Radial localization of a giant cell tumor: Case report and review of the literature

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Abstract

Introduction: Giant cell tumors (GCT) of bone are rare borderline mesenchymal tumors of local malignancy. They affect the metaphyso-epiphyseal regions of the long bones as well as the axial skeleton. Its localization at the radius is little described in the literature and its main treatment remains carcinological surgery. The aim of this case report is to review the epidemiological, diagnostic and therapeutic aspects of this rare tumor entity.

Observation: A 52-year-old, right-handed female patient presented for consultation in January 2020 with the appearance of a mass on the back of her right hand, painful and gradually increasing in size. X-rays showed a lytic and blowing lesion of the distal third of the radius as well as the carpal bones with thickening and infiltration of the peri-articular soft tissues. A CT scan was then performed to assess the extent of the lesion, particularly in terms of the radial head and the joint. No secondary localization was found. She underwent three incomplete surgeries with local relapse each time. Histological examination of the resection specimen from the last surgery showed a giant cell tumor, with a deep tumor border. She was referred to radiotherapy consultation to discuss this adjuvant therapeutic modality, but before the multidisciplinary decision was taken, she had a local recurrence, for which a carcinological amputation was decided in agreement with the patient.

Conclusion: Distal radial localization of giant cell tumors is rare. Positive diagnosis remains histological, and treatment consists of carcinological surgery to avoid tumor recurrence.

Keywords: Giant cell tumor; Radius; Treatment; Radiotherapy.

1. Introduction

Giant cell tumors are rare mesenchymal tumors classified as intermediate, i.e. neither completely benign nor definitely malignant, due to their frequent local recurrence and the possibility of very rare pulmonary metastases. They affect the metaphyseal-epiphyseal regions of long bones, as well as the axial skeleton [1]. On imaging, plain films are characteristic enough to suggest the diagnosis in typical forms [2]. Treatment is mainly surgical, either by extensive curettage (although the risk of recurrence is significant, at around 30%), or by en bloc resection of the affected bone segment with reconstruction, or by amputation, which must comply with the rules of carcinological surgery in the same way as resection [3]. Radiotherapy is used as adjuvant treatment for axial localization or certain iterative local relapses. Its indication, since the advent of Denosumab, should be discussed anecdotally [4]. The aim of this case report is to review the epidemiological, diagnostic and therapeutic aspects of this rare tumor entity.
2. Observation

A 52-year-old, right-handed female patient presented for consultation in January 2020 with the appearance of a painful, progressively enlarging mass on the back of her right hand, accompanied by limited amplitude of the wrist joint. X-rays taken during the initial consultation showed a lytic, blowing lesion of the distal third of the radius and the carpal bones, with thickening and infiltration of the periarticular soft tissues (figure 1). A CT scan was then performed to assess the lesion, with particular emphasis on the radial head and joint. The extension study showed no secondary localization. She underwent two incomplete surgeries, each time with local relapse. The last conservative surgery was performed in September 2023. Histological examination of the resection specimen showed a giant cell tumor, with a deep tumor border. She was seen in radiotherapy consultation to discuss this adjuvant treatment modality, but before the multidisciplinary decision was taken, she had a local revision with swelling and an inflammatory appearance of the wrist and hand (figure 2). In agreement with the patient, it was decided to propose amputation, as conservative treatment was no longer possible.

Figure 1 X-ray of the right hand showing a tumour of the distal end of the radius

Figure 2 Local recurrence with swelling and inflammation of the wrist and hand

3. Discussion

The giant cell tumor (GCT) of bone was first described by Astley Cooper in 1818 [5]. But it was in 1940 that Jeff and Lichtenstein distinguished giant cell tumors as a separate entity from other bone tumors. A benign but locally aggressive tumor, they account for less than 5% of primary bone tumors [3]. They have a slight female predominance [6] and are generally located on the distal femur or proximal tibia. The distal end of the radius is not a frequent site, and only a handful of cases have been reported in the literature [7]. The usual clinical presentation is pain and swelling in relation to the extent of bone destruction. A pathological fracture sometimes reveals the lesion [8]. Radiographs show a lytic, often eccentric lesion with cortical extension. Histologically, they are characterized by two predominant cell types: large numbers of multinucleated osteoclastic giant cells and fibroblastic stromal cells [9].

Histological sections may show foci of necrosis. Three histological grades of aggressiveness (I to III) have been described: mildly aggressive benign, aggressive benign and malignant. The differential diagnosis is with aneurysmal cyst, osteoblastoma or bone metastasis [9]. The literature suggests the possibility of pulmonary metastatic extension, and although this is rare, it is justified to carry out an extension work-up. Our patient’s distant work-up showed no secondary localization. Sienerberock et al [10] noted in their series the occurrence of pulmonary metastases with an average delay of four years, often associated with local recurrence of the giant cell tumor at its initial site.

The main treatment for giant cell tumors of the extremities is surgical excision with monobloc resection and reconstruction. Ideally, this surgery should be performed in an expert center and should be carcinological. After complete resection, local control is achieved in 85-90% of cases [11], but incomplete resection is frequently associated
with tumor recurrence in up to 50% of cases. Adjuvant systemic treatment options are limited, although progress appears to be being made thanks to a better understanding of the molecular mechanisms involved in the development of giant cell tumors. As they are rich in stromal cells that express RANKL, a key mediator of osteoclast activation [12], there has been growing interest in monoclonal antibodies against RANKL, such as denosumab. A pilot study of 37 patients showed an 86% response rate and functional improvements, including pain reduction in 84% of patients with giant cell tumors treated with denosumab [12].

Radiotherapy is an alternative treatment for patients with axially located giant cell tumors not amenable to complete resection, although based on small patient series collected over long periods, with wide variations in fractionation, total dose and radiotherapy techniques. [13 - 15]. The results of the 2015 metaanalysis by Yifei Ma et al on giant cell tumors of the spine including 13 studies comprising 42 patients who received radiotherapy with doses ranging from 21 to 80 Gy suggests a response rate of 100%, a 97.6% OS, a 1-year local control (LC) rate of 85.4%, a 2-year LC rate of 80.2% and an overall LC of 79% [16].

4. Conclusion
Distal radial localization of giant cell tumors is rare. It is a borderline tumor with local malignancy. The radiological appearance suggests a benign tumor. The positive diagnosis is histological, and treatment is mainly surgical: monobloc resection with reconstruction and, in extreme cases, amputation in accordance with carcinological rule.

Compliance with ethical standards

Disclosure of Conflict of interest
The authors have declared no conflicts of interest

Statement of ethical approval
The present research work does not contain any studies performed on animals/humans subjects by any of the authors.

Statement of informed consent
Informed consent was obtained from all individual participants included in the study.

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