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Severe Pain Resulting from the Impingement between Patella and Tibia Secondary to Melorheostosis: A Case Report

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Abstract

Melorheostosis is a rare sclerosing bone dysplasia. We report a unique case of melorheostosis with involvements of femur, tibia, and patella, resulting in pain, limited range of motion and impingement between patella and tibia. An arthroscopic assisted open surgery approach was performed to debride the damaged cartilage, soft tissue, and removal of the lesion on patella. This procedure led to an improvement of knee function and remission of pain.

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Introduction

Melorheostosis is a rare sclerosing bone dysplasia which was first described in 1922 by Leti and Joanny [7]. As opposed to bone malignancies, this disease is relatively benign and has a character of non-hereditary. Melorheostosis most likely occurs in the appendicular skeleton as compared with the axial skeleton. Pain, limited range of motion, limb deformity and stiffness are the major symptoms of this disease, however, most of the patients are asymptomatic. As the disease progresses, the lesion can lead to a compression or impingement of the adjacent nerve, cartilage, and other soft tissues. The diagnosis of melorheostosis is characterized by the radiological features similar to that of a "Melting wax dripping down a candle" [1].

We report a unique case of melorheostosis with involvement of femur, tibia, and patella, resulting in pain, limited range of motion and impingement between patella and tibia. A combination of surgery with the assistance of arthroscopy was chosen as an ideal treatment option.

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Case report

A 40-year-old female was admitted to our hospital with a complaint of progressive left knee pain and with difficulty in walking. The patient reported that the pain started 6 years ago and was progressive in nature, worsening over the last 2 years. The patient reported extreme pain particularly on extension and flexion of the left knee joint. The pain was localized in the medial-inferior part of the patella. She also reported that the local skin temperature over the left knee joint was higher than the right side following a long walk.

On physical examination, the patient walked with a shuffling gait, requiring the assistance of a walking aid. The patient was not able to fully extend the left knee joint with a contracture of 5 to 7 degree.

Plain radiographic image of the left knee showed cortical hyperostosis in the periosteal of the femur and the endosteal of the tibia. The patella showed a nodular hyperostosis on the medial-inferior part. There are several focal increased signal intensities around the knee joint. The medulla canal of femur and tibia showed significant narrowing due to the melorheostosis compared with the right side (Figure 1a,b).

Magnetic resonance imaging was conducted to check the condition of cartilage and confirm the character of the lesion near the knee joint (Figure 2). Computed tomography and three-dimension reconstruction were made to evaluate the morphology and the nature of the lesions on the femur, tibia, and patella (Figure 3). Gammagraphy was conducted to exclude the lesions from other metastatic disease (Figure 4). The results of these radiological examinations confirm the diagnosis of melorheostosis.

As the conservative treatments such as analgesics, rehabilitation, and bisphosphonate failed to relieve pain and improve the range of motion, an arthroscopic assisted open surgery approach was performed.

Anteromedial and anterolateral portals were used to gain access to the knee joint. The hyperplastic synovium of the joint compartment was debrided with the shaver. The damaged cartilage was debrided and shaped with shaver and plasma knife. (Figure 5a,b)The impingement between the bony lesion on patella and tibia occurred when the knee joint was fully extended, but the lesion on the patella and tibia were hard to remove during the arthroscopic procedure without open surgery. A mini-open approach was therefore made to expose and remove the lesions on the patella and tibia. The surgery was performed with the help of tourniquet that lasted an hour. The sample was sent to the Pathology Department of our hospital for analysis. The sample was a fragment of 10*12*4 mm in size, separated from the patella and was extremely hard in consistency.

The range of motion was at 0° on extension and at 140° on flexion after the surgery. The postoperative radiological images confirmed the success of the surgery, the report from pathology result confirmed the diagnosis of melorheostosis.

After three months, the patient showed a normal range of motion with no pain.

