ANTICANCER RESEARCH 42: 2139-2144 (2022)
doi:10.21873/anticanres.15696

Clavicula Pro Humero Reconstruction for Malignant Tumor of the Proximal Humerus in Children and Adults

HIDEYUKI KINOSHITA[1], HIROTO KAMODA[1], YOKO HAGIWARA[1], TAKESHI ISHII[1], SEIJI OHTORI[2] and TSUKASA YONEMOTO[1]

[1]*Department of Orthopedic Surgery, Chiba Cancer Center, Chiba, Japan;*
[2]*Department of Orthopaedic Surgery, Graduate School of Medicine, Chiba University, Chiba, Japan*

Abstract. *Background/Aim: The effectiveness of clavicula pro humero (CPH) reconstruction for pediatric proximal humerus sarcoma has been reported in a small number of cases. We aimed to investigate the effectiveness of biological CPH reconstruction for malignant bone tumors of the proximal humerus in children and adults. Patients and Methods: This was a retrospective cohort study that included eight patients who underwent CPH reconstruction due to a malignant bone tumor around the proximal humerus. Postoperative parameters, including complications, postoperative upper limb function, and the period until bone fusion, were investigated. Results: Three patients had non-union and fracture of the clavicular segment. Among them, one patient underwent revision surgery for internal fixation and bone grafting. Five patients achieved bone fusion, and the overall mean Musculoskeletal Tumor Society score was 70%, which is comparable to previous reports. Conclusion: CPH reconstruction is an effective technique for malignant bone tumors of the proximal humerus in all ages.*

The proximal humerus is a common site in both sarcoma and metastatic malignant bone tumors (1). The standard treatment for malignant bone tumors around the proximal humerus is wide resection and reconstruction (2). These reconstructions include prosthesis, arthrodesis, and vascularized fibula graft, among which clavicula pro humero (CPH) reconstruction for malignant bone tumors was found to be effective (3). CPH reconstruction is a biological technique for the proximal humerus, with the ipsilateral clavicle being rotated, and was originally introduced as a technique for congenital upper limb deficiency (4). The effectiveness of CPH reconstruction for pediatric malignant tumors of the proximal humerus has been reported but only in a few cases (5). There are even fewer reports on the efficacy of CPH reconstruction in adults.

In the current study, the efficacy of CPH reconstruction for malignant bone tumors of the humerus in children and adults, including complications, postoperative upper limb function, and period until bone fusion, was investigated.

Correspondence to: Hideyuki Kinoshita, Department of Orthopedic Surgery, Chiba Cancer Center, 666-2 Nitonacho, Chuo-ku, Chiba 260-8717, Japan. E-mail: kinoshi1783@yahoo.co.jp

Key Words: Clavicula pro humero reconstruction, malignant bone tumor, non-union, sarcoma.

Patients and Methods

Study design and patients. Approval of the Chiba Cancer Center Review Board and written informed consent from each patient prior to inclusion were obtained before initiating the study (Approval number: R03-177). We retrospectively investigated our Institution's database to identify patients who underwent CPH reconstruction for malignant bone tumors of the proximal humerus between January 2010 and December 2020. We identified eight patients who underwent CPH reconstruction due to a malignant bone tumor around the proximal humerus at our Institution. Patient demographics, including sex, age at operation, follow-up period after surgery, body mass index, histology, and chemotherapy are presented in Table I. Of the eight patients, one was only followed-up for 1 year due to relocation (patient 5).

Surgical procedure. The incision extended from the proximal side of the clavicle, across the coracoid process, extending to the distal end of the humerus tumor and included the biopsy tract (Figure 1A). The tumor and surrounding soft tissue were carefully resected en bloc. All eight cases were resected intra-articularly. Surgical margins were negative in all cases. The clavicle was mobilized by releasing its medial tissue, leaving its attachments to the acromion intact. The floating clavicle was rotated by approximately 90˚ (Figure 1B). In reconstruction of the bone, as one case (patient 1) was an infant patient and the humerus was short, total resection of the humerus and radial nerve resection were performed. Elbow joint reconstruction using the rotated clavicle and fibular head and nerve reconstruction was necessary. Another case (patient 2) required a vascularized fibular autograft, which was harvested from the ipsilateral leg to augment the length of the clavicle segment. In the other six cases, the rotated clavicle was fixed to the distal humerus with a plate. Two patients required axillary

