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Case Report

Rapidly acquired valgus deformity of the knee after osteochondroma resection in multiple hereditary exostoses pediatric patients: A report of two cases ☆

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ABSTRACT

Patients with multiple hereditary exostoses (MHE) often develop leg length discrepancies and limb alignment deformity around the knee as part of the natural course of the disease. Limb alignment deformity occurring post-resection of an osteochondroma has been described in one case report and only pertaining to the proximal medial tibia location. Here we describe the case of 2 patients with MHE, a 7-year-old female who underwent resection of distal femur and proximal tibia osteochondromas and a 9-year-old female who had a distal femur osteochondroma resected. Both patients developed rapidly progressive valgus knee deformity requiring surgical intervention. Excision of osteochondromas near the physis of a skeletally immature patient can cause overgrowth from the involved side of the growth plate resulting in a rapidly progressing unilateral coronal plane deformity. Surgeons should be aware of this potential complication and closely follow growing patients with serial alignment radiographs and counsel the family regarding the potential of acquired limb deformity and subsequent surgeries.

Introduction

Multiple hereditary exostosis (MHE) is a rare disease with an estimated prevalence of 1 per 50,000 people [1]. It is characterized by the presence of several benign osteochondromas, typically located on the pelvis or metaphysis of the long bones, and most frequently found around the knee [2,3]. The osteochondromas usually develop in the first 10 years of life and grow slowly in mass until skeletal maturity [4]. Patients with MHE often develop leg length discrepancies and limb alignment deformity around the knee as part of the natural course of the disease [5]. Pathologic genu valgum occurs in a third of MHE patients and slowly develops over time, usually during

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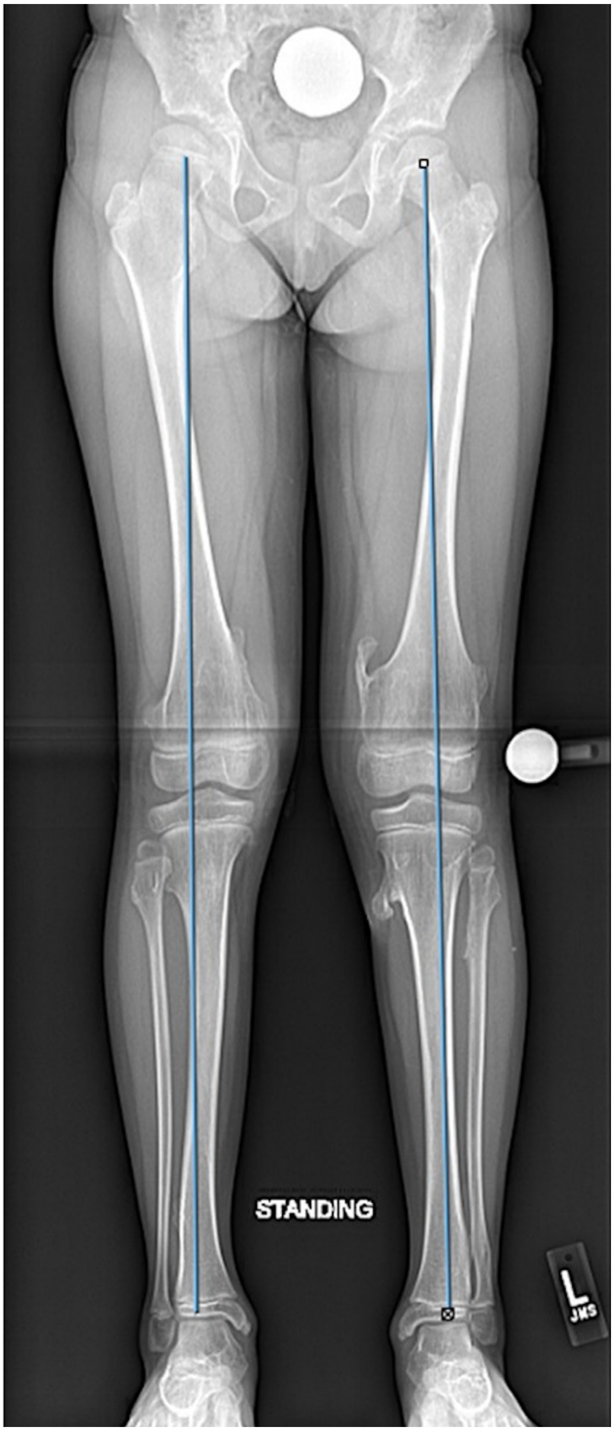

