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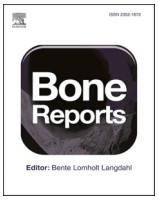

Case Report

Intra-articular osteolipoma of the elbow: A case report and a review of the literature

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ABSTRACT

Introduction: Lipomas are the most common benign mesenchymal tumors. The intra-articular localization is exceptional.
We report the first case of an intra-articular osteolipoma at the elbow.
Case report: A 36-year-old patient with no pathological history, presented to the emergency room with a two weeks history of a painless, progressively left elbow swelling in a context of apyrexia.
Physical examination revealed a hot, edematous left elbow. The gentle mobilization of the joint was painless. Biologie was normal. Initial radiograph showed a well-defined lobular image. Ultrasound revealed an anechoic intra-articular collection associated to a synovium hyperaemia and the MRI shows an aspect of synovitis. An arthrotomy was done via an internal approach of the elbow. Joint fluid was red. A free ossified intraarticular mass was discovered. Bacteriological samples were sterile. Histological examination concluded to an osteolipoma with no histological signs of malignancy. At the last 3-year follow-up, the patient didn't report any pain or functional discomfort. Elbow mobility was complete, with the absence of recurrences.
Discussion: Osteolipoma is defined as a histological variant of lipoma having undergone bone metaplasia. Allen found 06 cases in a series of 635 lipomas over five years. Pain has not been reported in the literature. In our case the pain was explained by the interposition of the tumoral mass between the articular surfaces. Histological examination confirms the diagnosis and recurrences are rare.
Conclusion: Osteolipoma is an extremely rare entity of mesenchymal tumors whose intra-articular location, although exceptional, does not worsen the prognosis.

1. Introduction

Lipomas are soft tissue benign tumors composed of mature fat tissue without atypical cells. They are frequently encountered in adults and account for 50 % of all soft tissue tumors (Murphy et al., 2004). The pathogenesis of lipomas is unknown. They generally consist of mature adipose tissue, sometimes associated with other mesenchymal elements such as muscle, fibrous, chondral, or bone tissue (Drevelegas et al., 2004). Multiple variants of lipomas have been described according to the type of tissue present: fibrolipoma, myolipoma, myxolipoma, angiolipoma, angiomyolipoma, etc.…

Osteolipoma is defined as a histological variant of lipoma having undergone bone metaplasia. Ossifying lipoma (osteolipoma) is the rarest subtype of lipomas, with the first case being reported in 1959 (Plaut et al., 1959). They can be either intra-articular or adjacent to bone tissue. Osteolipomas usually occur in the head and neck (Fletcher et al., 2013; Vanhoenacker et al., 2017).

Various hypotheses regarding the pathogenesis of these tumors have been discussed, however, the most predominant theory is the secondary ossification according to which repetitive trauma, metabolic changes or even ischemia would lead to metaplasia of the pre-existing fibrous elements within the lipoma to osteoblasts. Therefore, the term “ossifying lipoma” is preferred to “osteolipoma” (Obermann et al., 1999; Piattelli et al., 2001).

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The recommended treatment by most authors is surgical excision. Diagnosis of osteolipoma is made by histopathological examination.

We report a clinical case of an intra-articular osteolipoma at the elbow in a 36-year-old man initially treated as inflammatory arthritis.

2. Case report

A 36-year-old male patient, with no family and personnel medical or surgical history, presented to the emergency room with a two weeks history of a painless, progressive left elbow swelling in a context of apyrexia. The patient didn't report any previous old or recent trauma to the elbow.

On physical examination, the left elbow was swollen with increased local temperature. Elbow flexion was 100° with a flexion contracture of 30° (Normal range of motion: 0° to 150°). Gentle mobilization of the joint was painless within the described range of motion. There was no fever or lymphadenopathy.

Based on clinical examination, the diagnosis of septic arthritis of the left elbow was suspected.

The blood tests were slightly disturbed: erythrocyte sedimental rate at 20 mm/h (Normal value less than 18 mm/h), C reactive protein serum level at 25 mg/L (Normal value: less than 6 mg/L) and a white blood cells count within normal limits.

