Destructive lytic lesions in the small bones of the hands and feet can present a number of diagnostic challenges as both reparative and neoplastic lesions occur at these sites and are similar in clinical presentation, as well as radiologic and pathological appearance. In a review of 240 cases, Ostrowski and Spjut found 203 benign and 37 malignant lesions; 89 of the benign lesions were designated as florid reactive periostitis, bizarre parosteal osteochondromatous proliferation (Nora tumour), reaction to injury, or giant cell reparative granuloma (GCRG). This distribution and frequency of reactive lesions are unique to these sites, which are frequently exposed to trauma and injury. In this series, there were 26 lesions that were giant cell rich, of which 24 were designated as GCRGs and 2 as giant cell tumours (GCTs), highlighting the rarity of GCTs at these sites. However, the authors noted that many of their GCRG cases had originally been diagnosed as GCTs. We describe a case of GCRG seen in a young man.

Case report

A 25-year-old, right-hand dominant man presented with pain in the left hand that had been present for 8 months. For the past 5 months, he had noticed steadily progressive pain and swelling over the dorsum of the hand. He was otherwise healthy. He had been placed on light duties at work.

Physical examination revealed swelling over the dorsum of the left ring metacarpal. He had full flexion of his digits but an extensor lag of 20° at the ring metacarpophalangeal joint.

A plain radiograph of the hand (Fig. 1) showed an expansile, well-defined lytic lesion in the head of the fourth metacarpal, extending into the proximal metacarpal shaft. Subsequent magnetic resonance imaging confirmed the presence of a destructive lesion in the metacarpal that contained several fluid–fluid levels (Fig. 2). Bone scanning demonstrated uptake at only the left ring finger metacarpal.

He underwent open biopsy and subsequently an en-bloc resection and reconstruction. Initially, it was hoped that the articular surface could be preserved. However, at the time of operation, there was nothing structurally significant remaining of the distal three-quarters of the metacarpal. After resection of the tumour the reconstruction was performed with iliac crest bone grafting and fascial arthroplasty. By 7 months postoperatively, he had returned to unrestricted activities. His range of motion at the metacarpophalangeal joint was 15° to 50°, and his grip strength on the third setting of the Jamar scale (Salmon Preston, Bowlingbrook, Ill.) was 48 and 32 kg for the right and left hands respectively.

The initial biopsy yielded several fragments of tissue showing a fibrous stroma with sheets of giant cells, many containing multiple nuclei with occasional mitotic figures and focal osteoid formation. No areas of cystic degeneration or hemorrhage were noted, although some foci were more suggestive of reparative granulation tissue. The findings were consistent with the radiologic diagnosis of a GCRG, although sev-
eral of the features were considered atypical, including the appearance of the giant cells and the areas of the stroma that more closely resembled a GCT.

The resected mass measured 5 × 3 × 2 cm and showed a fragile cortical shell at one edge as well as areas of cystic degeneration and hemorrhage rendering the tissue a dark red to mauve colour with a spongy consistency (Fig. 3). Sections from the solid areas showed a highly cellular lesion with a fibrous stroma and many giant cells, some containing 60 to 100 nuclei (Fig. 4). Mitotic activity was not noted in the giant cells and was maximally recorded at 2 to 3 per 10 high-power fields in the stroma. There was a zonal pattern of giant cell distribution often surrounding areas of hemorrhage, and rare giant cells were identified within vascular spaces (Fig. 5). In the cystic areas, large blood-filled spaces were evident with fewer giant cells and sparse fibrous tissue, in keeping with an aneurysmal bone cyst. Osteoid formation was present focally. Slides submitted for a second opinion yielded diagnoses of GCT from one consultant and GCRG from another. The final consensus diagnosis was GCRG, based on the zonal pattern and areas of hemorrhage with areas of osteoid formation. The aneurysmal bone cyst component was considered secondary to degenerative changes within the lesion.

