

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

 **Gozde Ercan Zeybek**,¹  **Sevinc Kalin**,²  **Betul Sozeri**³

¹Department of Pediatrics, University of Health Sciences Umraniye Training and Research Hospital, Istanbul, Turkey

²Department of Pediatric Radiology, University of Health Sciences Umraniye Training and Research Hospital, Istanbul, Turkey

³Department of Pediatric Rheumatology, University of Health Sciences Umraniye Training and Research Hospital, Istanbul, Turkey

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 five years duration, followed by the development of wide spread lipoma arborescens associated with the uncontrolled treatment of juvenile idiopathic arthritis.

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

Cite this article as: Ercan Zeybek G, Kalin S, Sozeri B. Progressive bilateral lipoma arborescens of the knee caused by uncontrolled juvenil idiopathic arthritis. *North Clin Istanbul* 2020;7(5):512–515.

Lipoma arborescens is an uncommon lesion which comprise of 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 commonly 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, in this case study, we described a slowly progressive lipoma arborescens in a younger patient who was not being under the regular treatment of juvenile idiopathic 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 had 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. Routine blood investigations were carried out. All tests were within normal limits. Ultrasonography examination showed bilateral

Received: August 03, 2018 *Accepted:* February 18, 2019 *Online:* November 21, 2019

Correspondence: Gozde ERCAN ZEYBEK, MD. Saglik Bilimleri Universitesi, Umraniye Egitim ve Arastirma Hastanesi, Pediatri Klinigi, Istanbul, Turkey.

Tel: +90 531 685 04 84 e-mail: gozdeercan91@hotmail.com

© Copyright 2020 by Istanbul Provincial Directorate of Health - Available online at www.northclinist.com



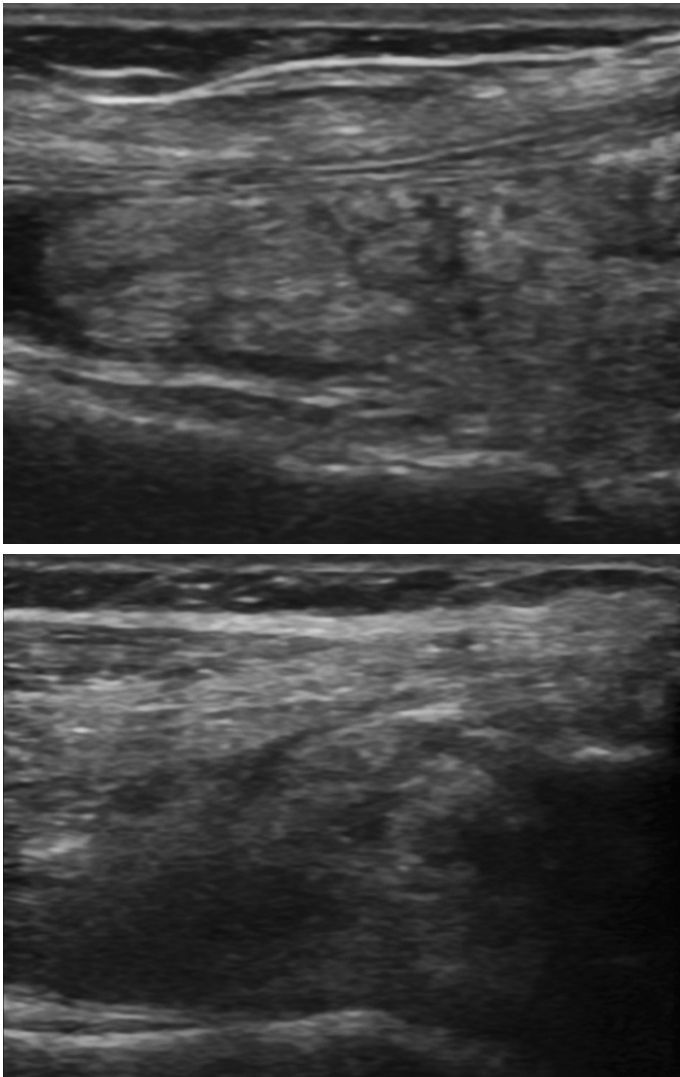


FIGURE 1. Both ultrasonography images show frond-like villous projections of the synovium and massive effusion in the suprapatellar pouch, the left image is the first presentation of the patient to our clinic, right image is features of the suprapatellar pouch after the treatment. Mild decrease in villous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

