

Case Report

Severe bilateral cystic osteochondral lesion of the talus: A case report

Otaviano de Oliveira Júnior¹ , Carollina Gomes de Oliveira¹ , Thiago Barroso Ferreira Lima Brito Campos¹ ,
Nathália Barroso Ferreira Lima Brito Campos¹ 

1. Faculdade Ciências Médicas de Minas Gerais, Belo Horizonte, MG, Brazil.

Abstract

Osteochondral lesion of the talus (OLT) affects the articular cartilage and subchondral bone and may progress to osteonecrosis and cystic degeneration. We report the case of a patient with severe, bilateral OLT with expansive cystic lesions, with no history of trauma or joint instability, but with subtle varus deformity of the hindfoot. Following imaging tests, an osteochondral lesion was suspected, with a differential diagnosis of cystic neoplasm. Treatment consisted of an arthroscopy to assess the ankle joint, followed by medial access with an osteotomy of the medial malleolus for osteochondroplasty of the OLT (curettage, autologous bone grafting, and coverage with a biological membrane), combined with axis alignment via a valgus osteotomy of the calcaneus. The anatomopathological examination confirmed OLT. The patient showed good clinical progress on follow-up imaging. This case contributes to the literature by describing a rare, bilateral, severe presentation of OLT with no relevant predisposing factors, as well as a favorable response to complex surgical treatment. It reinforces the disease's variability, which can range from mild forms to challenging conditions.

Level of evidence IV; Case Report.

Keywords: Osteochondral lesion; Bone cysts; Osteonecrosis; Talus; Case report.

Introduction

Osteochondral lesion of the talus (OLT) is a condition of the articular cartilage on the talar surface, often associated with changes in the subchondral bone⁽¹⁻⁴⁾. Its pathophysiology involves chondral damage, followed by infiltration of synovial fluid and compromised blood supply, leading to osteonecrosis of the subchondral bone and cystic degeneration⁽³⁾. Evidence indicates an association between OLT and microtrauma due to chronic ligament instability of the ankle or misalignment of the mechanical axis (hindfoot and ankle), in addition to acute trauma^(2,3,5).

Osteochondral lesions of the talus are more prevalent in men over 30 years of age^(2,3). Its main symptoms usually appear six to 12 months after an initial traumatic event, such as fractures or ankle sprains, and include pain, swelling, joint

stiffness, and limited movement with progressive functional loss, aggravated by physical activity or weight-bearing^(1,4). Occasionally, mechanical complaints of clicking and joint locking are also reported in cases with osteochondral loose bodies^(2,3). The presentation is predominantly unilateral, while bilateral involvement is less common, occurring in about 15% of patients with OLT. Among those with bilateral lesions, only about one-third present symptoms in both ankles⁽⁶⁾.

Imaging exams are essential for diagnosis, treatment definition, surgical planning, and follow-up^(1,2,4). Radiographs help assess mechanical axis alignment, but they show the lesion only in advanced stages^(1,3). Magnetic resonance imaging (MRI) allows assessment of chondral damage, adjacent bone edema, and associated soft tissue lesions, although it may overestimate the extent of the OLT when bone edema is present^(1,4). Computed tomography (CT)

Study performed at Faculdade Ciências Médicas de Minas Gerais, Belo Horizonte, MG, Brazil.

Correspondence: Otaviano de Oliveira Júnior. Alameda Ezequiel Dias, 275, Centro, Belo Horizonte, 30130-110, MG, Brazil. **Email:** ooliveirajr@gmail.com.
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allows precise analysis of bone morphology, lesion size, and location, including characterization of subchondral bone changes and cystic lesions, and can be complemented by imaging of the ankle in maximum plantar flexion^(1,7). Computed tomography in plantar flexion assists preoperative planning by allowing assessment of OLT accessibility and, together with its location and extent, helps define the access route, which may involve an arthroscopic approach, arthrotomy, or, when adequate exposure of the lesion is needed, malleolar or tibial osteotomy^(1,7,8).

This is a report of a rare case of bilateral OLT associated with osteonecrosis and extensive cystic degeneration of the talus, with an unusual clinical and radiographic presentation and a challenging initial diagnosis.

Case description

A 44-year-old male patient, a recreational soccer player, previously healthy, presented with pain in his left ankle and progressive limitation of sports practice after returning to play following previous surgery for reconstruction of an anterior cruciate ligament tear in his right knee. The patient denied joint instability, a history of trauma or previous interventions on the ankles, as well as any relevant personal or family medical history. The main systemic risk factors associated with osteonecrosis were evaluated and ruled out, including corticosteroid use, alcohol abuse, coagulopathies, and dyslipidemia, as well as the presence of apparently active metabolic, rheumatic, or autoimmune disease.

On physical examination, a mild, symmetrical varus deformity of the calcanei was observed, with a reducible Coleman deformity and negative Silverskiöld tests. The patient reported diffuse left ankle pain that did not prevent jumping with single-leg support, with no edema or apparent ankle instability.