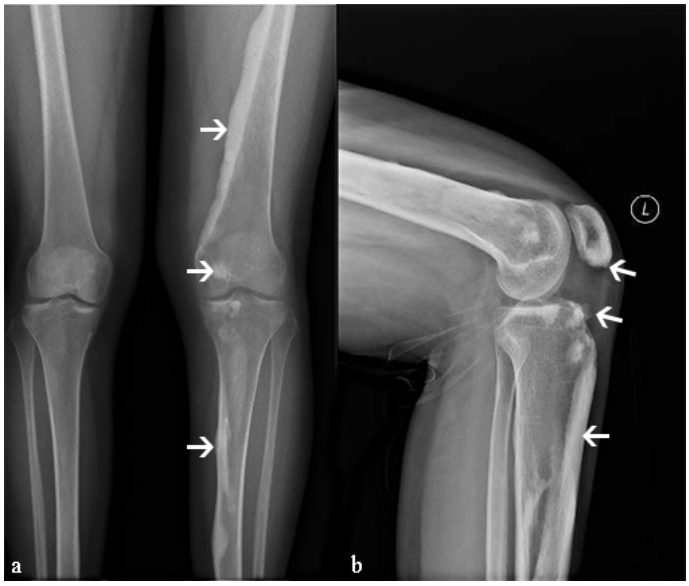

Figure 1: Preoperative x-ray image of the left leg showing characteristic "Dripping candle wax" sign on the medial aspect of the femur from mid-shaft to distal femur and a nodular hyperostosis on the medial-inferior part of the patella.

Figure 2: MR imaging scan of the left knee.
(2a) Sagittal T2 slightly flexed image showing signs of calcifications and edema around the knee joint.
(2b) Sagittal T1 image shows several focal hypodense lesions of irregular boarders.
(2c) Coronal T2-weighted image shows the characteristic "dripping candle wax" sign as an area of hypodense region on the medial aspect of the left femur.
(2d) Axial T2 shows an area of hyperdense region on the anterior side of proximal tibia due to impingement.

Figure 3: Represent a three-dimension reconstruction CT image of the left knee joint showing the morphology of the melorheostosis lesions.

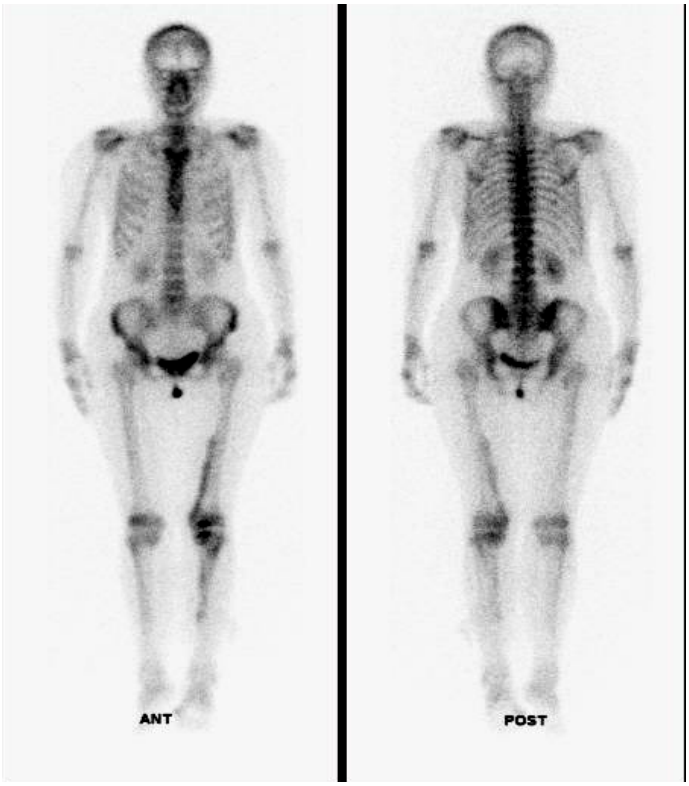

Figure 4: Gammagraphy showing the abnormal distribution of radioisotope on the femur, tibia, and patella with an increased uptake on the site of pathology.

