Left Knee Pain and Bilateral Knee Swelling in an Adolescent

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History and Physical Examination

A 14-year-old boy was referred by the rheumatology department to our orthopaedic service for evaluation of new-onset left knee pain and swelling during the past 2 months. The patient reported no history of injury or trauma and had no prior left knee problems. He had no mechanical symptoms, such as locking, catching, and giving way. He also had not experienced fevers, chills, weight loss, or night pain, but he did notice worsening of his symptoms when he was more active, whereas rest improved his symptoms. He did not have pain at rest.

The patient had presented 2 years earlier to his pediatrician with painless right knee swelling. He then underwent an outside orthopaedic and rheumatology evaluation. A review of the medical record from these earlier visits revealed a mild right knee effusion, a stable knee examination, no pain on palpation of his right knee, and a level gait. He was able to jump on each foot and arise from

a squat without pain, and he had no swelling or pain symptoms in other joints. Laboratory evaluation at that time revealed an erythrocyte sedimentation rate of 2 mL/hour, C-reactive protein of 0.11 mg/dL, and negative antinuclear antibody, HLA-B27, and rheumatoid factor tests, and a white blood cell count of $6.4 \times 10^3/\text{mm}^3$. An arthrocentesis was performed, which was not suggestive of Lyme disease, crystalline arthropathy, or infection. Gram stain and cultures were negative. Six months later, he was evaluated by rheumatology and his examination was unchanged.

His father had a positive history for enthesopathy (plantar fasciitis), but his family history otherwise was negative for reactive arthritis, rheumatoid arthritis, inflammatory bowel disease, psoriasis, ankylosing spondylitis, systemic lupus erythematosus, scleroderma, dermatomyositis, Raynaud disease, and sarcoidosis. The rheumatology service made a diagnosis of oligoarticular juvenile idiopathic arthritis. The patient was treated with cortisone injections; however, they did not provide relief of his swelling. At the time of our evaluation, he still was experiencing painless right knee swelling, although his primary complaint now was left knee pain and swelling. His medical history otherwise included several minor surgical procedures, including a tonsillectomy and adenoidectomy, mole removal, and tooth disimpaction. He had been taking NSAIDs for his left knee pain, but was taking no other medications and reported no drug allergies.

On physical examination, he appeared as a well-developed, well-nourished male. He ambulated with a normal, nonantalgic gait. Examination of his left lower extremity showed full, painless hip ROM. His knee ROM was 0° to 140° with some discomfort at terminal flexion and extension. He had a small joint effusion and infrapatellar fat pad

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Fig. 1A–B (A) The AP radiograph of the left knee appears normal, however, (B) the lateral view shows a small joint effusion (arrow).

hypertrophy, resulting in soft tissue fullness to his knee. No erythema was present. His knee was stable to varus and valgus stress and had firm end points on anterior and posterior drawer testing. McMurray testing was negative. He was unable to perform a single left-leg squat because of pain. Examination of his right lower extremity revealed painless full ROM. He had a similar clinical fullness to the knee, but was ligamentously stable and able to perform a single right-leg squat. All other joints were evaluated and were normal. No laboratory studies were performed at the time of our examination; however, he did undergo radiography (Figs. 1, 2) and MRI (Figs. 3, 4) of both knees.

Based on the history, physical examination, laboratory studies, and imaging studies, what is the differential diagnosis?

Imaging Interpretation

The AP radiograph of his left knee (Fig. 1A) is normal. There is no periosteal reaction, erosions, fractures, or focal bony lesions. The articular surfaces are smooth in contour and the joint spaces are maintained. The lateral radiograph of the left knee (Fig. 1B) shows normal alignment with soft tissue fullness in the suprapatellar bursa, suggesting a small joint effusion. There are no intraarticular bodies. The AP radiograph of the right knee (Fig. 2A) appears normal, however the lateral radiograph (Fig. 2B), shows soft tissue fullness in the suprapatellar bursa, suggesting a small joint effusion. No other abnormalities are seen on the radiographs.