To assess pain and function, the Visual Analog Scale (VAS), ranging from 0 to 10, and the American Orthopaedic Foot and Ankle Society (AOFAS) score, with a reference value of

100 for normality, were used. On the first evaluation, the left ankle had VAS pain of 5 and an AOFAS of 56; the right ankle had VAS pain of 0 and an AOFAS of 95.

Diagnostic imaging began with an MRI of the left ankle, which showed an extensive osteochondral lesion in the medial talar dome, affecting the entire anteroposterior medial length and measuring approximately 26 × 10 mm, with a large lobulated cyst measuring 29 × 21 × 13 mm and osteochondral fragmentation. Subsequently, bilateral radiographic images of the ankles were taken in anteroposterior, anteroposterior with 15° internal rotation, and lateral views, all weight-bearing, as well as axial and Saltzman axial views of the calcanei, which showed subtle and symmetrical varus of the calcanei, with a Saltzman angle estimated at approximately 5°, in addition to cystic images in the talar dome bilaterally, more prominent on the left (Figure 1). The investigation was complemented with a CT scan of the ankles, and on the symptomatic left side, a large image suggestive of a multilobulated subchondral bone cyst was found in the talar dome with cortical fragmentation and sclerotic margins reaching the inferior wall of the subtalar region, measuring 18 × 11 × 28 mm (Figure 2).

The patient was referred for an oncological evaluation to investigate a tumor lesion, which was ruled out based on the characteristics described. Given the clinical and imaging findings, the decision was made, together with the patient, to surgically address the lesion on the left side, which was symptomatic at that initial stage.

The surgical treatment consisted of an arthroscopic examination of the joint to evaluate the articular surface and identify changes not accessible through the osteotomy approach, such as possible loose bodies or other changes in the anterolateral gutter that were not identified. Next, an open osteochondroplasty of the OLT was performed on the left ankle, and a Chevron-type osteotomy of the medial malleolus of the tibia was performed to improve access to the lesion. The varus axis was also corrected through a Dwyer-type valgus osteotomy of the calcaneus, with about 1 cm of lateral wedge resection.



Figure 1. Bilateral ankle radiographs in anteroposterior view and anteroposterior view with 15° internal rotation showed expansile cystic lesions in the medial talus, more pronounced on the left side, as well as subtle hindfoot varus on the Saltzman view.

The osteochondroplasty of the OLT consisted of debridement of the detached cartilage, curettage of the cystic contents, and collection of material for biopsy, along with bone grafting and application of a biological membrane to the cystic defect (Figure 3). The osteotomy was subsequently fixed with two cross screws. The autologous bone graft used to fill the cyst was obtained from the lateral resection

wedge of the calcaneus after the valgus osteotomy using the Dwyer technique. However, due to the extent of the cystic defect, supplementation with an ipsilateral iliac crest graft was necessary. After filling the cyst, a biological membrane (Chondro-Gide® model, Manufacturer: Geistlich Pharma AG, Wolhusen, Switzerland) was applied to correct the chondral defect.

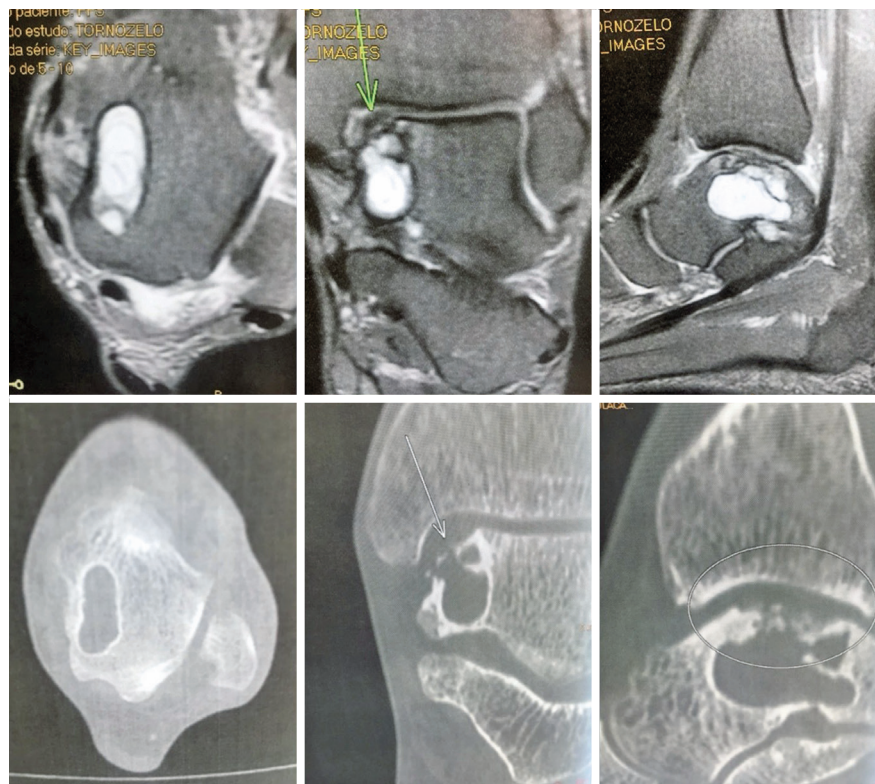


Figure 2. Magnetic resonance imaging and computed tomography scans of the left ankle demonstrate an osteochondral lesion of the talus with an extensive cyst.