Fig. 1 – Case 1: Preoperative standing hip-to-ankle radiograph displaying normal limb alignment and large exostoses on the left medial distal femur and proximal tibia. Left leg mechanical lateral distal femoral angle (mLDFA) measured 81º and mechanical medial proximal tibial angle (mMPTA) measured 90º.

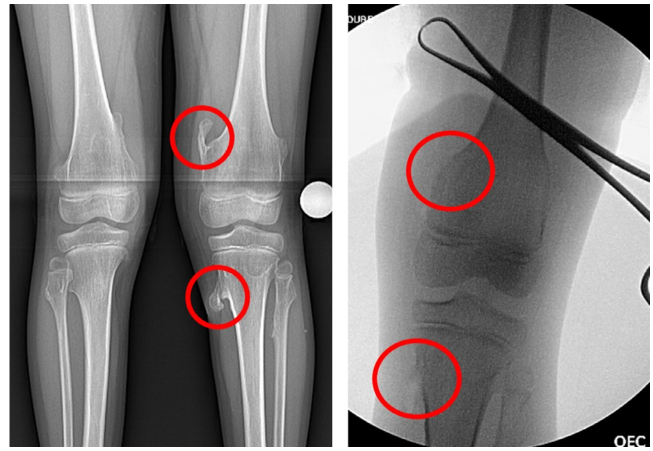

Fig. 2 – Case 1: Red circles indicate the left limb medial distal femoral and proximal tibial osteochondromas prior to excision (left) and intraoperatively after excision (right) (Color version of the figure is available online.)

the peak growth ages from 10 to 14 years [6,7]. Symptomatic osteochondromas can be treated by surgical excision with a low rate of complications [8]. However, Denduluri et al. reported on 3 patients that experienced a rapidly progressive tibial valgus deformity within the first 2 years after excision of medial proximal tibial osetochondromas [9].

We aim to add and expand upon this literature by reporting on 2 patients with MHE, one who underwent excision of medial distal femoral osteochondroma in isolation and one with concomitant excision of a medial proximal tibial osteochondroma and a medial distal femur osteochondroma. Both patients developed a rapidly progressing valgus deformity on their operative leg and were successfully treated with subsequent hemiepiphysiodesis.

Case presentation

Case 1

A 7-year-old female presented for MHE follow-up with bilateral pain over the medial distal femur, left worse than right, that occurred after activity. On clinical exam, her gait slightly favored her left leg. She had palpable lesions on both the medial distal femora and medial proximal tibiae. The lesions on the right leg were smaller and did not appear to be symptomatic on exam. Radiographs confirmed bilateral bony lesions on medial distal femora and proximal tibiae with normal limb alignment (Fig. 1). The patient was taken to the operating room for removal of symptomatic left medial distal femoral and proximal tibial osteochondromas. Careful attention was made to completely remove the cartilaginous cap of both osteochondromas. The successful resection was confirmed with intraoperative fluoroscopic images (Fig. 2).