frond-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 the different signal intensity of fat on all sequences (Fig. 2, 3).

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

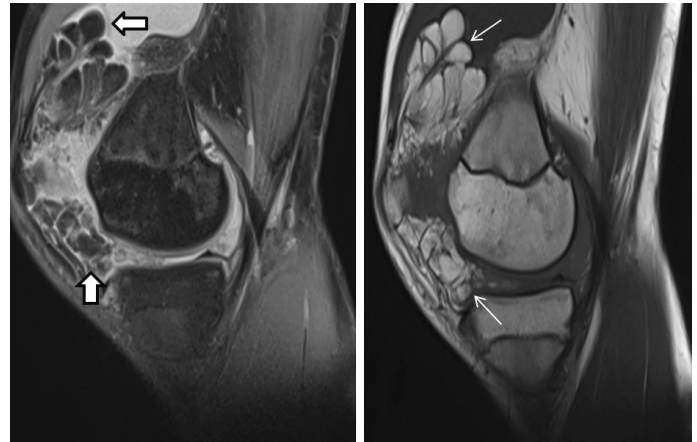


FIGURE 2. Saggital fat saturation T2A images (thick arrow) and saggital T1A images (thin arrow) shows the fat-containing frond-like synovial mass and suprapatellar projections outlined by concurrent joint effusion in the infrapatellar fat pad and quadriceps fat pillow. The lesion follows the signal intensity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

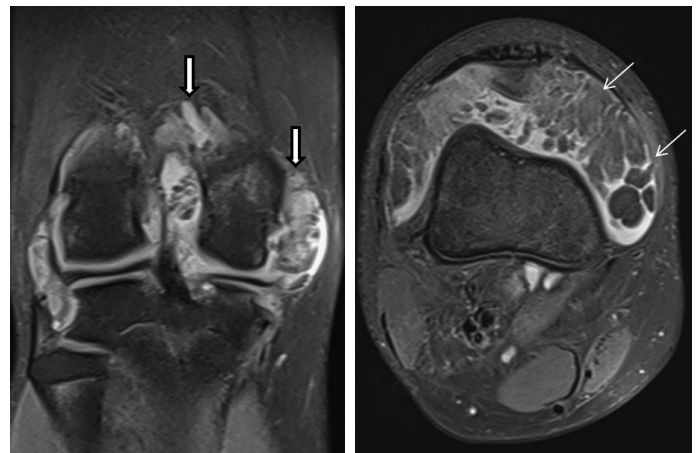


FIGURE 3. The axial (thick arrow) and coronal (thin arrow) fat saturation T2A images we also see synovial hypertrophy and bursal effusions in chronic JIA patient.

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

DISCUSSION

We presented the clinical progression and outcome of a JIA patient complicated with LA. She had bilateral knee joints involvement and slowly progressed into 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 [3]. It is mainly reported in the knee joint although the shoulder, elbow, hip, ankle and wrist joints have been and typically described as unilateral disease in the literature [1, 2, 3, 4]. 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 [5]. 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]. The involvement of both knees has been reported in up to 20% of affected patients in some studies [1, 2, 7]. Laboratory findings are generally unremarkable. We did not find any notable findings as is the case with our patient. 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 typically display a high echopattern, similar to that of adjacent subcutaneous fat and within 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 a 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 [3]. 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 by the time. The development of synovial LA in bilat-

