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Reverse total shoulder arthroplasty in a patient with osteogenesis imperfecta type I complicated by a proximal humeral enchondroma: a case report and review of the literature

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Case report

A 60-year-old right hand–dominant woman employed as a nurse was diagnosed with osteogenesis imperfecta type I as a child after sustaining multiple fragility fractures including 3 to her proximal right humerus that had resulted in a residual varus angulated deformity. The patient did not require operative intervention for these fractures. Approximately 4 years ago, the patient had the insidious onset of right shoulder pain and decreased range of motion. Subsequent magnetic resonance imaging (MRI) showed a large, full-thickness rotator cuff tear. Six months later, the patient underwent arthroscopic repair with biceps tenotomy at an outside facility. Unfortunately, this procedure was complicated by postoperative stiffness that was recalcitrant to conservative measures. She ultimately required a capsular resection and manipulation under anesthesia 18 months after the index surgery. However, because of a continued lack of symptomatic improvement, the patient obtained a repeated MRI study, which demonstrated a retear in the rotator cuff. Of note, it was during the initial workup that a 5-cm enchondroma was discovered in the proximal right humerus.

One year after being diagnosed with a second rotator cuff tear, the patient presented to our clinic with complaints of right shoulder pain and significantly decreased range of motion. On examination, the patient demonstrated active elevation to 30° with frank escape, external rotation to neutral, and internal rotation to the abdomen. Passively, her elevation improved to 70°. She noted pain at the end of all ranges of motion with diffuse weakness, most markedly with internal and external rotation. Radiography demonstrated a superior-riding humeral head with a suspected proximal humeral head enchondroma (Fig. 1). Computed tomography confirmed an articulation between the acromion and humeral head (Fig. 2). These images were interpreted by a musculoskeletal radiologist to be most consistent with an enchondroma with potential aggressive characteristics. However, it was noted in consultation that the lesion did not appear to involve the endosteum, no periosteal reaction was appreciated, and no lysis was present within the mineralized segment of the tumor. Thus, radiographically, the lesion met classic criteria for a benign process. A decision was thus made to proceed operatively for a reverse total shoulder arthroplasty but a resection arthroplasty if the implant was unable to be properly seated. Appropriate consent was obtained. In addition, the patient was seen preoperatively in consultation by an orthopedic oncologist for intraoperative assistance in excision of the enchondroma.

Under general anesthesia, the patient was placed in the beach chair position. A deltopectoral approach was performed. Adequate visualization was able to be obtained despite intensive scarring due to earlier operations. After the humeral head osteotomy was performed, an intramedullary trocar pointed reamer was unable to be passed distally through the lesion because of its impermeability. The solid nature of this lesion was unanticipated, given the benign-appearing radiograph, and as a result, immediate intraoperative frozen section pathologic examination was obtained after curettage was performed (Fig. 3). The specimens were interpreted as benign, reinforcing the diagnosis of a benign enchondroma, and the instrumentation portion of the case subsequently proceeded

This case report did not require review through the Mayo Clinic Institutional Review Board.

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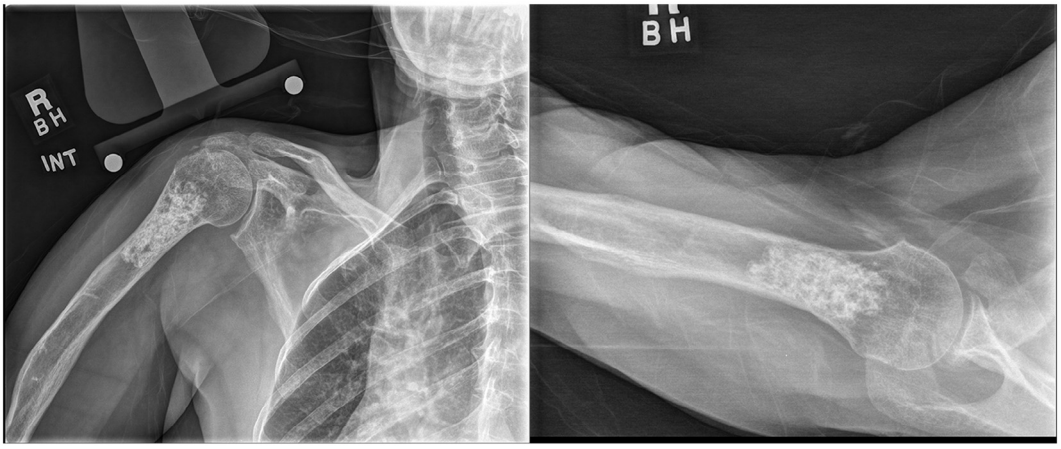

