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Case Report

# Allograft transplantation and shoulder replacement in management of proximal humeral osteosarcoma: A case report

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## Abstract

**Background:** Restoring and preserving the arm function in patients' with osteosarcoma is a big concern and the surgeons always aim to improve the quality of the patients' life.

**Case presentation:** A 16-year-old female patient had no special medical record history. She had felt pain in the upper arm for 2 weeks before hospitalization, the pain increased when resting and swollen the area of the proximal humerus. The patient had no history of trauma. X-rays and CT images of the patient's arm indicate a tumor in the shaft and proximal humerus (on the left side). The biopsy result confirmed the diagnosis: sarcoma of the proximal humerus. We decided to change the surgical method, did not use the shoulder disarticulation as usual. The patient was assigned to widely resect the tumor and proximal humerus allograft-prosthetic composite reconstruction to preserve arm function

**Conclusion:** Short-term results after surgery have shown that is a reasonable treatment to preserve the function of the arm. However, careful consideration should be made about the factors that affect the outcome of the treatment to avoid recurrence of the tumor.

**Keywords:** Osteosarcoma, proximal humerus, allograft, shoulder replacement

**INTRODUCTION**

Osteosarcoma is a sarcoma, which derived from osteoblasts, chondroblasts, the cells of the connective tissue of the bone, most commonly found at the proximal tibia, distal femur, proximal humerus, distal radius [1].

In the past, surgical treatment of osteosarcomas in Vietnam often led to limb amputation. This affects the limb function, aesthetics, as well as affect patient psychology, especially in young patients.

Recently, with the advancement of modern medicine, the treatment of osteosarcomas not only aims to prolong survival but also ensure aesthetic and functional rehabilitation for the patient [2].

However, reconstructing the defect of bone after removing the tumor is still a challenge to preserve the limb length, aesthetics and function, especially in the upper limb [3].

This article we would like to introduce a clinical case with osteosarcoma of the humerus, have been treated by allograft-prosthesis composite reconstruction of the proximal humerus.

**CASE PRESENTATION**

A 16-year-old female patient had no special medical record history. She had felt pain in the upper arm for 2 weeks before hospitalization, the pain increased when resting and swollen the area of the proximal humerus. The patient had no history of trauma. X-rays and CT images of the patient’s arm indicate a tumor in the shaft and proximal humerus (on the left side). The biopsy result confirmed the diagnosis: sarcoma of the proximal humerus. The patient was treated with 3 cycles of chemotherapy. Biopsy re-evaluation after chemotherapy treatment showed: Necrotic tumor tissue >90%. MRI images before surgery: the tumor did not spread to soft tissue around. The patient was consulted and indicated to remove the tumor and preserve humerus.

Clinical examinations before the surgery: The shoulder and elbow ranges of motion, as well as movement and the feeling of the patient’s left arm, were normal, compared to the right arm. The skin of the left shoulder and the left arm of the patient was normal. Evaluation of CT and X-rays showed that the tumor spreads to the proximal humerus and goes to the middle one-third of the humeral shaft (Fig. 1 and 2).

The complete removal of the tumor requires completely removed of the proximal humerus and cut it down to middle one-third of the humeral shaft. The bone segment should be removed is 18 cm in length. Meanwhile, our artificial shoulder joint only has a maximum stem is 13 cm lengths. We have decided to carry out the partial shoulder joint replacement and long-bone allograft. The allograft bone that we use was a segment of the adult Vietnamese tibia shaft, taken at the tissue bank of Hanoi Medical University.



**Fig. 1.** X-ray images before surgery

The shoulder joint was revealed through the deltopectoral approach extended to the anterolateral approach of the arm to reveal the humerus. The proximal humerus which contained the tumor was removed as planned (Fig. 3), after that, the frozen section was performed to ensure the safe resection margins. The removed part was S345 according to the classification of Musculoskeletal Tumor Society-MSTS [4,5], it complied with the recommendation that needs to remove at least 1.5 sites (Fig. 4)

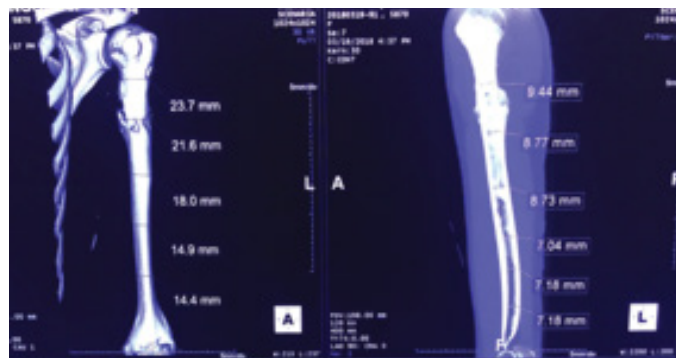
The allograft bone was measured and the length of the graft was as same as the length of the removed humeral segment (Fig. 5). Because the cartilage surface of the glenoid cavity of the scapula (in general) is good, there is no sign of invasive destruction of the tumor so we decided to perform partial shoulder replacement.

