Popliteal artery pseudoaneurysm and secondary ipsilateral deep vein thrombosis caused by an exostosis in a mature adult

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ABSTRACT

Exostosis (also known as osteochondroma) is the most common benign bony lump of young people, usually arising at the distal femur. Vascular complications associated with exostoses are rare and include true aneurysm or pseudoaneurysm formation, deep vein thrombosis, arteriovenous fistula, and arterial insufficiency of the limbs. Few case reports describe pseudoaneurysms resulting from exostoses in mature adults, and no consensus has been reached regarding the optimal therapy. We report the case of a 51-year-old male patient complaining of persistent right thigh pain with a pulsatile mass and right calf swelling, without a history of trauma or hereditary multiple exostoses. The diagnosis was confirmed by computed tomography angiography, which showed a pseudoaneurysm of the popliteal artery resulting from an exostosis on the lateral aspect of the distal femur. A Doppler ultrasound examination confirmed popliteal vein thrombosis caused by the compression of the pseudoaneurysm. Surgical treatment consisted of removing the exostosis, excision of the pseudoaneurysm, and an end-to-end anastomosis. The deep vein thrombosis was treated with rivaroxaban for 3 months. The patient was discharged after 6 days and followed up for 6 months with satisfactory results. (J Vasc Surg Cases Innov Tech 2024;10:101375.)

Keywords: Deep vein thrombosis; Exostosis; Osteochondroma; Popliteal pseudoaneurysm

Exostosis or osteochondroma is the most common benign bone tumor, with an incidence of 10% to 15%.[1] Most osteochondromas are asymptomatic, and complications occur in ~4% of cases, including skeletal deformities, joint disorders, growth abnormalities, malignant degeneration, neurologic complications, fractures, overlying bursa formation, and vascular compromise.[2] Vascular complications are rare, with popliteal artery pseudoaneurysms the most common.[3,4] Vascular injuries are frequently observed in the first 30 years of life, with most cases occurring in adolescence. We reported the case of a 51-year-old man presenting with a popliteal pseudoaneurysm and secondary ipsilateral deep vein thrombosis (DVT) due to an exostosis on the distal femur. The patient provided written informed consent for the report of his case details and imaging studies. The ethics committee of the First People's Hospital of Lianyungang approved the present study (approval no. LW-20221014001-01).

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CASE REPORT

Clinical presentation. A 51-year-old man presented with a 2-month history of persistent swelling of the right calf and thigh pain with a pulsatile mass, without intermittent claudication. He had no history of vigorous exercise or blunt trauma to the region. A pulsatile mass was found in his distal medial thigh with palpable pedal pulses. In addition, examination of the right leg disclosed enlargement of the calf and a positive Homans sign. All blood test results were unremarkable.

Radiologic evaluation. An arterial and venous Doppler ultrasound examination detected a large popliteal pseudoaneurysm and subsequent mass effect causing an acute DVT (Fig 1). Computed tomography angiography (CTA) confirmed the presence of a pseudoaneurysm due to direct injury from an exostosis at the posterior distal femur (Figs 2 and 3; Supplementary Video 1). The pointed lesion was intact with a sharp bony spur, suggestive of an exostosis. There was no radiographic evidence of an arteriovenous fistula, distal arterial thrombosis, or other bone deformity. The pseudoaneurysm measured 61.7 × 57.9 mm, with persistent flow arising from the popliteal artery. The interior contained a large mural thrombus occupying a part of the pseudoaneurysm. Adjacent inflow and outflow arteries were patent, with three-vessel runoff to the ankle. In the absence of worrisome imaging features of a thickened cartilaginous cap, irregular or lobulated margins, irregular or scattered calcifications, internal lytic areas, and erosion or destruction of adjacent bones,[5] the exostosis was considered benign, and we decided on operative treatment.

