Lipoma Arborescens of both Knees- Case report and Literature Review

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Abstract

Introduction: Lipoma arborescens (LA) is a rare, benign intra-articular lesion most commonly found in the knee, characterised by villous proliferation of the synovium .It generally presents as a longstanding, slowly progressive swelling of one or more joints associated which may or may not be associated with pain.MRI is the investigation of choice, with images clearest on fat-supressed or STIR sequences.

Case Report: We present a 35 year old male patient, who presented with a three year history of bilateral knee pain and swelling. Magnetic resonance imaging (MRI) scans of his knee showed the characteristic features of lipoma arborescens. A 99technetium bone scan revealed increased uptake in both knees. The patient underwent bilateral arthroscopic synovectomies and made an uneventful recovery. The samples sent for histology were reported as being characteristic of lipoma arborescens.

Conclusions: Lipoma arborescens is a rare, benign intra-articular tumour which may mimic a number of other diagnoses. MRI should be considered to exclude this pathology as well as other uncommon intra-articular pathology. Treatment with synovectomy is frequently curative.

Keywords: lipoma arborescens, knee, synovectomy.

Introduction

Lipoma arborescens (LA) is a rare, benign intra-articular lesion most commonly found in the knee, characterised by villous proliferation of the synovium [1]. It can be mono-, bi- or polyarticular and can affect patients of all ages (although it is commonest in the fifth decade and above) [1]. It often remains undiagnosed for a prolonged period as it mimics the more common arthritis, with secondary degeneration further clouding the picture clinically and radiographically. Synovectomy appears to be curative [2]. MRI is the investigation of choice, with images clearest on fat-supressed or STIR sequences

We present a rare case of bilateral lipoma arborescens and undertake a comprehensive review of the literature available.

Case Report

A 35 year old male patient presented with a three year history of bilateral knee pain and swelling. The pain and swelling were spontaneous in onset and there was no documented history of an associated injury. The pain and swelling had gradually worsened. There were no signs of mechanical obstruction in the knee. The patient had been investigated elsewhere, where the radiographs had been reported as normal and he had physiotherapy without any benefit.

On examination the patient was systematically well. He had bilateral swellings of the knee, with quadriceps wasting. He lacked the last five degrees of extension and flexed to one hundred and twenty degrees. There was no tenderness on palpation and he had an effusion and thickened synovium mainly in the suprapatellar region. The knee was stable.

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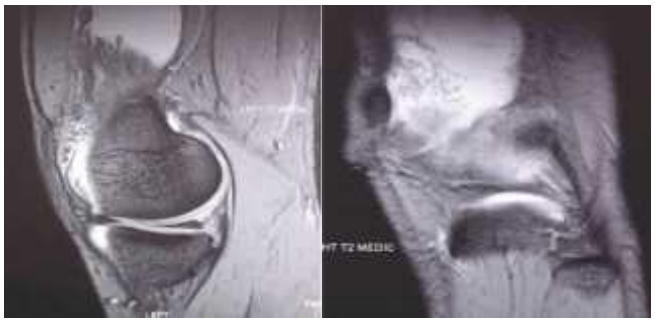

Figure 1, 2: MRI T2 weighted images showing multiple villous synovial proliferations in the spurapatellar areas

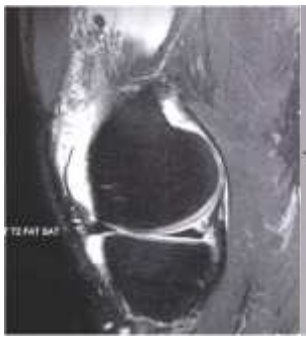

Figure 3: MRI T2 Fat saturated image

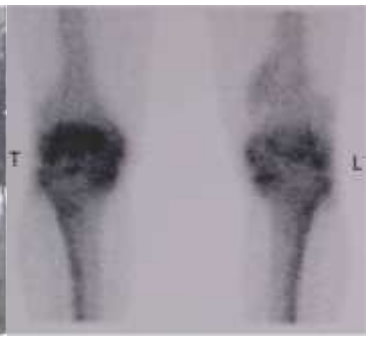

Figure 4: 99Technetium bone scan showing increased uptake in both knees

Plain radiographs were unremarkable. Magnetic resonance imaging (MRI) scans of his knee showed multiple villous lipomatous synovial proliferations and a 'frond-like' synovial proliferation of fat signal intensity (Figs. 1, 2 and 3). A 99technetium bone scan was done to exclude infection, which revealed increased uptake in both knees (Fig. 4). The patient underwent bilateral knee arthroscopic synovectomies (Fig. 5) and made an uneventful recovery. The samples sent for histology were reported as being characteristic of lipoma arborescens. Nine months following the arthroscopies the patient had no pain or swelling in his knees. He had regained a full range of movement and was asymptomatic.

