GIANT–CELL REPARATIVE GRANULOMA OF THE TIBIA ASSOCIATED WITH FEMORAL ENCHONDROMA

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SUMMARY

GIANT–CELL REPARATIVE GRANULOMA OF THE TIBIA ASSOCIATED WITH FEMUR ENCHONDROMA

Introduction: Occurence of giant cell reperative granuloma (GCRG) ouside the jaw, skull, or facial bones is unusual. In this paper was discussed an enchondromal case accidentally discovered in the femoral supracondylar region together with a tibially-located GCRG.

Patients and Methods: A 60-year-old female patient was referred to us with complaints of moderately tender swelling at the right leg. When whole body scintigraphic scanning was performed for this lesion, distal femoral lesions were incidentally discovered. The patient was admitted to surgery and incisional biopsies were performed on both masses. Upon receipt of the pathology report, the patient was readmitted to surgery during which the enchondroma was treated locally with intralesional resection and an allograft was performed. The GCRG was treated with a marginal resection, posterior iliac crest, allograph and intramedullar interlocking nailing. No recurrence was noted after 18th postoperative month in this patient who is being continuously followed-up.

Conclusion: This is the second reported case of giant cell reperative granuloma associated with enchondroma. In addition to the clinical features, the pathologic findings and differantial diagnosis were also discussed. A long-term follow-up is considered necessary, however, due to the recurrence risk involved.

Key Words: Giant cell reperative granuloma, Enchondroma, Tibia.

ÖZET

TİBİADA YERLEŞİM GÖSTEREN DEV HÜCRELİ REPARATİVE GRANÜLOMA İLE BİRLİKTE FEMURUN ENKONDROMU

Giriş: Giant cell reparative granuloma çene, kafa ve fasial kemikler dışındaki bölgelerde nadir olarak görülmektedir. Bu çalışmada, tibia'ya yerleşmiş GCRG ile beraber rastlantısal olarak tanısı konulan femur suprakondiler bölge enchondroma'sı değerlendirildi.

Hastalar ve Metod: Altmış yaşındaki bayan hasta sağ bacağındaki şişlik ve ağrı şikayeti ile geldi. Tibia'daki kitle için tüm vücut sintigrafisi çekildiğinde femur distalindeki kitle rastlantısal olarak saptandı. Her iki kitle için insizyonel biyopsi yapıldı. Patoloji raporundan sonra hasta tekrar cerrahiye alınarak enchondroma intralezyonal küretaj ve allogreftle, Tibia'daki GCRG marginal rezeksiyon, iliac greft ve allogreft uygulayarak kırık proflaksisi için intramedüller interlocking ile tedavi edildi. Postoperatif 18 ay sonrasında nüks görülmedi.

Sonuç: Bu olgu GCRG ile birlikte görülen literatürde bildirilen 2. Olgudur. Nadir görülen bu kitlenin klinik özellikleri, patolojik bulguları ve ayırıcı tanısı tartışıldı. Recuurens ihtimali nedeniyle uzun süreli takip gerekmektedir.

Anahtar Kelimeler: Giant cell reperative granüloma, Enchondroma, Tibia.

INTRODUCTION

The GCRG, first defined by Jaffe in 1953 and mostly involving the facial bones, is a benign osteal lesion[1]. It was reported in the subsequent years[2] as a granuloma case particularly in the short tubular bones of hand and foot. The occurrence in the long tubular bones is somewhat rare. Its association with other bone tumours was reported in only two cases[3,4].

In this study, a patient is presented with GCRG associated with enchondroma.

CASE REPORT

A 60-year-old female patient has been reported with complaints of moderately tender swelling at the right

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leg. Physical examination revealed a bony mass moderately tender to palpation in the proximal tibia medially. Function of the right knee was normal.

X-ray films revealed a large cystic expansile lesion that destructed the tibial cortex (Figure 1). When whole body scintigraphic scanning was performed for this lesion, distal femoral lesions were incidentally discovered (Figure 2). Computer tomography demostrated an eccentric lesion with cortical destruction (Figure 3). In magnetic resonance image, the tumor had clearly broken through the cortex to form a soft-tissue mass (Figure 4). The right femoral metaphysial lesion was shown as a centrally-localized radiolucent and scalloped one. There were no sclerotic rim and cortical destruction. Laboratory studies including alkaline phosphatase, calcium and phosphorus tests were conducted and the results were within normal limits.