The initial radiological assessment included an antero-posterior and lateral views of the elbow and an ultrasound examination.

The plain x ray showed an ossified, well-circumscribed solitary lesion on the anterior aspect of the distal humerus with a long axis measuring 1.5 cm suggestive of a cartilaginous tumour (chondroma). Malignancies such as extraskeletal chondrosarcoma were unlikely because of the age and the atypical localization. However, the diagnosis can't be ruled out.

In addition to that lesion, a clearly delineated lucent lesion of 3 mm bordered by a peripheral sclerotic rim without periosteal reaction was seen in the proximal ulna. This lucency is located at the level of the trochlea, with a central calcification (Fig. 1). This lesion was classified 1a according to the Lodwick classification (Lodwick et al., 1980).

Ultrasound revealed an anechoic intra-articular collection associated to a synovial hyperemia.

The suspected diagnosis at this stage was inflammatory arthritis of the elbow. The patient was put on symptomatic treatment and the elbow was immobilized in a long arm splint.

Two weeks later, and because of the persistence of pain and swelling, a magnetic resonance imaging (MRI) was requested. MRI showed a non-enhancing T1 hypertense lesion with fatty spot suggesting the diagnosis of osteolipoma with reactive synovitis located on the posterior aspect of the elbow (Fig. 2). In addition, two subchondral cysts in the capitellum and the olecranon were identified (Fig. 3). MRI excluded any signs of malignancy such as the absence of black pepper sign, enhancement and any aggressivity towards vessels, nerves and adjacent soft tissue. Hence, an extraskeletal chondrosarcoma was rule out and the diagnosis of the chondroma located in the anterior coronoid fossa was retained (Fig. 2).

Differential diagnoses at this stage were localized pigmented villo nodular synovitis and lipoma arborescens. The diagnosis of myositis ossificans under the triceps tendon in the olecranon fossa was less likely given the intra articular location of the lesion and the absence of signs of myositis on MRI. In tumoral calcinosis, calcification are periarticular and MR T2 weighted sequences shows high signal lesion. This is not our case.

The patient was operated by a senior orthopedic surgeon. Under general anesthesia, the patient was put in the supine position with the left upper limb on an arm board. A tourniquet was used. The elbow joint was approached medially after dissecting and protecting the ulnar nerve (Fig. 4). We noticed the presence of two whitish lesions located on the posterior synovium. A bacteriological sample was taken and an excisional biopsy of the two synovial lesions was performed. Free intra-articular bone formation was also sent for pathological examination. The postoperative course was uneventful.

Bacteriological samples were sterile.

