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CASE REPORT / OLGU SUNUMU

Giant cell tumor of the distal tibia: Report of a rare case

Distal tibianın dev hücreli tümörü: Nadir bir olgu sunumu

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ABSTRACT

Giant cell tumor of bone is an aggressive lesion, although benign. Foot and ankle involvement is rare. Herein, we presented a case of a 26-year-old woman complaining of increasing pain and swelling of the left ankle followed by limitation of joint motion. Imaging was consistent with the diagnosis of giant cell tumor of bone, which was confirmed by open biopsy. The lesion was treated with segmental en-bloc resection and ankle arthrodesis with good functional outcome. We also discuss clinical, radiological, and therapeutic characteristics of this pathology with the light of the literature. *J Clin Exp Invest 2013; 4 (4): 512-516*

Key words: giant cell tumor of bone; tibia; ankle; arthrodesis; bone transplantation

ÖZET

Kemiğin dev hücreli tümörleri iyi huylu olmalarına rağmen agresif lezyonlardır. Ayak ve ayak bileği tutulumu ise nadirdir. Biz burada, sol ayakta şişlik ve ağrı artışı şikâyeti nedeniyle eklem hareketi kısıtlanan 26 yaşında bayan hastayı sunduk. Görüntüleme yöntemleri kemikte dev hücreli tumor tanısı koydurdu ve bu tanı açık biyopsi ile doğrulandı. Lezyon segmental en-blok rezeksiyon ve bilek artrodezi yapılarak tedavi edildi ve iyi fonksiyonel sonuç alındı. Biz aynı zamanda yazımızda bu patolojinin klinik, radyolojik ve tedavi karakteristiklerini literatür bilgileri ışığında tartıştık.

Anahtar kelimeler: Kemiğin dev hücreli tümörü, tibia, ayak bileği, artrodez, kemik transplantasyonu

INTRODUCTION

Giant cell tumor of bone (GCTB) is an osseous neoplasm that is histologically benign but clinically shows local aggression and a high rate of recurrence [1-5]. It is thought to originate at the metaphyseo-epiphyseal junction and may extend into the metaphysis [1-3,6]. Numerous terms, including myeloid sarcoma, tumor of myeloplaxus, osteoblastoclastoma, and osteoclastoma have been used to depict GCTB [2]. It accounts for about 5% of all primary bone tumors in adults and predominantly occurs in the third and fourth decades of life with a slight predilection for females [1,3,5]. Involvement of the foot and ankle is rare and comprises less than 4% of all GCTBs [7]. Lesions with this localization are known to be unpredictable in their behaviour [8]. GCTB of hand and foot are more aggressive and aggressive treatment is recommended [9,10].

Herein, we present a rare case of GCTB of the distal tibia treated with segmental en-bloc resection and ankle arthrodesis. We also review the pathologic features, clinical manifestations, radiological appearance, and treatment of the GCTB.

CASE REPORT

A 26-year-old woman presented to our department with 1-year history of left ankle pain and swelling with no prior trauma. Increasing pain and limitation of motion in the joint during last three months necessitated the use of crutches. Physical examination revealed increased volume of the distal tibial metaphysis and moderate soft tissue swelling. The range-of-motion of the ankle was significantly reduced.

Plain X-rays (Figure 1 a, b), computed tomography (CT) (Figure 1 c, d), and magnetic resonance imaging (MRI) (Figure 1 e, f) presented a large eccentric, expansive lesion in the medial aspect of the distal tibia suggestive of a giant cell tumor of bone. Laboratory tests were in normal ranges. An open biopsy was performed and a giant cell tumor of bone was diagnosed. Microscopically, the lesion was presented by proliferating uniform oval mononuclear cells scattered around the background of numerous osteoclast-type giant cells. According to the radiological classification of Campanacci et al., the tumor was classified as a grade 2 lesion [6].

