Acute popliteal pseudoaneurysm rupture secondary to distal femoral osteochondroma in a patient with hereditary multiple exostoses

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ABSTRACT

A 21-year-old man with known hereditary multiple exostoses presented with a 24-hour history of atraumatic bruising and swelling of the posterior thigh. A leaking popliteal pseudoaneurysm was diagnosed on ultrasonography and an emergency saphenous vein bypass graft procedure performed. The patient required a post-operative blood transfusion but otherwise made a full recovery. Vascular complications from osteochondromas are rare and include vessel displacement, stenosis, occlusion, arteriovenous fistulas and pseudoaneurysm formation. Pseudoaneurysms usually present as an enlarging mass behind the knee. Acute rupture of an occult popliteal pseudoaneurysm caused by a distal femoral exostosis has not been reported previously.

KEYWORDS

Hereditary multiple exostoses – Popliteal – Pseudoaneurysm – Osteochondroma

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Osteochondromas constitute 25–50% of all benign bone tumours and are considered a developmental lesion rather than a true neoplasm.¹ Their pathognomonic feature is that of continuity with the underlying bone cortex and medullary canal, and they are composed of cortical and medullary bone with an overlying hyaline cartilage cap. They may be sessile or pedunculated and, when occurring multiply, may be associated with the autosomal dominant condition hereditary multiple exostoses (HME).

Complications arise in 4% of osteochondromas² and include osseous deformity, growth abnormality, fractures and pressure on surrounding structures (nerves, vasculature, bursa). Malignant transformation occurs in 1% of solitary osteochondromas and in 3–5% of patients with HME.¹ Vascular complications include displacement of surrounding vessels, stenosis or occlusion, arteriovenous fistula formation and development of pseudoaneurysms.

This is the first known report of acute pseudoaneurysm rupture secondary to an osteochondroma located on the distal femur.

Case history

A 21-year-old man presented to the emergency department with a 24-hour history of atraumatic bruising and swelling of his left thigh (Fig 1). He admitted to a brief, sharp pain behind his left knee while walking five days prior to the onset of symptoms. His past medical history included HME. He had undergone multiple operations previously to

Figure 1 Bruising overlying posterior aspect of thigh and proximal calf (marked)

Figure 2 Lateral plain film x-ray of left knee with posteriorly projecting exostosis on distal femur (arrow)

Figure 3 Colour Doppler ultrasonography showing turbulent blood flow in the pseudoaneurysm

Figure 4 Computed tomography angiography showing popliteal pseudoaneurysm (arrow) and surrounding haematoma

remove the more prominent of these bony projections from his arms and ankles.

On examination, the thigh appeared swollen in comparison with the contralateral side and was firm in consistency. The bruising extended from the proximal posterior aspect of the patient's left thigh to the proximal calf. There was a generalised fullness in the popliteal fossa but no pulsatility was detected. He was normotensive, and his heart rate and respiratory rate were also within normal limits.

Anaemia was detected on a full blood count with a haemoglobin of 10.1g/dl (normal range: 13.5–17.5g/dl). All other blood tests were unremarkable. Plain film x-rays revealed the presence of multiple exostoses around the knee joint, including one that projected posteriorly on the distal femur (Fig 2). Urgent ultrasonography detected a ruptured popliteal pseudoaneurysm with a large surrounding haematoma (Fig 3). This was in close proximity to the distal femoral exostosis. Computed tomography angiography allowed for accurate localisation of the pseudoaneurysm to aid in operative planning (Fig 4).

The patient was taken immediately to theatre where an S-shaped incision was made over the popliteal fossa. On deeper dissection, an extensive haematoma was found in

the area of the subsartorial canal that was tamponading the ongoing haemorrhage. The popliteal artery was isolated and the haemorrhage controlled using vascular slings. The artery was found lacerated and overlying the sharp apex of the distal femoral exostosis, which was removed using bone nibblers. A 7cm long great saphenous vein graft was used to bypass the damaged artery and arterial flow was restored. The patient required a 3-unit post-operative blood transfusion but made an otherwise unremarkable recovery and was discharged on the fifth post-operative day.

Discussion

Also known as familial osteochondromatosis or diaphyseal aclasis, HME is characterised by the development of multiple osteochondromas. The earliest report of a family to be affected was by Boyer in 1814⁵ and this is thought to occur with an incidence of 1:50,000 to 1:100,000 in Western populations.⁴ It is an autosomal dominant condition caused by mutations in three different genes. The EXT1 gene on chromosome 8q23–q24 and the EXT2 gene on chromosome 11p11–p12 are most commonly affected with abnormalities in the EXT3 gene on

chromosome 19p less frequently reported.²

Pseudoaneurysm secondary to osteochondroma was first reported in 1953 by Paul.⁵ It is the most common vascular complication (64%) and most frequently involves the popliteal artery.⁶ This is due to the high frequency of osteochondromas in this location and the fact that they lie in close proximity to the fixed position of the popliteal artery as it leaves the adductor canal. At this point there is a relative lack of mobility of the overlying artery, which is at risk of abrasion by the bony lesion. Normal movement or repetitive trauma can cause chronic abrasion, which can lead to laceration of the arterial surface.¹

The presence of a popliteal pseudoaneurysm in association with osteochondromas has been reported extensively in the literature. Such patients usually present with an enlarging mass behind the knee. It may be pulsatile in nature with a history of repetitive or traumatic activity. This complication most commonly affects young patients who are approaching skeletal maturity and occurs in solitary and multiple lesions in equal frequency. At this developmental stage the soft cartilage cap covering the exostosis undergoes ossification and becomes firm and often sharp as seen in the case presented here. Acute rupture of a popliteal pseudoaneurysm has only been reported twice: once from a tibial osteochondroma⁷ and once from a fibula osteochondroma.⁸

Conclusions

This case highlights this rare vascular complication of osteochondromas and is the first reported case of acute popliteal pseudoaneurysm rupture secondary to a distal femoral lesion.

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