Histological examination (Fig. 5) showed a non-ulcerated synovium,

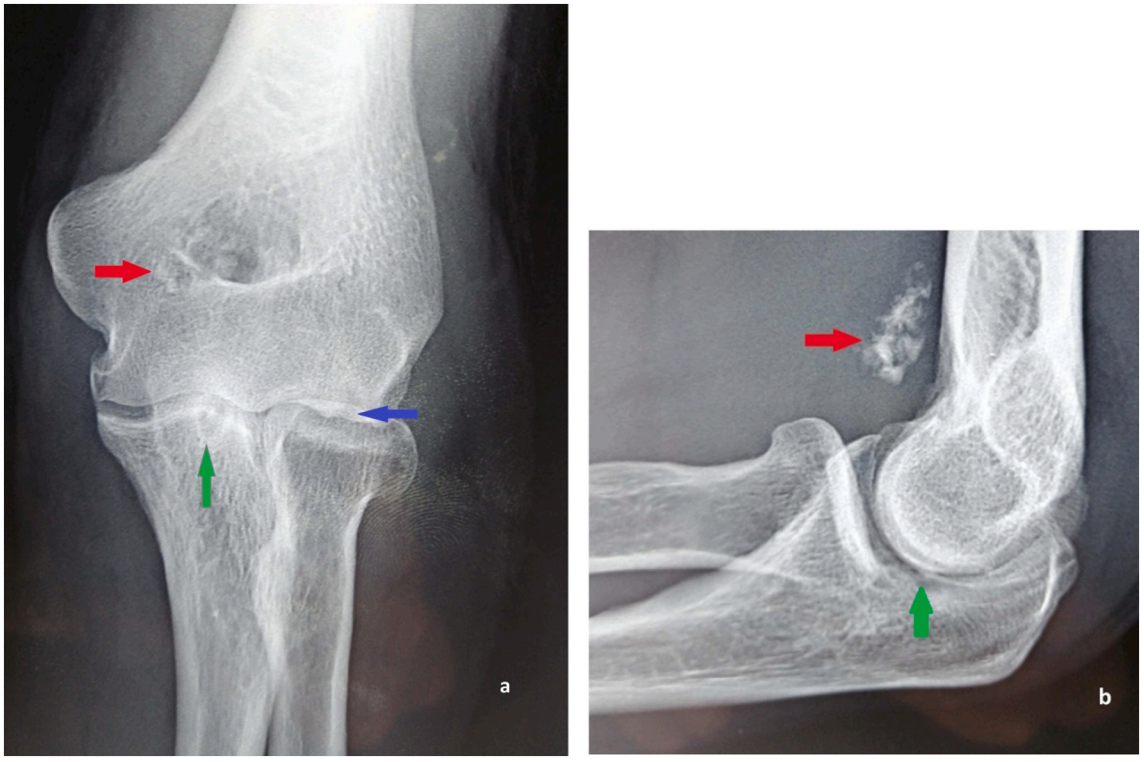

Fig. 1. X-Rays of the left elbow (a and b) showing a 1.5 cm ossification located on the anterior surface of the distal humerus (red arrow), in association to two well-circumscribed subchondral lucent lesions: one measuring 3 mm in the capitellum (blue arrow), the other measuring 5 mm with peripheral sclerotic reaction and a central calcification located in the olecranon (green arrows).

