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CASE REPORT

MONOARTICULAR CHRONIC SYNOVITIS IN A CHILD

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SUMMARY

Lipoma arborescens is a villous lipomatous proliferation of the synovial membrane characterized by chronic and painless synovial effusion. The aetiology is unknown. It has to be included in the differential diagnosis of chronic monoarticular disease in childhood. Magnetic resonance imaging provides a highly efficient tool for the diagnosis of this very rare condition. This is indeed the fourth paediatric case reported. Rather than resorting to the often inconvenient surgical synovectomy commonly recommended, we chose to treat the knee of this 13-yr-old boy with intra-articular osmic acid.

Key words: Lipoma arborescens, Osmic acid, Juvenile monoarthritis, Knee magnetic resonance imaging.

CLINICAL HISTORY

DR is a 13-yr-old boy who was admitted to our hospital because of chronic pain and swelling of his left knee persisting over a few years. He is the younger of two children. The parents and sister are well. His past medical history was unremarkable, without any history of lower limb trauma, chronic fever or tuberculosis, tick bite or erythema migrans, psoriasis, inflammatory bowel disease, uveitis, low back pain or a family history of spondylarthropathy.

He began to complain of his left knee 4 yr before admission. The affected joint always remained swollen, but was not very painful. Non-steroidal anti-inflammatory drugs only provided mild pain relief. He was then referred to our hospital by his general practitioner for further evaluation.

On physical examination, the boy presented an isolated and large left knee effusion without redness or warmth. His rectal temperature was 37.6°C. There was no clinical overgrowth of the left lower limb, micrognathia or hepatosplenomegaly. The ophthalmological examination was normal.

Laboratory findings, including erythrocyte sedimentation rate, C-reactive protein, full blood count, differential immunoglobulin measurements and routine biochemistry, were normal. IgM rheumatoid factor (Rose–Waaler test) and antinuclear antibodies were absent, as well as IgM and IgG antibodies against Borrelia burgdorferi.

Needle aspiration of the knee yielded a moderately yellow, cloudy synovial fluid containing 6990 white blood cells/mm³ (neutrophils 9%; lymphocytes 86%; monocytes 5%). A mucin clotting test was not performed. Cultures of the synovial fluid remained negative for common pathogens and tuberculosis.

X-rays of the knees showed chronic synovitis changes of the left side, including overgrowth of the epiphyses and suprapatellar soft-tissue swelling (Fig. 1). Osteopenia, periosteal reaction and joint space narrowing were absent. Echography of the left knee disclosed a sonolucent effusion and the presence of hyperechoic villous proliferations suggestive of fat in the suprapatellar pouch of the left knee (Fig. 2). Magnetic resonance imaging (MRI) of the left knee was performed on a 1.0 T unit utilizing coronal, sagittal and transverse spin-echo imaging. Menisci, ligaments and cartilage surfaces appeared normal. There was a joint effusion associated with a frond-like villous appearance of the hypertrophic synovium, presenting signal characteristics isointense with the s.c. fat on all pulse sequences (Fig. 3).

An arthroscopic guided biopsy was performed: histology showed only a dense fibrous component of the subsynovial tissue and a moderate hyperplasia of the synovium. Because of the typical features of the MRI findings, lipoma arborescens (LA) was considered as the final diagnosis. Clinical survey excluded other lipomas, especially in the tendon sheaths of the hand, wrist, feet, ankle or on the anterior surface of the patella.

Surgical synovectomy is the most widely recommended treatment. However, we preferred chemical synovectomy with osmic acid which has an excellent therapeutic record, especially for various synovitides occurring in juveniles. After arthrocentesis of the knee, 10 ml of 1% lidocaine, 5 ml of osmic acid (2% solution, Merck, Darmstadt, Germany) and 1 ml of methylprednisolone acetate (40 mg/ml) were injected. Following injection, bed rest was observed for 24 h with a knee-resting splint. The procedure rapidly reduced pain and swelling in our patient.

After 1 yr, the patient remained asymptomatic with a normal examination and knee function.

DISCUSSION

Lipoma arborescens is a synovial disease characterized by marked villous proliferation of the synovial...
membrane and hyperplasia of the subsynovial fat [1], almost always occurring in adults. To the best of our knowledge, only three paediatric cases have been reported [2]. Typically, a slowly increasing, painless synovial thickening and effusion with intermittent exacerbations develop over a few years [1–9]. The knee joint is most commonly involved, with a predilection for the suprapatellar pouch [1–9]. However, cases have also been reported in the wrist, hip and shoulder [6]. Patients with LA usually have normal laboratory findings [1–9]. This condition has occasionally been associated with degenerative disease, chronic rheumatoid arthritis and diabetes mellitus [6, 8].

