

Arthroscopically assisted knee contracture release secondary to melorheostosis: a case report

Raúl Torres Claramunt · Xavier Pelfort López ·
Enric Cáceres Palou · Joan C. Monllau García ·
Lluís Puig Verdie

Received: 20 July 2009 / Accepted: 19 April 2010
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Abstract Melorheostosis is a rare non-hereditary bone disease characterized by a radiographic pattern of flowing hyperostosis along the cortex with sclerotomal distribution. We report a case of a patient with severe knee contracture and a restricted range of motion caused by intraarticular bone fragment and hyperostotic bone lesions secondary to melorheostosis. An arthroscopically assisted approach was used successfully in order to remove free bone fragments and to release the hyperostotic lesions in the bone cortex of the distal femur.

Keywords Melorheostosis · Arthroscopic release ·
Knee contracture

Introduction

Melorheostosis is a rare progressive non-hereditary bone disease characterized by a radiographic pattern of flowing

hyperostosis along the cortex with sclerotomal distribution. Melorheostosis was described by Léri and Joanny [5] in 1922. A mutation in LEMD3 [4] gene has been related to this and other diseases included in the group of Sclerosing Bone Disorders [1]. It has also been proposed that the lesion arises from an abnormality of the sensory nerve of the affected sclerotome [7]. Although several bones (polyostotic) can be involved, it tends to affect one bone (monostotic).

Symptoms include pain, deformity, stiffness, restricted range of motion (ROM) and progressive limb deformity. They seem to be related to soft tissue contracture and nerve compression caused by the bone disorder.

Treatment is conservative in most of the cases, including drug management, hydrotherapy, physical and occupational therapy, etc.

Surgery should only be considered in cases in which joint function is seriously affected or pain cannot be controlled by conservative measures.

In these cases, sympathectomy, capsulotomies, hyperostotic bone removal, total arthroplasty or extraction of loose bodies are some of the surgical options.

As far as we know, only one case of melorheostosis involving the knee has been reported to be have been initially treated arthroscopically [8]. A case of a patient with severe knee stiffness secondary to melorheostosis affecting the left femur in which an arthroscopically assisted approach was used is reported.

Case report

A 59-year-old woman with pain and progressive stiffness in the left knee without improvement after 2 years of conservative treatment is presented.

R. T. Claramunt (✉) · X. P. López · E. C. Palou · L. P. Verdie
Hospital del Mar, Passeig Marítim 25-29,
08003 Barcelona, Spain
e-mail: RTorresClaramunt@parcdesalutmar.cat

X. P. López
e-mail: 92858@parcdesalutmar.cat

E. C. Palou
e-mail: 94734@parcdesalutmar.cat

L. P. Verdie
e-mail: 92825@parcdesalutmar.cat

J. C. M. García
Hospital de Sant Pau i de la Santa Creu,
Carrer Sant Antoni Maria Claret 167,
08025 Barcelona, Spain
e-mail: jmonllau@santpau.cat

There was no history of knee trauma or infection. Despite the conservative treatment, pain increased in the latter months and limited ROM to 0°/10°/60°.

Plain radiographs of the affected knee showed cortical hyperostosis in the entire left femur that particularly affected the knee joint (Figs. 1, 2). No other images suggesting generalized disease were found. Several loose bodies filled up both the anterior and the posterior knee compartments as well as the lateral gutter. These images looked like hardened wax that had dripped down the side of a candle. A CT scan and MRI study was made confirming the diagnosis. A scintigraphic study was reported as normal.

As all non-surgical treatments were ineffective and due to the knee pain and diminished ROM, an arthroscopically assisted approach was indicated.

The treatment was carried out in order to remove both free bone fragments and the hyperostotic lesions and to release the joint.

Five portals anteromedial, anterolateral, superomedial, posteromedial and posterolateral were used to gain access

to the whole joint. First, the fibrous tissue of the anterior compartment, intercondylar notch and medial and lateral gutters were released. Then, the bone fragments were removed in order to improve knee flexion. An enlarged anterolateral portal or mini-open approach was necessary to eliminate the largest bone fragments.

Afterwards, a posterior transeptal approach was performed using posteromedial and posterolateral portals. Close to the posterior cruciate ligament, a single large bone fragment that filled up the area extending from the posterior capsule to the posterior intercondylar notch was found (Fig. 3). This fragment was released through the anterolateral and posteromedial portals by using a bone burr and a radiofrequency device. Finally, a mini-open posterolateral approach was needed to remove it. The entire procedure was performed under tourniquet control.

An Outerbridge grade III–IV chondral lesion was also observed on the medial femoral condyle and was debrided with a shaver. No other associated injuries were found in the joint. The achieved ROM after finishing the procedure

Fig. 1 Preoperative radiological images showing ectopic bone formation in the knee joint as well as femoral cortical hyperostosis



