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Multifocal intraarticular lipoma arborescens of the hip and knee: A case report and review of literature

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Abstract:

Lipoma arborescens (LA) is a rare clinical condition presenting mostly as an intra-articular lesion characterized by villous proliferation of the synovial membrane and hyperplasia of sub synovial fat. We report a case of a multifocal LA localized in the knees and the hips in a 20-year-old man symptoms being present for the last 3 years and initially mimicking an inflammatory arthropathy. On physical examination a discreet joint swelling of the left knee with effusion, and limited movements in the left hip were observed. Laboratory tests presented normal acute phase reactants of inflammation as well as the rheumatoid factor, CK, and negative results for antinuclear and anti-CCP (ELISA) antibodies. Magnetic resonance imaging of the knees and hips showed articular effusion and synovitis, and a pattern of LA. The histopathologic study confirmed the diagnosis. Arthroscopic synovectomy for the knee and open synovectomy for the hip brought some improvement to joint mobility and pain. Although rare, this condition must be differentiated from inflammatory arthropathy, particularly in the absence of response to clinical treatment, and absence of positive biochemical markers, since the therapeutic strategy is radically different.

Keywords:

Arthritis, lipoma arborescens, synovium, villous proliferation

Introduction

Lipoma arborescens (LA) is a rare clinical condition presenting mostly as an intraarticular lesion characterized by villous proliferation of the synovial membrane and hyperplasia of sub synovial fat. We report a case of a multifocal LA localized in the knees and the hips in a 20 year old man symptoms being present for the last 3 years and initially mimicking an inflammatory arthropathy.

Case Report

A 20-year-old male patient presented with a 3-year history of left knee pain and swelling and pain in the left hip for the past 2 years. The pain and swelling in the left knee was spontaneous in the onset and had gradually worsened. The patient had two episodes of transient knee swelling of the knee which resolved spontaneously over a period of few days, but the recent massive enlargement led to decreased range of motion (ROM) and prompting him to seek medical attention. There was no history of joint trauma, morning stiffness, constitutional symptoms, or family members with swollen joints. Left hip pain was associated with episodes of locking mainly while flexing the hip but without any swelling around the joint. The ROM of the left hip was near normal, except for some restriction in flexion and extension with cracking sensation at the end of left hip flexion.

On examination, the patient had diffuse boggy swelling of the left knee. He was able to flex from 0° to 100°. There was medial joint line tenderness on palpation, crepitus, effusion, and thickened synovium

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mainly in the suprapatellar region. The remainder of the musculoskeletal evaluation and the general physical examination were normal. Left hip motion was as follows: flexion/extension: 100/0, abduction/adduction: 40/25, and external rotation/internal rotation: 40/20. Patrick's test was positive. Laboratory tests presented normal acute-phase reactants of inflammation and negative serological markers. Culture of joint fluid was negative.

Radiography of the knees included anterior–posterior and lateral view and was normal apart from a thickening of the periarticular soft tissue. Visualized bones appeared normal with maintained joint space. Radiographs of the hip joints presented a mild decrease in articular space in the left hip joint.

Magnetic resonance imaging (MRI) of the knee showed high signal intensity frondlike synovial thickening in the suprapatellar bursal region with effusion on T1 and low signal intensity on T2 images [Figure 1a and 1b]. Fat-suppressed proton density images revealed complete suppression of signal intensity of villous projections in the suprapatellar bursal region. There was associated mucoid degeneration in the anterior cruciate ligament with increased joint effusion without bone and cartilage erosion. MRI of the left hip [Figure 2a and b] revealed diffuse villous synovial proliferation with some fat signals. There was mild joint space narrowing with evidence of bone erosion at the anterosuperior aspect of the left femoral head.

The patient underwent arthroscopic synovectomy of the left knee and open synovectomy using the Smith-Peterson approach for the left hip. Intraoperative findings revealed multiple globular and villous projection of synovial covered tissue throughout hip and knee joints [Figure 3]. Arthrotomy of the left hip [Figure 4] showed a small, yellowish mobile synovial tumor with minimal cartilaginous abrasion of the femoral head but no invasion to the bone tissue. Excision of the lesion was performed along with subtotal synovectomy [Figure 5]. Histological analysis revealed elongated and hypertrophied villous projections of synovial folds distended by mature fat cells without atypia or malignancy. A dense and focally nodular lymphocytic and plasma cellular infiltrate was seen in the synovium, which along with fatty proliferation was evidence of coexistent chronic synovitis. After the surgery, pain decreased rapidly and the patient recovered normal hip and knee mobility. No recurrence of symptoms has been observed at last follow-up of the patient.