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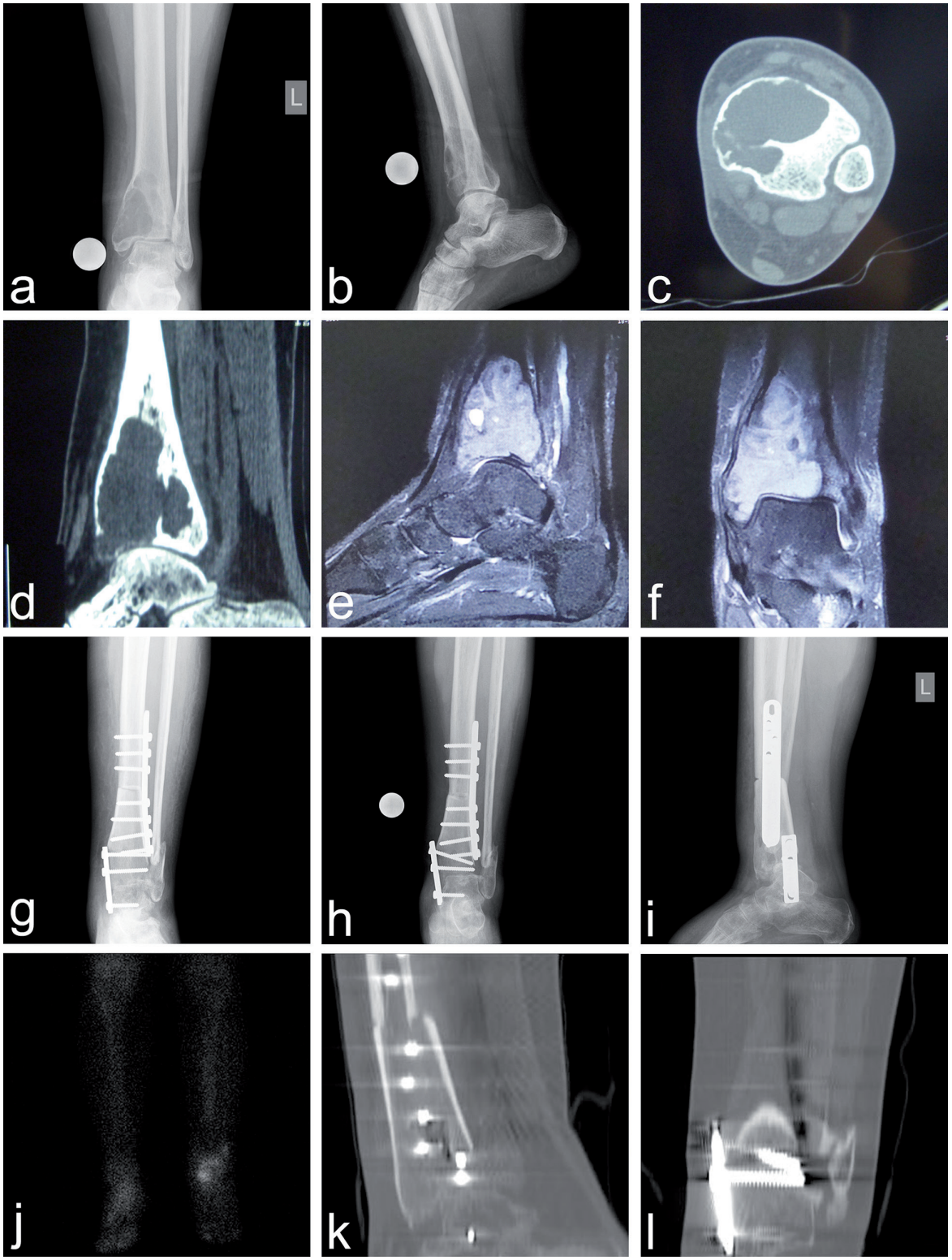

Figure 1. (a) Preoperative antero-posterior and (**b**) lateral radiographs revealed a large eccentric, expansile lesion in the medial aspect of the distal tibia; (**c**) and (**d**) CT presented thinned, inflated cortex, without periosteal reaction; (**e**) and (**f**) MRI presented thinned, expanded cortex, without periosteal reaction; (**g**) antero-posterior radiograph at the 6-month presented well arthrodesed ankle with no evidence of recurrence; antero-posterior (**h**) and (**i**) lateral radiograph 19-months after surgery presented osteolysis of the distal end of the allograft and proximal displacement of the talus with the tension band plate; (**j**) bone scans revealed increased bone metabolism in the region representing vascularization and resorption of the allograft; (**k**) and (**l**) CT presented no evidence of recurrence at 27-months after surgery

The condition, its prognosis, the possible treatment modalities and complications were discussed with the patient. Among the surgical alternatives, we considered curettage with reconstruction using an allograft, bone cement and an autograft, resection and arthrodesis, or joint replacement. Decision was made, with the patient, to perform en-block resection of the distal tibia, reconstruction with a structural tibial allograft, and ankle arthrodesis. Precise preoperative planning was used concerning the resection and arthrodesis. Using an anterior approach the left distal tibia was exposed extraperiosteally and resected at 9.5 cm proximal to the ankle joint. The articular surface of the talar dome was removed and a distal oblique fibular osteotomy was performed. The allograft was cut to size and fixed with a 4.5 mm DCP. Thereafter, for arthrodesis of the ankle, an oblique 4.5 mm compression cancellous screw from the metaphysis of the allograft to the talus was inserted. During the insertion of the second cancellous screw a partial fracture of the allograft occurred. After that, the ankle was arthrodesed with a tension band plate on the medial side. No adjuvant agents were used. The wound was closed over a deep suction drain. A short leg cast was applied and the leg was non-weight bearing for 6 weeks. On suture removal, which was performed through a window in the plaster cast, an area of partial-thickness skin necrosis was detected on the dorsum of the foot, measuring 3 cm by 5 cm. The lesion was managed with enzymatic debridement and healed for 3 weeks. At 6-months follow-up, the patient had a smooth healed scar with a painless and well arthrodesed ankle with no evidence of recurrence (Figure 1g). At 19 months after surgery, there was osteolysis of the distal end of the allograft and proximal displacement of the talus with the tension band plate on the medial side of the ankle with the foot remaining plantigrade (Figure 1 h, i). Bone scans revealed increased bone metabolism in the region representing resorption of the allograft (Figure 1j). Thereafter, the patient underwent treatment with bisphosphonates. On CT, no signs of continuing bone resorption and no evidence of recurrence was detected at 27-months follow-up (Figure 1 k, l). The patient had pain-free stable ankle and unlimited ambulation. Her subtalar and midtarsal motion measured 0° of dorsiflexion/supination and 15° of plantar flexion/pronation.

