rheumato

MDPI

Case Report

Lipoma Arborescens Might Be an Unusual Cause of Knee Pain in Adolescents: A Case Report

Lorenzo Moretti [1,†], Davide Bizzoca [2,3,*,†], Andrea Michele Abbaticchio [4], Alessandro Geronimo [4], Giuseppe Solarino [4] and Biagio Moretti [4]

[1] Orthopaedic and Traumatology Unit, AOU Consorziale Policlinico, Piazza Giulio Cesare 11, 70124 Bari, Italy
[2] UOSD Spine Surgery, AOU Consorziale Policlinico, Piazza Giulio Cesare 11, 70124 Bari, Italy
[3] PhD Course in Public Health, Clinical Medicine and Oncology, DiMePre-J, University of Bari "Aldo Moro", Piazza Giulio Cesare 11, 70124 Bari, Italy
[4] Orthopaedic and Traumatology Unit, DiBraiN Department, School of Medicine, University of Bari "Aldo Moro", Piazza Giulio Cesare, 70100 Bari, Italy
* Correspondence: davide.bizzoca@uniba.it
† These authors contributed equally to this work.

Abstract: Lipoma arborescens (LA) is a rare benign soft tissue tumor characterised by a hyperproliferation of villi and fat cells in the joint synovium. It is most frequently localized in the knee as reported here. This is a case report of a 16-year-old adolescent, affected by type I diabetes mellitus, who reported left knee pain and functional limitation to medical attention. She performed a physical examination, MRI and biopsy using an arthroscopic approach, leading to the LA diagnosis and classification. The LA has been thus treated with an arthroscopic synovectomy, which is the treatment of choice for LA, characterized by a low recurrence rate.

Keywords: lipoma arborescens; knee pain; adolescent; arthroscopy

Citation: Moretti, L.; Bizzoca, D.; Abbaticchio, A.M.; Geronimo, A.; Solarino, G.; Moretti, B. Lipoma Arborescens Might Be an Unusual Cause of Knee Pain in Adolescents: A Case Report. *Rheumato* **2023**, *3*, 196–200. https://doi.org/10.3390/rheumato3030014

Academic Editors: Bruce M. Rothschild and Chang-Hee Suh

Received: 12 January 2023
Revised: 21 June 2023
Accepted: 8 August 2023
Published: 14 August 2023

1. Introduction

Lipoma arborescens (LA) is a rare benign tumour characterised by fat cell proliferation in subsynovial tissue, associated with a villous proliferation of the synovium [1]. Originally described by Hoffa in 1904, LA still has an unknown aetiology [1]. LA most frequently affects the knee, where cases of bilateral involvement have been reported [1,2]. Rare is the involvement of other synovial joints such as the shoulder, elbow, wrist, hip and ankle [3].

Clinical presentation usually consists of an insidious onset of painless swelling of the affected joint, usually persisting for many years, followed by progressive pain accompanied by intermittent episodes of joint effusion [3]. Intermittent worsening pain and swelling of the involved joint may be related to the trapping of hypertrophied fatty villi between the moving joint surfaces [4–8].

To date, about 200 cases of LA have been described the in literature. Even if the spread of MRI (magnetic resonance) has led to an increase in diagnosed cases of LA, it is estimated that an LA has been diagnosed for approximately 0.25% of all MRIs performed for knee pain [4]. This report describes a case of LA of the left knee in an adolescent girl with type I diabetes mellitus.

2. Case Report

A 16-year-old adolescent (BMI = 26.9) volleyball player reported left knee pain with a functional limitation for about three months. In her medical history, she reported a diagnosis of diabetes mellitus type I at the age of nine and she has been on insulin pump treatment. She denied trauma and a family health history of arthritis and psoriasis. To control the pain symptoms, she was taking paracetamol 1000 mg tablets. She had a previous

rheumatological evaluation which excluded autoimmune arthritis and it placed the clinical suspicion of villonodular synovitis, recommending an orthopaedic consultation.

Clinically, the left knee was swollen, with appreciable distension of the suprapatellar bursa; a diffuse tenderness was palpated. There were no signs of instability in the varus or the valgus stress test; the anterior and posterior drawer tests, Lachmann tests, and McMurray tests were negatives. Full ROM, but painful beyond 90° of flexion. Patellar tracking was normal. Visual Analog Scale (VAS) = 5; Lysholm Score (LS) = 45. Moreover, clinically there was the bilateral presence of areas of necrobiosis lipoidica, linked to the underlying metabolic pathology (Figure 1).

