Functional Improvement After Humeral Shaft Nonunion in a Patient With Glenohumeral Ankylosis

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Abstract

Functionally limiting heterotopic ossification about the shoulder represents an uncommon clinical entity, which has been most commonly reported as a consequence of prolonged immobilization in intensive care unit patients. Severe cases may result in complete glenohumeral ankylosis, with resultant upper extremity motion through the scapulothoracic joint, and significant functional consequences.

We report the case of a 72-year-old male with spontaneous glenohumeral ankylosis who suffered a humeral shaft fracture with resultant painless nonunion. Motion through the nonunion site caused significant subjective functional improvements, increased range of motion, and the ability to complete his activities of daily living.

Patients with limited shoulder range of motion may be at higher risk for humeral fractures and nonunion. These patients, however, may experience improved function due to increased upper extremity range of motion through the nonunion site.

Heterotopic ossification (HO) is the formation of normal bone in an abnormal soft tissue location due to an alteration of the normal regulatory mechanisms of osseous formation.[1] Heterotopic ossification involving the shoulder is an uncommon clinical scenario which has been mostly encountered as a consequence of shoulder arthroplasty.[2] A 2005 retrospective review in the *Journal of Shoulder and Elbow Surgery* demonstrated an overall incidence of HO to be approximately 15% for patients undergoing both hemiarthroplasty and total shoulder arthroplasty.[2] However, the overwhelming majority of the cases represented low grade or clinically insignificant ossification primarily involving the soft tissues about the inferior glenoid margin.[3] Heterotopic ossification about the shoulder in the absence of surgical intervention has been previously reported in the critical care literature as a consequence of head injury or neurologic insult resulting in permanent or prolonged immobilization and the creation of an osteogenic environment.[4]

Humeral shaft fractures are a relatively common orthopedic injury and represent approximately 3%-5% of all fractures.[5] Nonsurgical management of these fractures continues to be the mainstay of treatment and has, in most clinical scenarios, produced adequate results.[5] Nonunion after nonsurgical management of a humeral shaft fracture is defined as the lack of radiographic and clinical union by approximately 24-32 weeks after the initial injury.[6] Fortunately, this circumstance is encountered infrequently, as published reports have documented union rates of greater than 90% for fractures treated with functional bracing.[5]

After a thorough review of the literature using PubMed and Medline, we are not aware of any reports of painless nonunion of a humeral shaft fracture in a patient with a prior ankylosis of the ipsilateral shoulder. The purpose of this case report was to document a patient's functional improvement after nonunion of a humeral shaft fracture in the setting of ipsilateral scapulohumeral ankylosis. The patient provided written informed consent for print and electronic publication of this case report.

Case Report

A 72-year-old right-hand dominant male school-bus driver presented to our clinic in 2001 for decreased range of motion of his right shoulder. He had a history of prolonged intensive care unit (ICU) stay for sepsis after bowel resection in 2000.

Over the course of approximately 6 months post-ICU discharge, he developed the insidious onset of increasing right shoulder stiffness and mild discomfort in his right shoulder with terminal range of motion. At initial orthopedic presentation in August of 2001, the patient had approximately 20° of active and passive internal rotation, and external rotation actively and passively to neutral. Abduction and forward flexion were 50° and 60°, and equivalent both actively and passively. Upper extremity motion was attributed to the scapulothoracic

