Pulmonary metastasis of recurrent giant-cell tumor in proximal humerus: a case report
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INTRODUCTION
Giant-cell tumor (GCT) of the bone is a benign neoplasm which commonly occurs in the distal femur, proximal tibia, and distal radius. The proximal humerus is a relatively rare site of occurrence, with a prevalence rate of 4%. We report a case of a 40 years-old male with a recurrent GCT of the proximal humerus. Wide excision and hemiarthroplasty of the shoulder was performed. However, the tumour recurred. Asymptomatic metastasis to the right lower lobe of the lung was found on plain radiograph. We suggest that multiple surgeries would increase the risk of local recurrence of GCT that will lead to pulmonary metastasis.


Case report
A 40 years old, smoker, the male was...
referred to our orthopaedic outpatient clinic due to an increasing mass on his left shoulder. He has occasional (intermittent) pain on his left shoulder while working or at rest. Visual Analog Scale (VAS) was graded at 5 to 6. He has no motor weakness of his upper extremity. There was no history of trauma, fever or pain elsewhere of his body, and he claimed he was otherwise healthy. Past medical history, he claimed that he underwent some tumour resection and fixation in some local district hospital three years ago. Physical examination revealed a deep-seated round mass, measuring 25 x 20 x 10 cm, located at his proximal left shoulder. It was hard in consistency, non-mobile and attached to the underlying structure. The colour of the skin was the same as the surrounding, no redness or warmth of the skin. He could achieve the only flexion of 30 degrees, extension 10 degrees, abduction 45 degrees while adduction was full. His range of motion was limited by pain (Figure 1).

![Figure 1](image1.jpg)

Figure 1. A round mass was located on the left shoulder with a size 25 x 20 x 10 cm, showing hard consistency and non-mobile feature

Plain radiographs revealed bulging radiolucent mass with some sclerotic rim at medial side, extending from middle to proximal shaft of the left humerus. The rush nail was well fixated from proximal to distal of the humerus. There was no sign of plate and screw breakage (Figure 2).

![Figure 2](image2.jpg)

Figure 2. A soft tissue mass in the left shoulder having predominantly radiolucent density with some sclerotic rim on the medial side; The rush nail was well fixated from proximal to distal of the humerus.
CT angiography was ordered to evaluate primary vascular status, feeding artery, and the nature of the lesion. The axillary and brachial artery showed good perfusion to the distal part of the limb. The tumour was graded as Campanacci grade 3 (Figure 3).

The patient was then admitted for wide resection. The shoulder was approached through the site of the previous incision with elliptical skin incision. Some of the deltoid muscle was taken with the mass but the biceps brachii and brachialis muscles are preserved. A shoulder hemiarthroplasty using Austin Moore prosthesis with 37 mm head diameter then performed (Figure 4). The postoperative biopsy result was Giant cell tumour with no sign of malignancy.

![Figure 3. The main feeding artery from the axillary artery to the mass; The main artery showed good perfusion to the distal of the upper limb.](image1)

![Figure 4. A) Wide resection of the recurrent GCT. B) Shoulder hemiarthroplasty using Austin Moore Prostheses](image2)

However, patient developed surgical site infection six months after surgery. He had debridement and removal of the implant performed in our hospital. A culture result of *Staphylococcus Aureus* was found, and specific antibiotic treatment was given for two weeks. Four months after subsequent surgery, the patient presented a new recurrent mass at his left shoulder. The previous history before the mass was not significant. The clinical finding
showed that there was a hypertrophic scar on the lateral side of the humerus, and an irregular round mass of hard consistency and nonmobile, measuring 15x10x7 cm. At that time, MRI was performed, and the T1 weighted image showed low intensity (Figure 5).

While T2 weighted image showed heterogeneous mixed high and low signal intensity in the lesion, further workup of recurrent GCT was carried out, and the patient was admitted for wide excisional biopsy (Figure 6).

Three months after the last wide excisional biopsy, the patient came to our outpatient clinic with a chief complain of growing mass on his left humerus. Clinical presentation at the time revealed an outgrowth mass of tumour extending from the proximal to the distal part of the humerus. There is multiple areas of ulceration with some necrotic patches but no pus discharge. He could hardly move his left shoulder due to pain. The plain radiograph showed massive growth of the tumor with a mixed area of lucency and ectopic calcification. We also took the chest x-ray and it showed a coin lesion at inferior lung field of right hemithorax. He was diagnosed with recurrent GCT with lung metastasis (Figure 7). Further management was shoulder disarticulation. Left lung metastasis will be observed strictly since it was asymptomatic at that time.
Giant cell tumour is a benign, locally aggressive, neoplasm with ability to recur and metastasize. The usual site for GCT is around the knee and away from the elbow. The proximal humerus is a rare site for GCT. Xu and colleagues demonstrated that age, gender, tumour location, pathologic fracture, and types of surgery were not risk factors for recurrence. Soft tissue involvement and GCT grade 3 and up are risk factors for recurrence in GCT. Local recurrence is one of the significant factors to induce pulmonary metastasis. Giant cell tumour patients who have more than two tumour recurrences showed a risk of pulmonary metastasis equal to or even higher than that of patients with malignant GCT of the bone. Other authors proposed that surgical manipulation could facilitate tumour migration, thereby promoting lung metastasis. The mechanism of metastasizing in this benign lesion was considered the same as a malignant lesion. The suggested mechanism is local vascular permeation and emboli formation that can be induced by multiple surgical manipulations. Thus, the above facts concurred with our finding why the patient was more vulnerable to pulmonary metastasis after several operations and tumour recurrence.

The patient had to undergo multiple operations because of the demand from the patient to keep his arm for social-economic reason. The used of hip Austin Moore Prostheses implant instead of the shoulder implant has been carried out because the government is only cover for the hip implant.

In summary, preventing local recurrence of GCT for minimizing pulmonary metastasis could be done by avoiding multiple surgeries. We hope this report may have some value in understanding pulmonary metastasis of the GCT.

CONFLICT OF INTEREST
The authors have none to declare.

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REFERENCES


