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CASE REPORT

Soft tissue recurrence of an osteoid osteoma: an exceptional observation

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Abstract

We report the observation of the soft tissue recurrence of an osteoid osteoma (OO) in a 26-year-old man initially complaining of post-traumatic pain and swelling of the right ankle. A first arthroscopic resection was performed after the misdiagnosis of "bone irregularities" observed on computed tomography (CT) and magnetic resonance imaging (MRI). The diagnosis of OO was made by histological analysis of the resection material. The patient became asymptomatic for 5 years until the symptoms progressively recurred. Follow-up MRI and CT studies demonstrated a nodular bony focus within the periarticular soft tissues of the ankle. The lesion was removed, and histological analysis confirmed the diagnosis of a whole viable OO. This observation likely resulted from the displacement of the initial lesion during the initial arthroscopic procedure. This case report highlights the possibility of recurrence of OO in the soft tissues.

Keywords Bone · Soft tissue · Tumor, osteoid osteoma · Computed tomography · MRI

Introduction

Osteoid osteoma (OO) in the lower limb most often involves the diaphyseal cortices [1]. Although OO of the feet is rare (2–11%), it represents 19.4% of benign tumors in the foot and ankle, with predominant involvement of the talus and calcaneus, particularly of the talar neck [2–4]. Depending on the location of OO, patients may present with local erythema, swelling, tenderness, bone deformity, gait disturbances, or muscle atrophy [5, 6]. The wide range of symptoms makes clinical diagnosis sometimes challenging, particularly in cases associated with traumatic or surgical history [6]. Intra-articular lesions are not exceptional, explaining particular imaging findings, such as lack of or presence of limited reactive sclerosis and periosteal reaction on computed tomography (CT) [5, 7] and prominent synovial hypertrophy and joint effusion, which can mimic degenerative, inflammatory, or infectious conditions [8].

Percutaneous CT-guided treatments have replaced surgical en bloc resection and most often consist of radiofrequency thermoablation, microwave ablation, cryoablation, and laser photocoagulation [2, 6, 9, 10]. These less invasive methods achieve results similar to those obtained with open surgery but do not provide histopathological proof [11, 12]. Articular OOs can be treated either by arthroscopic excision, open surgical resection, or percutaneous thermal ablation [13]. Arthroscopic treatment is often proposed for talar neck OOs [14–16].

Curative treatment requires complete resection/destruction of the nidus. Incomplete treatment leads to recurrence, usually in the initial location [17]. Recurrence at distance is very uncommon [18]. Treatment with radiofrequency thermoablation is associated with a failure rate of about 8%, while all types of treatment are cumulatively associated with a 10–16% failure rate [19]. Treatment failure has been reported at an average of 9 months post-treatment; however, recurrences may be observed up to 48 months [19].

We report an unusual observation of an OO of the talar neck presenting recurrence within the soft tissues several years after initial arthroscopic treatment. Only one previous report describing a recurrent OO in the soft tissues after primary treatment was found in the literature [7].

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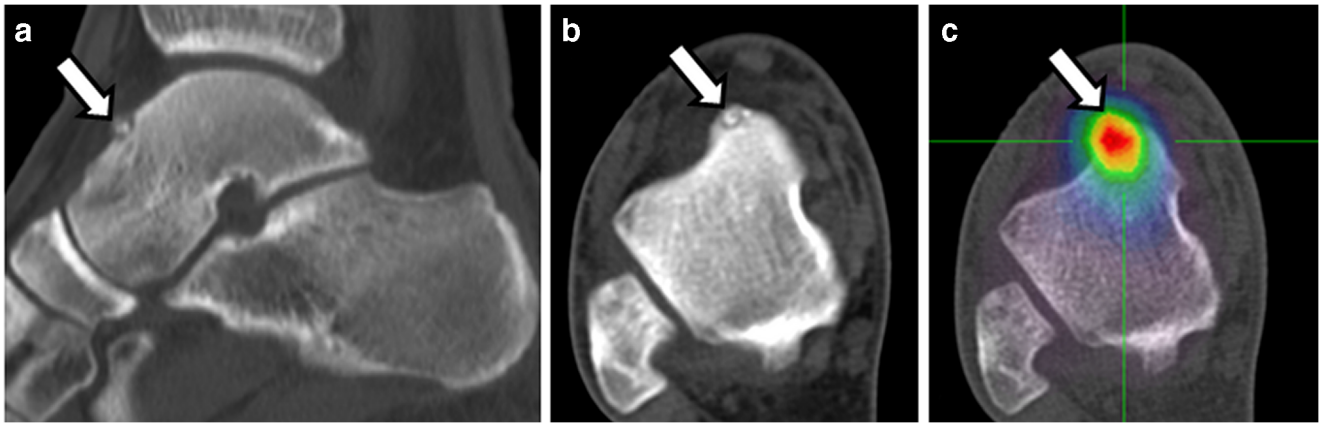

