JSES Reviews, Reports, and Techniques 5 (2025) 339–343

Contents lists available at ScienceDirect

JSES Reviews, Reports, and Techniques

journal homepage: www.jsesreviewsreportstech.org

Periarticular humerus resection and reconstruction with an orthoplastic approach: a case report

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ARTICLE INFO

Keywords:
Humeral reconstruction
Free fibular flap
Limb salvage
Osteosarcoma
Case report
Orthoplastic

Osteosarcoma (OS) is a rare malignancy found in younger patients, with an incidence of approximately 5 cases per million children.[1] Around 10% of OS cases occur in the humerus, with 90% of such cases occurring in the proximal humerus.[6] Reconstruction following humeral OS resection presents a unique challenge. Many strategies have been implemented utilizing megaprostheses, allograft-prosthetic composites, osteoarticular allograft, and vascularized autograft.[2-3,8] Autograft tissue, such as a vascularized fibular flap, presents an option for proximal humerus OS reconstruction in the pediatric population.[4] We discussed the case of a female aged 8 years who presented to a tertiary sarcoma referral center with a right humerus OS requiring complex periarticular upper extremity reconstruction. Through careful collaboration between the orthopedic surgery, plastic and reconstructive surgery, and pediatric hand surgery services, a single-stage operation was performed to achieve humeral reconstruction with a glenoid-articulating free fibular flap and radial nerve reconstruction with musculocutaneous nerve autograft. At 6 years following surgery, the patient remains disease-free and has regained good function of her right upper extremity, modest shoulder forward flexion, and radial nerve function.

Case report

The patient is an 8 years old right hand dominant healthy female who presented to our institution's pediatric emergency department after feeling "a pop" in her right arm while climbing a rope during gym class. Prior to her injury, she reported intermittent right arm pain for two weeks prior. On initial examination, the patient had right upper arm swelling and a palpable mass in the triceps region. She also had full range of motion (ROM), strength, and sensation of her right upper extremity. Radiographs were obtained which revealed an aggressive lesion of the right proximal humerus with periosteal reaction (Fig. 1).

The patient was referred to our institution's orthopedic oncology clinic, where subsequent workup included right upper extremity magnetic resonance imaging and chest computed tomography (CT) scan. The magnetic resonance imaging demonstrated abnormal bone marrow involvement spanning ~85% of the humerus to ~1 cm proximal to the trochlea, as well as significant soft tissue infiltration of the posterior arm compartment (Fig. 2). The chest CT demonstrated multiple pulmonary nodules concerning for bilateral lung metastases. A CT-guided needle biopsy of the humerus lesion revealed high-grade OS. The patient subsequently began a course of neoadjuvant chemotherapy comprised of two cycles of cisplatin, doxorubicin, and high-dose methotrexate.

Treatment goals and management

Surgical treatment goals to address the patient's humerus OS included the following: (1) wide resection of humeral OS with

No ethics approval was required for the conduct of this study.

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https://doi.org/10.1016/j.xrrt.2024.10.004

Figure 1 Initial right humerus radiographs obtained during patient's emergency department encounter demonstrating an aggressive lesion involving the proximal to middle humeral shaft.

