Reconstructive Surgery in Primary Malignant and Aggressive Benign Bone Tumor of the Proximal Humerus

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——— Abstract ———————————————————————————————————

Primary malignant bone tumors of the proximal humerus have traditionally been treated by forequarter amputation. However, with the increased interest in limb salvage operations, efforts have been made to improve reconstructive surgery and some methods have become available for tumor control and preservation of a useful distal limb. This report describes three reconstructive techniques used for reconstruction of the humerus following primary tumor excision. We followed 11 patients treated by reconstructive surgery following tumor excision for primary malignant and aggressive benign bone tumors in the proximal humerus. The average follow-up period was 35.6 months. The histologic diagnosis included osteosarcoma (9), chondrosarcoma (1) and giant cell tumor (1). The options for reconstructive surgery following tumor excision were six prosthetic arthroplasties with low heat treated autobone, four arthroplasties with Ender nail and bone cement, and one arthroplasty with custom-made tumor prosthesis. We performed a retrospective analysis regarding functional status, as well as local recurrence, distant metastasis and complication. The functional status at final follow-up averaged 16 points (53.3%) overall: 17 points (56.7%) in the six prosthetic arthroplasties with low heat treated autobone; 15 points (50.0%) in two of four arthroplasties with Ender nail and bone cement (the two others died); and 16 points (53.3%) in the one arthroplasty with custom-made tumor prosthesis. Local recurrence was not observed in any of the cases. The complications noted were one nonunion between reimplanted, low heat treated autobone and the normal distal humerus and two metal failures. Each of these techniques for reconstructive surgery resulted in a relatively good outcome, although somewhat better results were found in the case of prosthetic arthroplasty with low heat treated autobone.

Key Words: Malignant bone tumor, proximal humerus, reconstructive surgery

INTRODUCTION

The proximal humerus is one of the most frequent sites of malignant bone tumor occurrence. The incidence rate of the proximal humerus in classic osteosarcoma is almost 10% overall. Malignant bone tumor at this site has resulted in severe bone destruction and instability of the upper extremity, decreasing and even obliterating the functional capacity of the shoulder joint, and even joints below that. Therefore, difficulties in treating bone tumor at this site have arisen from the fact that both a complete removal of the tumor and preservation of the functional capacity should be performed. In the earlier days,

radical amputation was mainly performed and thus caused poor results regarding function and cosmesis.

Interest in limbsalvage operations has risen for the treatment of malignant bone tumor in the proximal humerus and as a result, various reconstructive techniques for the replacement of the bone defect and preservation of joint function have been sought following wide excision. As for these reconstructive techniques, auto or allobone graft, vascularized fibular graft or arthrodesis, as well as arthroplasty using various prosthesis, have been reported.[1-6]

In this study, 11 cases of primary malignant and aggressive benign bone tumor of the proximal humerus treated using three different techniques were compared and analyzed. The cases were followed up for a minimum of 2 years. The surgical techniques included: arthroplasty using low heat treated autobone with Neer prosthesis (Global shoulder system: Depuy, Warshaw, USA); arthroplasty using Ender nails with bone cement (Depuy, Warshaw, USA); and arthroplasty using a custom-made tumor prosthesis

(Link, Hamburg, Germany).

MATERIALS AND METHODS

Materials

Eleven cases that underwent reconstructive surgery for primary malignant and aggressive benign bone tumor at the proximal humerus between August 1993 and May 1997 as well as having a follow-up period of more than 2 years were selected as the subjects of this study.

Pathologic diagnoses of all 11 cases were confirmed by preoperative incision biopsy; nine cases were osteosarcoma, one case was chondrosarcoma and the remaining case was giant cell tumor. The male to female ratio was 7 : 4 and the average age was 23.4 years (range 10-52 years).

Preoperative plain chest x-ray, chest computed tomography (CT), bone scan and magnetic resonance image (MRI) were performed. Preoperative evaluation was done by Enneking's Surgical Staging system,[6] which showed that 6 of 9 cases of osteosarcoma were stage IIB and the remaining three were stage III. The chondrosarcoma case was stage IIA and the giant cell tumor case was stage 3.

Surgical techniques

Cases with no distant metastasis and no direct invasion of the neurovascular bundle were primarily selected as indications for reconstructive surgery. In cases with distant metastasis, reconstructive surgery was performed if, as a result of chemotherapy, prolonged survival was expected and maintenance of daily living activities was desired.

