CASE REPORT / OLGU SUNUMU

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SURGICAL MEDICAL SCIENCES / CERRAHİ TIP BİLİMLERİ

Lipoma Arborescens: A Rare Cause of Synovial Mass of the Knee at Pediatric Age with 10 Year Follow-up

10 Yıllık Takip ile Pediatrik Yaşta Dizde Sinovyal Kitlenin Nadir Bir Nedeni Lipoma Arborescens

Mustafa Onur Karaca, Mehmet Can Gezer, Hüseyin Yusuf Yıldız

Ankara University Faculty of Medicine, Department of Orthopedics and Traumatology, Ankara, Türkiye

Abstract

Lipoma arborescens (LA) is a benign lesion that occurs in the synovial joints and consists of villous lipomatous proliferation of mature fat cells in the subsynovial tissue, often involving the knee joint. In addition to the knee joint, shoulder, elbow, and hip joint involvements have also been reported in the literature. The most common site of involvement in the knee joint is the suprapatellar pouch. Patients usually present with painless joint range of motion and swelling. Laboratory parameters are within normal limits. LA can be divided into two subtypes as primary and secondary according to the age of the patient or the underlying rheumatological diseases. The primary type is mostly seen in pediatric patients and there is no underlying chronic inflammation. In the secondary type, there is underlying chronic inflammation or trauma. In this case study, we described the operation process and 10-year follow-up of an 8-year-old pediatric patient who applied to our clinic with knee swelling.

Key Words: Lipoma Arborescens, Pediatric Patient, Follow-Up

Öz

Lipoma arboresans (LA), sinovyal eklemlerde görülen ve sıklıkla diz ekleminin tutan olgun yağ hücrelerinin subsinovyal dokuda villöz lipomatöz proliferasyonundan oluşan iyi huylu bir lezyondur. Diz eklemi dışında omuz, dirsek kalça eklem tutulumları da literatürde bildirilmiştir. Diz ekleminde en sık tutulan yer subrapatellar bursadır. Hastalar genellikle ağrısız eklem hareket kısıtlılığı ve şişlik ile başvurular. Laboratuvar parametreleri sıklıkla normaldir. LA hastanın yaşına ya da alta yatan romatolojik hastalıklara primer ve sekonder olarak 2 tipe ayrılabilir. Primer tip daha çok genç hastalarda görülür ve alta yatan kronik bir enflamasyon yoktur. Sekonder tipte ise alta yatan kronik enflamasyon veya travma vardır. Biz bu olgu çalışmamızda, kliniğimize dizde şişlik nedeniyle başvuran 8 yaşındaki pediatrik hastanın operasyon sürecini ve 10 yıllık takibini anlattık.

Anahtar Kelimeler: Lipoma Arboresans, Pediatrik Hasta, Takip

Introduction

Lipoma arborescens (LA) is characterized by villous hypertrophy, which is usually localized to the knee joint, in which connective tissue cells turn into fat cells. It is a rare benign intra-articular lesion that often causes nonspecific knee pain in advanced age.

Cases of this lesion have been examined in the literature, but the long-term has not been shown much. In this study, we tried to present the results of the 10-year follow-up of our 8-year-old patient with the diagnosis of LA.

Case Report

An eight-year-old female came with a one-month history of progressive swelling and occasional pain of the left knee. Physical examination revealed severe effusion and non-tender mass at the suprapatellar pouch with 0°-110° range of motion without pain, and there was no patellar tilt, fever or erythema.

Address for Correspondence/Yazışma Adresi: Mehmet Can Gezer
Ankara University Faculty of Medicine, Department of Orthopedics and Traumatology, Ankara, Türkiye
Phone: +90 312 508 26 96 **E-mail:** mehmetcangezer9121@gmail.com **ORCID ID:** orcid.org/0000-0001-7034-2522
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Chest radiograph and knee X-ray were normal, complete blood count was normal, erythrocyte sedimentation rate 8 mm/h, urine analysis normal, C reactive protein 0.32 mg/L, antinuclear antibody test negative, rheumatoid factor normal, immunoglobulins normal, human leukocyte antigen-B27 negative, and her celiac profile and α-1-acid glycoprotein were normal. She had pyeloplasty surgery for the left kidney in her infancy, and she had a nonfunctional kidney on the right side at her past history. There was no history of throat infection, limb trauma, recurrent or chronic pyrexia, tuberculosis or romathological disease. There was a family history of Behçet's disease of her uncle.

