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Case Report

Reverse total shoulder arthroplasty with allograft bone block augmentation following proximal humerus sarcoma resection: a novel technique and case report of two patients

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ABSTRACT

There are several options for endoprosthetic replacement of the proximal humerus following oncologic resections. Reverse total shoulder arthroplasty (RTSA) has gained popularity in recent years for reconstruction, and bone allograft can be used to restore bone stock and improve implant survival. Patient A (67 years female) and patient B (62 years female) with sarcoma of the proximal humerus underwent tumor resection with wide margins and reconstruction with cemented reverse total shoulder arthroplasty. The construct was augmented with allograft bone blocks to decrease burden of osteotomy length and bone defect and improve prosthesis coverage and stability. RTSA with bone block augmentation is functionally stable and effective following massive tumor resection. Controversy still exists on the ideal construction method and more research is needed for assessment of outcomes and appropriate patient selection.

Keywords: Proximal humerus tumor, Sarcoma, Reverse total shoulder arthroplasty, Allograft-prosthesis composite, Bone augmentation

INTRODUCTION

The proximal humerus is the third most common site of primary bone sarcoma whereas soft tissue sarcomas of the upper extremity are rare and comprise less than 1% of all malignant tumors.[1,2] Sarcomas are associated with significantly high mortality rates in cases of distant metastasis or neurovascular invasion with bone and soft tissue involvement; hence why an early intervention is deemed critical.[3]

Limb salvage surgery is the cornerstone of treatment of proximal humerus sarcoma. Wide resection with tumor-free surgical margins is crucial for limb preservation.[4] Radical resection, however, requires concomitant extensive bone and soft tissue sacrifice and reconstruction henceforth. Multiple treatment options exist for endoprosthetic replacement of the proximal humerus following oncologic resections, including but not limited to hemiarthroplasty (HA), reverse total shoulder arthroplasty (RTSA), allograft-prosthetic composite (APC), osteoarticular allograft implantation, fibula autograft transplantation, among others.[2,5]

Reverse total shoulder arthroplasty has gained popularity in recent years for reconstruction in aims to preserve a stable functional shoulder. Following tumor resection, a large bone defect requires bone allograft to restore bone stock and improve implant survival.[6] The cases at hand present a novel way of bone graft augmentation with RTSA reconstruction. Both oral and written informed consents were obtained regarding the case study and future publications.

CASE REPORT

Patient A is a 67-years-old hypertensive female presented in 2022 for two and half months of right shoulder pain increasing in severity and limiting range of motion of shoulder and wrist. X-ray of the shoulder showed a lytic

lesion of the left proximal humerus (Figure 1) which on magnetic resonance imaging (MRI) was found to be aggressive, expansile lesion involving the proximal humerus extending from the epiphysis of the proximal shaft causing cortical destruction and erosion laterally with significant irregular calcified matrix (Figure 2). Biopsy of the humerus showed chondrosarcoma. On physical examination, patient had severe tenderness and marked limitation of shoulder adduction and internal rotation.

Figure 1: X-ray of the right shoulder of patient A showing aggressive, expansile lesion involving the proximal humerus extending from the epiphysis causing cortical destruction and erosion laterally. There is significant irregular calcified matrix.

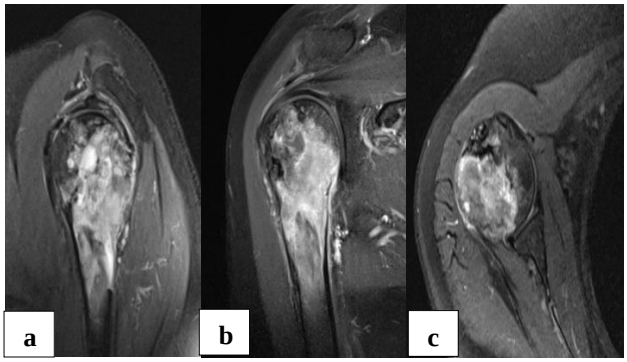

Figure 2 (a-c): MRI of the right shoulder of patient A showing aggressive, expansile lesion involving the proximal humerus causing cortical destruction and erosion with areas of both narrow and wide zone of transition and irregular calcified matrix. The lesion has variable signal with no lobulated bright signal areas.