Fig. 1 Sagittal (**a**) and transverse (**b**) reformatted images of the spiral computed tomography (CT) obtained at the initial presentation show the presence of a small calcified lesion adjacent to the anterior aspect of the articular surface of the talus (talar dome) (arrows). Corresponding SPECT-CT image (**c**) shows that this focus presents an intense uptake of the tracer. Although the diagnosis of OO may seem evident on the CT image, this lesion was misdiagnosed as nonspecific osseous irregularities

Case report

A 26-year-old male consulted a physician in 2011 for persistent anterior pain and ankle swelling, evolving over 2 years after a motorcycle accident that was followed by a sports injury 1 year later. The pain was mainly located in the anteromedial aspect of the ankle and was relieved by anti-inflammatory drugs and wearing an ankle brace. He was advised to limit the sports practice. The clinical examination revealed a stable ankle, symmetrical mobility, and pain at palpation of the dorsal aspect of the talus.

A single-photon emission tomography/computed tomography (SPECT/CT) revealed an intense focal isotope uptake that was diagnosed as an osteochondral fracture or post-traumatic bony "spurs" (Fig. 1). A magnetic resonance imaging (MRI) study showed bone marrow edema with low signal intensity (SI) on T1-weighted images and high SI on fat-saturated T2-weighted images within the bone marrow on the medial aspect of the talus, associated with a tibiotalar joint effusion and synovitis (Fig. 2). Based on these findings, (peri) articular post-traumatic lesions were suspected.

A subsequent arthroscopy revealed a synovial proliferation of inflammatory appearance in the anteromedial aspect of the ankle, which was treated by synovectomy. The underlying bone showed a reddish tumefaction along the medial edge of the neck of the talus, which the surgeon described as an "osteochondral lesion." This lesion was biopsied and then curetted with a surgical mill. Microscopic examination of the resected tissue surprisingly indicated the presence of OO fragments (Fig. 3). The pathologist described a richly cellular and vascularized lesion with numerous osteoblasts arranged around spans of non-lamellar bone; however, complete or incomplete resection could not be confirmed. The postoperative course was favorable, and no postoperative imaging was performed. The patient returned to his daily professional and sports activities.

Five years after the arthroscopy procedure, the symptoms progressively reappeared and worsened. A medial exploratory

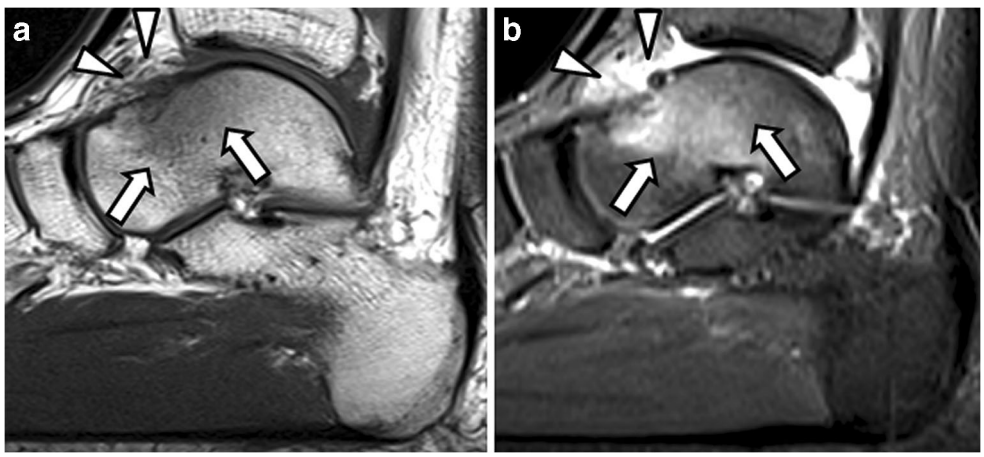

Fig. 2 Sagittal T1- (**a**) and fat-saturated T2-weighted (**b**) magnetic resonance (MR) images obtained at the initial presentation show the presence of bone marrow edema within the upper midportion of the talus (arrows) and synovitis of the tibiotalar joint (arrowheads). The nidus of the OO is seen on the anterior aspect of the articular surface of the talar dome