Table I. *Patient demographics and disease characteristics of patients undergoing reconstruction for malignant tumor of the proximal humerus.*

Characteristics	Value
Sex	
Male	4
Female	4
Age at operation, years	
Mean±SD	39±24.7
Follow-up period after surgery, months	
Mean±SD	35.5±16.2
Body mass index, kg/m^2	
Mean±SD	21.2±2.9
Histology	
Chondrosarcoma	3
Osteosarcoma	2
Undifferentiated pleomorphic sarcoma	1
Renal carcinoma	1
Thyroid carcinoma	1
Chemotherapy	
Perioperative	3
Postoperative	1
No	4

SD: Standard deviation.

nerve resection. Suture repair of the coracoid process fragment was performed at the time of closure. A *latissimus dorsi* free flap was required for closure in one case (patient 5). Shoulder joint orthosis was used for 4-6 weeks after postoperative immobilization. Shoulder and elbow range of motion exercises were then initiated under the guidance of a physical therapist. Two patients (patients 4 and 7) underwent low-intensity pulsed ultrasound to induce bone fusion.

Parameters for investigation. Intraoperative and postoperative results included operative time, amount of bleeding, surgical margin, postoperative complications, revision surgery, and disease status. The surgical margin was microscopically categorized: A positive margin (R1 resection) was defined as the presence of tumor cells at the closest margin, and a negative margin (R0 resection) was defined as the absence of tumor cells at the margin. Furthermore, the characteristics of eight patients and details of CPH reconstruction, including nerve resection, length of humeral defect, clavicular segment, and interposition material, were investigated. In addition, the time until bone fusion, complications, local recurrence, revision surgery, and Musculoskeletal Tumor Society (MSTS) score were evaluated. The MSTS score for limb salvage evaluation was a maximum of 5 for pain, function, emotional acceptance, positioning of the hand, manual dexterity, and lifting ability, respectively, and the sum of all items was calculated as a percentage of the total 30 points.

Statistical analysis. Patient data were consecutively entered into our database. All data were collected from central electronic medical records at our Institution. All analyses were performed using SAS software, version 14.2 (SAS Institute, Inc., Cary, NC, USA).

Results

Patient demographics. Data regarding patient demographics and disease characteristics are presented in Table I. Four patients were male, and four were female. The mean age at operation and follow-up period after surgery was 39 years (range=10-69 years) and 35.5 months (range=12-69 months), respectively. The mean body mass index was 21.2 kg/m^2 (range=16.7-25.3 kg/m^2). The histological tumor subtypes were chondrosarcoma (three cases), osteosarcoma (two cases), undifferentiated pleomorphic sarcoma (one case), renal carcinoma (one case), and thyroid carcinoma (one case). There were three and one patient who underwent perioperative and postoperative chemotherapy, respectively.

Intraoperative and postoperative results. The complete intraoperative and postoperative results are presented in Table II. The mean overall operative time for CPH reconstruction was 380 min (range=281-559 min). The mean overall blood loss during CPH reconstruction was 414 ml (range=150-980 ml). The surgical margins of all cases were R0, suggesting complete excision, both grossly and microscopically. Postoperative complications included non-union in two cases and fracture of the clavicular segment in two cases. Of the two patients with fractures of the clavicular segment, one patient with non-union and fracture did not want to undergo revision surgery because of lack of pain. The other patient had revision surgery for internal fixation and bone graft, followed by bone fusion 41 months after primary surgery. One patient had local recurrence of osteosarcoma, necessitating an interscapulothoracic amputation. At study end, the disease status was disease-free for three cases and died of disease for four cases. One patient was lost to follow-up because of relocation.

Table III shows the characteristics of each of the eight patients and details of CPH reconstruction. As the osteosarcoma and chondrosarcoma of the proximal humerus were large in patients 1 and 2, vascularized fibular autografts were needed for reconstruction. In addition, as patient 1 was 10 years old and the humerus was short, total resection of the humerus and radial nerve resection were performed, and leading elbow joint reconstruction using the fibular head and nerve reconstruction was necessary. Table IV indicates the results of postoperative bone fusion, complications, local recurrence, and function in each of the eight patients. The period until bone fusion after primary surgery ranged from 5 to 41 months and the mean MSTS score was 70% (range=60-80%). Figure 2 shows the postoperative radiographs of each of the eight patients. Furthermore, Figure 3 presents postoperative radiographs of three cases with complications, including non-union and fracture of the clavicular segment in patients 2, 4 and 6. The proximal portion in reconstruction with vascularized fibular autograft of patient no. 2 was non-