Patient B is a 62-years-old female previously healthy presented to our care in September 2024 for persistent left shoulder pain, swelling and restriction of movement. The history extends to 2 years prior to presentation when the left shoulder pain started with shoulder movement and medical advice at a different institute recommended pain control and multiple cortisone injection (x8) which did not improve her pain. Since then, the patient reports a gradual restriction in shoulder arc of motion that she could no longer move her left upper extremity and needed right hand assistance in moving the left extremity.

Investigation was initiated at the same institute and MRI in June 2024 revealed a mass in her left shoulder. A biopsy of the tumor showed spindle cell tumor consistent with low grade fibrosarcoma; surgical excision of the tumor followed, cement was applied and proximal humerus fixed with plates and screws (Figure 3). She didn't undergo any chemotherapy or radiotherapy after the surgery. 1 month later, and on repeat imaging, MRI showed heterogeneously enhanced large aggressive bone tumor of the proximal humerus with infiltration of the shoulder joint capsule measuring 6.5×6.5×5.7 cm abutting but not engulfing the neurovascular bundle; in addition to edematous changes of rotator cuff muscles (supraspinatus, infraspinatus, subscapularis), and to a lesser extent the deltoid and teres major muscles (Figure 4). On physical examination, sensation was intact but she was tender to palpation of humerus with limited range of motion of shoulder globally and on elbow extension.

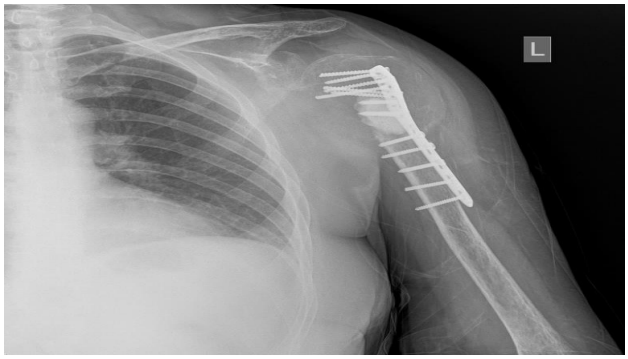

Figure 3: X-ray of left humerus of patient B preoperatively showing prior lateral plate and screw fixation of the proximal humerus extending from the head to the mid shaft. There is a large area of bone loss and destruction proximally at the level of the proximal meta-diaphysis corresponding to the tumor. Presence of cement of 3.6 cm in craniocaudal extent with overlying medial bone loss. No definite calcified matrix could be identified.

In view of the clinical history and MRI findings attributed to sarcoma in patient A and either residual tumor or recurrence in patient B, discussion between medical team and the patient/family yielded a decision to proceed with wide margin resection, i.e. 1 cm margins, of left proximal humerus tumor and reconstruction using cemented reverse total shoulder arthroplasty and allograft bone block augmentation. Preoperative laboratory tests were normal. Intraoperatively, and after deltopectroral approach in patient A and elliptical Incision including previous surgical incision in patient B, we performed thorough dissection of the cephalic vein, deltoid muscle, axillary nerve and artery, musculocutaneous nerve, rotator cuff muscles and conjoint tendon which was sacrificed along with remarkable portions of coracobrachialis, biceps and rotator cuff muscles as part of wide margin resection and dissection around the tumor. The tumor was then dissected and stripped circumferentially, including ligation of vascular feeders, and resected with wide margins. In

patient B, previously applied proximal humerus hardware was identified, and distal screws were removed then humerus shaft osteotomy was performed at 11 cm from proximal humerus tip and resected en bloc along with the hardware (Figure 5). In patient A, an osteotomy was performed at 6.5 cm from the tip of the humeral head. Intra-op frozen section for tumor margins were negative in both patients.