Resection margins were decided based on the preoperative MRI, bone scan and plain x-ray. In addition, a frozen biopsy examined at the operation field was also taken into consideration. The deltoid muscle and axillary nerve were preserved as much as possible during the complete excision of tumor mass. The extent of rotator cuff excision was decided based on the extent of the tumor dissemination. Furthermore, the extent of the resection was classified according to the system derived by the Musculoskeletal Tumor Society.[7] On postoperative pathological reports for excised tumor mass, S34B marginal excision was per-

formed on the giant cell tumor, while wide excision was performed for the remaining cases. Eight cases had S345B wide excision and two cases had S2345B wide excision.

The resection margin was 3 cm in all cases and the average resection length was 15.8 cm from the humeral head distally. No residual tumors were observed in any of the resection margins in the postoperative pathological reports.

Preoperative chemotherapy was performed in all osteosarcoma cases and postoperative chemotherapy was performed in eight cases with the exception of one that did not respond to preoperative chemotherapy. The giant cell tumor and the chondrosarcoma cases did not undergo chemotherapy.

Our indication for Ender nail and cementation was patients with advanced stage or extraarticular resection, and that for prosthetic arthroplasty was good response to preoperative chemotherapy and young patients without distant metastasis.

Result analysis

Postoperative functional evaluation was performed using the six criteria of functional analysis of the Musculoskeletal Tumor Society and the total points were presented as a percentage (Table 1). Local recurrence, distant metastasis and complications were analyzed as well.

RESULTS

Surgical techniques

Among the 11 cases, five osteosarcoma cases and one chondrosarcoma case underwent prosthetic arthroplasty using low heat treated autobone and Neer prosthesis (Fig. 1 and 2), four osteosarcoma cases underwent arthroplasty using Ender nail and bone cement (Fig. 3) and the remaining giant cell tumor case used a custom-made tumor prosthesis.

Functional evaluation

The functional scores from the last follow-up had an average of 16.4 points (54.8%) in the remaining nine cases, with the exception of two patients who died. Seven osteosarcoma cases had an average of 16
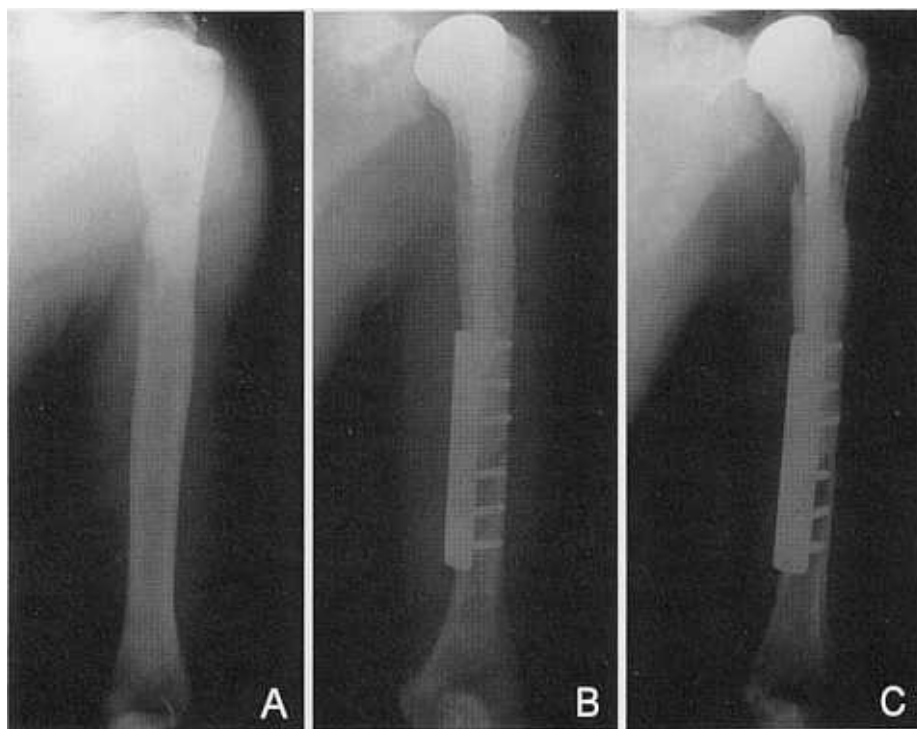
Fig. 1. Radiography of a 28-year-old male with osteosarcoma (IIB). A: A preoperative view B: An immediate postoperative view shows the reconstructive status with Neer prosthesis and low heat treated autobone after wide excision. C: A view at postoperative 2 years shows multiple bone absorptions at low heat treated autobone.
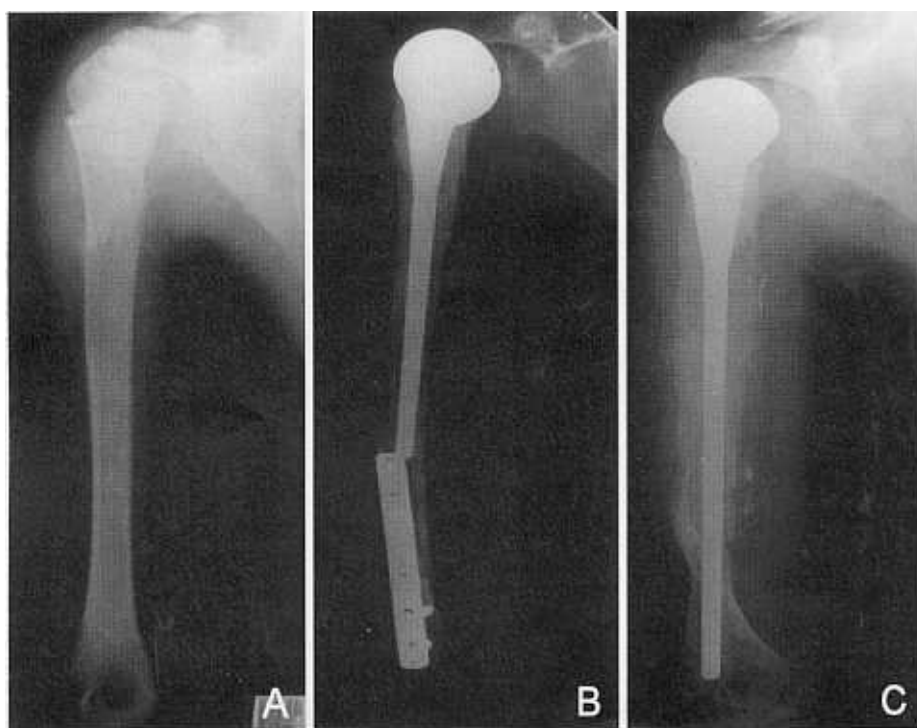