The patient's surgical recovery was uneventful; she regained full range of motion and resumed her activities. However, on 2-month postoperative radiographs she was noted to

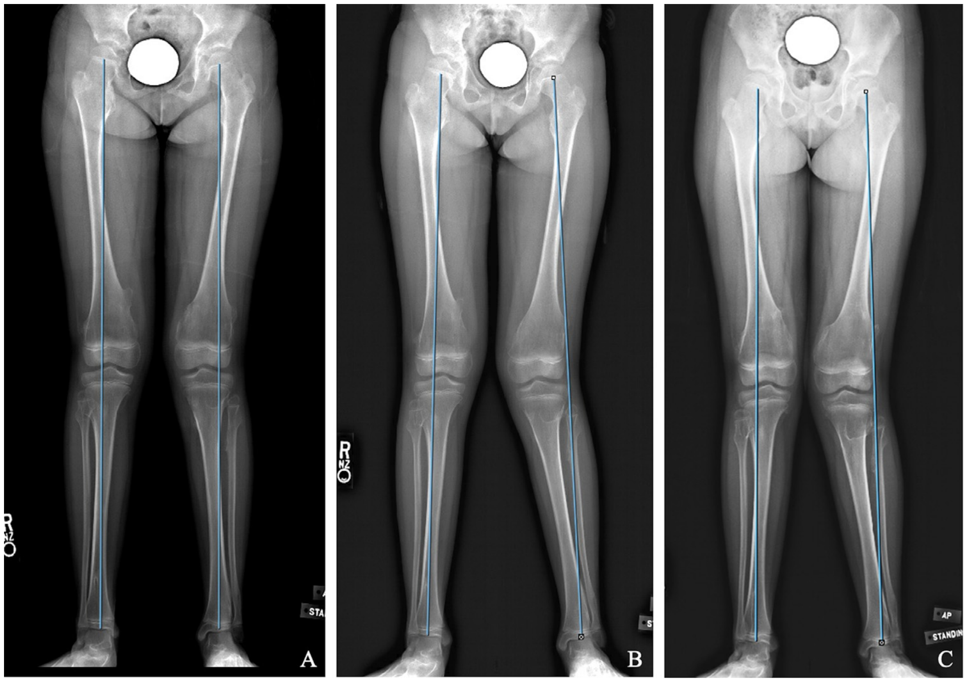

Fig. 3 – Case 1: Standing hip-to-ankle radiographs showing progressive left valgus deformity 2 months (A), 8 months (B) and 24 months (C) post medial distal femoral and proximal tibial osteochondroma excision. Left leg mLDFA measured 85, 81, and 82º respectively and mMPTA measured 91, 92, and 94° respectively.

