



Case Series

Intra-articular lipoma arborescens of the knee: A report of two cases with bilateral localization



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ABSTRACT

INTRODUCTION: Lipoma arborescens is a very rare pseudo-tumoral lesion of unknown etiology, characterized by lipomatous infiltration of subsynovial stroma, bilateral involvement is exceptional, only eight cases are reported in the literature.

PRESENTATION OF CASE: We report the very rare case of two men presented at our department with complaints of swelling of both knees and intermittent joint effusion. Surgical biopsy revealed Lipoma arborescens in both cases. Open total synovectomy was performed. Follow-up evaluation showed no signs of recurrence.

DISCUSSION AND CONCLUSION: Lipoma arborescens is a rare entity, the unilateral form affecting the knee is the most common, with a predilection for suprapatellar recess. Magnetic resonance imaging (MRI) is the examination of choice showing multiple villous proliferation of the synovium and fat-like cells, with a fatty signal on all sequences. Treatment by open or arthroscopic synovectomy offers the best outcomes.

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1. Introduction

Lipoma Arborescens is a very rare pseudo-tumoral lesion, characterized by a lipomatous infiltration under the synovium with a diffuse substitution of the synovial tissue by mature adipocytes [1], the multiplication of synovial villi distended by the fat takes on a tree-like appearance.

The area of predilection is the suprapatellar pouch in the knee joint [2], bilateral involvement is extremely rare (eight cases in the literature) [3]. We report two cases.

This case series has been reported in line with the PROCESS criteria [4].

2. Case report

2.1. Case 1

A 26-year-old man, followed for pachydermoperiostosis for 10 years, has had inflammatory arthritis of both knees for 4 years.

The physical examination found significant bilateral swelling of the knees with filling of the suprapatellar areas, condylar ramps and bilateral joint effusion. Joint motion of the knees was slightly reduced, without inflammatory signs. The symptoms were more marked on the left side. The examination of the joint fluid for crystals and bacteria was negative.

Radiography showed a rarefaction of the bone matrix, with the presence of a multi-lamellar periosteal reaction, and osteoarthritis lesions. MRI showed significant synovial thickening (Fig. 1a), enhanced after gadolinium injection with fatty infiltration of the synovium in hyperintense suppressed at STIR, with a large-scale joint effusion. The surgical biopsy found synovial hyperplasia without atypia, taking a villous appearance, infiltrated by mature adipocytes with presence of hemorrhagic suffusion in places, and lymphocytic infiltrate.

The patient underwent total open synovectomy on the left side three years ago and underwent the same treatment for the right knee after two years, after worsening of symptoms. Control at one year showed the disappearance of pain and swelling, with no signs of recurrence at both knees, with complete resumption of daily activities.

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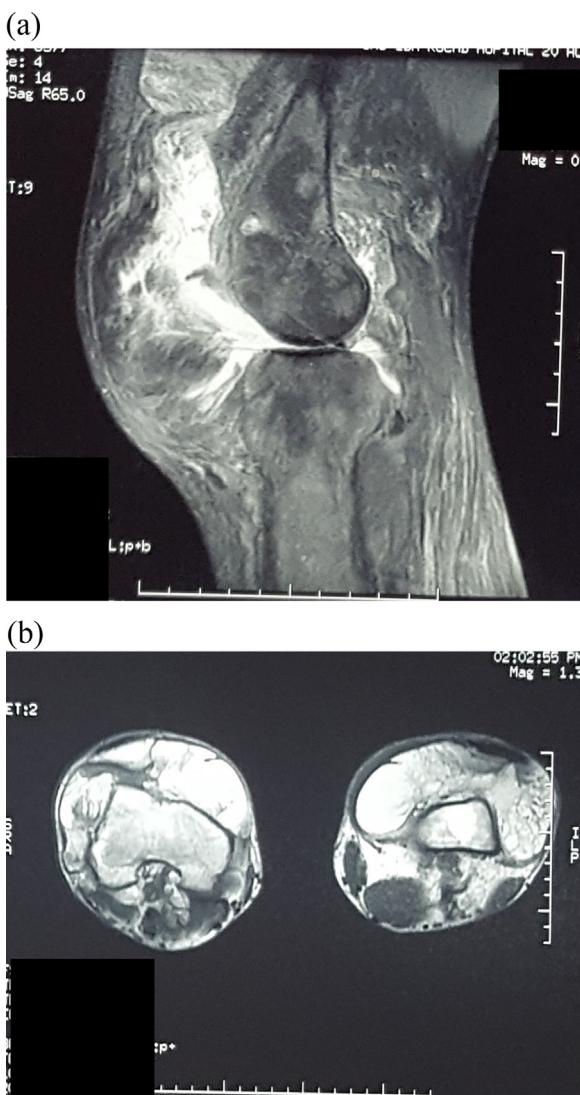