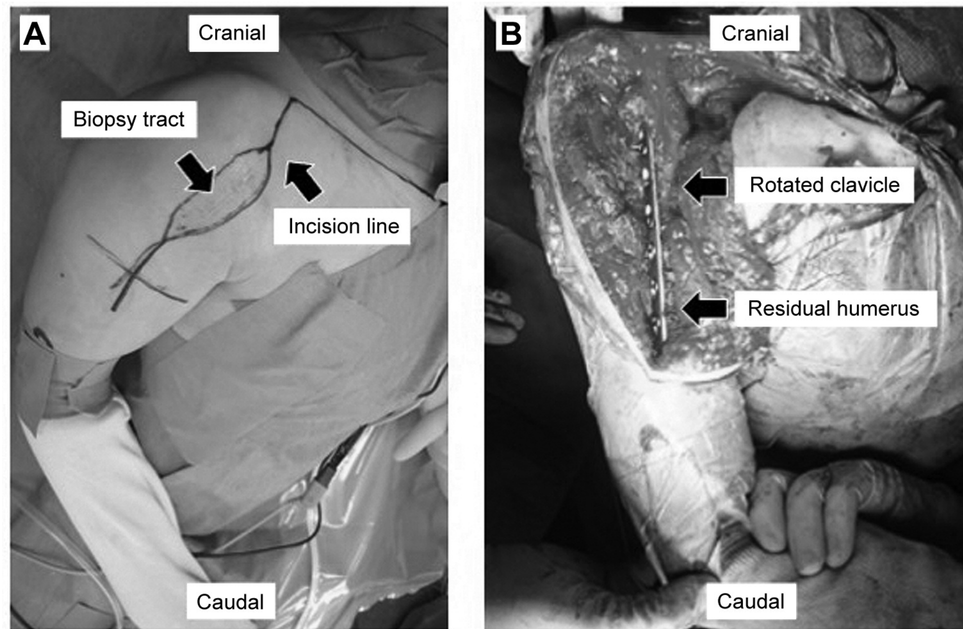

Figure 1. *Intraoperative photographs of the clavicula pro humero reconstruction. A: Frontal view of the preoperative skin incision line. B: Intraoperative photograph of the clavicula pro humero reconstruction. The rotated clavicle and residual humerus were fixed by an internal plate.*

union. In contrast, the distal portion achieved bone fusion at 25 months after surgery (Figure 3A). The 10-year-old patient (no. 4) had a fracture of the clavicular segment 3 months after surgery, leading to revision surgery for internal fixation and bone grafting (Figure 3B). Patient 6 had a fracture of the clavicular segment and non-union at 5 months after surgery. However, there was no need for revision surgery because of a lack of both pain and disability (Figure 3C).

Discussion

CPH reconstruction is a biological technique used for reconstruction of the proximal humerus after wide resection of a malignant tumor, with removal of the rotator cuff and part or all of the deltoid muscles. Other reconstructions for malignant tumors of the primal humerus include prosthesis, arthrodesis, and vascularized fibula graft (6). Recently, a reverse shoulder prosthesis without an allograft for proximal humerus malignant tumors was reported to achieve a stable shoulder and functionally satisfying results (7). However, complications of prosthesis of the proximal humerus after wide resection include infection, shoulder subluxation, and prosthetic loosening (8). Among them, infection at the prosthesis site is a severe complication, sometimes leading to several reoperations and prosthesis removal. CPH reconstruction has a lower risk of infection due to its biological property as compared to prosthesis. In fact, there were no infections as complications in the current study. The range of motion of the joints in this study tended to be smaller than in previous reports, with an average for shoulder abduction in this study and previous reports of 30˚

Table II. *Intraoperative and postoperative results for reconstruction for malignant tumor of the proximal humerus.*

Characteristics	Value
Intraoperative outcomes, mean±SD	
Operative time, min	380±91
Bleeding, ml	414±263
Surgical margin, n	
R0	8
R1	0
Postoperative complications, n	
Non-union	2
Fracture	2
Revision surgery, n	
Internal fixation and bone graft	1
Interscapulothoracic amputation	1
Disease status, n	
CDF	3
DOD	4
Unknown	1

CDF: Continuous disease-free; DOD: died of disease; SD: standard deviation.