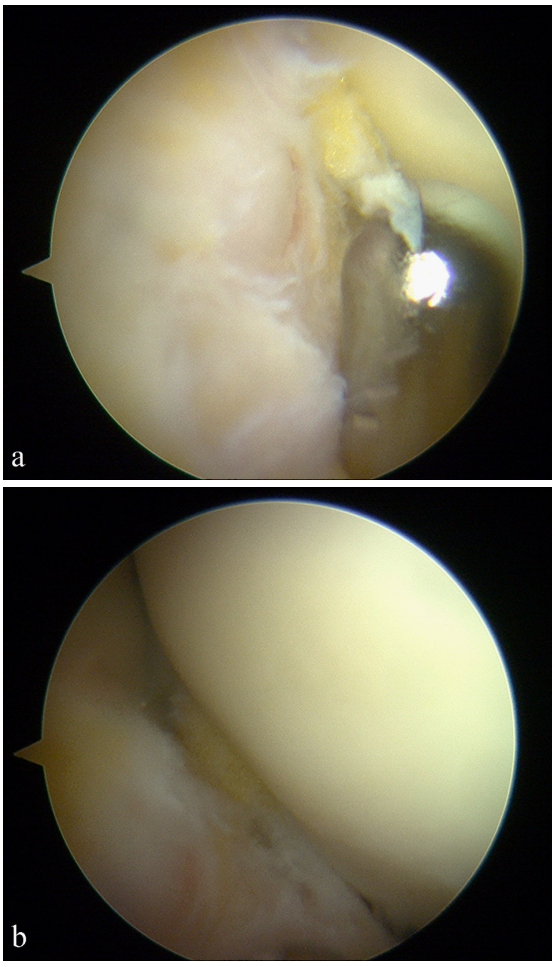

Figure 5: An intraoperative image of arthroscopic view from the anterolateral portal.
(5a) shows the bony lesion on the patella and the damaged synovium in the knee joint.
(5b) shows the impingement between bony lesion on patella and tibia.

Discussion

The reason we presented this case is because of the rarity of this disease and the novelty of the methods of treatment. The prevalence of this disease was reported about 1 per million between 1950 and 1979 [2]. The technique of arthroscopic surgery was selected to debride the damaged cartilage and soft tissues in the management of this case. The outcome of this patient shows an open surgery with the assistance of arthroscopy can be a useful way to manage this rare disease occurring in the knee joint.

Melorheostosis is a benign disease with a slow progression. It can appear at any age, but it usually presents in childhood or adolescence [3]. The presentation of this disorder including pain in the bone or joint, limb deformity, joint stiffness, contracture, and limited range of motion. However, pain and stiffness are always the major symptoms.

The etiology of melorheostosis still remains obscure. The mutation in LEM domain-containing protein 3 (LEMD 3) gene was initially regarded as the cause of melorheostosis, but further studies confirmed the mutation of LEMD 3 gene is also related to osteopoikilosis and Buschke-Ollendorff syndrome [4].

The melorheostosis is radiologically diagnosed. The classical radiological feature of melorheostosis includes the irregular, eccentric hyperostosis of the periosteal or endosteal surfaces of the cortex of the bone. It was first described as “Melting wax

flowing down the side of a candle." The osseous of melorheostosis can be monostotic or polyostotic, and the distribution to unilateral limb is more common than the distribution to bilateral limb which is quite rare [4]. The lower limbs are mostly affected than the upper limbs while the axial skeleton and the skull are rarely affected. As far as we know, there is no case that reported the melorheostosis in the patella, and cortical hyperostosis in the periosteal of the femur and the endosteal of the tibia until now.

Most patients with melorheostosis do not require any surgical intervention. Conservative treatment is the first choice for majority of the patients. It includes the use of analgesics, rehabilitation and rest. Bisphosphonate also can be a choice of pain control. Surgical intervention is indicated for some patients with the symptoms of neural compression, bone impingement, or pain that failed to respond to conservative treatment. Due to the symptoms and the hopes of this patient, an arthroscopic assisted open surgery approach was selected.

Few cases of arthroscopic treatment of melorheostosis have been reported. Claramunt et al [5]. reported a case of melorheostosis with the treatment of arthroscopy. Five portal and a mini-open incision was conducted to evacuate the bone fragment in the knee joint. Minarro et al. [6] used arthroscopy to treat the patient with melorheostosis lesion in the intercondylar notch and Hoffa fat. As far as we know, only this two cases adopted the arthroscopic technique to treat patients with knee lesion due to melorheostosis.

In this case, an arthroscopic assisted open surgery approach was carried out to remove the bone calcification on the patella and tibia. The damaged synovium and cartilage were debrided with the help of shaver and plasma knife. The factor that results in the impingement between the patella and tibia was eliminated. The patient achieved a good range of motion and feels no pain at three months after surgery.

Therefore, an arthroscopic assisted open surgery approach was proposed to the patient with melorheostosis involving the knee joint because of it's less invasive nature and conveniency in dealing with the damages of the deep joint. The combination of arthroscopy and open surgery significantly reduces the duration of surgery as compared with arthroscopic surgery alone. Patients would get a better early function of the knee joint and suffers less postoperative pain due to shorter tourniquet time.

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