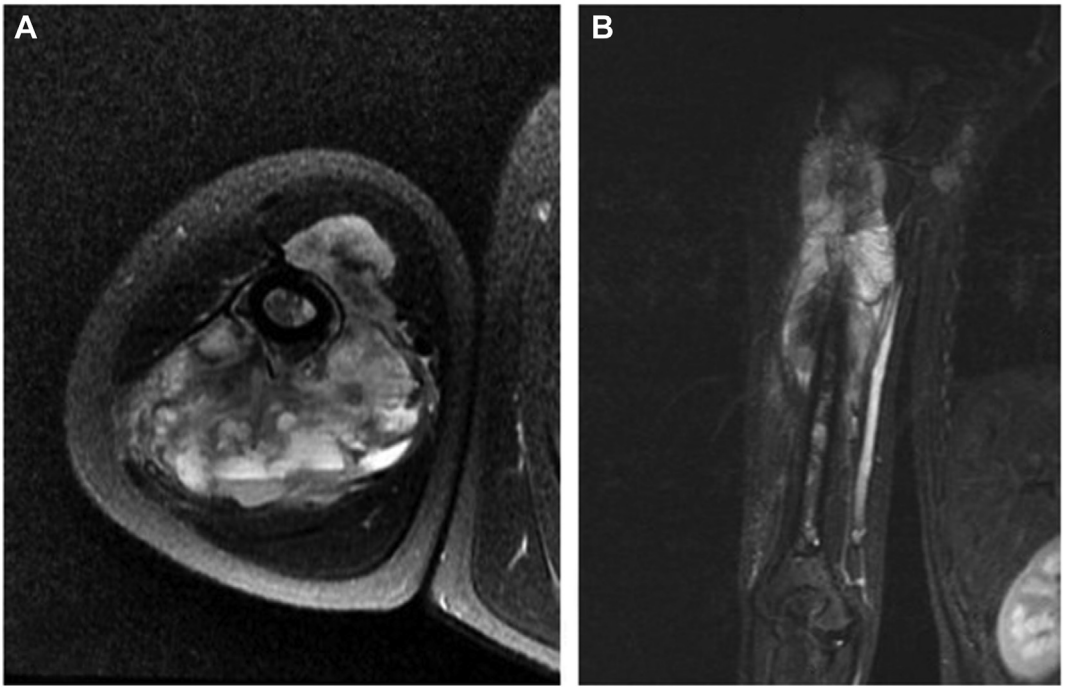

Figure 2 T2 weighted fast spin echo (FSE) MRI scan of the right upper extremity demonstrating extent of OS including soft tissue infiltration of the posterior arm compartment. (**A**) Axial image; (**B**) Coronal image; *MRI*, magnetic resonance imaging; *OS*, osteosarcoma.

negative surgical margins, and (2) limb salvage with preservation of a functional right upper extremity. Four surgical options were proposed, including (1) complete humerus resection with forearm suspension to the chest wall, (2) complete humerus resection with megaprosthetic reconstruction, (3) wide humeral resection with vascularized free fibular autograft reconstruction, or (4) wide humerus resection with allograft reconstruction. Forearm suspension was considered a lesser functional option, while concerns were raised with common complications observed in megaprosthetic reconstruction such as infection and failure by other mechanisms. After thorough discussion, the decision was made to proceed with option 3, presuming that negative margins could be obtained. It is hard to plan preoperatively our planned zone of resection to ensure negative margins. However, if negative margins are difficult to obtain intraoperatively, it is paramount we address the disease of the bone and remove more humerus until successful negative margins. If there is still doubt on the margins, our reconstructive options may change and a megaprostheses reconstruction would likely be more favorable given the caution to not perpetuate any disease progression with an autologous reconstruction option. In addition, given the infiltration of disease into the posterior arm compartment near the radial nerve, the possibilities of radial nerve resection and reconstruction were discussed. In preparation for surgery, the reconstructive plastic surgery service was contacted

