfunction or even articular fractures. In as many as 40% of patients, these lesions may be asymptomatic [6]. The literature contains only a few case reports of ganglion cysts in the talus [7-12].

We present a case of 2 separate intraosseous ganglion cysts of the talus in a 51-year-old man that were treated with a unique precise surgical targeting technique. To the best of our knowledge, this is the first reported case of distinct bifocal ganglion cysts of the talus. Additionally, we describe the chronologic

response to previous treatments, the diagnostic workup, the histological characteristics of the lesion, and a new precise targeting technique to address both lesions that does not expose the ankle joint and preserves the articular cartilage of both distal tibia and talonavicular joints. The patient provided informed consent for the data from his case to be submitted for publication.

Case Report

In November 2013, a 51-year-old man presented to our hospital with chronic pain in his left ankle joint. There was a subtle history of injury 1 year ago. The pain had mild to moderate intensity and was located on the anterolateral aspect of the ankle. It was associated with weight-bearing, aggravated on activity, and relieved on rest. The patient was able to perform activities of daily living but further activity caused great discomfort. There was no history of fever or any similar complaint in another joint. On palpation, there was minimal joint effusion. Ankle joint movements were within normal limits, without locking or clicking sensation or any signs of ankle instability. No muscle atrophy, erythema, redness, or signs of inflammation around the ankle joint were observed. Neurovascular examination of the foot was normal. Plain radiographs of the ankle joint revealed a well-defined oval radiolucent lesion on the lateral aspect of the talar dome (Figure 1A, 1B). The patient was treated conservatively with cast