She underwent arthrocentesis, which yielded a clear 50 mL fluid with negative cultures, including Ziehl-Neelsen staining and culture on Löwenstein-Jensen medium. Also there was no monosodium urate or calcium phospate crystalls detected in the fluid. She was referred to rheumatology to be studied but none of the diseases were detected.

Magnetic resonance imaging (MRI) showed a high signal intensity mass-like lesion containing lobulated frondlike fat component projecting into the suprapatellar pouch and mild effusion consistent with LA (Figure 1).

There was also synovial thickening at the posterior side of the knee. Excision of the yellowish brown mass inside the suprapatellar pouch was performed via median parapatellar arthrotomy. Pathologic examination revealed LA (Figure 2). At 3-month follow-up, he was free of pain and effusion with range of flexion 0°-130°. Control MRIs taken at the third month and one year showed mild synovitis and some residual tissue adjacent to posterior capsule. After ten years of follow-up, she had no signs of LA or osteoarthritis with a Visual analogue scale score of 0 and The Musculoskeletal Tumor Society score of 30 (Figures 1-5).

Discussion

LA is a pseudotumorous lesion of the synovium that is less common in women, frequently seen in the third and fifth decades, accompanied by synovial membrane thickness and subsynovial lipid hyperplasia (1). It usually proceeds painlessly and the most common symptom is intermittent effusion (2). Although it frequently affects the knee joint, it has also been seen in the shoulder, elbow, hip and wrist joints (3,4). The suprapatellar pouch is the most common site in the knee (1,5).

LA is usually seen in middle-aged men and laboratory findings are normal (6). In the literature with more than 200 cases, less than 15 pediatric patients have been reported (7,9). Unilateral affection is more common like our patient, less than 20% cases have bilateral involvement (8,9). While LA cases are often associated with inflammatory joint diseases, trauma or arthritis, no etiological cause was found in our case (10).

Painless swelling is the most common symptom in LA like our patient. It does not often cause limitation of motion, but in our case, there was limitation in knee flexion due to swelling (11). Conditions such as juvenile rheumatoid arthritis, Lyme disease, acute rheumatic fever, hemophilia, mycobacterial tuberculosis, which cause chronic atraumatic swelling, should be kept in mind in such cases (11).

In the literature, it was thought that osteoarthritis or compression of hypertrophic synovitis between the moving joint surfaces caused the cases accompanied by limitation of movement (12,13).

There is no specific laboratory finding for LA. Joint aspirations due to both signs of infection and effusion are usually normal like our case (9,14).

In the radiological examination of LA, an increase in density can be observed in the suprapatellar bursa on direct

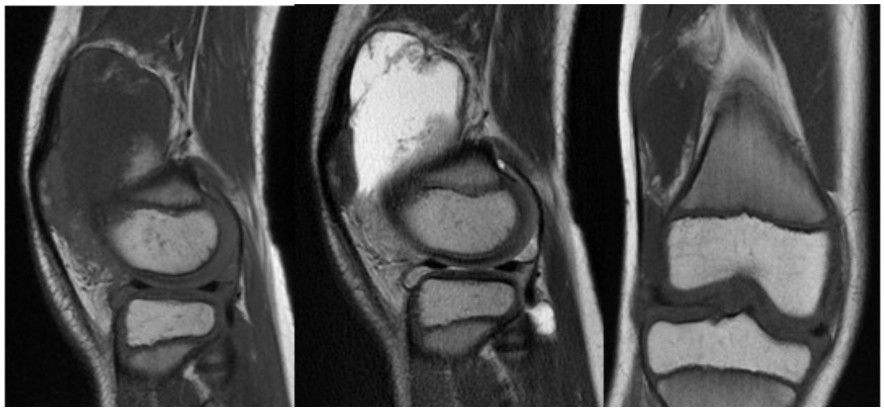

Figure 1: In sagittal T1 and T2 and coronal T1 sections, hypertrophy is observed in the suprapatellar bursa and posterior joint location with synovial thickening and adipose tissue