Fig. 2 Preoperative MRI images

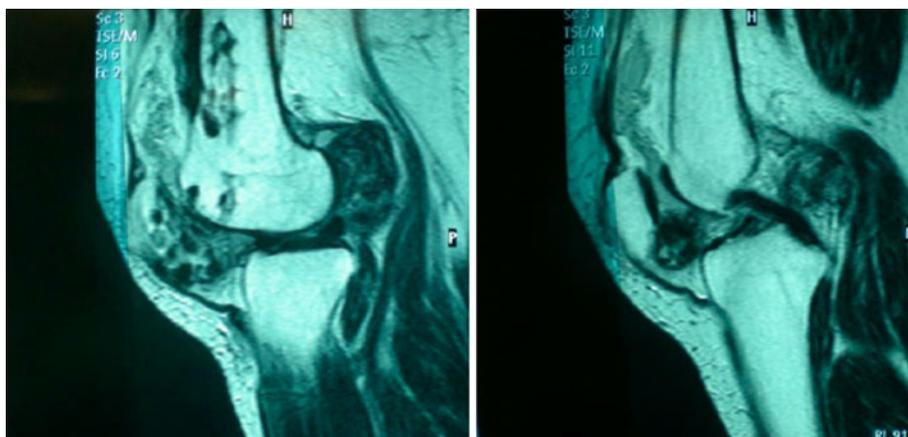
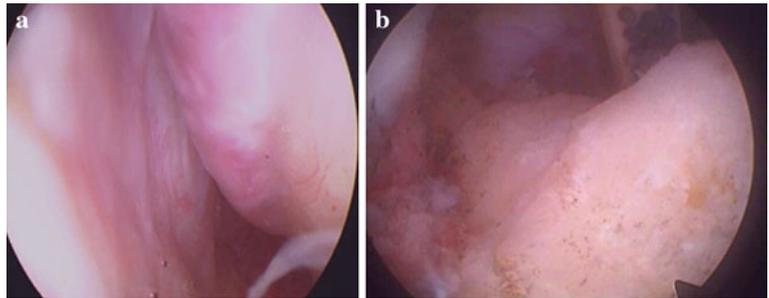


Fig. 3 **a** Arthroscopic view from the posteromedial portal. The bone fragment adhered to the medial condyle and deforming the posterior capsule can be seen. **b** Arthroscopic view from the posterolateral portal. A radiofrequency device was used to release the bone fragment from the posteromedial portal



was 0°/0°/130°. The postoperative radiological and CT images confirmed success of the surgery. Pathology confirmed the diagnosis of melorheostosis.

A rehabilitation programme was initiated 1 day after surgery. Progressive weight-bearing and continuous passive motion was immediately encouraged. Three weeks after surgery, the patient was able to walk with total weight bearing and ROM was 0°/0°/110°.

At the last follow-up examination, carried out at 15 months, ROM was 0°/5°/90°, while tolerable pain in the medial compartment was reported by the patient.

Discussion

The arthroscopic technique is a well-accepted method for loose body removal and soft tissue joint release. The most important finding of the present study is that an arthroscopically assisted technique can be also useful in the management of this rare condition.

Melorheostosis seriously affects soft tissues and bones of the lower extremities and progresses slowly. Cortical lesions are progressive and may extend into nearby joints causing loss of motion. Freyschmidt et al. [2] divided melorheostosis into five groups, based on their radiological patterns.

They are classified as osteoma like, classic, myositis ossificans like, osteopathia striata like and mixed. Accordingly, the present case would be classified as a mixed type (myositis ossificans like and classic).

Most cases of melorheostosis are benign and do not require surgery [9].

However, tendon lengthening, correction of bone deformities or joint arthroplasties are sometimes necessary. In the present case, conservative management was undertaken over a long period of time without improvement. Due to the patient's age and hopes, an arthroscopic technique was elected.

Few cases of surgical release of melorheostosis have been reported. Gong et al. [3] reported a case of melorheostosis in which the patient had a 90° fixed elbow contracture. An anterior and posterior compartment soft tissues open surgical release to permit complete ROM was successfully carried out. Moulder et al. [6] reported one

case of knee affection that was effectively treated by TKR. As far as we know, only Rooney et al. [8] have used arthroscopy to treat this condition.

In the present case, an arthroscopic arthrolysis including bone removal was used. Intensive postoperative rehabilitation made it possible for the patient to achieve good ROM at 3 weeks postoperatively. However, ROM progressively diminished over the following months leading to stabilization by the 6th month. At the last follow-up, this ROM was the same. The soft tissue contracture characteristic of the disease might have been the main reason for ROM diminishment and the knee pain that the patient complained of at the last visit.

The ease of use and low morbidity of the arthroscopic approach led, in this particular case, to a reasonable level of pain and ROM in a short period of time without compromising the future of the joint.

Therefore, arthroscopy has been shown to be effective at freeing a joint, at least temporarily, in this progressive disabling disorder.

References

1. de Vernejoul MC (2008) Sclerosing bone disorders. *Best Pract Res Clin Rheumatol* 22:71–83
2. Freyschmidt J (2001) Melorheostosis: a review of 23 cases. *Eur Radiol* 11:474–479
3. Gong HS, Lee KH, Oh JH, Chung JH, Baek GH, Chung MS (2008) Successful elbow contracture release secondary to melorheostosis. A case report. *J Bone Joint Surg Am* 90:1106–1111
4. Hellems J, Preobrazhenska O, Willaert A et al (2004) Loss-of-function mutations in LEMD3 result in osteopoikilosis, Buschke-Ollendorff syndrome and melorheostosis. *Nat Genet* 36:1213–1218
5. Léri A, Joanny J (1922) Une affection non décrite des os: hyperostose “en coulee” sur toute la longueur d’un membre ou “mélorhéostose”. *Bull Mem Soc Hosp Paris* 46:141–145
6. Moulder E, Marsh C (2006) Soft tissue knee contracture of the knee due to melorheostosis, treated by total knee arthroplasty. *Knee* 13:395–396
7. Murray RO, Mc Credie J (1979) Melorheostosis and the sclerotoses: a radiological correlation. *Skeletal Radiol* 4:57–71
8. Rooney RC, Fernicola PJ, Pitcher JD (1996) Arthroscopic excision of intra-articular ossifications. A case report and review of the literature. *Am J Orthop* 25:437–440
9. Wiater JM, King JC, Louis DC, Smith JS (1998) Melorheostosis of hand: a comprehensive review. *Hand Surgery* 3(1):123–130