Surgical treatment. With the patient under general anesthesia, the popliteal artery was exposed through an

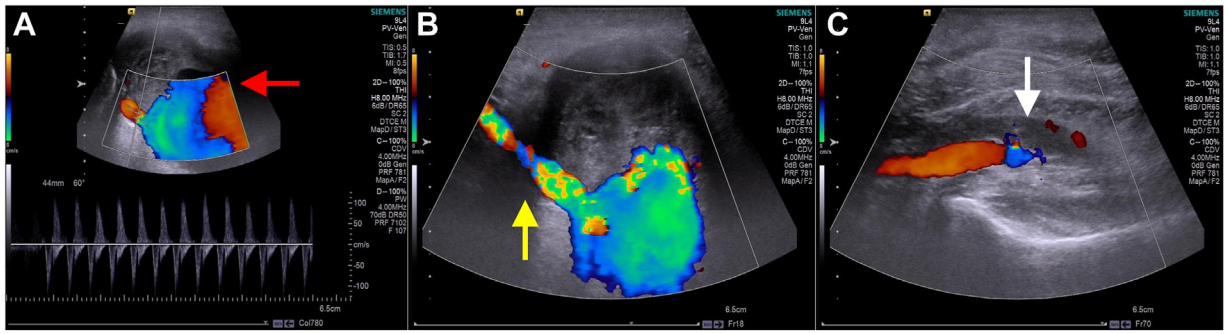

Fig 1. A,B, Color Doppler ultrasound scans showing classic yin-yang sign (*red arrow*) in the body of the pseudoaneurysm and revealing the pseudoaneurysm next to the popliteal artery (*yellow arrow*). **C,** Venous duplex ultrasound scan showing the noncompressible thrombosed popliteal vein (*white arrow*).

anteromedial longitudinal incision. After systemic heparinization, the popliteal artery was clamped proximally and distally. The pseudoaneurysm sac was opened longitudinally, and clots were evacuated. A 5-mm-diameter hole (Supplementary Fig 1) was found on the anterolateral aspect of the popliteal artery, close to the tip of the exostosis (Supplementary Fig 2). The exostosis was resected at its base, and the remaining bone edges were smoothed (Supplementary Fig 3). This specimen was sent for anatomic pathologic analysis. Arterial reconstruction was performed with an end-to-end anastomosis. The thick pseudoaneurysm membrane was partially preserved to avoid direct contact between the raw bone surface and the artery.

Anatomic pathology. Malignant transformation is only seen in 1% of solitary osteochondromas.[2] Increased cellularity, binucleate cells, and nuclear atypia are evidence of malignant transformation.[6] The results from postoperative histologic evaluation confirmed the diagnosis of a benign exostosis (Fig 4).

Clinical outcome. The postoperative recovery was uneventful, and the patient was discharged on postoperative day 6. Enoxaparin was continued for the duration of hospitalization (100 IU/kg subcutaneously every 12 hours). After discharge, the patient received rivaroxaban for 3 months (15 mg twice daily for 21 days, followed by 20 mg once daily thereafter). At his 1-month follow-up visit, the patient was able to return to a normal level of activity. At postoperative month 3, a Doppler ultrasound investigation was performed and no swelling, no DVT, and no arterial occlusion was observed. Follow-up CTA at 6 months postoperatively demonstrated excellent results (Figs 2 and 3; Supplementary Video 2).

DISCUSSION

Exostosis or osteochondroma is the most frequent benign tumor, accounting for 10% to 15% of all bone tumors.[7] It is an overgrowth of cartilage and bone and ordinarily stops growing when a child is fully grown. As the cartilaginous cap ossifies at the end of growth, it becomes a sharp, bony spike that can cause perforation of the adjacent vessel. Vascular complications are rare and mostly due to femoral osteochondromas.[4] Arterial damage accounts for 91% of vascular complications, with popliteal pseudoaneurysms the most common (49%).[3,4]

The popliteal artery is the most affected because osteochondromas are frequently located in the distal femur (66.2%).[4] Also, the artery is confined within Hunter's canal, leaving it vulnerable to injury from chronic friction. A popliteal pseudoaneurysm is usually diagnosed in adolescence and early adulthood, with a mean age of 20 years.[8] As reported in the literature, most patients with a popliteal pseudoaneurysm present with persistent consist pain and a pulsatile mass.[9] More than one third of cases occur secondary to strenuous physical exercise or trauma.[10,11] The compression of the surrounding vessels due to these pseudoaneurysms can lead to lower limb ischemia or venous thrombosis. In our patient, the popliteal artery damage complicated with DVT is typical but occurred much later in life (at 51 years of age), without a history of recent trauma.

