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Isolated Osteoblastoma of the Cuboid Bone: A Case Report and review of the literature

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Declaration of interests

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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Abstract

Osteoblastoma is a relatively rare, benign, bone-forming tumor, commonly observed in the second and third decades of life. Spine and the long tubular bones are the most common sites of involvement. Osteoblastoma is infrequently seen in other sites, including the bones of hand and foot. We report a rare case of a 35-year-old man that presented an osteoblastoma of the cuboid bone. The patient was treated with surgical resection and grafting. After the intervention, the patient recovered with no clinical and radiological evidence of recurrence after one year of follow-up.

Several cases of osteoblastoma-like variant of osteosarcoma of the cuboid have been previously reported, but, to our knowledge, this is the first case of conventional and isolated osteoblastoma involving the cuboid bone reported in the literature.

Keywords: bone-forming, bone tumors, midfoot, pathology, pain, rare disease

Introduction

Osteoblastoma is a locally aggressive, benign, bone-forming tumor, histologically similar to osteoid osteoma. It is a relatively rare lesion accounting for approximately 1% of all primary benign bone tumors. Osteoblastomas are typically larger than 1.5–2.0 cm in diameter. The lesion has an extremely rich vascular supply and macroscopically it appears usually red to red-brown in color, with a gritty cut surface due to the woven bone produced [1, 2]. It is commonly seen in the second and third decades of life, with a male to female ratio of 2:1 [3]. Spine and the long tubular bones are most frequently affected [4], while osteoblastoma of the bones of hand and foot is uncommon. To date, a few cases of osteoblastoma of the talus have been reported [5, 6, 7].

We describe a rare case of osteoblastoma located in the cuboid bone. To our knowledge, this is the first case reported in the literature of conventional and isolated osteoblastoma of the midfoot involving the cuboid bone.

Case Presentation

A 35-year-old male presented with severe mechanical pain on his left midfoot. He denied any previous significant trauma and pain at rest. Pain was present from 9 months despite non-steroidal anti-inflammatory drugs (NSAIDs) taking, and symptoms progressively worsened over the last 2 months.

Clinical examination did not reveal any swelling in the midfoot, but walking was very painful and caused him to limp. Foot movements were painful, especially active and passive ankle extension and flexion. The total American Orthopaedic Foot & Ankle Society (AOFAS) midfoot scale score was: 5/100. Laboratory investigations showed normality of acute-phase reactants (erythrocyte sedimentation rate, C-reactive protein), blood cell count with differential count, alkaline phosphatase, kidney and liver function.

Radiographs of the left foot showed a round shape, 2 cm, lesion with internal calcifications surrounded by a clear halo, in the superior aspect of the cuboid bone (Fig. 1A). Computed tomography (CT) of the ankle and foot confirmed a 2,5 cm expansive lesion of the superior border of the cuboid, with multiple small calcifications, causing cortical expansion and erosion (Fig. 1B, C). Enhanced Magnetic resonance imaging (MRI) revealed an isointense lesion with areas of decreased intensity corresponding to foci of calcification with marked enhancement after gadolinium injection. Enhancement of the surrounding soft tissues and edema of the postero-lateral aspect of the third cuneiform was also detectable (Fig. 1D, E). Biopsy of the lesion was performed, and histologic examination revealed findings consistent with the diagnosis of osteoblastoma (Fig. 1F) and phasebone scan with technetium-99 showing increased uptake of the cuboid bone(Fig. 1G).

The patient subsequently underwent complete surgical resection of the cuboid bone. Under general anaesthesia, the patient was placed in a supine surgical position. The lesion was approached through a dorso-lateral incision (Fig. 2A, B, C). The complete surgical resection of the cuboid was subsequently performed and the cavity filled with bone graft taken from the superior side of the iliac bone. Platelet rich

plasma (PRP) injection was carried out to encourage healing and to reduce inflammation.

The inflamed soft tissue on the lateral side of the midfoot was also excised. The histological examination confirmed the diagnosis of osteoblastoma, showing a bone-forming tumor composed of anastomosing trabeculae of woven bone, surrounded by osteoblasts without significant cellular atypia, intermingled with osteoclasts, in a rich vascular fibrous stroma (Fig. 3A, B, C); no necrosis nor mitotic activity were seen. There was no evidence of infiltration of soft tissues surrounding the neoplasm.

Postoperatively, the patient was managed in below-knee aircast for 3 months with non-weight bearing to avoid graft mobilization. Postoperative radiographs showed that the bone tumor was replaced by bone graft (Fig. 4A, B, C). After 3-month follow-up, he regained the normal range of motion of the left foot and weight-bearing was permitted.

