

Case Report

## Acute Lower Extremity Ischemia due to the Popliteal Pseudoaneurysm in a 16-year-old Boy with Multiple Exostoses

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**Osteochondromas or exostoses are the most common benign bone tumors. This is a case of a 16-year-old boy with multiple osteochondromatosis which caused the popliteal pseudoaneurysm and a subsequent distal embolism. Vascular complications, especially acute limb ischemia, caused by osteochondromas are very rare. This abnormality should be searched for when dealing with young patients with acute limb ischemia.**

**Keywords:** multiple exostoses, pseudoaneurysm, vascular complications of exostosis

### INTRODUCTION

A 16-year-old boy was referred to our department with an acute right lower limb ischemia. He had previous history of leukemia at 3-year-old and thyroid cancer at 15-year-old. He was well until when he presented with an acute onset of right foot pain, numbness, and sensation of coolness. Computed tomography (CT) angiogram revealed a pseudoaneurysm of right popliteal artery with an embolism in the trifurcation of below-the-knee popliteal artery and also osteochondromas in his distal femur and proximal tibia (Fig. 1).

The patient underwent emergency operation. The popliteal artery was exposed through a medial approach and found overlying the sharp apex of the distal femoral exostosis (Fig. 2). After controlling of proximal and distal vessels, the pseudoaneurysm, which included non-organized fresh thrombus, was opened. The irregular surface of a sessile exostosis was palpated through the

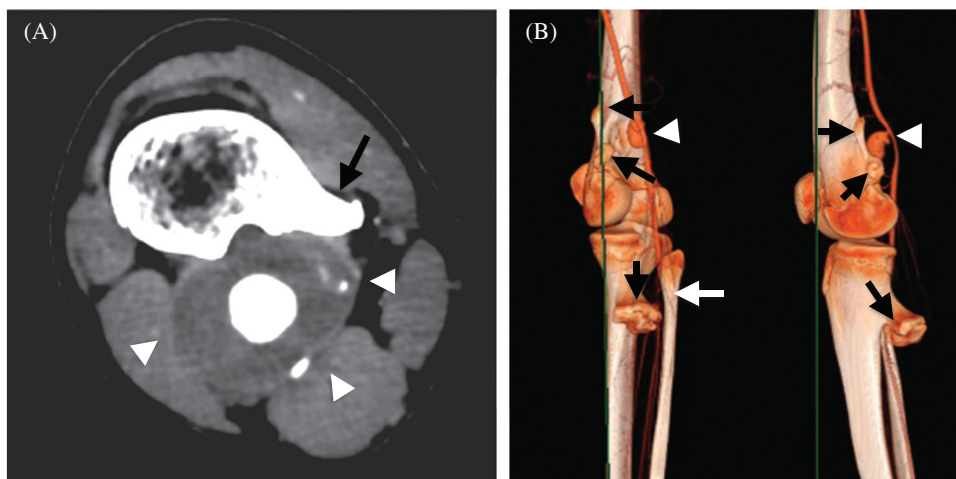
lumen of the aneurysm. The aneurysm was resected and vein graft interposition was performed (Fig. 3A). An embolectomy of the distal popliteal artery was performed using traditional Fogarty embolectomy catheter via a transverse arteriotomy at the level of anterior tibial artery. Intraoperative angiogram showed an embolism at the dorsalis pedis artery and thrombus aspiration was additionally performed. Completion angiogram revealed good results (Fig. 3B). All three exostoses (two in femur and one in tibia) were also removed. Pathohistological examination confirmed these bone tumors as osteochondromas.

### DISCUSSION

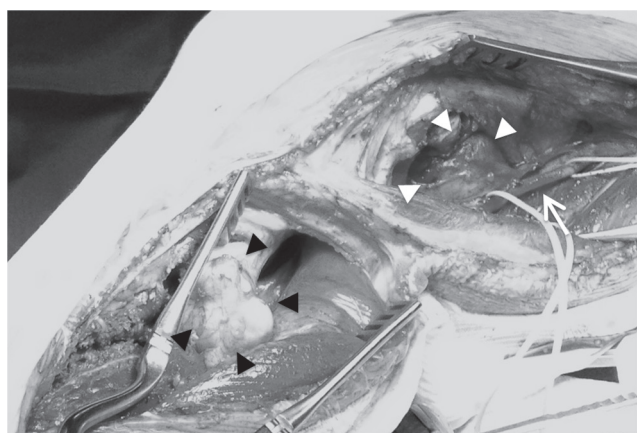
Osteochondroma, also referred to as osteocartilaginous exostosis or plainly exostosis, is the most common benign bone tumor. It can present as solitary osteochondroma or as multiple osteochondromatosis. These tumors account for 10%–15% of all bone tumors, while representing 36% of all non-malignant bone tumors.<sup>1)</sup> The most common sites for these tumors are the distal femur, proximal humerus, and proximal tibia. Osteochondromas are usually asymptomatic and most commonly present as a painless bump. They rarely cause vascular complication, such as arterial occlusion, pseudoaneurysm formation, or venous thrombosis. Pseudoaneurysm formation secondary to osteochondroma was first reported by Paul in 1953.<sup>2)</sup> It is the most common (64%) vascular complication,