CTA is the preferred diagnostic imaging modality for this complication. Angiography alone can underestimate the size of a thrombosed pseudoaneurysm. Magnetic resonance angiography is usually not the first choice in the diagnosis of an arterial lesion. Duplex ultrasound is helpful to evaluate the blood flow but can miss the presence of an exostosis. When examining the CTA images, bone abnormalities are often ignored by vascular surgeons because vascular complications are extremely rare. Yoon and Park[12] described a case of graft perforation by a spinal bony spur without any major trauma. Dregelid et al[13] reported a pseudoaneurysm of the abdominal aorta due to an osteophyte on the first lumbar vertebra. These reports imply that attention should be given when vessels and stent grafts are exposed to direct contact with any potentially harmful structures, such as fracture fragments, osteophytes, and osteochondromas. Also, lifelong follow-up with serial imaging seems necessary to evaluate the development of these lesions. For patients with osteochondromas, Tepelenis et al[5] recommended regular screening with plain radiographs every year or every other year.

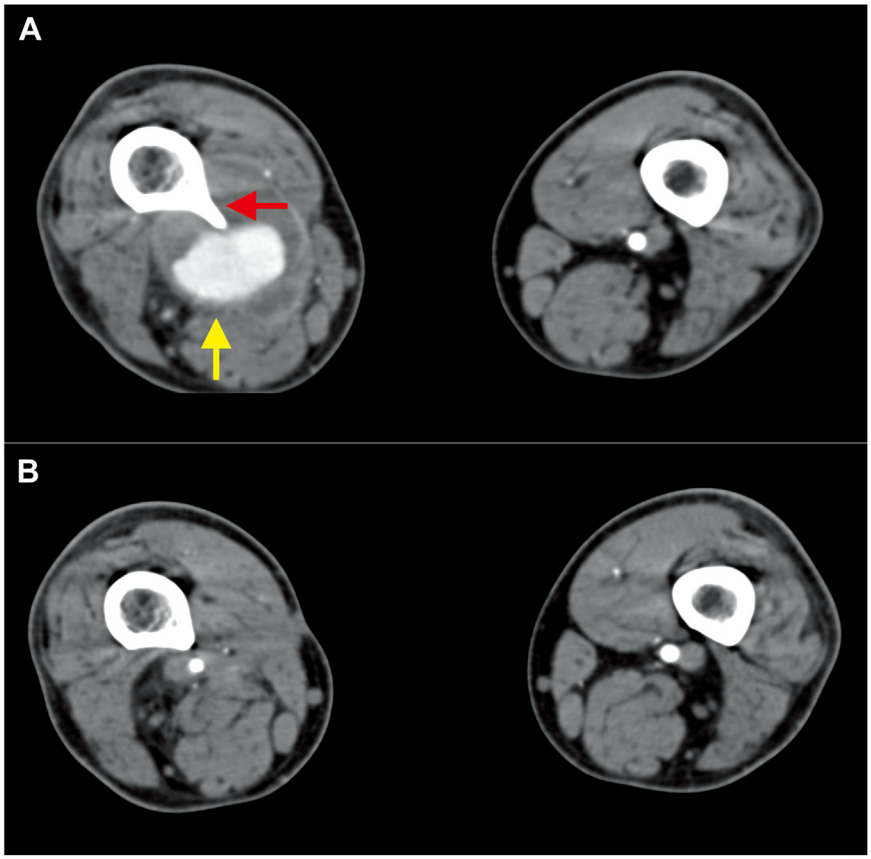

Fig 2. A, Computed tomography angiography (CTA) showing an exostosis (*red arrow*) located at the distal femur with a popliteal artery pseudoaneurysm (*yellow arrow*). **B,** Axial images obtained 6 months after surgery showing no recurrence of the exostosis and complete resolution of the pseudoaneurysm.