Follow-up MRI and CT scans were performed at 3, 6 and 12 months, and showed progressively osseointegration and decreased bone marrow and soft tissue edema (Fig. 5A, B, C) and (Fig.6A, B, C, D) and (Fig.7A, B, C). No radiologic evidence of recurrence was observed at the final 12-month follow-up visit (Fig. 7A, B, C). At 1-year visit, the patient was asymptomatic, and the clinical evaluation showed a slight limitation of supination (Fig. 8B) with an AOFAS midfoot scale score of 85/100 points.

Discussion

Osteoblastoma was firstly described by Jaffe and Mayer in 1932 [8], when the Authors reported a case of “an osteoblastic osteoid-tissue- forming tumor of the metacarpal bone”. In 1956 the term benign osteoblastoma was coined by Lichtenstein [9] to emphasize its benign nature and histologically conspicuous proliferating osteoblasts. It usually affects adolescents or young adults, with 70–80% of cases diagnosed in the first three decades of life. Pain is the most common presenting symptom, being present in 80–90% of patients [4]. Our patient was 35 years old, and pain and limping were the only symptoms. Unlikely osteoid osteoma, the pain is less responsive to salicylates and NSAIDs, and it is usually absent at rest. Though histologic features of osteoblastoma minimally differ from osteoid osteoma, the former usually achieves a larger size and has a more aggressive course [10]. Macroscopically, the lesion has a rich vascular supply, and is composed of trabeculae of osteoid and woven bone. The osseous spicules are irregularly arranged, and are lined by a single layer of osteoblasts, a phenomenon termed “osteoblastic rimming” [2]. Mayer et al [2, 11] described a subgroup of osteoblastoma, characterized by prominent epithelioid osteoblasts, previously known as aggressive osteoblastoma.

As illustrated in Fig.3 A, B, C, the histologic findings were consistent with the diagnosis of benign osteoblastoma.

To the best of our knowledge, this is the first case reported in literature of osteoblastoma localized in cuboid bone. In the literature, two cases of osteoblastoma-like osteosarcoma (OBLOS) involving the cuboid bone have been described [6 ,7]. Weiliang Wu et al. [7] reported the case of an osteoblastoma-like osteosarcoma of the cuboid bone and skull in a 12-year-old boy, while Kumar et al.

[6] described the case of an OBLOS of the cuboid bone in a 32-year-old man. OBLOS and osteoblastoma share similar clinical and radiological characteristics, but OBLOS behaves like osteosarcoma. Differential diagnosis is mainly histologic. OBLOS can be distinguished from osteoblastoma by the presence of permeation of the surrounding tissue, lack of maturation toward the edges [12], and by increased mitotic activity with atypical mitotic figures. On the contrary, like in our case, in osteoblastoma the mitotic activity is low or absent [13]. In our case, the lesion was considered benign because pathologic examination revealed osteoblastic proliferation, with no significant cellular atypia and low mitotic activity.

The radiologic appearance of the osteoblastoma can be confusing and makes the diagnosis difficult. Acrometastases of the foot, although very rare, have previously been described by Maccauro et al. [14] and must be considered in the differential diagnosis when an osteolytic bone lesion with ill-defined margins is present. Frequent radiologic appearance of osteoblastoma is that of a lytic lesion with or without matrix mineralization, surrounded by a narrow or broader zone of sclerosis - unlike most of the aggressive lesions- or, if expansive, a thin bony shell, periosteal reaction may be present [3, 4, 15]. CT and MRI can give additional information regarding the precise tumour origin, intraosseous and soft tissues extension [16].

In our case we found a round shape 2,5 cm expansive lesion of the superior border of the cuboid, with multiple small calcifications; cortical expansion and erosions enhanced MRI scans showed an hypo-isointense lesion with areas of decreased intensity corresponding to foci of calcification with avid enhancement after gadolinium injection due to the high blood supply. Enhancement of the surrounding soft tissues and edema of the postero-lateral aspect of the third cuneiform was also seen.

The treatment of choice of osteoblastoma is complete excision of the lesion usually followed by bone grafting or bone cement. In a report of 20 cases, Saglik et al. [17] found that osteoblastoma can be treated successfully with curettage, but wide excision should be considered along with careful follow-up over the long-term owing to the possibility of recurrence or malignant transformation. Our patient underwent complete surgical resection of the cuboid followed by iliac bone grafting and PRP injection. This treatment was found to be effective as no recurrence was found after one-year follow-up.

Conflict of interest

The authors declare that they have no conflict of interest.