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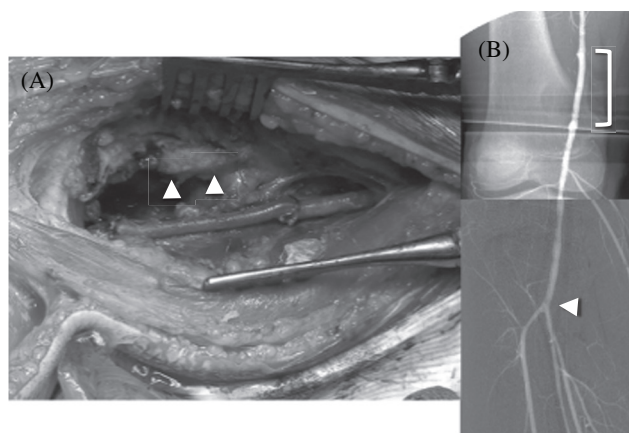
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**Fig. 1** Axial (A) and volume rendering (VR) (B) images of preoperative computed tomography (CT) scan. The white arrow heads indicate the pseudoaneurysm arising from the popliteal artery. The black arrows indicate multiple osteochondromas on distal femur and proximal tibia. A distal embolism is detected at the trifurcation of the below-the-knee popliteal artery (white arrow).



**Fig. 2** Intraoperative photograph. The pseudoaneurysm (white arrow heads) arises from the popliteal artery (white arrow). There are multiple exostoses on proximal tibia (black arrow heads) and distal femur (already removed).



**Fig. 3** (A) Intraoperative photograph after the vein graft replacement and the excision of osteochondroma on distal femur (white arrow heads). (B) Completion angiogram shows good results after the vein graft replacement and the embolectomy at the distal site (white arrow head).

mostly involving the popliteal artery.<sup>3)</sup> The popliteal artery, adjacent to the distal femur, is fixed proximally at Hunter's canal and distally at the trifurcation. Thus, both the proximal and distal portions of the popliteal artery have little mobility and can be displaced and stretched over an adjacent osteochondroma.<sup>4)</sup> Therefore, pseudoaneurysm due to an osteochondroma tends to occur in the popliteal region. In fact, we found 78 reported cases of pseudoaneurysm caused by osteochondroma, of

which located 6 in the brachial, 5 in the superficial femoral, 65 in the popliteal (2 in below-the-knee popliteal), and 2 in the genicular branch artery. Male sexes were 86% and their average age was  $19.5 \pm 9.6$  years old (median: 17). This complication most commonly affects young patients who are approaching skeletal maturity. At this developmental stage the soft cartilage cap covering the exostosis undergoes ossification and becomes firm and often sharp. It occurs not only in solitary but also in

multiple lesions like this case and solitary exostosis were more popular (80%) in the reported cases developing pseudoaneurysm. Also known as familial osteochondromatosis or diaphyseal aclasis, Hereditary multiple exostoses is characterized by the development of multiple osteochondromas. The diagnosis can be made when radiologically at least two osteochondromas of the juxta-epiphyseal region of long bones are observed. In the majority of patients, a positive family history and/or mutation in one of the EXT genes can be detected.<sup>5)</sup> The earliest report of a family to be affected was by Boyer in 1814<sup>6)</sup> and this is thought to occur with an incidence of 1:50000 in general population.<sup>7)</sup> It is an autosomal dominant condition caused by mutations in three different genes. The EXT1 gene on chromosome 8q23–q24 and the EXT2 gene on chromosome 11p11–p12 are most commonly affected with abnormalities in the EXT3 gene on chromosome 19p less frequently reported.<sup>8)</sup> The patient in this reported case presented multiple osteochondromas (two in the distal femur and one in the proximal tibia), but did not have any familial history.

The most popular symptoms in the reported cases were acute or subacute onsets of pain or painful mass (73%), and of which five cases caused an extended rupture. A rapid expansion (rupture) of pseudoaneurysm is a serious complication because it may need some blood transfusion and also may be miserable if it causes a neurological deficit.<sup>9)</sup> Cases which present acute limb ischemia due to distal embolism are also very rare. There are only three cases previously reported. The reason is that since the pseudoaneurysm itself presents a remarkable symptom, they will be diagnosed before developing distal embolism.

Although in this endovascular era, the open surgery is a better option. There might be a few cases that can be treated by endovascular technique, such as embolization,<sup>10)</sup> but usually adjacent osteochondroma needs to be excised and also the chronically injured artery itself needs to be treated. Moreover, the artery, in which the pseudoaneurysm occurs frequently, is located in non-stenting zone. As for the open surgery, although those diseased lesions can be treated by either medial approach or posterior approach, in most cases, medial approach seems to be easier due to the medial deviation of the artery caused by osteochondroma and subsequent pseudoaneurysm. Distal embolism in this case was treated directly via a transverse arteriotomy at the level of anterior tibial

artery, but embolisms in these lesions can be treated from proximal using devices such as an over-the-wire embolectomy catheter. We decided those treatment strategies because the osteochondroma in tibia had also to be excised.

## CONCLUSION

This case report highlights a rare vascular complication of osteochondromas. Vascular complications, especially acute limb ischemia, caused by osteochondromas are very rare. Care should be taken in treating young patients with acute limb ischemia.

## DISCLOSURE STATEMENT

None.

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