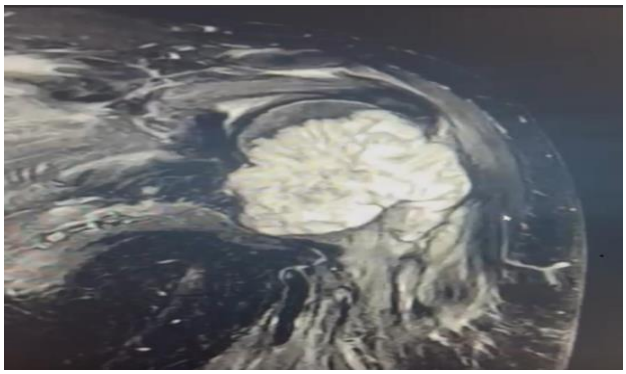

Figure 4: MRI of left humerus of patient B preoperatively showing grossly large aggressive bone tumor heterogeneously enhanced of the proximal humerus with infiltration of the shoulder joint capsule measuring 6.5×6.5×5.7 cm. There are edematous changes of rotator cuff muscles (supraspinatus, infraspinatus, subscapularis), and to a lesser extent deltoid and teres major.

Figure 5: Intraoperative image of patient B showing proximal humerus tumor with wide margins resection including soft tissues and plate previously applied plate and screws measuring around 14 cm.

In aims to restore shoulder functionality and in the context of intact deltoid and axillary nerve, arthroplasty using reverse total shoulder prosthesis followed. The latter started by excision of remnants of capsule and labrum, glenoid preparation and application of glenoid baseplate. After incremental reaming of the humerus, a cemented revision stem was inserted with 20-23° of retroversion. However, due to long humerus osteotomy and residual bone defect, bone augmentation was deemed necessary and 2 femoral head allografts were used and reamed sequentially, and circumferentially fitted around the humeral stem at bone-prosthesis interface and additionally fixed in situ using 2 K-wires in patient B (Figure 6). To achieve a stable modular prosthesis with adequate soft tissue tension at low dislocation risk, an eccentric glenosphere was inserted and a polyethylene retentive humeral tray was used. When reduction was performed, we noticed a stable construct with good range of movement and soft tissue tension. Then the deltoid and remaining parts of biceps, coracobrachialis and rotator cuff muscles were reattached onto prosthesis (Figure 7).

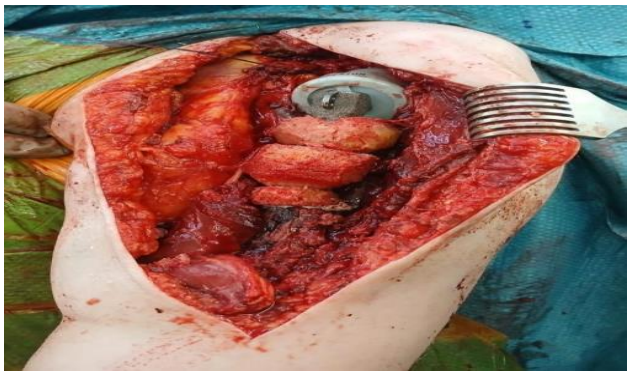

Figure 6: Intraoperative image following proximal humerus resection showing reverse total shoulder prosthesis augmented with allogenic bone blocks starting at osteotomy site and covering prosthetic stem.

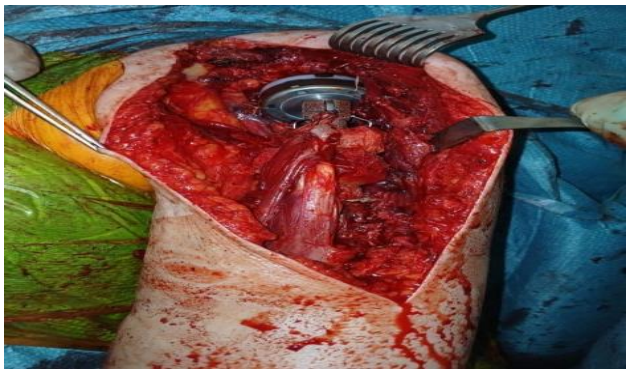

Figure 7: Intraoperative image showing fixation of bone blocks with 2 K-wires and attachment of deltoid, biceps, coracobrachialis and rotator cuff muscles onto prosthesis.

