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METACHRONOUS MULTICENTRIC GIANT CELL TUMOR OF THE UPPER LIMB

ÜST EKSTREMİTENİN FARKLI ZAMANLI ÇOK MERKEZLİ DEV HÜCRELİ TÜMÖRÜ

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ABSTRACT

Metachronous multicentric giant cell tumor (GCT) of bone is a rare entity. The high recurrence rate after curettage may mean that more aggressive surgical management is mandatory, including en-bloc resection when indicated. We report a young woman presenting with metachronous recurrent benign GCT located at the right proximal humerus and a second lesion at the ipsilateral distal radial metaphysis. The case was successfully treated with an aggressive surgical approach (en-bloc resection).

Key words: Giant cell tumor, bone, multicentric, en-bloc resection

ÖZET

Kemiğin farklı zamanlı çok merkezli dev hücreli tümörü (DHT) nadir bir durumdur. Küretajdan sonra yüksek tekrarlama oranı, endikasyon olduğunda en-blok rezeksiyonu da içeren çok daha agresif cerrahi uygulamalar zorunludur anlamına gelebilir. Biz sağ humerus üst uçta farklı zamanlı tekrarlayan iyi huylu DHT ile birlikte ikinci lezyonu aynı taraf radius distal metafizde yerleşik olan genç bir kadını sunduk. Hasta agresif cerrahi yaklaşımla (en-blok rezeksiyon) başarılı bir şekilde tedavi edildi.

Anahtar kelimeler: Dev hücreli tümör, kemik, çok merkezli, en-blok rezeksiyon

INTRODUCTION

Giant-cell tumor (GCT) of bone was first described by Cooper and Travers in 1818. It consists approximately 4%-5% of all primary bone tumors (2). In its most common presentation, GCT is a solitary neoplasm occurring in the epimetaphysis of a long bone in a mature young adult (4, 9). The presence of more than one primary GCT in the same patient is very rare. They represent less than 1% of all GCTs (2, 7, 8). The mechanism by which GCT involves multiple locations is unknown. Multicentric GCT, in contrast to unifocal GCT, has a tendency to involve the hands, feet, and metaphysis/diaphysis of long bones and to occur in a slightly younger population (8). Less than 60 cases of multicentric GCT have been reported in the literature (2, 3, 7). Most multicentric GCT are synchronous, occurring within a poorly defined time of the initial tumor (3). Sixty-eight percent of cases of multicentric GCT occurs within a duration of shorter than 4-years disease-free interval (3).

We present a case of metachronous multicentric and recurrent benign GCT of the upper limb in a young woman successfully treated with an aggressive surgical approach.

CASE

An 18 year- old female had undergone curettage and bone grafting for a destructive lesion at the right proximal humerus in another centre. Previous plain radiographs demonstrated a large lytic lesion. Histological examination of the biopsy materials had showed benign GCT. She did well until June 1996 (17 months after surgery) when she had a local recurrence of the tumor (Figure 1A). A triple-phase bone scan of the entire body showed the known solitary lesion. En-bloc resection of the proximal part of the humerus was carried out with reconstruction by custom made replacement arthroplasty (Figure 1B).

On routine follow-up 4.5 years after the last operation, a radiolucent lesion was detected at the right distal radial metaphysis (Figure 1A). Computed tomography scans showed a well circumscribed, subarticular radiolucent lesion involving the distal radial physis. Examination of a biopsy speci-

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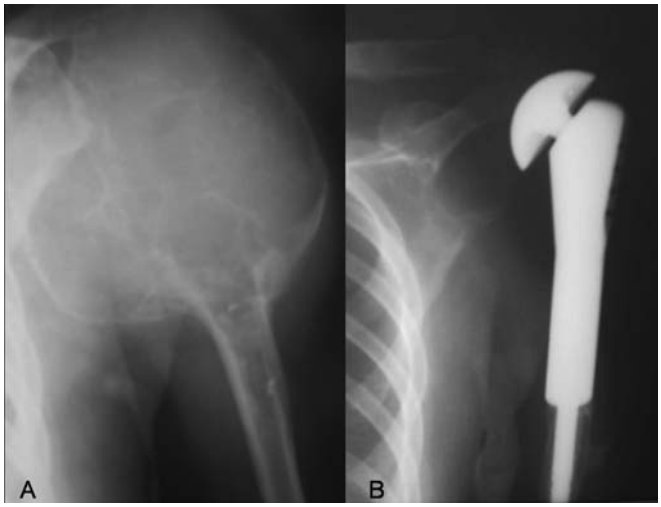

Figure 1. (A) Antero-posterior radiograph after curettage and bone grafting showing marked destruction and loss of cortex in the proximal part of the right humerus.
(B) Postoperative antero-posterior radiograph of the right shoulder after resection and reconstruction with hemiarthroplasty.