radiographs (Figure 3). Although ultrasonography and computed tomography are used for diagnosis, MRI is the gold standard (15-17). Arborescent or frond like proliferations of the thickened synovium containing globules or lobules of fast signal intensity on images obtained with all pulse sequences, associated joint effusion, absence of magnetic susceptibility effects originating from hemosiderin, and potential chemical shift artifacts at the fat fluid interface are the characteristic MRI features of LA (15,16). In our case, synovial thickening in the suprapatellar bursa and posterior joint space and villous hypertrophy in the adipose tissue were observed on MRI, and although pigmented villonodular synovitis, synovial osteochondromatosis, and synovial hemangioma were considered in the differential diagnosis, LA was considered as a preliminary diagnosis.

LA is such a rare disease that definitive treatment guidelines are lacking. The most common treatment method is synovectomy. Open or arthroscopic synovectomy can be applied as a treatment method. When synovectomy is performed with arthroscopy, residual tissue may remain, and adequate excision of arthroscopic synovectomy may not be achieved in large masses (8,14). Another treatment option for LA have is intra-articular injection of radio-active compounds such as Yttrium 90 or steroids. However, since LA may have adhesion to the surrounding tissues, cartilage and other surrounding tissues may be damaged with this method (19).

The recurrence rate has been reported very little in the literature. In a review by Wang et al. (20), 28 studies that performed arthroscopic synovectomy were examined and the recurrence rate was found to be 2.8%. However, most of the patients in these studies were adult patients. In addition, the follow-up period of the patients in this study ranged from 3 weeks to 84 months. In our case, the patient was followed from childhood to adulthood both clinically and radiologically for 10 years, and there was no clinical or radiological recurrence in the patient (Figures 4 and 5).

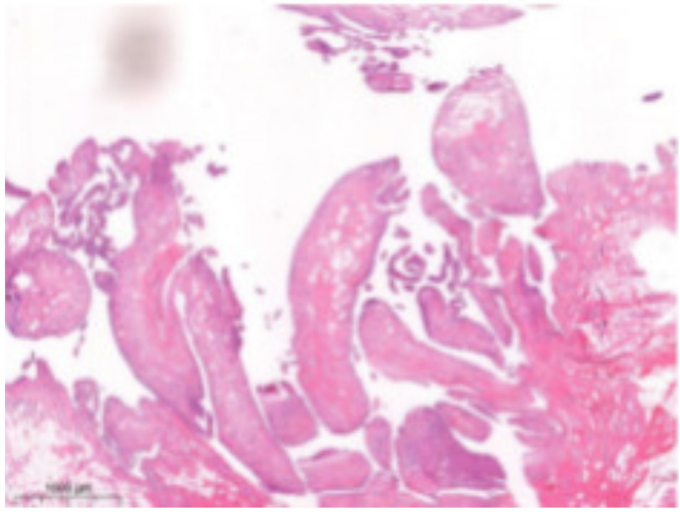

Figure 2: Microscopic image of the lesion. Dense fat cells covered with synovial connective tissue extending in villous structure and mononuclear cells with capillary proliferation are seen in places (H&E, x10)

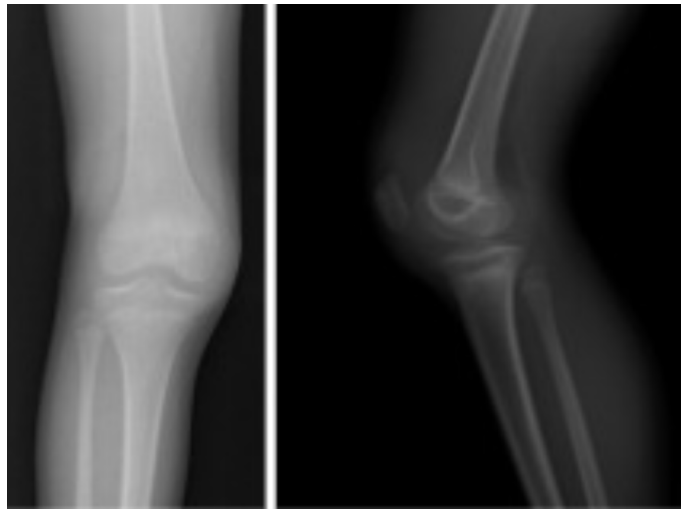

Figure 3: X-ray images of the case, an increase in density is observed in the suprapatellar bursa in the lateral radiograph

