PSEUDOANEURYSM OF POPLITEAL ARTERY SECONDARY TO HEREDITARY MULTIPLE EXOSTOSES: CASE REPORT

Nabeel MS Qattan, FRCS; Muaffak Jawdat, Facharzt; Ali Al-Sabti, FRCS

The popliteal artery is the most common site of peripheral aneurysmal formation. Arteriosclerosis, syphilis and mycotic infection are the most common etiological factors while in young adults, the aneurysm is often of the false type and caused by trauma.^{1,2}

Osteochondroma around the knee joint can give rise to such pathology that was first described by Brailsfords in 1953^{3,4} followed by a more detailed account by Paul in the same year.⁵

Since that time, approximately 27 cases have been reported in the English literature, including ours.^{6-10,16,20-22}

Case Report

A 23-year-old Saudi male was referred from another hospital with a history of severe pain and swelling in the right lower limb which occurred during jogging. The patient experienced difficulty in walking due to the pain and the decreased movement of the knee, but no pain at rest. There was a family history of hereditary multiple exostoses (HME).

Physical examination showed diaphoretic skin, marked swelling of the right posterior lower extremity distal to the mid thigh primarily behind the knee. There was increased warmth and tenderness and palpable popliteal pulse. A thrill was reported early at the referring hospital, but disappeared after arrival at our hospital. All pedal pulses were palpable. The ankle/brachial pressure was 0.8. We recommended medical advice and treatment to the members of the family who were suffering from HME but the family refused.

Investigation

Plain x-ray showed multiple osteochondroma over the left shoulder and left knee (Figure 1).

Computed tomography (CT) and angiogram showed a false aneurysm at the popliteal artery with bony spikes originating from both femurs (Figures 2 and 3). This was

FIGURE 1. Plain x-ray of the right femur.

FIGURE 2. Computed tomography scan of both knees.

From the Departments of Surgery (Drs. Qattan, Jawdat) and Orthopedics (Dr. Al-Sabti), Riyadh Armed Forces Hospital, Riyadh.

Address reprint requests and correspondence to Dr. Qattan: Department of Surgery, Riyadh Armed Forces Hospital, P.O. Box 7897, Riyadh 11159, Saudi Arabia.

Accepted for publication 4 December 1993.

performed in the referral hospital. Magnetic resonance imaging (MRI) performed at our hospital confirmed the presence of a large pseudoaneurysm which showed areas of decreased signal, signifying flow and a small area of hematoma seen medially (Figure 4).

Operation

Through a medial approach, the exostosis was removed, the aneurysmal sac was excised and the defect in the artery was repaired by saphenous vein patch. The patient had an uneventful postoperative course. He was followed up in the clinic for six months, after which he failed to appear. On the last visit, his pedal pulses were palpable and the ankle/brachial pressure was 1.

Discussion

In 1786, HME was first described by Hunter and Palmer in lectures on the principles of surgery¹² as a skeletal disorder that primarily affects endochondral bone during growth. Virchow named the disorder multiple exostoses¹³ in 1876. Keith suggested the name diaphyseal aclasis.¹⁴ Other names such as osteogenic disease, chondral osteogenic dysplasia, chondral osteoma, dyschondroplasia, deforming chondroplasia, multiple hereditary osteochondromata, multiple cartilaginous exostoses, exostatic dysplasia and exostosis have been recommended for the term HME. It is an autosomal dominant disease with the prevalence estimated in the United Kingdom as nine per 1,000,000.^{15,23}

In general, chronic pulsation of any vessel on a sharp, bony spike may cause erosion of the arterial wall, and the blood leaks out and becomes encased by the surrounding tissues, which produce the false aneurysm.

Review of patients previously described with a false popliteal aneurysm caused by exostoses, whether single or multiple (HME), reveals that the patients were young with a mean age of 20 years, range from nine to 45 years, with male predominance. Half of the cases had multiple exostoses in other locations.

The patient is presented with an increasingly painful mass and a bruit is audible on auscultation or a thrill palpated over the swelling. Although this is not usually the case, in this particular case the thrill disappeared and it was absent in other reported cases⁸⁻¹¹ where they were misdiagnosed initially. In the Marcove et al. case,¹⁰ the aneurysm biopsied was mistaken for a bony tumor, while in the Denman et al. case,¹⁷ biopsy of the aneurysm was mistakenly performed because of the absence of the thrill and palpable impulse. This happened because the slow blood flow in the aneurysmal sac caused a thrombosis which eventually led to compression of the artery. This may explain why a bruit and thrill cannot be elicited in some cases and disappears in others.

FIGURE 3. Angiogram of the right popliteal artery.

FIGURE 4. Magnetic resonance imaging of the right knee.

Usually the distal pulses are palpable but in a case reported by Shah,⁶ there were symptoms of peripheral arterial insufficiency occurring due to embolism from the aneurysm. Ultrasound and CT are helpful in establishing the diagnosis. Ferriter et al.¹⁶ stated that a CT scan with contrast media will show filling of a pseudoaneurysm and thus confirm the diagnosis.

