JSES Reviews, Reports, and Techniques 4 (2024) 499–503

Contents lists available at ScienceDirect

JSES Reviews, Reports, and Techniques

journal homepage: www.jsesreviewsreportstech.org

Acute compartment syndrome following allograft-prosthetic composite reverse shoulder arthroplasty for osteosarcoma of the proximal humerus: a case report

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ARTICLE INFO

Keywords:
Compartment syndrome
Reverse shoulder arthroplasty
Osteosarcoma
Radial nerve palsy
Allograft-prosthetic composite
Pediatric

Acute compartment syndrome (ACS) is a surgical emergency. It occurs when there is an increase in pressure within a closed fascial compartment leading to decreased circulation and eventual ischemia and irreversible necrosis.[4,15] Although it is considered a clinical diagnosis, measurements of intracompartmental pressures > 30mmHg is used as a threshold to aid in diagnosis.[4,11] Without prompt diagnosis and surgical intervention, ACS can result in severe morbidity or even mortality.[11]

Although ACS more commonly affects the lower extremity, ACS of the upper extremity can result in profound disability for patients.[8] Fractures are the most common causes of ACS of the upper extremity, while use of anticoagulation therapy, hematoma formation, pectoralis muscle tendon rupture, biceps tendon rupture, and prolonged lateral decubitus position have been reported as etiologies as well.[3,10,12-14] Tumors are rare causes of compartment syndrome.[2,16] Because ACS of the upper extremity is an orthopedic surgical emergency, it is critical for accurate clinical identification to prevent extensive damage to tendons, muscles, and nerves.[10]

Though extremely rare, postoperative hematoma after reverse shoulder arthroplasty (RSA) can lead to compartment syndrome within 24 hours.[10] Another risk factor for ACS is length of procedure, with a longer operative time increasing the risk for developing ACS.[17] Timely recognition and surgical intervention for compartment syndrome is critical, but diagnosis can be delayed due to lack of familiarity with uncommon presentations of compartment syndrome such as in the upper arm.

In this manuscript, we describe a novel clinical situation of ACS in a 16 year-old patient four days after allograft-prosthetic composite (APC) RSA for proximal humerus osteosarcoma for the purpose of educating providers on this rare presentation of ACS of the upper extremity.

Clinical history

An otherwise healthy 16-year-old right-hand dominant male was initially evaluated at an outside institution for indolent onset of right shoulder and arm pain. Imaging was obtained and was concerning for a pathologic lesion (Fig. 1), and so he was referred to our institution. Advanced imaging and a biopsy were consistent with primary osteosarcoma of the proximal humerus (Fig. 2).

On physical examination, he had no overlying skin changes aside from mild swelling at the biopsy site without signs of erythema. His sensation and motor function were both intact. His strength and range of motion (ROM) were full but painful compared to his left shoulder.

He was indicated for and subsequently underwent initial neoadjuvant chemotherapy and followed by wide surgical resection. Due to the large size of the necessary proximal humerus resection, which was to be distal to his deltoid tuberosity, APC-RSA was planned for his reconstruction. An allograft with soft tissue attachments was used to optimize postoperative function.

Institutional review board approval was not required for this case report.

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https://doi.org/10.1016/j.xrrt.2024.03.009