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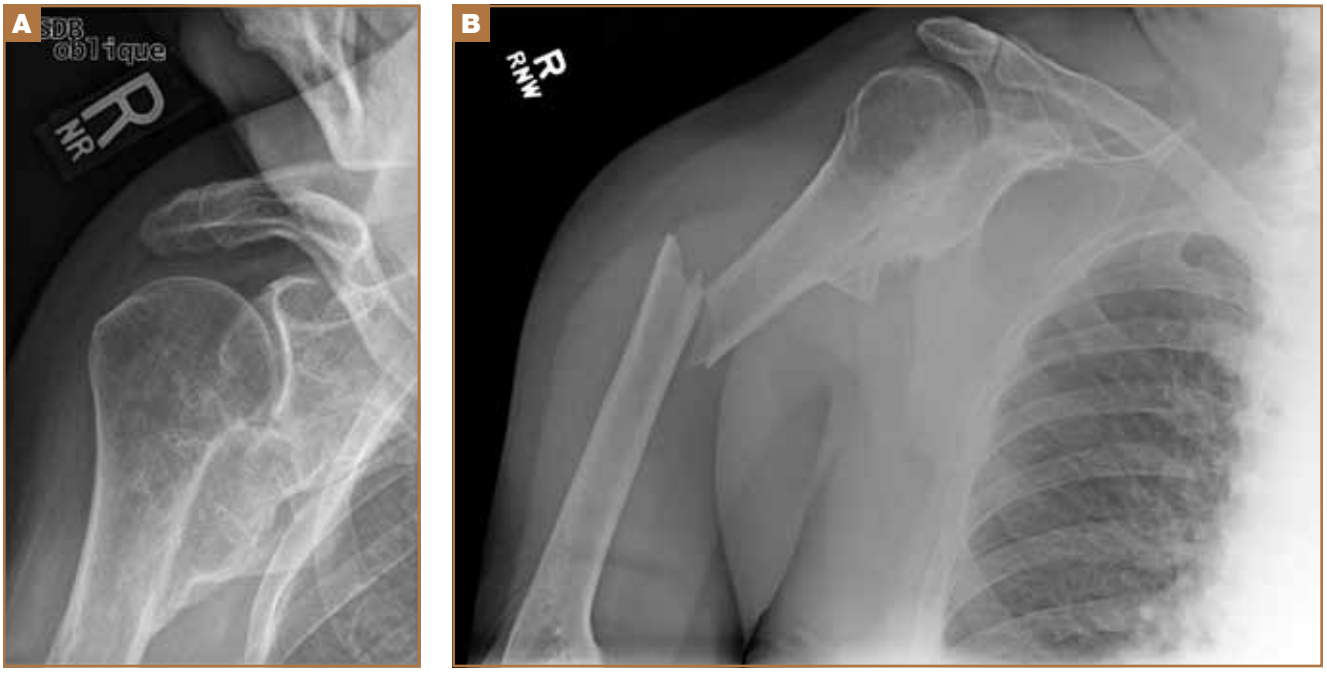

Figure 1. Anteroposterior view (A) of the glenoid prior to humeral shaft fracture. There is extensive bridging heterotopic ossification traversing the scapulohumeral interval. Anteroposterior view (B) of the humeral shaft at the time of initial humeral shaft fracture evaluation. There is significant varus angulation and displacement. The bridging HO in the scapulohumeral interval is still noted.

articulation as no motion was appreciated at the glenohumeral articulation on clinical examination. Elbow range of motion was full. In addition, the patient was noted to have an ulnar nerve sensory deficit and weakness to 4+/5 intrinsic and grip strength. Motor and sensory function was otherwise normal.

The patient underwent radiographic evaluation with plain x-rays (**Figure 1A**) and computed tomography (CT) scan, which demonstrated preservation of the glenohumeral joint space but extensive, dense bony union between the inferior and posterior glenoid neck and the proximal shaft of the humerus. In addition, it was noted fluoroscopically that attempted motion through the glenohumeral articulation resulted in motion exclusively at the scapulothoracic joint.

The patient had significant functional deficits. He was unable to reach his face or mouth, and had difficulty dressing himself and driving, which necessitated an extended period away from his work as a school bus driver. Due to these concerns, surgical takedown of the heterotopic bone with postoperative radiation therapy was offered in 2007, 6 years after presentation. An extensive discussion regarding the risk of neurologic injury was also included. Due to the patient's concern for potential neurologic injury the patient elected to proceed with nonoperative management.

