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Multiple osteochondromatosis complicated with a popliteal artery pseudoaneurysm

Multipl osteokondromatozise bağlı gelişen popliteal arter psödoanevrizması

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In this article, we present a 21-year-old male patient who presented with swelling and pain located to right lower thigh and knee. Physical examination and subsequent diagnostic work-up revealed a pseudoaneurysm associated with an osteochondroma at lower thigh. This complication should be considered in young patients with a mass at knee and lower thigh region.

Key words: Osteochondromatosis; popliteal artery; pseudoaneurysm.

Bu yazıda, sağ uyluk alt kısmı ve dizde şişlik ve ağrı yakınması ile başvuran 21 yaşında bir erkek hasta sunuldu. Fizik muayene ve takibinde yapılan tanısal çalışmalarda uyluk alt bölgesinde yer alan osteokondrom ile ilişkili bir psödoanevrizma tespit edildi. Bu komplikasyon diz ve uyluk alt bölgesinde bir kitle ile başvuran genç hastalarda düşünülmelidir.

Anahtar sözcükler: Osteokondromatozis; popliteal arter; psödoanevrizma.

Osteochondromas are the most frequent benign bone tumors, observed in 1-2% of the population.[1-4] These bone abnormalities appear during a growth period as a result of subperiostal displacement of adjacent epiphyseal plane cartilage.[1,2] As common benign bone tumors, they frequently occur in the proximal humerus, tibia, and distal femur.[3] Osteochondromas are usually asymptomatic and can be detected accidentally on plain radiographs of bones. However multiple osteochondromas may easily be diagnosed with physical examination and plain radiographs.[5] Complications such as neurological compromise, skeletal defects or vascular lesions occur in about 4% of osteochondromas.[1,2,4,6,7] Most vascular complications appear in the second decade of life. Vascular complications are four times more frequently in men than in women.[1,2,4,6] Nearly 90% of vascular lesions resulting from osteochondromas affect the arteries, and up to 60% result in false aneurysms.[1,2,4] Bone tumors which lead to the development of false aneurysms are usually located in the distal femur metaphysis, less often in other bones.[1,4] The aim of this case report is to highlight the possibility of arterial complications resulting from osteochondromas.

CASE REPORT

A 21-year-old male was admitted to department of orthopedics and traumatology with pain and development of a mass in the lower third of his right thigh during the last month. There was no history of trauma. There were two small masses located in the distal third of both femurs and bulging masses in the upper part of both tibias (Figure 1). Since multiple osteochondromas mainly have a familial characteristic, the patient and family were questioned. However this was the index case. There were no

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sensory and motor deficits and peripheral pulses were present. Radiography of the involved bones revealed spiked bone tumors located in both tibias and femurs, identified as osteochondromas. Computed tomography (CT) scan identified a right popliteal arterial false aneurysm (Figure 2) and its close relationship with the bone and the tumor.

The patient consulted with the cardiovascular surgery department and an elective surgery was planned. We used a posterior approach and found a 10x8 cm pseudoaneurysm developed from the popliteal artery that was closely related to a spike of femoral osteochondroma (Figure 3). After clamping the artery and opening the pseudoaneurysm, local thrombus was removed. Distal embolectomy was not needed. The femoral artery was repaired with a 6/0 polypropylene suture. Osteochondromas located in the right femur and tibia were completely removed after vascular repair. Pathological examination of the specimens confirmed the diagnosis of osteochondroma.

The postoperative course of the patient was uneventful. The patient was discharged successfully from the hospital on the seventh postoperative day without any complication.

DISCUSSION

Osteochondromas are bone tumors developed during adolescence and located in bone metaphysis, most frequently in the distal femur.[1,4] Occasionally they can be located in the tibia, fibula, scapula, clavicle, rib, vertebra, talus, and patellar tendon. They can occur as solitary (in about 90% of cases) or multiple tumors (in about 10%). Multiple osteochondromas are 10 times more likely to degenerate into malignant chondrosarcomata than solitary ones.[1,2,4,6,8]

Because of the possibility of multicentricity of the exostoses in up to 10% of the patients, a general screening is mandatory, basically with plain radiographs of the upper and lower limbs and the chest.[4] However, Crandall et al.[9] reported that this step must be mandatory only in patients whose parents are affected.

