Aneurysmal Bone Cyst of the Cuboid. Case Report and Review of the Literature

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ABSTRACT

A rare clinical case of an aneurysmal bone cyst located at the level of the cuboid in a 13-year-old boy is presented. The diagnosis was reached through clinical evaluation, imaging studies (radiograph-MRI), and a pathological anatomy analysis. Its treatment consisted of thorough intralesional curettage, high speed burring, electrocautery, and filling with lyophilized bone allograft with cortical/cancellous chips. Despite the late diagnosis, we would like to highlight the favorable clinical evolution of the patient, with ad integrum remission of the symptoms and return to his sport activities, without elements of local recurrence.

Keywords: aneurysmal bone cyst.

Level of Evidence: IV

Quiste óseo aneurismático en el cuboides de un niño de 13 años. Presentación de un caso clínico

RESUMEN

Se presenta un caso clínico poco frecuente de un quiste óseo aneurismático localizado en el cuboides de un niño de 13 años. Se llegó al diagnóstico mediante la tríada de síntomas, estudios por imágenes (radiografía, resonancia magnética) y anatomía patológica. El tratamiento consistió en el abordaje y curetaje minuciosos dentro de la lesión, el fresado de alta velocidad, la electrocauterización y el relleno con aloinjerto óseo liofilizado con chips cortico-esponjosos. Pese al diagnóstico tardío, la evolución clínica fue favorable con remisión completa de los síntomas y retorno a las actividades deportivas, sin recidiva local.

Palabras clave: Quiste óseo aneurismático.

Nivel de Evidencia: IV

INTRODUCTION

Jaffe and Lichtenstein published the first description of an aneurysmal bone cyst (ABC) in 1942.[1,2,3] It is a rare condition and represents about 1% of all primary bone tumors. Although it can occur at any age, it is most common between the ages of 10 and 20, with around 75% of cases corresponding to individuals aged <20.[1,2,3] It can manifest as a primary lesion (70%) or as a secondary lesion to an existing lesion.[1,2,3] It is defined as a benign cystic lesion of the bone composed of blood-filled spaces separated by connective tissue septa containing fibroblasts, osteoclast-like giant cells, and reactive bone tissue.[1,2,3]

Its clinical presentation can go unnoticed, and it can develop with pain, inflammation, or without symptoms depending on whether it sits on a weight-bearing bone or not.

In some cases, it can present as a pathologic fracture.[1,2,3,4,5]

It should be noted that it is a locally destructive process and has high recurrence rates.

ABC treatment is determined by the patient's age, location, extent, degree of aggressiveness, and size. There are currently several treatment modalities available, such as intralesional curettage, resection plus bone grafting, local

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adjuvants, such as high-speed drilling, electrocautery, phenol, liquid nitrogen, and embolization, which have the advantage of expanding the zone of necrosis of tumor tissue cells.

There are different opinions about which is the best method of treatment for the pediatric population as opposed to adults, because it is based on immature bone and can appear in areas adjacent to the growth plate.[1,2,3]

The first description of an ABC in the cuboid bone dates from 1977 and, since then, few cases of this clinical presentation have been published in this very atypical topography, either in its primary form or associated with other lesions, such as chondroblastoma.[6,7,8,9,10,11,12,13] Tables 1 and 2 detail the cases published to date.

Table 1. Published cases of aneurysmal bone cyst.

Year	Title	Authors
1977	Aneurysmal bone cysts: a clinicopathological study of 105 cases	Ruiter DJ et al.
1990	Aneurysmal bone cyst of the cuboid	Kashuk KB et al.
1999	Le kyste osseux anévrysmal du cuboide: etude d'un cas et reveu de la litterature	Essadki B et al.
2003	Aneurysmal bone cyst of the cuboid	Verrina F et al.
2010	Curettage of aneurysmal bone cysts of the feet	Chowdhry M et al.
2016	A rare case of aneurysmal bone cyst of cuboid bone in a 10-year-old girl	Bojovic N et al.

Table 2. Published cases of cuboid aneurysmal bone cyst/chondroblastoma

Year	Title	Authors
2005	Chondroblastoma with associated aneurysmal bone cyst of the cuboid	Sessions W et al.
2007	Chondroblastoma of the cuboid with an associated aneurysmal bone cyst: a case report	Sepah YJ et al.

In our Service (Centro Hospitalario Pereyra Rosell), curettage and filling with bone allograft is the most widely used therapeutic option.[3]

The aim of this article is to report our outcomes in this extremely rare clinical case.

It is critical that the interdisciplinary team that performed the diagnostic research on this type of patient remains in charge of the condition's definitive treatment. The team should include traumatologists, pediatricians, imaging specialists, pathologists, and pediatric oncologists.

CLINICAL CASE

After repeated emergency consultations for persistent pain in the neck of the foot and left foot as a result of multiple traumatisms, a healthy 13-year-old boy from the city of Minas, Uruguay, was referred to our external polyclinic service.

The patient suffered from pain with inflammatory characteristics (it did not subside with non-steroidal anti-inflammatory drugs or at rest, with a nocturnal component, without elements of general adaptation syndrome) and had a slow-growing tumor located in the midfoot that had evolved over seven months.

During the initial physical examination, the patient was found to be in good general condition, free of systemic symptoms, and requiring crutches for standing and ambulation. When asked to bear weight on the affected limb, he would use support and an antalgic gait to avoid the outer region of the foot.

The evaluation of the hip, knee and ankle revealed no alterations in passive/active range of motion and the distal neurovascular examination was normal.

The tumor was located on the external side of the midfoot, in the sector of the cuboid bone, it measured 3 x 3 cm, and had ill-defined limits, an ovoid shape, an irregular surface, a stony consistency, was immobile, associated with deep planes, and painful to the touch (Figures 1 and 2).

