

## Research Paper

## Soft tissue recurrence of giant cell tumor of the bone: Prevalence and radiographic features

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## ABSTRACT

**Aim:** Recurrence of giant cell tumor of bone (GCTB) in the soft tissue is rarely seen in the clinical practice. This study aims to determine the prevalence of soft tissue recurrence of GCTB, and to characterize its radiographic features.

**Methods:** A total of 291 patients treated by intralesional curettage for histologically diagnosed GCTB were reviewed. 6 patients were identified to have the recurrence of GCTB in the soft tissue, all of whom had undergone marginal resection of the lesion. Based on the x-ray, CT and MRI imaging, the radiographic features of soft tissue recurrence were classified into 3 types. Type I was defined as soft tissue recurrence with peripheral ossification, type II was defined as soft tissue recurrence with central ossification, and type III was defined as pure soft tissue recurrence without ossification. Demographic data including period of recurrence and follow-up duration after the second surgery were recorded for these 6 patients. Musculoskeletal Tumor Society (MSTS) scoring system was used to evaluate functional outcomes.

**Results:** The overall recurrence rate was 2.1% (6/291). The mean interval between initial surgery and recurrence was  $11.3 \pm 4.1$  months (range, 5–17). The recurrence lesions were located in the thigh of 2 patients, in the forearm of 2 patients and in the leg of the other 2 patients. According to the classification system mentioned above, 2 patients were classified with type I, 1 as type II and 3 as type III. After the marginal excision surgery, all patients were consistently followed up for a mean period of  $13.4 \pm 5.3$  months (range, 6–19), with no recurrence observed at the final visit. All the patients were satisfied with the surgical outcome. According to the MSTS scale, the mean postoperative functional score was  $28.0 \pm 1.2$  (range, 26–29).

**Conclusions:** The classification of soft tissue recurrence of GCTB may be helpful for the surgeon to select the appropriate imaging procedure to detect the recurrence. In addition, the marginal resection can produce a favorable outcome for the patients.

## 1. Introduction

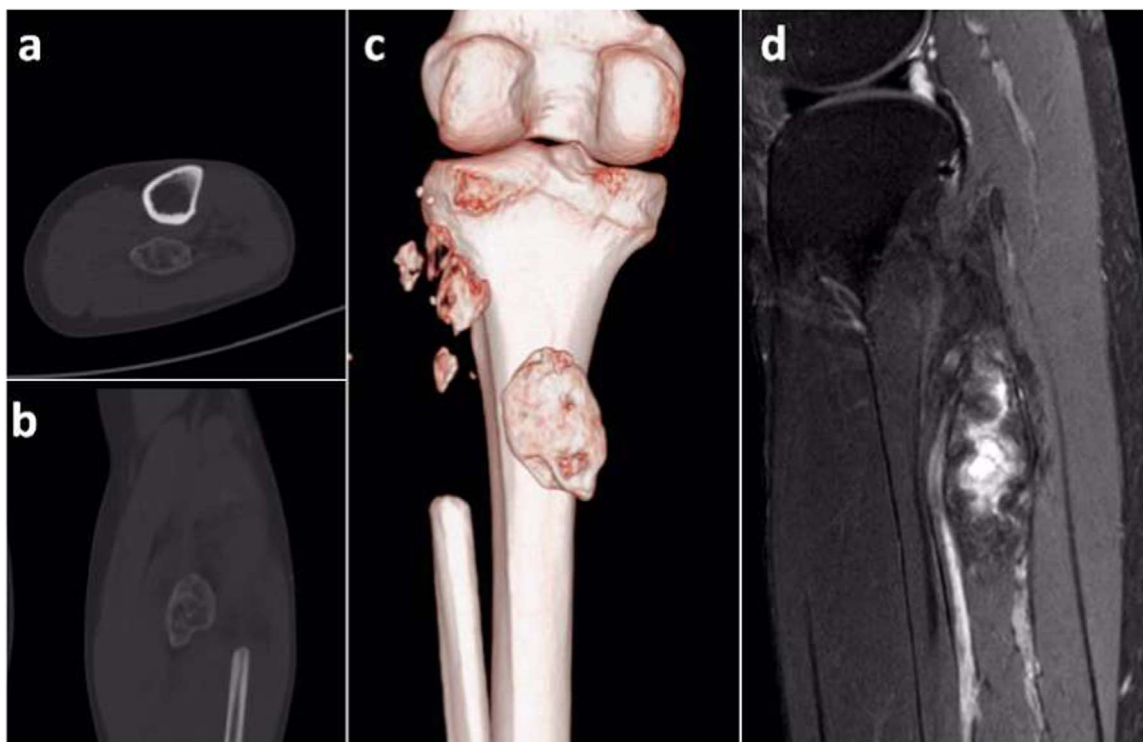
Giant cell tumor of bone (GCTB) is a neoplasm typically occurring in the epiphyses of long bones and representing approximately 5% of all bone tumors [1]. Despite its benign histopathology nature, GCTB was reported to have a high rate of local recurrence as well as occasional pulmonary metastases, thereby implicating the aggressiveness of the tumor [2–4]. For most cases, the postoperative recurrence could be observed within 24 months after the surgery. Depending on the type of surgical procedure and local presentation of the tumor, the recurrence rate of GCTB could range from 2.5% to 45% [5,6]. Patients treated with intralesional curettage were reported to have higher recurrence rate