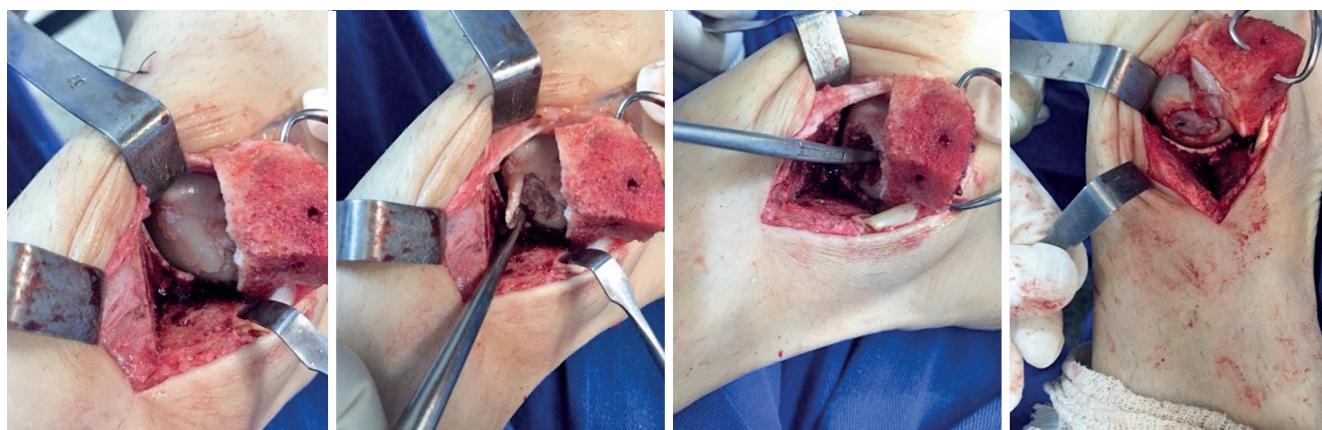


Figure 3. Intraoperative appearance of the left talus during osteochondroplasty.

The pathological anatomy examination revealed inflammatory fibroconnective tissue, typical of osteochondral necrosis, with no evidence of neoplasia (Figure 4). Despite the extent of the cystic component, no frozen-section histopathological analysis was performed during the procedure. This decision was based on the prior oncological evaluation, which found no evidence suggestive of neoplasia, and on the bilateral presentation and clinical-radiographic findings, which were considered more compatible with a biomechanical/degenerative etiology. Therefore, the decision was made to perform definitive reconstructive treatment during the same procedure, filling the defect with a bone graft and covering it with a membrane.

The surgery was performed under spinal anesthesia combined with sedation and regional analgesic block, with the administration of three doses of cefazolin sodium for antibiotic prophylaxis. For suturing, the arthroscopic portals were closed with simple sutures using 2-0 Nylon (Mononylon®/ETHILON®; Ethicon, Johnson & Johnson MedTech). In the other wounds, the deep layers were closed with 2-0 Polyglactin (Vicryl®; Ethicon, Cincinnati, USA) and 3-0 Polyglecaprone (Monocryl®; Ethicon, Cornelia, USA), and the skin was closed with Donati sutures using 3-0 Nylon (Mononylon®/ETHILON®; Ethicon, Wilmington, USA).

In the postoperative period, the patient remained non-weight-bearing for six weeks, using crutches and an orthopedic boot for immobilization. Physical therapy began after the surgical wounds healed, about three weeks after the procedure, encouraging early mobility of the operated joint and conventional muscle-strengthening exercises, with weight-bearing allowed after six weeks post-surgery. The patient had monthly outpatient check-ups during the initial follow-up, then every six months. Initially, the patient chose

to postpone surgery on the contralateral ankle, which was asymptomatic at the time, preferring to wait for the outcome of the procedure on the left side and to perform surgery on the contralateral side only if there was clinical worsening.