Discussion

Medline and Embase were searched using the terms "lipoma arborescens", "villous lipomatous proliferation" and "arborescent lipoma". References from each paper were cross-referenced and the search was broadened using the "related articles" function. All abstracts were read by a single author (ADL) and relevant papers were retrieved. In total, 72 papers were retrieved, which contained reports of 152 patients. The papers were largely case reports with some small series. The earliest reports were both in 1952, the most recent paper is from December 2009.

Lipoma arborescens is undoubtedly rare, but the availability of MRI has led to a marked increase in the numbers of reported cases over recent years. The exact incidence is unclear, but Vilanova et al's review of 12,578 consecutive knee MRIs found 32 patients with LA [1] and Iovane et al found 9 out of 6387 [2]. This gives an incidence of between 0.14% and 0.25% of scanned knees; the incidence within the asymptomatic population will be much lower.

Most cases of LA have been described in single-case reports although there are a few larger series. In the 120 cases that we have identified, there was a slight male preponderance (67:53) and patients have a mean age of 45.6 years of age. The youngest case described was 9 years old [3](although the authors suggest that the patient underwent her first resection at the age of four) and the oldest was 90 [4].

Lipoma arborescens generally presents as a longstanding, slowly progressive swelling of one or more joints associated which may or may not be associated with pain [5]. A proportion of patients may present with symptoms of secondary degeneration such as crepitus, joint line tenderness and restriction of range of movement. Depending on the anatomical site of the disease, some patients may present with exacerbations secondary to interspersion of villi within

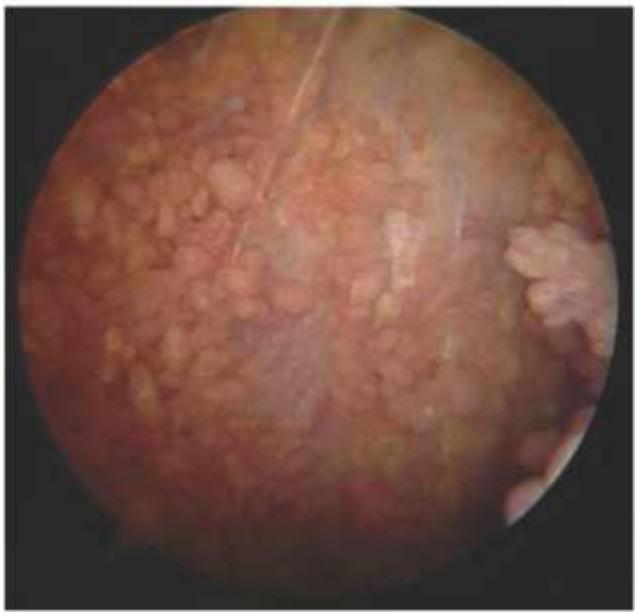

Figure 5: Arthroscopic image showing villous appereance

the joint space [6]; mechanical symptoms of locking and instability of the knee have been described by the same mechanism [6].

The most common anatomical site by far is the knee, and specifically the pre-patellar pouch, although cases have been described in many other synovial joints including the hip [8], shoulder [9], elbow [10], wrist [11] and ankle [12]. Bilateral involvement is uncommon, but when bilateral joints are involved they usually occur at the same time [13]. In very rare cases, LA has been reported to affect multiple joints, mimicking rheumatoid arthritis [14].

Examination of the knee reveals a boggy, supra-patellar swelling, occasionally with a palpable mass. Aspiration of the joint demonstrates clear, yellow synovial fluid devoid of crystals and cells on microscopy and sterile on culture, although the presence of a haemarthrosis does not exclude the diagnosis [15]. Signs of secondary osteoarthritis may be the dominant feature on examination. Typically, haematological investigations are normal with the exception of a mildly raised ESR. Aside from soft tissue shadows, signs of secondary degeneration are the only features on plain radiographs. MRI is the investigation of choice, with images clearest on fat-supressed or STIR sequences [16]. The characteristic appearances are of multiple villous lipomatous synovial proliferations and a 'frond-like' synovial proliferation of fat signal intensity [17]. These features may occur either separately or together, with a joint effusion present in all cases. Other associated findings are degenerative changes with mensical tears, synovial cysts, bony erosion and chondromatosis [1]. Cases have been described with abnormality or absence of the meniscus [18].