Fig. 1. (a) MRI revealed significant synovial thickening, enhanced after gadolinium injection with fatty infiltration of the synovium, and joint effusion (Patient 1). (b) MRI showing bilateral hypertrophy of the villous synovial fringes of a fatty signal and bilateral joint effusion (Patient 2).

2.2. Case 2

A 22-year-old man, presented with bilateral painful swelling of the knees gradually increasing in volume for three years, with an intermittent joint effusion.

The physical examination found bilateral joint effusion and quadriceps recess filling, the knee had full range of motion, was stable, and showed no inflammatory signs.

Radiography was normal, the usual biological examinations were normal, the examination of the articular fluid was negative. MRI showed bilateral hypertrophy of the villous synovial fringes of a fatty signal and bilateral joint effusion (Fig. 1b).

Surgical biopsy macroscopically found diffuse digitiform synovial hyperplasia, the histological study found papillary synovial proliferation with adipocyte axis, made of mature adipocytes with fine fibro-vascular septa (Fig. 2).

The patient underwent an open total synovectomy on the most symptomatic right side (Fig. 3), and the extracted tissue was then histologically analyzed and confirmed the diagnosis. Currently and after 14 months of follow-up, the knee was painless, stable, and without signs of recurrence.

3. Discussion

Described by Arzimanoglu in 1957 [5], Lipoma Arborescens is a rare benign intra-articular lesion of unknown etiology [1], characterized by the substitution of synovial tissue by mature adipocytes, also called villous lipomatous proliferation or synovial lipomatosis [2].

In 46 cases of knee located lipoma arborescens found in the literature [6,7], the involvement was bilateral in only 8 cases, the average age is 46 years for men, 37 years for women [2]. The unilateral form affecting the knee is the most common, with a predilection for supra patellar recess [8,9]. The hip, wrist, elbow, ankle and bursae may also be affected [10].

There are two types of arboreal lipomas: primary, idiopathic, typically affecting middle-aged, and a more common secondary variety affecting elderly patients with coexisting inflammatory arthritis, such as rheumatoid arthritis, gout, and psoriatic arthritis [3,11,12].

The clinical examination finds symptoms of chronicity, such as progressive soft swelling, often very abundant joint effusion, pain, stiffness and episodes of joint blockage [1], which is exacerbated when the proliferation is interposed between articular surfaces [8].

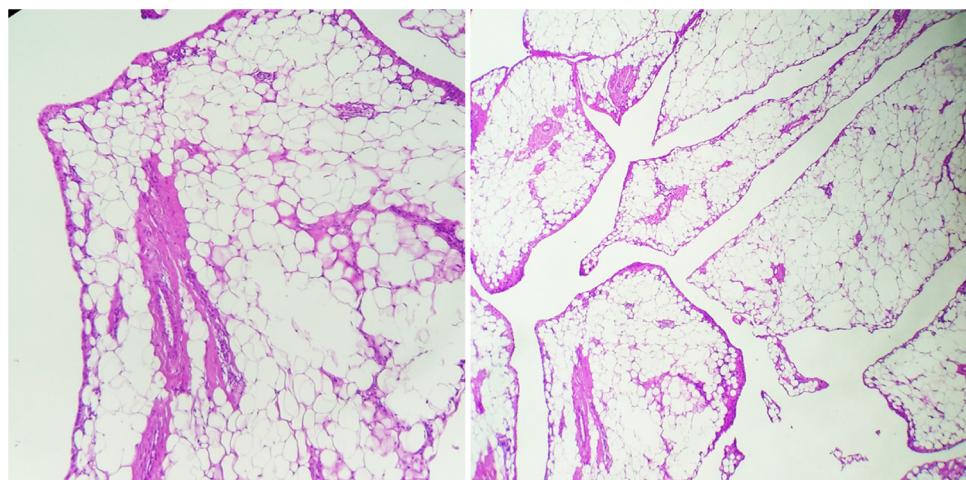


Fig. 2. Histopathological examination showing a synovial proliferation with adipocyte axis. Synovial tissue is completely replaced by the villous proliferation of mature fat cells.

