

Double localization of Giant Cell Reparative Granuloma of the bone in the same finger

Differential diagnosis and treatment



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Double localization of Giant Cell Reparative Granuloma of the bone in the same finger. Differential diagnosis and treatment

AIM: This article reports a case of GCRG seen in a young man with a rare double localization in the same finger and illustrates the degree of diagnostic difficulty and treatment.

MATERIAL OF STUDY: A 16-year-old man presented with dull pain and swelling of the left middle finger. Plain radiography of the hand showed expansive and lucent lytic lesions with circumferential cortical destruction in the small bones of the proximal and middle phalanges. He underwent en-bloc resection and reconstruction.

RESULTS: Histologic findings were consistent with the radiologic diagnosis of GCRG, although several of the features were considered atypical, including the appearance of the giant cells and the areas of the stroma that more closely resembled a giant cell tumor (GCT). At 6 months postoperatively, the morphologic recovery was complete and he had returned to unrestricted activities. His range of motion at the involved joint was the almost completely recovered, and his grip strength on the third setting of the Jamar scale was 48 and 42 kg for the right and left hands respectively.

CONCLUSIONS: GCRG is a rare intraosseous lesion that must be considered in the differential diagnosis of hand and foot lesions, such as giant cell tumor, brown tumor, giant-cell-rich osteosarcoma.

KEY WORDS: Giant-cell reparative granuloma, Hand, Giant cell tumor

Introduction

Giant-cell reparative granuloma (GCRG) is a rare, non-neoplastic intraosseous lesion associated with subperiosteal hemorrhage, first described by Jaffe¹ in 1953 believed to be limited to the mandible and maxilla. Since then, GCRG have been reported in other skeletal bones. The lesion most commonly occurs in the small tubular

bones of the hands and feet². Ackerman and Spjut³ described 2 cases of "giant cell lesions" occurring in the hands in 1962, and in 1980, Lorenzo and Dorfman⁴ added the term "reparative" to this title for 8 additional cases they reported in the hands and feet. These lesions were postulated to represent a reactive process to intraosseous hemorrhage, although a history of trauma to the affected site was infrequent.

We describe a case of GCRG seen in a young man with a rare double localization in the same finger and illustrate the degree of diagnostic difficulty and treatment.

Case report

A 16-year-old man presented with dull pain and swelling of the left middle finger. Plain radiography of the hand showed expansive and lucent lytic lesions with circum-

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ferential cortical destruction in the small bones of the proximal (Fig. 1A, upper arrow) and middle (Fig. 1A, lower arrow) phalanges. The patient's medical history did not include previous history of hand trauma. The open surgery highlighted the presence of bone destructive lesions in the phalanges of the middle finger that contained fluid and showed a fragile cortical shell as well as areas of cystic degeneration and hemorrhage rendering the tissue a dark red to mauve color with a spongy consistency (Fig. 1B, 1C). He underwent en-bloc resection and reconstruction (Fig. 2). The key points of the surgery were: Z incision, approach trans-tendon, curettage, hydroxyapatite for graft, stabilization by Kirschner-wire, reeducation motion.

Histology showed fragments of tissue richly vascularized with mononuclear spindle-cell stroma and scattered osteoclast-like multinucleated giant cells and histiocytes clustered around hemorrhagic patches and reactive osteoid formation. Histologic findings were consistent with the radiologic diagnosis of GCRG, although several of the features were considered atypical, including the appearance of the giant cells and the areas of the stroma that more closely resembled a giant cell tumor (GCT). At 6 months postoperatively, the morphologic recovery was complete (Fig. 3A) and he had returned to unrestricted activities. His range of motion at the involved joint was the almost completely recovered

(Fig. 3B), and his grip strength on the third setting of the Jamar scale (Salmon Preston, Bowlingbrook, Illinois) was 48 and 42 kg for the right and left hands respectively.