Fig. 2. Radiography of a 10-year-old boy with osteosarcoma (IIB). A: A preoperative view B: A view at postoperative 5 months shows periprosthetic fracture at distal end portion of prosthesis due to trauma after reconstructive surgery with Neer prosthesis and low heat treated autobone. C: A view at postoperative 3 years shows reoperative status with long-stem prosthesis and cement.

points (53.3%), the giant cell tumor case had 16 points (53.3%) and the chondrosarcoma case had 20 points (66.7%).

Functional scores according to the method of reconstructive surgery were as follows: the six cases using low heat treated autobone and Neer prosthesis
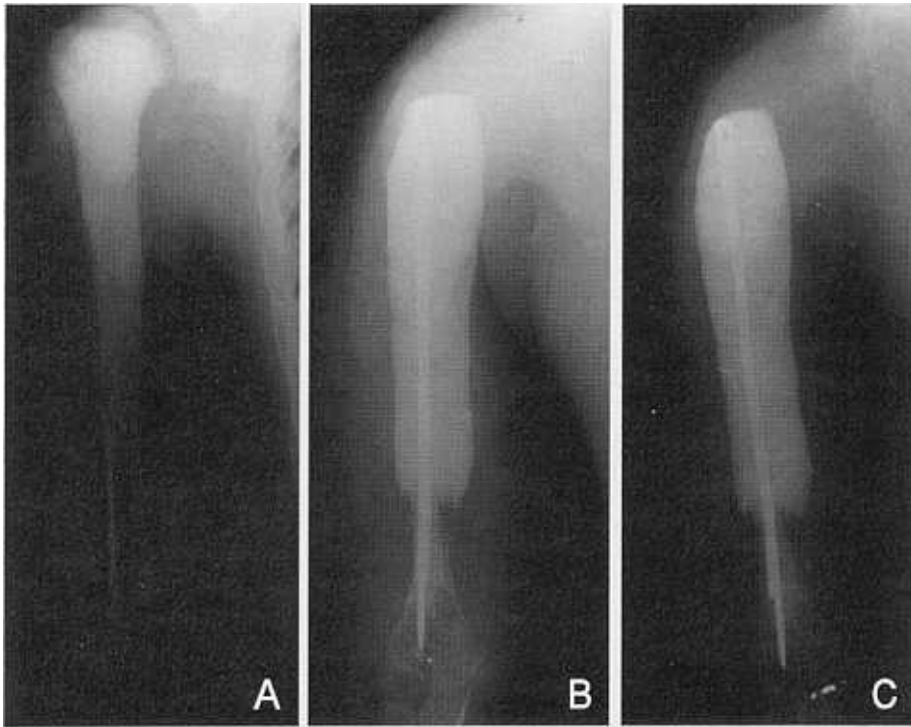
Fig. 3. Radiography of a 10-year-old boy with osteosarcoma (III). A: A preoperative view B: An immediate postoperative view shows reconstructive status with one Steinmann pin and one Ender nail and cement after wide excision. C: A view at postoperative 6 months.