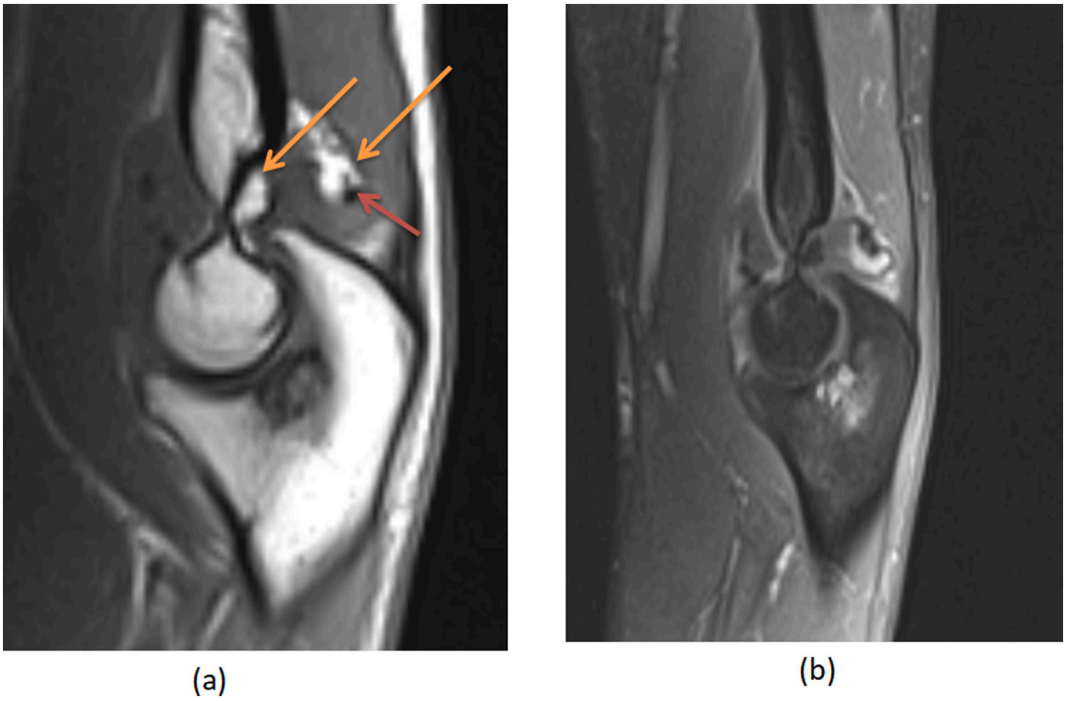

Fig. 2. Two intra-articular lesions of the posterior olecranon fossa (orange arrows) on T1 weighted sequences (a) containing lesions that attenuate on fat-saturated sequence (b). We also note the presence of dark spots due to calcification (red arrow).

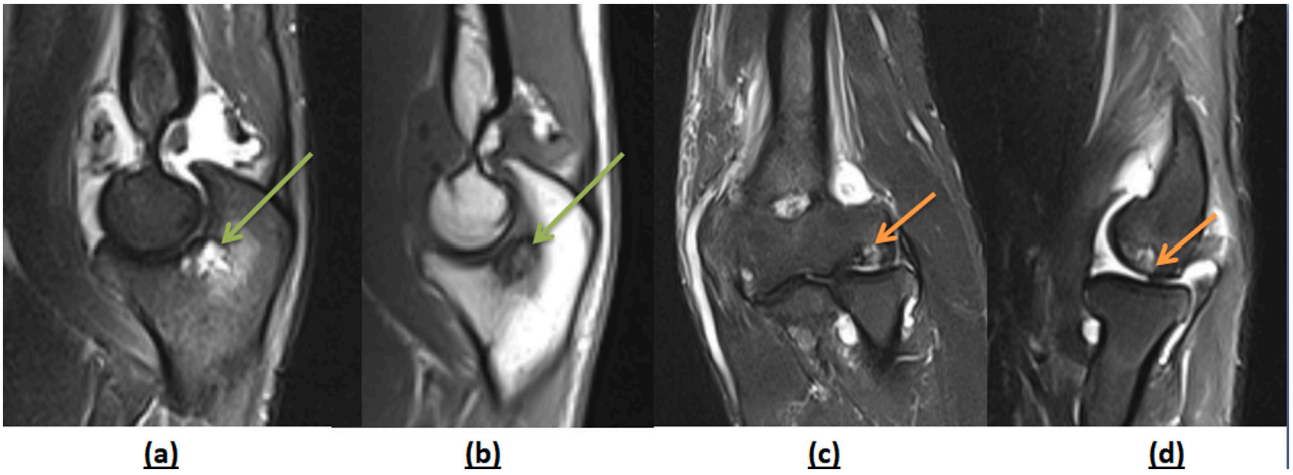

Fig. 3. The sagittal T2 fat suppressed (a), sgittal T1 (b), coronal T2 fat-saturation (c) and sagittal T2 fat suppressed (d) sequences of the MRI showing two subchondral fluid filled lesions at the level of the olecranon (green arrow) and the capitellum (orange arrow) (HypoT1 and HyperT2) surrounded by a discreet bone oedema suggesting subchondral geodes.

slightly hyperplastic in appearance. This epithelial lining was underpinned by a fibro-adipose partition with a fairly marked vascular congestion. It was associated to an inflammatory lymphocyte and plasma cell with moderate density with no histological signs of malignancy. Furthermore, it showed an ossified tissue fragment of 15 mm × 8 mm × 5 mm, associated to mature adipocytes lobules with regular nuclei. The lobules were dissociated by many fine and regular bone lamellae evoking an osteolipoma.

At the last three year follow-up, the patient didn't report any pain or functional discomfort. The patient had a 10° flexion contracture with full flexion of the elbow. No recurrence was reported.

3. Discussion

The conventional lipoma is made up of adipocytes, however, other mesenchymal elements may be associated, for example: cartilage (chondrolipoma), fibrous tissue (fibrolipoma), myxoid tissue (myxolipoma), bone tissue (osteolipoma) (Fletcher et al., 2013).