Figure 1 Anteroposterior and lateral radiographs demonstrating a superior-riding humeral head with a proximal humerus enchondroma.

without hesitation. As a result of curettage, the majority of the mass was successfully removed, but a small portion of residual lesion was left circumferentially in abutment of the inner aspect of the humeral cortex to prevent an iatrogenic fracture. Fluoroscopy was able to confirm that approximately 95% of the lesion was removed (Fig. 4). Subsequently, the glenoid was carefully prepared (Fig. 5), and excellent fixation was obtained with the baseplate using one central 6.5×25-mm screw, one 4.75×20-mm peripheral screw, and three 4.75×15-mm peripheral screws. After trial components demonstrated excellent stability and good range of motion, a permanent 6-mm micro stem was implanted using antibiotic-impregnated polymethyl methacrylate. After placement of the permanent humeral tray, bearing surface, and glenosphere, appropriate soft tissue closure was obtained.

The patient had an uneventful postoperative course and was discharged to home with a shoulder sling and passive range of motion protocol. She was seen at 6 weeks postoperatively, when she was advanced to pulley and wand exercises focused on external rotation and elevation, with progression to gentle isometric strengthening at 8 weeks postoperatively.

At 5 months postoperatively, the patient had minimal pain and her active forward elevation was >90°, with radiographs demonstrating well-aligned implants (Fig. 6).

Discussion

Osteogenesis imperfecta is a qualitative collagen disorder of heterogeneic origin that has numerous orthopedic manifestations, most notably pediatric fragility fractures.[22] Whereas extensive literature has evaluated the pediatric manifestations of this disease and a multitude of varying treatment modalities,[7,16,24] there has been a deficiency in the literature regarding the adult orthopedic manifestations of this disease.

The limited literature that does address the adult orthopedic manifestations and treatment options of osteogenesis imperfecta has typically focused on both techniques and outcomes used in hip and knee arthroplasty. However, many of the outcomes and conclusions surrounding lower extremity arthroplasty discuss techniques and pitfalls unique to these patients, and as such, these conclusions can likely be extrapolated to shoulder arthroplasty in this population. Anecdotal evidence suggests that total knee arthroplasty is an acceptable treatment for end-stage osteoarthritis in a properly selected patient with osteogenesis imperfecta.[14] Roberts et al recently reviewed the literature with regard to lower extremity arthroplasty outcomes in patients with osteogenesis imperfecta and found that the quality of subchondral bone found in these patients led to poor outcomes if it is used with cementless fixation,