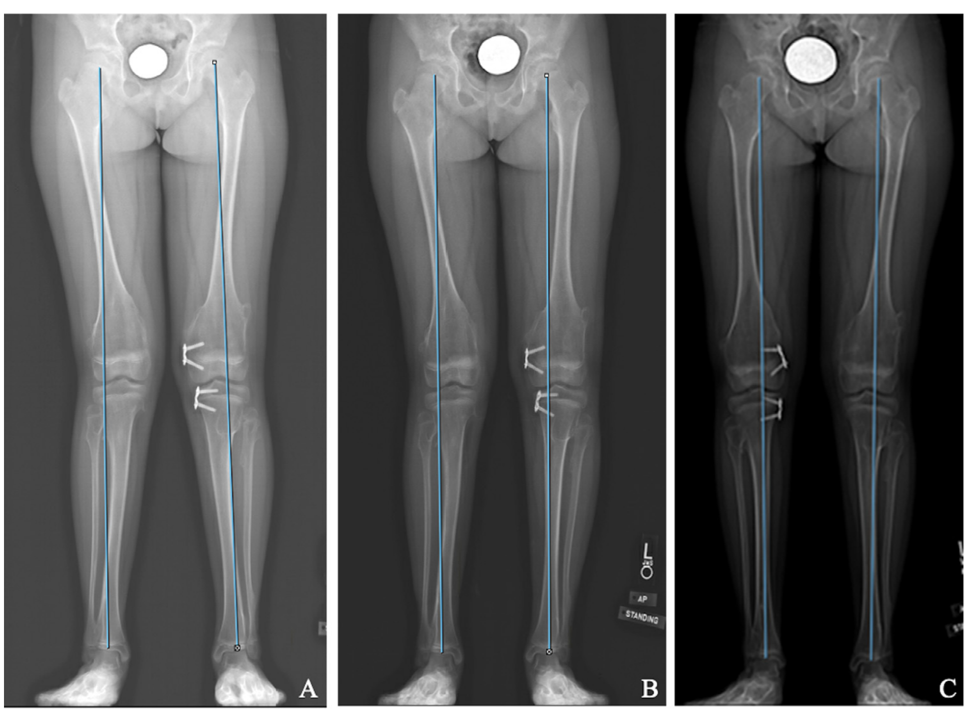

Fig. 4 – Case 1: Standing hip-to-ankle radiographs showing progressive correction of left valgus deformity 8 months (A) and 12 months (B) after implant mediated guided growth procedure. (C) Standing hip-to-ankle radiographs showing satisfactory left limb alignment 5 months post removal of implant mediated guided growth hardware on the left limb. Left leg mLDFA measured 88, 93, and 88º respectively and mMPTA measured 91, 86, and 90º respectively.

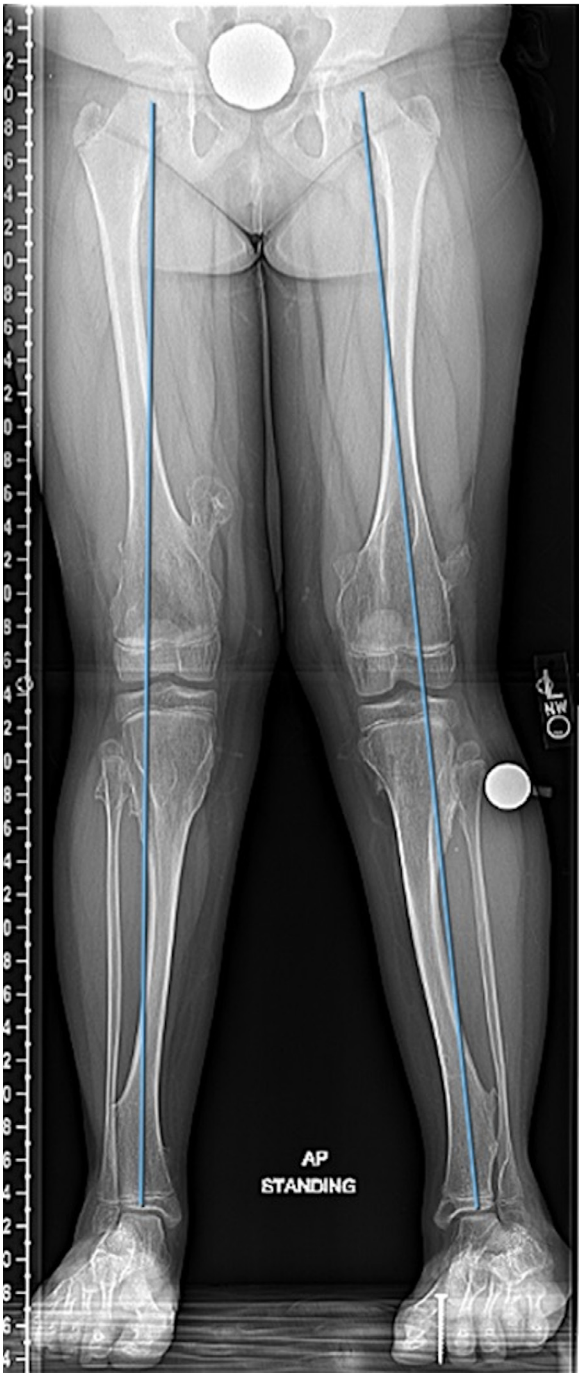

Fig. 5 – Case 2: Preoperative standing hip-to-ankle radiograph displaying left leg with valgus alignment, and right leg with normal alignment and a large exostosis on the right medial distal femur. Right leg mLDFA measured 89º and mMPTA measured 95º.