The patient was admitted to surgery and incisional biopsies were performed on both masses. Specimen taken from the femur was bluish white in colour and specimen taken from tibia showed a brownish tinge. Both specimens were sent to pathological examination.

The microscopic examination of the specimens revealed mononucleic chondrocytes displaying local hyalinisation with a lobular appearance in a single lacuna of the femoral mass. The microscopic characteristics of the tibial mass were new osteal lamellæ encircled by osteoblasts, multinuclear giant cells denser at the hæmorrhagic regions of stroma consisting of fibroblasts, histiocytes and inflammatory cells (Figure 5).

Upon receipt of the pathology report, the patient was readmitted to surgery during which the enchondroma was treated locally with intralesional resection and an allograft was performed. The GCRG was treated with a marginal resection, posterior iliac crest, allograft and intramedullar interlocking nailing (Figure 6).

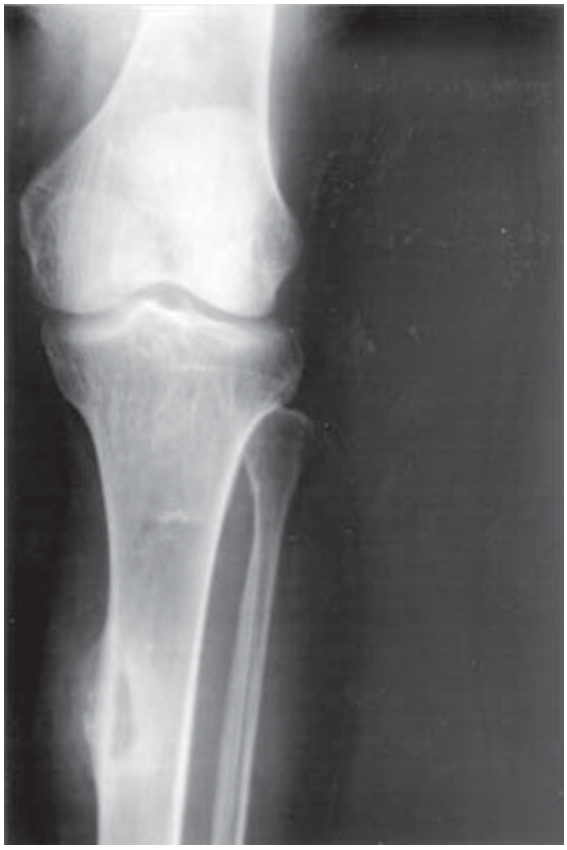

Figure 1a

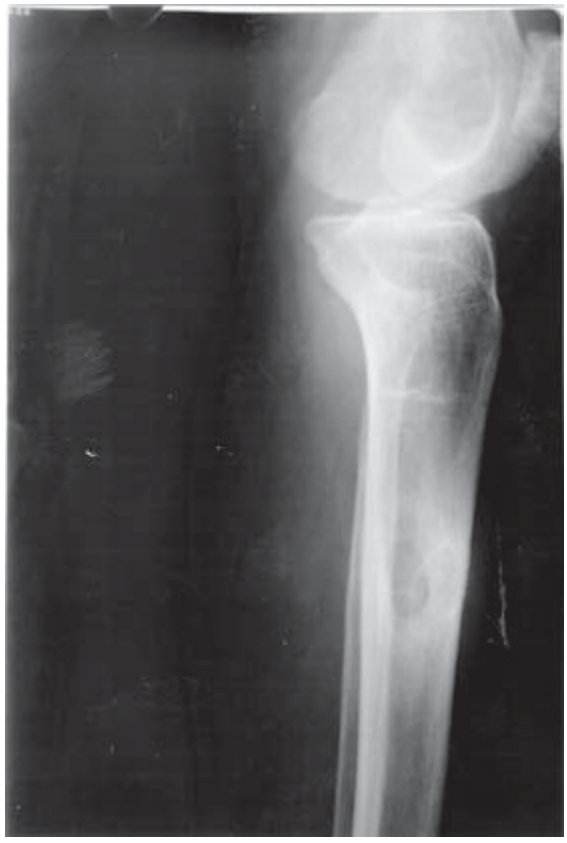

Figure 1b

Figure 1: Anteroposterior (A) and lateral (B) plain radiographs of the distal femur and proximal tibia.are showing a large cystic expansive lesion that destructed the tibial cortex.