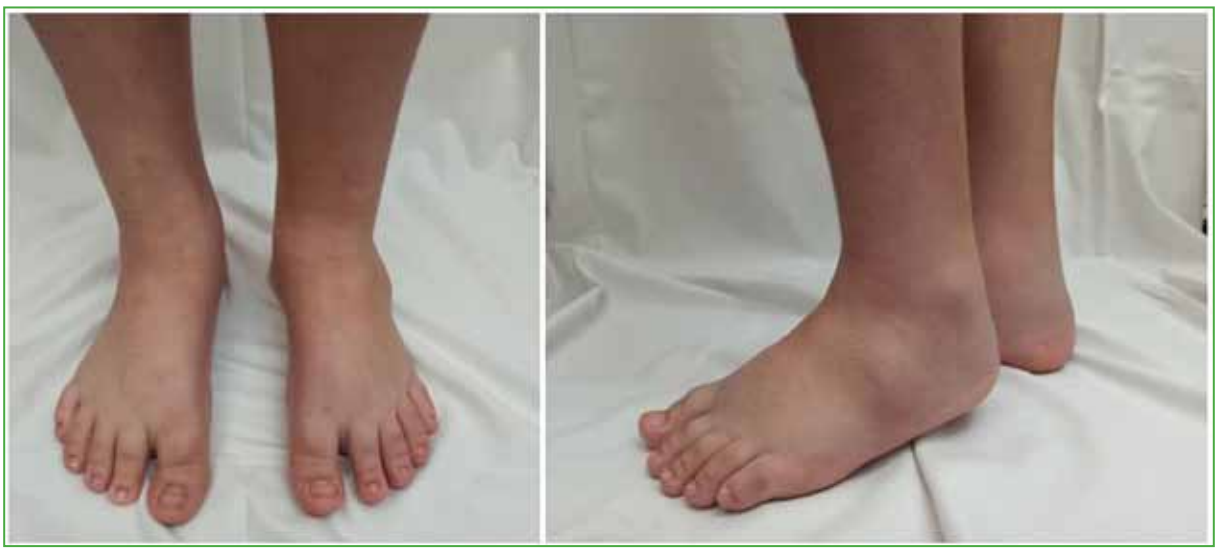

Figure 1. A and B. Clinical appearance of the patient's foot at the time of consultation. Tumor mass at the level of the external side of the midfoot at the level of the topography of the cuboid bone.

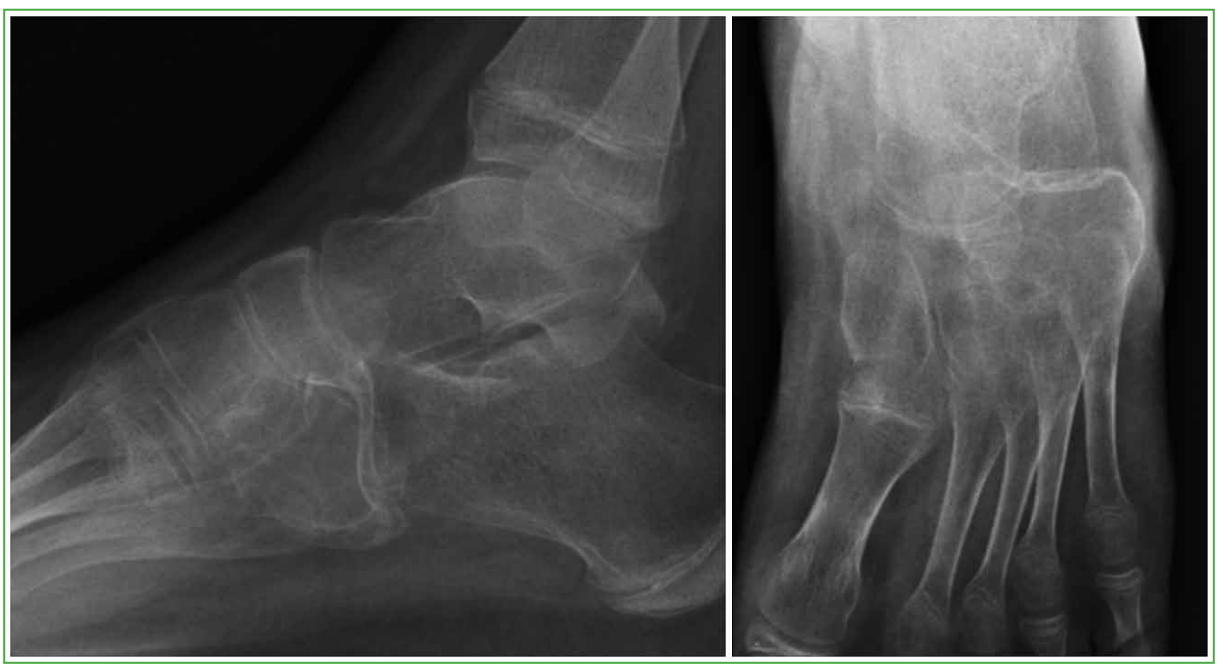

Figure 2. Anteroposterior and lateral foot radiograph. Due to diffuse osteopenia, an alteration of the normal morphological bone structure of the midfoot is evident; at the cuboid level, a lytic image is visible, with poorly defined margins, a narrow transition zone, hyper-inflated, with cortical thinning, and no soft tissue component.
Stage 2 Enneking.

Faced with a tumor of bone origin, he was evaluated using our institution's standard protocol, which included radiographs of the afflicted area, paraclinical blood tests for general and infectious evaluation, and magnetic resonance imaging (Figure 3).