Six years after left ankle surgery, the patient appeared to be progressing satisfactorily (VAS 1; AOFAS 90), but with worsening right ankle pain (VAS 5; AOFAS 66) (Figure 5). Computed tomography was performed for evaluation, revealing a grotesque cystic, expanding lesion in the medial talus on the right side, measuring 23 x 14 x 21 mm (Figure 6). Given the contralateral result, where the patient showed significant clinical improvement despite residual degenerative joint changes, we decided to repeat the same surgical technique (Figures 7 and 8). The only difference between the sides was the need to use three screws to fix the right medial malleolus osteotomy in a cross configuration, as well as the types of screws provided by the suppliers and health insurance at the time. No adverse events were observed during treatment or follow-up.

Currently, eight years after treatment of the left ankle and two years after treatment of the right ankle, both are showing favorable clinical progress (VAS 1 during physical exertion and AOFAS score of 90 bilaterally), with significant improvement in pain and joint function and a return to high-impact sports involving jumping, sprinting, and rapid changes of direction. Imaging studies (Radiographs, CT, and MRI) showed near-complete incorporation of the bone graft into the cystic lesions, consolidation of the osteotomies, and no signs of failure of the fibrocartilaginous repair tissue (Figures 9, 10, and 11). Despite persistent cystic changes and degenerative changes in the left tibiotarsal and subtalar joints, these findings did not have significant clinical repercussions during long-term follow-up.

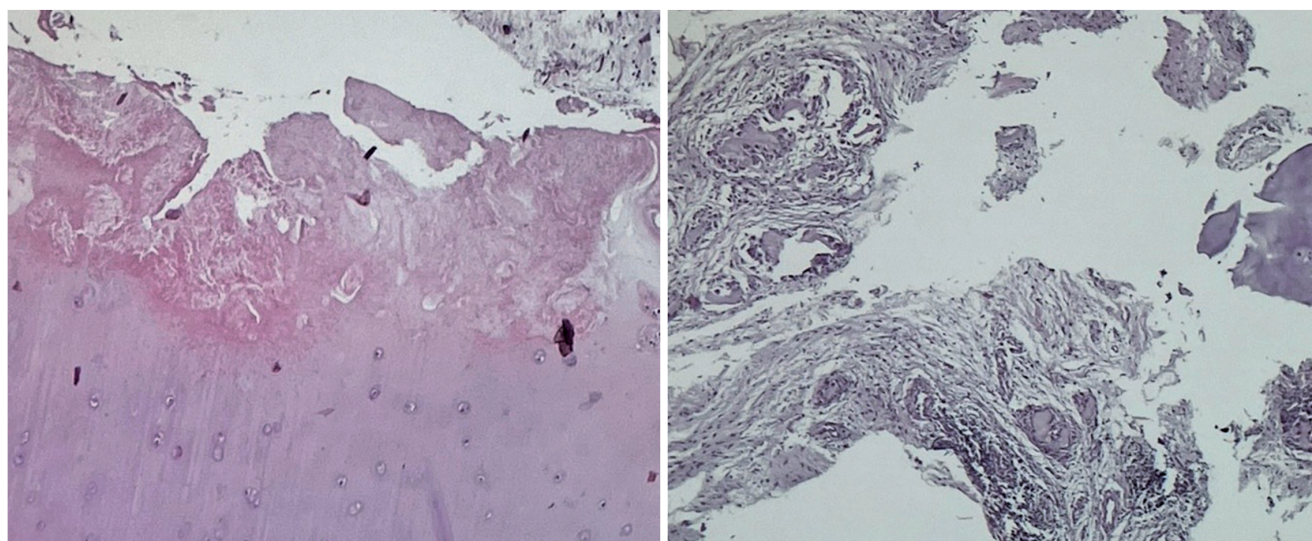


Figure 4. Histopathological analysis of the biopsy specimen from the left talus, consistent with an osteochondral lesion of the talus.



Figure 5. Clinical appearance of the left ankle and foot six years after surgery and the right side preoperatively.

The patient signed the Informed Consent Form, confirming his participation and authorizing the publication of this case report. The study was approved by the Institutional Review Board.

Discussion

This case is noteworthy due to the unusual presentation of an OLT in its severe, bilateral form, associated with exuberant cystic degeneration, in a patient with no clear history of trauma and no reported joint instability. This set of findings makes the case relevant, as it differs from the presentation most described in the literature, where OLT is usually unilateral and smaller or has cystic degeneration⁽⁶⁾.

The etiology of OLT is still not fully understood⁽¹⁾. There is consensus that joint instability and trauma are associated with their development, but there is no evidence that deviation of the mechanical axis is a direct causal factor, although the literature suggests a strong relationship⁽³⁾. In this context, eccentric stress on the articular cartilage, medial in varus and lateral in valgus, may favor repetitive microtrauma and degeneration of the subchondral bone, contributing to OLT^(3,9). In this case, although mild hindfoot varus probably does not explain the severity of the condition in isolation, it may have contributed as a biomechanical factor, promoting chronic medial overload and progressive subchondral degeneration.