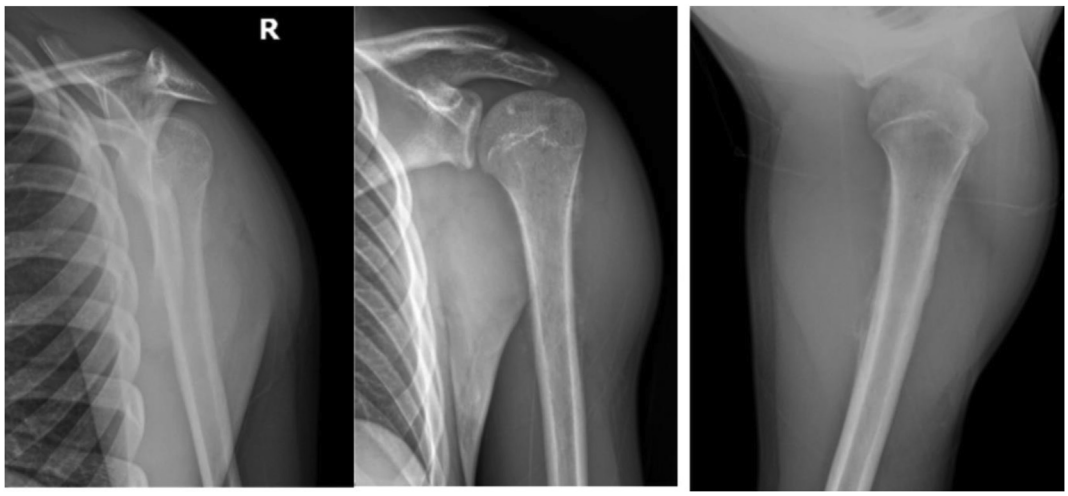

Figure 1 Initial three-view radiographs.

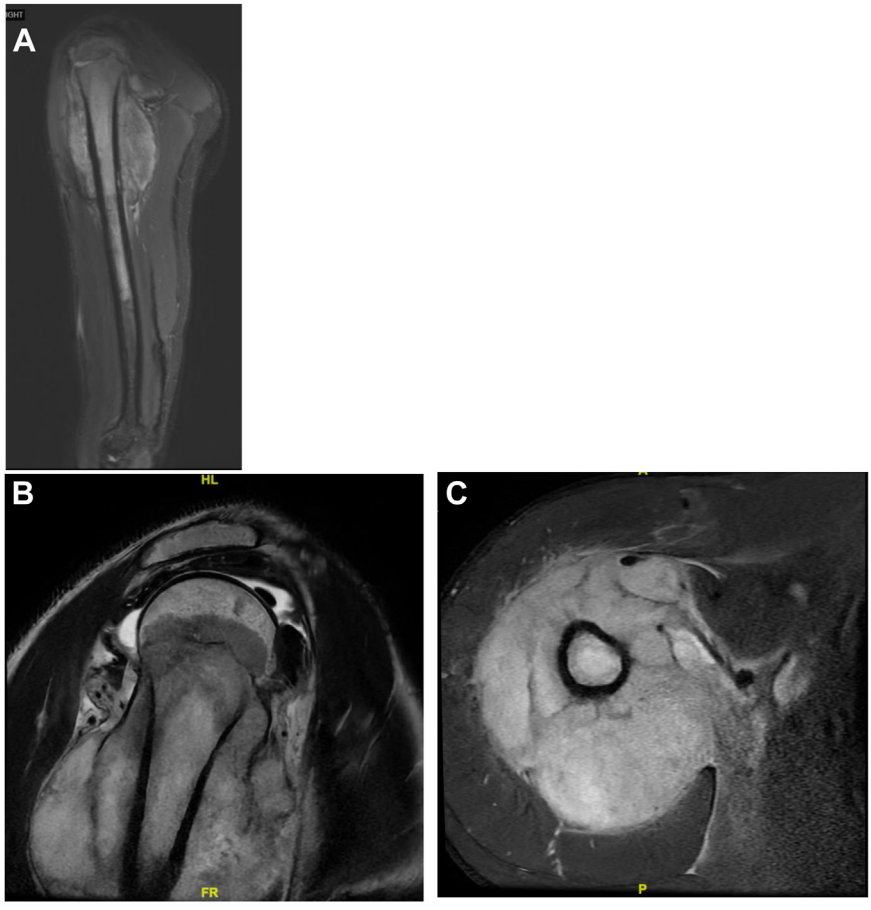

Figure 2 MRI of the right Humerus and Shoulder. (**A**) Coronal T2 weighted MRI of the R humerus demonstrating. (**B**) Sagittal T2 weighted MRI of the R proximal humerus again demonstrating extensive soft tissue involvement. (**C**) Axial T2 weighted MRI of the R proximal humeral shaft demonstrating soft involvement around the shaft of the humerus. *MRI*, magnetic resonance imaging.

Index surgical procedure

The patient was positioned in beach chair position. The patient was then prepped and draped in usual sterile fashion and antibiotics were administered as per protocol.

First the wide tumor resection was performed. A deltopectoral approach was utilized, and the biopsy site was excised. Initial dissection and exposure was focused on preserving musculotendinous attachments for eventual transfer to the allograft. The long head of the biceps, the rotator cuff tendons, the deltoid insertion, and the latissimus dorsi and teres major tendons were all tenotomized and tagged for this purpose.