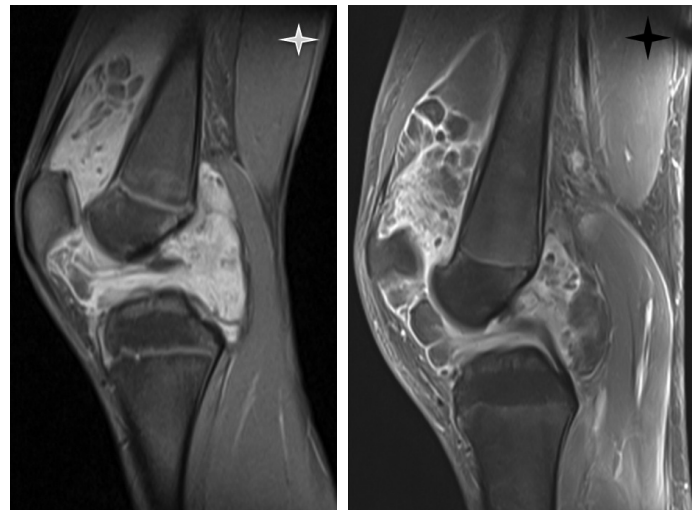


FIGURE 4. We see the progression of the disease between 2013 (white star) to 2018 (black star) years in sagittal fat saturation images.

eral knees and synovial fronds with fat signals became more common in five years on the patient (Fig. 4).

Lipoma arborescens does not require an aggressive surgical treatment unless it is symptomatic despite conservative management [1]. The primary treatment of the LA is to reduce the further progression of secondary type and its associated symptoms. However, in advanced primary and difficult cases of secondary LA, surgery may 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 that was suffering from bilateral persistent knee swelling, diagnosed with JIA for five years. The underlying mechanism of persistent swelling leads us to the diagnoses of LA as a result of imaging. In the literature, so many LA cases indicate a benign slow progress, but fewer reported cases that have been complicated by JIA. This case highlights LA associated with JIA and due to slow progression; extremely widespread synovial fronds were detected.

Acknowledgements: We wish to thank the patient and her parents for consenting to the publication of this case report, Dr. Ismail Islek for participated in discussion of the differential diagnoses.

Informed Consent: Written informed consent was obtained from the patient for the publication of the case report and the accompanying images.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: This report did not receive any spesific grant from funding agencies in the public, commercial or nor-for-profit sectors.

Authorship Contributions: Concept – GE, SK, BS; Design – GE, SK, BS; Supervision – GE, SK, BS; Data collection and/or processing – GE, BS; Analysis and/or interpretation – BS; Litarature review – GE, BS; Writing – GE, SK, BS; Critical review – BS.

REFERENCES

1. Sanamandra SK, Ong KO. Lipoma Arborescens. Singapore Med J 2014;55:5–11.
2. Al-Ismail K, Torreggiani WC, Al-Sheikh F, Keogh C, Munk PL. Bilateral lipoma arborescens associated with early osteoarthritis. Eur Radiol 2002;12:2799–802.
3. Mohammad HR, Chaturvedi A, Peach C. An unusual case of lipoma arborescens. Ann R Coll Surg Engl 2016;98:e126–9.
4. Kamran F, Kavin K, Vijay S, Shivan G. Bilateral lipoma arborescens with osteoarthritis knee: Case report and literature review. J Clin Orthop Trauma 2015;6:131–6.
5. Vilanova JC, Barceló J, Villalón M, Aldomà J, Delgado E, Zapater I. MR imaging of lipoma arborescens and the associated lesions. Skeletal Radiol 2003;32:504–9.
6. Plotkin BE, Varma R. Lipoma Arborescens of the Knee in a 17-Year-Old Man. Radiol Case Rep 2015;3:164.
7. Yan CH, Wong JW, Yip DK. Bilateral knee lipoma arborescens: a case report. J Orthop Surg (Hong Kong) 2008;16:107–10.