had 17 points (56.7%); of the four cases using Ender nail and bone cement, two cases expired whereas the other two had 15 points (50.0%); and one case using a custom-made tumor prosthesis had 16 points (53.3 %). In view of the functional scores in each category, the scores were the highest in pain relief, but lowest in function and emotional acceptance (Table 1).

Local recurrence and distant metastasis

The follow-up period ranged from 24 months to 60 months (average 35.6 months) and no local recurrence developed during this period. Among the 11 cases, three cases of osteosarcoma had distant metastasis at initial presentation (stage III). Two of these cases (case 4 and 5) had lung metastasis and the other (case 10) had multiple bone metastases. Both cases that had lung metastasis expired during the follow-up, of which one case (case 4) had brain metastasis at postoperative 7 months. One osteosarcoma case (case 11) had lung metastasis at postoperative 18 months, and underwent wedge resection.

Complications

There were complications in three cases.

In case 11, which was the osteosarcoma case treated with low heat treated autobone and Neer prosthesis, a loosening and displacement of the plate and screws developed due to bone absorption at the junction portion of the low heat treated autobone and the normal distal humerus at postoperative 30 months. After the hardware was removed, an internal fixation with two Steinmann pins at the distal humerus was accomplished along with a wiring of the distal portion of the inserted prosthesis. Bone cement was also added to the bone defect site.

In case 7, which was also the osteosarcoma case treated with low heat treated autobone and Neer prosthesis, the fracture developed due to trauma at the distal tip portion of the inserted prosthesis at postoperative 5 months. First, cast immobilization was completed. However, a nonunion persisted for 5 months after trauma. Therefore, curettage and internal fixation with plate and wiring were performed along with brace immobilization. However, pain during exercise and evidence of a nonunion persisted for 14 months following trauma and therefore allobone graft and cast immobilization was performed following removal of the previously inserted plate and bone cement. After allobone graft and cast immobilization, nonunion and false motion persisted at the previous

Table 1. Details of Reconstructive Surgery in 11 Patients

Case	Sex /Age (years)	Diagnosis (stage)	Extent of resection	Type of surgery	Chemotherapy		Follow-up (months)	Local recur-rence	Distant metastasis (follow-up)	Functional analysis (%)
					Preop.	Postop.				
1	M/19	Giant cell tumor (3)	S34B	Custom-made tumor prosthesis	—	—	42	No	No	16 (53.3)
2	F/23	Osteosarcoma (IIB)	S345B	Neer prosthesis Low heat treated autobone	ADR CDDP	same	24	No	No	16 (53.3)
3	F/24	Osteosarcoma (IIB)	S345B	Neer prosthesis Low heat treated autobone	ADR CDDP	same	55	No	No	16 (53.3)
4	F/25	Osteosarcoma (III)	S345B	Ender nail & cement	ADR CDDP	same	8	No	Brain	Expired
5	M/10	Osteosarcoma (III)	S2345B	Ender nail & cement	ADR CDDP	MTX, cytoxan, bleomycin Acti-D	14	No	No	Expired
6	M/52	Chondrosarcoma (IIA)	S345B	Neer prosthesis Low heat treated autobone	—	—	40	No	No	20 (66.7)
7	M/10	Osteosarcoma (IIB)	S345B	Neer prosthesis Low heat treated autobone	ADR CDDP	VP-16 CDDP	40	No	No	20 (66.7)
8	M/28	Osteosarcoma (IIB)	S345B	Neer prosthesis Low heat treated autobone	ADR CDDP	same	31	No	No	16 (53.3)
9	M/16	Osteosarcoma (IIB)	S2345B	Ender nail & cement	ADR CDDP	same	38	No	No	20 (66.7)
10	M/33	Osteosarcoma (III)	S345B	Ender nail & cement	ADR CDDP, MTX Ifosphamide	—	60	No	No	10 (33.3)
11	F/17	Osteosarcoma (IIB)	S345B	Neer prosthesis Low heat treated autobone	ADR CDDP	same	40	No	Lung	14 (46.7)

ADR, adriamycin; CDDP, cisplatin; MTX, methotrexate; Acti-D, actinomycin-D; VP-16, etoposide.

fracture site during follow-up and as a result, reopera-tion was done with a long stem prosthesis and bone cement at postoperative 3 years (Fig. 2).