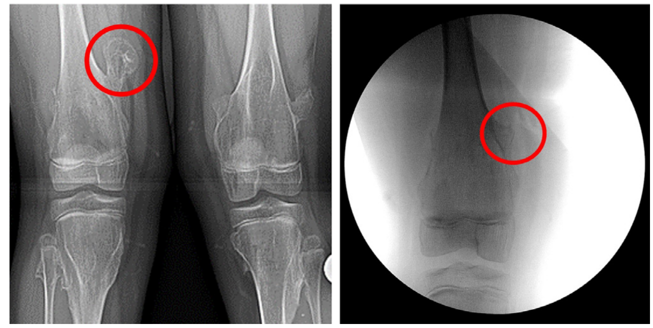

Fig. 6 – Case 2: Red circles indicate the right medial distal femoral osteochondroma prior to excision (A) and intraoperatively after excision (B) (Color version of the figure is available online.)

have increased valgus alignment of her left distal femur which had further progressed on radiographs taken 8 months postoperatively (Fig. 3A and B). A guided growth procedure was recommended at the time but the family refused. The patient returned 2 years postoperatively with progressive valgus deformity of both the left femur and tibia leading her to trip while running (Fig. 3C). At this point, the family consented to surgical correction, and patient underwent hemiepiphysiodesis of the left distal femur and proximal tibia with 8 plates.

The patient showed improved alignment 8 months after the growth modulation procedure (Fig. 4A). At 12 months postoperatively, radiographs showed slight over correction into varus alignment (Fig. 4B) at which time the patient had both the left femoral and tibial plates removed. The 8-plates were in place for a total of 11.7 months. At latest follow-up 5-months post hardware removal, the patient was back to full activity, and radiographs revealed good alignment of the left extremity (Fig. 4C). Of note, in the interim the patient underwent hemiepiphysiodesis on the right limb to correct a gradually progressive valgus deformity that is commonly seen in the setting of MHE.

Case 2

A 9-year-old female with a known diagnosis of MHE presented for a slight "knock kneed" appearance of the left limb. At that time, the presence of a large right medial distal femoral lesion was also noted. Radiographs revealed valgus alignment of the left limb and confirmed a large distal femoral osteochondroma on the right side, among additional exostoses (Fig. 5). The patient was indicated for surgery and underwent concomitant excision of the right distal femoral osteochondroma and a hemiepiphysiodesis of the medial proximal tibia and medial distal femur on the left side to correct the valgus deformity. Intraoperative fluoroscopy was used to confirm the complete excision of the osteochondroma cartilaginous cap of the medial distal right femur (Fig. 6).

Similar to the first case, the patient's immediate postoperative course was uneventful, but she was noted to have slight increased valgus alignment of her right distal femur at her 2-month postoperative visit (Fig. 7A). The deformity of the right limb rapidly progressed by the 4-month postoperative visit (Fig. 7B). The patient thus underwent hemiepiphysiodesis of the right distal femur and proximal tibia with 8 plates to correct her growth deformity.

Radiographs showed improved valgus deformity 4-months after the guided growth procedure, and 6-month postoperative radiographs revealed correction into slight varus