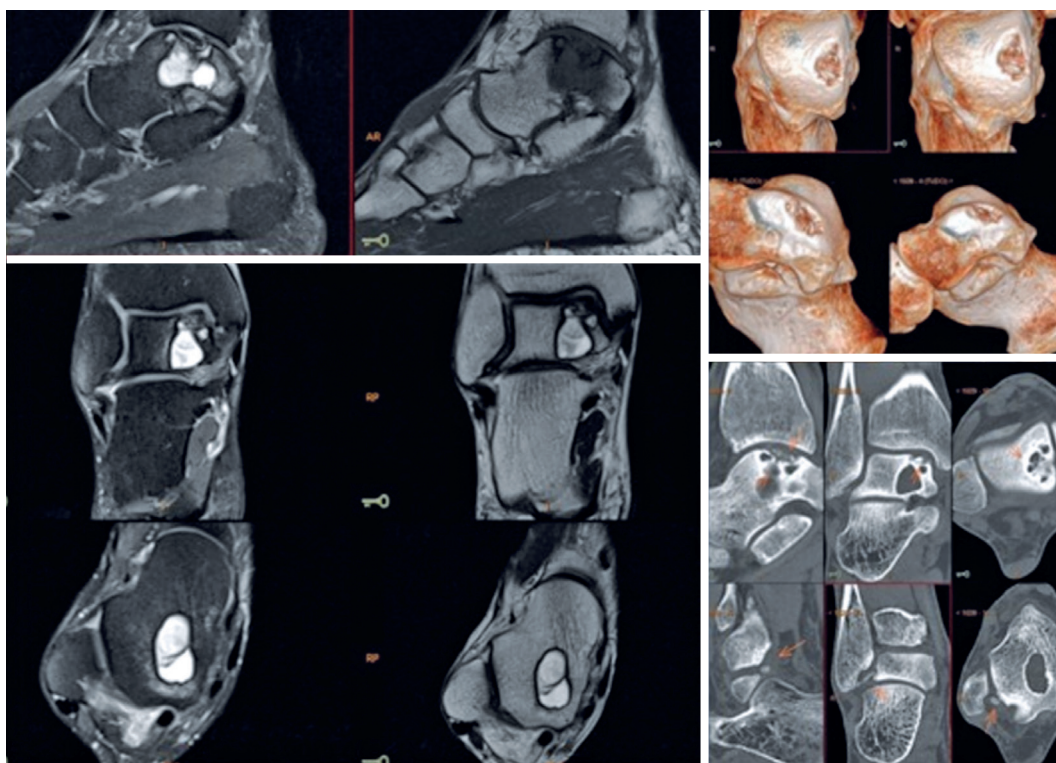


Figure 6. Magnetic resonance imaging and computed tomography scans of the right ankle demonstrating an osteochondral lesion of the talus.