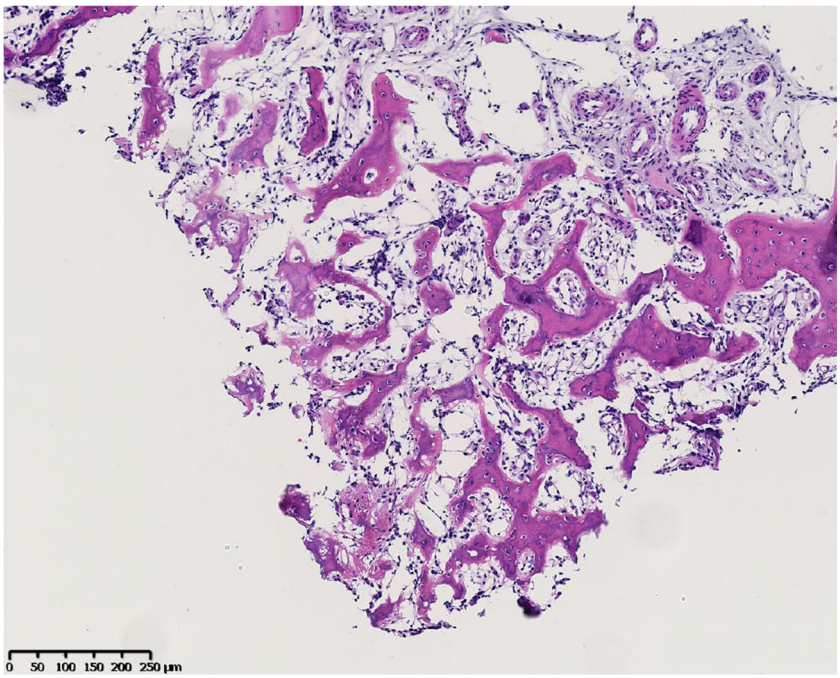

Fig. 3 Pathologic examination (hematoxylin and eosin staining) after the initial surgery. Histologic analysis of a bone fragment resected during the initial surgical procedure reveal an interlacking network of osteoid trabeculae showing variable mineralization and richly vascularized stroma, typical for a fragment of OO

arthrotomy was performed without an imaging update, and resection of inflammatory tissue and joint release were performed. Nonspecific fibrosis within the resected tissue was observed by the pathologist.

One year later, the patient was referred to our hospital. Since the last surgery, he had experienced variable and inconsistent pain in the anteromedial aspect of the ankle. The pain continued to increase, especially at night, and eventually prevented the patient from participating in sporting activities. The clinical differential diagnosis included mechanical impingement, scar pain, or recurrence of an OO. Follow-up MRI and CT scan suggested the presence of an OO in the soft tissues, approximatively 10 mm from its "previous retrospective" location on the neck of the talus. CT showed a rounded calcified nodule, and MRI revealed a richly vascularized lesion with characteristic early and intense enhancement on dynamic MRI sequences surrounded by inflammatory tissue (Figs. 4 and 5).

Open surgical resection directed to the soft tissues located at the anterior aspect of the talus neck was performed under local anesthesia. The calcified nodule and surrounding reactive tissue were completely removed, in addition to performing a synovectomy. A radiograph of the removed tissue confirmed resection of the whole calcified lesion (Fig. 6). Macroscopic and microscopic histological analyses confirmed the presence of a typical 11 × 8 mm whole OO in the fibrous tissue (Fig. 7). There were no postoperative complications. The patient's pain disappeared immediately after the operation. After 3 weeks, he returned to work and participated in sporting activities without limitation. At 3-year follow-up, there were no signs of recurrence.