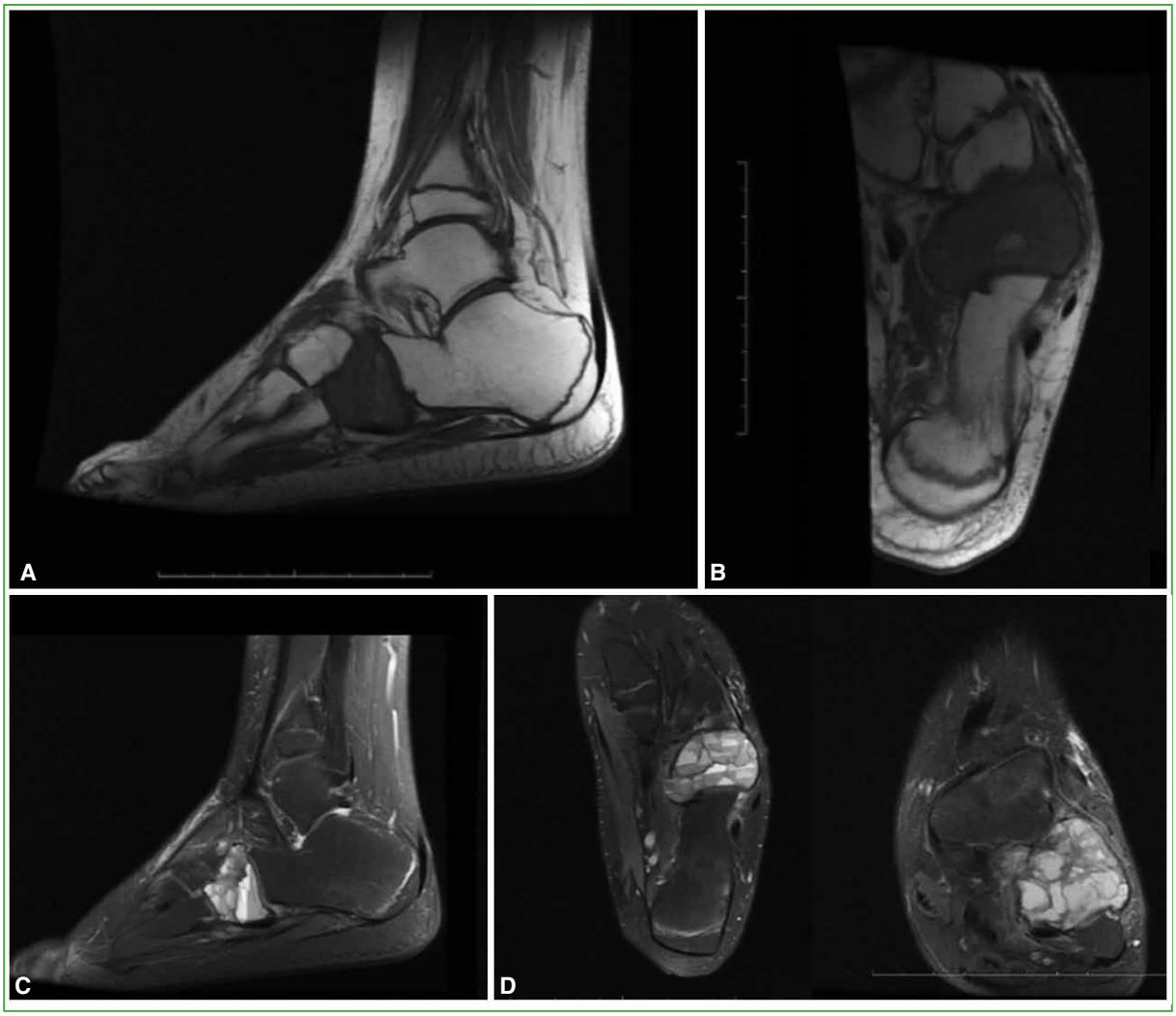

Figure 3. MRI of the neck of the foot and foot. T1 (A/B) -T2 (C/D) weighted images show the expansive cystic lytic lesion at the cuboid level with fluid-fluid levels within it, the presence of internal septa with a soap-bubble appearance, without changes at the soft tissue level or at the level of the calcaneus/cuboid, cuboid/4th and 5th metatarsal joints.

After the corresponding imaging studies, and despite the fact that they were highly suggestive of ABC, it was imperative to complete the diagnostic triad with a planned biopsy of the lesion (Figure 4). In published studies, it is argued that definitive treatment should not be administered without a diagnosis.

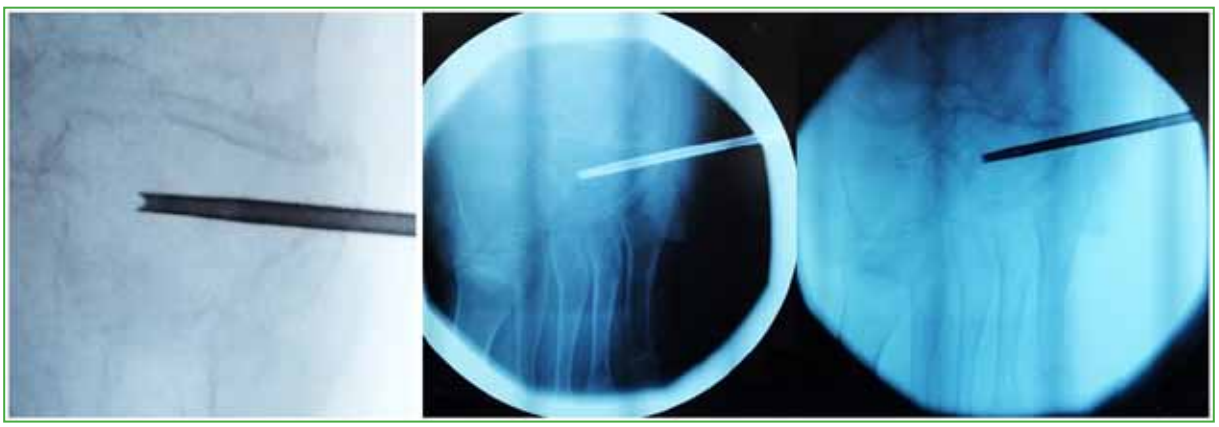

Figure 4. Intraoperative images of the percutaneous biopsy with bone trocar.

The procedure was carried out in the surgical unit under general anesthesia under strict aseptic conditions. The percutaneous biopsy was carried out through an approach on the long axis of the bone (longitudinal incision) as the last diagnostic step in order to confirm the nature of the tumor through pathological anatomy studies and bacteriological culture (Figure 5). Eight red-brown fragments measuring 0.4 x 0.5 and 0.2 x 0.2 cm were sent for examination. The microscopic analysis revealed fibrous wall flaps covered with histiocytic tissue and osteoclast-type giant cells, fibrous collagenic flaps, associated edematous fibrous tissue, and extensive hemorrhage in biopsy sections of bone. The histopathological diagnosis was ABC.

Figure 5. Puncture area and hemostasis with nylon suture.

Definitive treatment

Once the diagnostic triad was completed, we proceeded to complete the resection of the cyst in a second surgical stage (Figures 6-11).