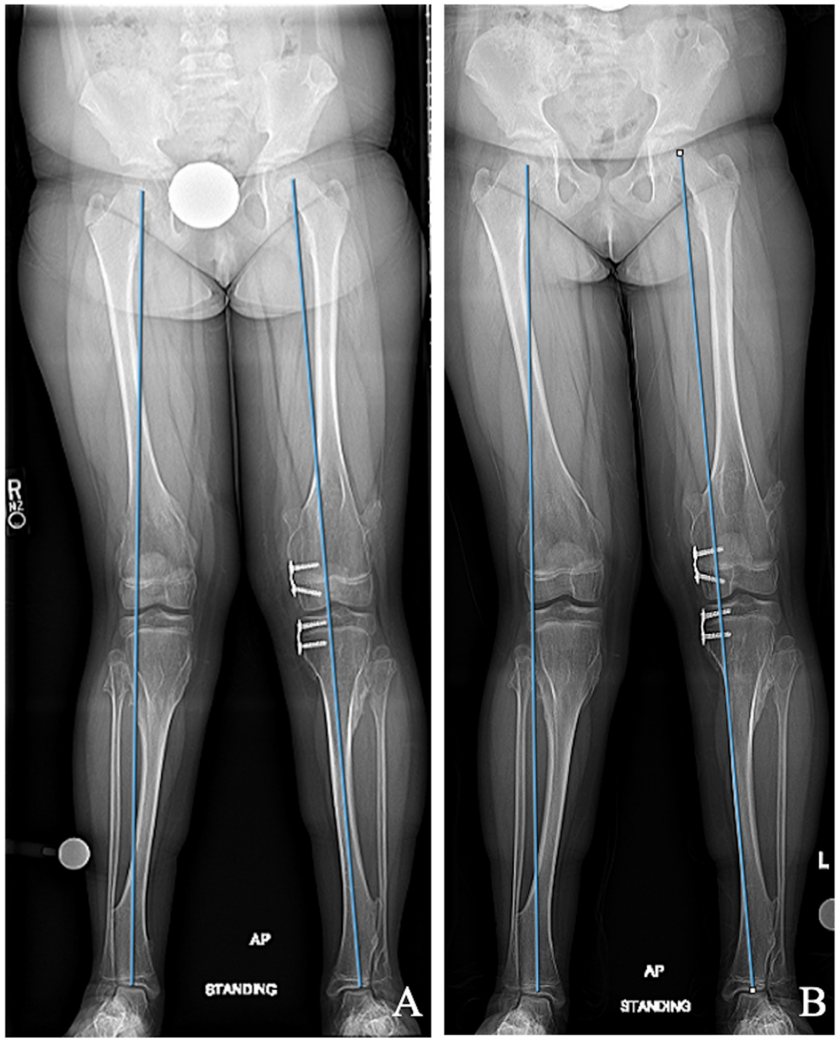

Fig. 7 – Case 2: Standing hip-to-ankle radiographs showing progressive right valgus deformity 2 months (A) and 4 months (B) post medial distal femoral osteochondroma excision. Right leg mLDFA measured 89 and 87º respectively and mMPTA measured 96º at both time points.

(Fig. 8A and B). At that point, the patient had the metaphyseal screws removed on both the right femur and tibia, but the 8 plates were retained in the event future correction is required ("sleeper" plate technique [10].) The functional 8-plate construct was in place for a total of 6.7 months. On her latest follow-up 4-months post screw removal, the patient had full range of motion, and radiographs revealed good alignment (Fig. 8C).

Discussion

Pathologic genu valgum occurs frequently in MHE, and is thought to be the result of the exostosis mass affecting the growth of the bone [11]. To our knowledge, this is the first report of a valgus limb deformity secondary to excision of a medial distal femoral osteochondroma either isolated, or in combination with excision of a proximal tibial osteochondroma. Only one other study in the literature reports a similar phenomenon after isolated excision of medial proximal tibial osteochondromas in 3 patients [9]. Similarly to our study, the patients in the former study did not present with angular deformity prior to the resection and, in all 3 cases, rapidly progressive valgus deformity was noted between 6, and 14 months after the resection procedure. However, there is no report of this occurring after the resection of distal femoral exostoses, as found in the present study.

Given the lack of deformity prior to surgical excision of the osteochondromas, and the fast progression of deformity post excision, it is likely the deformity was secondary to the procedure rather than the disease. A comparable rapid onset valgus deformity is seen is children after a traumatic fracture of the proximal tibial metaphysis also known as a Cozen's fracture. The inferred mechanism behind this phenomenon is that the increased blood flow to the injured area caused by the body's natural response to trauma stimulated the surrounding physis [12,13]. In our patients' scenario, resection of the exos-