In case 9 which was treated with Ender nails and threaded Steinmann pins, bone cement showed metal failure at the junction portion of the normal humerus and the inserted bone cement. The hardware had to be removed and Ender nails and bone cement had to be reinserted.

In this study, six cases that received reconstructive surgery reimplanting low heat treated autobone show-ed bone absorption to different degrees at the low heat treated autobone on postoperative follow-up plain X-ray. In two out of six cases, prosthetic loose-ning and displacement occurred at the distal end. However, no cases showed dislocations or subluxation of the prosthesis in the shoulder joint and wound infections.

DISCUSSION

Great progress has been made since the mid-1980s in the treatment of malignant bone tumors as a result of evolving limb reconstruction techniques, the adju-vant chemotherapy protocol and improved criteria for the selection of patients.[1-5,8] Important criteria that should be considered in the operative treatment of

malignant bone tumor of the proximal humerus are preservation of functional capacities of the shoulder and elbow joints, along with complete removal of the tumor.[9] The limb salvage of malignant upper extremity tumors was first attempted by Tikhoff-Linberg interscapulothoracic resection, which was described by Linberg in 1928. Marcove et al. reported that the main principle for resection was that the tumor did not involve the axillary artery or brachial plexus and that there was no lymph node involvement in the Tikhoff-Linberg procedure.[10,11]

Reconstruction can also be carried out using autograft or allograft. Avascular fibular autografts have been used for many years, but they tend to fracture if the defect is longer than 12 cm and the resulting fracture may not unite.[12] Since the first success of the replacement of a massive bony defect in humans using a vascularized fibular graft was achieved by Taylor et al.[13] in 1974, it has been recommended for defects greater than 6 cm when used singly or for defects greater than 12 cm when used to supplement an avascular allograft as an adjunct in an attempt to promote early osseous union.[14] The advantage of rapid bony union regardless of the graft length has been reported in this treatment modality. However donor site complications of the vascularized fibular graft have been recently reported[15] and microvascular surgical techniques are essential for reconstruction.[16] Another autograft reconstruction method is autoclaved or pasteurized autobone graft.[17,18] Freiberg et al. and Smith and Struhl have reported no local recurrence in the replantation of autoclaved bone.[19,20] The advantages of autoclaved autogenous bone are ease of procurement, absence of problems that are associated with storage and dimensional requirements, ensured sterility, and probable avoidance of immunological response. However, the bone morphogenetic potential and biomechanical strength of the reimplanted bone is decreased, and autoclaved bone resorption is relatively frequent. Smith and Struhl[20] reported that autobone graft will improve the bony union between reimplanted, autoclaved autogenous bone and host bone, and emphasized the need for careful delineation of the indication. Low heat treated autobone can overcome these disadvantages of the autoclave method.[18] In the experimental study, autoclaved ilium of rabbits resulted in a degeneration of bone material, and the grafts were absorbed with little new bone formation and union. In contrast, the

structures of pasteurized bones were preserved and the grafts were gradually replaced with viable bone in a manner similar to that seen in fresh bone grafts. Additionally, pasteurized bone was superior to autoclaved bone in the aspect of mechanical strength and preservation of bone morphogenetic proteins.[21,22]

With the advent of newly developed prosthesis and surgical techniques, not only ablation of the neoplasm, but function of the shoulder joint, preservation of the upper extremity length and cosmesis were also of concern to oncology surgeons. Ross et al. suggested the criteria for prosthetic replacement of proximal humerus tumors as follows.[23] First, the tumor should be of low histological grade. Second, it should be intraperiosteal or have limited extraperiosteal spread without evidence of metastasis. Third, it should be confined to the upper humeral shaft with a sufficient lower half for secure fixation. The complete macroscopic excision of the tumor and minimal previous local surgery were also necessary for salvage procedure. Malawer et al. suggested that a limb sparing procedure should not be performed in a patient who has a pathological fracture, particularly if the fracture is around the pelvis.[24,25]

Currently the indications have changed, and in cases with no distant metastasis and no direct invasion of the neurovascular bundle, limb salvage procedures can be performed. In addition, it can be done for pain relief and functional improvement of the upper extremities in restricted cases of stage III.