Osteolipoma is defined as a histological variant of lipoma having undergone bone metaplasia, other authors classified the lesion into “osteolipoma” and “ossifying lipoma” according to the predominance of the fatty component compared to the bone component (Vanhoenacker et al., 2017).

The World Health Organization (WHO) classifies soft tissue tumors and bone tumors into 14 types (Fletcher et al., 2013), ossifying lipoma is the rarest subtype of lipoma, the first case was reported in 1959 (Plaut

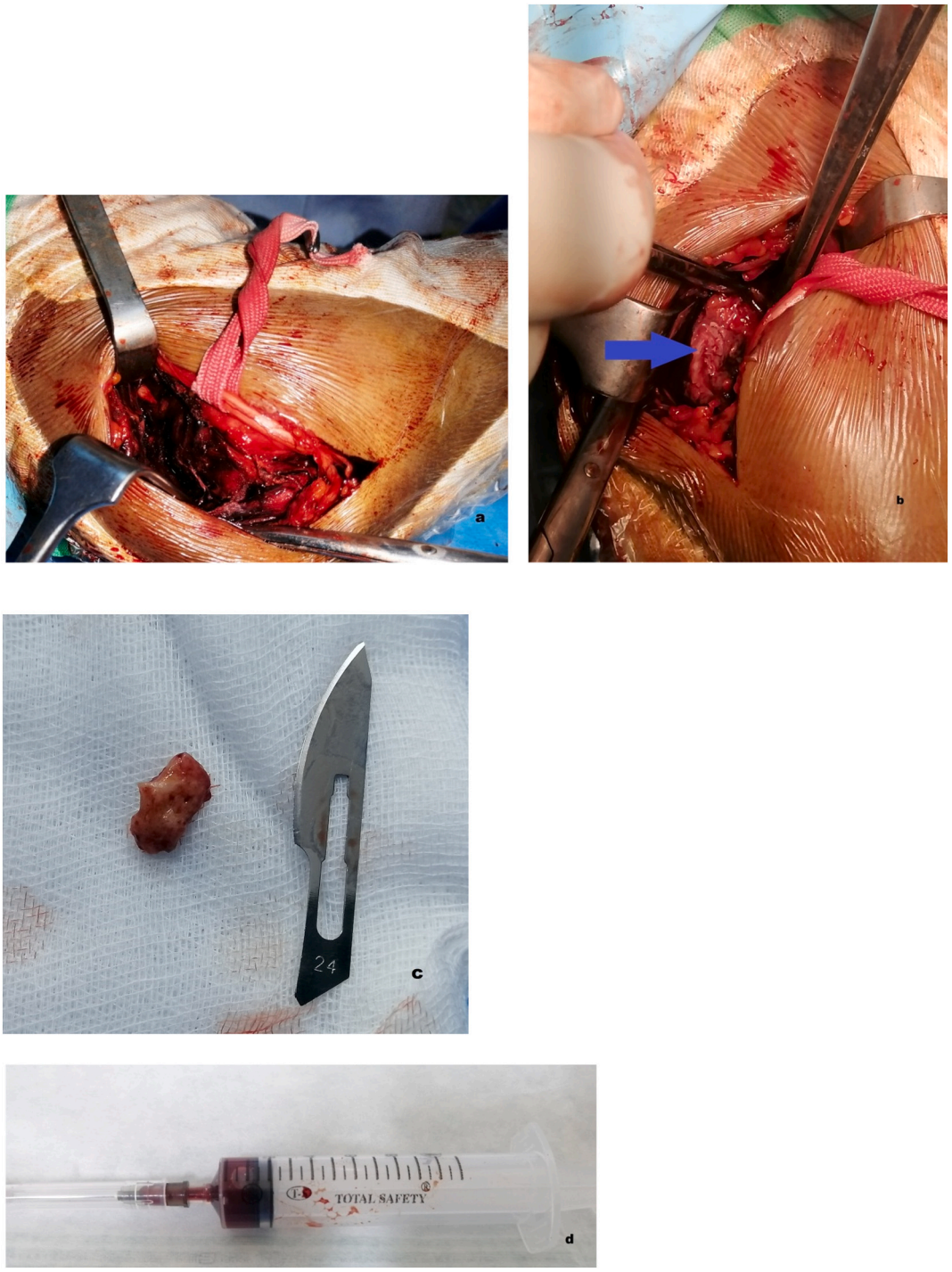

Fig. 4. Using a medial approach to the elbow (a), we found a well-defined whitish lesion (b) located at the level of the posterior synovium (blue arrow). (c) 1.5 cm free intra-articular fragment. (d) hematic joint fluid aspirated.

et al., 1959).

In an interesting series of 635 lipomas studied over a period of 05 years, only 6 cases with ossification were found (Allen, 1981).

The head and neck are the most frequent localization of reported osteolipomas (Obermann et al., 1999; Piattelli et al., 2001; Lodwick et al., 1980; Allen, 1981; Jaiswal et al., 2005). Most cases of lipoma are with osseous tissue connected with bone (inside a bone or adjacent to it) (Friedman et al., 2013; Murphey et al., 2005; Wurlitzer et al., 