The tibial allograft was attached to the rest of the humerus with 8 holes dynamic compression plate (Fig. 6). When we were screwing into the graft, we drilled ingeniously to avoid drilling into the prosthesis stem. After the shoulder joint had been replaced, the incision was drained and closed.

After the surgery, the movement and sensory of the patient have recovered well. A week after surgery, the patient was able to lift a light object by the surgical arm. X-rays image after surgery of the patient shown that the prosthesis was positioned correctly, the humeral shaft and the graft were well fixed, ensure the axial anatomy of the arm (Fig. 7).

After surgery, the patient was instructed rehabilitation to recover the function of the arm and continuously treated by chemotherapy. The MSTS point was 23 at 2 weeks after surgery and the shoulder active abduction and adduction was 45° and 48° respectively.

Two months after the surgery, the patient felt painful and was swollen at the proximal humerus. At the upper one-third of the humerus, there are numerous bulges spreading to the most one-thirds



**Fig. 2.** CT images before surgery



**Fig. 3.** Tumor resection

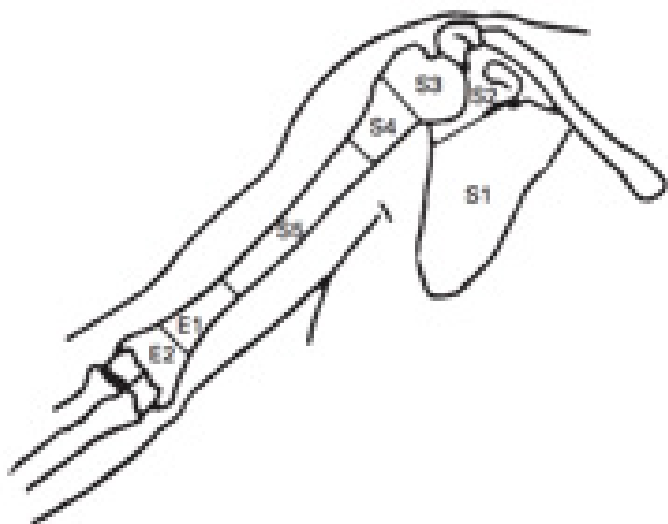


Fig. 4. S345 Part removed from arm



Fig. 5. Measured allograft bone

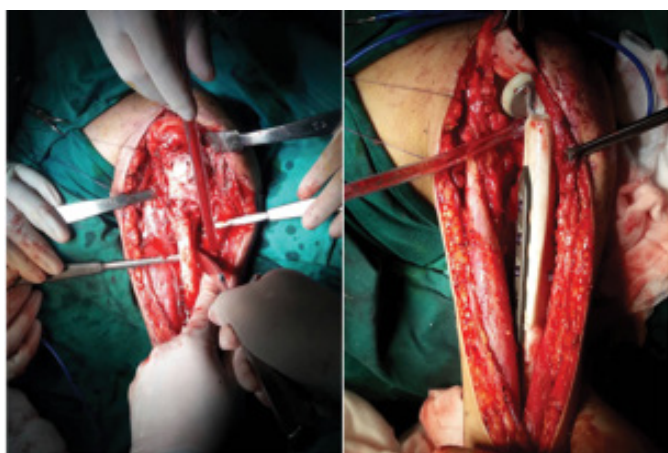


Fig. 6. Tibial allograft attached to the rest of humerus with 8 holes dynamic compression plate

upper arm, they were hard, immobile, unclear boundary and painful when palpated. The subclinical result did not detect new metastasis tissue. The patient was diagnosed recurrence of osteosarcoma and then underwent second surgery: extended resection of the recurrent cancer tissue resected 2 cm more at the humeral shaft. Frozen sections during the operation revealed no tumor cells in the resection margins.

