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Multicentric osteolytic lesion of the middle finger of the hand. Case report

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SUMMARY: **Multicentric osteolytic lesion of the middle, finger of the hand. Case report.**

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Giant cell-rich osteolytic lesions may have overlapping clinical, radiologic, and histopathologic features, with an important degree of difficulty of diagnosis and treatment. We report a case of double osteolytic lesion at the middle-finger in a young man without previous history of hand trauma. He underwent en-bloc resection of the bone lesions and reconstruction by graft of hydroxyapatite, resulting in a good morpho-functional result. Histological diagnosis was giant cell reparative granuloma (GCRG), although several 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.

GCRG is a benign rare intraosseous lesion and the true nature is controversial and unknown. The theories are that it could be a reactive lesion, a developmental anomaly or a benign neoplasm. It appears as an osteolytic lesion that must be considered in the differential diagnosis of other "critical" bone lesions similar in clinical, as well as radiologic and pathological appearance. Further characterization studies are helpful and necessary for the proper management.

RIASSUNTO: **Lesione osteolitica multicentrica del dito medio della mano.**

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*Le lesioni osteolitiche a cellule giganti possono avere sovrapposizioni cliniche, radiologiche e istopatologiche, con un alto grado di difficoltà di diagnosi e trattamento. Riportiamo il caso di un giovane con due lesioni osteolitiche non traumatiche del dito medio. Il paziente è stato sottoposto a resezione en-bloc delle lesioni e ricostruzione con innesto di idrossiapatite, con un buon risultato morfo-funzionale. La diagnosi istologica è stata di granuloma riparativo a cellule giganti (*giant cell reparative granuloma, *GCRG), anche se molte delle caratteristiche risultavano atipiche, tra cui l'aspetto delle cellule giganti e dello stroma che assomigliava piuttosto a quelli di un tumore a cellule giganti.*

Il GCRG è una rara lesione intraossea benigna la cui natura rimane ad oggi sconosciuta. Si ipotizza che si tratti di una lesione reattiva, di un'anomalia di sviluppo o di una neoplasia benigna. Appare come lesione osteolitica che va considerata in diagnosi differenziale con altre lesioni ossee "critiche" simili nella presentazione clinica così come nell'aspetto radiologico e patologico. Ulteriori studi di caratterizzazione sono utili e necessari per un corretto trattamento.

KEY WORDS: Hand - Osteolytic lesions - Giant cell reparative granuloma - Surgery.
Mano - Lesioni osteolitiche - Granuloma riparativo a cellule giganti - Chirurgia.

Introduction

The giant cell-rich osteolytic lesions including giant cell-rich osteosarcoma (GCRO), giant cell tumor (GCT) of bone, aneurysmal bone cyst (ABC), and giant cell reparative granuloma (GCRG), may have overlapping clinical, radiologic, and histopathologic features. This leads to an important degree of difficulty of diagnosis and treatment (1, 2).

We report a case of osteolytic double lesion at the middle-finger observed in a young man. This double localization at level of the same finger was never reported in literature.

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 lytic lesions with circumferential cortical destruction in the small bones of the proximal and middle phalanges (Fig. 1 A). The patient's medical history did not include hand trauma.

The open surgery highlighted the presence of bone destructive lesions in the phalanges of the middle finger that contained fluid (Fig. 1 B). We performed *en-bloc* resection and reconstruction by graft of

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Fig. 1 - Double osteolytic lesion of the middle finger of the hand. A) plain radiography showed expansive and lytic lesions with circumferential cortical destruction in the small bones of the proximal (upper arrow) and middle (lower arrow) phalanges of the middlefinger. B) open surgery highlighted the presence of bone destructive lesions containing fluid in the phalanges of the middle-finger. C) Six-months follow-up with morphologic recovery and absence of pathological lesions.

hydroxyapatite.

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. Histological diagnosis was giant cell reparative granuloma (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).

Surgery resulted in good morpho-functional outcome (Fig. 1 C).

Discussion

Destructive lytic lesions in the small bones of the hand can present a number of diagnostic challenges as both reparative and neoplastic lesions occur at these sites. Wold et al. noted that the clinical and radiologic features did not distinguish between the two entities, and a history of trauma was inconsistent. They found that collagenized, fibrous connective tissue with osteoid formation, stromal hemorrhage, and zonal clustering of giant cells were the most consistent histological features of GCRG, but state that a clear-cut distinction between GCT and GCRG is not always possible (3). Nevertheless, the distinction between GCT and GCRG is important as GCT carry a small but real risk of metastasis. Although they cannot be reliably distinguished from the clinical or radiologic features, the histological findings in most cases will provide the correct diagnosis. However, a small proportion of GCT especially those associated with a pathologic fracture may show the identical histological findings of a GCRG, so a close clinical follow-up is a must in those circumstances (Fig. 1 C).

In the future a substantial help to simplify the approach to these lesions is developing with the cytogenetic. Gleason et al. analyzing the karyotype of cases of giant-cell-rich bone lesions, suggested that at least a subset of GCRG may be neoplastic and that these lesions differ cytogenetically from classic GCT of bone or solid ABC, although the latter entity is otherwise not distinguishable from reparative granuloma (1). 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.

One understands even further the importance of these scientific insights, when GCRG is confused with GCRO (a rare subtype of osteosarcoma) that requiring more aggressive surgery and adjuvant therapies. Recently, Fu et al. had a histological diagnosis of GCRG from a biopsy at level of a mandibular mass but after segmental mandibulectomy the postoperative histological diagnosis showed GCRO (4).

GCRG is a benign rare intraosseous lesion that most commonly occurs in the small tubular bones of the hands and feet (2), first described by Jaffe (5) in 1953 believed to be limited to the mandible and maxilla. Since then, GCRG have been reported in other skeletal bones. The true nature of this lesion is controversial and remains unknown; the three competing theories are that it could be a reactive lesion, a developmental anomaly or a benign neoplasm. Furthermore, the actual aetiology of GCG is still unclear, although inflammation, haemorrhage and local trauma have all been suggested; it has also been hypothesized that GCG may have a genetic aetiology (6). It appears as a destructive osteolytic lesion that must be considered in the differential diagnosis of other critical bone lesions that are similar in clinical presentation, as well as radiologic and pathological appearance (2, 7-9). Jadu et al. pose the question of the role of radiology in predicting the degree of aggressive behaviour of these lesions before surgery, suggesting a role of the CT characteristics (6). However, there are many efforts to be made to simplify the differential diagnosis, which is important for good medical and surgical practice. Certainly, further characterization studies are helpful and necessary for the proper management.

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