1973). Osteolipoma independent of bone tissue has been reported in very few cases (Yabe et al., 2006). The intra-articular location is exceptional. However, our review of the English-language literature using the keywords "osteolipoma" and "ossifying lipoma" and "elbow" on PubMed and Google Scholar finds no cases reported. Described cases of intra-articular osteolipoma were rare; the most common intra-articular location is the knee (Yabe et al., 2006; Huynh et al., 2017; Fritchie et al., 2012; Pudlowski et al., 1979; Zaizi et al., 2020; Demark, 2021) (Table 1).

The pathogenesis of osteolipoma is still not clear. Two main theories exist for the pathogenesis of osteolipomas (de Castro et al., 2010; Demiralp et al., 2009). First, these tumors appear to be of mesenchymal origin, which is derived from pluripotent cells than it may be called as benign mesenchymoma (de Castro et al., 2010).

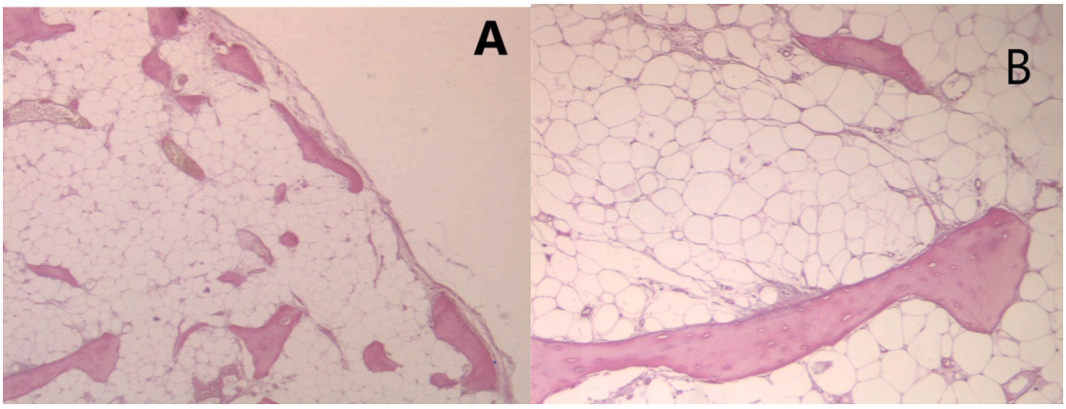

Fig. 5. A/ Thinly encapsulated proliferation of mature adipocytes with haphazardly distributed bone lamellae (H&E, 40×).
B/ Mature fat cells with uniform nuclei, varying slightly in size and shape. Osseous component is made of thin, mature and vital lamellar bone structures. No mature bone marrow elements are found (H&E, 100×).