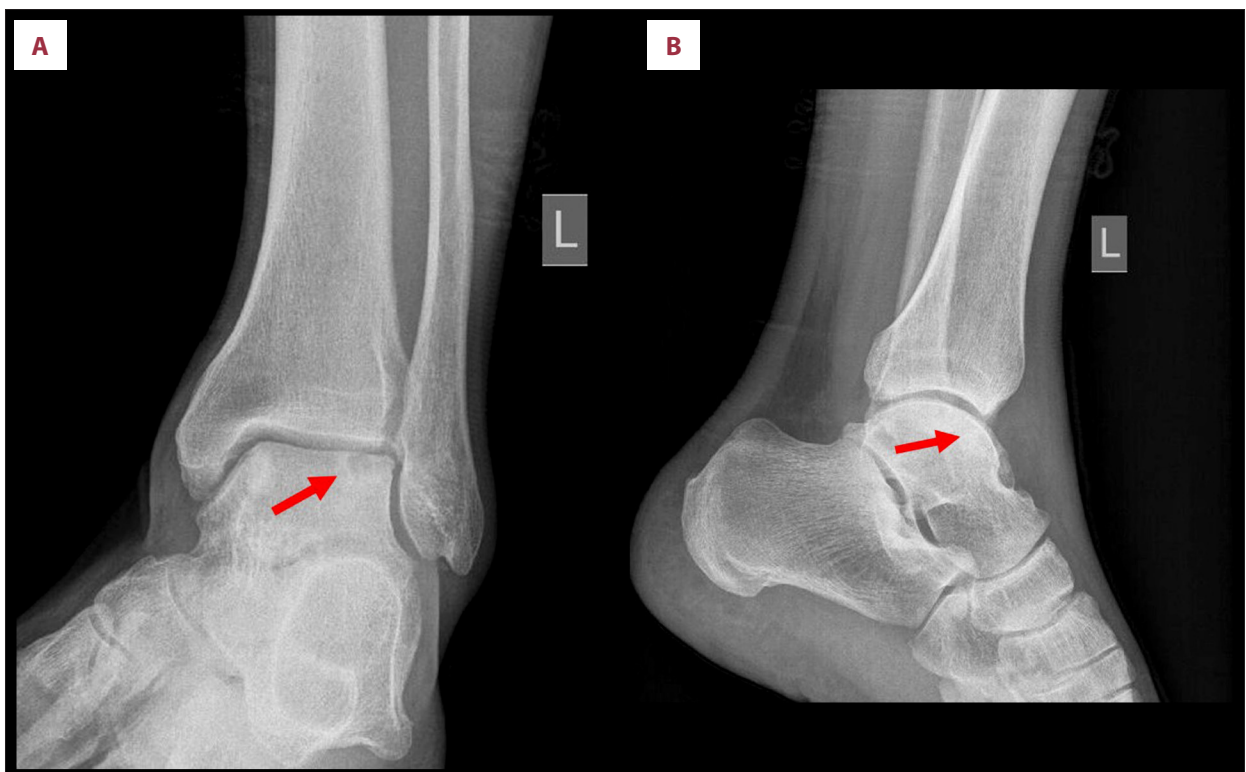


Figure 1. Anteroposterior (A) and lateral (B) radiographs of the left ankle revealing a radiolucent lesion (red arrow) in the lateral aspect of the talar dome.

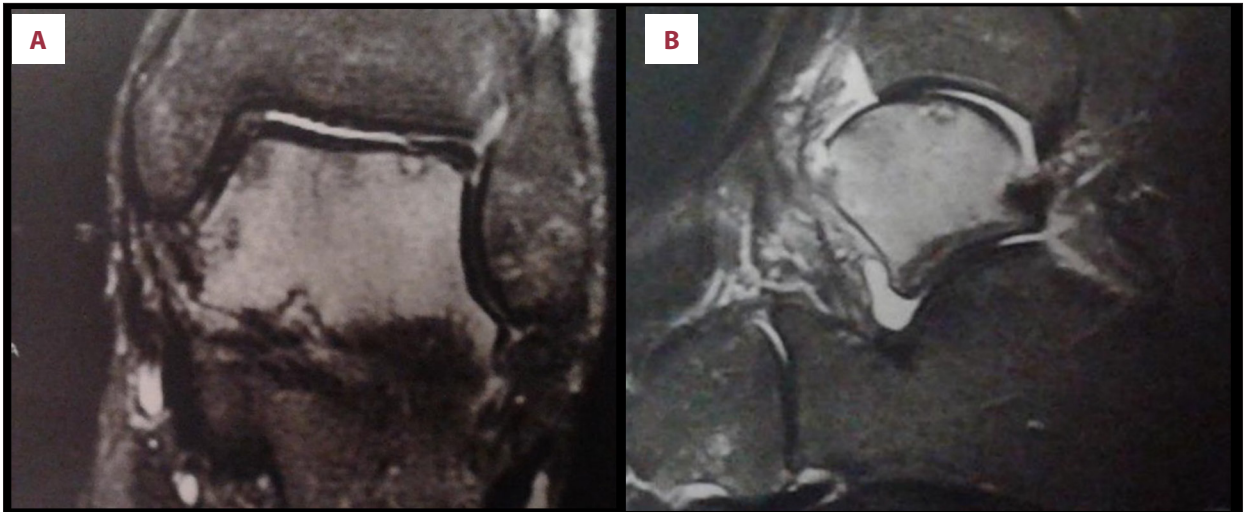


Figure 2. T2-weighted fat-saturated (A) coronal and (B) sagittal magnetic resonance images showing extensive bone marrow edema of the talus, subchondral lesions, and joint effusion.