Surgical treatment for patients with vascular complications of osteochondromas is recommended as an urgent procedure.[3] For patients with a popliteal artery pseudoaneurysm secondary to exostosis of the femur, knowledge of the anatomy might determine the best approach. The pseudoaneurysm can involve the proximal popliteal artery because the exostosis will usually locate in the metaphysis of the distal femur.[2] If the pseudoaneurysm extends upward to the adductor canal, the medial approach will provide the best access for proximal extension.[14] However, if the lesion does not extend proximally beyond the adductor hiatus, the posterior approach might be the preferred approach because of the superior rates of patency in the long term and minimal short-term complications.[14] The present case was complicated by ipsilateral DVT. Our patient underwent popliteal artery pseudoaneurysm repair with a medial approach to reduce the movement of the affected limb (the posterior approach required the patient to be positioned prone on the operating table). In addition, harvesting of the saphenous vein is easier through the medial approach.

Before vascular reconstitution, the most important aspect is to obtain proximal and distal control of the artery. A simple and feasible solution is to place a groin incision to obtain proximal control of the femoral artery.[15] However, this method will prolong the operating time and increase the risk of wound infection. A minimally invasive approach is to insert a balloon catheter into the femoral artery via the contralateral approach and then inflate the balloon.[16] However, the best combination of dilation pressure, balloon diameter, and duration of indwelling has not been reported. This might result in the complications associated with balloon tamponade, such as arterial dissection, rupture, distal embolization, acute kidney injury, and puncture site complications.[17] A pneumatic tourniquet is frequently used during upper and lower limb surgery to reduce bleeding.[18] However, complications such as thigh pain, nerve injury, compartment syndrome, and systemic complications (ie, DVT, hypertension, arrhythmia) can occur.[18] Minimizing the blocking time and optimizing the cuff pressure might reduce the incidence of complications.[19]

Resection of an osteochondroma is necessary to avoid potential complications. Depending on the extent of vessel wall damage, direct repair, end-to-end anastomosis, autologous saphenous vein grafting, or a prosthetic bypass could be necessary. Direct repair of the artery wall should only be attempted if the injury involves ≤30% of

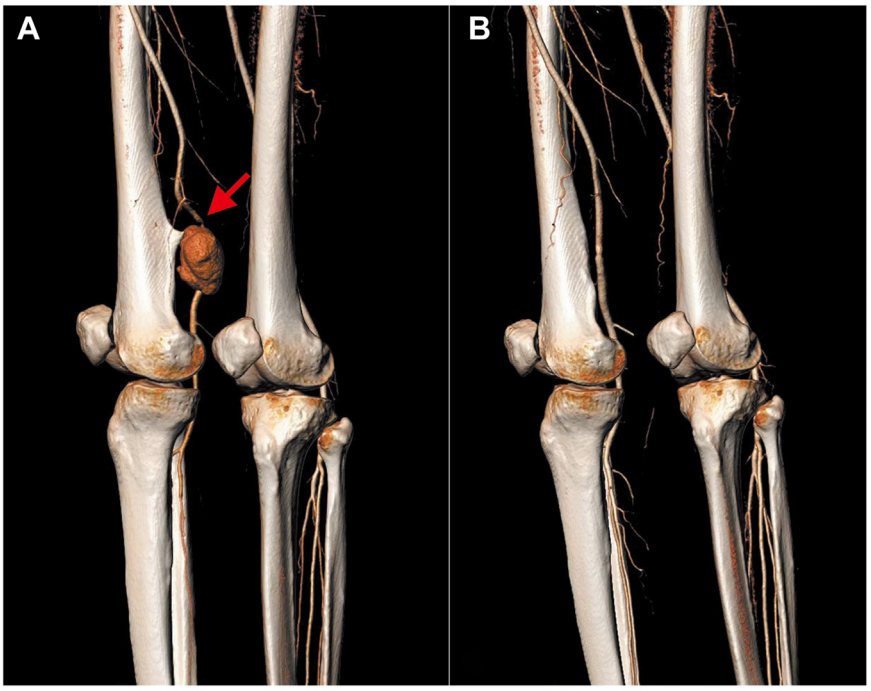

Fig 3. A, Three-dimensional computed tomography angiography (CTA) showing a bone spicule penetrating the pseudoaneurysm (*red arrow*). **B,** Three-dimensional CTA performed at 6 months postoperatively demonstrating excellent patency after revascularization and smooth bone surfaces.