Table 1
Summary of intra articular osteolipomas, clinical and radiologic data.

Authors	Age	Sex	Localization	Size	Management	Results	Recurrence
Yabe et al. (2006)	45	F	Sterno-clavicular joint	2.5 × 1.5 cm	Excision biopsy	Not described	No recurrence
Huynh et al. (2017)	64	M	Knee	3.8 × 3.4 × 1.5 cm	Excision biopsy	Not described	No recurrence
Fritchie et al. (2012)	51	M	Lateral anterior infra patellar tendon	4.2x4x2.8 cm	Excision biopsy	Satisfactory	No recurrence
Fritchie et al. (2012)	31	F	Infra patellar tendon	5.2 × 4.3 × 4.1 cm	Excision biopsy	Satisfactory	No recurrence
Pudlowski et al. (1979)	53	M	Suprapatellar bursa	5.5 × 4.5 × 2.5 cm	Excision biopsy	Not described	No recurrence
Zaizi et al. (2020)	47	F	Ankle	2 cm	Excision biopsy	Satisfactory	No recurrence
Demark (2021)	33	F	The left thumb carpo metacarpal joint	1.5 × 0.6 × 1.3 cm	Excision biopsy	No pain or limitations	No recurrence

M: male F: female.

According to the second theory, ossification may also have been induced by poor nutritional supply in the center of a large lipoma after repetitive trauma, metabolic changes, or ischemia leading to transformation of fibroblasts into osteoblasts (Demiralp et al., 2009; Yang et al., 2013).

Radiologically, these lesions generally appear as a zone of radiological clarity surrounded by a dense zone corresponding to ossification, which is the case in our observation.

The differentiation between ossification and calcification represents a challenge for the clinician. Therefore, the radiological differential diagnosis of a calcified or ossified intra-articular mass includes benign tumors (hemangioma, synovial chondromatosis, calcified synovitis, synovial chondromatosis, calcified myositis, myositis ossificans) and malignant tumors (osteosarcomas, synovial sarcoma) (Pudlowski et al., 1979). For these reasons, exploration by MRI is key to guide the diagnosis and to plan the therapeutic management. MRI describes the osteolipoma as a heterogeneous mass in T1 and T2 sequences with macroscopic zones of fat and bone signal and absence of enhancement with contrast product (Pudlowski et al., 1979).

Definitive diagnosis of osteolipoma is made by histological examination. According to the WHO classification of tumors of soft tissue and bone (Sbaraglia et al., 2021 Apr), ostéolipoma is a benign tumour composed of mature adipocytes showing areas of bone formation. In our case, the presence of a fibrous capsule surrounding the adipocytic tissue favors the neoplastic nature of this lesion and the bone trabeculae form an intrinsic part of this tumour. These features help rule out simple calcification of the posterior fat pad.

The treatment is not yet well codified; however, most authors recommend surgical excision (Pudlowski et al., 1979). Our case responded well to surgical treatment.

Osteolipomas have the same prognosis as simple lipomas and no recurrence has been reported (Yabe et al., 2006; Huynh et al., 2017; Fritchie et al., 2012; Pudlowski et al., 1979; Zaizi et al., 2020; Demark, 2021). The review of the literature confirms the benign evolution for this lesion. In fact, all the operated intra-articular osteolipomas didn't show any recurrence. The functional results of biopsy excision were satisfactory in most of described cases including ours (Table 1).

4. Conclusion

Osteolipomas are extremely rare and may involve intra/juxta-articular locations that should always be kept in mind when a lesion with adipose tissue associated with ossification is encountered. A multidisciplinary management between surgeon, radiologist and pathologist is necessary, because a large differential diagnosis should be evaluated in advance.

CRediT authorship contribution statement

All the authors participated in the design, performance, analysis and drafting of this manuscript.

Ethical approval

Ethical approval was granted by the Ethical Committee of MTM hospital.

Consent

Written informed consent was obtained from the patient for publication of this case report.

Declaration of competing interest

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.

Data availability

No data was used for the research described in the article.

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