SUBPERIOSTEAL GIANT-CELL REPARATIVE GRANULOMA

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We present a case of subperiosteal giant-cell reparative granuloma followed over six years showing the complete evolution from the early phase of subperiosteal haematoma to the end stage of an ossified haematoma. Such lesions, although they are histologically similar to true giant-cell tumours, can be distinguished by the patients’ age, their location on the diaphysis, and by their radiological and histological features.

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Subperiosteal reactive processes have been given various names. Their nomenclature is based on their location and the most impressive clinical, radiological or histological findings (Bloodgood 1910; Cone 1928; Potts 1940; Shipley 1940; Coley and Miller 1942; Present 1945; Hodgson and Frantz 1947; Lichtenstein 1950; Thompson 1954; Norman and Dorfman 1970; Spjut et al 1970; Picci et al 1986; Wold et al 1986). There has often been confusion between such lesions and true giant-cell tumours. We present a case of subperiosteal giant-cell reparative granuloma which was followed through the stages of its evolution.

CASE REPORT

A 40-year-old man presented with a two-month history of the insidious onset of dull, aching pain in the right calf with no history of injury. Physical examination revealed a deep non-mobile mass on the posterior aspect of the midshaft of the tibia. There was no vascular or neurological deficit and the previous medical history was unremarkable.

The lateral radiograph showed an oval saucer-like depression in the posterior cortex surrounded by a slight peripheral periosteal reaction (Fig. 1a). CT demonstrated erosion of the outer surface of the cortex (Fig. 1b) and MRI revealed a subperiosteal lesion the signal from which was consistent with fluid (Fig. 1c). There was no intramedullary abnormality. Bone scanning showed a focal increase in radioisotope uptake. The differential diagnosis, at this stage, included subperiosteal haematoma and subperiosteal aneurysmal bone cyst.

Incisional biopsy revealed that the periosteum was elevated over a solid haemorrhagic mass. Histological examination demonstrated a mononuclear spindle-cell stroma containing a large population of osteoclast-like multinucleated giant cells (Fig. 2). In some areas these cells were randomly and evenly distributed; in others they showed nodular clustering around haemorrhagic patches. Haemosiderin deposits and foci of early bone formation were identified. The lesion was richly vascular with areas of erythrocyte extravasation.

The large multinucleated, randomly distributed giant cells gave the overall impression of a giant-cell tumour; but the subperiosteal location in the shaft excluded that diagnosis. Moreover, the nodular clustering of giant cells around haemorrhagic areas with haemosiderin deposits and reactive bone formation is not typical of a true giant-cell tumour, and the mononuclear stromal cells were of the reactive or reparative type rather than true neoplastic cells.

These histological findings have, however, been described in giant-cell reparative granuloma (GCRG) (Lorenzo and Dorfman 1980). Subperiosteal aneurysmal bone cyst (ABC) may also show solid areas of fibrous stroma containing scattered giant cells, and both GCRG and ABC are associated with haemorrhagic processes. The brown tumour of hyperparathyroidism is microscopically indistinguishable from GCRG but the normal calcium, inorganic phosphate, and alkaline phosphatase levels in our patient made hyperparathyroidism also an unlikely diagnosis. Based on the microscopic findings of reactive fibroblastic tissue with giant cells and haemosiderin deposits, the final diagnosis of GCRG was made. Three months after the initial diagnosis the radiograph revealed periosteal ossification (Fig. 3) and two years later the lesion had matured into a 'subperiosteal ossified haematoma' (Fig. 4). The patient continued to be asymptomatic for the next six years by which time the bone showed only cortical thickening.
Figure 2a – A mononuclear spindle-cell stroma containing evenly distributed multinucleated giant cells (haematoxylin and eosin x50). Figure 2b – High-power view shows mononuclear cells, which look benign, with scattered multinucleated giant cells (haematoxylin and eosin x200).
DISCUSSION

Geschickter and Copeland (1930) reported a haemorrhagic process involving the skull and jaws, which they described as 'subperiosteal giant-cell tumor'. Jaffe (1953) described a benign non-neoplastic process associated with intraosseous haemorrhage limited to the mandible and maxilla and called it 'giant-cell reparative granuloma'. Spjut et al (1970) reported two cases of lesions in the phalanges, histologically similar to GCRG in the facial bones, and used the term 'giant-cell reaction' of the small bones of the hands. Lorenzo and Dorfman (1980) reviewed eight cases in which the bones of the hands and feet were the sites of lesions histologically identical to those described by Jaffe and Spjut. These were reported as GCRG of the short tubular bones of the hands and feet.