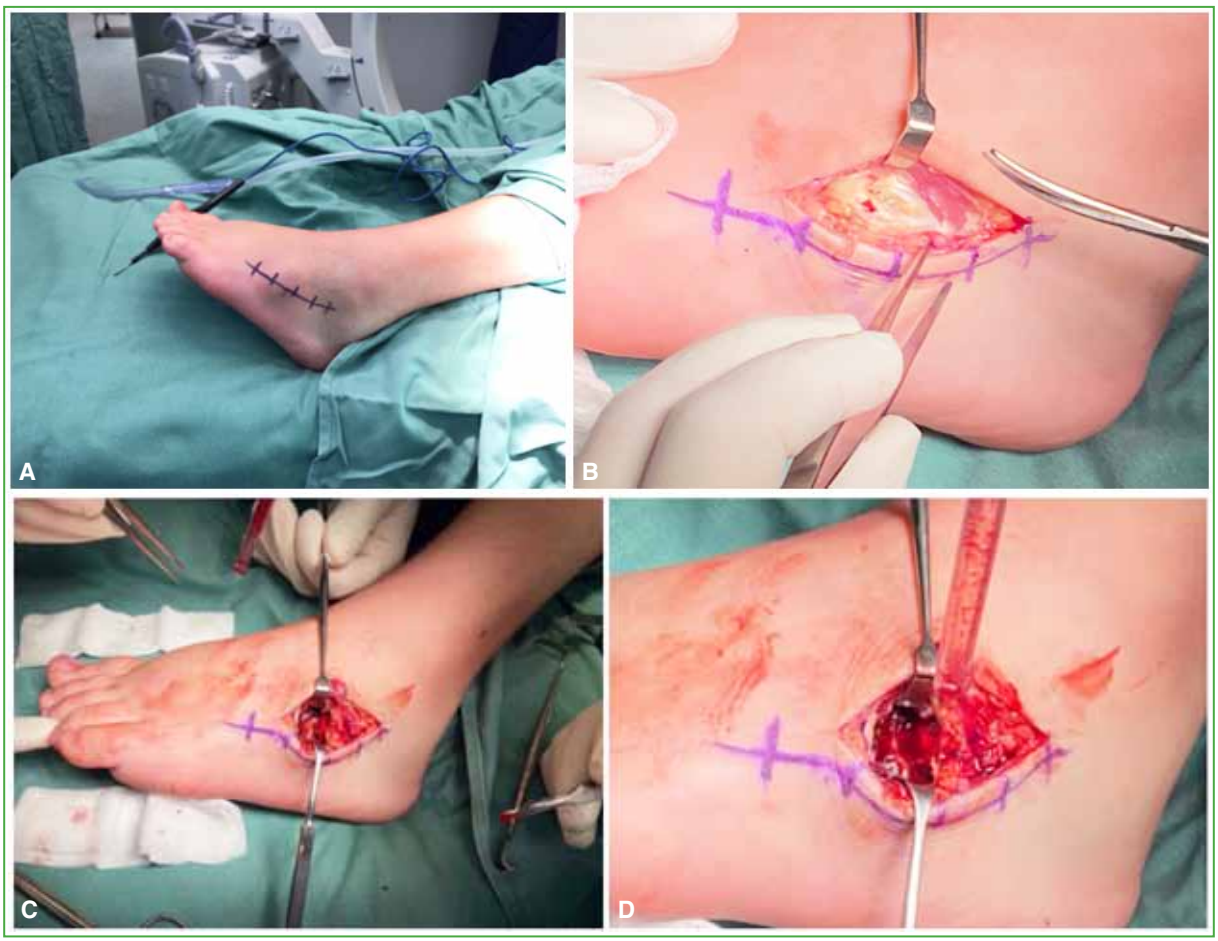

Figure 6. **A/B** Longitudinal approach centered on the topography of the cuboid bone. A bone window is made with a fine chisel to open the cyst. **C/D** The cavity is meticulously curetted using a simple curette to remove the internal connective tissue membranes and adherents to the walls.

