



Case report

Severe valgus deformity of the knee with permanent patellar dislocation associated with melorheostosis: A case report and review of the literature



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ABSTRACT

We present a case of an 8-year-old boy diagnosed with melorheostosis who was suffering from severe genu valgum, permanent dislocation of the patella, knee flexion contracture and leg length shortening. Soft tissue contracture of the limb and subsequent joint deformities were reported to represent clinical manifestations of pediatric melorheostosis. As the epiphyseal plate had not closed, patellar reduction was achieved by soft tissue surgical stabilization, including lateral retinacular release, medial retinaculum plication, and transfer of the lateral half of the patellar tendon. At 4 years postoperatively, as a result of improved limb alignment and knee flexion contracture, the leg length shortening has improved, and the patient does not limp and participates in sports activities. Surgical intervention should be performed as early as possible, because genu valgum and external rotation of the tibia may deteriorate with age, rendering the patellar dislocation irreversible in patients with melorheostosis before epiphyseal closure.

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1. Introduction

Melorheostosis is a rare disorder with characteristic hyperostosis in a sclerotomal distribution [1,2]. The clinical features of melorheostosis differ between children and adults. Restricted range of motion, limb shortening, and soft tissue contracture of the limb and subsequent joint deformities represent clinical manifestations of melorheostosis before epiphyseal closure, which is the case with childhood onset. Initial symptoms often comprise joint pain and gait disturbance [3]. On the other hand, extraperiosteal ectopic ossification is frequently seen in adult-onset cases, and not only joint contractures and deformities, but also osteosclerotic foci themselves can form a cause of pain [4]. We present the case of an 8-year-old boy diagnosed with melorheostosis who was suffering from permanent dislocation of the patella, severe genu valgum, and flexion contracture. As the epiphyseal plate had not closed, patellar reduction was successfully achieved by soft tissue surgical stabilization, including lateral retinacular release, medial retinaculum plication, and transfer of the lateral half of the patellar ligament.

2. Case report

When the patient was 2 years old, firm thickening of the skin with tethering of underlying fascia was noted in the lateral thigh,

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but no definitive diagnosis was reached. From 4 years old, valgus deformity of the left knee gradually progressed, and he displayed a left knee pain and a limp at 6 years old. By 8 years old, valgus deformity had progressed further and extension of the left knee was restricted, exacerbating limping gait with severe pain. He was therefore referred to our hospital for specialist treatment. On examination, severe valgus deformity of the left knee was seen and the range of motion was -30° for extension and 135° for flexion. The patella showed lateral deviation throughout the range of motion (Fig. 1). No swelling or joint effusion was evident. Based on the spino-malleolus distance, leg length inequality was 5.5 cm. Limping gait was attributed to the leg length inequality and restricted knee extension. Kujala score was 52. The plain radiography revealed severe genu valgum, and the patella was completely dislocated laterally. Intracortical hyperostosis appeared as streakiness along the femur and tibia (Fig. 2). The femoro-tibial angle (FTA) had decreased from 175° to 158° over the previous 2 years. The femur was apparently in contact with the tibia only in the lateral compartment. On magnetic resonance imaging (MRI), the tibia was externally rotated about 40° relative to the femur. Fibrous bands with low signal intensity on T1-weighted imaging in the axial plane extended from the lateral femoral intermuscular septum to the iliotibial tract (Fig. 3).

According to the characteristic osteosclerosis in one leg on plain radiography, melorheostosis and associated contracture of the knee were diagnosed. Because of the severe gait disturbance due to permanent patellar dislocation, restriction of knee extension, and genu valgum, surgical treatment was carried out to restore these deformities.



Fig. 1. Macroscopic findings of the left knee at 8 years old, showing severe genu valgum and the patella palpable lateral to the femoral condyle. The tibia is externally rotated compared to the femur.

2.1. Operative findings

As the epiphyseal plate was not closed, soft tissue release and stabilization were applied as the main strategies to reduce the patella and maintain the proper position. The thickened iliotibial band was cut in a Z fashion (Fig. 4A). The vastus lateralis, which had deviated posterolaterally as a result of external rotation of the tibia, was dissected from the intermuscular septum and moved anteriorly. No ectopic ossification was identified in the soft tissue. In accordance with the method described by Fujikawa et al. [5], the lateral half of the patellar ligament was dissected from its patellar attachment (Fig. 4B), and rectus snip was performed to lengthen and mobilize the extensor mechanism, which allowed deep knee flexion after the reduction of patella. The rectus snip involved oblique cut across the proximal portion of the rectus femoris tendon. To maintain a reduced position, medial plication was performed, involving dissection of the medial retinaculum and vastus medialis obliquus (VMO) and then reattachment more

medially onto the patella (Fig. 4C). VMO has been widely applied as a major dynamic stabilizer of the patella. Overall, the order of procedures performed was: 1) cutting of the thickened iliotibial band and lateral retinaculum; 2) cutting the joint capsule; 3) dissecting the lateral half of the patellar ligament; 4) rectus snip; and 5) anteromedial transfer of the medial retinaculum and VMO over the patellar surface.

2.2. Postoperative course

The knee was immobilized at 30° flexion in a plaster cast for 2 weeks after surgery. Thereafter, the range-of-motion training and walking with partial weight-bearing within hinged dynamic knee brace were started. At 4 weeks postoperatively, full weight-bearing was permitted. As of 5 years postoperatively, the range of motion has improved to 0° extension and 140° flexion, and Japanese-style sitting has been allowed as tolerated. Radiography showed that the patella positioned in the center of the femoral groove, and the FTA was 175° (Fig. 5A). In addition,

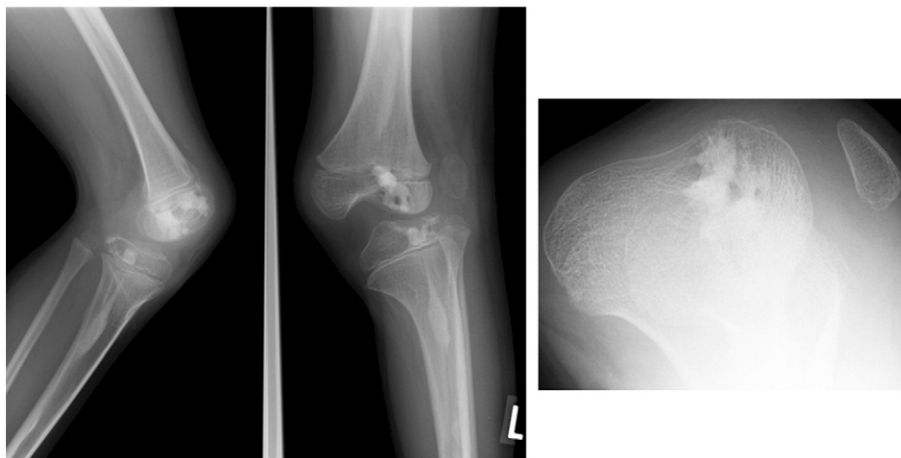


Fig. 2. Radiographs of the knee at 8 years old. Patellar dislocation and osteosclerosis of the femur and tibia are evident.

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