than those undergoing wide resection of the tumor [7,8].

Compared with local recurrence in bone, recurrence in the soft tissue is more rarely seen in clinical practice. It has been documented that the soft tissue recurrence of GCTB most frequently arises in the area adjacent to curettage site, probably due to the contamination during surgical removal of the tumor [9–12]. For an early detection of the recurrence, Balke et al. [5] suggested MR imaging be performed in case of any suspicious findings. To date, there were a limited number of literatures that described the presentation of soft tissue recurrence [13–15]. A peripheral rim of ossification surrounding the mass was reported as an indicator of soft tissue recurrence of GCTB [13,15]. Nevertheless, some studies have shown a low detection rate of the

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**Fig. 1.** A 44-year-old female patient with giant cell tumor in the proximal left fibula. (a–c) CT scan of the left leg at the 17th month after surgery showed soft tissue nodular lesions with eggshell-like peripheral hyperdense rim. (d) On the cross-sectional fat-suppressed T2-weighted MR image, soft tissue masses revealed inhomogeneous high signal intensity.

ossification on the plain radiography [14,16]. Obviously, radiographic characteristics of soft tissue recurrence of GCTB have not been well recognized in previous studies, and a good understanding of them would facilitate accurate diagnosis and appropriate treatment.

In the present study, we retrospectively reviewed a cohort of GCTB patients treated with intralesional curettage in our center. Radiographic data and clinical outcome of the patients with soft tissue recurrence of GCTB were analyzed. The purpose of our study was to determine the prevalence of soft tissue recurrence of GCTB and to characterize its radiographic features.

## 2. Methods

### 2.1. Patients

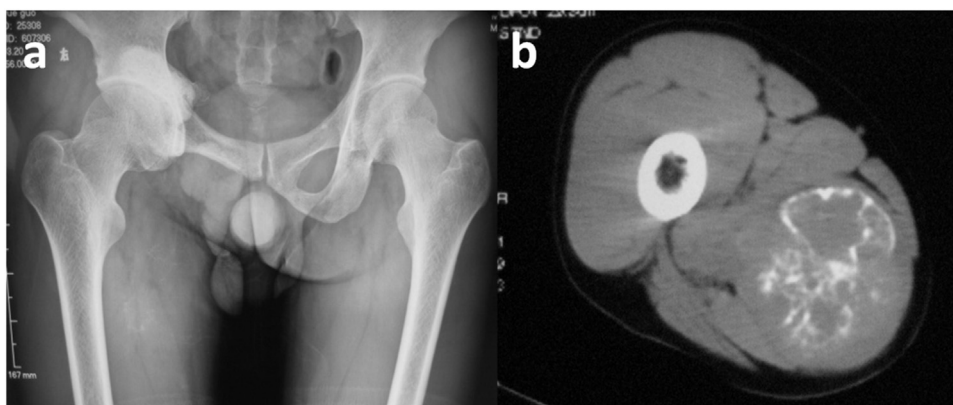
Between January 2002 and December 2014, a total of 291 patients with histologically diagnosed benign GCTB were treated by intralesional curettage at our department. All the patients had a minimum follow-up of 2 years with complete radiographic data. Radiological

images and clinical data were retrospectively reviewed and the baseline characteristics were recorded for each patient, including gender, age, location of the tumor, tumor grade and tumor size that was measured as the greatest dimension in centimeters from x-ray films. Specifically, tumor grade was classified as grades I, II, and III according to the Campanacci method as previously reported [17].