In January 2011, approximately 4 years after his most recent orthopedic visit, the patient tripped, fell onto his right side, and presented to an outside institution where a middle-third diaphyseal humerus fracture was diagnosed (**Figure 1B**). The fracture was completely displaced, varus angulated, and short oblique in nature. He was splinted and followed-up 6 days later, where he was noted to be neurologically unchanged from his previous examination. Plain radiographs of the humerus at the time of this initial follow-up demonstrated the fracture to be in satisfactory alignment (10° varus and apex posterior angulation, 50% displacement). Subsequent follow-up at approximately 2 weeks and 4 weeks after the injury demonstrated an unchanged neurovascular examination and preserved fracture alignment, with the appearance of early healing callus. Approximately 4 weeks after the initial injury, the patient was transitioned to a molded fracture brace. Subsequent follow-up at approximately 9 weeks post-injury showed continued improvement in the patient's level of discomfort, preserved fracture alignment, and increased fracture healing (**Figure 2**). At that time, the patient's brace was discontinued.

Figure 2. Anteroposterior view of the humeral shaft approximately 9 weeks after the injury. There is significant healing callus present and improvement in fracture position.

Approximately 12 weeks

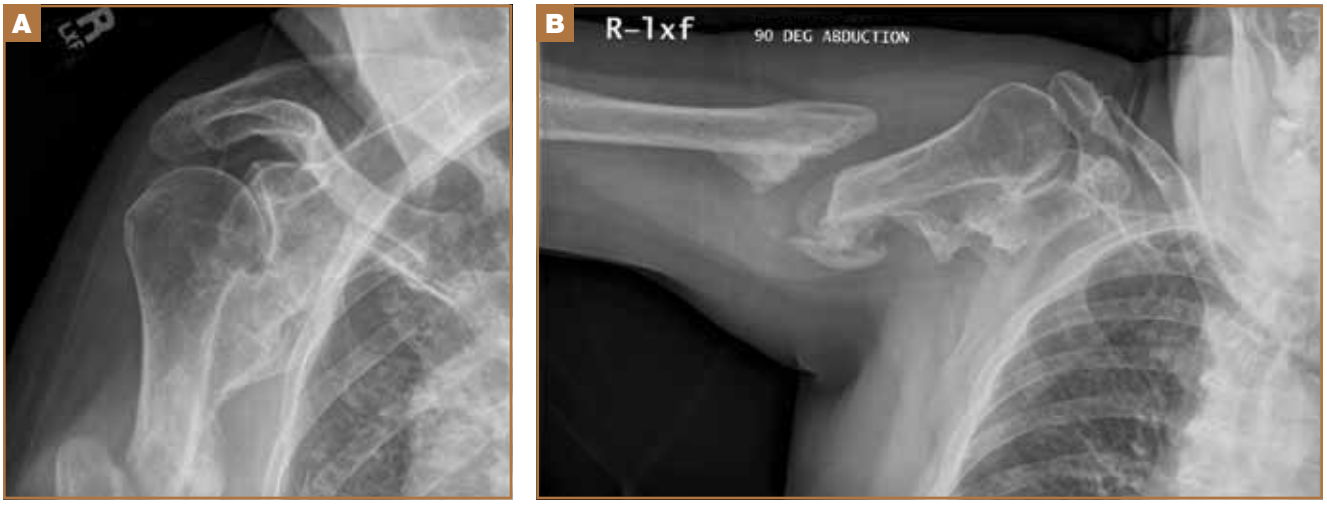

Figure 3. Anteroposterior view (A) of the glenoid at 10 months after the injury. The extensive bridging callus is still present. Anteroposterior view (B) of the shoulder and proximal humeral shaft in 90° of abduction at 10 months after the injury. There is noted displacement at the nonunion site, motion through the scapulothoracic articulation, and no significant change in position at the glenohumeral joint.