A coronal T1-weighted image of the anterior aspect of the left knee (Fig. 3A) shows an intraarticular frond-like proliferation of high signal intensity (bright) and projects into the suprapatellar bursa. The signal characteristics follow the same signal as the subcutaneous fat, suggesting the projections contain fatty tissue. The sagittal proton density image (Fig. 3B) shows the intraarticular fatty projections along with the high signal intensity joint fluid. No intraarticular bodies were seen. The articular cartilage is normal. An axial proton density fat-saturated image was obtained at the level of the suprapatellar bursa (Fig. 3C). The frond-like projections are low signal (dark), again following the signal intensity of the subcutaneous fat and confirming that the projections contain fat content. The intraarticular projections appear contiguous with the synovium. A small joint effusion is present, correlating with the abnormality seen on the lateral radiograph (Fig. 1B). The bone marrow signal is normal. A coronal T1-weighted image of the anterior aspect of the right knee (Fig. 4A) shows an intraarticular frond-like proliferation (arrows) of high signal intensity (bright) and projects into the suprapatellar bursa, similar to the left knee. The signal characteristics follow the same signal as the subcutaneous fat, again, suggesting the projections contain fatty tissue. A sagittal proton density image (Fig. 4B) shows the intraarticular fatty projections along with the high signal intensity joint fluid. No intraarticular bodies are seen and the articular cartilage is normal. An axial proton density fat-saturated image was obtained at the level of the suprapatellar bursa (Fig. 4C). The frond-like projections are low signal (dark), again following the signal intensity of the

Fig. 2A–B (A) The AP radiograph of the right knee appears normal, but as with the left knee, the (B) lateral view shows a small joint effusion (arrow).

subcutaneous fat. The intraarticular projections appear contiguous with the synovium. A small joint effusion is present, correlating with the abnormality seen on the lateral radiograph (Fig. 2B). Normal bone marrow signal is present.

Differential Diagnosis

- Pigmented Villonodular Synovitis
- Intraarticular lipoma
- Lipoma arborescens
- Osteochondritis dissecans
- Juvenile idiopathic arthritis
- Infection
- Synovial Chondromatosis

A biopsy specimen was taken of the frond-like intraarticular fat in the left knee (Fig. 5).

Based on the history, physical examination, laboratory studies, imaging studies, and histologic picture, what is the diagnosis and how should the patient be treated?

Histology Interpretation

Grossly, the specimen consists of fibrofatty tan-white tissue. Microscopic examination of the specimen shows villi lined by hyperplastic synoviocytes with mature adipose tissue displacing normal fibrovascular stromal elements.

A mild and focal lymphoplasmacytic infiltrate is present in the stromal cores.

Diagnosis

Bilateral knee lipoma arborescens (synovial lipomatosis).

Discussion and Treatment

Lipoma arborescens is extraordinarily rare in children, and bilateral presentations are even more unusual. There are only 10 reported cases involving pediatric or adolescent patients [1, 3, 5, 8, 13, 15, 21, 23, 25, 27], and, to our knowledge, there are only five reported cases involving pediatric patients with bilateral affected joints, all of which involved the knees [5, 13, 21, 23, 27]. Because many of the symptoms are nonspecific, this unique condition is even more likely to be overlooked. The knee is the most commonly affected joint, although other joints can be affected. Patients typically present with a subtle onset of joint swelling. There is no associated erythema or warmth. As the swelling continues, it often is accompanied by intermittent episodes of joint pain. While mechanical symptoms are possible, the mass effect of the lesion more commonly results in restricted ROM. The tumor is most commonly located in the suprapatellar pouch when affecting the knee. If large enough, a soft, nontender mass can be palpated.

Fig. 3A–C (A) Coronal T1-weighted and (B) sagittal proton density MR images of the left knee show the frond-like projections in the suprapatellar region (arrows). Joint fluid is present. The signal intensity of the projections is the same signal as the subcutaneous fat. (C) An axial

proton density fat-saturated image shows a small joint effusion with intraarticular frond-like projections (*) adjacent to the synovium (arrows). The intraarticular projections are low signal (similar to the subcutaneous fat), confirming that the projections are fatty.

Symptoms may progress slowly during several years. Patients also might have joint line pain and crepitus, as the lesion can be associated with degenerative joint changes. Laboratory tests typically are normal. Synovial fluid analysis does not show an elevated white blood cell count or other significant findings.