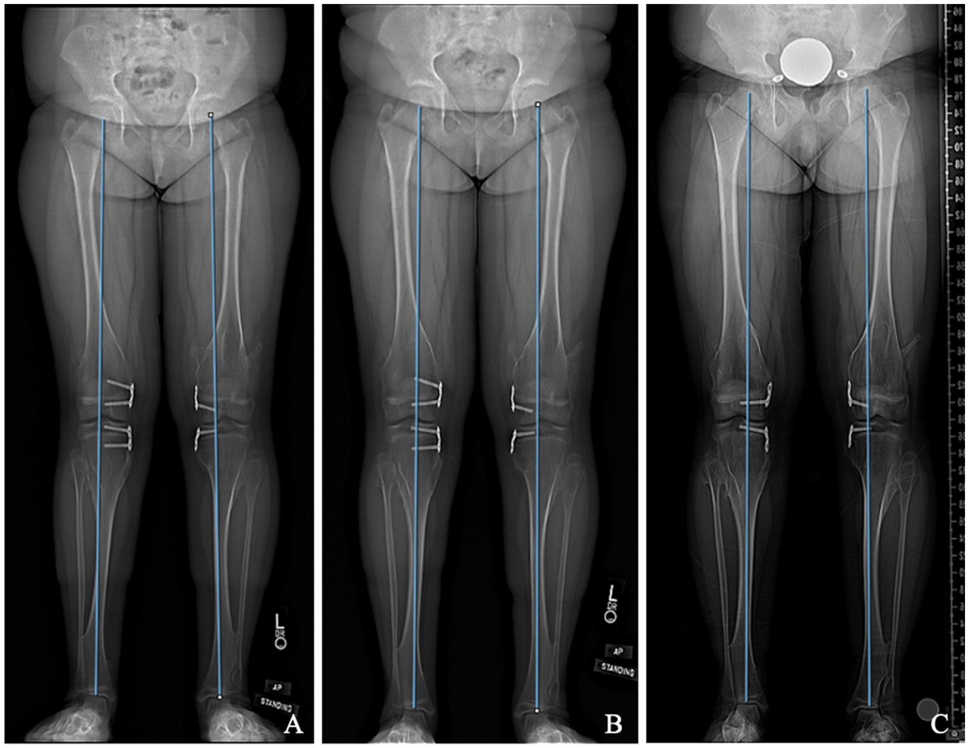

Fig. 8 – Case 2: Standing hip-to-ankle radiographs showing progressive correction of right valgus deformity 4 months (A) and 6 months (B) after implant mediated guided growth procedure. (C) Standing hip-to-ankle radiographs showing satisfactory right limb alignment 4 months post removal of metaphyseal screws. Right leg mLDFA measured 89, 93, and 91º respectively and mMPTA measured 93, 92, and 90º respectively.

toses may have acted as surgical trauma resulting in hyperemia and an increased blood flow to the medial distal femoral and proximal tibial physis, thus stimulating asymmetric bone growth.

Resection of a symptomatic osteochondroma is considered a standard procedure for MHE patients. Surgical excision tends to produce favorable outcomes with a recurrence rate of 2% and complication rate of 13% [14,15]. The previously reported complications include neuropraxia, arterial laceration, compartment syndrome, and fracture. However, based on the 2 cases reported in this study and the other 3 cases in the literature, we suggest advising the patient and parents of the potential for postoperative angular deformity. Additionally, surgeons should be aware of this potential complication, and carefully observe skeletally immature patients with serial hip to ankle radiographs after excision of an osteochondroma near the knee.

In this report of 2 cases, hemiepiphysiodesis was successful in correcting the valgus deformity that arose after osteochondroma excision. However, it should be noted that MHE patients tend to have an abnormal physes, and correction times may vary in this population. A previous study found that while hemiepiphysiodesis can be successful in correcting the limb alignment in the setting of MHE, correction times may be prolonged by half a year when compared to patients with idiopathic deformities [16]. While other studies have found that post-traumatic valgus deformity of the proximal tibia may correct spontaneously, no comparable study exists for valgus deformity of the distal femur [17,18]. Treatment with hemiepiphysiodesis produced favorable outcomes in the present patients, but due to the limited number of cases, and lack of long term follow-up, no formal treatment recommendations can be made.

Conclusion

Resecting an osteochondroma located near the growth plate in the distal femur or proximal tibia in a growing child may be associated with a later genu valgum deformity. The treating orthopedist should provide the patient and the family advice regarding this potential complication and observe the patient following the excision.

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