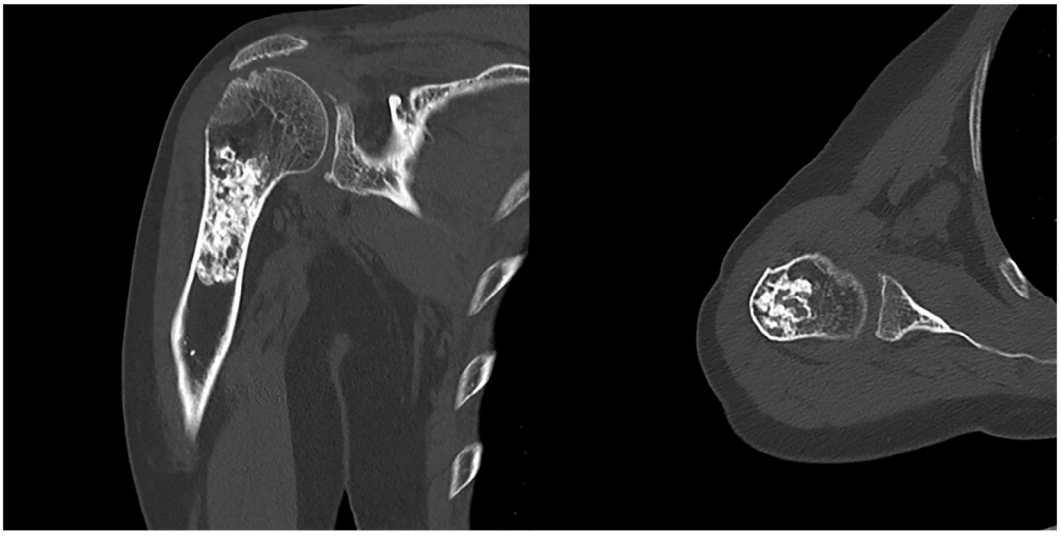

Figure 2 Coronal and axial computed tomography scans demonstrating articulation between acromion and humeral head as well as proximal humerus enchondroma.

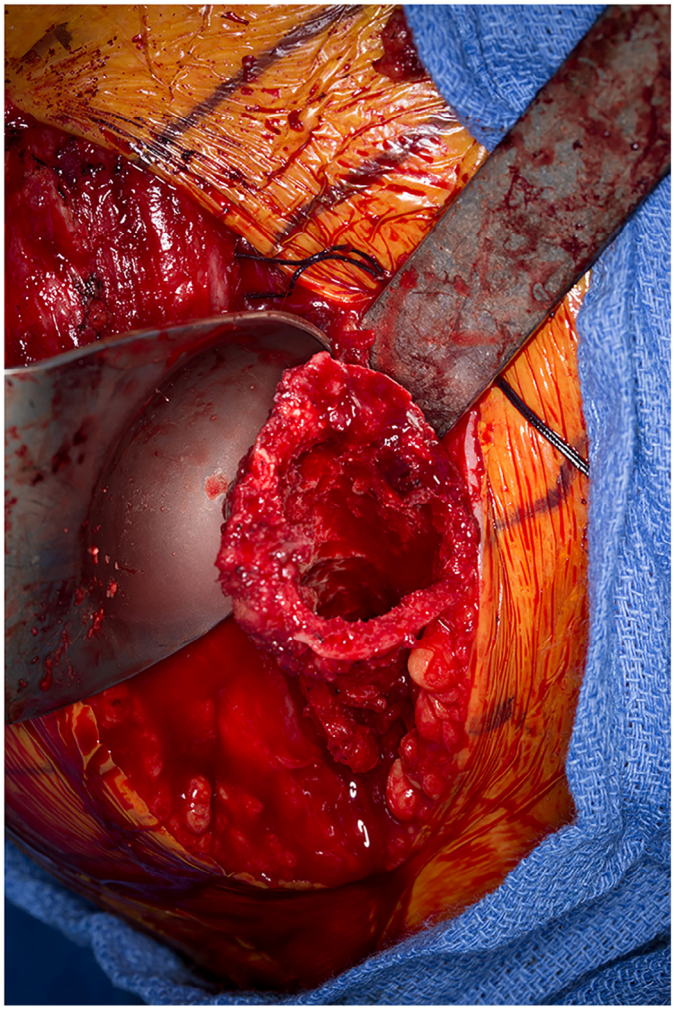

Figure 3 Intraoperative photograph showing appearance of humeral canal after enchondroma was removed by curettage.