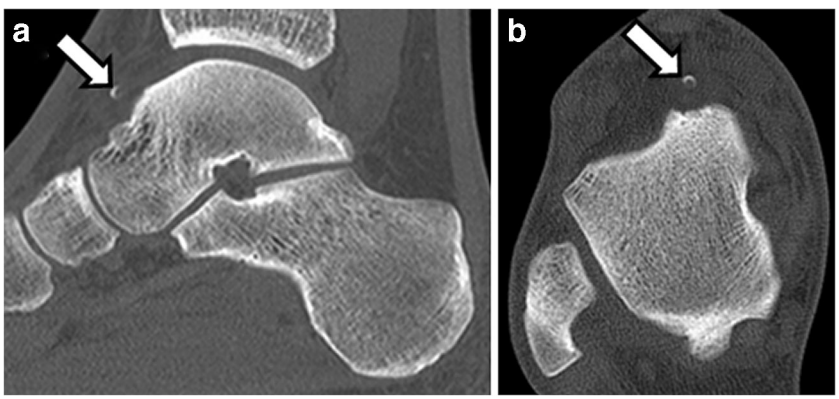

Fig. 4 Sagittal (**a**) and transverse (**b**) reformatted follow-up computed tomography (CT) images by the time of recurrence (at the same levels as Fig. 1) show "mobilization" of the OO within the soft tissues on the anterosuperior aspect of the talus (approximately 1 cm from its previous location) (arrows)