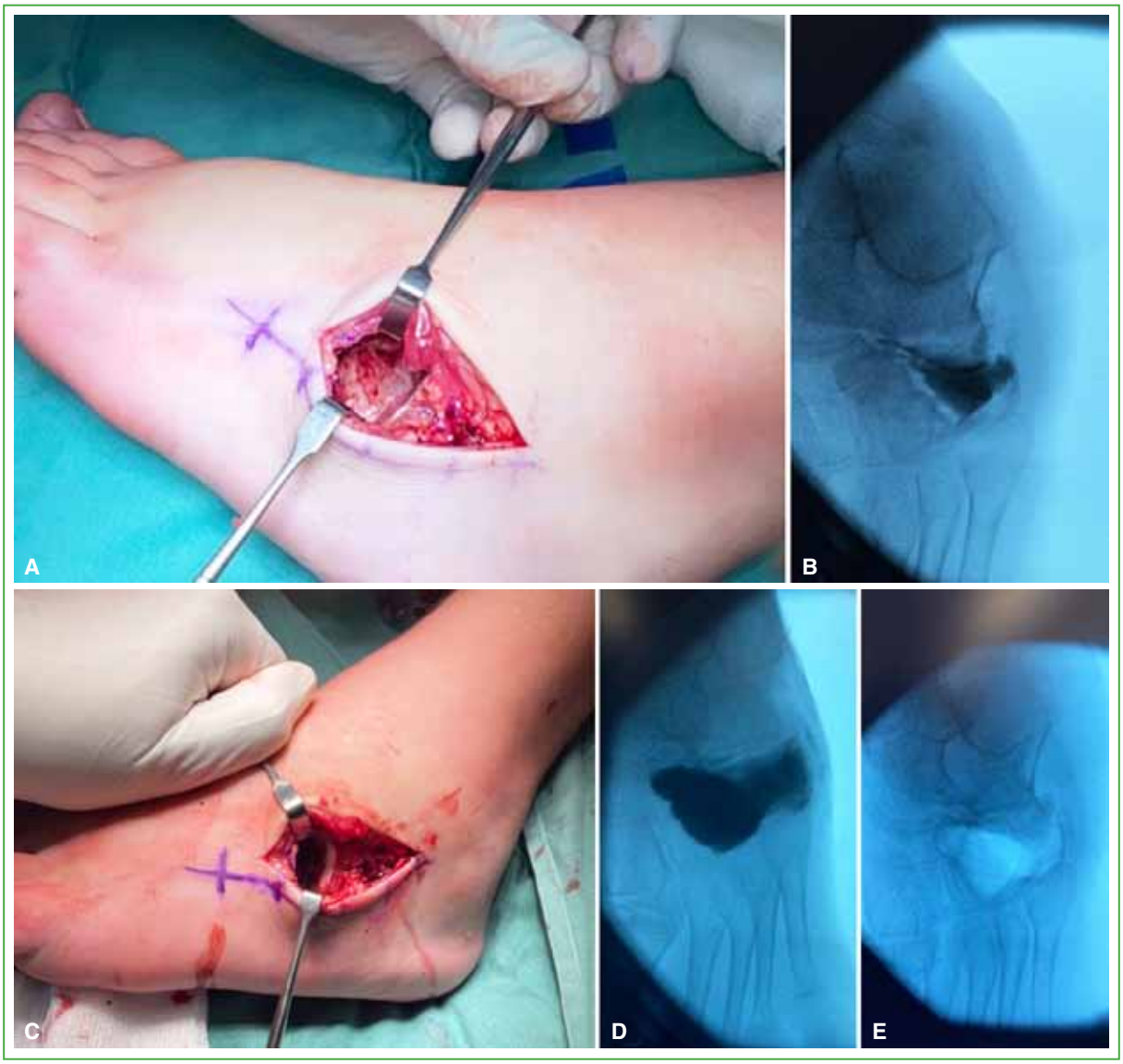

Figure 7. **A/B** Curettage of membranes, use of contrast medium to evaluate the resection of the cavity, the septated cyst can be seen. **C/D/E** After extraction of all the membranes, cautious curettage is required to avoid fracturing the cortical walls. It is then assessed with contrast, the difference between the **B-D**images can be appreciated.

Figure 8. The allograft of lyophilized bone from the INDT bank was rehydrated with saline solution for 20 minutes and cut before placement.

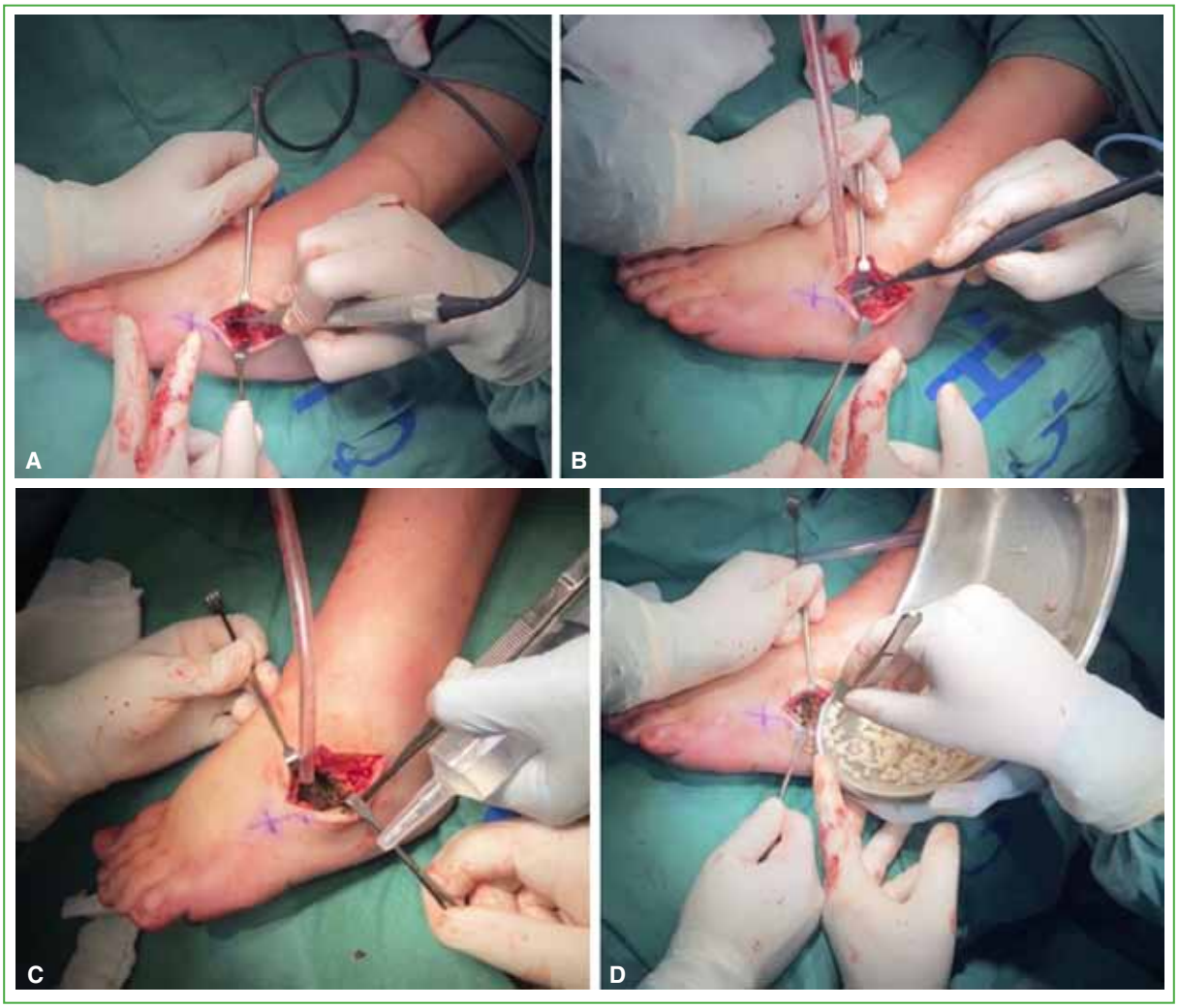

Figure 9. **A-B** High-speed drilling, electrocautery in spray mode as an adjuvant method for the excision of the membranes. **C-D** Cavity filling with lyophilized bone allograft with cortico-cancellous chips.