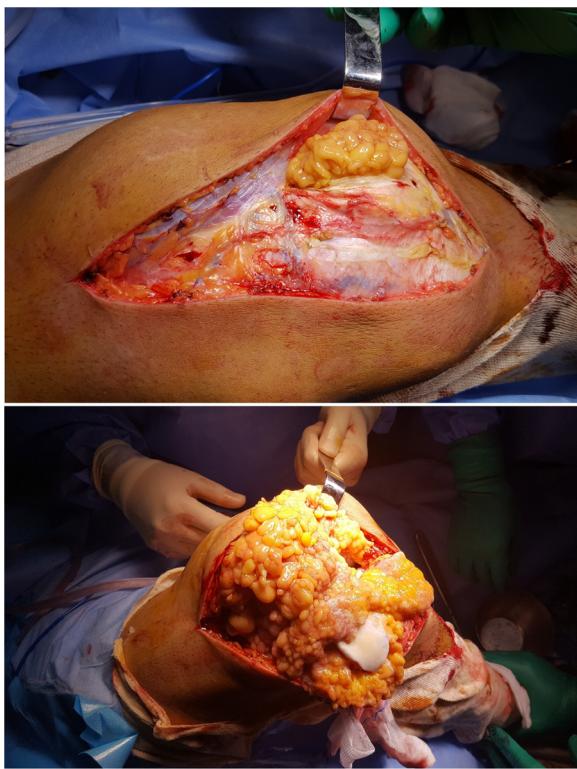


Fig. 3. intraoperative images showing the appearance of lipoma Arborescens (patient 2).

Synovial fluid is usually acellular, sometimes moderately inflammatory or hemorrhagic [2–12].

Plain radiographs show an increase in the density of the soft parts of the knee, it can show bone erosions or osteoarthritis lesions [9], it is also useful for eliminating any differential diagnosis. MRI is the examination of choice showing multiple villous proliferation of the synovial and frond-like structures with a fatty signal on all sequences which are suppressed on FAT-SAT, with joint effusion, and the absence of effects of magnetic susceptibility of hemosiderin [3–5].

The differential diagnosis includes villonodular synovitis, chondromatosis, rheumatoid arthritis, synovial hemangioma, and amyloid arthropathy [1–13].

Surgical or percutaneous biopsy can guide the diagnosis showing microscopically a synovial villi completely infiltrated by mature adipocytes, the synovial coating is inflammatory or hyperplastic with focal perivascular infiltration and chronic mononucleated inflammatory cells [2,12,14].

Treatment with intra-articular injections of radioactive compounds, such as Yttrium 90 or steroids, relieves symptoms during moderate periods [13] but with the risk of cutaneous necrosis and epiphyseal osteonecrosis, the surgical approach consists of an open synovectomy or through arthroscopy if the lesion is located in the anterior compartment of the knee with less morbidity [2–15]. The macroscopic histological study shows broad-based polyps or villi. These villi are filled with adipocytes and sometimes congestion [12].

In our patients the important development and the diffuse aspect of the lesions justified open surgery. Recurrence after synovectomy is rare [10], Though Afonso [16] reported a case of recurrence one year after synovectomy.

4. Conclusion

Lipoma Arborescens is a rare benign lesion. MRI allows to evoke the diagnosis by showing the fatty nature of the synovial proliferation and specifies its extension. Early synovectomy offers the best functional outcome.

Conflict of interest

The authors declare having no conflicts of interest for this article.

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No sources of funding to declare.

Ethical approval

This research did not require ethical approval due to the institute not requiring it for this type of study.

Consent

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

Author contributions

Hatim Garnaoui: Corresponding author writing the paper.

Ayoub Rahmi: study concept.

Abdeljebbar Messoudi: supervised the writing of the manuscript.

Abderrahim Rafaoui: study concept.

Mohamed Rafai: revision of paper.

Abdelhak Garch: correction of the paper.

Hasna Elkhiriaoui: data collection.

Samira Benayad: data collection.

Salwa Belhaj: data collection

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