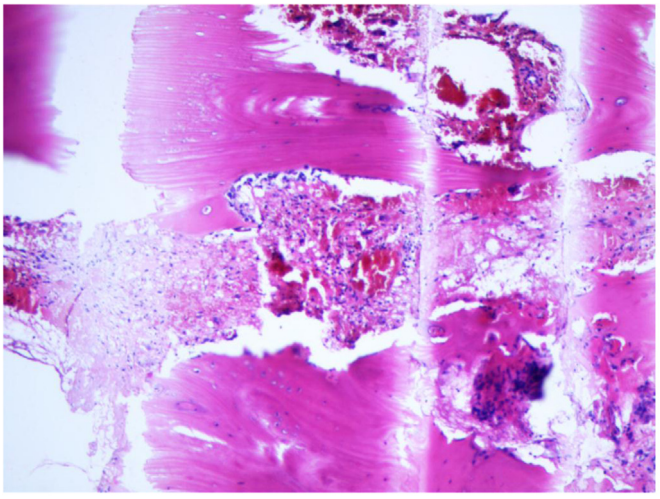

Fig 4. Histopathologic evaluation of the surgical specimen showing a bone tumor with interstitial hemorrhage but no prominent cartilage cap.

the circumference.[20] A tension-free end-to-end anastomosis can be used if the resected segment is ≤2 cm.[21] Otherwise, femoropopliteal bypass should be performed. The great saphenous vein should be preferentially used for femoropopliteal bypass because of its superior long-term patency.[22] A prosthetic vascular conduit (polytetrafluoroethylene or Dacron) is an acceptable alternative if the great saphenous vein is not available.[23]

Endovascular treatment is not recommended, especially in the popliteal location. Although several studies have described long-term results comparable to those after open repair,[24] concerns remain regarding stent fractures and long-term patency rates.[25] In addition, the popliteal artery pseudoaneurysm is frequently observed in the first 30 years of life. It is unacceptable for young people to receive lifelong antiplatelet therapy.

The prognosis after surgery is excellent. As reported by Chen et al,[4] local recurrence of an osteochondroma after resection is low at 1.8%. Only two reports have described pseudoaneurysm formation caused by residual tumor.[26,27]

CONCLUSIONS

We have reported a case of a popliteal artery pseudoaneurysm complicated with DVT, which was successfully treated by exostosis resection, repair of the injured artery, and anticoagulation therapy. A popliteal artery pseudoaneurysm caused by an exostosis in mature adults has been rarely documented in the literature. This case can provide a reference for the surgical treatment of popliteal artery pseudoaneurysms in middle-age adults.

DISCLOSURES

None.

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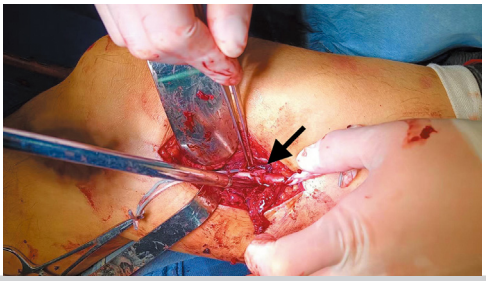
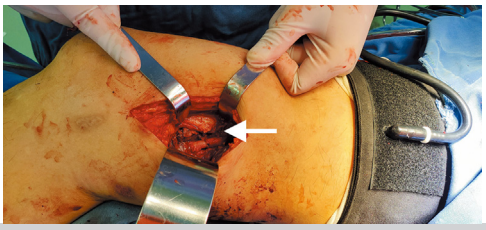
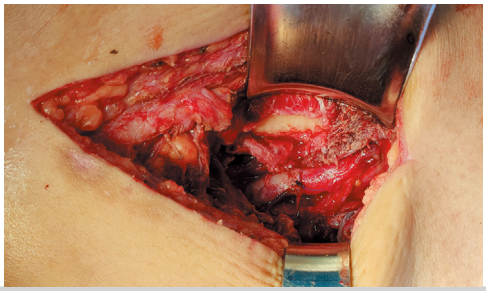
Supplementary Data



Supplementary Fig 1. Intraoperative photograph showing a hole in the anterior wall of the popliteal artery (*black arrow*).



Supplementary Fig 2. Intraoperative photograph depicting the popliteal artery in close contact with a spike of the femoral exostosis (*white arrow*).



Supplementary Fig 3. Intraoperative photograph showing complete removal of the exostosis and excision of the pseudoaneurysm.