The literature confirms that the angiography is very important for diagnosis of pseudoaneurysm and to visualize the distal run-off, except in the Marcove et al. case,¹⁰ where it showed occlusion of the popliteal artery without any evidence of an aneurysm due to compression of the hematoma on the artery. None of the cases reviewed had MRI performed. This was done in our case, which

confirmed the diagnosis. Magnetic resonance angiography is not available in our hospital and it does not seem to have the diagnostic accuracy to replace conventional angiography in the lower extremities as yet.²⁴ In all instances, popliteal aneurysms, because of osteochondroma, have been false aneurysms except in one case when it was an arterial venous fistula.¹⁹

Absence of distal pulses and ischemia of the foot may occur due to emboli from the aneurysmal sac as in the case of Shah.⁶ Surgical treatment was always the treatment of choice and excision of the sac with direct repair or saphenous vein graft is the most common practice, although Gortex graft has been used,⁸ while in only one case was the artery ligated.¹⁸ Surgical treatment has been excellent and with no reports of amputation.

References

1. Bouhoustos J, Martin P. Popliteal aneurysms: a review of 116 cases. *Br J Surg* 1974;61:469-75.
2. Blair D, Vermilion MD, Shirley A, et al. A review of one hundred and forty-seven popliteal aneurysms with long-term follow-up. *Surg* 1981;90:1009-13.
3. Brailsford JF. *Radiology of bone and joints*, ed 5. Baltimore, Williams and Wilkins, 1953;241.
4. Anastasi GW, Wertheimer HM, Brown R. Popliteal aneurysm with osteochondroma of the femur. *Arch Surg* 1963;87:636-9.
5. Paul M. Aneurysm of popliteal artery from perforation by cancellous exostoses of the femur: report of a case. *J Bone Joint Surg* 1953;35B:270-1.
6. Shah PJR. Aneurysm of the popliteal artery secondary to trauma from an osteochondroma of the femur: a case report and review of the literature. *Br J Surg* 1978;65:786-8.
7. Schoene HR, Berthelsen S, Changwoo AHN. Aneurysm of the femoral artery secondary to osteochondroma. *J Bone Joint Surg* 1983;55A:847-9.
8. Hasselgren P, Eriksson B, Lukes P, Seeman T. False popliteal aneurysm caused by exostoses of the femur. *J Cardiovasc Surg* 1983;24:540-2.
9. Vallance R, Hamblen DL, Kelly IO. Vascular complications of osteochondroma. *Clin Radiol* 1985;36:639-42.
10. Marcove RC, Lindeque BG, Silane MF. Pseudoaneurysm of the popliteal artery with an unusual arteriographic presentation. *Clin Orthop Rel Res* 1988;234:142-4.
11. Harrington I, Campbell V, Valazques R, Williams T. Pseudoaneurysm of the popliteal artery as a complication of an osteochondroma. *Clin Orthop Rel Res* 1991;270:283-7.
12. Hunter JIN, Palmer JF. *The works of John Hunter, FRS. Vol 1 - London: Library of the Royal College of Surgeons of Ireland* 1835;262-6.
13. Virchow R. Ueber die Entstehung des Enchondroms und seine Beziehungen zur Endochondrosis und exostoses cortilaginea. *Monatsberichte der Koniglichen Preussischen Akademie der Wissenschaften* 1876:760.
14. Keith A. Studies on the anatomical changes which accompany certain growth disorders of the human body. *J Anat* 1920;54:101-15.
15. Hennekan RCM. Hereditary multiple exostoses. *J Med Genet* 1991;28:262-6.
16. Ferriter P, Hirschy J, Kessler H, Scott WN. Popliteal pseudoaneurysm: a case report. *J Bone Joint Surg* 1983;65A:695.
17. Denman FR, Shindler TO, Hampton J, Hanson LC. Aneurysm of the popliteal artery caused by osteochondroma of the femur. *J Bone Joint Surg* 1959;41A:1526-8.
18. Masson AF, Pullan JM. Aneurysm complicating exostoses. *Br J Surg* 1966;53:929.
19. Lesser AJ, Greenley CE. Femoral popliteal arteriovenous aneurysm caused by fractured osteochondroma of the femur. *JAMA* 1958;167:1830.
20. Woolson ST, Maloney WJ, James DR. Superficial femoral pseudoaneurysm and arterial thromboembolism caused by an osteochondroma. *J Paediatr Orthop* 1989;9:335-7.
21. Leve L, Kalideen JM. Popliteal false aneurysm complicating osteochondroma - a case report. *S Afr Med J* 1979;55:1087-8.
22. Kover JH, Schwalbe N, Levowitz BS. Popliteal aneurysm due to osteochondroma in athletic injury. *NY State J Med* 1970;12:3001-3.
23. Voutsinas S, Wynne-Davies R. The infrequency of malignant disease in diaphyseal acialis. *J Med Genet* 1983;20:345-9.
24. Borrello JA. MR angiography versus conventional x-ray angiography. *Radiol* 1993;187:615-7.