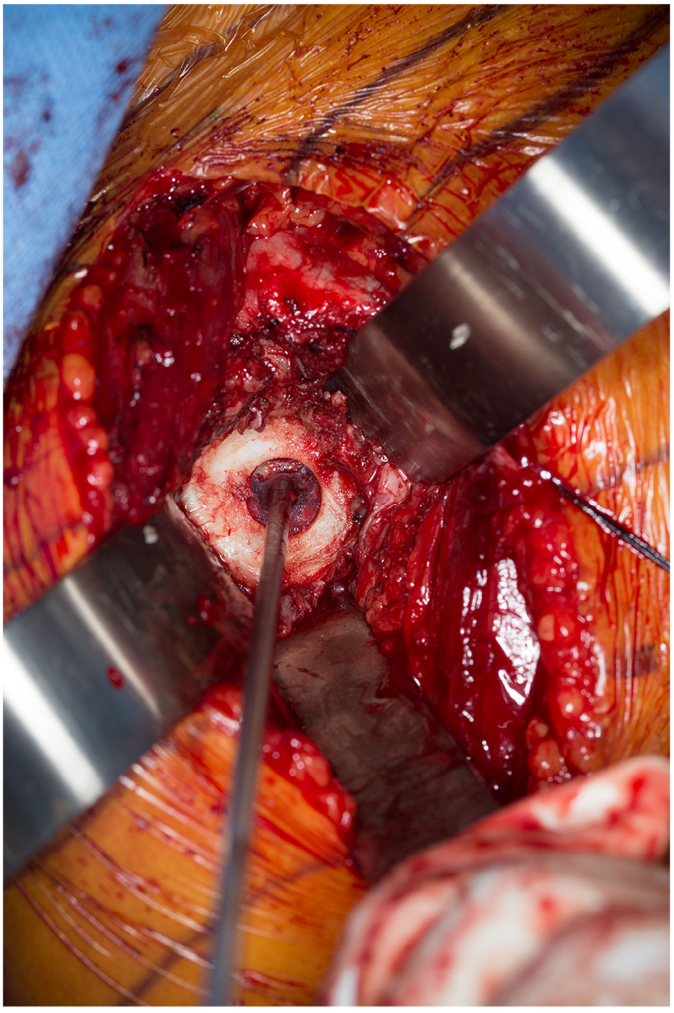

Figure 5 Intraoperative photograph demonstrating glenoid preparation. Care was taken to avoid iatrogenic fracture, given the patient's history of osteogenesis imperfecta.

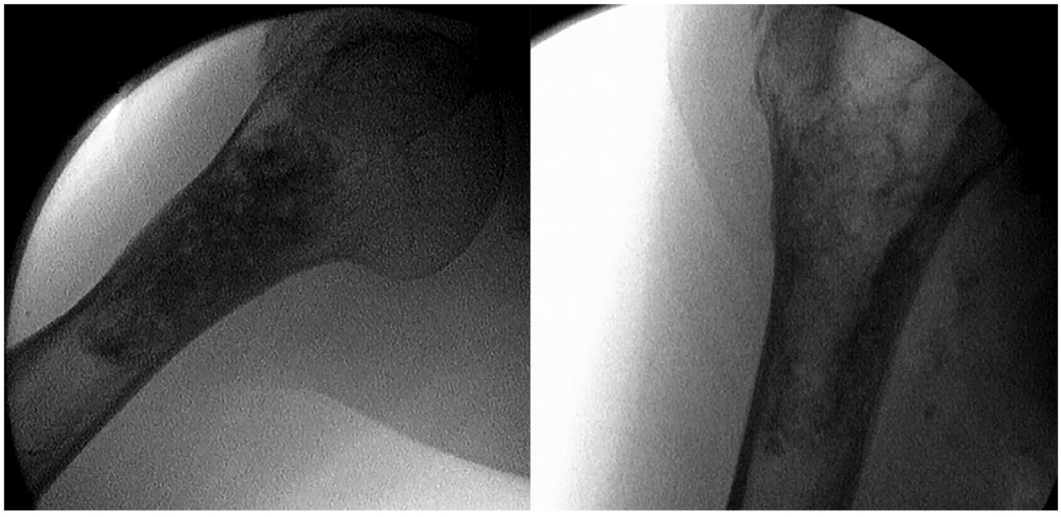

Figure 4 Intraoperative fluoroscopy showing anteroposterior images before (*left*) and after (*right*) curettage.

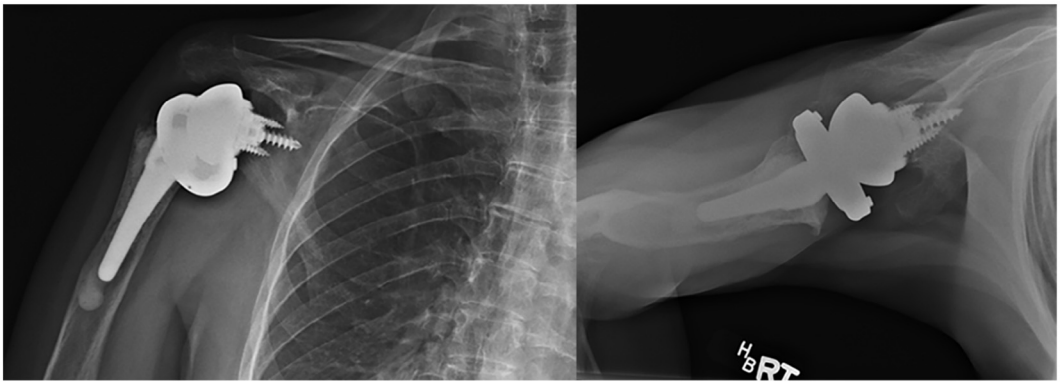

Figure 6 Radiographs at 5 months postoperatively showing excellent implant alignment.