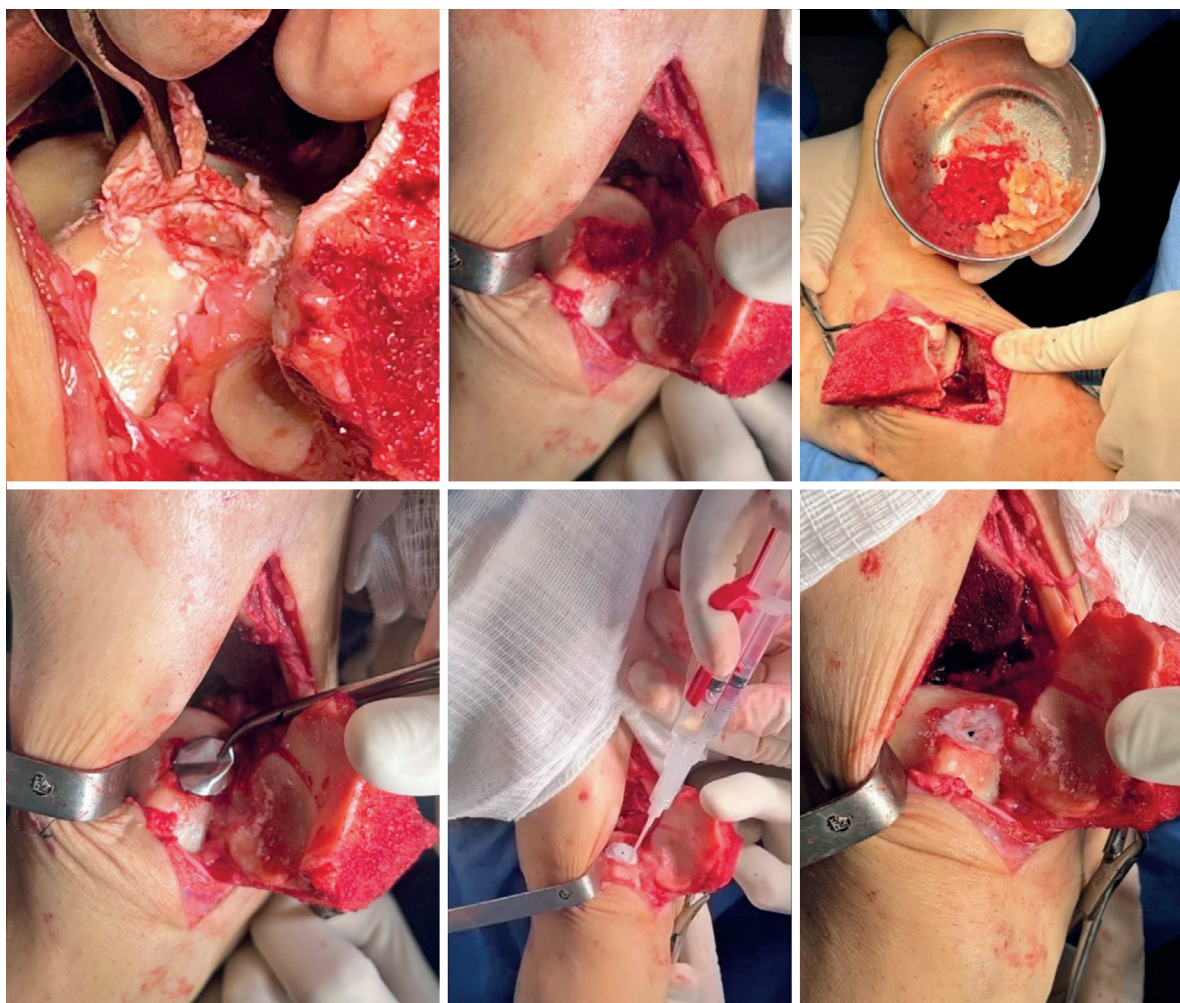


Figure 7. Intraoperative appearance of the right talus during osteochondroplasty, showing the osteotomy and curettage of the lesion, followed by bone grafting and biological membrane coverage.

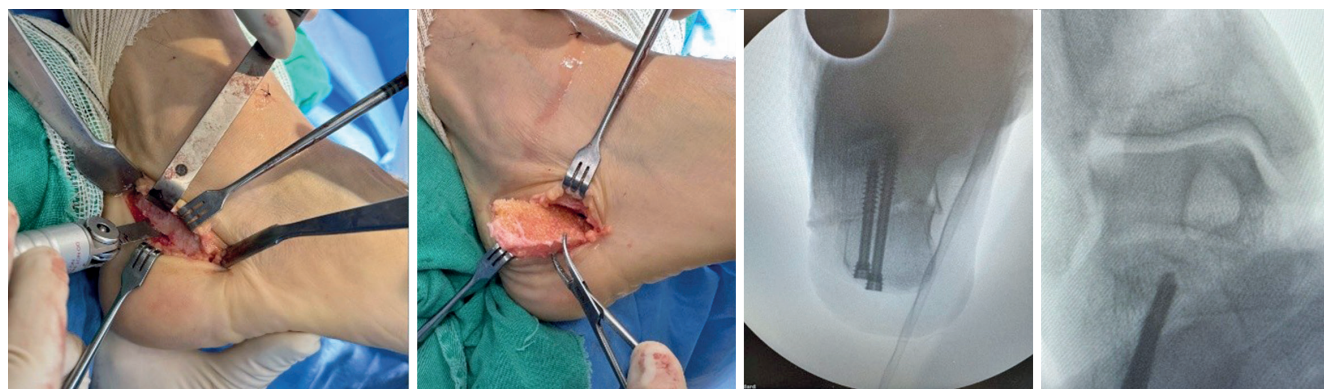


Figure 8. Intraoperative aspect of the Dwyer osteotomy of the right calcaneus, with fluoroscopic imaging confirming fixation using two screws.

