🍃 Case Report 🐔

Popliteal Artery Pseudoaneurysm Associated with Osteochondroma

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Osteochondroma is the most common benign bone tumor, which can sometimes cause vascular complications. Here we report two rare cases (a 48-year-old woman and a 28-yearold woman) presenting with pain and a pulsatile mass in the popliteal region. Computed tomography revealed pseudoaneurysm in the popliteal artery, which was closely associated with a protrusion of a femoral osteochondroma. Surgical repairs were performed, and the patients remained asymptomatic during follow-up. Therefore, considering the potential risk of vascular complications, close observation is mandatory in patients with femoral osteochondroma.

Keywords: popliteal artery pseudoaneurysm, osteochondroma, benign bone tumor

Introduction

Osteochondroma is the most common benign bone tumor and frequently appears in the distal femur in young individuals. Although several osteochondromas are asymptomatic, they can sometimes cause vascular complications, which most often involve the popliteal artery. Here we present two rare cases of popliteal artery pseudoaneurysm associated with osteochondromas in the distal femur.

Case Report

Case 1

A 48-year-old woman presented with pain and a pulsatile mass in the right popliteal region without any antecedent trauma. She had previously undergone bilateral excision

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no sensory or motor deficits and no distal pulse deficits (right ankle-brachial pressure index was 1.1). Radiography of the involved bones revealed a spiked bone tumor located in the distal femur. Multidetector computed tomography (CT) revealed a pseudoaneurysm in the right popliteal artery $(10 \times 8 \text{ cm})$, which was closely associated with a sharp protrusion of the femoral osteochondroma (Fig. 1). She was diagnosed with pseudoaneurysm of the popliteal artery associated with osteochondroma, and a surgical intervention was planned. She was administered epidural anesthesia and underwent surgical treatment using a posterior approach through the popliteal fossa with an S incision. This approach was chosen because we could directly oversee the popliteal pseudoaneurysm and osteochondroma through the popliteal fossa, and the approach was in line with the surgeon's preference and experience. The pseudoaneurysm had developed from a small 1-mm hole in the anterior part of the popliteal artery. The surface of the femoral osteochondroma was noticeably sharp, rigid, and firm and was closely related to the pseudoaneurysm. The osteochondroma was completely removed through the lumen of the aneurysm, and the remaining bone edges were subsequently smoothed. Although there was only one small hole in the arterial wall causing the pseudoaneurysm, the popliteal artery was of poor quality. Including the pseudoaneurysm hole, the short segment of the popliteal artery was resected and an end-to-end anastomosis was performed using 6/0 Prolene sutures. The postoperative course was uneventful, and her peripheral pulse was maintained postoperatively. The patient has remained asymptomatic during 5 years of follow-up.

of tibial osteochondromas at the age of 17 years. She had

Case 2

A 28-year-old woman with pain in the right popliteal region without any antecedent trauma was referred to our hospital. She had a history of multiple osteochondromas and had previously undergone excision of a right middle

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Fig. 1 (a) Multidetector computed tomography reveals a pseudoaneurysm in the right popliteal artery (10×8 cm). (b) The pseudoaneurysm (arrowheads) is closely associated with a sharp protrusion of the femoral osteochondroma.



Fig. 2 Computed tomography scan reveals a pseudoaneurysm in the right popliteal artery (4×6 cm), which is related to a protrusion of the femoral osteochondroma.

phalanx osteochondroma at the age of 19 years. Because her father also had multiple osteochondromas, hereditary multiple osteochondromas were suspected. Her peripheral pulse was palpable, and no sensory or motor deficits were detected. CT scan revealed the presence of a pseudoaneurysm in the right popliteal artery (4×6 cm), which was related to a protrusion of a femoral osteochondroma (Fig. 2). She was diagnosed with pseudoaneurysm of the right popliteal artery, and a surgical intervention was planned. She was administered general anesthesia and was placed in the dorsal position for the surgery. A medial incision was made above the knee, and the area was explored. This approach was chosen because it is the most common surgical technique for popliteal aneurysm repair, which is technically easy and safe to perform. The pseudoaneurysm had developed from a 2-mm hole in the anterior part of the popliteal artery. The surface of the osteochondroma was blunt and conical-shaped and was closely related to the pseudoaneurysm. The osteochondroma was completely removed through the lumen of the aneurysm, and popliteal arterioplasty was performed using 5/0 Pronova simple sutures for the hole. The pathological examination of the resected bone protrusion confirmed osteochondroma. The patient was discharged on postoperative day 11 and has been doing well during the 3 months of follow-up.

