

Progressive bilateral lipoma arborescens of the knee caused by uncontrolled juvenil idiopatic arthritis

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ABSTRACT

Lipoma arborescens (LA) is a chronic, slowly progressive intra-articular lesion characterized by villous lipomatous proliferation of the synovium. Most cases have been described in elderly patients with degenerative or post-traumatic joint disease, but in several case reports it has been considered to be related to inflammatory joint diseases. Here we report a case of 17 years old female firstly presenting with bilateral swelling in both knees of 5 years duration, followed by the development of wide spread lipoma arborescens associated with uncontrolled treatment of juvenil idiopatic arthritis.

Keywords: Inflammatory joint disease; lipoma arborescens; lipomatous proliferation.

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Lipoma arborescens is an uncommon lesion which comprise from subsynovial villous proliferation of mature fat cells, generally involving the suprapatellar pouch of the knee joint. Clinical presentation includes painless swelling of the joint, followed by progressive pain with intermittent episodes of joint effusion for many years [1]. The aetiological types of lipoma arborescens are primary and secondary, depending on the age of onset and underlying causes [1]. The secondary type is more common associated with a reason of chronic irritation, such as degenerative disease, trauma, meniscal injury or synovitis, and is usually seen in elderly patients [1, 2]. The less common primary type is idiopathic and seen more in younger age. Here we describe a slowly progressive lipoma arborescens in a younger patient who is not being under regular treatment of juvenile idiopatic arthritis (JIA).

CASE REPORT

A 17-year-old female presented to the pediatric rheumatology department with a 5-year history of persistent knee swelling. The patient has no history of trauma prior. In medical history, she was diagnosed as JIA due to bilateral knee swelling and 5th metacarpal joint pain in 14-year-old. At the time of diagnosis, she was treated with sulfasalazine (SSZ) in the previous center. There was no improvement with SSZ. On physical examination, there were large-sized bilateral knee joint effusions and limitation in range of motion. The redness or warmth on the joint was absent. Bilateral sacroiliac joints were tender on palpation. The autoantibodies and the rheumatoid factor were negative. The routine blood investigations were done. All test were within normal limits. Ultrasonography examination showed bilateral frond

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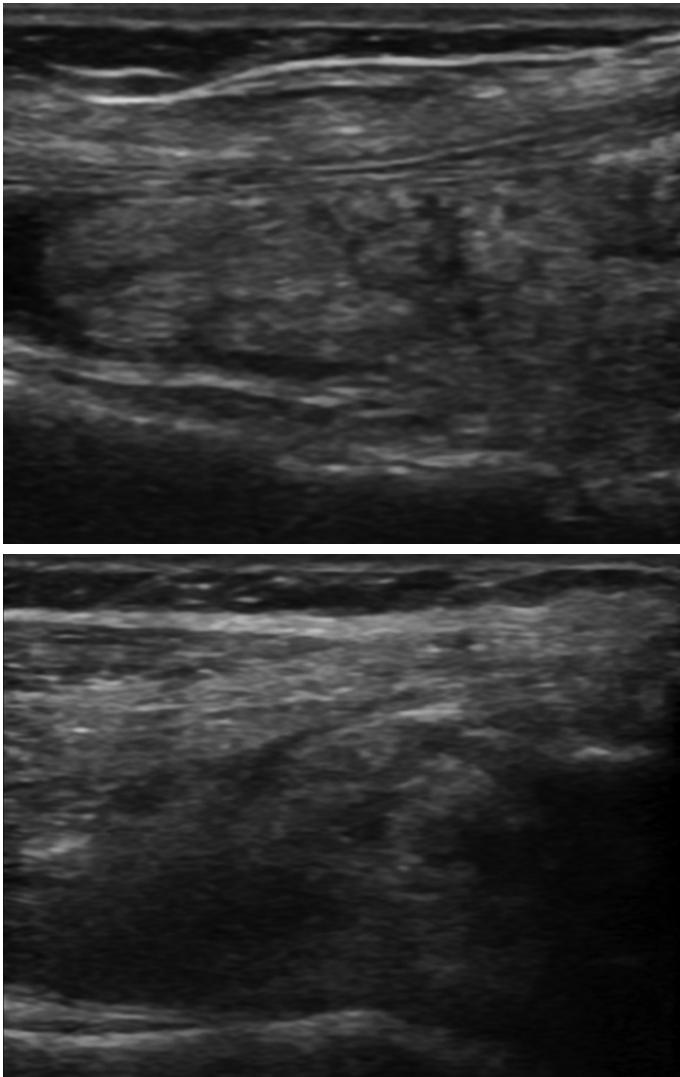