Marcove et al. reported 17 patients whose upper extremities were salvaged with Tikhoff-Linberg procedure and who had a significant amount of hand-elbow function.[10] However, the results of implementing this surgical method brought about worse consequences when compared to the reconstructive method executed in the current series in regard to the aesthetic field and limb length preservation. Furthermore, the range of motion of the shoulder joint was significantly decreased due to the fusion of the distal humerus to the rib, performed for stability of the shoulder joint after resection.[11] It should also be considered that shortening of the upper extremity causes severe limitations of daily activities such as sports activities and impairs the use of using hand-operated devices such as a computer, particularly in the dominant side.

The functional results of limb salvage procedures depend on the size of the tumor, the extent of the

resection, the reconstructive technique, design of the prosthesis, efforts at rehabilitation, and the motivation and cooperation of the patient.[24] The choice of reconstruction technique should be based on the extent of the resection and the needs of the patient. With regard to the positioning of the hand and lifting ability, the functional results after reconstruction with an osseous arthrodesis or an osteoarticular allograft were better than those with a proximal humeral replacement prosthesis or a functional spacer.[26] The proximal humeral replacement prosthesis and spacer such as Ender nail and cementation are the least time consuming procedure of the available options and the deep infection rate is much lower than with the intercalary allograft and vascularized fibular graft.[26,27] Moreover, it offers immediate distal fixation and the ability to administer radiation therapy in the early postoperative period. The Ender nail and cementation technique also has the advantage of cost effectiveness. In this series, the average medical expense of the Ender nail and cementation was less than that of prosthesis implantation (420 dollars versus 7085 dollars). Furthermore, the operative time was decreased by an average of one hour in cases using Ender nail and bone cement compared to other methods.

Additionally, the spacer can be converted to other options and it is acceptable to the Korean situation in that the allobone graft is only available by importation. If the patient is elderly, financially limited and the expected survival time is short, then the Ender nail and cementation method would be the treatment of choice. The functional score was low in this series, but that could be because of the short follow-up period. The functional score was 26 points (87%) by Malawer et al.[24] and this was the score at which the reconstruction was performed by modular, custommade and Neer prosthesis in 29 patients with a median follow-up of three and one-half years and the resection margins were not in detail. The functional score by O'Connor et al.[26] was 52% in two patients by S34B or S345B resection and proximal humeral replacement prosthesis, while in six patients with S234B or S2345B resection and functional spacer, the functional score was 54%. Either the Dacron aortic graft or Mersilene tape used for soft tissue reconstruction by Asavamongkolkul et al.[28] showed that the functional rating was 76% in 17 proximal humeral prosthetic replacements. For Jeon et al.[27] of Korea, the functional score was 22.5 (75%) overall, while in

the cases of S345B resection that were most frequent in our series, it was 21.7 (72%). We considered that the functional evaluation results would differ according to the length of follow-up, active range of motion exercise and the degree of muscle and soft tissue resection. By minimizing the amount of resection of muscle and soft tissue within the complete removal of the tumor and by appropriate reconstruction, complications like subluxation or dislocation after arthroplasty will decrease and the postoperative joint function can be preserved maximally following reconstructive surgery.[9,28,29] Damron et al. reported that the function of the distal portion (elbow, forearm, and hand) generally was excellent, regardless of the type of upper extremity shoulder reconstruction.[30] Thus, the ultimate choice of limb salvage technique in upper extremity reconstruction comes down to the preference of surgeon and patient. Therefore, patient age, occupation, prognosis and financial status of the patient should be considered in the selection of surgical reconstruction of the upper extremity.

In this study, we gained relatively positive results for each technique used in the limb reconstructive surgery and better results than others in arthroplasty using low heat treated autobone and prosthesis. However, there was no statistical significance because of the small number of cases. In patients with a poor prognosis, reconstructive surgery with Ender nail and bone cement was thought to be cost-effective. Finally, in the surgical treatment of malignant bone tumor of the proximal humerus, the patient's age and functional condition, the stage and progression of tumor, the extent of bone destruction, and the degree of soft tissue invasion must all be considered. The appropriate surgical technique must be selected based on these factors and it is considered that the remaining function of the preserved joint, dissociation of prosthesis, nonunion and fracture between reimplanted autobone and normal host bone, tumor recurrence and complication must be analyzed through long term follow-up.

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