Discussion and Commentary

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 appearance⁵. Therefore, GCRG must be considered in the differential diagnosis of hand and foot lesions, such as giant cell tumor, brown tumor, giant-cell-rich osteosarcoma, aneurismal bone cyst. Particularly, 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 giant cell tumor (GCT) and GCRG⁶. Biscaglia et al. analyzing 900 GCT found only 29 cases involving the bones of the hands and feet, confirming the rarity of this lesion at these sites and highlighted the histologic overlap with GCRG in 4 cases (14%) and the presence of a secondary aneurismal bone cyst in 7 cases (24%)⁷. Moreover, Wold and colleagues noted that the clinical and radiologic features did not distinguish between the 2 entities, and a history of trau-

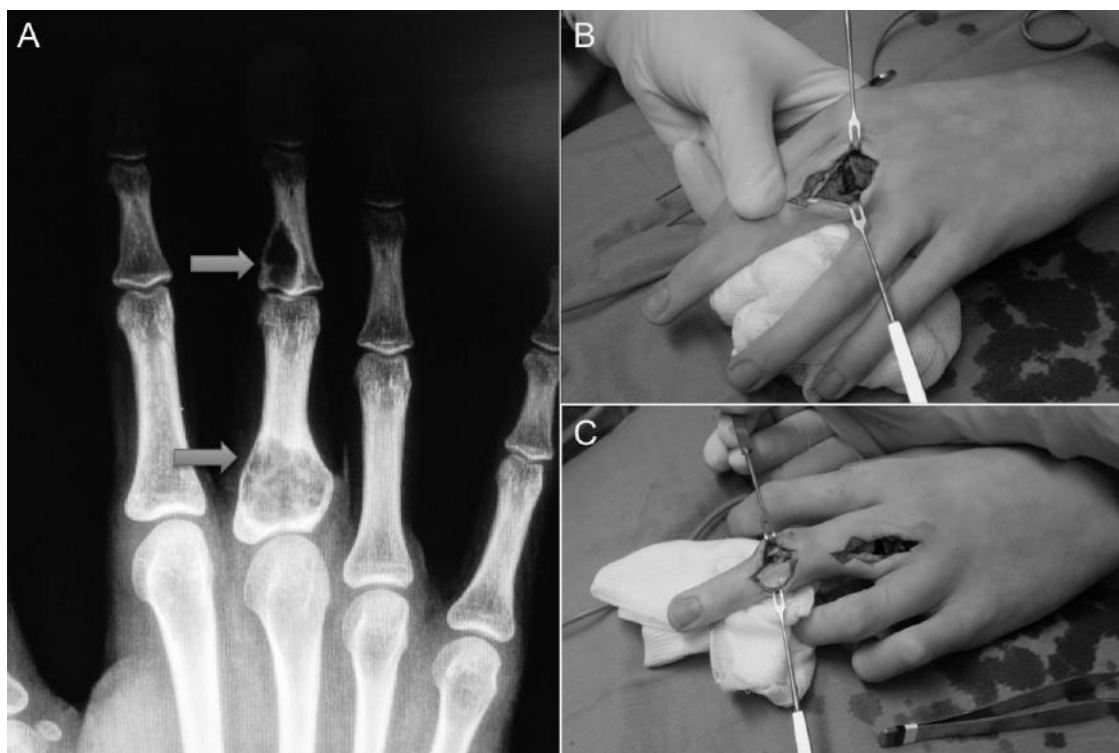
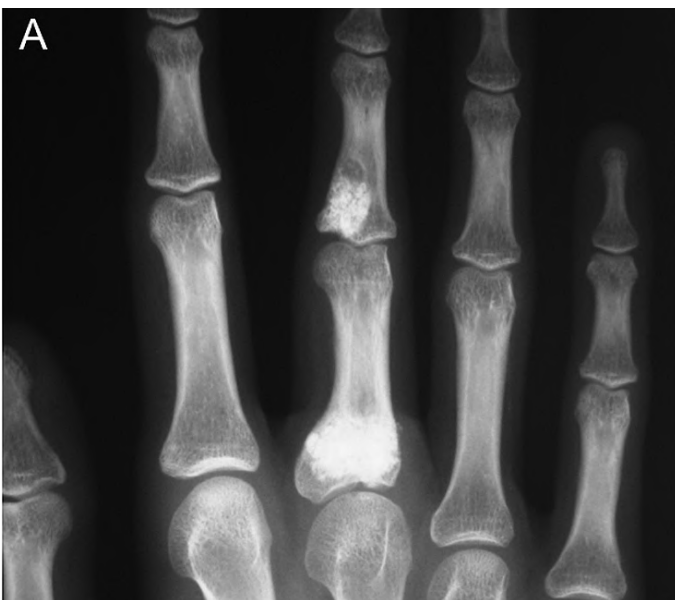