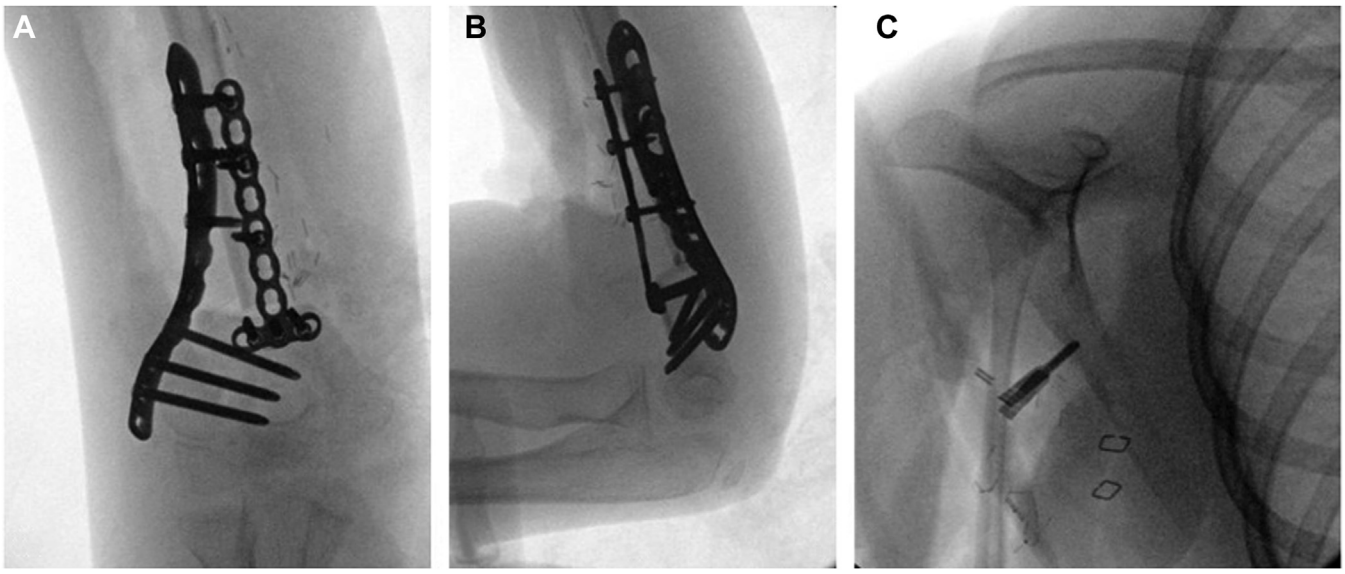

Figure 3 Intraoperative fluoroscopic images depicting surgical fixation technique. (**A**) Anterior-posterior view of preliminary fixation of distal humeral fragment to fibular autograft. (**B**) Lateral view of preliminary fixation of distal humeral fragment to fibular autograft. (**C**) Anterior-posterior view of shoulder demonstrating reconstructed glenofibular articulation.

regarding the vascularized free fibular graft humeral reconstruction, and the pediatric hand service was contacted regarding the anticipated radial nerve resection and reconstruction.

Wide resection was performed by the orthopedic oncology service in the supine position, utilizing an extended deltopectoral approach ending past the elbow flexion crease. Bony resection of the distal humerus was performed using an oscillating saw, with negative margins confirmed on intraoperative frozen section. Proximally, a 7 cm span of radial nerve was involved within the tumor, which was consistent with preoperative imaging. The radial nerve was divided proximally and distally and tagged for subsequent reconstruction. The rotator cuff tendinous insertions were also divided from the proximal humerus to aid resection. The proximal extent of the tumor resection was completed with an extensive posterior musculature dissection, and frozen section pathology margins were negative for tumor involvement.

While the oncologic resection took place, the reconstructive plastic surgery team simultaneously harvested a pedicled fibula free flap from the patient's contralateral left lower extremity. The residual lateral collateral ligament was inserted into the incisure fibularis of the tibia with suture anchors. After harvest was complete, the vascularized fibula graft was placed in the defect of the resected humerus. A 2.4 mm locking T-plate was utilized to fix the fibula graft to the distal humerus fragment. An additional 2.7/3.5 mm locking distal lateral plate was applied to the fibula graft and the lateral distal humerus (Fig. 3, *A* and *B*). The fibular head was then reduced such that the fibular articular cartilage and glenoid were articulating as a neo-glenofibular joint (Fig. 3, *C*). The rotator cuff tendons were then fixed to the fibular head using suture anchors medially and laterally. #5 Ti-Cron suture (Medtronic, Dublin, Ireland) was used to further secure the anchored tendons. After appropriate alignment was confirmed on fluoroscopy, the fibula graft was secured with additional screws through the previously placed distal plates. Anastomosis of the pedicled free fibula to the circumflex humeral vessels was then performed by the plastic surgery service.

After fixation was complete, the pediatric hand team harvested a 10 cm segment of the musculocutaneous nerve to utilize as nerve autograft for radial nerve reconstruction. The harvested nerve was divided into two separate 5 cm cable grafts to span the radial nerve defect. The cable nerve grafts were inset under operative microscopy with interrupted 8-0 nylon sutures. AxoGuard nerve wrap (Axogen, Alachua, FL, USA) was then secured over the proximal and distal nerve anastomoses with 8-0 nylon sutures. After tension-free nerve repair, a layered closure was performed, followed by application of a right upper extremity posterior slab splint, and left lower extremity knee immobilizer brace prior to leaving the operating room.