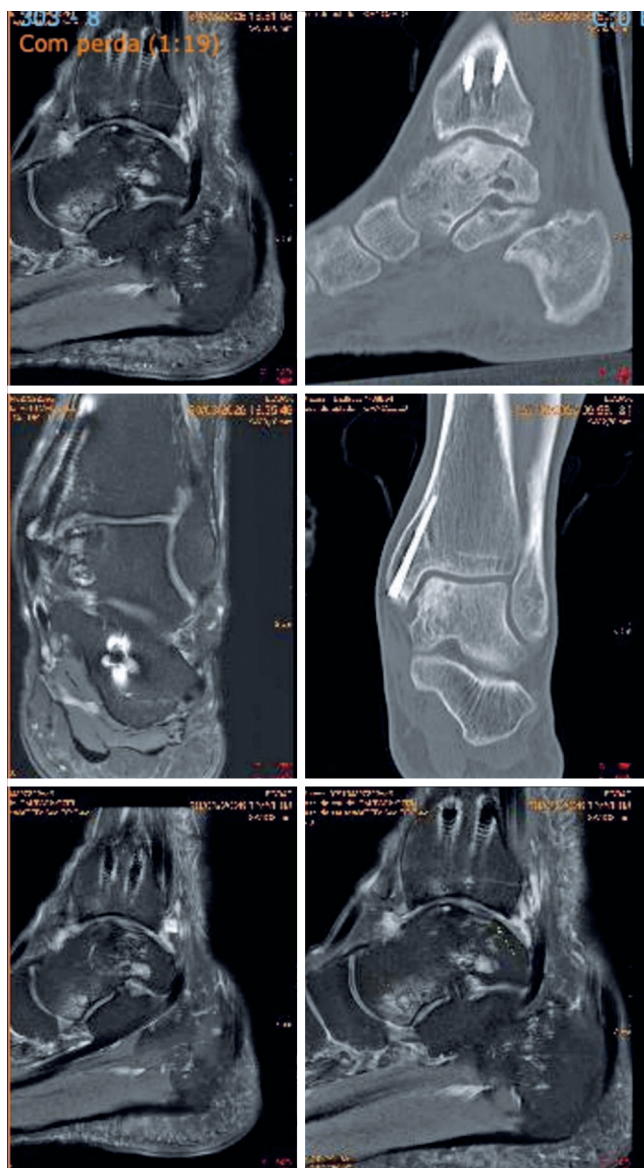


Figure 9. Magnetic resonance imaging and computed tomography scans of the left ankle eight years post-surgery.

Osteochondral lesions of the talus do not have a single, standardized classification system and can be characterized by linear dimensions, surface area, or volume, which limits direct comparison between studies^(1,8). In addition, different assessment methods use their own classifications: the Berndt and Harty classification is based on radiographs, the Ferkel classification on CT, the Hepple classification on MRI, and the Pritsch classification on arthroscopic findings. For topographic description, Raikin's nine-zone classification can be used to locate the lesion in the talar dome, not to grade its severity^(1,7,10). In the present case, although complex, they fall under Berndt and Harty type V (Radiograph), Hepple type V

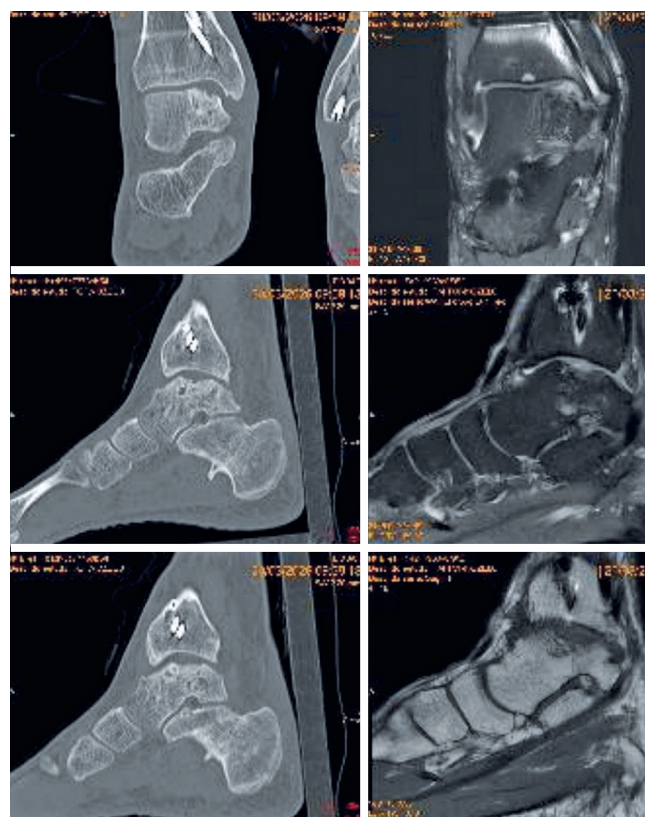


Figure 10. Magnetic resonance imaging and computed tomography scans of the right ankle two years post-surgery.



Figure 11. Bilateral radiographs of the ankles, eight years after surgery on the left and two years after surgery on the right ankle.

(MRI), Ferkel stage 3 (CT), Pritsch grade III (arthroscopy), and Raikin zones 1, 4, and 7 (topography).

Some studies have evaluated OLT dimensions. One study involving 142 symptomatic patients found lesions with mean dimensions of $6.9 \times 9.4 \times 5.4$ mm (0.35 cm^3)⁽¹⁾. Another study of 161 patients with bilateral OLTs reported a mean volume of $0.52 \pm 0.64 \text{ cm}^3$, with smaller lesions in asymptomatic ankles and larger ones in symptomatic ankles⁽⁶⁾. Brulc et al.⁽¹¹⁾, evaluating 99 patients who underwent surgical treatment, found larger lesions, with a mean area of $2.0 \pm 1.1 \text{ cm}^2$ and 51% of defects greater than 2 cm^2 . Although the methodological differences between the studies prevent a direct quantitative comparison, these data demonstrate the wide dimensional variability of the OLTs and help contextualize the magnitude of the osteochondral involvement observed in the present case, in which the lesions were considerably larger than those described in this paragraph as measured by CT (left side $18 \times 11 \times 28$ mm, 5.54 cm^3 ; right side $23 \times 14 \times 21$ mm, 6.76 cm^3)^(1,6,11).