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Figure Legends

Figure 1: Figure.1 X-ray, Computed tomography, MRI and hystologic images of the right foot at initial admission. laterl **(A)**. X-ray image showing round shape 2 cm lesion with internal calcifications surrounded by a clear halo, in the superior aspect of the cuboid bone, axial **(B),** and sagittal **(C)**. computed tomography images showing expansile lesion of the superior border of the cuboid, with multiple small calcifications and cortical erosion and periosteal reaction. **(D),** TSE T1W sagittal MRI image showing an hypointense lesion in the superior aspect of the cuboid bone. **(E)** T1W FS post-contrast MRI on sagittal plane showing an

hyperintense lesion with areas of decreased intensity corresponding to foci of calcification. Enhancement of the surrounding soft tissues and edema of the postero-lateral aspect of the third cuneiform can also be seen. Scan view **(F)**of bioptic fragments some of which showing normal trabecular bone tissue, others showing the tumoral proliferation admixed with necrotic areas. Ematoxylin and eosin stain. Original magnification: 10xtrabecular bone tissue, others showing the tumoral proliferation admixed with necrotic areas. Ematoxylin and eosin stain. Original magnification: 10x**.**

Phasebone scan with technetium-99 showing increased uptake of the cuboid bone**(G)**

Figure 2: Photos **(A,B,C)**show complete surgical resection of the cuboid through a lateral-dorsal incision; the cavity was then filled with iliac crest bone graft fixed with two K-wires

Figure 3: (A) Pathologic examination reveals osteoblasts and osteoclast-like giant cells, with ectatic blood vessels and anastomosing irregular bony trabeculae rimmed by a single layer of osteoblasts. (**B, C)** On higher magnification, areas with predominance of osteoblasts without significant cellular atypia **(B)** and areas with osteaoclast-like giant cells (**c**). A, b, c: ematoxylin and eosin stain. Original magnifications**: A)** 100x; **B,C)** 200x

Figure 4: Plain radiographs on lateral **(A)**, oblique **(B)** and antero-posterior **(C)** view show complete surgical resection of the cuboid and iliac bone crest graft fixed with two K-wires

Figure 5: Clinical and radiological evaluation after 3 months. Sagittal STIR image showing bone edema of the graft, of the lateral aspect of the third cuneiform and of the navicular bone **(A)**. Post-contrast coronal T1 FS image showing enhancement of the soft tissues surrounding the graft **(B)**. CT scan on axial plane showing initial osseointegration **(C)**. Photo of the left foot shows a scar in the lateral aspect of the left foot **(D)**.

Figure 6: Clinical and radiological evaluation after 6 month. Plain radiograph on oblique plain and CT scans on sagittal and coronal plane showing progressive osseointegration **(A, B, C)**. Post-contrast T1 FS image on sagittal plane showing bone enhancement of the graft, of the calcaneus, of the talus and of the third metatarsal bone for algodystrophy changes **(D)**. Photo of the left foot shows a scar in the lateral aspect of the left foot **(E)**.

Figure 7: Radiological evaluation after 12-month. Plain radiograph on oblique plain and CT scan on coronal plane show osseointegration of the graft **(A, B).** STIR MRI image on sagittal plane showing residual bone edema of the graft and of the calcaneus. No recurrence is evident **(C).**

Figure 8: Clinical evaluation after 12 months a limitation of supination movement of the foot **(B)**.

Highlights

- Osteoblastoma is a locally aggressive, benign, bone-forming tumor
- The bones of hand and foot are uncommon localizations
- Osteoblastoma of the cuboid bone is extremely rare