Other imaging modalities give varying results. Ultrasonography is accurate in determining the extent and location of the lesion in the various synovial surfaces of the knee and has the advantage of easy accessibility and low cost [19]. CT is fairly nonspecific, showing a degenerative picture with synovial swelling in affected joints [20]. Arthrography was used in the diagnosis of earlier cases but this has been superseded by newer modalities [21].

Arthroscopy reveals multiple globular and villous projections of synovial-covered tissue, restricted to the affected area of the joint. Again, the joint often shows signs of degeneration [22].

Microscopy of resected tissue reveals hypertrophic synovial villous proliferation with diffuse replacement of the subsynovial tissue with hyperplastic mature fat cells and an infiltrate of chronic inflammatory cells.

The aetiology of lipoma arborescens is unclear. It appears that in a subset of patients, there is an antecedent history of local joint trauma [23] or diabetes [24]; four cases have been described in the context of psoriatic arthritis [25-27]. However, in most reported cases there is no pre-existent pathology. Four cases have been described in the context of psoriatic arthritis. It has been postulated that morphologically distinct subtypes of LA exist in patients with and without a history of pre-existing inflammatory joint disease. One series of 12 patients found that previously normal joints demonstrated synovial fronds alone whilst the more typical villonodular picture were found in patients with a preceding history of joint disease [17].

At cellular level, adipocytes, osteoblasts, chondrocytes and myofibroblasts all have a common origin, and are believed to be derived from multipotent mesenchymal stem cells. It has been suggested that an inverse relationship exists between adipocyte differentiation and the osteogenic activity of bone marrow stromal cells, and that this is reciprocally regulated by bone morphogenetic proteins. From these observations, Ikushima et al [28] have put forward a hypothesis that LA is a rare form of a reactive lesion of the synovium in which the mesenchymal stem cells differentiate into adipocytes, whereas osteochondral differentiation of the mesenchymal stem cells results in synovial chondromatosis. They therefore suggested that LA and synovial chondromatosis might have a common aetiology.

The natural history of LA is poorly understood. LA appears to predispose to osteoarthritis although the cause for this is unknown. One theory put forward to explain osteoarthritis in LA suggests that chronic irritation of the synovium and underlying cartilage by the synovial fronds and long standing effusions leads to degenerative changes [28,29]. The severity of the osteoarthritic changes in the affected knees has been suggested to correlate with the duration of the symptoms [30].

LA can mimic nearly any intra-articular pathology, and

there are reports of cases being misdiagnosed as acute rheumatic fever [31] and rheumatoid arthritis [14]. The main differential diagnoses are true intra-articular lipoma (which is much rarer), Synovial chondromatosis or haemangioma, or pigmented villonodular synovitis (PVNS). The diagnosis is made on MRI [4].

PVNS, atypical synovitis of RA, and synovial chondromatosis can usually be differentiated by MRI, since these do not show fat lobules. PVNS masses contain haemosiderin and have a low signal intensity on T1 and T2 images. Signal intensity in RA masses is low on T1 and intermediate to high on T2 images and in synovial chondromatosis is low on T1 and high on T2 images. Chondromatous bodies may contain fat in the centre and have rim-like calcifications which have a low intensity on all sequences and are usually visible on plain radiographs. True intra-articular lipoma does not have the same frond-like appearance as LA, and usually occurs in the infrapatellar fat pad [20,27].

Synovectomy is the recommended treatment for LA and is usually curative, although recurrence following synovectomy has been reported [32]. Open synovectomy has been used in the treatment of this condition though arthroscopic synovectomy is now the treatment of choice. Synovectomy has been reported to result in complete and long standing alleviation of symptoms of LA in most patients but does not appear to halt the progression of secondary osteoarthritis [30].

Non-surgical alternatives to synovectomy appear to be successful, although there are very few reports of their use. Erselcan et al [33] successfully used yttrium-90-radiosynovectomy to treat one patient and chemical synovectomy with osmic acid has also been described with no recurrence of symptoms at one year [34].

Conclusion

In conclusion Lipoma arborescens is a rare, benign intra-articular tumour which may mimic a number of other diagnoses. In cases of unexplained chronic joint effusion, MRI should be considered to exclude this pathology as well as other uncommon intra-articular pathology. Treatment with synovectomy is frequently curative.

Clinical Message

Lipoma Arborescens is one of the differentials of knee pain and swelling and can present at varied age. Diagnosis is by MRI and synovectomy is most often curative

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