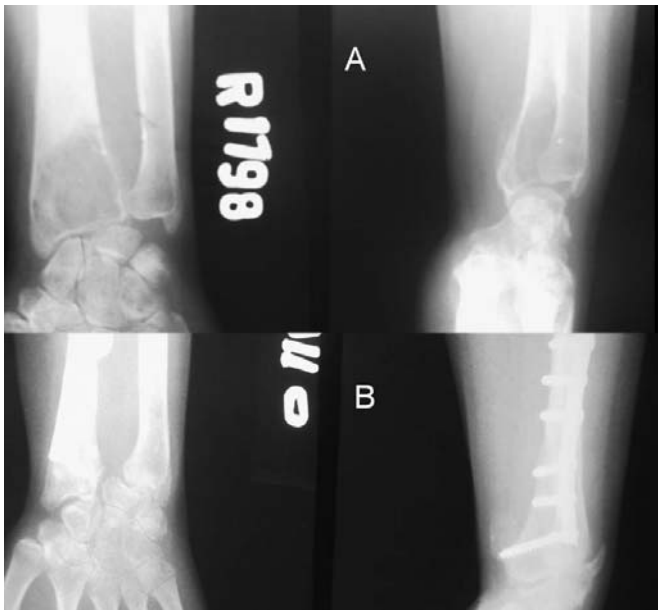

Figure 2. (A) Large lytic lesion in the distal part of the radius which extends almost to the joint surface. (B) Antero-posterior and lateral radiographs of the right wrist show successful treatment after en-bloc resection and free fibular graft reconstruction.

men confirmed another benign GCT. The lesion was treated by en-bloc resection of the distal radius and reconstruction with a free nonvascularized fibular head autograft (Figure 2B). During this time, results of blood chemistry studies, including levels of calcium, phosphorus, and alkaline phosphatase remained normal, and there was no roentgenographic evidence of systemic bone disease, such as hyperparathyroidism.

Six years after shoulder arthroplasty, we observed skin necrosis and soft tissue defect at the shoulder. The defect was reconstructed with a rotationplasty.

At the most recent follow-up of 3.5 years since the latest surgery, there was no recurrence of both tumors and a satisfactory functional outcome was achieved. Her most recent bone scan revealed uptake in all the locations of known previous disease, without evidence of additional lesions. Metastatic disease to the lungs was not observed on serial radiographs of her chest.

DISCUSSION

The presence of more than one primary GCT in the same patient is rare (2, 7, 8). Multiple bone involvement made therapeutic assessment difficult (1). Other primary osseous lesions also may have multicentric presentation (fibrosarcoma, osteogenic sarcoma, chondroblastoma, hyperparathyroidism, infection, eosinophilic granuloma, Paget's disease, multiple and metastatic carsinoma) (9). It must be differentiated from other multiple lesions that present certain similarities for the most part roentgenotraphically, by combined clinical, roentgenographic, chemical and histological studies (5, 6, 9).

In our patient, multifocal GCT presented no special problems in diagnosis. Each lesion exhibited the typical histopathological pattern of GCT, while most of the lesions also had roentgenographic features consistent with GCT. The patient has been followed-up by periodic roentgenograms and bone scans; no additional lesions have been detected. The bone scan has been useful, particularly for diagnosis of a recurrence (6), but it may not be reasonable to recommend routine use of the bone scan as a follow up test for patients with a solitary GCT.

Recurrence of solitary type GCT of bone has been reported to be expected in 10 to 60% of cases; the rate depends on the type of treatment, the anatomic location, and the cortical integrity of the involved bone (5, 10). Repeated curettage and grafting when applied to recurrent lesions was uniformly unsuccessful, necessitating wide local amputation. Multiple lesions tend to exhibit the same aggressive clinical behaviour as a solitary GCT. The multicentric variety is often of a higher stage at diagnosis and is more often associated with a pathological fracture than the unifocal tumor (4). En-bloc resection is the most successful surgical technique for treating both multicentric and solitary lesions (5). The recurrence in our case was treated by en-bloc resection and further recurrence was not evident.

Inconclusion, if multiple radiolucent lesions are present preoperatively, other diagnoses should be ruled out at first. Preoperative evaluation should include clinical, roentgenographic, chemical and histological studies. The high recurrence rate after curettage may mean that more aggressive surgical management is mandatory, including en-bloc resection when indicated.

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