Figure

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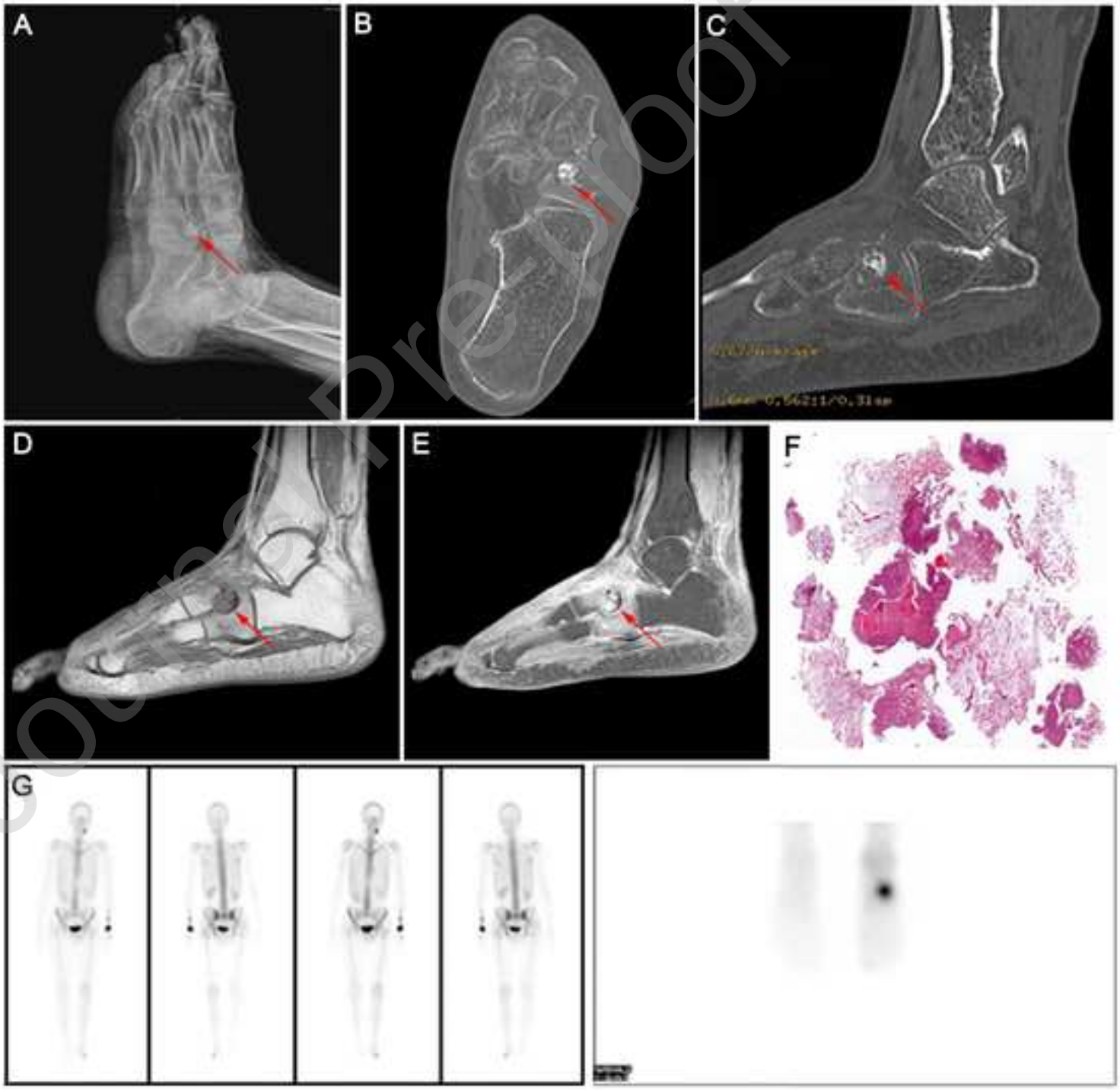

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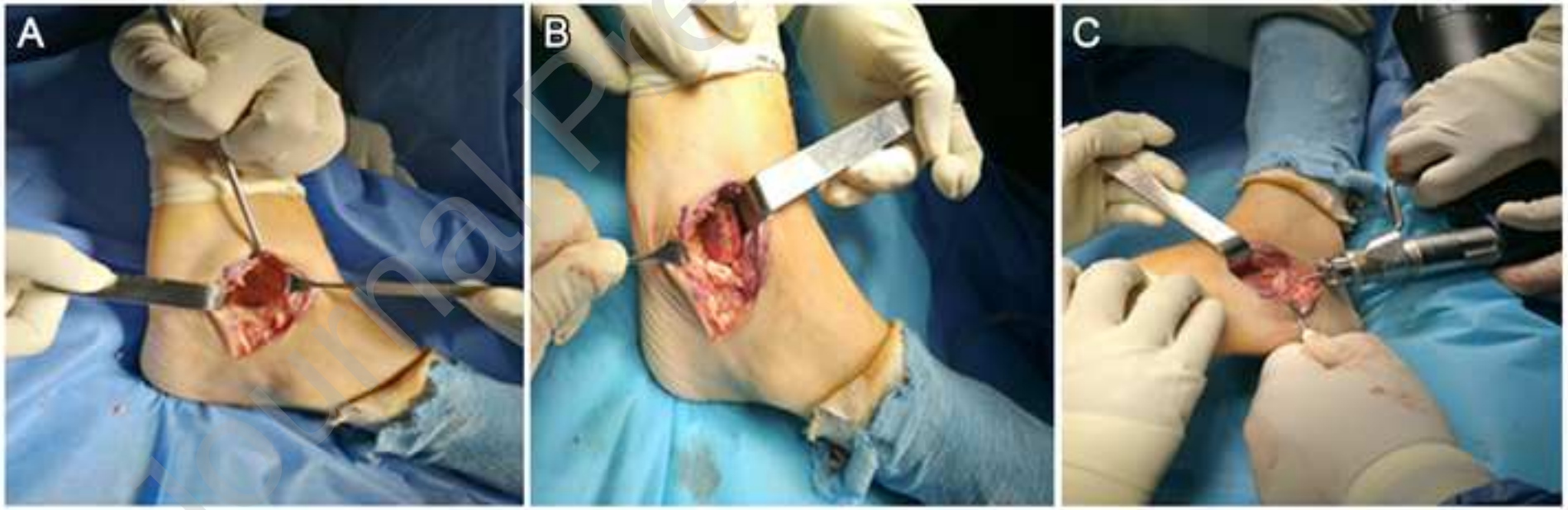