The presence of haemorrhage and haemosiderin deposits in the medullary cavity led to the belief that these lesions were caused by intraosseous bleeding. The occurrence of similar processes in the subperiosteal compartment has been described (Shipley 1940; Lichtenstein 1950; Thompson 1954; Steiner, Ghosh and Dorfman 1981; Dupree and Enzinger 1986) and the lesions have been given several names according to the stage of their maturation. Van Arsdale (1893) first reported a subperiosteal lesion described as 'ossifying hematoma of subperiosteal origin' in the midshaft of the humerus and Bloodgood (1910) recognised the condition as a bone cyst, due to periosteal reaction. Cone (1928) described the condition as subperiosteal haematoma resulting from trauma.

The striking appearance of the multinucleated giant cells has sometimes led to a mistaken diagnosis of true giant-cell tumour. Coley and Miller (1942) described the condition as 'atypical giant-cell tumor' and Present (1945) described a giant-cell lesion involving the diaphysis of the radius as a 'subperiosteal giant-cell tumor'. Thompson (1954) reviewed 32 reported cases of 'subperiosteal giant-cell tumor', most of them in patients who were too young to have had true giant-cell tumours. Their ages ranged from 10 to 28 years, with a definite male predominance, and there had been antecedent trauma in two-thirds. The presenting symptoms were dull, aching pain and swelling and the most common sites were the diaphyses of the humerus and tibia. Lichtenstein (1950) recommended the term 'subperiosteal ABC' to highlight the non-neoplastic nature of the condition, and he suggested that the aetiology was a vascular disturbance.

The radiological appearances vary with the stage of maturation. At first, there is a dome-shaped periosteal elevation caused by the collection of blood and the underlying cortical surface may be saucerised. Later, the periosteum begins to ossify, producing a bony shell which has been described as 'subperiosteal ossifying hematoma'. Johnson and Laurence (1935) described this stage as 'localised myositis ossificans', while Mirra (1989) preferred the term 'periostitis ossificans'. For lesions in the small bones of the hands and feet, Spjut and Dorfman

Figure 3 – The radiograph three months after presentation showed more distinct subperiosteal calcification.

Figure 4 – Two years later the lesion had matured into a fully ossified subperiosteal haematoma.
(1981) introduced the term ‘florid reactive periostitis’ to make the point that some of these lesions are associated with extensive periosteal reaction. Microscopically, they may appear aggressive and have been described as ‘pseudomalignant fibro-osseous tumours’ (Hutter et al. 1962; Dupree and Enzinger 1986).

The concept that needs to be emphasised is that all these processes are benign, non-neoplastic, and are the result of haemorrhages. Histologically, they show different features because they are at different stages of maturation. The lesions consist of granulation tissue with new collagen and neovascularisation within a non-neoplastic spindle-cell stroma. Numbers of histiocytes and giant cells surround the haemorrhagic areas, the giant cells containing multiple nuclei similar to those in the stromal cells. There may be haemosiderin deposits in the stroma and reactive osteoid and bone may also be present.

The lesions are probably due to a vascular disturbance, secondary to trauma to the nutrient artery of the bone. Subperiosteal haematomas contain potent osteoinductive agents, which cause mesenchymal cell proliferation and the formation of fibrous tissue, cartilage and bone as occurs in callus. The osteoblastic cells may provoke osteoclastic giant-cell proliferation, giving the impression of ABC or a giant-cell tumour. In addition, the haemorrhage may induce the formation of histiocytic giant cells functioning as foreign-body giant cells. In the hereditary bleeding disorders and coagulopathies, it is well known that large subperiosteal haematomas can cause sauerisation of the underlying bone and the formation of pseudotumours.

It is important to recognise the characteristic features of these lesions which differ from those of giant-cell tumours in many ways. The patient’s age, the location and the radiological and microscopic features all clearly distinguish GCRG from the true giant-cell tumour with its different prognosis and treatment. Giant-cell tumours are true neoplasms which erode the cortex and may extend beyond the bone into the soft tissues; GCRG never invades the surrounding soft tissues. Microscopically, GCRG demonstrates giant cells with cytoplasmic deposits of haemosiderin, a finding which has not been described in giant-cell tumours. These giant cells contain fewer nuclei than do those of giant-cell tumours and they cluster around haemorrhagic areas. Reactive osteoid, bone formation and fibrosis are all seen in GCRG and do not occur in giant-cell tumours.

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REFERENCES


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