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Case Report

Bilateral Lipoma Arborescens of the Knees in a Patient with Pachydermoperiostosis: A Case Report

Mariem Ghribi, Faten Frikha*, Fatma Rekik, Abir Derbel and Zouhir Bahloul

Department of Internal Medicine, Hedi Chaker Hospital Sfax, University of Sfax, Tunisia

***Corresponding Author:** Faten Frikha, Department of Internal Medicine, Hedi Chaker Hospital Sfax, University of Sfax, Tunisia.

Received: February 06, 2021; **Published:** March 29, 2021

Abstract

Introduction: Lipoma arborescens presents a rare cause of chronic, slow-growing lesion which is characterized by proliferation of the synovium, with replacement of the subsynovial tissue by mature fat cells. It is most commonly observed in the knee joint, however other joints involvement have also been seen. Herein, we describe the exceptional case of bilateral Lipoma arborescens of the knees in a patient with pachydermoperiostosis (primary Hypertrophic osteoarthropathy).

Case Presentation: We report the case of a 26-year-old male presenting with swelling and joint effusion of right and left knees. The diagnosis of bilateral lipoma arborescens of the knees was considered, based on Magnetic resonance imaging (MRI) and histological findings. He was also diagnosed with complete primary form of pachydermoperiostosis according to physical examination, and investigation (thickening of the skin, peculiar cutis vertices gyrate, hyperhidrosis, clubbing and periosteal reaction in bones). The patient was treated by an open synovectomy associated with colchicine and oral paracetamol with a good response.

Conclusion: Lipoma arborescens is a rare entity. Bilateral involvement of the knees is rare and its association with pachydermoperiostosis is extremely rare. MRI is the best imaging examination. Surgical treatment by synovectomy offers excellent outcomes.

***Keywords**: Lipoma Arborescens; Pachydermoperiostosis; Periostosis; Magnetic Resonance Imaging*

Introduction

Lipoma arborescens presents an uncommon disease of slow-growing and chronic benign lesion which is characterized by intra-articular proliferation of the synovium, with replacement of the subsynovial tissue by mature fat cells. It is a rare cause of chronic monoarticular arthritis that typically affects adults and its etiology still remains obscure [1-3].

Lipoma arborescens most commonly involves the knee joint, especially the suprapatellar pouch of this joint. Other sites may also be affected such as the hip, the shoulder, the elbow and the wrist. Clinically, patients present with pain and swelling in the affected joint. The diagnosis is based on the typical appearance on Magnetic resonance imaging (MRI) (villous proliferation of the synovium with a fatty signal), and the recommended treatment is synovectomy (by arthroscopy or by open surgery). Bilateral involvement of the knees is rare and its association with pachydermoperiostosis is extremely rare [4]. We report one case.

Case Presentation

A 26-year-old male, whose parents are second-degree cousins, was admitted to our Department with a 1-year history of slowly progressive swelling and mild pain in both knees. There was no history of prior trauma or surgery in his knees.

Clinical examination disclosed bilateral joint effusion in both knees with a restriction of motion. Skin examination revealed thickened skin with deep furrowed forehead. The skin of scalp had the appearance of a peculiar cutis vertices gyrate. Examination also revealed pan digital clubbing of all digits of hands. Patient reported hyperhidrosis of palms and soles. Examination of the cardiovascular, respiratory, and gastrointestinal systems revealed no significant abnormalities.

Laboratory analysis including erythrocyte sedimentation rate (ESR), C-reactive protein (CRP), blood cell count and thyroid function tests were normal. Liver function, serum creatinine, calcium, and uric acid level showed no significant abnormalities. The rheumatoid factor, anti-CCP, anti-DsDNA, ENA and antinuclear anti- bodies (ANA) were negative. Radiographic investigations were done to look for skeletal abnormalities. X-ray of knees depicted sub-periosteal new bone formation and cortical thickening in the femur and tibia, with preservation of the joint space.

X-rays of hands and feet showed cortical thickening with a periosteal bone reaction. The chest x-ray was normal. According to the data available from history, examination, and investigation (thickened skin, peculiar cutis vertices gyrate, hyperhidrosis, a periosteal reaction in the long bones, clubbing) after ruling out other secondary causes, the patient was diagnosed with complete primary form of pachydermoperiostosis (PDP). MRI of knees was performed. It revealed significant synovial thickening (Figure 1A and 1B) with fatty infiltration of the synovium in all sequences. There was also a joint effusion. Based on MRI findings, the diagnosis of a lipoma arborescens was suspected. An open synovectomy was performed and histological examination confirmed the diagnosis of bilateral lipoma arborescens of the knees. There was no signs of malignancy. He was treated also with paracetamol associated to daily colchicine. The response was very good and the patient returned to his normal activity after one month.