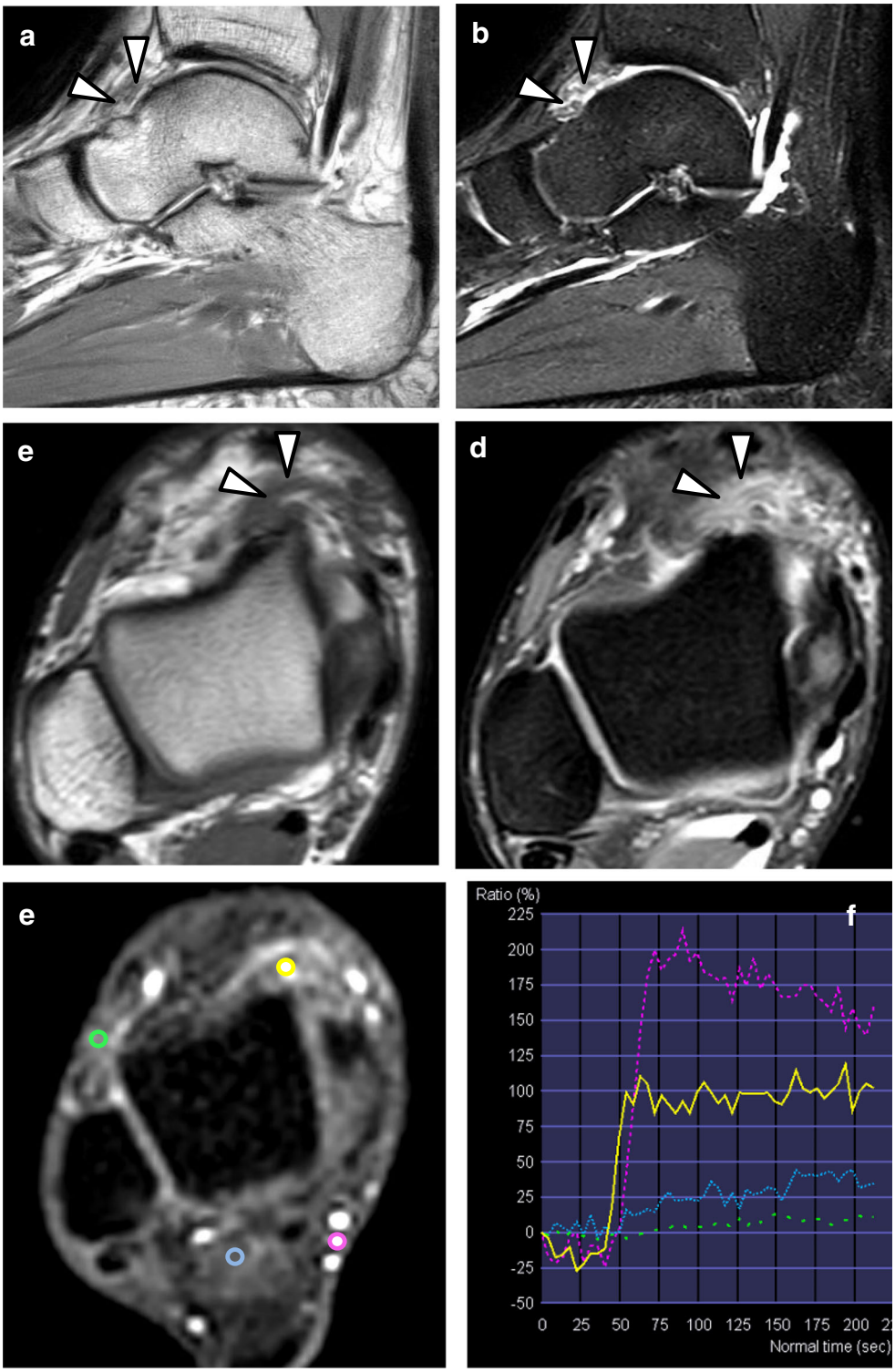

Fig. 5 (**a**, **b**) Sagittal T1- (**a**) and fat-saturated T2-weighted (**b**) magnetic resonance (MR) images obtained at the time of recurrence, 6 years after the previous MR examination, show the disappearance of bone marrow edema within the talus, lack of visibility of the nidus, but persisting inflammatory tissue on the anterior aspect of the tibiotalar joint (arrowheads). (**c**, **d**) Transverse MRI sections show the same tissue (arrowheads). (**e**, **f**) Transverse dynamic sequence (**e**) and enhancement curves (**f**) obtained after intravenous injection of gadolinium show early and intense enhancement within the center of the lesion (nidus, the yellow region of interest (ROI) and curve), paralleling the enhancement observed within the tibial posterior artery (rose ROI and curve). Slower and limited enhancement is seen within muscles (blue and green ROIs and curves)

Discussion

The present case reports a rare recurrence of an OO in the soft tissue 6 years after the initial arthroscopy following an early misdiagnosis. This case highlights the possibility of OO recurrence within the soft tissues. It underlines the importance of a careful analysis of the CT and MRI scans for the accurate and early diagnosis of an intra-articular OO, particularly in patients with a history of traumatic events, which may skew the clinical diagnosis of OO. Accurate diagnosis of an OO

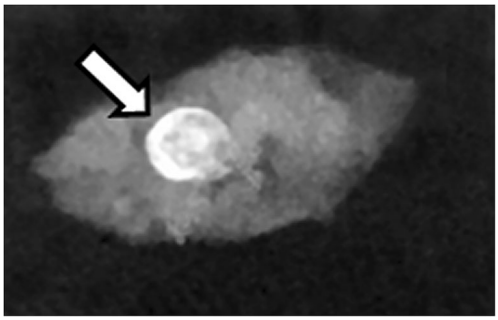

Fig. 6 Radiograph of the resected tissue performed during the surgical intervention shows the presence of a round calcified lesion within the resected tissue (arrow)

requires identification of the nidus on CT and analysis of the associated soft tissue and bone marrow changes, as well as vascularization of the lesion on MRI [5].

Early MRI and SPECT-CT demonstrated imaging features highly suggestive of an OO; however, these features were misinterpreted by the radiologist and nuclear medicine physician as bone irregularities, reactional edema, and synovial irritation related to the traumatic events mentioned on the imaging request. The initial radiologist may have had limited knowledge and experience regarding the typical appearance of OO in an articular location or may have been skewed by the history of traumatic events in the patient. The patient experienced pain after the traumatic events and associated his symptoms to those events. A recent case study reported the misdiagnosis of a talar neck OO in a patient presenting with a traumatic history [6].

The intra-articular location of OO, especially at the anterior margin of the articular surface of the talus, is not exceptional [3, 14, 16, 20, 21]. Imaging findings of intra-articular OO differ from those of the more frequent extra-articular locations [22]. Specifically, on CT, the reactive cortical thickening is minimal or absent in intra-articular OO, a finding believed to be due to a lack of periosteum, particularly of its deeper layer, the cambium, and only discrete reactive bone sclerosis can be seen within the cancellous bone [8]. In the present case, the CT displayed no reactive sclerosis, which can be misleading. Additionally, on MRI, the presence of severe synovial