FIGURE 1. Both ultrasonography images shows frond like villous projections of the synovium and massive effusion in the suprapatellar pouch, left image is first presentation of the patient to our clinic, right image is features of suprapatellar pouch after the treatment. Mild decrease in villous projections of the synovium is seen after treatment.

like villous projections of the synovium and massive effusion in the suprapatellar pouch (Fig. 1).

MRI examination of both the knees demonstrated frond-like synovial mass and synovial hypertrophy with large-size joint effusions, which showed different signal intensity of fat on all sequences (Fig. 2, 3).

She underwent both knee aspiration, which showed no sign of infection. Intra-articular triamsinolon hexacetonide applied both knee joints and also methotrexate 20 mg/weekly subcutan injection was given. The resolution of bilateral knee swelling and improvement in range of

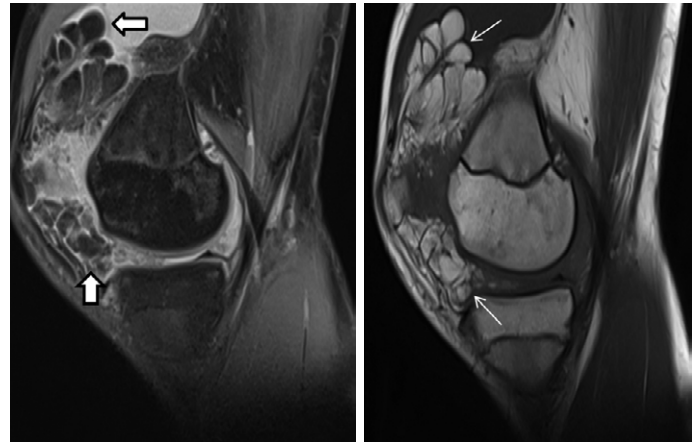


FIGURE 2. Sagittal fat saturation T2A images (thick arrow) and sagittal T1A images (thin arrow) shows fat-containing frond-like synovial mass and suprapatellar projections outlined by concurrent joint effusion in infrapatellar fat pad and quadriceps fat pillow. The lesion follows the signal intensity of fat on all sequences.

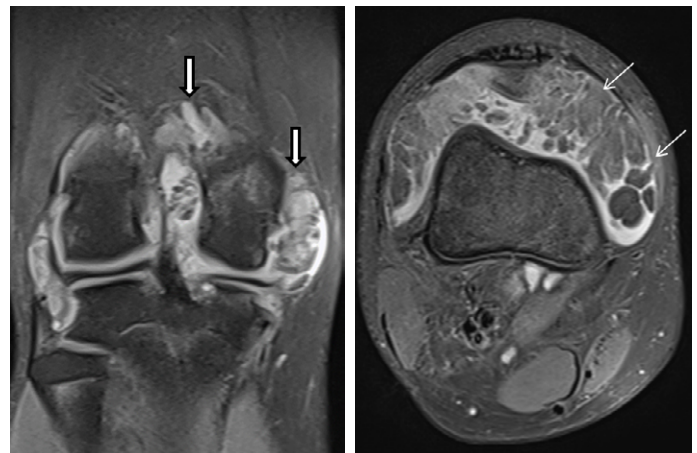


FIGURE 3. The axial (thick arrow) and coronal (thin arrow) fat saturation T2A images we also see synovial hypertrophy and bursal effusions in chronic JIA patient.

motion was detected. The informed consent was taken from the patient.

DISCUSSION

We presented clinical progression and outcome of a JIA patient complicated with LA. She had bilateral knee joints involvement and slowly progressed in to five years. Although we had seen the patient when she was seventeen years old, there have been complaints since the age of twelve.

Lipoma arborescens is an unusual condition of the synovial lining of bursae, joints and tendon sheaths [4]. It is mainly reported in the knee joint although the shoulder, elbow, hip, ankle and wrist joints have been and typically described as a unilateral disease in the literature [1, 2, 4, 5]. Lipoma arborescens is usually between the third and fifth decades, and it is more common in males [2]. The most common symptom is refractory knee effusion before and after excision of LA [3]. Although LA is often associated with trauma, osteoarthritis or rheumatoid arthritis, there have been cases with any underlying condition [6].