Outcome

Postoperatively, the patient recovered well without any acute flap complications and was discharged on postoperative day 12. Immediately postoperatively, her donor site lower extremity was placed in a knee immobilizer and a Pressure Relieving Ankle Foot Orthosis boot. The inpatient team did not note a foot drop present at the time. She underwent planned adjuvant chemotherapy and bilateral thoracotomies to treat her metastatic lung disease. In clinic, she was noted to have a left foot drop at her 2-week visit which resolved by 6 months. The foot drop was attributed to peroneal nerve manipulation during fibular graft harvest. At 1.5-year follow-up, the patient had regained sensorimotor function in the radial nerve distribution and was able to make a composite grasp with full elbow ROM. Due to limited shoulder ROM, targeted physical therapy was initiated at that time. There were no residual functional deficits of the flap donor site at the contralateral lower extremity. Patient was able to fully plantar and dorsiflex her ankle and had full sensation in the superficial and deep peroneal nerve distributions. By 3-year follow-up, the patient was cleared to participate in gym class with strict contact precautions and had regained sufficient radial nerve function to play the piano and clarinet. At most recent 7-year follow-up, she had full extension of her fingers and her right thumb, full extension of the elbow, and elbow flexion to 100 degrees (Fig. 4). Healing of the bone anastomosis is evident and there is no evidence of failure of the graft or fixation (Fig. 5). She has also participated in her school's marching band and has no residual deficits in the lower extremity. She

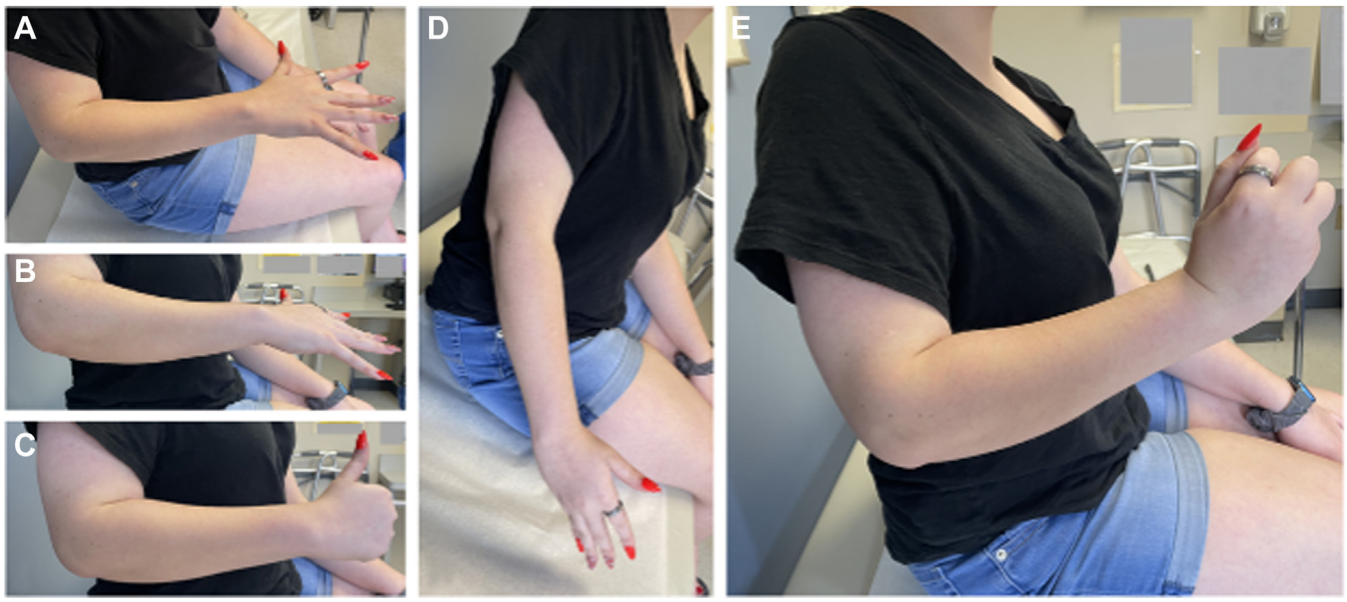

Figure 4 Photos taken in clinic at 7-years postop showing (**A** and **B**) full finger extension; (**C**) thumb extension; (**D**) elbow extension; and (**E**) elbow flexion.