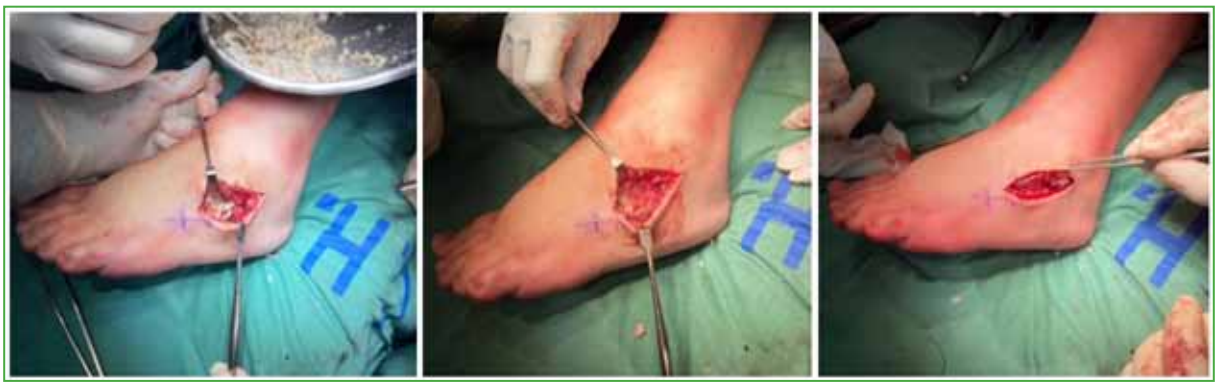

Figure 10. After filling the cavity, closure of the bone window and closure by planes is performed.

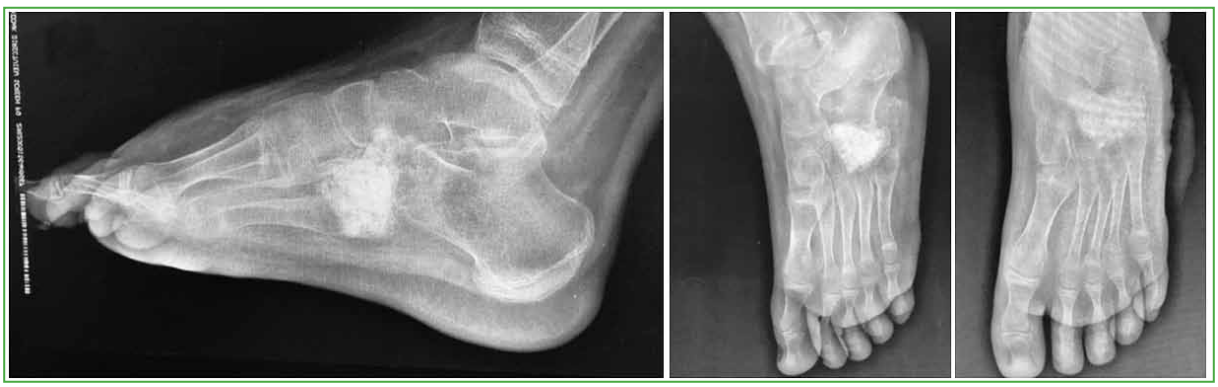

Figure 11. Postoperative foot radiographs, lateral, oblique and AP views. Graft filling can be appreciated without invading the cortices of the cuboid bone.

Postoperative period

The patient suffered no complications in the immediate postoperative period (Figure 11). He was discharged 48 hours later, with instructions to rest, avoid weight-bearing on the limb (walking on crutches), and take nonsteroidal anti-inflammatory drugs. Partial weight-bearing was allowed one month after surgery and the load began to be increased progressively every week.

One week after the operation, he consulted in the emergency department due to local swelling accompanied by serosanguineous exudate at the surgical site (Figure 12). He had no symptoms of other major physiological systems, no fever, and no elements of osteoarticular infection in the hip, knee, or ankle. The laboratory analysis revealed acute phase reactants in the normal range.

The symptoms disappeared after watchful waiting with physical measures (rest, nonsteroidal anti-inflammatory drugs).

Figure 12. Clinical images a week after surgery. Swelling and local serosanguineous exudate; acute phase reactant analyses were performed, which were normal.

Follow-up

One month following surgery, partial weight-bearing was permitted, with the load gradually increasing weekly.

We want to highlight that the follow-up of the patient was carried out bi-monthly for the first 6 months (Figures 13, 14, 15) and then after a year because he could not attend for personal reasons (Figures 16, 17).

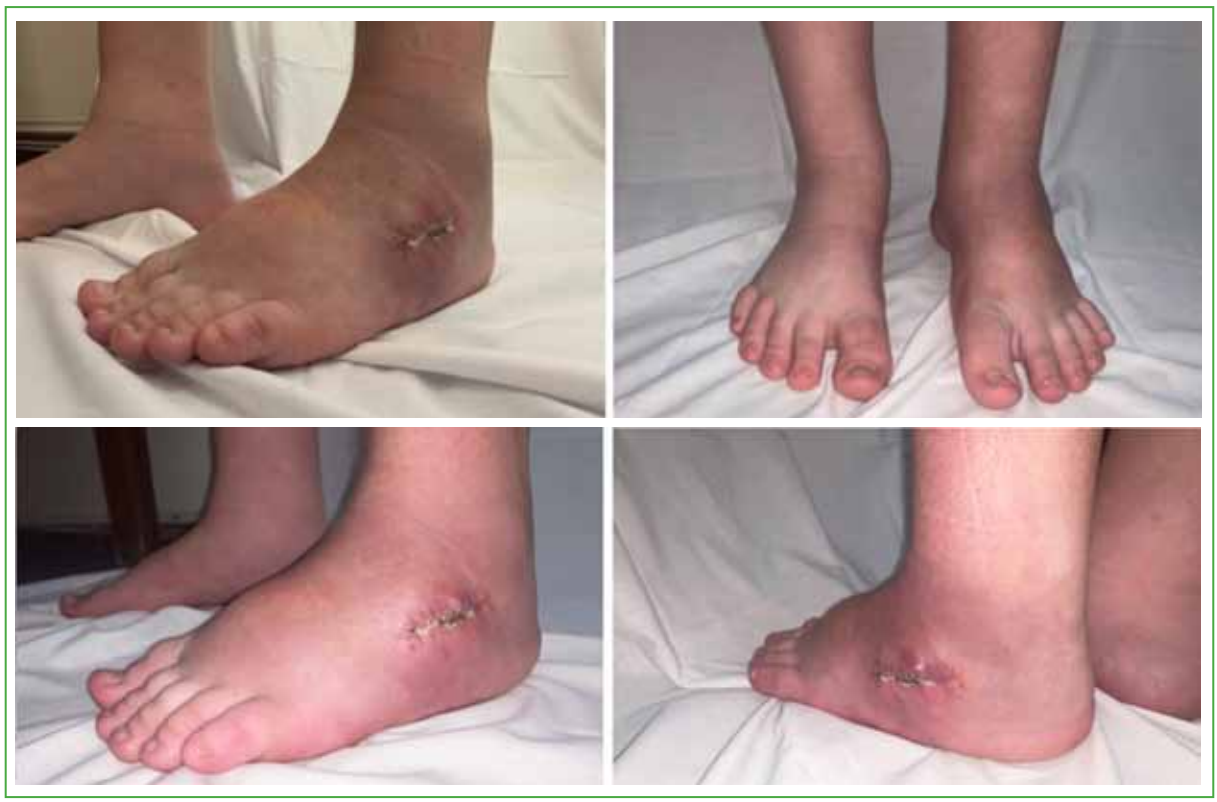

Figure 13. Clinical appearance after 2 months. Patient walking without crutches, without pain, reintegrating into all his activities of daily living.

