Intramuscular haemangioma adjacent to the bone surface with periosteal reaction

REPORT OF THREE CASES AND REVIEW OF THE LITERATURE

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We present three cases of intramuscular haemangioma adjacent to bone in the lower limb. All patients had local pain during the third decade. Plain radiographs showed an irregular or hypertrophic periosteal reaction on the shaft of the fibula and an intramuscular mass adjacent to the bone with inhomogeneous high signal intensity on MRI. These lesions mimic periosteal or parosteal tumours.

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Intramuscular haemangiomata are common, benign soft-tissue tumours, usually found in the limbs. When such a tumour is located adjacent to bone, a periosteal reaction may occur. It mimics a parosteal lesion, such as osteosarcoma, chondrosarcoma, ganglion or periostitis. We present three cases of histologically proven intramuscular haemangioma adjacent to bone in the lower limb with an extensive periosteal reaction, and describe the findings on MRI.

Case reports

Case 1. A 25-year-old woman presented with a 13-year history of swelling and intermittent pain in the right lower leg. There was no history suggestive of trauma or infection. On physical examination there was swelling, local heat and tenderness. All other clinical and laboratory studies were normal. A plain radiograph showed cortical thickening of the mid-shaft of the fibula with an irregular periosteal

reaction (Fig. 1a). CT demonstrated a reactive thickening of the posterolateral cortex of the fibular (Fig. 1b). No abnormality of soft tissue was apparent on either plain radiographs or CT. MRI revealed an ill-defined soft-tissue mass within peroneus brevis adjacent to the fibula, with an inhomogeneous high signal (Fig. 1c). An open biopsy was performed. At operation, vascular tissue with an ill-defined margin was seen within the muscle adjacent to the surface of the fibula. An excision of the tumour with a margin of normal muscle was performed together with a biopsy of the cortical surface of the fibula. The periosteum beneath the tumour was thickened and the cortical surface irregular. The histological findings confirmed the diagnosis of cavernous haemangioma (Fig. 1d); the cortical bone was histologically normal. The symptoms resolved after surgery and at follow-up five months later she was free from pain with no evidence of recurrences.

Case 2. A 26-year-old woman presented with a six-month history of pain in the right calf, particularly at night, with no history suggestive of trauma or infection. On physical examination, there was tenderness and local warmth laterally below the knee. A bruit was heard on auscultation of the calf. There was a full range of movement of the knee and ankle. A plain radiograph showed an irregular periosteal reaction, cortical thickening and erosion of the fibula, with a hypertrophic periosteal reaction of the lateral aspect of the mid-tibia (Fig. 2a). Axial MRI revealed a soft-tissue tumour extending from the fibular shaft towards the tibia, with an inhomogeneous low signal intensity on the T1-weighted and a high signal intensity on the T2-weighted image (Figs 2b and 2c). A bone-seeking isotope scintigram revealed a soft-tissue lesion with increased activity in the area of the tumour. The diagnosis of haemangioma was made at open biopsy. Since her pain was minimal, she received no further treatment but was reviewed regularly.

Case 3. A 20-year-old man presented with a five-month history of pain and swelling of the left lower leg after physical exercise. No swelling, warmth, pulsation or redness were found on physical examination although there was local tenderness. A plain radiograph showed erosion and cortical thickening of the distal fibula and phleboliths in the soft tissues (Fig. 3a). Coronal MRI showed a tumour around the fibula extending towards the tibia (Fig. 3b). Axial T2-weighted MRI showed an intramuscular tumour

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Fig. 1a

Fig. 1b

Fig. 1c

Fig. 1d

Case 1. Figure 1a – An AP radiograph of the right fibula showing cortical thickening and an irregular periosteal reaction over approximately 6 cm. Figure 1b – Axial CT shows the irregularity of the lateral fibular cortex. Figure 1c – Axial-gradient echo MRI (TR 550 ms; TE 22 ms) shows the soft-tissue mass with inhomogeneous high signal intensity. The intensity of the marrow is normal. Figure 1d – A photomicrograph of a resected soft-tissue tumour shows vascular channels of varying size within the muscle. The histological diagnosis was cavernous haemangioma (haematoxylin and eosin $\times 75$).