Although its clinical presentation can be ambiguous, the distinguishing features for this lesion are most easily found in its unique appearance on MR images [22]. Radiographs of the affected joint may appear normal. In some cases, degenerative joint changes or bony erosions may be present. MRI is the preferred imaging modality for lipoma

arborescens. The MRI findings in lipoma arborescens are characteristic, with villous or frond-like intraarticular projections that follow signal characteristics of fat in all pulse sequences, which are better seen on T1-weighted or fat-suppressed T2-weighted sequences [17, 26]. However, because of the rarity of the lesion, these findings may be overlooked on MR images if one is not aware of the diagnosis. The definitive diagnosis can be made by arthroscopic examination, intralesional excision, and biopsy; histologic samples can confirm the diagnosis.

Pigmented villonodular synovitis (PVNS) can commonly mimic other disorders. It presents with insidious

Fig. 4A–C (A) Coronal T1-weighted and (B) sagittal proton density MR images of the right knee show a frond-like mass of fat intensity in the suprapatellar region (arrows) with a small knee effusion. The signal intensity of the projections is the same signal as the subcutaneous fat. (C) The axial proton density fat-saturated image

shows a small joint effusion with intraarticular frond-like projections (arrow) adjacent to the synovium (arrowhead). The projections now appear dark, similar to the subcutaneous fat on this fat-saturated image, confirming the projections contain fat.

onset of knee stiffness and swelling [10]. On examination, nearly all patients present with an effusion, and nearly $\frac{1}{2}$ have a synovial mass. Patients also may present with a flexion contracture. MRI findings in pediatric patients with PVNS include hyperplastic synovium with a characteristic paramagnetic effect attributable to hemosiderin deposition, which results in low signal intensity on all sequences. However, despite a similar clinical presentation, this patient's MR images showed characteristic frond-like configuration of the lesion, which had the same signal

intensity as adipose tissue and is consistent with lipoma arborescens, but not typical of PVNS.

Although an intraarticular lipoma most commonly occurs in the knee and can present with similar symptoms, MRI findings for synovial lipomas are identical to those of lipomas in other locations, which include a homogenous, well-circumscribed, solitary lesion that matches the signal of adipose tissue on all sequences. These findings are inconsistent with findings observed on our patient's MR images.

Fig. 5A–C Histologic examination showed (A) multiple thickened, frond-like synovial villi with abundant stromal fat (Stain, hematoxylin & eosin; magnification $\times 20$). (B) A separate field of same specimen shows two synovial-lined villi with stromal infiltration by fat and mild lymphohistiocytic inflammation (Stain, hematoxylin & eosin;

magnification, $\times 40$). (C) An intermediate magnification image of a third field from same specimen highlights the above features. There is absence of hemosiderin deposition, multinucleated giant cells, or intense inflammation (Stain, hematoxylin & eosin; magnification, $\times 100$).

Osteochondritis dissecans should be considered in patients with vague knee symptoms and intermittent effusions [7]. However, the characteristic findings on MRI indicate a primary diagnosis of lipoma arborescens. While articular erosions might accompany lipoma arborescens, the findings of the actual lipoma arborescens lesion will be the predominant pathologic features observed on MR images. Erosions may vary in size and resemble arthritides such as rheumatoid arthritis, occurring more commonly on the inferomedial margin of the medial femoral condyle [20]. Osteochondritis dissecans, in contrast, more commonly affects the lateral aspect of the medial femoral condyle and has a distinct osteochondral fragment that may have fluid surrounding it in contrast to an articular erosion [7].

Juvenile idiopathic arthritis is a diagnosis of exclusion. The most common subtype is oligoarticular, affecting less than five joints. Although the majority of these patients have positive results for antinuclear antibody, they can have a negative result for rheumatoid factor, and a normal

laboratory examination [9]. Radiographs are nonspecific for early changes [18]. The most common MRI finding in patients with juvenile idiopathic arthritis is synovial hypertrophy in 65% of cases. Cartilage lesions and bone erosions are rare, occurring in less than 10% of cases [19]. The initial workup was unable to elicit a specific etiology for our patient's symptoms, but the characteristic MRI findings effectively ruled out juvenile idiopathic arthritis.