The patient initially presented with nonspecific symptoms in the left ankle, characterized by pain and functional limitation, despite the presence of bilateral structural lesions. His initial clinical evaluation included differential diagnoses such as osteoarthritis, ligament microinstability (anterolateral, rotational, or multidirectional), and tendinopathies. However, the initial MRI scan revealed extensive cystic lesions of the talus with cartilage fragmentation in the dome, leading to a differential diagnosis of osteoarticular neoplasia. The subsequent finding of bilaterality, in addition to the clinical presentation and the findings in the other radiographs and CT scans, including the presence of sclerotic margins in the cysts and cartilage fragmentation, supported the diagnosis of OLT^(6,12).

Osteochondral lesions of the talus should be treated individually, as no gold standard exists for surgical management, especially for chondral repair^(1,3). The conservative approach is mainly indicated for oligosymptomatic cases, newly diagnosed cases, or cases without cartilage detachment⁽³⁾. When surgery is indicated, treatment can be divided into three main strategies: repair, regeneration, and osteochondral replacement^(3,4).

Direct repair, generally indicated for lesions smaller than 1.0 cm^2 , involves debridement of the OLT and microperforations of the subchondral bone to stimulate bone marrow and reparative tissue formation^(1,4,5). For larger lesions, especially those over 1.5 cm^2 and associated with subchondral cysts larger than 3 mm, regenerative techniques can be used,

with filling of the bone defect with a graft and coverage with biological membranes, as in Autologous Matrix-Induced Chondrogenesis (AMIC), including different scaffolds of collagen, hydroxyapatite, hyaluronic acid, or platelet-rich fibrin^(2,3,5). The third option is osteochondral replacement with an autologous graft, traditionally obtained from the lateral femoral condyle, preferably as a single plug, or with an allograft in extensive or refractory lesions, although with greater complexity and risks of long-term incorporation^(1,3,5).

Consider correcting the mechanical axis through periarticular osteotomies, whether at the calcaneal or supra-malleolar level, when varus or valgus deviations represent significant risk factors^(1,3,6,10). Medial OLT correlates with hindfoot varus, emphasizing the need to assess and correct axis alignment with a periarticular osteotomy during surgical treatment⁽¹⁰⁾.

In this case, osteochondroplasty combined with chondral regeneration was chosen, as recommended and available at the time. After curettage of the OLT and cancellous bone grafting in all cysts, a biological membrane (ChondroGide® - Geistlich Pharma AG, Wolhusen, Switzerland) was applied according to the recommended technique and with fibrin glue, which induces the differentiation of mesenchymal stem cells, restoring the integrity of the newly formed fibrocartilage^(1,3,4). Bone grafting of the subchondral defect plays an important role in treating cystic OLTs and may be necessary when, after debridement, bone loss greater than 4 mm persists, especially when scaffolds are used⁽⁵⁾. In severe cases like this one, realignment of the mechanical axis by osteotomy is essential, as it helps redistribute joint load and prevent degenerative changes that may precede OLTs^(3,9).

Finally, the severe, bilateral progression of OLTs stands out, even without significant underlying causes. After all, the mild varus deformity of the hindfoot alone would not justify this level of degeneration. This report reinforces the importance of a comprehensive, individualized assessment of atypical osteochondral lesions, supporting early diagnosis, appropriate therapeutic planning, and advancing knowledge of rare and challenging conditions. It also highlights the disease's variability, which can range from mild forms to more challenging presentations. The authors consider curettage of the subchondral cyst with subsequent bone grafting, along with correction of the mechanical axis, the most important parts of the surgical treatment and decisive for success, with coverage of the chondral defect with a biological membrane as an adjunct.

Authors' contributions: Each author contributed individually and significantly to the development of this article: OJOJ *(<https://orcid.org/0000-0001-7766-9974>) Performed the surgeries, clinical examination, and data collection; CGO *(<https://orcid.org/0009-0008-3628-3408>) Conceived and planned the activities that led to the study, formatted the article, participated in the review process, and statistical analysis; TBFLBC *(<https://orcid.org/0009-0002-9921-2152>) Conceived and planned the activities that led to the study, survey of the medical records, and bibliographic review; NBFLBC *(<https://orcid.org/0009-0002-1160-3301>) Conceived and planned the activities that led to the study, interpreted the results of the study, formatted the article, participated in the review process. All authors read and approved the final manuscript. *ORCID (Open Researcher and Contributor ID) .

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