The tumor was noted to be intimate with the axillary and radial nerves and the brachial vessels. These were carefully preserved,

Figure 3 Immediate postoperative radiographs showing the reverse total shoulder arthroplasty and its allograft components.

and then the tumor was removed in its entirety (148cm^3). Approximately 10 cm of native distal humerus remained after the resection. Intraoperative margins, including intramedullary tissue, were confirmed to be negative of any gross osteosarcoma cells, and therefore the decision was made to proceed with APC-RSA.

To perform the APC-RSA (Zimmer Biomet, Warsaw, IN, USA), the glenoid was prepared and reconstructed, followed by preparation of the allograft on the back table, followed by reconstruction of the humerus with the allograft and then implantation and reduction of the final components. For the glenoid reconstruction, as there was no deformity, a nonaugmented mini baseplate with a central compression screw and peripheral locking screws was used. A large, 40 mm glenosphere was impacted onto the cleaned morse taper of the baseplate; this size glenosphere was used to maximize implant stability and impingement free ROM.

For the humeral reconstruction, as previously mentioned, a humeral allograft with soft tissue attachments was utilized for the purpose of reattaching the native previously tenotomized tendons to maximize his postoperative function. This allograft was thawed and then prepared on the back table, with its length based on preoperative templating. Once prepared, it was potted with a trial stem in place onto the prepared distal humerus at 30 degrees retroversion. The allograft with the trial stem was then compressed to the native distal humerus with a 4.5 narrow locking compression plate on the anteromedial aspect of the construct as well as a 3.5 locking compression plate on the posterolateral aspect (DePuy Synthes, Raynham, MA, USA). After each screw was placed, the trial stem was partially removed to confirm lack of incarceration by the screw. Next, the trial stem was removed, cement inserted and pressurized, and the final humeral stem inserted. There was no cement leakage at the APC interface with pressurization, confirming excellent fit of the allograft onto the native humerus.

Once the cement had hardened, multiple humeral tray and liner combinations were trialed until a final construct with optimal stability was chosen, impacted, and reduced. Finally, the patient's tenotomized tendons were reattached to the humeral allograft including the deltoid, pectoralis, latissimus and teres major, and the rotator cuff tendons.

Of note, there was no active bleeding at the conclusion of the surgery. Careful hemostasis was achieved. Two deep drains were placed due to risk of hematoma formation. No transexamic acid was used in this case. The soft tissues were closed in standard fashion. Two view postoperative x-rays confirmed appropriate alignment and positioning of the components (Fig. 3).

Postoperative course #1

The patient was admitted for standard postoperative management and monitoring. A preoperatively-placed but postoperatively-dosed supraclavicular peripheral nerve catheter was started in the recovery room after standard postoperative neurovascular check, which was unremarkable. The highest drain output, 280 cc, occurred postoperative day (POD) two. Drain output decreased daily with the final output being 25cc serosanguinous fluid on the day of discharge. The patient was discharged home with the drains in place on postoperative POD four, with the plan to remove this in clinic at one week postoperatively. The peripheral nerve catheter was pulled at home on POD three by the patient's parents, per standard protocol.

Acute compartment syndrome

On POD four in the early afternoon, the patient called the on call orthopedic surgery resident describing a "pop" in the shoulder during a bowel movement followed by acute worsening of pain and swelling, dressing saturation, and increased drain output over the next 30 minutes. He was instructed to present to our emergency department (ED) for evaluation.

Physical examination in the ED demonstrated a saturated dressing, active serosanguinous discharge from the incision site, and numbness and weakness with a noted inability to extend his right index finger. The numbness was localized in the radial distribution, with mild decrease in the median nerve distribution as well. Sensation was intact in the axillary, lateral antebrachial, and ulnar distributions. It was also noted that the patient had a swollen, tense right arm. Stat radiographs obtained in the ED showed no signs of dislocation or hardware failure in the APC-RSA (Fig. 4).

Given the evolving ACS with the new radial nerve palsy, the patient was brought immediately to the operating room for surgical fasciotomies, hematoma evacuation, and exploration. Intracompartmental pressures were not obtained due to the clarity of the diagnosis and time sensitivity of surgical intervention. After induction of general anesthesia, the patient was again positioned in the beach chair position and standard preoperative antiseptic preparation, draping, and time-out was again performed. His previous incision was opened as was the deep fascia. There was a large hematoma which was removed from the deep compartment of the arm. The APC-RSA was intact without complication. Despite the hematoma, there were no large bleeding vessels present. All minor bleeding sites were coagulated, a new drain was placed, and the soft tissues were closed once again.