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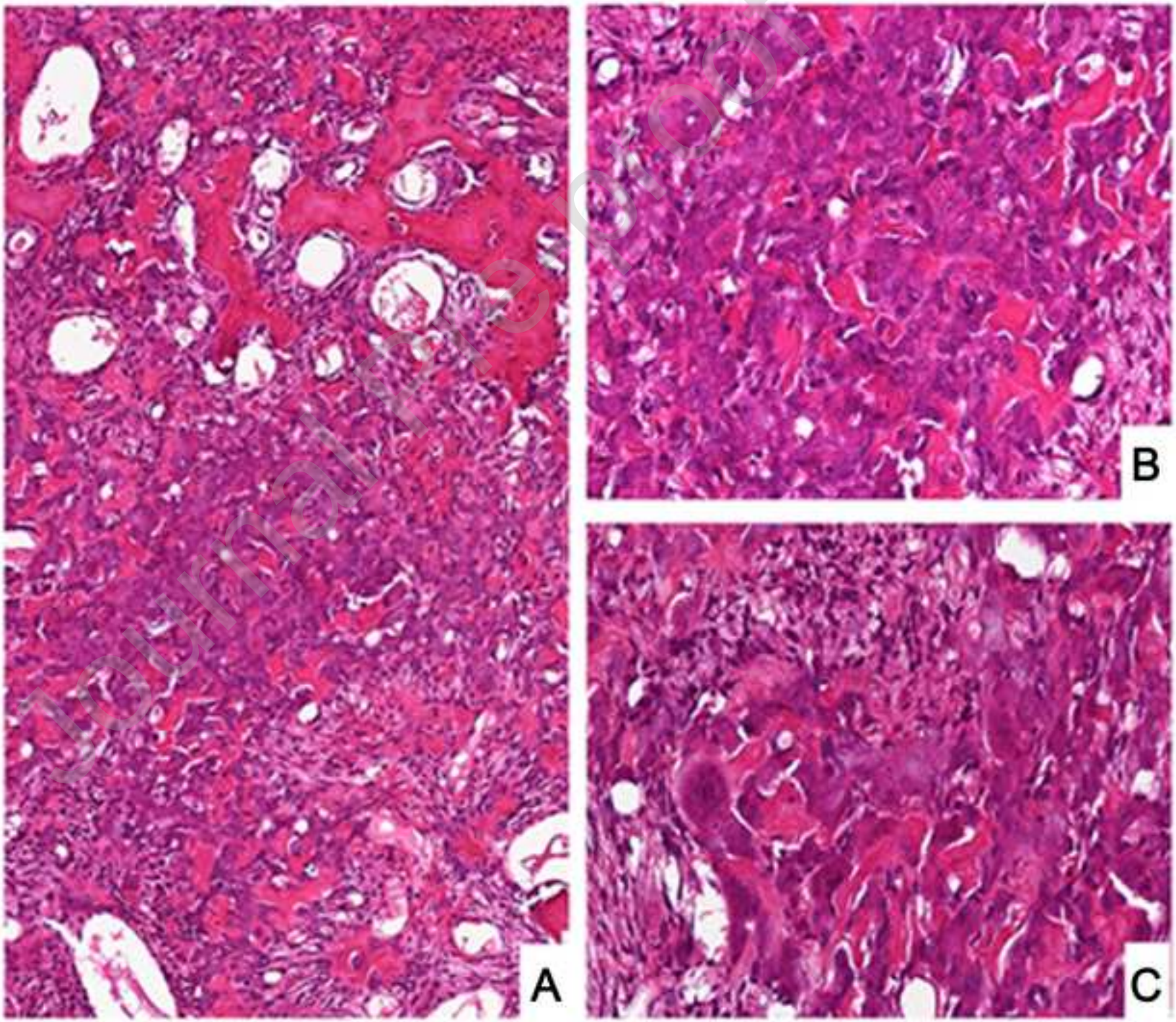