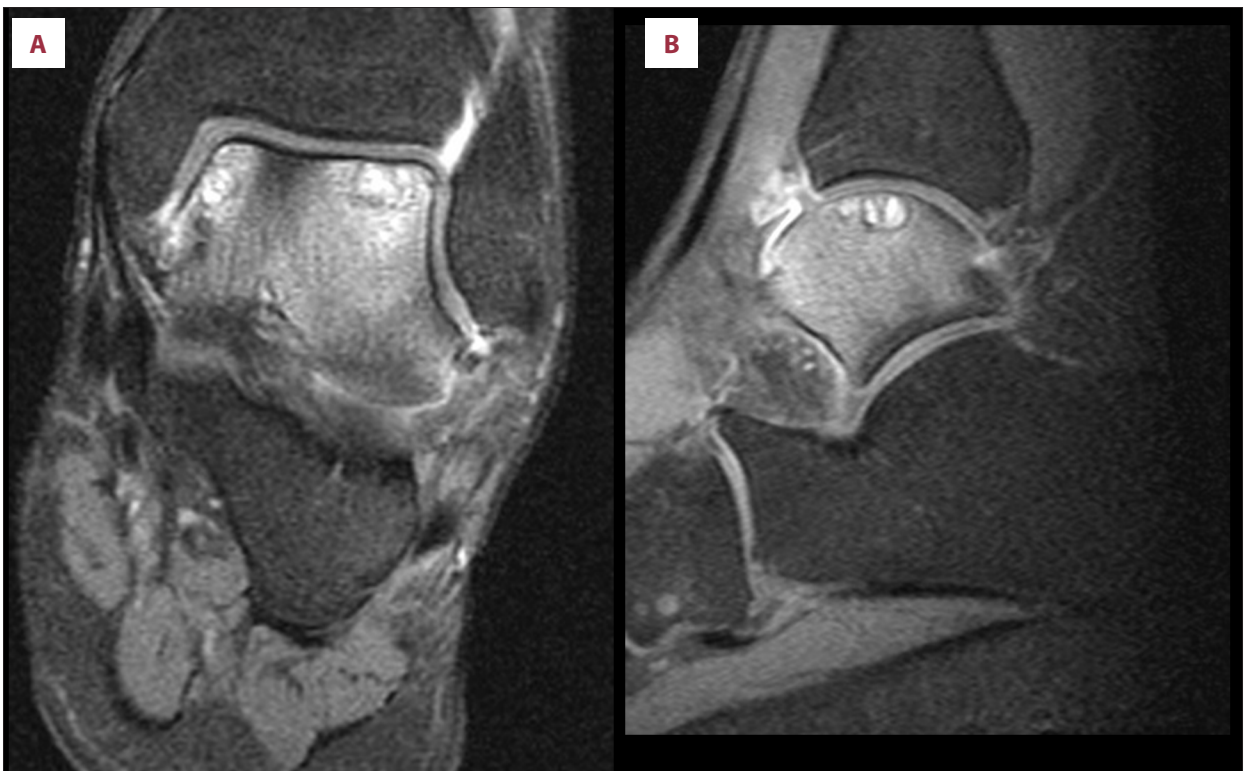


Figure 3. T1-weighted fat-saturated (A) coronal and (B) sagittal consecutive magnetic resonance images with gadolinium contrast 1 year postoperatively illustrating subchondral cystic lesions and extensive bone marrow edema of the talus and synovitis.

immobilization and nonsteroidal anti-inflammatory medication for 1 month, followed by partial weight-bearing and physiotherapy. No significant pain improvement was noticed and a magnetic resonance imaging (MRI) examination of the ankle was obtained, which showed a subchondral lesion at the anterolateral aspect of the talar dome with significant joint effusion and bone marrow edema of the whole talus (**Figure 2A, 2B**).

Because the patient was still symptomatic, an ankle arthroscopy was performed. Intraoperatively, a Ferkel-Cheng grade C articular lesion was found at the anterolateral aspect of the talus [13]. Debridement of the fibrillated cartilage of the talar dome and partial synovectomy was performed. Postoperatively, the patient was instructed to remain non-weight-bearing for 6 weeks but to have full range of motion of his ankle. He had