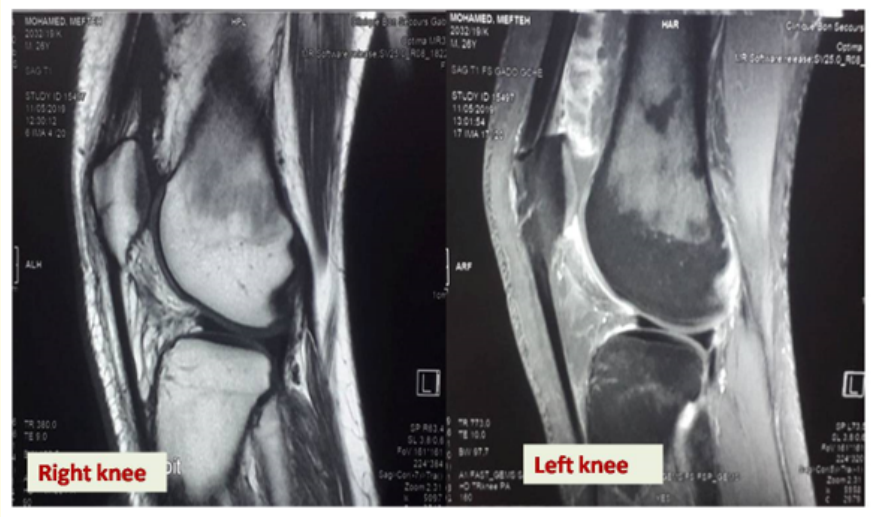

Figure 1: *MRI of right knee (Figure 1A) and left knee (Figure 1B) on sagittal T1-weighted revealed significant synovial thickening, with fatty infiltration of the synovium and joint effusion without any sign of menisci or ligaments' injury, or osseous erosions.*

Discussion

Lipoma Arborescens (diffuse articular lipomatosis) is a rare benign intra-articular lesion of unknown etiology, described first by Albert Hoffa, a German surgeon, in1904, then by Arzimanoglu in 1957 [1,5]. The term 'arborescens' is latin and means 'tree-like appearance', describing the frond-like morphology of this lesion.

It presents as an intra-articular lesion with subsynovial villous proliferation of mature fat cells mainly involving the knee. The most common form is the unilateral location involving the supra patellar recess of the knee joint. The shoulder, the elbow, the hip, the wrist, and the ankle may also be affected by this condition [6]. Bilateral and polyarticular involvements are not uncommon. We report a rare case with bilateral involvement of the knees.

Lipoma arborescens has been observed in men and women with equal frequency [7]. There are two types of lipoma arborescens: primary, idiopathic, typically affecting middle-aged, and secondary variety affecting patients with coexisting inflammatory arthritis, such as rheumatoid arthritis, gout, psoriatic arthritis [3,4,8], or patients with an underlying chronic irritation, such as degenerative disease, trauma, meniscal injury and is usually seen in elderly patients.

Our patient was a 26 year old male who presented with painless swelling of the 2 knees joints for one year with a concomitant diagnosis of a Complete form of pachydermoperiostosis.

The association of lipoma arborescens with pachydermoperiostosis like our case is extremely rare. Recently, Garnaoui H., *et al*. reported the case of a young male, diagnosed with pachydermoperiostosis for 10 years, in whom the diagnosis of lipoma arborescens of both knees was made [4]. In fact, the development of lipoma arborescens may be a reactive synovial fatty proliferation in response to chronic inflammatory attacks.

Clinically, the lesion consists of an insidious pain of the involved joint with swelling and intermittent episodes of effusion [5,9]. Exacerbation of symptoms are due to trapping of the lipoma villi in the joint space [5].

Laboratory tests including erythrocyte sedimentation rate, serum uric acid and rheumatoid factor are generally normal in the idiopathic form.

Many causes of intra-articular masses can mimic lipoma arborescens such as infectious conditions, non-infectious synovial proliferation, neoplastic and vascular diseases [10]. The main differential diagnosis includes chondromatosis, rheumatoid arthritis, villonodular synovitis, synovial hemangioma, and amyloid arthropathy.

Imaging especially the MRI examination is the gold standard to explore this disorder. It contributes to early diagnosis and to differentiate lipoma arborescens from other the intra-articular masses.

Radiography of affected joint demonstrate nonspecific soft tissue opacity. The absence of sclerosis and/or articular surface erosions can differentiate lipoma arborescens with gouty arthropathy or pigmented villonodular synovitis.

MRI remains the examination of choice revealing the villous proliferation of the synovial with a fatty signal on all sequences which are suppressed on FAT-SAT, accompanied by joint effusion [1]. This specific MRI findings can lead to the diagnosis of lipoma arborescens by distinguishing it from other intra-articular lesions.

Lipoma arborescens is a benign condition, which can be treated with conservative management and does not require surgical treatment unless become symptomatic. However, an arthroscopic examination is recommended to obtain tissue sampling for histological examination. The surgical treatment is either open or arthroscopic synovectomy. In our patient the diffuse aspect of the lesions with pain

and restriction of motion of both knees justified synovectomy by performing an open surgery. Recurrence of lipoma arborescens is generally rare [5].

Conclusion

Lipoma arborescens is a rare disease with only several cases reported in the literature. Bilateral involvement of the knees is rare and its association with pachydermoperiostosis is extremely rare. MRI remains the best examination for early diagnosis. Treatment by performing an arthroscopic or open synovectomy leads to good outcomes.

Conflict of Interest

The authors have no conflict of interest.

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Volume 12 Issue 4 April 2021