Discussion

Lipoma arborescens (LA), also known as synovial lipomatosis, LA is a benign indolent synovial proliferative disease and an uncommon cause of articular masses. The term "arborescens" describes the characteristic tree like morphology of this lipomatous villous synovial proliferation, which resembles a frond-like synovial mass.[1]It presents with monoarticular involvement in majority of the cases, although bilateral involvement of the hips [2], ankles [3], elbows [4], shoulders[5] has also been reported. Moreover, extensor and flexor tendon sheath involvement in the hands and tendons around the ankles has been described.[6,7] The involvement of multiple joints in the same patient is an even rarer occurrence, with only a few cases published.[2,6] The literature contains sporadic reports of cases of simultaneous involvement of the knees, wrists, and hands. It can affect patients of all ages (most common in the fifth decade) with equal predominance in men and women. It may be associated with rheumatoid arthritis (RA), degenerative arthritis, or prior trauma.[8,9] The reason why the fat proliferation occurs is not completely known. Although it can occur without antecedents, sometimes it is preceded by a traumatic or inflammatory process. Accordingly, it has been suggested that chronic synovitis may be the precursor for LA.[10]

Intraarticular LA presents with a chronic unilateral mass, joint pain, soft-tissue swelling, and effusion in the affected joint[7,11,12] and may restrict ROM. As in our patient,

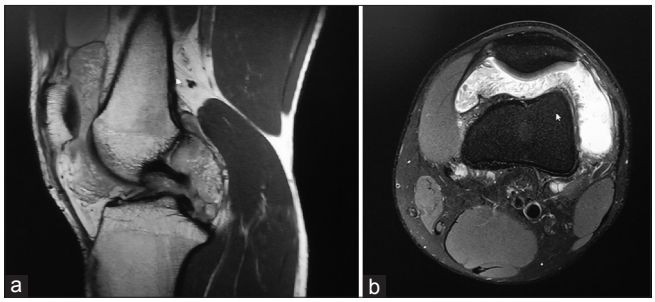

Figure 1: Showing sagittal T1 (a) and axial T2 (b) fat-saturated magnetic resonance images of the left knee show diffuse villous and frond-like intraarticular fatty masses in the suprapatellar bursa with associated marked joint effusion

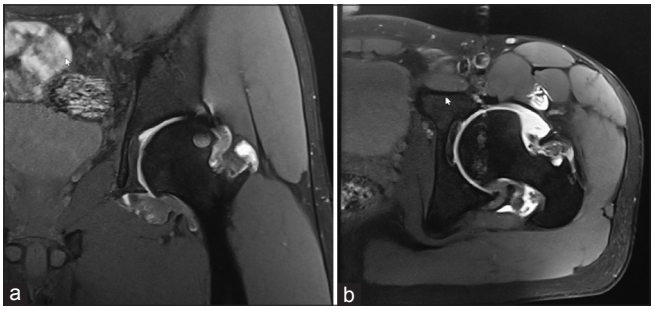

Figure 2: Showing coronal T2 (a) and axial T2 (b) fat-saturated magnetic resonance images of the left hip show diffuse villous and frond-like intraarticular fatty masses with associated marked joint effusion

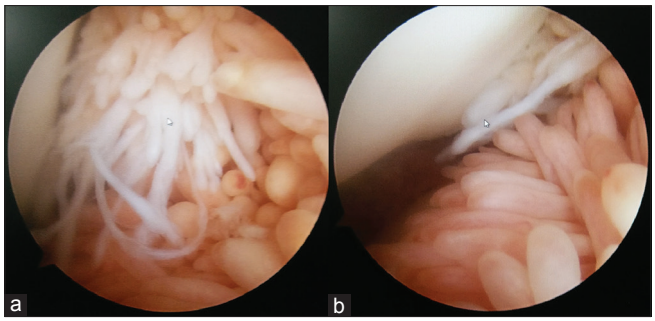

Figure 3: Arthroscopic appearance of lipoma arborescens shows yellowish-white, frond-like synovial proliferation in the synovium: (a) suprapatellar fossa, (b) posteromedical space in the knee

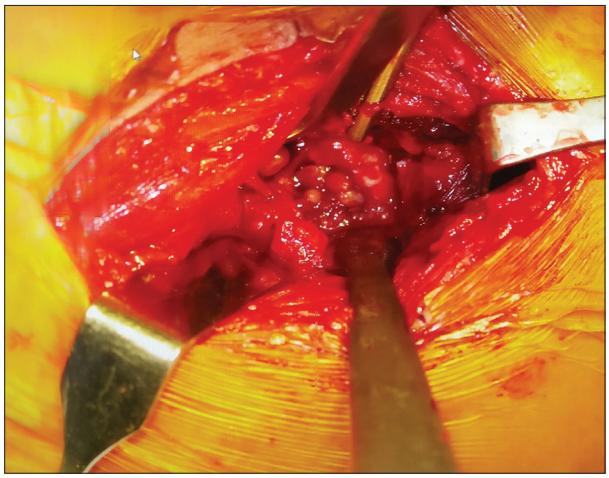

Figure 4: Appearance of lipoma arborescens shows yellowish-white, frond-like synovial proliferation in the synovium in the hip opened by the Smith-Peterson approach

