

Case Report

Lipoma Arborescens in Childhood: A Report of Two Sisters

Çocukluk Çağında Lipoma Arboresan: İki Kız Kardeşin Sunumu

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Lipoma arborescens (LA) is a rare benign lesion of synovial joints and bursae. The knee is the most common site of the involvement. Magnetic resonance imaging findings are typical in the diagnosis. The number of case reports of LA in children is significantly lower compared to the adults. In this article, two nine-year-old and 15-year-old sisters who were examined for the complaints of bilateral pain and swelling in knees were presented. Magnetic resonance imaging demonstrated typical features of LA in the right knees of both children. Pediatric patients with mono/oligoarticular effusion and synovial hypertrophy without systemic involvement, LA should be considered in differential diagnosis. Magnetic resonance imaging is a sensitive modality in the diagnosis of LA.

Key words: Familial tendency; juvenile oligoarthritis; knee; lipoma arborescens; magnetic resonance imaging.

Lipoma arborescens (LA) is a rare benign lesion of the synovial joints and bursae characterized by the replacement of the synovial tissue with mature fat cells, leading to villous proliferation. The etiology is unknown. Although it has been reported both in adults and in children, there are far fewer case reports of LA in children compared to adults. The knee is the most common site of involvement, but presentations in the wrist, shoulders, and hips have Lipoma arboresan (LA) sinovyal eklemler ve bursaların nadir benign bir lezyonudur. En sık tutulum yeri dizlerdir. Manyetik rezonans görüntüleme bulguları tipiktir. Çocuklardaki LA olgu sunumlarının sayısı, erişkinlere kıyasla oldukça azdır. Bu çalışmada, her iki dizde ağrı ve şişlik yakınması muayene edilen dokuz ve 15 yaşlarındaki iki kız kardeş sunuldu. Manyetik rezonans görüntülemede her iki çocuğun sağ dizinde LA için tipik özellikler gösterildi. Çocukluk çağındaki hastalarda sistemik tutulum olmadan mono/oligoartiküler efüzyon ve sinovyal hipertrofi varlığında, ayırıcı tanıda LA akla getirilmelidir. Manyetik rezonans görüntüleme, LA tanısında duyarlı bir yöntemdir.

Anahtar sözcükler: Ailesel yatkınlık; juvenil oligoartrit; diz; lipoma arboresan; manyetik rezonans görüntüleme.

also been described.^[1,2] Clinically, the most common finding is slow-growing, painless swelling. Most of the affected patients have no other rheumatologic problems. However, in some, osteoarthritis, rheumatoid arthritis (RA), gout, psoriatic arthritis, sarcoidosis, and joint trauma have been reported.^[1,2] Although plain radiography, ultrasonography (USG), direct arthrography, and computed tomography (CT) are helpful, the magnetic resonance imaging (MRI)

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results which have revealed LA have generally been considered diagnostic. A surgical synovectomy is the usual recommended treatment.^[1-6]

We report on the cases of two sisters, one being nine years old and the other 15 years old, who were evaluated because of bilateral pain and swelling in their knees. Although routine laboratory evaluations were within normal limits, an MRI demonstrated characteristic features of lipoma arborescens in the right knees of both children.

CASE REPORT

Case 1– A 15-year-old female admitted with a two-year history of bilateral knee pain and swelling that had been prominent in the right side for the last year. She had also complained of locking in the knee joint. Her past medical history was negative for tuberculosis, chronic fever, trauma, psoriasis, inflammatory bowel disease, uveitis, or back pain. She reported mild pain relief with nonsteroidal anti-inflammatory drugs (NSAIDs).

On physical examination, tender swellings that were more prominent at the suprapatellar level of both knees were detected. A patellar shock test was positive on the right side. No redness or localized heat increase was reported. There was no limitation in the range of movements in either knee; however, the right knee was painful during examination. The McMurray test, the Apley test (meniscal rotator test), and stability tests were negative.

Laboratory findings, including erythrocyte sedimentation rate (ESR), C-reactive protein (CRP), immunoglobulin M (IgM) rheumatoid factor (RF), full blood count, complements, Ig levels, and routine biochemistry tests, were within normal limits. Antinuclear antibodies (ANA), anti-double-stranded deoxyribonucleic acid (anti-dsDNA), Brucellar agglutination, and human immunodeficiency virus (HIV) tests were negative. Protein electrophoresis was also normal, but the urine and throat cultures were negative.