Immediate postoperative X-ray of the humerus showed well fitted and reduced cemented RTSA-allograft composite in patients A (Figure 8) and B (Figure 9) and both were placed in sling and swathe. Figure 10 shows the postoperative imaging of patient A at 10-months and 2-years interval and show further sclerosis of bone blocks with no clear evidence of bone resorption or hardware complication including mechanical failure, loosening or instability. 2 months postoperatively, patient A was doing pendulum exercises and had reached 80° passive abductions and 70° forward flexion, but lacked 30° of elbow full extension.

Figure 8: X-ray of right humerus of patient A immediately post resection of the proximal left humerus including the lytic lesion and replacement with cemented reverse total shoulder arthroplasty prosthesis with circumferential allograft bone blocks augmentation around the humeral stem.

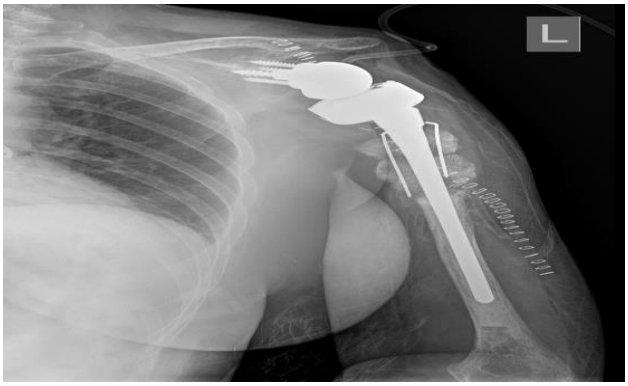

Figure 9: X-ray of left humerus of patient B immediately post resection of the proximal left humerus including the lytic lesion and replacement with cemented reverse total shoulder arthroplasty prosthesis with circumferential allograft bone blocks augmentation around the humeral stem and k-wire fixation of bone blocks.

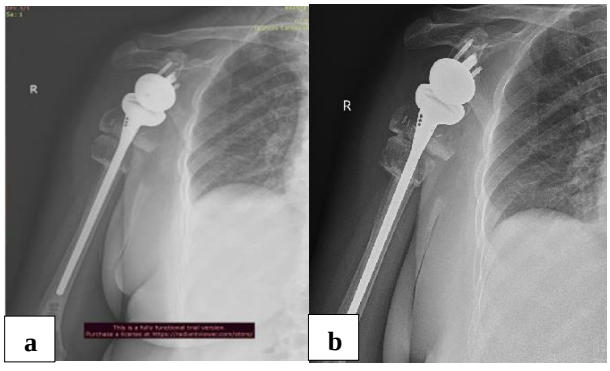

Figure 10 (a and b): X-ray of left humerus of patient A at 10 months (left) and at 2 years (right) post tumor resection and cemented RTSA and allograft bone blocks augmentation showing further sclerosis of bone blocks with no clear evidence of bone resorption or hardware complications.

At 4 months, pulley was started and at 6 months she had reached 90° abduction and forward flexion. At 18 months, patient had regained 70% of shoulder arc of motion and at last follow-up patient A continued to have acceptable shoulder range of motion, but slightly decreased compared to normal shoulder.

DISCUSSION

This study presents a review of the treatment options following shoulder sarcoma and associated outcomes and complications, and discloses two cases of proximal humerus sarcoma resection reconstructed with cemented reverse total shoulder arthroplasty and augmented with allograft bone blocks in a novel technique.

Previously, extremity sarcomas were treated routinely with amputation, but with the advent of imaging tools, chemoradiotherapy, and surgical techniques, most sarcoma patients are now candidates of limb salvage surgery.[2] Despite the large number of studies, the role of preoperative radiotherapy and chemotherapy on soft tissue sarcomas is still not conclusive.[4] Yet still, shoulder reconstruction with endoprosthesis is presumed to provide pain relief and durable implant survival, but the choice of implant for reconstruction is still controversial.[7]

Reverse total shoulder arthroplasty is a semi-constrained prothesis used in patients with rotator arthropathy and deficiency. The medial and distal pivot point allows for good shoulder function and range of motion driven by the deltoid.[5,8] As such in cases of rotator cuff deficit or resection with intact deltoid and axillary nerve, RTSA is a viable option; the inverted joint geometry leads to better recruitment of deltoid fibers and thus the reported preserved range of motion.[5,9] Several studies reported that RTSA is a stable construct that reproduces the shoulder after oncologic proximal humerus resection with good postoperative range of motion but slightly decreased compared to a normal shoulder.[10,11]