Table III. *Characteristics of eight patients and details of clavicula pro humero reconstruction.*

Patient number	Age at surgery, years	Histology	Chemotherapy	Nerve resection	Length of humeral defect, cm	Length of clavicular segment, cm	Interposition material
1	10	Osteosarcoma	Perioperative	Radial nerve	26	12	Vascularized fibular autograft, 14 cm
2	66	Chondrosarcoma	None	None	23	13	Vascularized fibular autograft, 10 cm
3	67	Thyroid carcinoma	None	Axillary nerve	13	13	None
4	10	Undifferentiated pleomorphic sarcoma	Perioperative	Axillary nerve	13	13	None
5	24	Chondrosarcoma	None	None	11	11	None
6	69	Renal carcinoma	None	None	13	13	None
7	49	Chondrosarcoma	Postoperative	None	11	11	None
8	17	Osteosarcoma	Perioperative	None	13	13	None

Table IV. *Results of postoperative bone fusion, complications, local recurrence, revision surgery and function after clavicula pro humero reconstruction.*

Patient number	Time to bone fusion, months	Complications	Local recurrence	Revision surgery	MSTS, %
1	8	None	Yes	IST amputation	60
2	Proximal: Non-union Distal: 25	Non-union	No	None	76
3	5	None	No	None	73
4	41	Fracture at clavicular segment	No	Internal fixation and bone graft	63
5	Unknown	None	No	None	66
6	Non-union	Non-union + fracture at clavicular segment	No	None	63
7	11	None	No	None	80
8	10	None	No	None	73

IST: Interscapulothoracic; MSTS: Musculoskeletal Tumor Society Score.

and 63˚, respectively (9). However, the results for the MSTS score of CPH reconstruction were comparable to those of previous reports, at 70% and 69%, respectively (3).

In the current study, postoperative complications occurred in three out of the eight patients with pseudoarthrosis and associated fractures. Malignant bone tumors often have poor postoperative bone healing due to the use of perioperative chemotherapy. In fact, four patients in this study also underwent perioperative chemotherapy, and in these patients, bone healing was not achieved during chemotherapy, but was achieved promptly after the completion of chemotherapy. Therefore, rehabilitation should be performed with attention to fracture during chemotherapy, and aggressive rehabilitation should be performed when bone healing is achieved after the completion of chemotherapy. Furthermore, we consider CPH reconstruction as a good indication for chondrosarcomas that are chemoresistant and do not receive perioperative chemotherapy. In addition, since bone healing was eventually achieved in the two patients who underwent low-intensity pulsed ultrasound, it may be necessary to consider these adjuvant therapies. Furthermore, the case of the 10-year-old patient (no. 4) in the current study had a fracture of the clavicular segment, requiring revision surgery for bone grafting; since the clavicle is extremely thin in pediatric patients, the diameter and number of screws had to be considered carefully. There was one case of postoperative local recurrence despite R0 resection, which required interscapulothoracic amputation. Preoperative evaluation of appropriate margins and careful wide resection of high-grade sarcomas may be necessary.

As CPH reconstruction was originally introduced as a treatment for congenital upper limb deficiency, it is often

