

## Popliteal artery pseudoaneurysm secondary to hereditary multiple exostosis

### Hereditär multiple ekzositoza sekonder popliteal arter psödoanevrizması

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#### ABSTRACT

A 25-year-old female patient with hereditary multiple exostosis was diagnosed with a pseudoaneurysm of the left popliteal artery. She previously underwent surgical excision of the exostoses located in the left distal femur and proximal tibia. Left leg pain occurred with swelling nine months after surgery, and radiographic examination revealed a popliteal artery pseudoaneurysm with an extensive destruction of the femur. In young patients, non-traumatic pseudoaneurysms of the distal femoral artery may be a complication of an exostosis, and those aneurysms may very rarely destruct the femur.

**Keywords:** False aneurysm; hereditary multiple exostosis; popliteal artery.

#### ÖZ

Hereditär multiple ekzositozu olan 25 yaşındaki kadın hastaya, sol popliteal arterinde psödoanevrizma tanısı kondu. Hastaya daha öncesinde sol femur distali ve tibia proksimali yerleşimli ekzositoları nedeniyle cerrahi eksizyon yapılmıştı. Cerrahiden dokuz ay sonra sol bacağına şişlik ile ağrılar ortaya çıktı ve radyolojik incelemede popliteal arter psödoanevrizması ile beraber femurda geniş bir harabiyet tespit edildi. Genç hastalarda distal femoral arterde travma dışı oluşan psödoanevrizmalar ekzositoz komplikasyonu olabilir ve bu anevrizmalar çok nadiren femur harabiyetine neden olabilir.

**Anabtar sözcükler:** Yalancı anevrizma; hereditär multiple ekzositoz; popliteal arter.

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.<sup>[1]</sup> The most common vascular complications are pseudoaneurysms, vascular compression, and arterial and venous thrombosis and pseudoaneurysms most frequently involve the popliteal artery.<sup>[1]</sup> Herein, we present a female patient with previous surgical excision of exostoses and a popliteal artery pseudoaneurysm which caused extensive destruction of the distal part of the femur.

#### CASE REPORT

A 25-year-old female patient was admitted to our clinic with a four-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 with swelling nine months after the initial operation. There was no history of trauma.

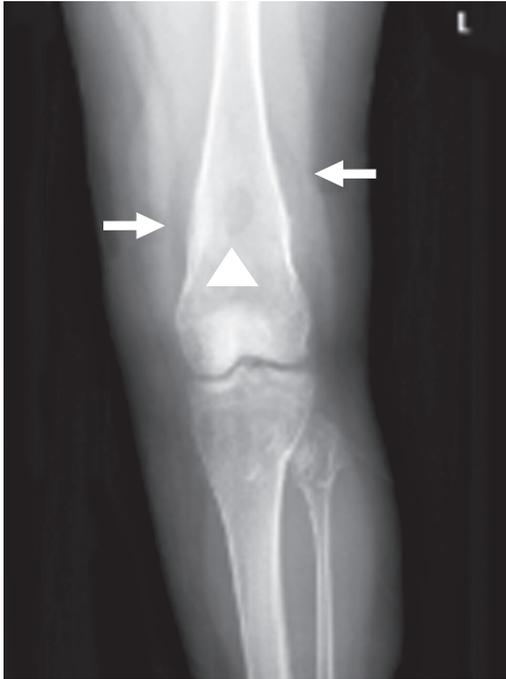
On physical examination, a pulsatile mass, 7 cm in diameter, was found in the left popliteal fossa.

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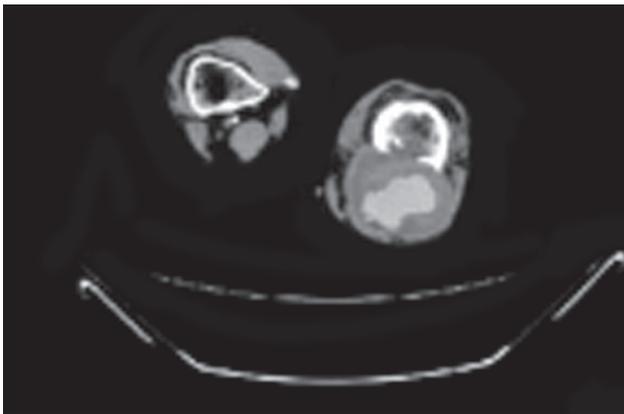
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**Figure 1.** Plain X-ray showing the destruction of distal femur like a hole (arrowhead), and a circular density around the hole (white arrows) suggesting an aneurysm.



**Figure 3.** Pseudoaneurysm and bony destruction in the volume-rendering image.



**Figure 2.** Computed tomography angiography showing pseudoaneurysm and bony destruction clearly on axial view.

Peripheral pulses were palpable. Plain X-ray showed a hole in the distal femur and a circular density around the hole and femur, suggesting a pseudoaneurysm (Figure 1). Computed tomography angiography (CTA) demonstrated multiple exostoses, and a popliteal artery pseudoaneurysm which led to extensive destruction of the femur (Figures 2,3).

The popliteal artery was exposed through a posterior approach. After controlling proximal and distal vessels,

the pseudoaneurysm with a size of 7×6 cm was opened. The integrity of the posterior side of distal femur was broken, and 2×4 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 destroyed bone edges were smoothed.

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

## DISCUSSION

Exostoses are observed in 1 to 2% of the overall population.<sup>[1]</sup> The majority of exostoses (90%) tend to be solitary. Multiple exostoses are 10 times more likely to degenerate into malignant chondrosarcomas compared to the solitary ones.<sup>[2]</sup> The skeleton should be screened after the diagnosis of an exostosis, basically with plain radiography to investigate the multi-centricity of the lesions.

Pseudoaneurysms are the most common vascular complications, and it has been considered that they develop due to continuous friction of the artery by the exostosis.<sup>[1]</sup> The vascular complications 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.<sup>[1]</sup> In our case, no exostosis was found on the bone around the pseudoaneurysm. Although she underwent surgery 15 months ago, her complaints began four months before her admission 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 the 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. In a similar way, pseudoaneurysm may develop in chronic contained rupture of an abdominal aortic aneurysm due to contact with lumbar spine which sometimes causing pressure necrosis of the vertebral body.<sup>[3]</sup> Although erosion of the exostosis has been described in the literature,<sup>[3-5]</sup> we were unable to observe this kind of bone destruction due to the popliteal pseudoaneurysm in our patient with multiple exostoses.

Computed tomography 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 pseudoaneurysms.<sup>[6,7]</sup> In our case, surgical treatment was thought to be necessary to decompress the mass effect of pseudoaneurysm. Medial supragenicular or posterior approach may be preferred for surgical repair popliteal pseudoaneurysms.<sup>[2,3,8]</sup> Even if both techniques have their own advantages, they are highly comparable.<sup>[9]</sup> 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.

#### Declaration of conflicting interests

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