6 patients were identified to have the recurrence of GCTB in the soft tissue, all of whom had undergone marginal resection of the lesion. Demographic data including period of recurrence and follow-up duration after the second surgery were recorded for these 6 patients. Under the approval of the local Institutional Review Board, all the patients signed the informed consent for participation in the current study.

### 2.2. Classification of radiographic features

Based on the x-ray, CT and MRI imaging, the radiographic features of soft tissue recurrence were classified into 3 types. Type I was defined as soft tissue recurrence with peripheral ossification (Fig. 1). Type II was defined as soft tissue recurrence with central ossification (Fig. 2).



**Fig. 2.** A 37-year-old male patient with giant cell tumor in the right ischium. (a–b) A centrally located ossification could be observed on both plain radiography and CT scan at the 9th month after surgery.



Fig. 3. A 34-year-old female patient with giant cell tumor in the distal right femur. (a) There was no evidence of ossification in the soft tissue on plain radiography (b) The coronal T2-weighted MR image showed inhomogenous intermediate to high signal intensity.

Type III was defined as pure soft tissue recurrence without ossification (Fig. 3).

### 2.3. Surgical outcome

Measurements of surgical outcomes included procedure-related complications and health-related quality of life. Musculoskeletal Tumor Society (MSTS) scoring system was used to evaluate functional outcomes concerning the following six domains: pain, functional activities, emotional acceptance, use of external support, walking ability, and gait.

## 3. Results

### 3.1. Baseline characteristics of the patients

Table 1 summarized the baseline characteristics of the initially screened 291 patients with GCTB. The average age of the patients was  $33.1 \pm 10.5$  (13–78). The average follow-up period was  $53.2 \pm 21.7$  months (31–95). The tumors were located in the lower extremities of

**Table 1**  
Baseline characteristics of the initially screened patients with GCTB.

Characteristic	Patients (n = 291)
Age (years)	
< 20	28
20–40	198
> 40	65
Gender	
Male	135
Female	156
Tumor location	
Distal femur	76
Proximal tibia	47
Proximal femur	34
Sacrum	29
Proximal humerus	28
Distal radius	25
Proximal fibula	21
Lumbar vertebra	19
Other locations	12
Campanacci grading	
1	57
2	186
3	48

211 patients (72.5%), in the upper extremities of 57 patients (19.6%) and in the trunk of 23 patients (7.9%). The tumors size was averaged 3.6 cm, ranging from 2 to 7 cm. According to the Campanacci staging system, 57 cases were classified as grade I, 216 as grade II and 18 as grade III.

6 patients had the recurrence of GCTB in the soft tissue after the initial treatment. The overall recurrence rate was 2.1% (6/291). As shown in Table 2, the mean interval between initial surgery and the incidence of recurrence was  $11.3 \pm 4.1$  months (range, 5–17). The recurrence lesions were located in the thigh of 2 patients, in the forearm of 2 patients and in the leg of the other 2 patients. The most common complaint was a growing mass reported by all 6 patients. 3 patients reported mild to moderate pain around the lesion. No patient reported obvious clinical dysfunction.

### 3.2. Radiographic features of the soft tissue recurrence

Radiologic evaluation in all patients excluded bone involvement in the recurrence. According to the classification system mentioned in the Section 2, 2 patients were classified as type I, 1 as type II and 3 as type III. As shown in Fig. 1, soft tissue nodular lesions with an eggshell-like peripheral hyperdense rim could be observed through CT scan for patients with type I. For internal soft tissue, inhomogenous intermediate to high signal intensity could be observed on fat-suppressed T2-weighted MRI. Histological examination of the excised mass presented an ossified shell surrounding the soft tissue mass. For patients with type II (Fig. 2), a centrally located ossification could be observed on both plain radiograph and CT scan. For patients with type III (Fig. 3), there was no evidence of ossification in the soft tissue masses. Fat-suppressed T2-weighted MRI showed inhomogenous intermediate to high signal intensity.

### 3.3. Surgical outcome

After the marginal excision surgery, all patients were consistently followed up for a mean period of  $13.4 \pm 5.3$  months (range, 6–19), with no recurrence observed at the final visit. There was no case of procedure-related complication. All the patients were satisfied with the surgical outcome. According to the MSTS scale, the mean postoperative functional score was  $28.0 \pm 1.2$  (range, 26–29).