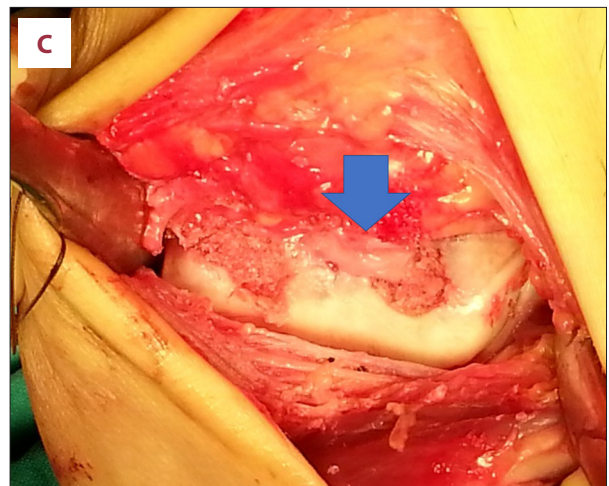
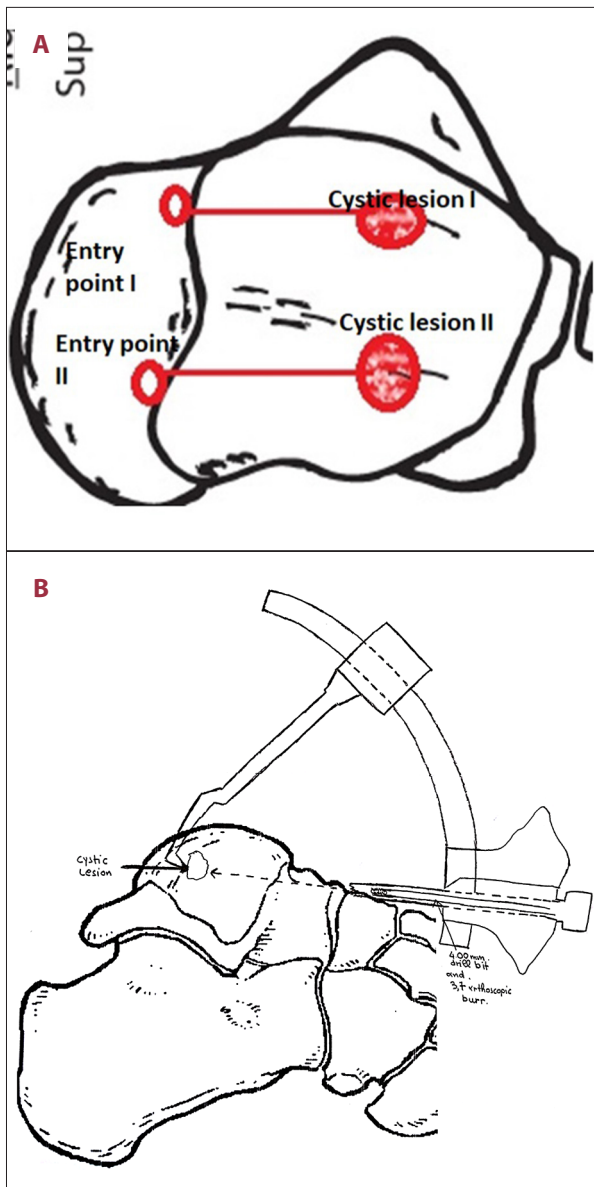


Figure 4. Schematic illustration of our surgical approach. (A) Two entry portals in the medial and lateral side of the talus were created to avoid branches of the anterior tibial artery. (B) Usage of the anterior cruciate ligament tibial guide. (C) Thick gelatinous fluid was extruded from both cysts (blue arrow).

accommodate the handle of the anterior cruciate ligament [ACL] tibial aiming device), 2 small entry points just under the end of articular cartilage of the body of the talus were created, avoiding branches of anterior tibial artery (**Figure 4A-4C**).

Under fluoroscopic guidance and using the ACL tibial aiming device, the ACL guide wire was inserted toward the anterolateral lesion and then, using a second entry point 1.5 cm medially, toward the anteromedial talar dome lesion. Upon the lesions being reached, a thick gelatinous fluid was extruded from both cysts with the use of a 4-cm cannulated drill bit, which confirmed their correct location (**Figure 4C**). Complete curettage was performed using a 3.7-cm soft tissue arthroscopic saver. The cavity was filled with autogenous cancellous bone graft obtained from the left iliac crest.

significant pain improvement for 6 months after surgery and returned to work without restrictions.