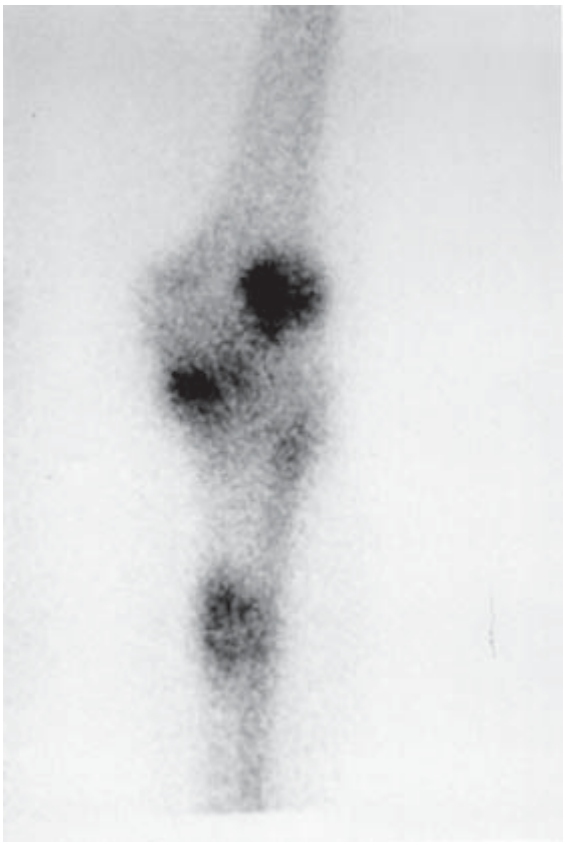

Figure 2: In the scintigraphic scanning, tibial lesion and distal femoral lesions are demostaring significantly high osteoblastic activity.

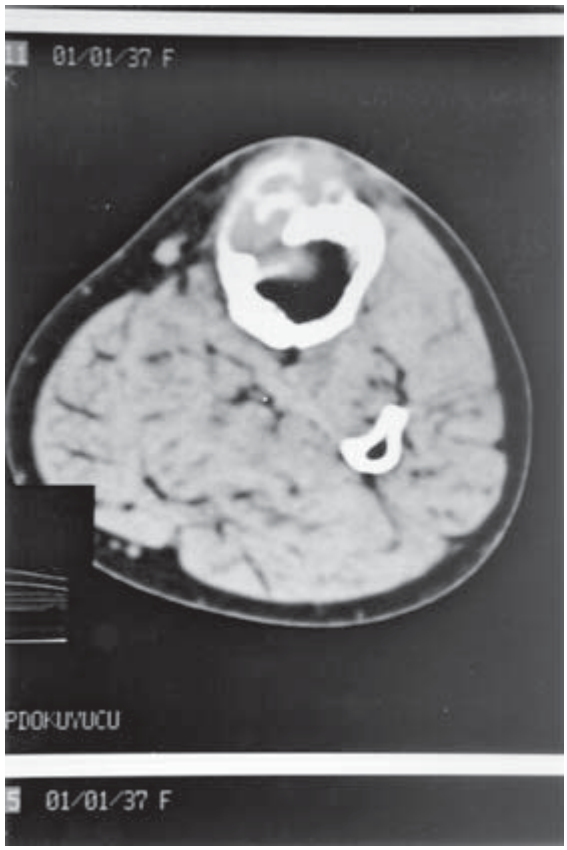

Figure 3: Computed tomography is demonstrated an eccentric medullar mass with cortical destruction involving soft tissue.

The follow-up examination made in 18th postoperative month showed that the patient was asymptomatic. The physical examination indicated no tenderness to palpation of the operation sites. The knee functions were normal and laboratory studies were within normal limits.. Roentgenograms showed consolidation of the bone grafts and diminished site of the tibial expansile lesions as compared to the preoperative roentgenograms.

DISCUSSION

In this paper is discussed an enchondromal case accidentally discovered in the femoral supracondylar region together with a tibially-located GCRG.