Discussion

Jaffe\(^2\) in 1953 originally described GCRGs in the jaw or facial bones as non-neoplastic lesions distinct from GCTs. Ackerman and Spjut\(^3\) described 2 cases of “giant cell lesions” occurring in the hands in 1962, and in 1980, Lorenzo and Dorfman\(^4\) added the term “reparative” to this title for 8 additional cases they reported in the hands and feet.\(^4,4\) These lesions were postulated to represent a reactive process to intraosseous hemorrhage, although a history of trauma to the affected site was infrequent. The defining histologic features were a moderately to highly cellular stroma composed of spindle to oval cells, multinucleated giant cells often distributed in a zonal fashion surrounding areas of hemorrhage with areas of reactive osteoid formation.\(^4,4\)

Hemorrhagic areas that resembled aneurysmal bone cysts were noted in the 2 of the 8 cases reported by Lorenzo and Dorfman and in 6 of an additional 20 cases described later by Ratner and Dorfman.\(^5\) The association with areas of aneurysmal bone cyst, an uncommon lesion in the hands and feet was also noted in a series of 5 cases of GCRG reported by Glass and associates.\(^6\) Other cases of aneurysmal bone cyst with predominantly solid areas and multinucleated giant cells had been earlier described by Tillman and colleagues\(^7\) and later reported by others\(^8\) as the “solid variant” of aneurysmal bone cyst. All these lesions appear to be related expressions of the same pathogenic mechanism with overlapping clinical, radiologic and histologic features.

A series of 52 osteolytic lesions of the bones of the hands or feet demonstrated that the clinical and radiologic findings proved of limited diagnostic value in distinguishing GCTs and GCRGs.\(^9\) In another study of 900 GCTs from the Instituto Rizzoli, Biscaglia and associates\(^10\) found only 29 cases involving the bones of the hands and feet, confirming the rarity of this lesion at these sites. They noted the histologic overlap with GCRG in 4 cases (14%) and the presence of a secondary aneurysmal bone cyst in 7 cases (24%).

Wold and colleagues\(^11,12\) also noted that the clinical and radiologic features did not distinguish between the 2 entities, and a history of trauma was inconsistent. They found that collagenized, fibrous connective tissue with osteoid formation (100% cases), evidence of stromal hemorrhage (93%), and a zonal clustering of giant cells (73%) were the most consistent histologic features of GCRGs.

The Atlas of Tumor Pathology, fascicle on tumours of the bones and joints, also indicated that a clear-cut distinction between GCTs and GCRGs is not always possible and that, in general, GCRGs tend to have their giant cells aggregated around areas of hemorrhage in a zonal pattern and that giant cells with more than 2 dozen nuclei are uncommon.\(^13\)

In a large series of 90 GCRGs, including 33 cases from the hands and feet, osteoid was noted in 83%, and areas resembling an aneurysmal bone cyst were present in 29%. None of the lesions showed malignant transformation and none was associated with pulmonary metastases.\(^14\) Of these 33 lesions, 18% recurred\(^13\) compared with 39% in the Mayo Clinic series\(^15\) and 36% in the Instituto Rizzoli series,\(^16\) although a higher rate of 50% was reported in a smaller study of 8 patients by Lorenzo and Dorfman.\(^4\) Recurrence rates of GCTs treated by curettage and grafting have historically been reported as high as 40% to 60% although newer surgical treatments are associated with lower rates of recurrence (2%–25%).\(^17\) Thus, the recurrence rate cannot be used to differentiate between GCTs and GCRGs.

Conclusions

The distinction between GCTs and
Giant cell granuloma of the hand

Giant cell granuloma (GCG) is important as giant cell tumors (GCT) carry a small but real risk of metastasis, which is not the case with GCRGs. Although they cannot be reliably distinguished from the clinical or radiologic features, the histologic findings in most cases will provide the correct diagnosis. However, a small proportion of GCTs especially those associated with a pathologic fracture may show the identical histologic findings of a GCRG, so close clinical follow-up is warranted in those circumstances. This degree of diagnostic difficulty is illustrated by our case.

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References