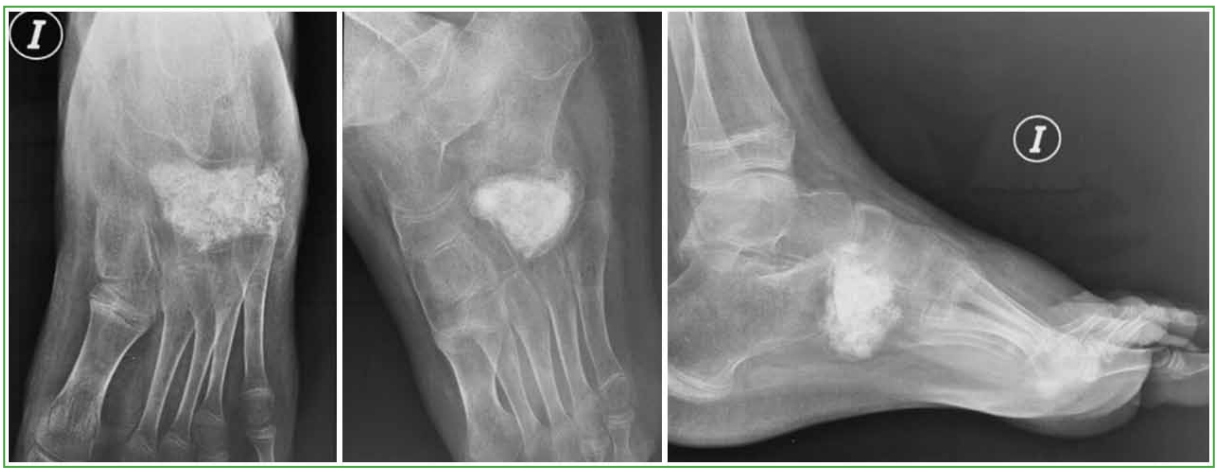

Figure 14. Foot radiographs after 2 months, AP, oblique, and lateral views. The bone graft can be seen at the level of the cuboid, without fractures, preservation of its joint relations with the calcaneus at the level of the hindfoot and the 4th and 5th metatarsals at the level of the forefoot. The skeleton progressively begins to reossify when given mechanical stimulation.

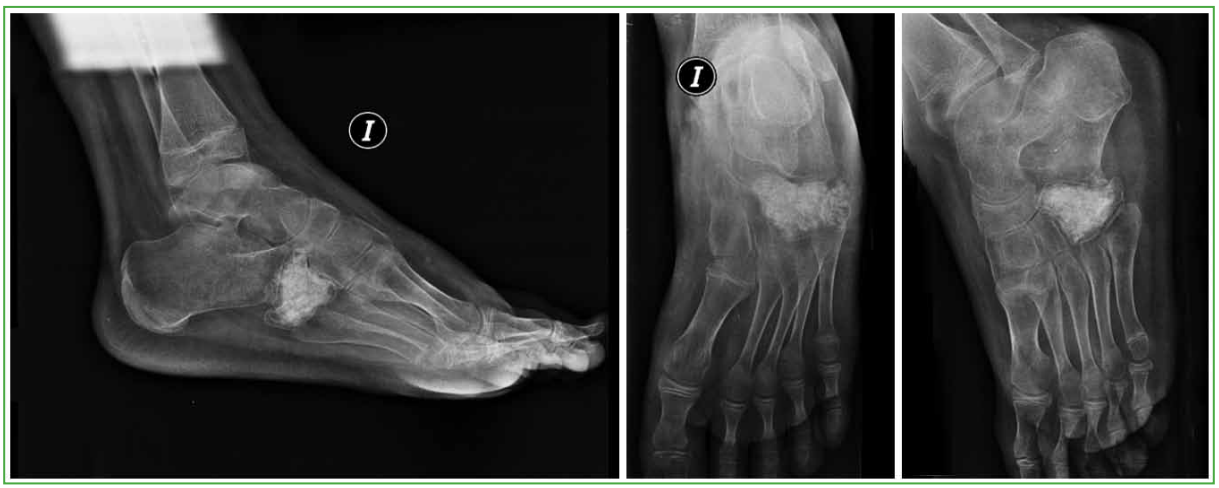

Figure 15. Radiographic postoperative control at 4 months. Lateral, AP, and oblique foot views.