Enchondromæ are classically long and oval lesions settling at the centres of tubular bones, generally locate at the smaller bones of hands and feet. Their other localisations are the metaphyses and diaphyses of such long bones as femur and humerus. They are generally asymptomatic and discovered accidentally or following a fracture[5,6]. Those larger than 3 cm or displaying symptomatic lesions require surgical intervention. Treatment for the long bone enchondromæ is generally curettage and allographic fill of the cavity[5]. In our patient, the localisation was the femoral distal metaphysis, discovered only in the bone scintigraphy when performed because of her complaints of swelling at the tibia. The enchondroma was curetted and filled with an allograph. There was only one reported case[3] of GCRG together with an enchondroma.

The GCRG, considered as a tumor-like lesion by several authors, is observed mostly in the maxillary, cranial and facial bones. A study of the available literature suggests a frequency of localisation as hand phalanxes, metatarsal, metacarpal, tarsals and carpals. Phalanxes in the hands and metatarsæ in the feet are the usual locations[7-13]. Two femoral, one humeral and one tibial cases were also reported[3,4,14,15]. In addition to the foregoing, occurrences together with one fibrous dysplasia and one enchondroma were also presented. Our case

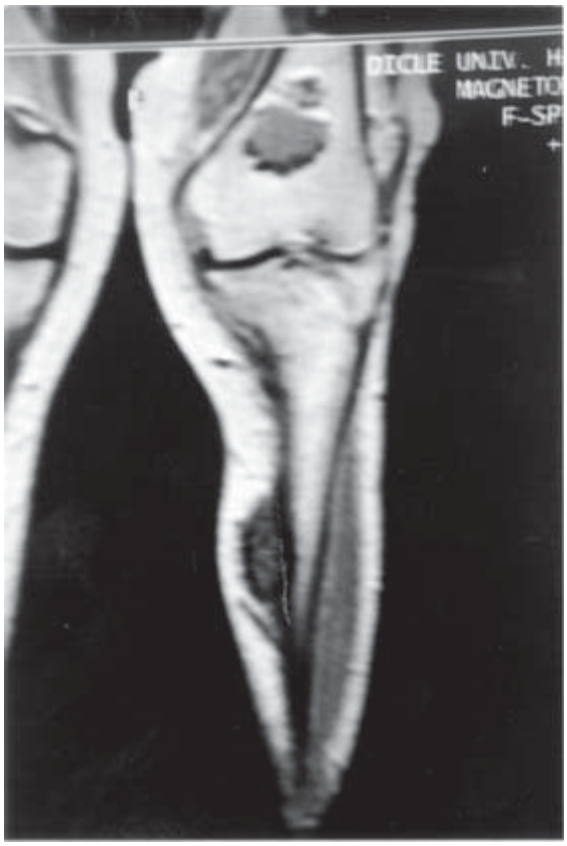

Figure 4. In magnetic resonance images, the right femoral metaphyseal lesion is shown as a centrally-localized hypointense and scalloped one. There are no sclerotic rim and cortical destruction. The tibial lesion has clearly broken through the cortex to form a soft-tissue mass.

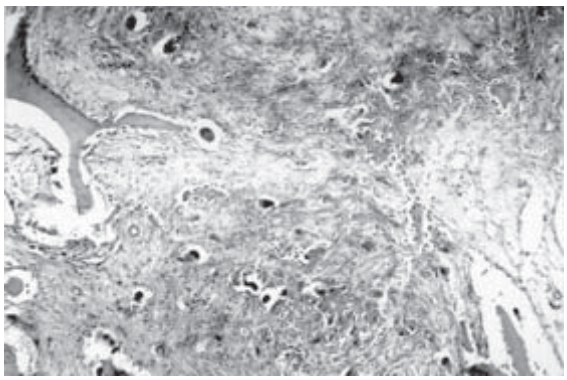

Figure 5. The microscopic characteristics of the tibial mass are new osteal lamellæ encircled by osteoblasts, multinuclear giant cells denser at the hæmorrhagic regions of stroma consisting of fibroblasts, histiocytes and inflammatory cells(H&E stain, X 200).