However, his symptoms recurred at the 1-year follow-up. A second MRI scan was obtained, revealing diffuse synovitis of the ankle joint, extensive edema, and 2 well-defined intraosseous cystic lesions in the anteromedial and anterolateral talar dome (**Figure 3A, 3B**). Blood examination, including total lymphocyte count, erythrocyte sedimentation rate, C-reactive protein, rheumatoid factor, and serum uric acid, were unremarkable.

The patient was treated surgically using a single 6-cm longitudinal incision through the upper one-fifth of the talonavicular joint to avoid damage to the precarious blood supply network of the talus. After a slight subluxation of the joint (to

All the extruded material for each lesion separately was sent for pathological examination and tissue culture. All cultures were negative, excluding low-grade bone infection. Histological examination of the extruded fluid revealed a loose connective tissue with foci of myxoid degeneration and the presence of small newly formed vessels. No synovial lining, hemosiderin-laden macrophages, or cholesterol clefts were seen. The final diagnosis was ganglion cysts (**Figure 5A, 5B**).

There were no intraoperative or postoperative complications. Postoperatively, the ankle was immobilized with a short leg cast for 6 weeks. Partial weight bearing was permitted thereafter and full weight bearing was allowed after 10 weeks. The clinical outcome at 6 months after surgery was excellent; the patient was able to do all his activities with full range of motion

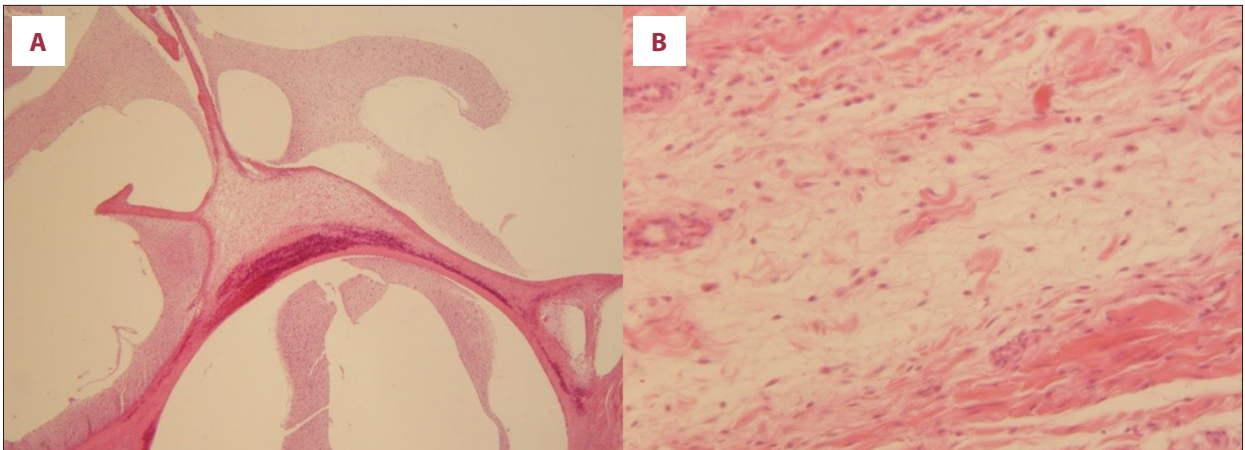


Figure 5. Photomicrographs at magnification of (A) $\times 20$ and (B) $\times 40$ depicting the loose connective tissue with myxoid change and the presence of blood vessels and a small number of lymphocytes.