Postoperative course #2

On POD one, the patient was able to extend his right index finger again, and sensation in the median nerve distribution returned to

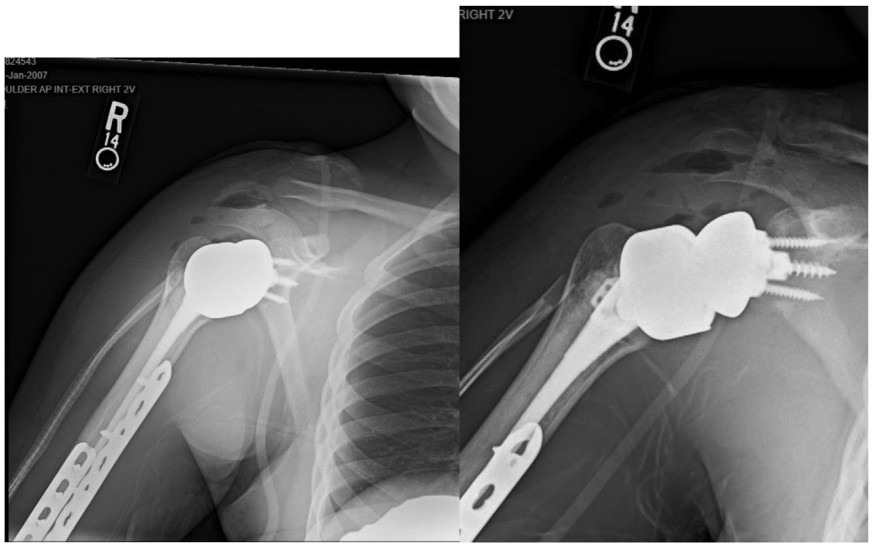

Figure 4 POD 4/5 Anteriorposterior and lateral views of the R shoulder obtained in the ED after a "pop" was heard while rolling in bed. *POD*, postoperative day; *ED*, emergency department.

normal. There continued to be mild numbness in the radial nerve distribution. On POD two, the patient reported a return to normal, full sensation in his right hand including the radial distribution, and he was discharged POD three with the drain in place. At his first follow-up appointment (POD 7), he reported minimal pain and normal sensation. The drain was removed. At his second postoperative appointment (POD 14) he continued to report little to no pain as well as intact sensation. At his third follow-up appointment (POD 21), the staples were removed, and the patient was cleared to restart chemotherapy. The patient then presented for 28-week postoperative follow-up. He had been working with physical therapy and ROM was 90° active forward flexion and 90 ° active abduction. Passive ROM check showed 130 ° forward flexion, and 130 ° abduction (Fig. 5, *A*). Active external rotation with the shoulder adducted was approximately 40 ° (Fig. 5, *B*). Figure 6 shows postoperative radiographs at that visit, which demonstrated a well healed APC-RSA without evidence of complication.

Figure 5 (**A**) Most recent postoperative visit demonstrating range of motion – forward flexion, external rotation. (**B**) Most recent postoperative visit demonstrating shoulder abduction.

Discussion

In this article, we report the case of a 16 year old patient who developed upper extremity ACS secondary to hematoma formation four days after APC-RSA for proximal humerus osteosarcoma for the purpose of educating shoulder surgeons on this rare presentation of upper extremity ACS. Urgent surgical intervention, within 2 hours of the initiation of symptoms, fortunately resulted in an excellent clinical outcome. At the most recent, four-month follow-up, our patient had minimal pain, normal sensation, full motor function, and was making progress with therapy in terms of shoulder ROM. We believe that any delay in his diagnosis or surgical intervention could have severely compromised his outcome and long-term prognosis.

Surgical intervention, along with chemotherapy, has been the mainstay treatment of osteosarcoma.[12] However, failure of suture suspension, megaprosthesis dislocation, wound dehiscence and neurologic complications can develop postoperatively.[1,18] Compartment syndrome is typically associated with traumatic injury but can be seen in cases of nontraumatic surgical intervention with large amounts of soft tissue damage and intraoperative blood loss.[7,9] Vaynrub et al reports that a risk factor for nontraumatic compartment syndrome in cancer patients is length of operation.[17] Resultant reperfusion can lead to acute inflammation and systemic responses from inflammatory cytokines and reactive oxygen species.[5]