Synovial fluid aspiration was obtained from the right knee. The fluid was yellow in color and nonpurulent in nature. In a mucin coagulation test, there was strong mucin positivity. A microscopic evaluation demonstrated 200 leukocyte/mm³, 20 erytrocyte/mm³. Aspirated synovial fluid culture samples for common pathogens and tuberculosis were negative.

X-rays of the knees were normal except for mild degenerative changes. Sonographic examinations of

the knees were performed by the Logiq 9 ultrasound imager (GE Medical Systems, Milwaukee, Wis) with a high frequency linear array probe. These revealed globular and villous hyperechoic structures projecting into the suprapatellar effusion in the right knee. There were focal areas of hyperechogenicity suggesting fatty deposition (Figure 1). Magnetic resonance imaging was performed using a 1.5-T MR unit (Symphony Vision, Siemens Medical Systems) with an extremity coil for knee studies. The sequence included T1- and T2-weighted spin echo, T1-weighted gradient echo, and T2-weighted fat-suppressed spin echo in the axial, coronal, and sagittal planes.

Magnetic resonance imaging revealed a large effusion and numerous frond-like projections which were prominent in the suprapatellar compartment of right knee. The intensity of these frond-like synovial projections, which were suppressed in fatsaturated sequences, was similar to the intensity of fat (Figures 2a, b). An effusion and a mass-like lesion were also detected in the popliteal bursa of the right knee. (Figures 3a, b) In addition, mild osteochondral changes occurred in both knees which were more pronouced on the right side (Figure 2a). Suprapatellar effusion was also detected in the left knee. No meniscal or ligament pathology was detected in either knee.

The presence of the large effusion and the numerous frond-like synovial projections in conjunction with the mass-like lesion, the intensity of which was suppressed in fat- saturated sequences in the right knee, led us to the diagnosis of LA. Since the MRI of the left knee did not meet the criteria for the LA diagnosis, the patient was referred to the orthopedics department



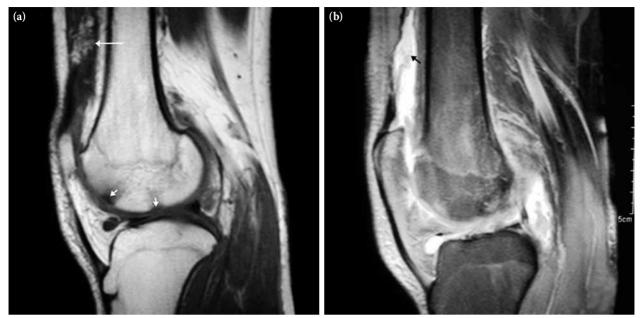


Figure 2. (a) Mild osteochondral changes (short arrows) and numerous frond-like projections (long arrow) in the suprapatellar compartment of the right knee in T1-weighted spin echo in the sagittal plane. (b) Suppression of intensity of frond-like synovial projections (arrow) in the right knee in T2-weighted fat suppressed spin echo in the sagittal plane.

for a surgical synovectomy of the right knee and for diagnosis of the problem in the left knee. During preoperative evaluation, echocardiography revealed atrial septal defect and the patient was referred to pediatric cardiology department for further evaluation.

Case 2- A nine-year-old female presented with bilateral knee pain and swelling which had persisted

for one year. The pain and swelling were more prominent in her right knee, and she described recent aggravation of the pain. Her past medical history was unremarkable.

On physical examination, there were tender swellings and local heat increases in both knees. A patellar shock test was positive on the right side. There



Figure 3. (a) The mass-like lesion (arrow) in the popliteal bursa of the right knee in T1-weighted spin echo in the sagittal plane. **(b)** Suppression of intensity of the mass-like lesion (arrow) in T2-weighted fat suppressed spin echo in the sagittal plane in the right knee.

was no limitation in the range of movement in either knee; however, the right knee was painful during the examination. The McMurray test, the Apley test, and stability tests were negative.

Laboratory findings were within normal limits, and the patient's urine and throat cultures were negative. Synovial fluid aspiration was obtained from the right knee. The fluid was yellow in color and noninflammatory in nature. A microscopic evaluation demonstrated 500 leukocyte/mm³. Aspirated synovial fluid culture samples for common pathogens and tuberculosis were negative.