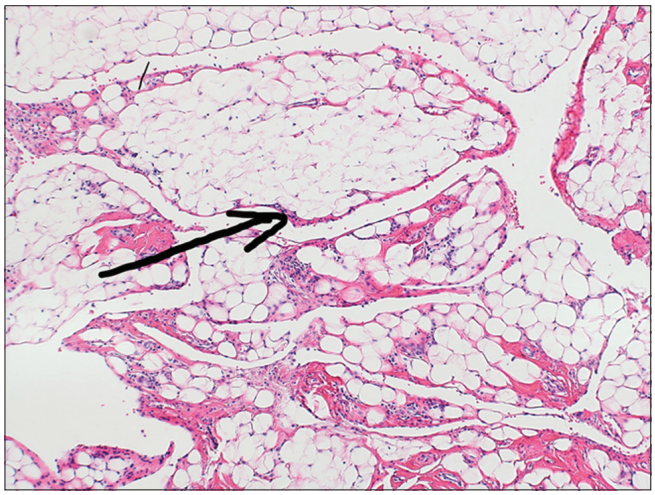

Figure 5: Histopathological study of the knee synovial tissue revealing: (a) several villous structures with synovial lining cells, which contain a stroma that exhibits increased mature adipose tissue. (b) villi with intense nonspecific inflammatory synovitis

intermittent exacerbations may occur with increases in pain and decreases in motion and may be related to the trapping of hypertrophied fatty villi between the moving joint surfaces. There are two types of LA, namely primary and secondary, depending on the age of onset and underlying precipitating condition.[3,8] More common secondary type is associated with an underlying chronic irritation, such as degenerative disease, trauma, meniscal injury, or synovitis and is observed in elderly patients. The less common primary type is idiopathic and occurs in patients of younger age group between the second and third decades of life.[7] Because of the young age of our patient and the absence of any predisposing conditions, our patient had a primary form of LA.

LA is identified more frequently after the age of 50 years. However, in the present case, where joint manifestations started before the age of 20 years did influence us to treat the condition more aggressively. The involvement of more than one large joint resembled an inflammatory arthropathy, but the normal erythrocyte sedimentation rate and serological markers ruled it out. Inflammatory synovitis[10] in LA if present should be an indication for much more aggressive treatment to avoid joint damage. Precise diagnosis is established by MRI of the affected joints. The main findings of MRI[13] are as follows: synovial mass with arborescent architecture, sign of intensity similar to fat in all the sequences, in Short tau inversion recovery (STIR) (fat suppression), and artifacts of the "chemical-shift" type at the fat – liquid interface in the articular space.[6] Soler *et al.*[14] suggested three common morphological patterns of presentation on MRI: multiple villous lipomatous synovial proliferation, isolated frond-like fat subsynovial mass, and mixed pattern. Our case report had features of multiple villous lipomatous synovial proliferation.

The differential diagnosis of LA should include other diffuse pathologies of the synovium, such as pigmented villonodular synovitis (PVNS), synovial chondromatosis, synovial hemangioma, and RA. PVNS is a benign inflammatory lesion with extensive intraarticular vascular synovial proliferation with characteristic low signal intensity on all sequences due to the presence of iron, although high signal areas due to fat, effusion, edema, and inflammation can be seen. Synovial chondromatosis is characterized by the formation of multiple cartilage nodules, which may detach and become free intraarticular bodies. MRI shows homogeneous lobulated intraarticular mass isointense to muscle on T1 and hyperintense to muscle on T2 with or without osteochondral bodies of low signal intensity. Synovial hemangioma is a rare benign vascular malformation that occur in relation to joint and show intermediate signal intensity on T1- and T2-weighted images with areas of low signal intensity due to phleboliths or fluid void within a linear and punctate lesion of high signal intensity corresponding to fibrous septa between the vascular channels and fatty tissue within the lesion. RA is a chronic systemic connective tissue disease. MRI

shows pannus with intermediate to low signal intensity on T1 and T2. Subchondral edema and effusion appear hypointense on T1 and hyperintense on T2.

LA usually does not require aggressive surgical treatment unless symptomatic despite conservative management. The primary treatment should be directed toward mitigating any underlying precipitating condition to reduce further progression and its associated symptoms. In advanced primary cases and difficult cases of secondary LA, surgery can be considered. The surgical treatment of choice is either open or arthroscopic synovectomy. The lesion, especially in the knees, is amenable to arthroscopy due to less soft-tissue trauma and faster rehabilitation. Recurrence after synovectomy, whether open or arthroscopic, is uncommon. Radiosynovectomy with yttrium-90 colloid[15] has also been successfully used.

Conclusion

LA affecting more than one joint simultaneously is a rare entity and can mimic RA or early undifferentiated form of arthritis. This rare entity should be ruled out in arthritides that remain undifferentiated after conventional clinical, biochemical, plain radiographic evaluations, and lack of response to disease-modifying anti-rheumatic drugs. MRI due to its multiplanar capabilities and high-contrast resolution allows correct evaluation of size and grade, accurate treatment planning, and effective follow-up. Either open or arthroscopic synovectomy is usually recommended in LA. If treatment is not provided in a timely manner, then the condition can progress to cause early osteoarthritis and resulting pain and inflammation in the joints can decrease the quality of one's life.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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