Giant cell reparative granuloma of the proximal tibia: a case report

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ABSTRACT
Giant cell reparative granulomas (GCRGs) are non-neoplastic inflammatory lesions, usually of the jaw or gingiva or small bones of the hands and feet. We report one such case in the right proximal tibia of a 45-year-old man. Radiological studies showed a lytic lesion with marginal sclerosis in the epiphysis and metaphysis. After open biopsy, a preliminary diagnosis of a benign giant cell tumour was made. One month after admission, the lesion was curetted and filled with cancellous bone and hydroxyapatite. Based on the histology of the curetted lesion, the diagnosis was changed to a GCRG. The patient had an uneventful postoperative course, with no evidence of local recurrence and metastasis. He died from gastric cancer 2 years later.

Key words: giant cell tumors; granuloma, giant cell

INTRODUCTION
Giant cell reparative granulomas (GCRGs) are non-neoplastic inflammatory lesions that most frequently affect children and young adults. They were first described in 1953 as lesions reactive to intra-osseous haemorrhage in the maxilla and mandible.1 They usually occur in facial bones or the small bones of the hands and feet,2 rarely in long bones. Radiological and histological features of GCRGs may overlap those of other giant cells containing lytic lesions, such as giant cell tumours (GCTs), aneurysmal bone cysts, and the brown tumour of hyperparathyroidism.

CASE REPORT
In July 2004, a 45-year-old man presented with a 2-month history of gradually increasing dull pain of the right leg. He had no history of trauma. The tibial tuberosity was tender but not inflamed. The range of movement and stability of the knee were normal,
as were the laboratory findings. Radiography and computed tomography showed a lytic lesion with marginal sclerosis in the epiphysis and metaphysis of the right proximal tibia (Fig. 1), but there was no cortical destruction. Magnetic resonance imaging (using the muscle signal as a reference) showed a multinodular lesion that contained almost iso-high but focal low signal intensity in a T1-weighted image, iso but focal high signal intensity in a T2-weighted image, and high signal intensity in an enhanced T1-weighted image, with cystic change in the proximal part of the lesion (Fig. 2). An angiograph showed that the tumour was fed by branches of the popliteal artery (Fig. 3). Technetium 99m scintigraphy showed increased isotope uptake.

Gross examination of the lesion after open biopsy revealed friable soft tissues with a tan-grey surface and occasional haemorrhagic foci. Microscopically, the lesion consisted of fibrous stromal tissues with proliferation of spindle-shaped or ovoid-shaped cells. Multinucleated giant cells were scattered throughout the lesion. Atypism and mitosis were not seen in the stromal cells. The lesion was preliminarily diagnosed as a benign GCT.

One month after admission, the tumour was curetted and filled with cancellous bone and hydroxyapatite. The specimen showed multiple haemorrhagic areas, reactive osteoid and bone formation. Multinucleated giant cells were mostly seen around these areas; their nuclei were round, but those of the stromal cells were ovoid or spindle-shaped (Fig. 4). The stroma contained abundant collagen, many spindle cells, inflammatory mononuclear cells, haemorrhages and haemosiderin pigments, and occasional newly formed reactive osteoid or bone trabeculae rimmed by osteoblasts. Atypical mitoses were not seen. Based on these findings, the diagnosis was changed to a GCRG. The chromosomal analysis

Figure 1 (a) Radiograph shows a lytic lesion with marginal sclerosis of the right proximal tibia. (b) Computed tomographic scan reveals a lytic lesion with no destruction of the cortex.

Figure 2  Magnetic resonance images of the multinodular lesion showing (a) heterogeneous signal intensity in a T1-weighted image, (b) almost iso signal intensity in a T2-weighted image, and (c) high signal intensity in an enhanced T1-weighted image.
revealed no cytogenetic abnormalities. The patient had an uneventful postoperative course, with no evidence of local recurrence and metastasis. He died from gastric cancer 2 years later.

DISCUSSION

Up to 74% of patients with GCRGs are under 30 years of age, whereas approximately 80% of patients with GCTs are between 20 and 50 years of age. Most GCRGs are of an intramedullary origin and occur in the jaw (24%), small bones of the hands and feet (36%), craniofacial bone (9%), and vertebrae (8%) but rarely in long bones. Only one case of GCRG occurring in the tibia has been reported. Of 44 GCRG cases reported in Japan, 27 occurred in the jaw bones, 8 in the craniofacial bone, and 3 in the small bones of the hands and feet.

Radiographically, GCRGs are indistinguishable from GCTs, aneurysmal bone cysts, chondroblastomas, and metastatic carcinomas. GCRGs involving the temporal bone can be easily misdiagnosed as GCTs. GCRGs have low-iso signal intensity in both T1- and T2-weighted images, and high but low focal signal intensity in enhanced T1-weighted images.

GCRGs have a recurrence rate after curettage of 22 to 50%; eradication typically requires no more than 2 excisions. In contrast to GCTs, sarcomatous transformation or aggressive lesional spread has not been reported in GCRGs.

In patients with GCTs or GCRGs, the most common symptoms are non-specific pain and swelling. Their clinical courses are similar. Pathological examination is the diagnostic tool of choice. Microscopically, the stroma of GCRGs consists of many collagen fibres and spindle-shaped stromal cells. Multinucleated giant cells are unevenly distributed and form clusters, particularly around haemorrhagic foci. Their nuclei are round whereas those of the stromal cells are spindle-shaped. The multinucleated giant cells of GCRG contain fewer nuclei than those of GCT. Haemosiderin, haemorrhagic foci, and reactive bone formation in fibrous stroma are sometimes found in GCRGs. The osteoid or bone formation is more frequently observed in GCRGs than GCTs. In most cases of GCT, multinucleated giant cells are evenly scattered and the nuclei of giant cells and stromal cells are similar.

The aetiology of GCRGs remains unknown. The multinucleated giant cells of GCRGs are formed from monocyte-like and macrophage-like osteoclast precursors that differentiate into osteoclasts under the influence of osteoblast-like stromal cells. These phenomena are similar to those seen in GCTs. Cytogenetic abnormalities including translocations between sex chromosomes and autosomes, t(X;4)(q22;q31.3) have been reported in people with GCRGs, but in our patient a chromosomal analysis detected no aberration.
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