X-rays of the knees revealed minimal degenerative changes. Sonographic examination of both knees was performed by the EUB 6000 (Hitachi Medical Systems, Japan) with a high frequency linear array probe, and this revealed villous hyperechoic structures projecting into the suprapatellar effusion and a hyperechoic pseudo-mass lesion in the suprapatellar bursa in the right knee (Figure 4). Magnetic resonance imaging was performed using a 1.5 Tesla MR unit (Intera, Philips Medical Systems, the Netherlands) with an extremity coil for knee studies. The sequence included T1- and T2-weighted turbo spin echo (TSE) images in the fat-suppressed Short Tau Inversion Recovery (STIR) sequence in the axial, coronal, and sagittal planes. Magnetic resonance imaging revealed a large effusion (Figure 5), a hyperintense masslike lesion, and numerous frond-like projections in the suprapatellar compartment of the right knee

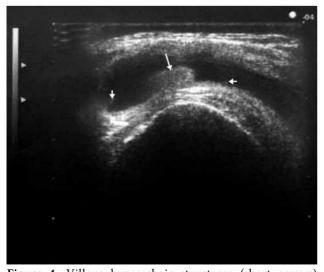


Figure 4. Villous hyperechoic structures (short arrows) and the hyperechoic pseudo-mass lesion (long arrow) projecting into the suprapatellar effusion in the right knee in ultrasonografi examination.

(Figure 6a). The intensity of these frond-like synovial projections and the mass-like lesion, which were suppressed in the fat-saturated STIR sequences, was similar to the intensity of fat (Figure 6b). Chemical shift artifact was also detected (Figure 5). The signal intensities of these lobules were isointense with fat on the T1- and T2-weighted TSE images. These findings confirmed the fatty nature of the lesion and led us to the diagnosis of LA in the right knee. There were also mild osteochondral changes in both knees prominent in the right side. No meniscal or ligament pathology was detected in either knee.

A synovectomy is accepted as the treatment of choice for LA. Post-surgical recurrences are uncommon.^[1] We referred our patients to the orthopedics department, but the family did not agree to the surgical synovectomy. Nonsteroidal anti-inflammatory drugs (NSAIDs) provided significant relief in the symptoms in both cases. We advised avoidance of weight-bearing activities and trauma to both knees and also suggested follow-up appointments every three months at one of our outpatient clinics.

DISCUSSION

The non-inflammatory conditions that may lead to suprapatellar or joint swelling/tenderness during childhood consist of trauma, bleeding disorders, or



Figure 5. The large effusion with the hyperintense masslike lesion (long arrow) and chemical shift artifact (short arrow) in the suprapatellar compartment of the right knee in T2-weighted turbo spin echo in the sagittal plane.

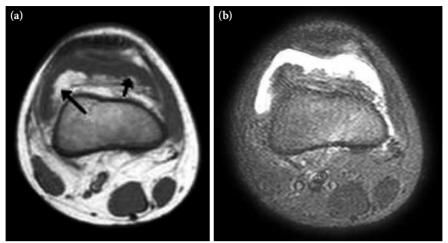


Figure 6. (a) The hyperintense mass-like lesion (long arrow) and numerous frondlike projections (short arrow) in the suprapatellar compartment of the right knee in T1-weighted turbo spin echo in the axial plane. **(b)** Suppression of intensity of frondlike synovial projections and the mass-like lesion in fat-saturated (STIR) sequence in the axial plane in the right knee.

tumors.^[7,8] Due to the absence of trauma history and the presence of a normal coagulation profile, bleeding disorders and trauma in the etiology of our cases was unlikely. Synovial disorders that might cause swelling in the knees are as rare as benign and malignant tumors.^[7,8] Lipoma arborescens is a rare intraarticular disorder characterized by villous lipomatous proliferation of the synovial membrane. It is usually seen in adults between the ages of 30 and 50 years old, and it generally involves suprapatellar recess of the knee joint. It can also be observed during childhood, sometimes bilaterally.^[7-12]

