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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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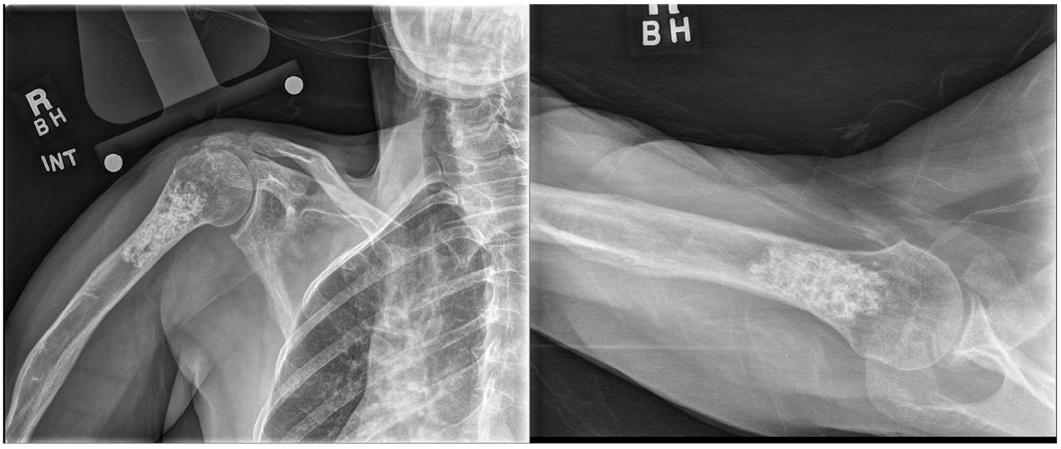

Figure 1 Anteroposterior and lateral radiographs demonstrating a superior-riding humeral head with a proximal humerus enchondroma.

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,

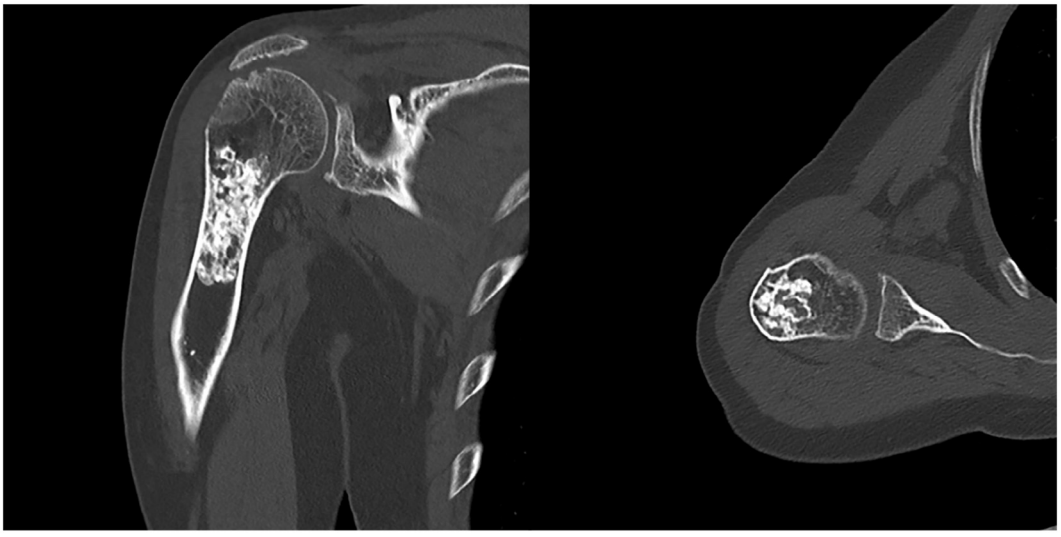

Figure 2 Coronal and axial computed tomography scans demonstrating articulation between acromion and humeral head as well as proximal humerus enchondroma.

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