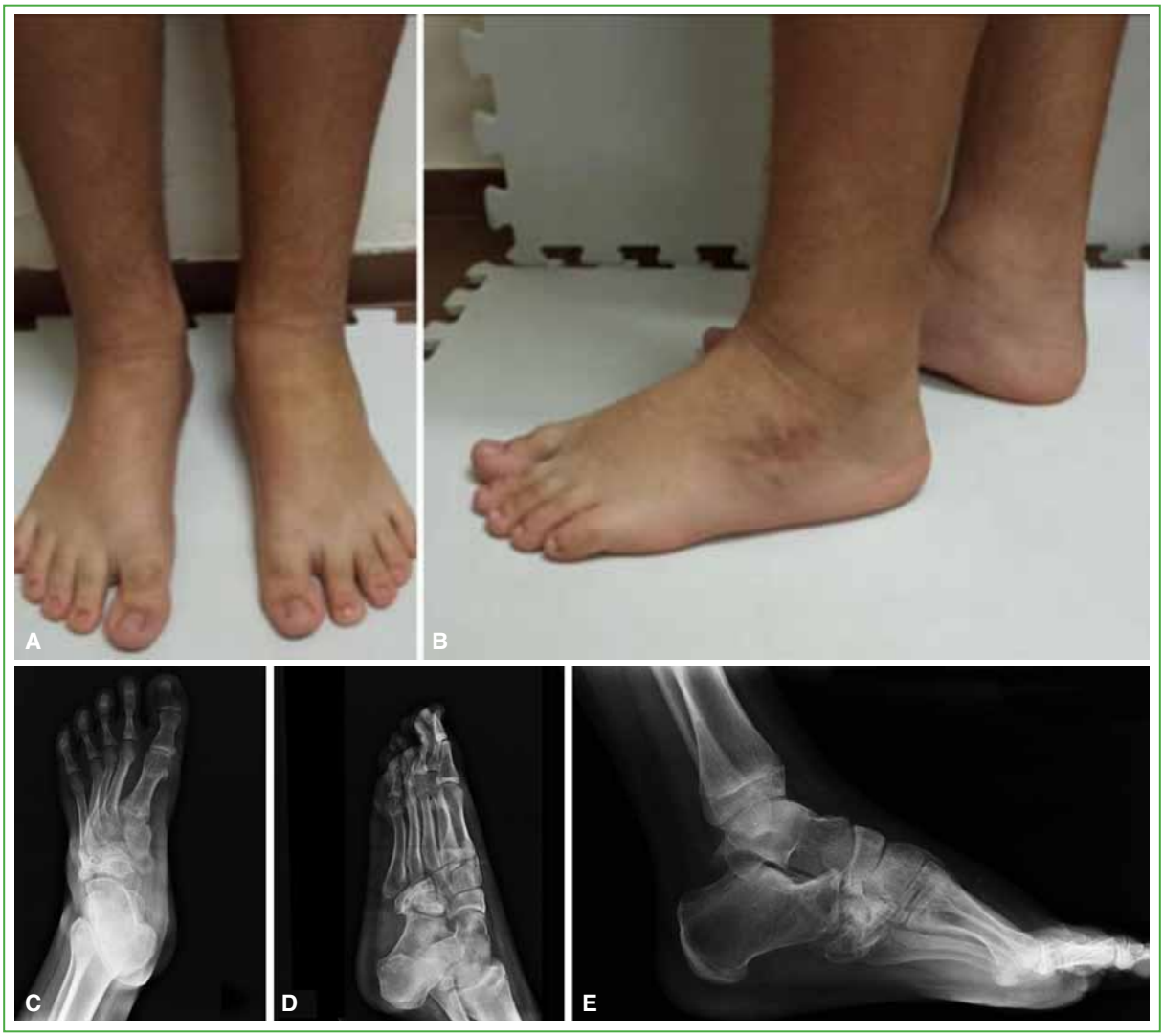

Figure 16. **A/B** Clinical - Radiographic control at 12 months **C/D/E**.

Figure 17. Clinical control / Walking without crutches, without pain.

DISCUSSION

The location of an ABC in the foot is rare and, in the cuboid bone, it is exceptional. Although it is suggested that its etiology is unknown, one of the theories that is used today is that ABC may correspond to intraosseous arteriovenous malformations surrounded by a thin layer of periosteum, and beyond the fact that it can appear in any bone, the vast majority manifest at the metaphyseal or metaphyseal-diaphyseal level of long bones. It is estimated that the incidence in the bones of the foot is 5-9% and its incidence in the cuboid is unknown.[2,3,6,7,8,9,10,11,12,13]

It should be noted that this patient had a late diagnosis. He was referred to our center after multiple consultations for foot neck trauma in the emergency department where he was not evaluated with radiographic approaches and was treated with physical rest, nonsteroidal anti-inflammatory drugs, and local ice, as if he had a sprain.

Our initial clinical-radiological approach was correlated with the results of the biopsy, and it was possible to administer a successful treatment.

The evolutionary phase of the ABC can be classified according to the Enneking stages; in our case, it corresponded to stage 2 (active).

The indications for surgery after diagnosis are: pain, pathological fracture, risk of fracture, large cysts located in weight-bearing areas. The symptoms that prevailed in our patient were: pain and inability to use the limb for weight-bearing, standing, and walking.

Multiple therapeutic modalities for ABC have been described; the traditional method of intralesional curettage plus grafting has a recurrence rate ranging between 20% and 41.6%.[14,15,16,1718] In our center, it was 37.5%.

At present, several techniques such as phenol, alcohol, polymethylmethacrylate, high-speed drilling, electrocautery, liquid nitrogen, and argon laser coagulation are used as adjuvant procedures to curettage in order to increase surgical oncological margins and prevent recurrence.[1,2,3,6-13,19,20,21,22,23,24,25]

It should be noted that the graft filling in the bone cavity is of vital importance, and that it can be carried out with bone autograft or allograft. In our patient, we opted for curettage and scraping of the membranes, electrocautery in spray mode, and high-speed drilling, in order to increase the margin of tumor cell necrosis. Lyophilized bone allograft was used for filling.

The recurrence is diagnosed by classification based on the radiological result of the treatment, according to the Capanna scheme that specifies four possible types of therapeutic response: grade 1, cured; grade 2, incomplete healing; grade 3, recurrence; grade 4, no response. Grades 1 and 2 are defined as success, while grades 3 and 4 represent therapeutic failure.[1,2,3,4,5,6,7,8,9,10,11,12,13] Our case was grade 1. Recurrence in this type of location is not described.

Our patient was monitored bi-monthly during the first semester, and then after a year, because the patient could not attend the controls for personal reasons.

We finally managed to contact him after a year in our external polyclinic service, his clinical evolution was very favorable, he did not suffer pain in the ankle and the neck of the foot, the symptoms had completely disappeared and he had resumed his school and sports activities.

CONCLUSIONS

ABC is uncommon in foot bones, with the metatarsal being the most afflicted and, in rare cases, the cuboid. This condition poses diagnostic difficulties and its location may go unnoticed. Clinical suspicion should always be maintained in young patients who suffer low-energy trauma and come to the emergency department with long-standing pain associated with a tumor process.

Despite the late diagnosis, the indication of correct therapy made it possible to timely prevent the natural progression of the disease with one of its possible complications, such as fracture, and an improvement in the patient's quality of life.

Given the scarcity of published cases, we believe it is important to share our experience in managing the lesion and recommend that the treatment of ABC with intralesional curettage, high-speed drilling, electrocautery, and filling with lyophilized bone allograft with cortico-cancellous chips is safe for patients.

Conflict of interest: The authors declare no conflict of interest.

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