One of the most critical etiologies of joint pain and swelling that must be excluded on initial examination is infection, including unusual infections such as Lyme disease. Given the chronicity of our patient's symptoms, waxing and waning symptom severity, bilateral involvement, and lack of constitutional symptoms, an infectious etiology seemed unlikely. Further confirming this was that at his initial presentation 2 years earlier, he underwent a thorough workup that excluded this diagnosis.

Another entity that may cause a similar clinical examination of knee fullness, pain, and possibly limited ROM is

synovial chondromatosis. This disease results from cartilage metaplasia and causes many pebble-like loose cartilage bodies in the joint. However, while the clinical examination may be similar, the distinct calcific “bag of coins” finding indicating the presence of multiple loose bodies on imaging distinguishes this disease from lipoma arborescens [12].

Lipoma arborescens is a benign tumor with unknown etiology. A association between the lesion and degenerative joint disease was reported in elderly patients, although a causal relationship between the two is not yet understood [26]. Symptoms usually start as painless swelling of the knee and may progress to include pain and mechanical symptoms or limited ROM. The onset of symptoms has been reported to take from days to nearly 30 years [20]. Treatment should be based on the patient’s symptoms, although definitive treatment usually is a synovectomy. It is important to arrive at a proper diagnosis, as the severity of erosive changes to the joint and arthritic symptoms has been correlated to the duration of the disease [14]. Treatment with steroid injection has been reported to provide symptomatic relief, although not definitive resolution [2]. Traditionally, an open synovectomy has been performed. However, successful use of arthroscopic synovectomy, as performed in our patient, has been described [11, 16, 24]. Recurrence after surgery is rare [4, 6, 24].

Owing to his continued left knee pain, the patient and his family elected that he undergo arthroscopic evaluation. A large soft tissue mass was seen in the suprapatellar pouch (Fig. 6). An intralesional biopsy was performed arthroscopically. Histologic analysis confirmed the preoperative diagnosis of lipoma arborescens. The lesion was excised arthroscopically. In addition to the primary lesion, there was a small amount of hypertrophic synovitis in the suprapatellar pouch and the intercondylar notch. An arthroscopic synovectomy was performed. The articular surfaces were observed carefully and probed. No cartilaginous defects or soft spots consistent with an osteochondritis dissecans lesion were seen.

His postoperative course was uneventful. By 3 months, he had resolution of his left knee pain, no effusion, and had advanced to full activities. We again discussed the MRI findings in his right knee with the patient and his family, which most likely indicate that he has bilateral lipoma arborescens. As he has only minor swelling and no pain of his right knee, he elected not to have surgical treatment at this time. The patient’s father was contacted via telephone 8 months postoperatively. He reported the patient was a member of the high school track team in the spring and had begun training for the fall football season. He was having no difficulty with his left knee, and his right knee remained asymptomatic. The patient will return for evaluation after the football season, and we will continue to monitor his

Fig. 6 An arthroscopic image shows the suprapatellar pouch with the frond-like tissue proliferation being removed with a pituitary forceps.

right knee for progression of size or change in symptoms, as he might require surgical treatment in the future.

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References

1. Afonso PD. Lipoma arborescens. *Arthritis Rheum.* 2012;64:2054.
2. Azzouz D, Tekaya R, Hamdi W, Montacer Kchir M. Lipoma arborescens of the knee. *J Clin Rheumatol.* 2008;14:370–372.
3. Bansal M, Changulani M, Shukla R, Sampath J. Synovial lipomatosis of the knee in an adolescent girl. *Orthopedics.* 2008;31:185.
4. Blais RE, LaPrade RF, Chaljub G, Adesokan A. The arthroscopic appearance of lipoma arborescens of the knee. *Arthroscopy.* 1995;11:623–627.
5. Cil A, Atay OA, Aydingoz U, Tetik O, Gedikoglu G, Doral MN. Bilateral lipoma arborescens of the knee in a child: a case report. *Knee Surg Sports Traumatol Arthrosc.* 2005;13:463–467.
6. Coventry MB, Harrison EG Jr, Martin JF. Benign synovial tumors of the knee: a diagnostic problem. *J Bone Joint Surg Am.* 1966;48:1350–1358.
7. Crawford DC, Safran MR. Osteochondritis dissecans of the knee. *J Am Acad Orthop Surg.* 2006;14:90–100.
8. Donnelly LF, Bisset GS 3rd, Passo MH. MRI findings of lipoma arborescens of the knee in a child: case report. *Pediatr Radiol.* 1994;24:258–259.
9. Eisenstein EM, Berkun Y. Diagnosis and classification of juvenile idiopathic arthritis. *J Autoimmun.* 2014;48–49:31–33.
10. Flandry F, Hughston JC, McCann SB, Kurtz DM. Diagnostic features of diffuse pigmented villonodular synovitis of the knee. *Clin Orthop Relat Res.* 1994;298:212–220.
11. Franco M, Puch JM, Carayon MJ, Bortolotti D, Albano L, Lallemand A. Lipoma arborescens of the knee: report of a case managed by arthroscopic synovectomy. *Joint Bone Spine.* 2004;71:73–75.
12. Giancane G, Tanturri de Horatio L, Buonomo PS, Barbuti D, Lais G, Cortis E. Swollen knee due to primary synovial chondromatosis in pediatrics: a rare and possibly misdiagnosed condition. *Rheumatol Int.* 2013;33:2183–2185.