In childhood, the differential diagnosis of chronic monoarticular synovitis includes tuberculous or other infections (such as Lyme disease), haemophilia, pigmented villonodular synovitis, synovial chondromatosis and many of the chronic juvenile arthritides [2].

Radiological investigations on plain films rarely show erosions [6], but osteopenia, periosteal reaction and joint space narrowing have not been described. The other radiological techniques show the synovial thickening with villous proliferations [1–9]. These appear as non-specific filling defects on arthrography [4]. Their fatty nature can, however, be demonstrated by ultrasound, suggesting the diagnosis by a hyper-echoic appearance of the fronds [3, 8]; by computed tomography with fat attenuation of the fronds [5, 9] and by MRI with signal intensity of the villous proliferations similar to that of fat on all pulse sequences [2, 6–9]. Arthroscopy discloses an important hyperplasia of the synovial membrane sometimes presenting typical fatty-like globules [10].

Because of the fatty nature of the lesion, MRI seems to be the most sensitive and specific technique for the diagnosis, also allowing the exclusion of pigmented
villonodular synovitis, synovial chondromatosis and synovial haemangioma [2, 6–9].

Whereas a variable amount of adipose cells is present in the underlying connective tissue of the normal synovial membrane [11], LA is characterized by a complete replacement of this subsynovial tissue by a fibrous tissue and mature fat cells which extend into the proliferative villous projections. The overlying synovium may present aspecific changes with thickening and chronic inflammation [2–4, 8]. These findings are quite distinct from synovial fat necrosis associated with ischaemic pancreatic disease [12], from collections of lipid-laden macrophages sometimes associated with pigmented villonodular synovitis [13] and also from the histological features of intra-articular lipoma [14].

The aetiology of LA is unknown. As the disease has been occasionally associated with rheumatoid arthritis [15], one might hypothesize that it develops as some sort of inflammation-induced reaction. Trauma has also long been suspected to be a triggering agent of various types of synovitis [16]. Such traumatic or inflammatory events could not be found in this case, which we think arose de novo as in the vast majority of LA cases.

LA is such a rare condition that precise treatment guidelines are not available. Most authors recommend partial synovectomy, with recurrence reported only in one case [5]. We could not find any data about non-surgical treatments such as intra-articular steroid injections, or yttrium-90 and osmic acid synoviorthesis. We selected osmic acid synoviorthesis as the initial treatment because it has a long safety record in juveniles, as well as being effective in other conditions characterized by synovial membrane proliferation and thickening, such as rheumatoid arthritis [17] or pigmented villonodular synovitis [18]. Its efficacy was recently confirmed and quantitated by MRI [17]. Cartilage histopathological changes following intra-articular osmic acid injection were reported >20 yr ago [19].

The clinical consequences of the superficial cartilage alterations, however, remain unclear, but appear to be devoid of any harmful consequences. Arthroscopic synovectomy of the knee is effective, but entails more inconvenience and expense. Its indication is in patients who have failed to respond to osmic acid or radiation synovectomy [20].

**Fig. 3.**—(a) Parasagittal MR imaging of the left knee showing suprapatellar effusion and villous proliferations of the hypertrophic synovium presenting central high signal intensity on T1-weighted images (600/14) (curved arrows). (b) Parasagittal MR imaging of the left knee showing central decreased signal intensity of the villous proliferations surrounded by the enhanced overlying synovium on contrast-enhanced T1-weighted fat-suppressed MR images (540/14) (arrowheads). (c) Parasagittal MR imaging of the left knee showing intermediate signal intensity of the villous proliferations with chemical shift artefacts along the frequency encoded direction on proton-density images (2200/20) (curved arrows). These artefacts are represented by a dark edge at the interface between the fluid and the fat because of respective intrinsic variation in resonance frequency.
This fourth paediatric case of LA highlights the importance of considering the condition within the differential diagnosis of chronic juvenile arthritis. As in adults, MRI proved highly appropriate to establish the diagnosis. Chemical synovectomy (osmic acid) deserves further evaluation in this condition. At 1 yr follow-up, our young patient is doing well, with normal function and clinical examination of his knee.

REFERENCES