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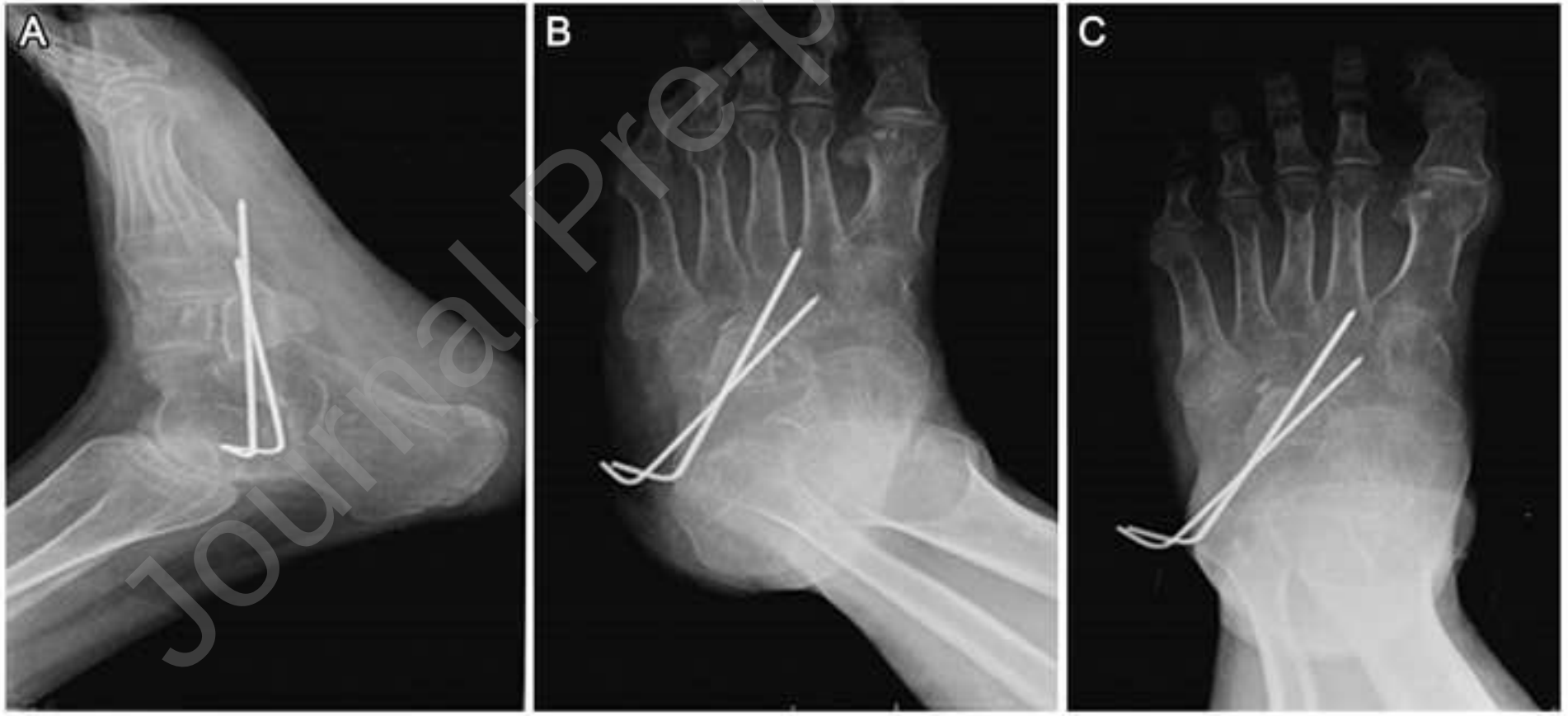

Figure

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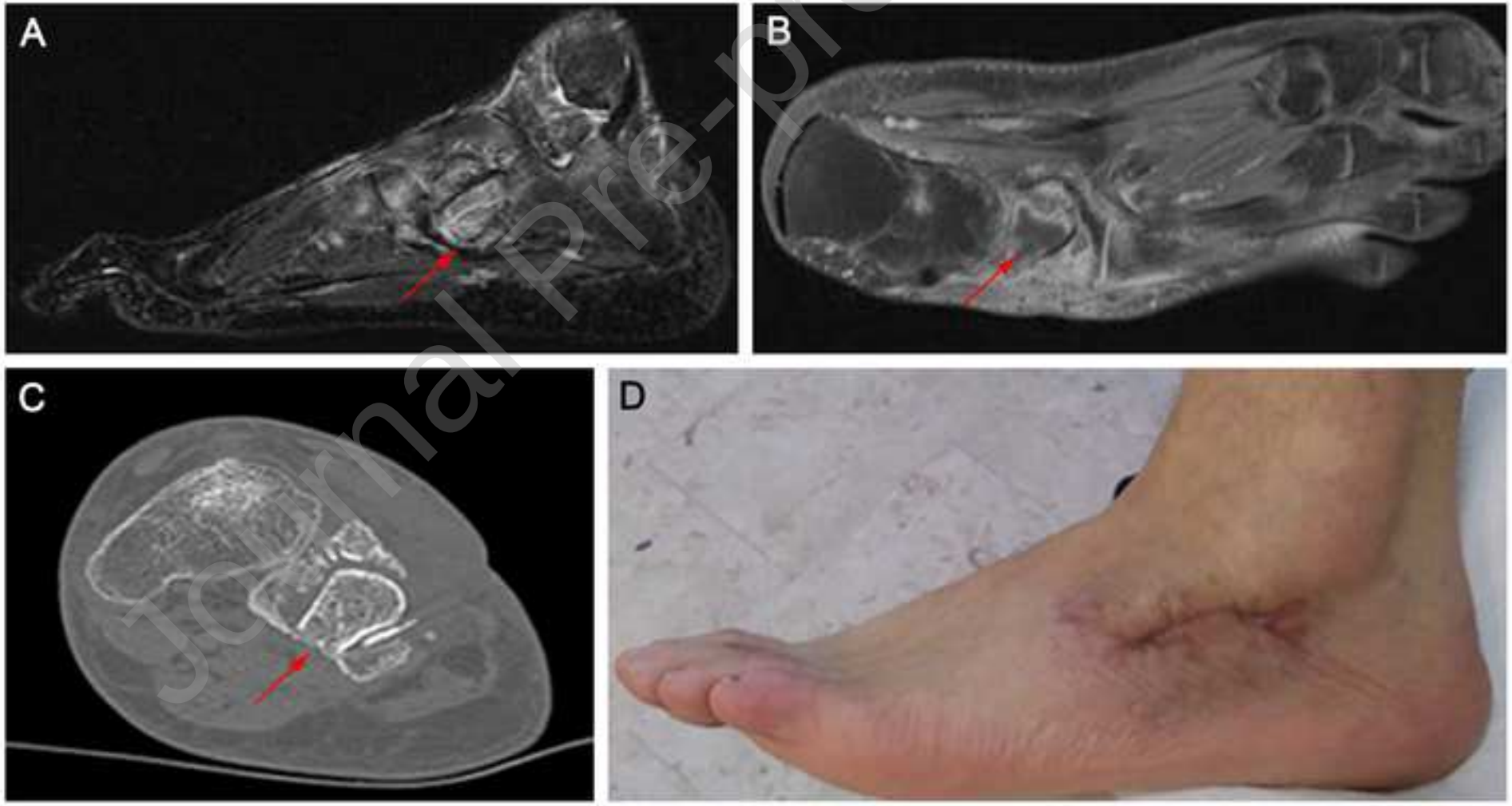

