Exostoses are the developmental abnormalities of the growth plate, and they are the most frequent benign bone tumors. They are usually seen during adolescence, and usually occur in the femur. Although they tend to be asymptomatic, vascular complications may occur. They are four times more frequent in men than in women. The most common vascular complications are pseudoaneurysm, vascular compression, arterial and venous thrombosis, and the pseudoaneurysms most frequently involve the popliteal artery. Herein, we present a female with previous surgical excision of exostosis and a popliteal artery pseudoaneurysm that caused extensive destruction of the distal part of the femur.
CASE

A 25-year-old female was admitted to our clinic with a 4-month history of pain and swelling of the medial side of her left thigh, just above her knee. The radiographic examination performed by the department of orthopedics about 15 months ago revealed multiple exostoses of both femur and tibia, and surgical excision of three exostoses was performed due to severe pain. The pain of her left thigh recurred along with swelling, nine months after the first operation. There was no history of trauma.

On physical examination, a pulsatile mass 7 cm in diameter was found in the left popliteal fossa. Peripheral pulses were palpable. Plain X-ray showed a hole in the distal femur, and a circular density around the hole and femur, corresponding to the pseudoaneurysm (Figure 1). Computerized tomography angiography (CTA) demonstrated multiple exostoses, and a popliteal artery pseudoaneurysm which led to extensive destruction of the femur (Figure 2-3).

The popliteal artery was exposed through a posterior approach. After controlling proximal and distal vessels, the pseudoaneurysm with a size of 7x6 cm was opened. The integrity of posterior side of distal femur was broken, and 2x4 cm defect was seen. There was a hole 5 mm in diameter on the anteromedial aspect of the popliteal artery, and it was repaired with a vein patch. The irregular surfaces of the remaining destructed bone edges were smoothened.

The postoperative course of the patient was uneventful, and she was discharged from the hospital on the forth postoperative day, without any complications.

DISCUSSION

Exostoses are observed in 1-2% of the population. They usually tend to be solitary (90%). Multiple exostoses are 10 times more likely to degenerate into malignant chondrosarcomas compared to the solitary ones. The skeleton should be screened after diagnosing an exostosis, basically with plain radiography to investigate multicentricity of the lesions.
Pseudoaneurysms are the most common vascular complications, and it has been believed that they develop due to continuous friction of the artery by the exostosis. The vascular complication usually appear in the second decade of life due to the ossification of the cartilage cap of the exostosis, which then becomes a firm and rigid spike.

In our patient, no exostosis was found on the bone around the pseudoaneurysm. Although she had surgery 15 months ago, her complaints began 4 months before she admitted to our clinic.

Pseudoaneurysm may be a late complication of the previous surgery, or develop as a result of a recurrent exostosis. However, in our patient, it was almost impossible to define the exact pathophysiological mechanism. Due to presence of a number of exostoses in those patients, we consider that those bone tumors may be primarily responsible for the etiology. However, the enlarging pseudoaneurysm-induced necrosis was seen at the distal part of femur, and no exostosis could be found during surgery. This bony destruction is similar to that observed in chronic-contained rupture of an abdominal aortic aneurysm in which a pseudoaneurysm develops in contact with the lumbar spine, sometimes causing pressure necrosis of the vertebral body.3 Although erosion of the exostosis has been described in the literature,3-5 we could not encounter this kind of bone destruction due to the popliteal pseudoaneurysm in a patient with multiple exostoses.

CT scans are currently the preferred modalities to demonstrate the relationship between the exostosis and the aneurysm or artery. Covered stent grafts or surgery may be performed to treat the pseudoaneurysms.6,7 In our patient, surgical treatment was thought to be necessary to decompress the mass effect of pseudoaneurysm. Medial suprageniculate or posterior approach may be preferred for surgical repair the popliteal pseudoaneurysm.2,3,8 Even if the both techniques have their own advantages, they are highly comparable.9 Multiple surgical strategies may be used for repair. We preferred vein patch angioplasty since the arterial defect was small, and it was easy to be repaired with this technique.

In conclusion, pseudoaneurysms may develop as a result of exostoses of the bones, and since the recurrence of exostosis is possible, it should be considered as a cause of recurrent symptoms and occurrence of complications even after surgical resection. A pseudoaneurysm, which creates a constant progressive pressure by expansion, may destruct the exostosis and bone over time. Surgical treatment is recommended as an urgent procedure once the diagnosis is established to prevent rupture and further destruction of the bone. The reconstruction of the popliteal artery has a satisfactory postoperative course.

Conflict of Interest
Authors declared no conflict of interest or financial support.

REFERENCES