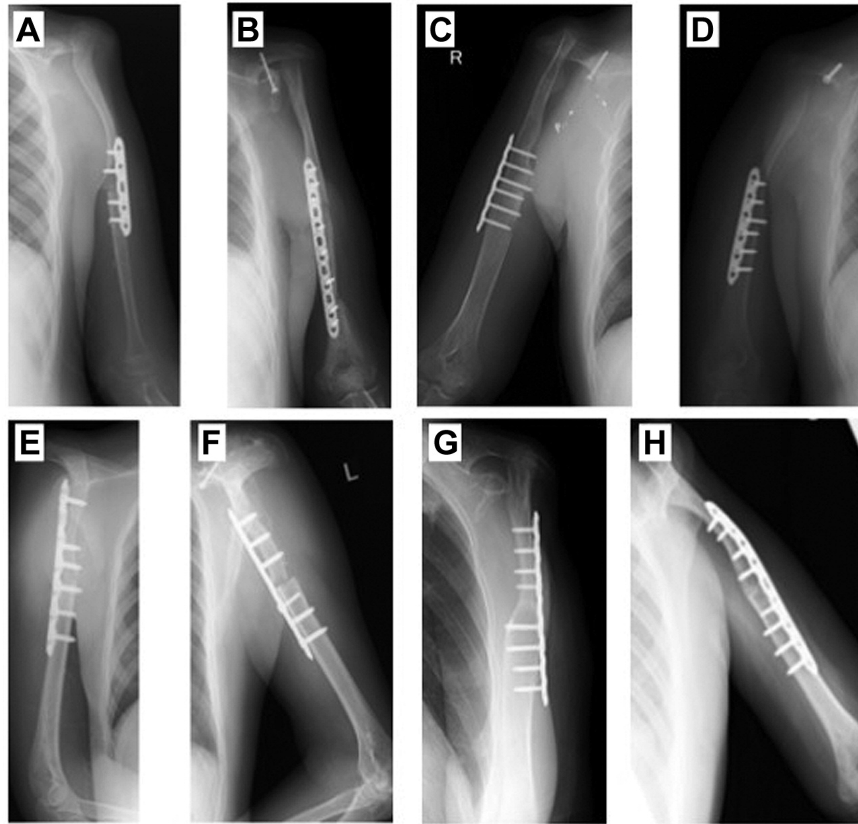

Figure 2. *Postoperative x-ray of patients 1-8, respectively. A: The total humerus was removed and reconstructed with only the clavicle being rotated and with vascularized fibular autografts. The elbow joint was reconstructed with a fibular head. B: Clavicula pro humero reconstruction was performed with a rotated clavicle, vascularized fibular autograft and residual humerus. C-H: Reconstruction was performed with only a rotated clavicle and residual humerus.*

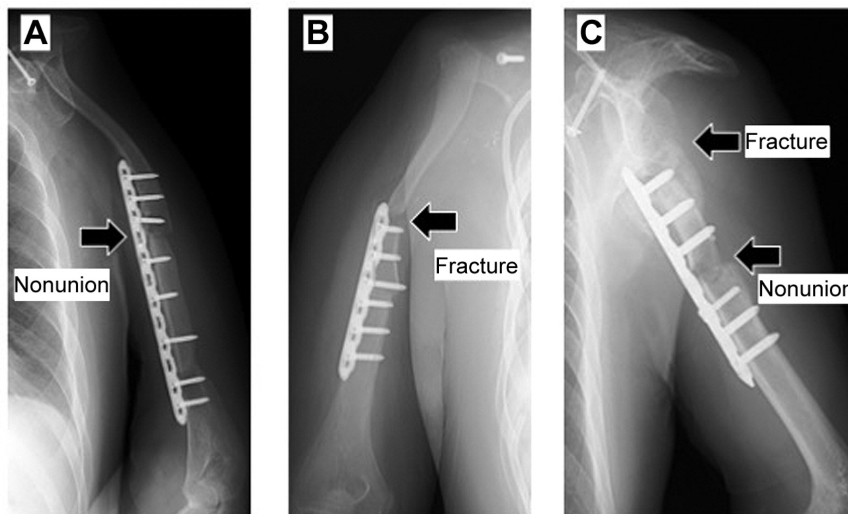

Figure 3. *Postoperative X-ray of three cases with complications. A: Patient 2: Proximal portion in reconstruction with vascularized fibular autograft did not achieve union but the distal portion achieved bone fusion at 25 months after surgery. B: Patient 4 had a fracture of the clavicular segment at 3 months after surgery, leading to revision surgery for internal fixation and bone graft. C: Patient 6 had a fracture of the clavicular segment and non-union at 5 months after surgery. However, there was no need for revision surgery due to a lack of both pain and disability.*

performed in children for malignant tumors of the proximal humerus (10). There have been a few reports on the effectiveness of CPH reconstruction in children but there have been even fewer reports including adults. Since cancer metastasis often occurs in the elderly, but sarcoma is also common in adolescents and young adults, evaluation of CPH reconstruction in malignant bone tumors of the humerus in various generations is considered necessary. In this study, we examined the efficacy of CPH reconstruction from children to the elderly and demonstrated its effectiveness. This study included one of the largest numbers of reported cases of CPH reconstruction in different generations (3, 11, 12).

However, this study has several limitations. Firstly, this was a retrospective study with a small number of cases. Secondly, due to the small number of cases, we were unable to compare the results by group.