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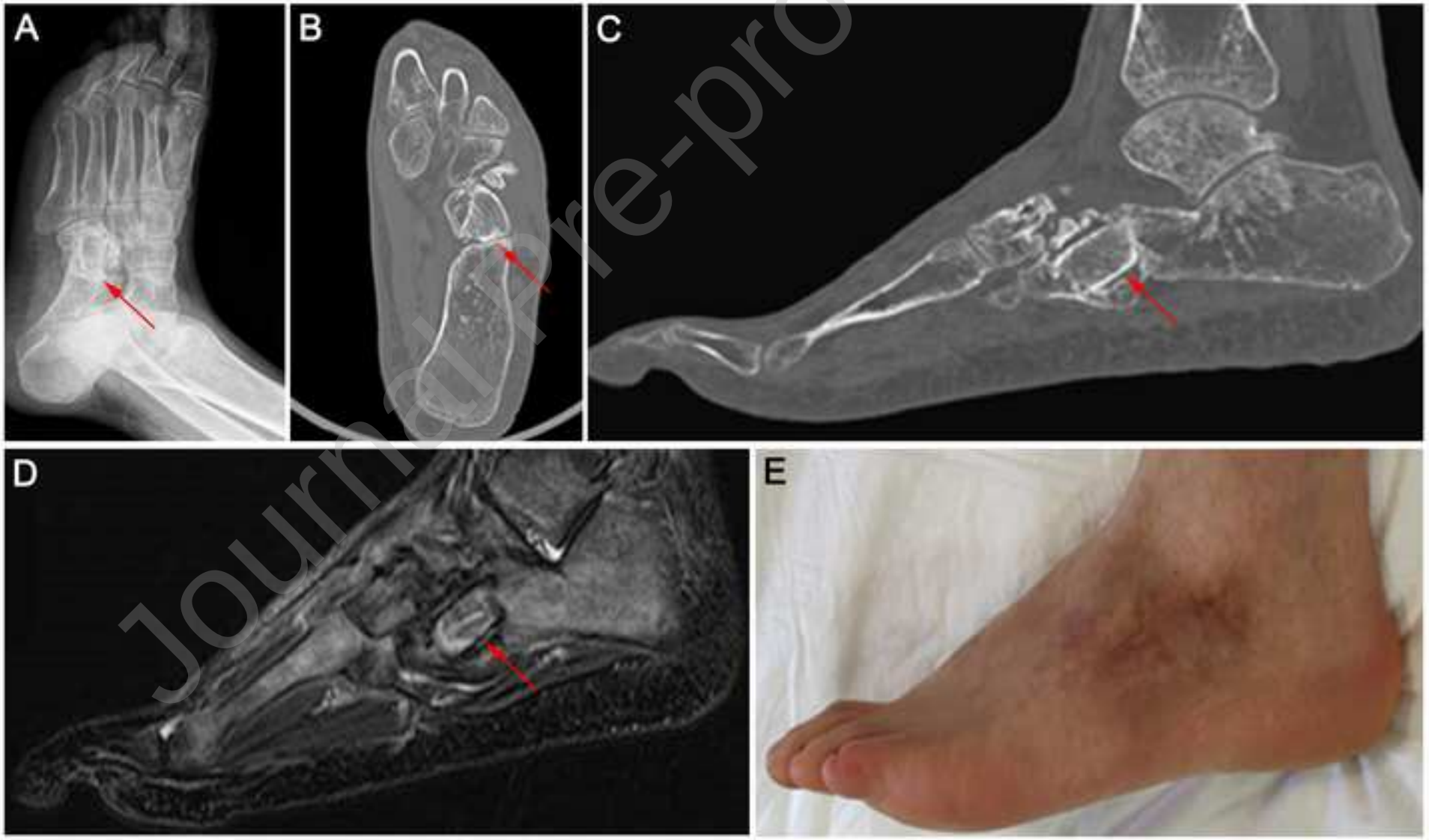