and as a result, it was recommended that components be fixated with cement.[20] In addition, it was noted that intraoperative iatrogenic fracture was common in this population and that care should be taken especially at the time of component impaction to prevent this complication.[20] This is important to note as limited cohorts have demonstrated markedly poor outcomes in both hip and knee arthroplasty in this population if an intraoperative fracture occurs.[8,23] In the largest published cohort, Papagelopoulos and Morrey retrospectively evaluated the Mayo Clinic Total Joint Registry to determine the outcomes of lower extremity arthroplasty in patients with osteogenesis imperfecta and found no complications in this population with the use of cemented total hip or knee arthroplasty at a minimum follow-up of 21 years.[15]

Anecdotal evidence exists to suggest that upper extremity arthroplasty may be an effective treatment option for post-traumatic injuries in patients with osteogenesis imperfecta. Scalise and DeSilva described the successful outcome of a total elbow arthroplasty in a patient with osteogenesis imperfecta who sustained an intra-articular distal humerus fracture.[21] Similarly, Mnif et al reported the successful outcome of a primary cemented total shoulder arthroplasty at 3 years postoperatively after a fracture-dislocation.[13] However, there exists no such evidence to describe upper extremity arthroplasty either in the setting of primary osteoarthritis or with the use of a reverse total shoulder prosthesis.

Enchondromas are the second most common benign cartilaginous tumor and are typically found in the metaphysis of long bones, including the proximal femur and humerus.[11] Hong et al reviewed 477 shoulder MRI scans obtained for routine reasons and found a 2.1% rate of incidental enchondromas, 90% of which were found in the humerus.[6] It has been reported that solitary enchondromas in the proximal shoulder can become symptomatic.[10] Treatment, which consists of curettage followed by either bone grafting or polymethyl methacrylate, has been indicated in those who become symptomatic.[5,9,11] This treatment has been met with acceptable results.[5,11]

The outcomes of cemented arthroplasty in oncologic reconstruction have been well described in the lower extremity reconstruction literature.[1,12,18,19] Randall et al discussed the role of ingrowth in the setting of possible residual disease and subsequently advocated for cemented fixation of the implant to limit the possibility of implant failure.[19] The use of cement was similarly echoed in the upper extremity by Marco et al, who suggested reconstruction with cemented implants after excision of cartilaginous lesions.[11] Reverse total shoulder arthroplasty for proximal humerus reconstructions after tumor resection has recently been found to have good functional outcomes, although a higher rate of complications has been noted as opposed to hemiarthroplasty.[2]

Our patient presented after a failed rotator cuff repair, and given her osteogenesis imperfecta and proximal humeral enchondroma, she presented a unique treatment challenge. A typical patient with rotator cuff arthropathy in the absence of other glenohumeral disease represents an excellent candidate for a reverse total shoulder prosthesis.[3,4,17] However, given the poor bone quality, an experienced musculoskeletal oncologist assisted with the curettage of the proximal humeral enchondroma to prevent iatrogenic fracture or cortex violation, and the humeral component was fixated with polymethyl methacrylate to limit the chance of aseptic failure. The aforementioned physical therapy regimen was undertaken, and the patient reported excellent range of motion and significant improvement of pain at 5 months postoperatively.

Conclusion

This case highlights a challenging patient who underwent a reverse total shoulder arthroplasty in the setting of osteogenesis imperfecta type I in addition to a proximal humeral enchondroma with superb clinical and radiographic outcomes at 5 months postoperatively.

Disclaimer

John W. Sperling reports that he has received royalties from Biomet. All the other authors, their immediate families, and any research foundations 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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