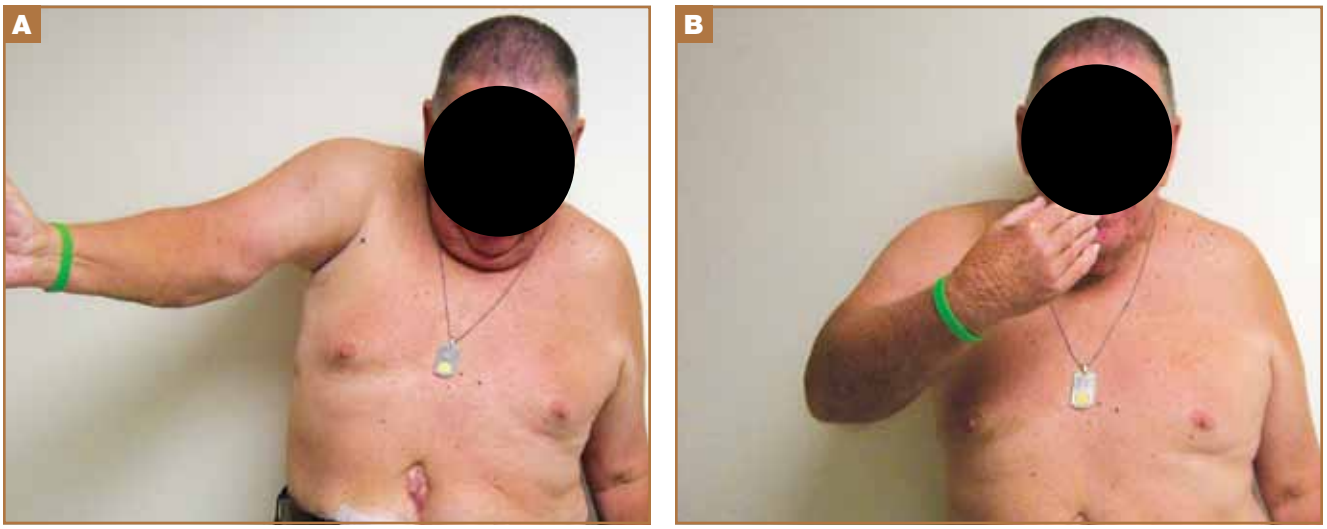

Figure 4. Patient demonstrating active abduction in the scapular plane at 10 months after humeral shaft injury (A). Patient demonstrating ability to reach face and mouth at 10 months after humeral shaft injury (B).

post-injury, the patient suffered a fall onto his right upper extremity causing increasing pain at the site of his fracture. Plain radiographs demonstrated an increase in fracture displacement and angulation (approximately 100% displacement, 20° varus angulation), and an obvious disruption at the site of healing callus. The patient was placed back into a fracture brace, and x-rays 10 days later demonstrated acceptable alignment of the fracture (approximately 2° varus angulation and 40% displacement).

At that time, concerns for impending nonunion prompted the use of a bone stimulator to augment healing. Follow-up appointments at approximately 6 weeks and 12 weeks later (5 and 6.5 months postinjury) were notable for no discomfort at the site of injury, unchanged neurovascular examination, and radiographs significant for a persistent hypertrophic nonunion of his midshaft humerus fracture. At 6.5 months from the original injury, the fracture brace was discontinued.

The patient's most recent follow-up visit was at 10 months postinjury and demonstrated pain-free range of motion at the site of injury (**Figure 3A, 3B**). The patient could abduct his right arm to 90°, forward flex to 90° through the site of nonunion (**Figure 4A, 4B**). Functionally, the patient gained the ability to reach his face and mouth, he is able to dress himself, and has returned to work with limited deficits. A recent survey outlining the patient's functional state demonstrated a simple shoulder test score of 34 (scale 0-100) and a subjective shoulder value of 50 (affected shoulder functions approximately

50% of normal shoulder). The patient was able to complete 5/5 of his activities of daily living including getting dressed, bathing, toileting, doing housework, and preparing meals.

Discussion

Heterotopic ossification of the shoulder without recent surgery is an uncommon circumstance that has been most commonly reported in patients with underlying neurologic injury. Of the patients with HO in the shoulder, only a small subset develop bony ankylosis with functional deficits. The largest reported series of heterotopic ossification in a nonsurgical population involved 447 patients with underlying neurologic injury including both quadriplegia and paraplegia.[7] Overall, 110 patients (25%) developed HO, involving 321 total joints. Of these 321 joints, 37 cases involved the shoulder (11.5%) and 5 of these 37 resulted in bony ankylosis as diagnosed by the presence of a bony bridge on radiographs.