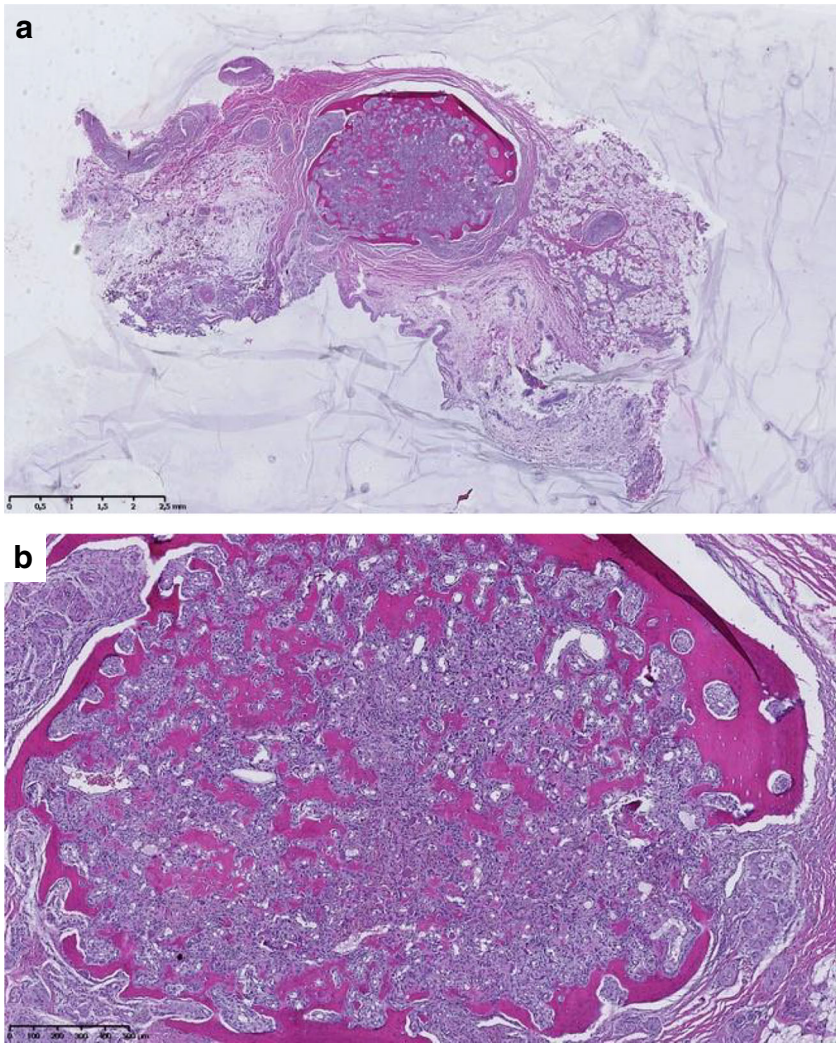

Fig. 7 (**a**, **b**) Pathologic examination (hematoxylin and eosin staining) after resection of the soft tissue lesion: macroscopic (**a**) and microscopic (**b**) appearance of the lesion reveal interlacking network of osteoid trabeculae showing variable mineralization and of a richly vascularized stroma. The nidus is completely surrounded by fibrous tissue

hypertrophy and a joint effusion accompanying intra-articular OO may be at the forefront and mimic degenerative, inflammatory, anterior impingement syndrome, or even septic arthritis. Moreover, a small nidus may be masked by extensive bone marrow and soft tissue edema, and traumatic injury or infection may then be suspected [8]. MRI is shown to have limited value in detecting the nidus when compared with CT [8, 23]; Jordan et al. reviewed 223 cases of foot and ankle OO, of which 34% missed the diagnosis based on MRI [24].

The present case showed a very late reappearance of symptoms after the first surgical procedure, which is beyond the usual time (9–48 months) of recurrence [19]. The lack of diagnostic imaging prior to the second intervention did not allow the diagnosis of OO recurrence. However, CT and MRI examinations performed in our center demonstrated features that suggested a "transplanted" viable OO in the soft tissues, which was confirmed by histopathological analysis following an open surgical resection [25–27]. Comparing the initial and 6-year follow-up images, it is likely that the first surgical intervention displaced the OO to the soft tissues.

Recurrence of OO at its initial location after primary treatment is not uncommon [17].

Observation of a soft tissue recurrence is exceptional [7]. Several hypotheses might explain this observation. First, the initial misdiagnosis may have caused suboptimal planning of the initial treatment. Second, the choice of arthroscopy may have complicated visualization and resection of the whole lesion and/or have caused loss of material/seeding during the procedure. Third, the articular location of the OO might have favored the soft tissue recurrence as the synovial cavity could provide a natural space for the lesion to persist.

Although arthroscopic resection is presented as the treatment of choice for OO by some authors [13], Dubuc et al. reported the deep recurrence of intra-articular OOs of the talar neck within the underlying cancellous bone after initial arthroscopic resection, which may have resulted from either marginal recurrence or may have been favored by the location of the OO within the joint capsule [18]. Malghem et al. reported a histologically confirmed case of OO recurring in the soft tissues beneath the femoral neck several months after surgical resection of its initial acetabular location [9]. This observation provides insight into the soft tissue recurrence at the intracapsular location. Similarly, the articular location of the OO might have favored the soft tissue recurrence.

In conclusion, our case is the second observation illustrating the delayed soft tissue recurrence of an OO after mobilization from its initial articular location during a previous arthroscopic procedure and responsible for delayed recurrence of symptoms.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

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