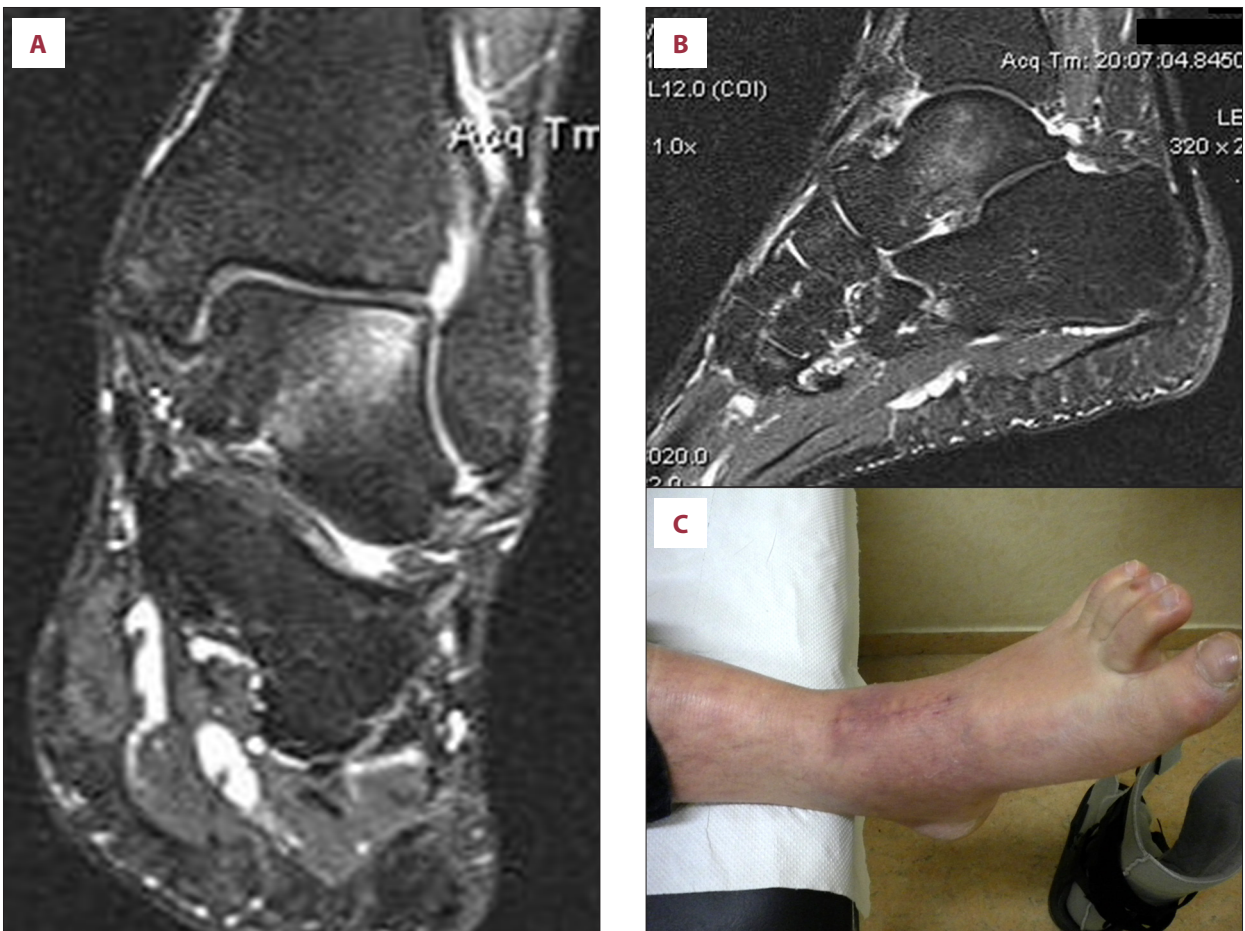


Figure 6. T1-weighted (A) coronal and (B) sagittal magnetic resonance images 2.5 years after surgery show mild bone marrow edema of the talus and only 1 nondiscrete small subchondral lesion. (C) Final clinical follow-up was excellent.

and no pain. He returned to his professional obligations without any discomfort. There was no recurrence at the final follow-up 5 years postoperatively, and the American Orthopaedic Foot and Ankle Society score was 90. The radiographs did not show any degenerative changes or signs of recurrence. The last MRI demonstrated almost complete filling of the subchondral cystic lesions without any signs of articular surface collapse. There was mild bone marrow edema of the talus and 1 non-discrete small subchondral lesion (Figure 6A-6C).