DISCUSSION

GCTB is described as a locally invasive tumor with a high rate of recurrence and a possibility of mainly pulmonary metastases or transformation into a malignancy [1-4]. The diagnosis of this tumor requires precise assessment of the clinical findings, imaging modalities, and histopathologic evaluation [2,10,11].

In the current literature, GCTB is described as a predominantly osteoclastogenic stromal cell tumor of mesenchymal origin [12]. It is composed of three cell types - the spindle-like stromal cells, mononuclear monocyte cells, and multinucleated giant cells [12-16]. The multinucleated giant cells which mimic osteoclasts are principally responsible for the extensive bone resorption that is characteristic of GCTB [16]. However, the stromal cells are the main neoplastic component of GCTB and have been shown to express and secrete a variety of chemotactic factors to enlist pathologic components [12,16]. Mononuclear monocyte cells are considered to be either reactive macrophages or osteoclast precursors [12,16].

The main clinical symptoms are non-specific and include pain of variable severity, local swelling, tenderness of the affected area, and limited range of motion of the adjacent joint [2,3,17]. The duration of symptoms usually varies from two to six months. Rarely, a pathologic fracture may be the first symptom [2,3,17].

Imaging studies are essential for the diagnosis of GCTB [2,11,17]. On conventional radiographs this tumor typically presents as a purely lytic eccentric lesion, with expansion and thinning of the cortex. Periosteal reaction is usually absent [1,3,7,17]. Campanacci et al. classified GCTB in three grades: grade 1 is static form with minimal involvement of the cortex; grade 2 presents with thinned and expanded cortex and in grade 3 the lesion penetrates the cortex and has a soft tissue component [6]. As with any suspicious bone lesion, full staging with MRI and CT should be undertaken [17,18]. CT is useful in the evaluation of the cortical bone and could clearly present the thinning of the cortex and subchondral bone, the pathologic fracture, the periosteal reaction, and the absence of matrix mineralization [3,19]. In the cases of cortical destruction and soft-tissue tumor extension MRI is superior to CT in delineation of GCTB. The tumor will appear with a non-homogenous signal on magnetic resonance imaging: low in T1-weighted images and high in T2-weighted images. Bone scintigraphy could also be used for evaluation of giant cell tumor of bone [2,3,17].

Various limb salvage techniques for the distal tibia have been described in literature: extended curettage with a large window, high speed burring, and filling of the cavity with bone cement or bone graft; resection and ankle arthrodesis; resection

and reconstruction with porous tantalum spacer; resection of the tumor followed by placement of an external fixator for segmental bone transport and endoprosthetic replacement [7,20-25]. Autografts and allografts are associated with high rates of healing when used for arthrodesis. In most cases, arthrodesis of the ankle joint provides excellent stability of the ankle and very good functional outcome [7]. However, due to the high pressure in the ankle joint after resection of the distal tibia and arthrodesis of the ankle, the allograft could collapse or fracture. Infections, non-union, osteolysis, iatrogenic fracture of the allografts were also reported [27]. In such cases these complications could compromise the ankle arthrodesis. In cases of bone resorption, bisphosphonates may be a reasonable option due to the reported evidence of inhibiting bone resorption in human and animal trials [26]. Rarely, in cases when surgery is not feasible, irradiation for the treatment of GCTB could also be an alternative option [28].

GCT of bone is a locally aggressive tumor with a high tendency to recur after removal. The rates of recurrence after simple curettage ranged from 12-65% as compared with 12-27% after curettage and adjuvant treatment and 0-12% after resection [1,4,29-31]. In cases of GCTB affecting the hand and foot the recurrence rate is higher in comparison with GCTB in more conventional sites [8,10]. GCTB could metastasize in up to 10% of patients. Most commonly the metastatic spread occurs after repetitive local recurrences [10,11].

In conclusion, GCTB of the foot and ankle is a rare lesion. Prognosis, treatment, and results are directly dependent on early diagnosis and adequate therapy. In this report, we present a rare case of GCTB of the distal tibia treated with en-bloc resection and ankle arthrodesis. This treatment modality leads to good results.

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