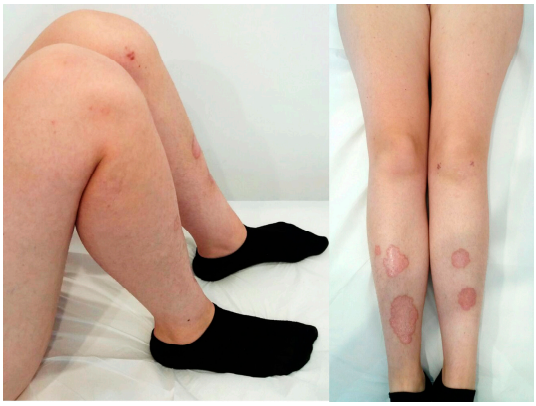

Figure 1. Patient's clinical presentation at baseline and bilateral presence of areas of necrobiosis lipoidica.

Necrobiosis lipoidica (NL) is a rare idiopathic granulomatous disease of collagen degeneration with a risk of ulceration [9]. This disease has typically been associated with diabetes and is sometimes confused with other diseases based on clinical presentation and associated locations, such as granuloma annulare, erythema nodosum, necrobiotic xanthogranuloma (NXG), chronic venous stasis ulcers, and sarcoidosis. Many components of the disease still require more research to define, such as the pathogenesis and cause as well as treatment options that can prove to be both efficacious and consistent. The incidence of NL in individuals with diabetes is only 0.3% to 1.2%; NL precedes diabetes in up to 14%, appears simultaneously in up to 24%, and occurs after diabetes is diagnosed in 62% of cases. Although NL may present in healthy individuals with no underlying disease, the most commonly associated conditions are thyroid disorders and inflammatory diseases, such as Crohn's disease, ulcerative colitis, rheumatoid arthritis, and sarcoidosis [9].

From a laboratory point of view, complete blood count, ESR, CRP, rheumatoid factor, and anti-CCP antibodies were within physiological limits. MRI of the left knee demonstrated diffuse villous and nodular hypertrophy of the synovial wall which presented in T2-weighted sequences a small signal of hyperintensity intrinsic to the villi; consistent with lipid accumulation (Figure 2).

In light of the clinical and imaging data, we opted for an arthroscopic synovectomy. During the procedure, significant and widespread villous synovitis was observed (Figure 3). A biopsy for histological examination followed by a synovectomy was performed during the arthroscopy. Histological examination showed the presence of synovial villi of different diameters, with the presence of a subsynovial lipid infiltrate, confirming the suspected diagnosis of lipoma arborescens of the synovial membrane.

At the 12-month clinical follow-up, MRI showed incomplete eradication of the LA (Figure 4). However, a complete recovery of the left knee range of motion was recorded (Figure 5); associated with an excellent clinical outcome: VAS = 0 and LS = 97.

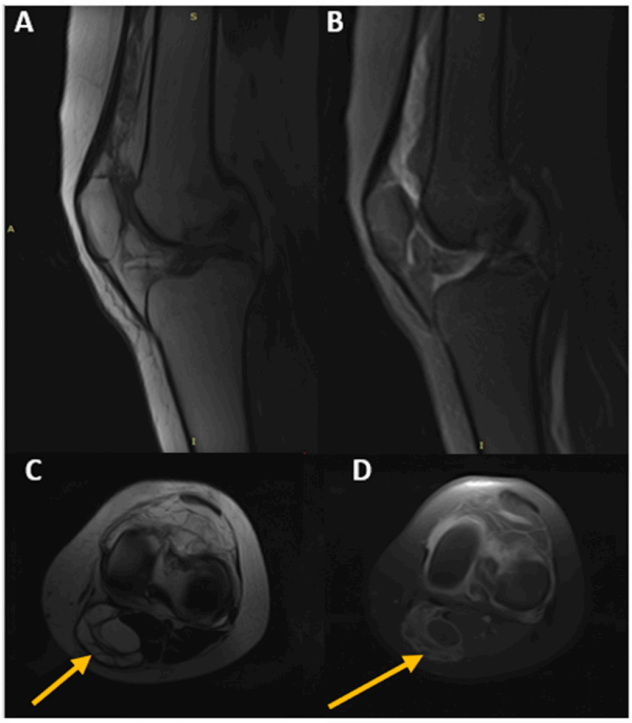

Figure 2. MRI at baseline. The arrow indicates the lesion. (**A**) T1 weighted sagittal. (**B**) STIR weighted sagittal. (**C**) T1 weighted axial. (**D**) STIR weighted axial.