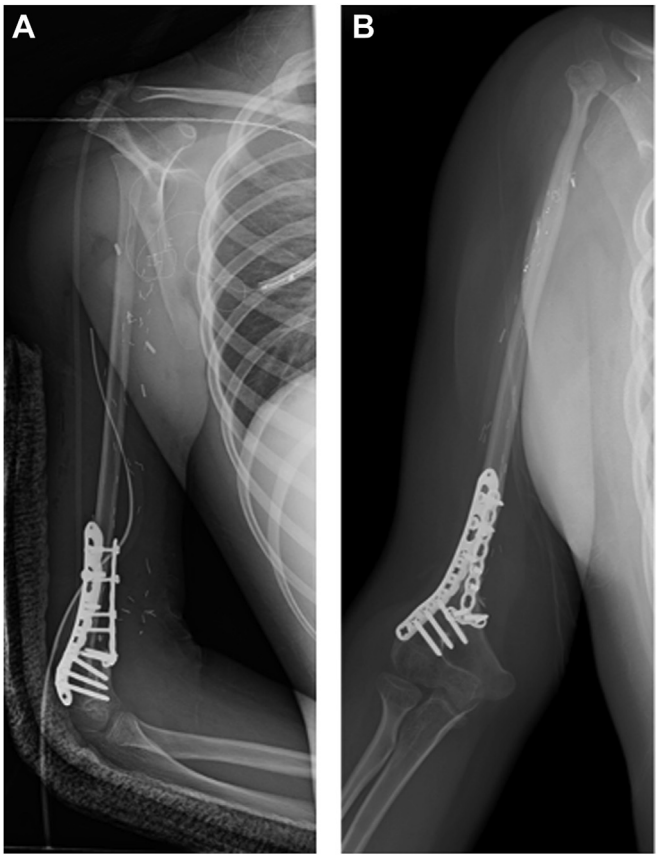

Figure 5 (**A**) Immediate postoperative radiograph depicting distal fixation of free fibular autograft and proximal glenofibular articulation. (**B**) 7-year follow-up clinical radiograph demonstrating stable hardware.

shows no signs of knee instability with her activities of daily life, no signs ankle instabilities, and is able to live her life as a teenager such as partaking in summer band camp.

Discussion

Multidisciplinary involvement of plastic surgery and orthopedic surgery services enabled a biological construct with reasonably restored shoulder function, maintained capacity for physical growth, and preserved/recovered upper extremity neurological function. Perceived strengths of our surgical technique include achieving sufficient stability by using two distal humeral plates for fixation of our vascularized fibular graft, as well as achieving good fixation of the rotator cuff tendons to the transferred fibular head. A notable complication of this procedure was the transient peroneal nerve palsy due to presumed manipulation during fibular graft harvest. Given that graft fractures are common following this procedure, additional allograft tissue could have been utilized as described in the modified Capanna's technique to strengthen our construct.[5] However, our patient recovered well without graft failure.

Currently, two of the most common techniques utilized for upper extremity reconstruction are vascularized fibular free flap reconstruction and endoprosthetic reconstruction. Each of these techniques restores some degree of function; however, each approach confers different complication risks including delayed union, nonunion, hardware failure, and periprosthetic fracture. Vascularized fibular free flap humeral reconstructions have a notably high risk of graft fracture estimated between 27-64%.[7,9] To address this high fracture risk, newer techniques which supplement autograft with allograft tissue have been described with satisfactory results.[5] Vascularized fibular free flap reconstruction is emerging as a viable option in proposed algorithms for treating proximal humeral pediatric tumors with intra-articular involvement.[4] Additional parameters to produce durable, long-term constructs should be investigated, as susceptibility to nonunion or graft fracture may differ based on the length of fibula transferred or the type of fixation used for graft placement. Understanding these more nuanced factors may notably improve outcomes for this procedure. Nevertheless, this collaborative approach enabled a successful oncologic outcome with acceptable function in this patient.

Conclusion

This case highlights the successful multidisciplinary approach involving orthopedic oncology, plastic surgery, and pediatric hand surgery for the treatment of high-grade OS of the humerus in a pediatric patient. By utilizing autologous vascularized fibular free flap reconstruction combined with meticulous fixation techniques, we were able to achieve limb salvage, maintain functionality, and restore nerve continuity with minimal long-term complications in a young patient. Despite the transient postoperative foot drop, the patient demonstrated excellent recovery, regaining significant sensorimotor function and returning to full participation in physical activities. This case underscores the importance of individualized surgical planning and interdisciplinary collaboration to optimize functional outcomes while addressing the oncologic needs in complex pediatric cases. Long-term follow-up revealed no graft or fixation failure, and no residual donor site morbidities, further supporting the durability of our surgical approach.

Disclaimers:

Funding: The authors have no financial disclosures. No funding was received for this article.
Conflicts of interest: The authors, their immediate families, and any research foundation with which they are affiliated have not received any financial payments or other benefits from any commercial entity related to the subject of this article.

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