Figure

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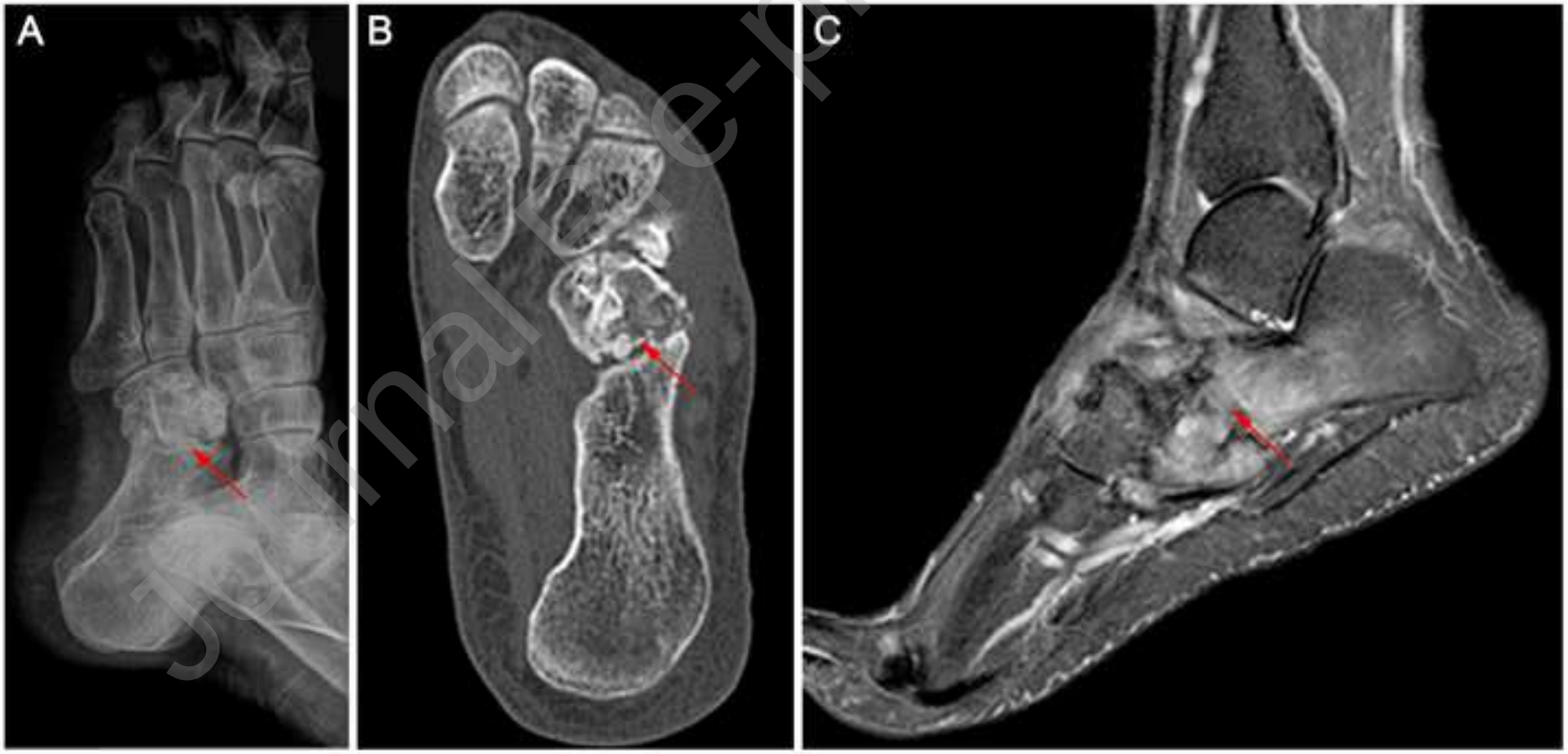

Figure

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