Discussion

Intraosseous ganglion talar cysts are rarely encountered in orthopedic practice. Despite their benign nature, these cysts may impose a significant burden on the affected patients, causing deep ankle pain that frequently increases with activity, local swelling, and reduced range of motion [14]. The radiographic appearance of these lesions usually involves a well-demarcated radiolucent defect, outlined by a rim of sclerotic bone [15]. Differential diagnosis of a cystic talar lesion should include subchondral cyst related to arthritis, enchondroma, chondroblastoma, giant cell tumor, osteoid osteoma, and unicameral bone cyst. In our case, there were no radiologic signs of osteoarthritis. MRI is the preferred imaging modality for differential diagnosis because it can identify any interaction between the intraosseous lesion and the joint space and can reveal the intrinsic structure of the ganglion. In our patient, 2 separate cystic lesions were found in the anteromedial and anterolateral talar dome with no interconnection between them or to the ankle joint.

Controversy exists in the literature regarding the pathogenetic mechanism of intra-osseous ganglia. Two different types of intra-osseous ganglion have been reported: (1) isolated, non-communicating soft tissue ganglion and (2) juxta-articular soft tissue ganglion communicating through a cortical defect. According to some authors, intraosseous ganglia are considered lesions that probably arise from the soft tissues and then by pressure can erode the adjacent bone [16]. Other authors have suggested that ganglia can arise from the bone itself or from juxta-articular soft tissues and the type of progression in either an outside-in or inside-out fashion may vary depending on the anatomical site [17]. Another theory is mucoid degeneration of intramedullary connective tissue that is probably preceded by focal ischemia or aseptic necrosis. However, other authors support that isolated intraosseous ganglia must be considered discrete lesions from their soft tissue counterparts eroding the bone and should also be distinguished from subchondral lesions associated with osteochondral injury or established osteoarthritis [18]. These authors have proposed other causative theories, such as proliferation of synovial remnants, local vascular disruption, aseptic necrosis, and local metaplasia [19].

Our case is interesting and in our view worth reporting because not only is the existence of an intraosseous talar ganglion rare, but the simultaneous ingrowth of a second similar lesion in the same bone without any connection to the first has never been reported. A probable cause is that these 2 ganglia arose from juxta-articular soft tissues within the bone in an outside-in fashion. The fibrillated cartilage and the intense synovitis created during the first arthroscopic debridement might have been the cause of these lesions. Different timing of progression possibly explains the absence of the second ganglion during the first MRI interpretation.

The approach we have described provides the following significant advantages: (1) avoidance of injury to articular cartilage; (2) avoidance of damage to the adjacent ligament tissue, peripheral blood vessel, and nerves; (3) convenience for treating the cyst, especially any located at the anterior ankle; and (4) shorter postoperative hospitalization time and less morbidity.

The generally suggested treatment of a symptomatic intraosseous ganglion includes discharging of the cyst content and curettage of the cystic lesion with abrasion of the surrounding sclerotic rim, along with bone grafting. Arthroscopic, endoscopic, and open approaches have been described in the management of intraosseous talar ganglion cysts [20]. The management of these lesions becomes more complicated in cases with concomitant joint involvement. In these cases, the cartilage lesion should also be addressed using osteochondral reconstruction techniques. We recommend this surgical approach and technique in cases of intraosseous talar lesions without communication to the talar dome articular surface, as in our patient.

Our main goal to support the subchondral bone while preventing overlying cartilage collapse was successfully achieved, as indicated by the final MRI findings, 5 years after surgery. We believe that the establishment of the bony canal of the talar neck under fluoroscopic guidance can achieve effective debridement and bone grafting with satisfactory long-term follow-up results. However, the present study had some limitations, including the single case report and the lack of a control group. In the future, multicenter studies could compensate for these deficiencies.

Conclusions

To the best of our knowledge, this is the first reported case of 2 distinct separate intraosseous ganglion cysts of the talus. We recommend this type of precise targeting technique in cases of intraosseous talar lesions with intact articular cartilage.

Department and Institution Where Work Was Done

Investigation performed at the Departments of Orthopaedic Surgery, Radiology, and Pathology of General University Hospital of Patras and the Department of Microbiology of General St. Andrews Hospital of Patras, Greece.

Conflict of Interest

None.

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