In Meshram et al's case series, there were two cases of revision reverse total shoulder arthroplasty (rTSA) that led to the development of ACS, one with rheumatoid arthritis and the other osteoarthritis. The first patient developed ACS POD one secondary to a large hematoma found on emergency computed tomography scan. The second patient developed multiple pulmonary emboli POD one after his revision rTSA and was started on low-molecular-weight heparin

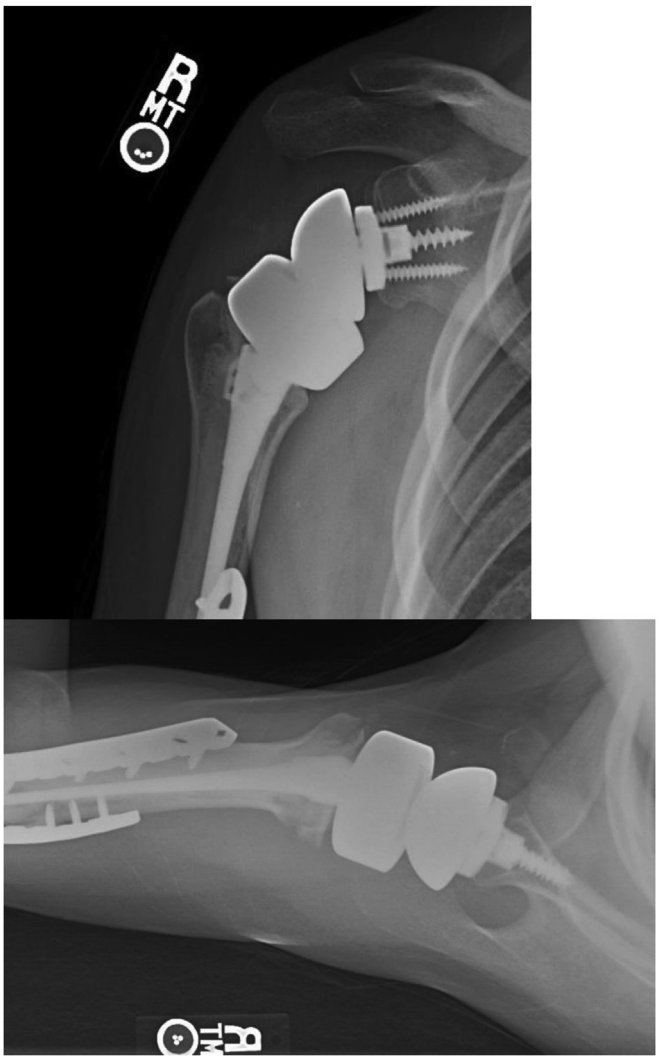

Figure 6 Postoperative radiographs at 28 weeks. There were no issues with hardware seen.

and bridge to warfarin on POD 10 and subsequently discharged home. On POD 14, the patient returned to the ED and due to concern for ACS underwent compartment releases from the anterior shoulder into the volar aspect of the forearm for a large hematoma. Our patient was not placed on any anticoagulation. However, he did present similarly to the second patient above, after discharge home. Providers should be aware of compartment syndrome symptoms after patient's discharge home, with a high index of suspicion.

Hendy et al analyzed 6421 shoulder arthroplasties and documented that rTSA were more likely to develop postoperative hematoma, and they were more likely to develop within 21 days postoperatively.[6] The majority was managed nonoperatively; however, about 7% of them were associated with periprosthetic joint infections.

Conclusion

Compartment syndrome is a potentially devastating operative complication that should be acted on immediately to prevent permanent damage to surrounding neurovascular structures. Good outcomes can be achieved with timely intervention and interdisciplinary follow-up care. This case describes an incidence of compartment syndrome following a reverse total shoulder arthroplasty and allograft prosthetic composite for treatment of osteosarcoma.

Disclaimers:

Funding: No funding was disclosed by the authors.

Conflicts of interest: Nicholas Bernthal, MD is a paid consultant for Biomet, Daiichi Sankyo, Deciphera, Onkos. He has received research support from Trellis, Zimmer, and the National Institutes of Health. He is also a board or committee member of the Musculoskeletal Tumor Society and the Orthopaedic Research and Education Foundation.

Andrew R Jensen is a paid consultant for FH Ortho and Zimmer Biomet. He is also a board or committee member of American Academy of Orthopaedic Surgeons.

The other 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.

Patient consent: Obtained.

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