Fig. 1: A) Plain radiography shows the double localization of GCRG of the bone in the same finger of the left hand with lytic lesions and circumferential cortical destruction in the small bones of the proximal (upper arrow) and middle (lower arrow) phalanges. B) and C) Open surgery highlighted the presence of bone destructive lesions with a fragile cortical shell as well as areas of cystic degeneration and hemorrhage rendering the tissue a dark red to mauve color with a spongy consistency.



Fig. 2: Reconstruction and stabilization by Kirschner.

Fig. 3: A) Complete morphologic recovery and B) good range motion at 6 months postoperatively.



ma 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 GCRG⁸. The Atlas of Tumor Pathology also indicated that a clear-cut distinction between GCT and GCRG is not always possible and that, in general, GCRG 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 uncommon⁹. Recurrence rates of GCT 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%)^{10,11}. The distinction between GCT and GCRG is important as GCTs carry a small but real risk of metastasis, which is not the case with GCRG. 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 circumstances⁵.

So, giant cell-rich lesions of bone, including GCT of bone, GCRG, and aneurysmal bone cyst (ABC), may have overlapping clinical, radiologic, and histopathologic features. Gleason et al. analyzing one previously published karyotype of a GCRG, which revealed a reciprocal translocation, t(X;4)(q22;q31.3), reported 3 cases of giant cell-rich bone lesions with novel karyotypes: one lesion located in the first metacarpal, a typical location for GCRG, was histologically consistent with a giant cell tumor and showed the following karyotype [46,XX,inv(2)(p13q21),t(inv2;11)(q21;q13)]; the second lesion, also a giant cell tumor of bone, in the sacrum showed the following karyotype [46,XX,r(9)(p24q34)[cp7]/46,r(16)(p13.3q24)[cp10]/46,XX]. The third lesion, a hard palate

mass, had the histopathologic features of a GCRG and a karyotype showing a reciprocal translocation, 46,XY,t(2;10)(q23;q24). These findings suggest that at least a subset of GCRGs may be neoplastic and that these lesions differ cytogenetically from classic giant cell tumors of bone or solid ABC, although the latter entity is otherwise indistinguishable from reparative granuloma¹².

Surely, further cytogenetic characterization of giant cell-rich bone lesions may improve the utility of karyotyping as a tool in their differential diagnosis and may shed light on the pathogenetic relationship between these lesions.

Riassunto

Il Granuloma riparativo a cellule giganti (GCRG) è una rara lesione intraossea non neoplastica associata ad emorragia subperiostale descritto per la prima volta in letteratura da Jaffe nel 1953 come lesione limitata alla mandibola ed alla mascella. Studi successivi hanno dimostrato che si verifica più comunemente nelle piccole ossa tubolari delle mani e dei piedi. Si ritiene che la patogenesi sia legata ad un processo reattivo di emorragia intraossea, anche se una storia di trauma per il sito interessato è infrequente. L'importanza del GCRG è data dalla difficoltà di fare diagnosi differenziale con patologie ossee di natura neoplastica della mano e del piede, in particolare il tumore a cellule giganti e l'osteosarcoma a cellule giganti. Infatti, tutte le lesioni distruttive litiche delle piccole ossa, sia che siano reattive sia che siano neoplastiche, hanno sovrapponibile presentazione clinica, quadro radiologico e aspetto patologico. Pertanto non di rado ci si può trovare di fronte ad una vera e propria sfida diagnostica. Questo articolo riporta il caso di un giovane uomo con una rarissima duplice localizzazione a livello dello stesso dito della mano sinistra e ne illustra il grado di difficoltà diagnostica ed il trattamento basato su resezione en-bloc e ricostruzione mediante approccio trans-tendineo, curettage, innesto di idrossiapatite, stabilizzazione con fili di Kirschner, rieducazione motoria. Sono inoltre discussi i risultati ottenuti e la neces-

sità ulteriore di caratterizzazione citogenetica come ulteriore strumento di diagnosi differenziale.

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