Discussion

Osteochondroma, a common benign bone tumor, occurs in approximately 1%-2% of the population and is more prevalent in young males. Osteochondromas, which develop from the cortical bone dysplasia in the epiphyseal plate, can injure the adjacent tissue by forming calcified sharp spikes.^{1–3)} They are located in the bone metaphyses and frequently appear in the distal femur or in the proximal tibia.

Many osteochondromas are asymptomatic, which are incidentally diagnosed during radiological examinations. Complications occur in approximately 4% of the cases, which include neurological compromise, skeletal defects, growth abnormalities, and malignant degeneration due to chondrosarcoma- or orthopedic-related complications.^{1,2,4,5} Although vascular complications are rare, the development of pseudoaneurysm, arteriovenous fistula, arterial rupture, arterial or deep venous thrombosis, and luminal stenosis due to extrinsic compression has been reported.⁴⁾ Pseudoaneurysms are the most common vascular complication (up to 60% of the cases with complications), which most frequently involve the popliteal artery.^{2,4,5)} Paul reported the first case of popliteal artery pseudoaneurysm associated with osteochondroma in 1953.6) Raherinantenaina et al. reviewed 101 cases of pseudoaneurysm associated with osteochondroma, which are similar to the present cases.7)

Osteochondroma-related arterial lesions occur either by repeated trauma resulting from knee movement in the presence of an osteochondroma spike or by direct trauma. During the growth period of osteochondromas, they are covered with a soft and pliable cartilage cap, which protects the adjacent artery. However, many authors have speculated that upon cessation of growth, the cartilage cap undergoes ossification and becomes a more firm and rigid bone spur.^{3,8)} When an osteochondroma is located adjacent to the relatively fixed popliteal artery (between the Hunter's canal superiorly and the popliteus muscle inferiorly), constant compression and pulsatile friction can cause continuous damage to the arterial wall, resulting in the formation of a pseudoaneurysm.^{2,5,7,9)} This appears to be the main pathological mechanism when no history of acute trauma exists. However, Matsushita et al. have speculated another mechanism in which a large osteochondroma is shown to cause arterial hole formation and pseudoaneurysm, and then the pressure necrosis via the pseudoaneurysm is shown to break the large osteochondroma into small spikes with rough surfaces.⁸⁾ In case 1, two bony spikes were present, which appeared to be too small to cause continuous damage to the arterial wall; thus, it is possible that pressure necrosis via the pseudoaneurysm break the large osteochondroma into small spikes like Matsushita's mechanism. Unfortunately, this case lacked clinical or histological data that would support this mechanism. Therefore, we suspect that in the present cases, a spiky femoral osteochondroma injured the arterial wall via local compression and continuous rubbing.

To prevent the occurrence of irreversible damage, such as that resulting from the occlusion of distal arteries, phlebitis, or venous thrombosis with a risk of pulmonary embolism, surgical repair of an osteochondroma-related vascular complication is recommended as an urgent procedure.^{2,7,8)} In previous reports, excision of osteochondroma and surgical repair of the pseudoaneurysm (primary closure, venous patch angioplasty, resection with end-to-end anastomosis, or vein bypass grafting) have been performed simultaneously.^{4–9,10} For instance, in cases with one small defect, primary closure of the artery is performed, whereas in cases with a defect larger than 5 mm, venous or synthetic patch angioplasty or resection with end-to-end anastomosis is preferred. Moreover, when end-to-end anastomosis cannot be performed, vein bypass grafting is recommended.^{2,7)} Raherinantenaina et al. have reported that the most common surgical repairs performed for osteochondroma-related vascular complications are vein bypass grafting (40%) and lateral suture (39%). In contrast, resection with end-to-end anastomosis is performed in only 14% of cases.⁷⁾ Although no long-term postoperative follow-up data exist for pseudoaneurysms associated with osteochondromas, the postoperative course was found to be uneventful in the present cases. Therefore, considering the potential risk of vascular complications, close observation is mandatory in patients with femoral osteochondroma.

Conclusion

Local compression and continuous rubbing of the popliteal artery by a spiky femoral osteochondroma were suspected to have caused arterial wall rupture in the present cases. Even without a history of trauma in the popliteal area, the potential risk of vascular complications in patients with an osteochondroma in the distal femur (or with multiple osteochondromas) should be considered when new symptoms appear, such as pain or a pulsatile mass.

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Disclosure Statement

The authors have no conflicts of interest to declare.

Author Contributions

Study conception: AT Data collection: AT Analysis: AT Investigation: AT Writing: AT Critical review and revision: all authors Final approval of the article: all authors Accountability for all aspects of the work: all authors

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