13. Haasbeek JF, Alvillar RE. Childhood lipoma arborescens presenting as bilateral suprapatellar masses. *J Rheumatol.* 1999;26:683–686.
14. Hallel T, Lew S, Bansal M. Villous lipomatous proliferation of the synovial membrane (lipoma arborescens). *J Bone Joint Surg Am.* 1988;70:264–270.
15. Huang GS, Lee HS, Hsu YC, Kao HW, Lee HH, Chen CY. Tenosynovial lipoma arborescens of the ankle in a child. *Skeletal Radiol.* 2006;35:244–247.
16. Kim RS, Song JS, Park SW, Kim L, Park SR, Jung JH, Park W. Lipoma arborescens of the knee. *Arthroscopy.* 2004;20:e95–99.
17. Martin S, Hernandez L, Romero J, Lafuente J, Poza AI, Ruiz P, Jimeno M. Diagnostic imaging of lipoma arborescens. *Skeletal Radiol.* 1998;27:325–329.
18. Munir S, Patil K, Miller E, Uleryk E, Twilt M, Spiegel L, Doria AS. Juvenile idiopathic arthritis of the axial joints: a systematic review of the diagnostic accuracy and predictive value of conventional MRI. *AJR Am J Roentgenol.* 2014;202:199–210.
19. Nusman CM, Hemke R, Schonenberg D, Dolman KM, van Rossum MA, van den Berg JM, Kuijpers TW, Maas M. Distribution pattern of MRI abnormalities within the knee and wrist of juvenile idiopathic arthritis patients: signature of disease activity. *AJR Am J Roentgenol.* 2014;202:W439–446.
20. Ryu KN, Jaovisidha S, Schweitzer M, Motta AO, Resnick D. MR imaging of lipoma arborescens of the knee joint. *AJR Am J Roentgenol.* 1996;167:1229–1232.
21. Sailhan F, Hautefort P, Coulomb A, Mary P, Damsin JP. Bilateral lipoma arborescens of the knee: a case report. *J Bone Joint Surg Am.* 2011;93:195–198.
22. Sanamandra SK, Ong KO. Lipoma arborescens. *Singapore Med J.* 2014;55:5–10; quiz 11.
23. Sharma SD, Bagri DR, Gupta RK, Sarna A. Lipoma arborescens. *Indian J Pediatr.* 2011;78:1016–1018.
24. Sola JB, Wright RW. Arthroscopic treatment for lipoma arborescens of the knee: a case report. *J Bone Joint Surg Am.* 1998;80:99–103.
25. Tiao WM, Yeh LR, Lu YC, Liang HL, Chen CK. Lipoma arborescens of the knee: a case report. *J Formos Med Assoc.* 2001;100:412–415.
26. Vilanova JC, Barcelo J, Villalon M, Aldoma J, Delgado E, Zapater I. MR imaging of lipoma arborescens and the associated lesions. *Skeletal Radiol.* 2003;32:504–509.
27. Xue J, Alario AJ, Nelson SD, Wu H. Progressive bilateral lipoma arborescens of the knee complicated by juvenile spondyloarthritis: a case report and review of the literature. *Semin Arthritis Rheum.* 2013;43:259–263.