Fig. 2a

Fig. 2b

Fig. 2c

Case 2. Figure 2a – An AP radiograph of the right fibula shows cortical thickening, depression and irregular periosteal reaction. There is hypertrophic periosteal reaction in the adjacent lateral aspect of the tibia. Figures 2b and 2c – Axial MRI (T1: TR 500 ms, TE 15 ms; T2: TR 1700 ms; TE 80 ms) revealed an intramuscular tumour. Inhomogeneous low-to-intermediate signal intensity is seen on the T1-weighted image (b) and high signal intensity on the T2-weighted image (c). The intensity of the marrow is normal.

Fig. 3a

Fig. 3b

Fig. 3c

Case 3. Figure 3a – An AP radiograph of the left fibula shows osteopenia, cortical thickening (arrow) and phleboliths (arrowheads). Figure 3b – Coronal T2-weighted MRI (TR 3500 ms; TE 100 ms) revealed an ill-defined, large soft-tissue mass with high signal intensity. Figure 3c – Axial T2-weighted MRI (TR 3500 ms; TE 100 ms) revealed an intramuscular tumour within tibialis posterior and soleus involving tibialis anterior, with inhomogeneous high signal intensity (arrows). The posterior half of the marrow of the fibula showed high signal intensity (arrowhead) suggesting local bony invasion by tumour.

within tibialis posterior and soleus, which extended into the anterior compartment. The fibular marrow showed increased signal intensity with the same MRI parameters (Fig. 3c), suggesting invasion into bone by the tumour. An open biopsy was performed and histological examination revealed a cavernous haemangioma. The pain remained minimal and no further surgery was carried out.

Discussion

A haemangioma is a common, benign soft-tissue tumour, usually occurring in the skin, subcutaneous tissue or muscle. Haemangioma of bone, including periosteal haemangioma, is rare. Our cases resembled those of periosteal or parosteal tumours from the plain radiographs, but the presence of a soft-tissue mass adjacent to the bone and the characteristic signal intensity on MRI suggested the correct diagnosis.¹⁻⁴ All cases were proven histologically to be haemangiomata. Since the bone was not involved with the tumour in cases 1 and 2, and there was a large soft-tissue mass with minimal bony involvement in case 3, we believe that the classification as intramuscular haemangioma adjacent to bone with periosteal reaction is the most suitable.

Cases of haemangioma in the periosteal region have been described as periosteal,⁵⁻⁹ subperiosteal,¹⁰ surface¹¹ and surface-based,^{12,13} but there has been little information to suggest that there is a correlation between the tumour and the periosteum. Only one report presented the histology of the lesion including that of the periosteum and bone.¹⁴ The

authors described the periosteum near the soft-tissue mass as demonstrating reactive changes, with no tumour invasion, and the bone as being normal, which is similar to the findings in our case 1.

In most reported cases, the diaphysis has been affected, most commonly the fibula, tibia or ulna.

This condition mimics other periosteal or parosteal lesions such as osteosarcoma, chondrosarcoma, a ganglion and periostitis. The plain radiological findings of haemangioma in the periosteal region have been described as cortical thickening or depression,^{5-8,10-12,14-17} periosteal reaction, a soft-tissue mass and osteopenia.

MRI is useful for identifying a soft-tissue mass adjacent to bone and distinguishing intramuscular haemangiomata from other soft-tissue tumours, both benign and malignant.¹⁻⁴ Phleboliths, as seen in case 3, are another radiological finding characteristic of haemangioma. Most reported patients had pain and a palpable mass.

Some authors recommend resection *en bloc*, including the affected bone, to reduce the incidence of local recurrence.^{7,12,13} We performed surgical resection in only one of our three patients, removing the soft-tissue mass only, because the adjacent bone showed no more than reactive changes to the overlying soft-tissue mass. Since the other two cases had only mild pain, formal resection was not undertaken. Haemangiomata do not enlarge and which are associated with minor local discomfort only, merely require regular review.

In summary, a haemangioma in the periosteal region is

rare. It occurs most commonly adjacent to the long bones of the lower limb. Since these lesions mimic periosteal or parosteal tumours they are likely to be misdiagnosed. The appearance on MRI gives the most diagnostic information.

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