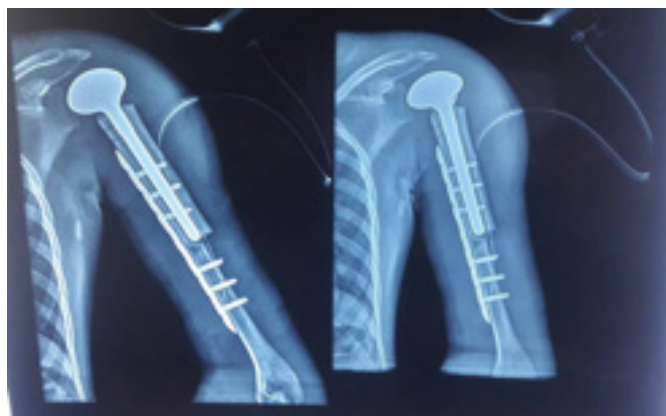


Fig. 7. X-rays images after surgery

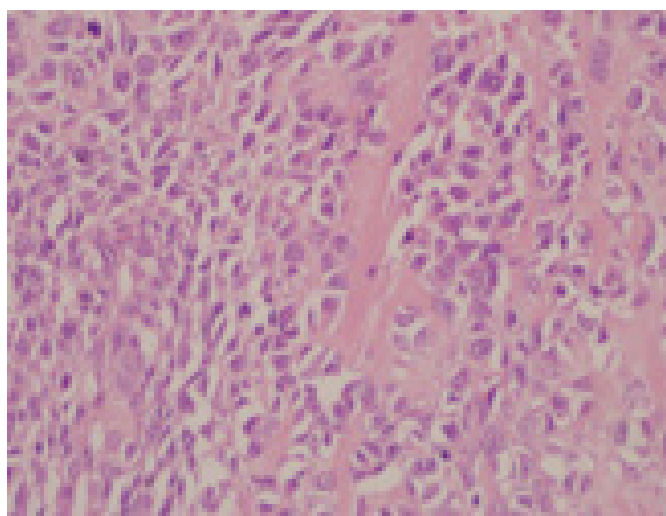


Fig. 8. Tumor biopsy

After surgery, the patient status was good. On day 14, the patient was transferred to the oncology department, Methotrexate therapy was administered at 9 doses for 3 months. However, after 3 weeks without chemotherapy, the tumor was recurrent in situ, grows rapidly, spreading over the proximal humerus, with swelling, heat, painful all over upper one-third of the humeral shaft. The tumor biopsy result was osteosarcoma (Fig. 8). The patient diagnosed with recurrence osteosarcoma of left arm, poor response to chemotherapy. Patients underwent shoulder disarticulation to prevent metastasis and the tumor from invading to the surrounding structures.

**DISCUSSION**

Osteosarcoma is common in the proximal humerus. In the early 1970s, osteosarcoma of the proximal humerus was usually treated with shoulder disarticulation, which would lead to complete loss of function of the arm. Today, with the development of preoperative chemotherapy, radiology, and surgical techniques as well as the prostheses, limb salvage surgery has become the optimal treatment option for the early and middle stage of osteosarcoma of the proximal humerus.

Preoperative chemotherapy helps to reduce the size and harden the tumor. Our patients received 3 cycles of chemotherapy after being diagnosed with osteosarcoma but the response was not very good.

Before performing a limb salvage surgery, there are many factors to consider, including whether the tumor can be removed completely or not, whether the soft tissue can be recovered or not, postoperative rehabilitation for patients, and complications may be encountered. The safe resection margin is the only way to reduce the local recurrence of the tumor. For the patient in this report, the section is determined on preoperative CT scan and actual tumor status during surgery. In

addition, the frozen section was performed to ensure safe resection margins.

While removing the tumor, the rotator cuff, capsular and glenoid labrum were not invaded by the tumor and were preserved as much as possible. The rotator cuff was sutured into the lateral holes of the prosthesis stem. We are very concerned about the strength of the shoulder, which is an important factor in the postoperative rehabilitation of the shoulder. The good recovery of the soft tissue around the joints will ensure the stability of the shoulder joint [6]. In a study that was thought to have the largest sample size of Abdeen et al., over 36 cases of shoulder allograft-prosthesis composite reconstruction, there are only 1 case of shoulder dislocation and 5 cases of subluxation of the joint (rate of instability 17%) [7].

In surgery for allograft bone transplantation, bone grafting between the allograft bone and the host bone is a very important factor. Farid Y's research showed that non-union is the most common complication (10% of patients) in surgery for joint replacement with allograft bone transplant [8]. In research by Hornicek, which we considered to be the largest sample size with 945 patients, the rate of nonunion between the allograft bone and the host bone was 17.3%. However, this rate was higher (27%) in the group of patients receiving chemotherapy. In the non-chemotherapy group, the rate of nonunion was only 11% [9]. In Abdeen's research, there are only 4 cases of delayed union (11%) and only one case of non-union (2.8%). A number of other researchers have reported the rate of non-union in allograft bone transplant of the proximal humerus is between 4%-50% [10-12].