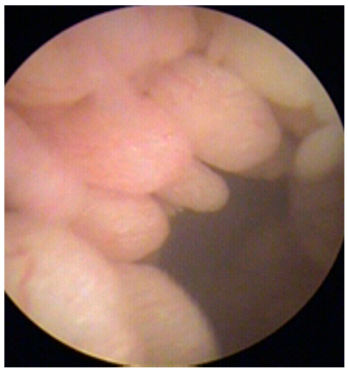

Figure 3. Villous synovitis was observed in the arthroscopy.

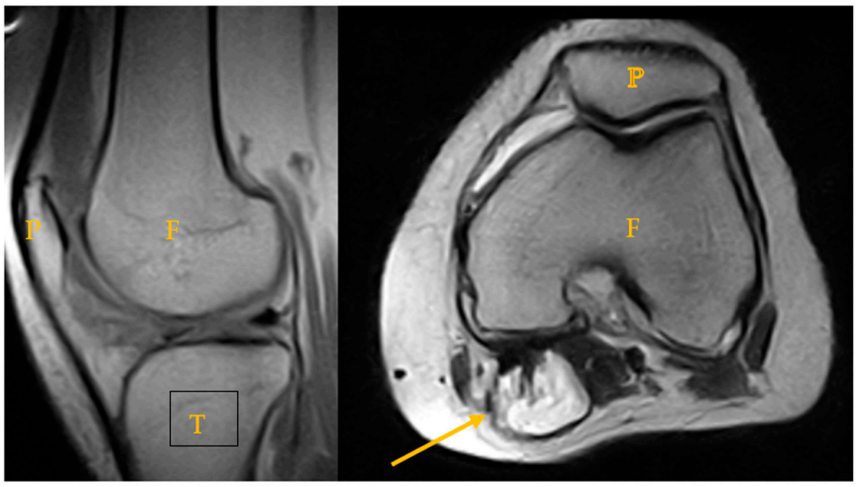

Figure 4. MRI scans at 12 months follow-up. The arrow indicates the incomplete eradication of the lesion. P = patella, F = Femur, T = Tibia.

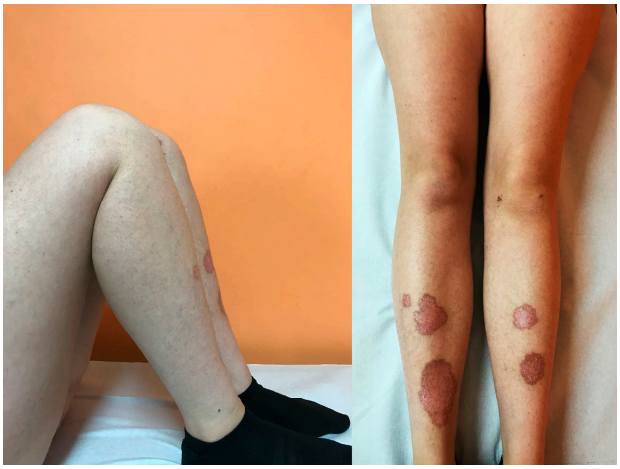

Figure 5. Clinical presentation at 12-month follow-up. In contrast to Figure 1 the left knee can now be bent to the same extent as the right knee (same range of motion).

3. Discussion

LA is a rare benign lesion that occurs mainly, but not exclusively, at the knee. It is possible to distinguish a primary LA (typical of young adults) and a secondary LA (more frequent in elderly patients) with concomitant inflammatory arthropathy [4].

As regards its etiopathogenetic origin, it remains unknown. As far as in the literature it has been seen that many lesions were secondary to traumatic or degenerative events [4], and also diabetes mellitus and the use of steroid drugs have been hypothesized to be associated with this pathology.

From a pathogenetic point of view, LA represents the consequence of the synovium's chronic non-neoplastic inflammatory process. In secondary LA, a synovial infiltration of chronic inflammatory cells can be found at the level of the villi [4]. In association with LA, it may be possible to experience a bone hypertrophic reaction, cartilage or meniscal lesions, and joint effusion [4,5]. In the differential diagnosis, it is appropriate to consider: rheumatoid arthritis, villonodular synovitis, synovial osteochondromatosis and synovial hemangioma [4–9].

The clinical case under examination concerns a sixteen-year-old patient affected by type I diabetes mellitus, with a concomitant LA of the left knee, who underwent arthroscopic synovectomy. Indeed, the literature reports that arthroscopic synovectomy is the treatment of choice in the presence of an LA. The recurrence rate after arthroscopic synovectomy is extremely low; in our case, no signs of LA recurrence were observed 12 months after surgery.