In conclusion, CPH reconstruction is an effective technique, not only for children but also for malignant bone tumors of the proximal humerus in older individuals.

Conflicts of Interest

The Authors have no conflicts of interest directly relevant to the content of this article.

Authors' Contributions

HK designed and performed experiments, analyzed data, and wrote the article; HK, YH, TI, SO and TY provided technical support and conceptual advice.

Acknowledgements

This work was supported by JSPS KAKENHI grants (19K16760) and the Hamaguchi Foundation for the Advancement of Biochemistry.

References

1 Kinoshita H, Yonemoto T, Kamoda H, Hagiwara Y, Tsukanishi T, Inoue M, Terakawa F, Ohtori S and Ishii T: Effectiveness of salvage knee rotationplasty on sarcoma around the knee in adolescents and young adults. Anticancer Res *41(2)*: 1041-1046, 2021. PMID: 33517313. DOI: 10.21873/anticanres.14860

2 Rödl RW, Gosheger G, Gebert C, Lindner N, Ozaki T and Winkelmann W: Reconstruction of the proximal humerus after wide resection of tumours. J Bone Joint Surg Br *84(7)*: 1004-1008, 2002. PMID: 12358362. DOI: 10.1302/0301-620x.84b7.12989

3 Tsukushi S, Nishida Y, Takahashi M and Ishiguro N: Clavicula pro humero reconstruction after wide resection of the proximal humerus. Clin Orthop Relat Res *447*: 132-137, 2006. PMID: 16467622. DOI: 10.1097/01.blo.0000201169.80011.ff

4 Sulamaa M: Upper extremity phocomelia. A contribution to its operative treatment. Clin Pediatr (Phila) *2*: 251-257, 1963. PMID: 13979184. DOI: 10.1177/000992286300200511

5 Calvert GT, Wright J, Agarwal J, Jones KB and Randall RL: Is claviculo pro humeri of value for limb salvage of pediatric proximal humerus sarcomas? Clin Orthop Relat Res *473(3)*: 877-882, 2015. PMID: 25057117. DOI: 10.1007/s11999-014-3814-4

6 Armangil M and Bilgin SS: Reconstruction of proximal humeral defects with shoulder arthrodesis using free vascularized fibular graft: surgical technique. JBJS Essent Surg Tech *3(2)*: e8, 2013. PMID: 30881739. DOI: 10.2106/JBJS.ST.L.00034

7 Guven MF, Aslan L, Botanlioglu H, Kaynak G, Kesmezacar H and Babacan M: Functional outcome of reverse shoulder tumor prosthesis in the treatment of proximal humerus tumors. J Shoulder Elbow Surg *25(1)*: e1-e6, 2016. PMID: 26234664. DOI: 10.1016/j.jse.2015.06.012

8 Shi SF, Dong Y, Zhang CL, Bao K and Ma XJ: [Prosthesis replacement of the proximal humerus after the resection of bone tumors]. Chin J Cancer *29(1)*: 121-124, 2010. PMID: 20038324. DOI: 10.5732/cjc.009.10287

9 Barbier D, De Billy B, Gicquel P, Bourelle S and Journeau P: Is the clavicula pro humero technique of value for reconstruction after resection of the proximal humerus in children? Clin Orthop Relat Res *475(10)*: 2550-2561, 2017. PMID: 28699149. DOI: 10.1007/s11999-017-5438-y

10 Okimatsu S, Kamoda H, Yonemoto T, Iwata S and Ishii T: Effectiveness of clavicula pro humero reconstruction for elderly patients: report of two cases. Case Rep Oncol Med *2016*: 4140239, 2016. PMID: 27847662. DOI: 10.1155/2016/4140239

11 Kitagawa Y, Thai DM and Choong PF: Reconstructions of the shoulder following tumour resection. J Orthop Surg (Hong Kong) *15(2)*: 201-206, 2007. PMID: 17709862. DOI: 10.1177/230949900701500216

12 Nishida Y, Tsukushi S, Yamada Y, Kamei Y, Toriyama K and Ishiguro N: Reconstruction of the proximal humerus after extensive extraarticular resection for osteosarcoma: a report of two cases with clavicula pro humero reconstruction. Oncol Rep *20(5)*: 1105-1109, 2008. PMID: 18949408.

Received January 27, 2022
Revised February 17, 2022
Accepted February 21, 2022