PSEUDOMALIGNANT HETEROTOPIC OSSIFICATION ("MYOSITIS OSSIFICANS")

RECURRENCE AFTER EXCISION WITH SUBSEQUENT RESORPTION

J. D. SPENCER, G. A. K. MISSEN

From United Medical and Dental Schools of Guy's and St. Thomas's Hospitals, London

An eight-year-old boy presented with massive pseudomalignant heterotopic ossification around the upper femur. The mass was completely excised because of severe pain, systemic illness and a flexion contracture at the hip.

Symptomatic improvement was swift, but two weeks later the mass had recurred and was even more extensive. During the subsequent 18 months of conservative management he has been free of pain and there has been progressive resorption and remodelling of the heterotopic bone. There is now no limitation of physical activity and movement at the hip is full.

A formerly healthy eight-year-old boy developed, over two months, massive swelling of the left upper thigh and a 70° flexion contracture at the hip. There was no history of significant trauma. He was brought to London for a second opinion because amputation through the hip had been proposed in his own country, where superficial biopsies had proved inconclusive. On admission to Guy's Hospital, he was febrile, dehydrated and in great pain. A smooth, hard, non-tender mass extended from the lateral aspect of the left upper thigh posteriorly into the buttock and thence into the adductor compartment.

Radiography revealed a calcific mass in the soft tissues around the upper femur (Fig. 1), while CT scans confirmed its extent and showed it to be well circumscribed; it appeared to be continuous with the greater trochanter and its inferior portion had a "water-dense" centre (Fig. 2). Chest radiography and CT scan were normal. A bone-scan revealed greatly increased uptake of isotope by the mass. Arteriography showed a mature pattern of arterial supply, derived predominantly from

the profunda femoris artery. There was neutrophil leucocytosis (9350 neutrophils/ μ l).

It was considered that the boy had pseudomalignant heterotopic ossification in soft tissues ("myositis ossificans") with pain due to stretching of muscle fibres and the development of a compartment syndrome. The mass was therefore exposed and a full-thickness biopsy of its wall taken; a bony outer shell was found enclosing a soft friable yellow centre. Microscopy confirmed the provi-

Pre-operative radiographs of the left upper femur showing the calcific mass extending medially, laterally and posteriorly around the bone.

J. D. Spencer, MRCP, MS, FRCS, Senior Lecturer/Honorary Consultant Orthopaedic Surgeon
G. A. K. Missen, DM, FRCPath, Consultant Histopathologist Emeritus
United Medical and Dental Schools of Guy's and St. Thomas's Hospitals, Guy's Campus, St. Thomas Street, London SE1 9RT, England.

Correspondence should be sent to Mr J. D. Spencer.

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Fig. 2

CT scan showing the mass in the soft tissues of the posterior left thigh, continuous with the femur. It has a radiolucent centre and a densely bony outer layer, well demarcated externally, though its surface-contour is irregular.

Fig. 3

Positive radiograph of a horizontal slice of variable thickness through the excised mass. 1, 2 and 3 – Ackerman's zones of progressive outward maturation: F – the site of femoral attachment; B – the biopsy site.

Fig. 4

Anteroposterior radiographs taken two weeks, six months and 18 months after the removal of the original mass. Massive recurrence of soft tissue ossification had taken place by two weeks, but subsequently there was progressive resorption and remodelling of bone, which developed a mature appearance.

sional diagnosis, showing three zones of progressively increasing maturation from central fibrin and granulation tissue, through a stratum of part-calcified osteoid, to the outer shell of young cancellous bone of woven type (Ackerman 1958). The entire mass was excised 10 weeks after the onset of the illness; its surface was well demarcated and its attachment to the greater trochanter easily divided. Portions of gluteus medius and of the adductors proved necrotic and were also excised.

The excised mass, after removal of an almost complete covering of soft tissue, was outwardly bony and essentially smooth; it measured 12×11×8 cm and weighed 605 g; a rough anterior bony area, 35 mm in diameter, indicated the site of detachment from the femur. Radiography of the specimen showed the progressive outward maturation (Fig. 3) and histology confirmed heterotopic ossification.

The patient recovered well with almost complete relief of pain, his flexion contracture improved to 20° and he could bear full weight. By two weeks after the operation, however, a new and larger calcific mass, largely amorphous radiologically, had developed (Fig. 4). He was otherwise well. At six months he could take part in all sports and the radiologically amorphous material had been transformed into an irregular, but sharply defined, mass of mature bone pierced by rounded lacunae. At 18 months there had been further resorption of bone. Hip movements were full and physical activity unlimited.

The pathology of heterotopic ossification of this kind, which may in its earlier stages give rise to equivocal radiological and histological appearances, is well described (Ackerman 1958; Angervall et al. 1969; Ogilvie-Harris and Fornasier 1980). Mistaken diagnoses of osteosarcoma have, on occasion, led to unnecessary amputation. A history of significant preceding trauma is

only occasionally obtained. A mass frequently develops in only one to two weeks – a useful clue, since sarcomata rarely grow so fast (Dahlin 1978). In the *fully developed* lesion, CT scanning is usually diagnostic.

Misinterpretation of adequate biopsy material is unlikely unless this consists solely of young, cellular and rapidly growing fibroblastic tissue, either from a young lesion less than three to four weeks old, or entirely from the soft centre of a lesion like the present one. There is sometimes a good case for biopsy-excision of the entire mass with its enclosing soft tissues – a course dictated in our patient by pain, systemic illness and a probable compartment syndrome.

Recurrence of the lesion following excision is rare. It perhaps reflects the activity of persistent sub-periosteal osteoblasts. In our patient, the rapidity and extent of the recurrence were remarkable. While its cause is not clear, the unusually large size of the mass and the need to divide a bony attachment were possible factors. The boy is now fit and there is radiological evidence of progressive resorption of the new bone.

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