The presence of HO in critically ill patients without underlying neurologic injury also represents a unique circumstance with only a few cases reported in the literature. In 1993, Clements and Camilli[4] reported 3 cases of patients with clinically significant HO and sepsis requiring prolonged ventilatory support. One of these patients had HO in bilateral shoulders resulting in significant limitation of shoulder motion. Furthermore, in a 1999 case report in the *Journal of Shoulder and Elbow Surgery*, Warner and colleagues[8] described spontaneous scapulohumeral ankylosis due to HO in a critically ill patient with sepsis.

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The management of functionally limiting HO of the shoulder is not well defined. Various methods of medical management including radiation, bisphosphonates, and non–steroidal anti-inflammatories have been described with no single method of treatment being universally accepted.[9] Surgical resection of the bridging bone has been reported with varying success. In 1986, Wenner[10] reported a case of ankylosis of the shoulder after scapulohumeral bridging HO, which was treated with surgical resection. Resection resulted in improvement in shoulder range of motion, but was complicated by an axillary neuropraxia and the redevelopment of HO in the scapulohumeral interval. Warner and colleagues[8] reported a case of surgical resection of HO in the scapulohumeral interval, which resulted in marked improvement in range of motion to near full, and no noted postoperative complications.

Little data exist regarding function after scapulohumeral HO resulting in ankylosis of the shoulder. However, studies evaluating function after surgical glenohumeral arthrodesis have been performed. This may or may not reflect the level of function after spontaneous ankylosis as glenohumeral fusions are purposefully performed in a shoulder position, which would maximize the patient's level of function. In addition, glenohumeral arthrodesis is performed for a variety of conditions, which may ultimately affect function distal to the level of the fusion (eg, brachial plexus injury, etc). A retrospective review of 71 patients who underwent shoulder fusion demonstrated that approximately 70% were able to perform daily activities adequately, including feeding, dressing, and personal hygiene.[1] However, only 21% of these patients were able to perform activities at shoulder height. In 1987, Hawkins and Neer[11] reported 17 patients who underwent glenohumeral arthrodesis for disorders that did not affect distal neurologic function. Only 3 of the 17 patients in the series were satisfied with their functional capabilities, and the majority of patients in the series had difficulty reaching their face and head for hygienic purposes.

This case report documents the improvement in function and upper extremity range of motion after diaphyseal humeral fracture nonunion in a patient with a prior ankylosis of glenohumeral motion due to bridging scapulohumeral heterotopic ossification. It is unclear whether the lack of motion at the glenohumeral joint predisposed this patient to a humeral shaft fracture or to a nonunion. It is possible that in the setting of trauma to the upper extremity, the energy—which normally would be dissipated by glenohumeral motion—could have been concentrated at the humeral diaphysis and caused this fracture. Furthermore, other than the short oblique nature of the fracture, the patient had no obvious risk factors for nonunion. The patient's exuberant callus formation would indicate the presence of a hypertrophic nonunion. It is quite possible that persistent motion at the fracture site, due to limitations at the glenohumeral articulation, may have prevented the secondary ossification of the healing callus, which had been present.

One critical element of this case is the lack of clinical symptoms (eg, pain) associated with the patient's humeral nonunion. If the site of the nonunion was documented as a significant pain generator, open reduction and internal fixation may be considered as a treatment alternative. In addition, surgical treatment of glenohumeral ankylosis, may be considered at the time of humeral fixation as the lack of shoulder motion may lead to subsequent rapid humeral hardware failure, and possible persistent nonunion as abnormal stress may be seen at the site of fixation. Furthermore, isolated fixation of his humerus may, at best, return the patient to the same diminished level of function (due to lack of motion) he had prior to his fracture.

Conclusion

The management of glenohumeral ankylosis due to heterotopic ossification in the scapulohumeral interval is controversial as the majority of cases occur in patients who are medically debilitated; the surgical management of this condition places the local neurovascular structures at significant risk. Furthermore,

patients with severely limited shoulder range of motion may be at higher risk for humeral fractures and subsequent nonunion. However, these patients may experience improved function due to increased upper extremity range of motion. As a result, a detailed discussion of the risks and benefits of the treatment of glenohumeral ankylosis, and the humeral fractures that may develop in this setting, should take place between the treating physician and the patient.

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This paper will be judged for the Resident Writer's Award.