Rotator cuff insufficiency and bony defect are major problems secondary to wide margin resection, that results in limited shoulder range of motion and higher rate of instability and dislocation. Amouyel et al reported that large resections of 170 mm versus 100mm are associated with increased risk of prosthetic instability.[12] Hence, APC is a method of reconstruction that overcomes the latter complications by allowing reattachment of rotator cuff tendons and capsule reconstitution and compensates the bone stock. It was shown to preserve an acceptable shoulder range of motion at a similar rate of complications and low incidence of implant revision.[13,14] However, delayed complications include delayed union, osteolysis, and resorption, fracture and fragmentation.[13-15] Hemiarthroplasty is another method of reconstruction but the drawbacks include limited shoulder strength and range of motion and proximal migration as compared to other methods of reconstruction.[13,16]

Similar to any surgical intervention, RTSA is not without complications. According to Henderson classification, complications arise from either mechanical and non-mechanical causes. The former includes soft tissue failure, aseptic loosening and prosthetic failure/fracture, whereas the latter include periprosthetic infection and tumor progression.[16,17] Instability remains the most important complications that warrants caution. As such, in cases of shoulder instability post resection secondary to tumor invasion of surrounding soft tissues or the need to sacrifice stabilizing structures including deltoid muscle/axillary nerve and rotator cuff muscles, a constrained RTSA implant is recommended. A study by Ayvaz et al showed that a constrained design helps prevent instability but at the expense of decreased range of motion, and a higher risk of aseptic loosening at humeral component/bone interface and revision.[6] Therefore, the choice of constrained vs non-constrained implant depends on intraoperative stability, soft tissue extent and anatomical and functional needs.

In our patients, we performed an osteotomy of humerus (6 cm and 11 cm), had to sacrifice parts of rotator cuff, protected the axillary nerve and did not sacrifice a lot of the deltoid muscle. Given the patients' age and functional status and to preserve a functional shoulder, we decided to proceed with cemented RTSA using a revision stem. To overcome the risk of instability following RTSA, the construct was lateralized and we used an eccentric glenosphere with retentive humeral tray to increase soft tissue tension and decrease risk of sublaxation and dislocation. To address the bony defect, an allograft was necessary and with the available femoral head bone grafts, an allograft bone blocks were augmented in a novel way to restore bone stock and improve implant survival. Postoperatively, patients undergoing RTSA with bone grafting require immobilization for few weeks with sling and swathe followed by gradual strengthening of the deltoid to maintain stability.[5] It is noteworthy that bone healing and callus formation can be delayed by postoperative chemotherapy and cement use especially at the bone junction.[13]

A study by Teunis et al comparing various surgical options found that APC have similar functional outcomes and survival rates compared with prosthesis, and is superior to osteoarticular allografts.[7] Rampam et al noticed a better functional outcome in patients with RTSA-APC compared to those with HA but there was no clear superiority when using RTSA-APC compared to HA-APC in rates of complication and functional outcomes.[16] Antal et al, however, noticed the best range of motion was among RTSA-autograft composite followed by fibular reconstruction whereas osteoarticular allograft had the highest complications and revisions rate followed by RTSA then HA (40%, 25%, 15% respectively); there was no significant difference in quality of life among the groups.[5] In young patients with good life expectancy and small humerus intramedullary cavity, fibula autograft seems to be a good option. Poor prognosis patients might benefit the most from a HA due to acceptable results and low complications and revision rates with the disadvantage of poor shoulder range of motion.[5,18,19] Hao et al found no difference between APC and endoprosthesis in postoperative forward elevation, external rotation, functional scores.[14]

CONCLUSION

Reverse total shoulder arthroplasty with bone block augmentation is functionally stable and effective following massive tumor resection in appropriately selected patients. Controversy still exists on the ideal reconstruction method and more research is needed to identify the best method of reconstruction tailored per patient tumor properties, extent of resection, expected shoulder function and the associated long-term outcomes and complications associated with the prosthetic implant.

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