The functional results of shoulder replacement for bone tumors in the literature are very different. Gebhardt et al. reported a very good result of the function in 67% of patients [10]. They argued that it is difficult to achieve the shoulder active abduction and adduction more than 45°. In another study of Getty and Peabody, average function points according to modified Musculoskeletal Tumor Society (MSTS) score was 70%; maximum of active abduction was 40°, active flexion and extension are also limited to similar angles [13].

Ross et al. reported that the active abduction, flexion, and extension of the shoulder joint reduced to an average of 30°, while the passive motion is almost normal, reflecting the loss of soft tissue function in this method [14]. In Abdeen's research, the mean MSTS score was 26. The mean active flexion and abduction are respectively 56° and 50°.

The MSTS score of the patient in this report was 23 at 2 weeks after surgery, the active flexion and abduction were respectively 45° and 48°, as same as Abdeen's findings.

In the first operation, the tumor was widely resected and the resection margins were safe, two months after the surgery, there was evidence of a local recurrence of the tumor. In the second surgery, we continued to resect more soft tissue and 2 cm of the humeral shaft. The frozen sections of all the resection margins were benign. After surgery, the patient continued to receive chemotherapy. However, osteosarcoma continues to recur.

According to Puri's study of 20 patients with humeral osteosarcoma who have replaced the whole humerus, the local recurrence rate was 26.3% and the 5-year survival rate was 52% [15]. Loh AH et al. also reported a study of 200 pediatric patients with an average age of 14.9 who was diagnosed osteosarcoma and underwent limb salvage [16]. These authors reported a 9% postoperative local recurrence rate, mean recurrence time was 1.4 years postoperatively. The average survival time was 11.8 months after recurrence. The authors also point out that, if the patients were excised and continued to conserve the limb, patients with the section >1 cm had a longer life expectancy than patients with the section <1 cm. In patients with amputation, the mean survival was

2.4 years after recurrence, more than that in the patients undergoing limb salvage surgery (0.86 years).

Carlos in 2004 also published a 30-year study of 26 patients with recurrent osteosarcoma. They believed that the prognosis of patients with recurrent osteosarcoma after surgery is poor, and radical resection of the tumor (amputation) after recurrence is necessary. The tumor recurrence time is also a prognostic factor of the patient. The faster of recurrence, the worse of the prognosis [17,18].

Bacci and his colleagues in 1998 also published a large-scale study of osteosarcoma 540 patients who underwent surgery and chemotherapy. The rate of local recurrence was 6%. They believed that the rate of recurrence of the tumor is not related to age, sex, tumor location and size, chemotherapy regimen and surgical methods. In general, these authors have identified two factors related to tumor recurrence prediction: the primary surgical resection margins biopsy and the response of the patient to chemotherapy. The group of patients responding well to chemotherapy had a 7-year survival rate of 95%, in the poor responding to the chemotherapy group, the incidence was 90%. Group of patients with safe resection margins had a 7-year survival rate of 97%, this rate in the group of patients with unsafe section was 71%.

In this case, the tumor recurred very quickly, with poor response to chemotherapy. So, at the third surgery, we had to remove the whole shoulder joint to remove the tumor thoroughly and avoid distant metastasis.

Another question is that with a large tumor of osteosarcoma, it is recommended that the amputation should be the optimal choice. According to a study of Poudel on 95 patients with osteosarcoma, the local recurrence rate was 15.8%, in which the initial tumor size in the recurrence group was smaller than the mean tumor size of the non-recurrent tumor group. They concluded that the tumor size was not a prognostic factor of amputation [19].

## CONCLUSION

Shoulder replacement with allograft transplantation is a relatively new surgery in Vietnam. Although this is not a complete method to avoid recurrence of cancer, however, the initial results of preserving the limb length, aesthetics and function after surgery are quite satisfactory. It brings the hope of preserving the limb for patients with osteosarcoma in general and patients with osteosarcoma of the proximal humerus in particular. However, the designation to preserve the limb of this method should be carefully considered before surgery to avoid recurrence of the tumor.

## ETHICAL APPROVAL

Ethical approval has been granted in this study.

## CONSENT

Written informed consent was obtained from the patient's legal guardian(s) for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

The patient received an explanation of the procedures and possible risks of the surgery and gave written informed consent.

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The authors declare that sponsors had no such involvement.

## CONFLICT OF INTERESTS

The authors of this article declare no conflict of interest.

## AUTHOR CONTRIBUTION

DTT contributed to perform the operation, data collection, analysis

and interpretation, manuscript drafting, revising, and approval for publishing;

MNH, SNTQ and TLK contributed to assist the operation, data collection, analysis and interpretation, manuscript drafting, revising,

and approval for publishing.

## GUARANTOR

Guarantor in this study is DTT.

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