Complications occur in 4% of osteochondromas as neurologic compromise, growth abnormality, or malignant degeneration to chondrosarcoma. Rarely, some osteochondromas can produce a vascular complication. The age of presentation of vascular complications is in the second decade of life, related to ossification of the cartilage cap of the osteochondroma present during the growth period. As soon as growth ceases, the cartilage cap begins its ossification and it becames a spike.[10] As the popliteal artery is fixed in Hunter's canal and in its trifurcation, local compression of the artery by an osteochondroma can stretch the vessel with the appearance of a pseudoaneurysm.[4]

A mass in the popliteal space is the most popular symptom. About 30% of patients had a history of trauma or vigorous exercise prior to the appearance of a pseudoaneurysm.[1] The development of pain in

Figure 1. Preoperative general view of the patient.

Figure 2. Coronal multidetector row computed tomography angiogram of the right lower thigh through the popliteal artery. The white arrows point to the pseudoaneurysm with different stages of blood elements and slight contrast extravasation giving the mass a 'whirl like' appearance (curved arrow). **(a)** Compression of the popliteal wartery (black arrows) with the pseudoaneurysm is well seen. **(b)** Multiple exocytoses involving the femur and tibia are also observed (arrow heads). F: Femur; A: Anterior; P: Posterior.

Figure 3. Intraoperative view of the pseudoaneurysm.

relation to an osteochondroma should be a consequence of a fracture or vascular compression. A patient with a false aneurysm can develop signs of a distal venous thrombosis (in about 2% of cases) and acute limb ischemia. The distal venous thrombosis appears as a result of compression of distal veins, caused by a false aneurysm's mass. The acute limb ischemia can be related to embolisation of intraaneurysm thrombus to the distal arteries or to rupture of a pseudoaneurysm.[1]

A pulsed duplex scan, which reveals a rounded heterogeneous mass adjacent to the damaged artery, is indicated if a vascular complication is suspected. The diagnosis of arterial injury is made if angiography shows aneurysm, pseudoaneurysm, or thrombosis.[1] Angiography may fail to show an aneurysm in the case of total thrombosis. Currently the diagnosis can be done with CT/magnetic resonance angiography techniques without the need for invasive angiography as in our case.

In the case of a popliteal false aneurysm surgery via a posterior approach is recommended. This approach allows good exposure to the popliteal neurovascular structure and allows an appropriate arterial repair depending on the size of the wall lesion. In cases of a single, less than 5 mm hole, primary closure of the artery is preferred. If the defect is larger than 5 mm a vein or the synthetic patch closure or partial artery resection followed by reconstruction with end-to-end anastomosis is preferred. When an end-to-end anastomosis cannot be performed, graft replacement with the autologous vein or synthetic graft is recommended.[1,2,5,6] In our case we preferred the posterior approach and primary repair of the artery.

When contraindications to open surgery appear, Wong recommends closure of a false aneurysm by a coil insertion into its lumen.[11] However this procedure is controversial because of the possibility of coil migration to the systemic circulation, leaving out the bone tumor, which can produce complications.

In conclusion, surgical treatment of vascular complications of osteochondromas is recommended as an urgent procedure to avoid irreversible damages such as occlusion of distal vessels by embolism or phlebitis with the risk of pulmonary emboli. The commonly uneventful postoperative course emphasizes the necessity of surgical treatment of those lesions. In patients with osteochondromas located in the distal femur, one should keep in mind the possibility of vascular complications. Opinions are controversial regarding the necessity of prophylactic resection of all osteochondromas in cases of multiple hereditary osteochondromas. However, surgical resection of osteochondromas appears to be indicated if it is in the vicinity of a vessel, if there is a risk of interference with a joint, if there is a fracture of the osteochondroma, and if there is suspicion of malignant transformation.

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