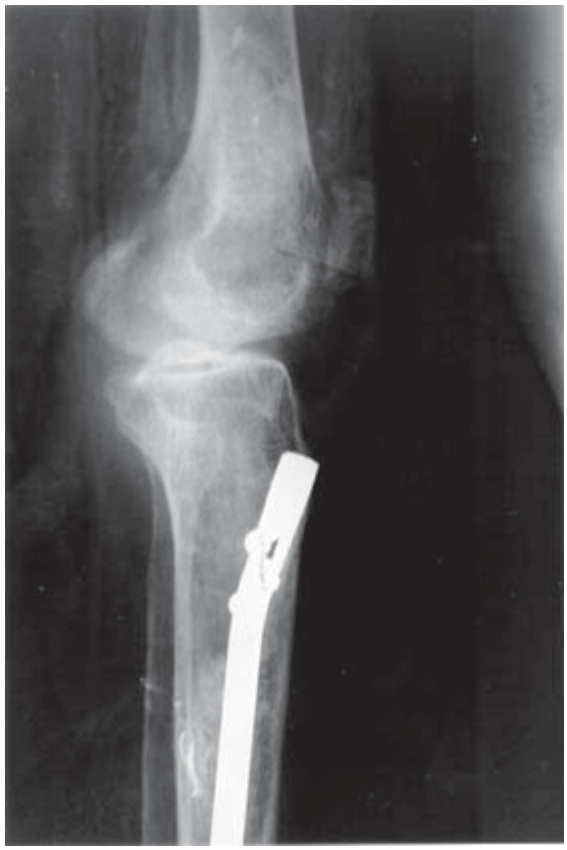

Figure 6. On the lateral radiograps intralesional curettage and greffonage in the femur and marginal resection, allograft filling of the cavity and intramedüller fixation of tibia are shown.

is the second patient with a tibial localisation. It is generally monophylic. The multiple involvements were reported in only two cases in the literature, one in the hand and the other at the foot[2,16]. While its preferred osteal location is the metaphysis, diaphysial involvements were found in a lesser degree. There are no reports on epiphysial involvement[7,13]. In our case, the localisation was at the diaphysis. Our case showed a diaphysial involvement. Though the second decade is suggested as the age of occurrence, the youngest age and the oldest age that we could determine in the literature was 3 and 76 years[2,7,12,13,17]. Our patient was 60 years old. The gender distribution was reported by some researchers as equal although some others suggest a male domination[2,7,13].

Several authors indicate the ethiologic cause to be the posttraumatic intraosseous hæmorrhage[2,9,13,17,18]. Yet there are a number of cases, like ours, in which no trauma were revealed in the anamnesis[7]. The characteristic histology of the GCRG includes a

cellular fibrous stroma with irregularly distributed multinucleated giant cells, many of which occur in clusters associated with foci of hæmorrhage. Occasionally, mononuclear inflammatory cell infiltration is present and osteoid formation is frequently found[2,7,9,10,12,13].

Since it was discovered in our case at a non-unique skeletal region, it should be differentiated from the multicentric giant cell and brown tumour.

A giant cell tumour usually occurs in the third or fourth decade of the life, at an average age of 33 years. This tumour typically affects the epiphysis but may also extend into the metaphysis. It is an eccentric, lythic and expanded lesion. Histologically, a giant cell tumor consists of homogeneous stroma with giant cells and mononuclear cells dispersed evenly throughout the tumor. It rarely contains osteoid or new bone. This contrasts with the GCRG in which the giant and mononuclar cells predominate in the hæmorrhage areas. This tumour has a high incidence of recurrence and, therefore, requires a more aggressive treatment.

The brown hyperparathyroidism tumour may be rœntgenographically and histologically indistinguishable from the GCRG. It can only be excluded by the absence of biochemical abnormalities (serum calcium, phosphorus and alkaline phosphatases) and of radiographic evidence of generalized skeletal rarefaction.

Treatment of GCRG consists of intralesional curettage and greffonage, although the recurrence rate varies between 30 and 70 percent. In our case, an allograft was made into the cavity resulting from the marginal resection. Since this part of the bone was weakened, it was fixed by a interlocing intramedullary nail. As for the enchondroma at the distal femur, an intralesional curettage and greffonage was applied to avoid the probability of a pathological fracture in the future. No recurrence was noted after 18th postoperative month in this patient who is being continuously followed-up. A long-term follow-up is considered necessary, however, due to the recurrence risk involved[3,13,17].

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