The etiology of LA is unknown. We found no information in the literature about the genetic basis of LA. In our case, detection of LA in both sisters is probably coincidental. There are some reports that have suggested trauma, chronic RA, and osteoarthritis (OA) in the etiopathogenesis.^[1,5,10] Some authors suggest that LA may induce secondary OA.^[3,12] Osteoarthritis is usually seen in advanced ages, but LA can be seen in any age group. In our cases, plain radiographs and MR imaging revealed mild osteoarthritic changes, however as our patients were in the childhood and had no history of trauma or any disorder that might lead to osteoarthritis, we think that osteoarthritis developed secondary to LA. Therefore, we believe that early diagnosis of LA is of crucial importance. There are some reports suggesting a correlation between the severity of OA and the duration of LA.^[3,9,10] However, the mild osteoarthritic changes can be attributed to the

presence of complaints for just two years in the first case and for one year in the second.

In radiological evaluation, plain radiographs may reveal focal radiolucent areas suggesting fat in the suprapatellar and articular recesses. There might be associated degenerative changes.^[1,3] Ultrasonography may show villous hypertrophic structures projecting into the suprapatellar effusion.^[3,4,7] Sonographic evaluation of the right knees in our cases revealed similar signs. We suggest that USG can be helpful in the evaluation regarding the extent and features of that joint disorder.

Because of its histological nature, MR imaging findings of LA are accepted as typical and help us for a confident preoperative diagnosis. Magnetic resonance imaging is highly accurate for the identification and characterization of LA and is the best imaging modality and the cornerstone of the preoperative diagnosis.^[13-15] Villous synovial proliferations with a signal intensity similar to that of fat in all sequences, mass-like subsynovial deposits, large effusions, potential demonstration of associated chemical shift artifact at the interlace of the synovial lesion and the effusion, and no evidence of hemosiderin deposition are included in the MRI results identifying LA.^[1,3,5,7] These typical results allow for a confident preoperative diagnosis and can exclude other possible clinical and radiological mimickers,, such as synovial osteochondromatosis, pigmented villonodular synovitis, synovial hemangioma, and synovial lipoma.^[14,15] In our cases, there was prominent suprapatellar effusion and

synovial hypertrophy. In the fat-suppressed MRI, the fatty nature of the focal areas was more clearly demonstrated. Pigmented villonodular synovitis (PVNS), synovial chondromatosis, RA, intraarticular lipoma, and synovial hemangiomas should be considered in the differential diagnosis of LA as they usually present with painless synovial thickening and effusion. In PVNS, there are focal areas of hemosiderin deposits characterized by significantly low signal intensity in both T1- and T2-weighted images. There is also contrast enhancement in PVNS. Synovial chondromatosis is defined by multiple cartilaginous nodules. These nodules have intermediate-to-high signal intensity in T2-weighted images and low-tointermediate signal intensity in T1-weighted images. Synovial hemangiomas appear as either intraarticular or extraarticular lesions of intermediate signal intensity on both T1- and T2-weighted images. There might be areas of focal low signal intensity. This suggests calcified phleboliths or fluid void in abnormal vessels. Synovial lipoma is another disorder that must be considered in differential diagnosis. In this disorder, MRI reveals a solitary, localized mass of adipose tissue without synovial changes. Chronic rheumatoid arthritis shows intermediate signal intensity on T1-weighted images and relatively decreased signal intensity on T2-weighted images within the synovium. This is associated with the formation of pannus. Lipoma arborescens is usually located in the suprapatellar recess. In contrast, PVNS, synovial hemangiomatosis, and lipoma are usually located in the infrapatellar fat pad of Hoffa.[1,10,17-19] According to the clinical and radiological/laboratory examinations and the MRI findings, we concluded that LA was the correct diagnosis in our cases. Although we referred our patients to orthopedics department for surgical synovectomy aiming not only to establish LA diagnosis conclusively, but also to treat the disease, family didn't accept the surgical synovectomy. Therefore LA diagnosis could not be confirmed histologically in our patients, though MR imaging findings were highly suggestive of LA in both cases.

In conclusion, in children or adolescent patients with mono/oligoarticular effusion and synovial hypertrophy without systemic involvement, synovial chondromatosis, PVNS, synovial hemangiomatosis, and LA should be considered in the differential diagnosis. Clinical findings with the help of MR imaging findings lead us to the presumptive LA diagnosis.

Declaration of conflicting interests

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