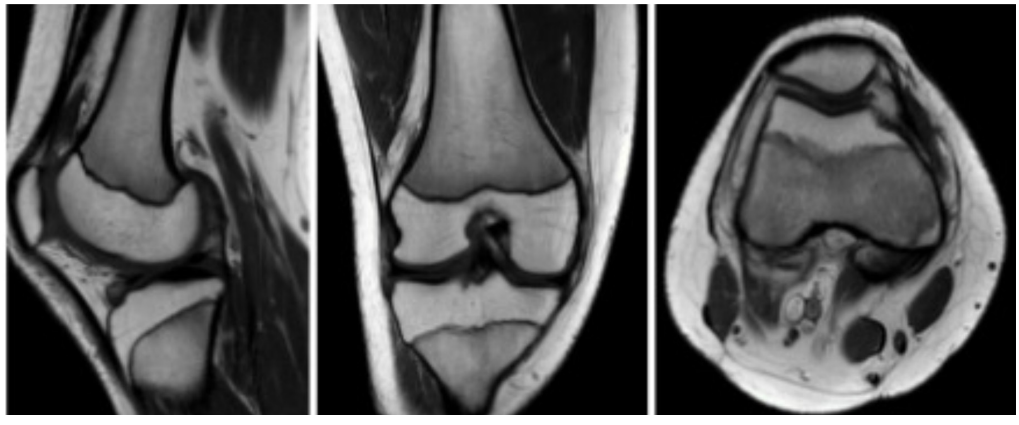

Figure 4: No recurrence was observed in the sagittal, coronal and axial images of the MR image of the case in the 6th postoperative year

MR: Magnetic resonance

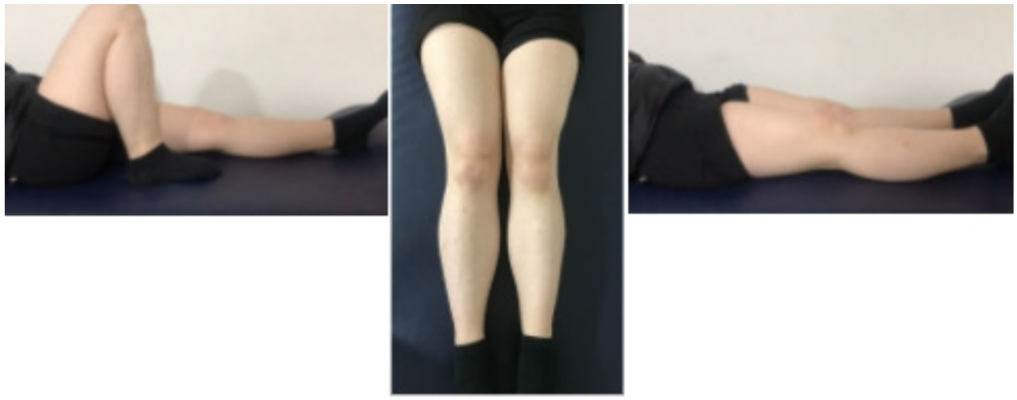

Figure 5: In the 10th postoperative year of the patient, knee joint roms were painless and fully open

LA should be considered in the differential diagnosis of pediatric patients with painless swelling of the knee joint. The patients who were treated by synovectomy had complete healing of the synovial lesion without osteoarthritis in the pediatric population.

Ethics

Informed Consent: Written informed consent was obtained from the patient for this case report.

Peer-reviewed: Externally peer-reviewed.

Authorship Contributions

Concept: M.C.G., H.Y.Y., Design: M.O.K., M.C.G., H.Y.Y., Data Collection and Processing: M.C.G., Analysis or Interpretation: M.O.K., M.C.G., Literature Search: M.O.K., M.C.G., Writing: M.O.K., M.C.G., H.Y.Y.

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