## 4. Discussion

Complete surgical elimination of GCTB has always been a

**Table 2**  
Demographic data of patients with soft tissue recurrence.

No.	Gender	Age	Location	Recurrence time (months)	Radiographic classification <sup>a</sup>	F/U (months)	MSTS score
1	Male	37	Right leg	11	II	13	30
2	Male	23	Right forearm	12	III	12	26
3	Female	26	Left leg	14	III	19	28
4	Female	58	Left thigh	11	I	12	29
5	Female	44	Left forearm	17	I	18	28
6	Female	34	Right thigh	5	III	6	29

<sup>a</sup> Type I was defined as soft tissue recurrence with peripheral ossification; Type II was defined as soft tissue recurrence with central ossification; Type III was defined as pure soft tissue recurrence without ossification.

challenging task, since local recurrence is frequently observed even in the hands of highly experienced orthopedic surgeons. Much less commonly, recurrence of GCTB within the soft tissue was presumed to originate from seeding at the time of primary curettage of the tumor [9–12]. The accurate incidence of soft tissue recurrence has yet to be determined. Cooper et al. [13] reported an incidence of 1.5% among 1100 GCTB patients at a single center (17/1100). Park et al. [15] reported 4 cases of soft tissue recurrence after reviewing 129 cases of GCTB (3.1%). In our study, the rate of soft tissue recurrence was 2.1%, mostly in line with those reported by previous studies [13,15]. Moreover, we observed that the recurrence mostly occurred within 12 months after the original surgery. Comparably, the recurrence time was at approximately 1 year after surgery in study of Akaike et al. [18], and ranged from 7 to 13 months in the study of Park et al. [15]. Therefore, early radiological detection of the recurrence lesion should be accentuated for the diagnosis and surgical intervention.

Knowledge concerning the radiographic features of the soft-tissue implants of GCTB remains a controversial issue. A peripheral rim of ossification has been described as a distinctive radiographic appearance. Cooper et al. [13] identified soft tissue recurrence with a peripheral rim of ossification in 17 cases. Occasionally, however, a soft-tissue recurrence may not be visible on plain radiographs when the recurrent lesion is asymptomatic and contains no ossification. In the study of Lee et al. [16], 4 patients with an isolated soft-tissue recurrence had negative findings on plain radiography. For each patient, a soft-tissue mass could be palpable on physical examination, and successfully detected as a heterogeneous signal pattern on MRI. Among the 4 patients reported by Park et al. [15], only 2 had ossification in the soft tissue. In another study of Ehara et al. [14], 1 out of 3 recurrence cases had no ossified periphery.

Current advances in radiological imaging have enabled more sensitive modalities to detect small soft tissue lesions. MR imaging can detect soft tissue masses and reveal the extent of the lesion, while the outcome may be nonspecific. Both plain radiography and CT scan are sensitive enough to detect ossification. For the first time, we proposed a new classification of the soft-tissue recurrence of GCTB based on a combination of the plain radiograph and MR imaging. In this study, 3 patients were identified as pure soft tissue recurrence without ossification (Type III), 2 as peripheral ossification (Type I) and the other 1 as central ossification (Type II). Our finding was consistent with those of Park et al. [15], who concluded that ossification of soft tissue is commonly identified at the periphery and occasionally in the central portion. Moreover, it appeared that there could be a high proportion of recurrence lesion without ossification in the clinical practice. Herein, for patients present suspicious soft-tissue mass on the plain radiography, MR imaging is strongly recommended to determine whether a recurrence of GCTB in the soft tissue has occurred. If necessary, additional diagnostic studies, particularly a biopsy performed under the guidance of computed tomography, can be used to exclude the possibility of soft tissue recurrence.

No studies have specifically documented the surgical treatment for soft tissue recurrence. In this study, we applied marginal excision to all the 6 patients. During a mean follow-up of  $13.4 \pm 5.3$  months, no

recurrence was observed. While some patients had complained of pain before surgery, a mean MSTS score of  $28.0 \pm 1.2$  indicated that the surgical outcome was satisfying.

## 5. Conclusions

Although our study was limited by the small sample size, we believe the classification system of soft tissue recurrence of GCTB may be helpful for the surgeon to select the appropriate imaging procedure to detect the recurrence, which mostly occurred within 12 months after the original surgery. The marginal resection of the soft tissue recurrence can produce a favorable outcome for the patients.

## Conflict of interest statement

No benefits in any form have been or will be received from a commercial party related directly or indirectly to the subject of this manuscript.

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