The patient had bilateral LA in knees associated with JIA when she was twelve. Bilateral involvement of LA is not common, particularly in the secondary forms [1]. Involvement of both knees have been reported in up to 20% of affected patients in some studies [1, 2, 7]. Laboratory findings are generally unremarkable, we didn't find any notable findings as is the case with our patient. A normal leukocyte count and erythrocyte sedimentation rate were usually seen. Joint fluid is generally negative for bacteria [7].

The differential diagnosis of LA encompasses pigmented villonodular synovitis (PVNS), true intra-articular lipoma, synovial chondromatosis and chronic inflammatory synovial proliferation like rheumatoid arthritis, tuberculous arthritis and gouty arthropathy [1]. Lipoma arborescens's villous fatty projections are typically display a high echopattern, similar to that of adjacent subcutaneous fat and with in the surrounding effusion on ultrasonography. The mass is usually soft in consistency and compressible, as opposed to the firm and noncompressible masses of PVNS [1].

Magnetic resonance imaging is the gold standard to diagnose LA. The pathognomonic features are hypertrophic adipose proliferation of the subsynovial tissues, especially using the fat-suppressed or short T1 inversion recovery sequences [7]. Absence of magnetic susceptibility effects of haemosiderin, and absence of enhancement on intravenous administration of contrast were seen in LA whereas rheumatoid arthritis on MRI is synovial proliferation with fibrosis with intermediate intensity on T1 and T2 imaging [4]. The MRI of the patient showed synovial fronds with fat signals on all sequences, joint effusion and absence of enhancement on intravenous administration of contrast.

Lipoma arborescens displays moderate progression

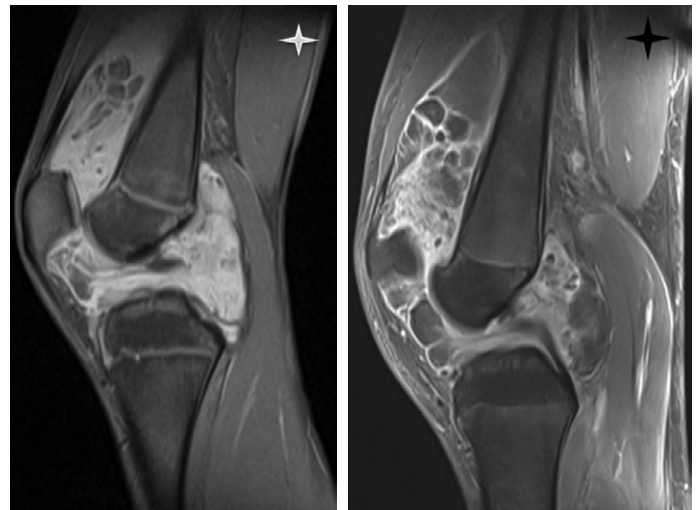


FIGURE 4. We see the progression of the disease between 2013 (white star) to 2018 (black star) years in sagittal fat saturation images.

by the time. The development of synovial LA in bilateral knees and synovial fronds with fat signals had been become more common in five years on the patient (Fig. 4).

Lipoma arborescens does not require aggressive surgical treatment unless it is symptomatic despite conservative management [1]. The primary treatment of LA is to reduce further progression of secondary type and its associated symptoms. However, in advanced primary and difficult cases of secondary LA, surgery might be considered. The surgical treatment of choice for LA is either open or arthroscopic synovectomy [1].

Although the patient showed moderate progression with both knee joints aspiration and one course of methotrexate treatment, we planned arthroscopic synovectomy for definitive treatment. We have obtained partial regression in her symptoms, however if she was given an earlier diagnosis her results would have been better.

Conclusion

This case revealed a patient who suffering from bilateral persistent knee swelling, diagnoses with JIA for 5 years. Underlying mechanism of persistent swelling lead us to the diagnoses of LA as a result of imaging. In the literature, so many cases of LA indicate a benign slowly progress but there are less reported cases have been complicated by JIA. This case highlight of

LA associated with JIA and due to slowly progression extremely wide spread synovial fronds was detected.

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