The patient under examination presents a series of atypical aspects, unlike the classic presentation of the LA. First of all, the average age of onset is between the fourth and fifth decade of life, while the patient presented is a 16-year-old young man [10]. Secondly, the presence of ROM limitation and joint pain and swelling does not occur so frequently in patients with LA [11]. Finally, cases of association of LA with type I diabetes mellitus are rare.

In our opinion, it should be considered that when the patient came to our observation, she had good glycemic compensation. This glycemic control was also maintained during the follow-up. However, the presence of areas of lipoid necrolysis in the lower extremities documents poor glycemic control in the past. In the literature, a clear association between diabetes and LA has not yet been documented. Only in a few cases, patients affected by LA presented a positive family history of diabetes mellitus or were affected by this metabolic pathology [2,3,8].

4. Conclusions

LA is a benign lesion that predominantly, but not exclusively, affects the knee. From a clinical point of view, it is described by knee pain, effusion, and joint swelling. The elective treatment is arthroscopic synovectomy, which is characterised by a low recurrence rate.

Author Contributions: Conceptualization, L.M. and D.B.; methodology, D.B.; validation, G.S. and B.M.; data curation, D.B.; writing—original draft preparation, A.M.A. and A.G.; writing—review and editing, D.B.; visualization, L.M.; supervision, G.S.; project administration, B.M. All authors have read and agreed to the published version of the manuscript.

Funding: This research received no external funding.

Institutional Review Board Statement: The study was conducted following the Declaration of Helsinki, and approved by the Ethics Committee of AOU Policlinico di Bari (Comitato Etico Interregionale Policlinico di Bar, Prot. N. 5836, 6 May 2019).

Informed Consent Statement: Written informed consent has been obtained from the patient(s) to publish this paper.

Data Availability Statement: Not applicable.

Conflicts of Interest: The authors declare no conflict of interest.

References

1. Utkan, A.; Ozkan, G.; Köse, C.C.; Ciliz, D.S.; Albayrak, A.L. Congenital absence of the medial meniscus associated with lipoma arborescens. *Knee* **2010**, *17*, 258–260. [CrossRef]
2. Sailhan, F.; Hautefort, P.; Coulomb, A.; Mary, P.; Damsin, J.P. Bilateral lipoma arborescens of the knee: A case report. *J. Bone Jt. Surg. Am.* **2011**, *93*, 195–198. [CrossRef] [PubMed]
3. Vilanova, J.C.; 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. [CrossRef] [PubMed]
4. Zhu, G.; Tian, X.; Du, D.; Lei, M.; Guan, L.; Wang, J.; Tan, Y.; Yang, C.; Zheng, X. Medical Image Analysis of Knee Joint Lipoma Arborescens and Arthroscopic Treatment. *Comput. Med. Imaging Graph.* **2018**, *66*, 66–72. [CrossRef] [PubMed]
5. Kulkarni, H.G.; Kulkarni, G.S.; Kulkarni, P.G. Lipoma Arborescens—Eyes See What Mind Knows! *J. Orthop. Case Rep.* **2017**, *7*, 59–62. [PubMed]
6. Loriaut, P.; Djian, P.; Boyer, T.; Bonvarlet, J.-P.; Delin, C.; Makridis, K.G. Arthroscopic treatment of localized pigmented villonodular synovitis of the knee. *Knee Surg. Sports Traumatol. Arthrosc.* **2012**, *20*, 1550–1553. [CrossRef] [PubMed]
7. Karantanas, A.H.; Zibis, A.H.; Papanikolaou, N. Comparison of echo planar imaging, gradient echo and fast spin echo MR scans of knee menisci. *Comput. Med. Imaging Graph.* **2000**, *24*, 309–316. [CrossRef] [PubMed]
8. Bernardo, A.; Bernardes, M.; Brito, I.; Vieira, A.; Ventura, F. Lipoma arborescente da sinovial. *Acta Med. Port.* **2004**, *17*, 325–328. [PubMed]
9. Sibbald, C.; Reid, S.; Alavi, A. Necrobiosis Lipoidica. *Dermatol. Clin.* **2015**, *33*, 343–360. [CrossRef] [PubMed]
10. Kalifis, G.; Maffulli, N.; Migliorini, F.; Marín Fermín, T.; Hovsepian, J.M.; Stefanou, N.; Hantes, M. Surgical management of upper limb lipoma arborescens a systematic review. *J. Orthop. Surg. Res.* **2022**, *17*, 138. [CrossRef] [PubMed]
11. Sanamandra, S.K.; Ong, K.O. Lipoma arborescens. *Singap. Med. J.* **2014**, *55*, 5–10; quiz 11. [CrossRef] [PubMed]