



Reverse shoulder arthroplasty in a patient with Paget's disease using the modified Ling technique



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ARTICLE INFO

Keywords:

Reverse
Shoulder arthroplasty
Paget's
Humerus
Ling technique
Reconstruction
Impaction graft

Paget's disease of bone is a chronic condition of increased bone resorption, formation, and remodeling that leads to distorted bony anatomy and ectatic cortical expansion.⁷ Despite being first described in 1877, the etiology of Paget's disease remains unclear.¹ Paget's disease is rarely found in patients under the age of 40 and doubles in incidence for every decade after 50, with a strong predilection for white patients.⁷ The estimated incidence is 1.5%-3% of patients in the United States over the age of 60, with seventy-five percent of cases being polyostotic and the remainder monostotic.⁷

Owing to the relatively high prevalence of Paget's disease involving the proximal femur compared with other periarticular locations, there has been significantly more experience performing total hip arthroplasty (THA) in patients with Paget's disease than other joint arthroplasty procedures.^{4-6,8,10,11} Studies have found that these patients have higher complication rates and worse outcomes than others without Paget's disease undergoing THA.⁵ The main challenge in obtaining a successful THA outcome in Paget's disease patients has been obtaining adequate femoral component fixation and implant longevity due to high rates of loosening and stem subsidence over time.^{5,8}

In an effort to optimize femoral component fixation in patients with Paget's disease, Dr. Robin S. Ling (inventor of the Exeter hip system) and colleagues described a technique for these patients that involves impaction grafting cancellous allograft bone into ectatic femoral pockets followed by cementing the femoral stem.^{8,9} This method has become known as the "Ling technique" and has been used in primary and revision THA surgeries for patients with Paget's disease has led to improved outcome over traditional techniques.⁸ The goal of the Ling technique is to achieve immediate femoral stem stability that does not predispose to stem subsidence over time.

As Paget's disease of the shoulder girdle is less common than of the hip, the collective experience of shoulder arthroplasty in patients with Paget's disease is much more limited. Just one case report, of an anatomic total shoulder arthroplasty (TSA) in a patient with Paget's disease involving the proximal humerus, has been described in the literature.¹³ There has not been a description of reverse shoulder arthroplasty (RSA) in a patient with Paget's disease or of the Ling technique used in the upper extremity for humeral component fixation.

Therefore, in this case report, we describe our experience treating a patient with glenohumeral osteoarthritis in the setting of an incompetent rotator cuff and Paget's disease involving the entirety of the humerus. An RSA was performed utilizing the Ling technique. The purpose of this case report is to demonstrate the successful application of the Ling technique to the upper extremity in conjunction with RSA, to review the relevant literature, and to provide recommendations for performing shoulder arthroplasty in patients with Paget's disease of the humerus.

Institutional review board approval was received from Mayo Clinic (12-007498).

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<https://doi.org/10.1016/j.xrrt.2020.100001>

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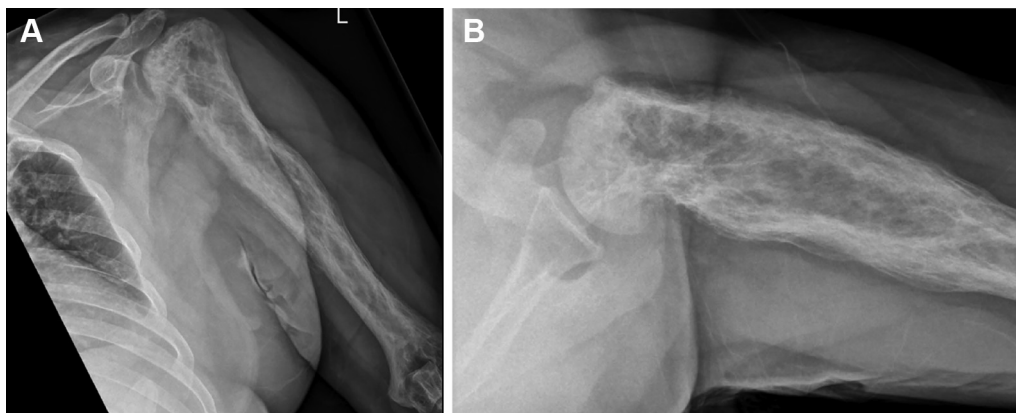


Figure 1 Preoperative AP (A) and axillary (B) radiographs demonstrating Paget's disease involving the entirety of the left humerus as well as humeral head deformity and proximal migration.

Case report

Preoperative evaluation

A 62-year-old right-hand dominant white male with a known history of Paget's disease affecting his left humerus presented to orthopedic clinic complaining of a one-year history of left-sided shoulder pain. The pain was worsened by manual activities and affected the patient's sleep. He had tried a course of nonoperative therapies, including oral anti-inflammatories and one intra-articular steroid injection, without persistent resolution of the pain. He had also attempted bisphosphonate therapy without resolution of his pain. His Paget's disease was initially diagnosed incidentally on plain radiographs obtained in his forties. He had no family history of Paget's disease and his disease was isolated to his humerus.

On physical examination, the patient had 80° of active forward elevation, 90° of passive forward elevation, 10° of external rotation, internal rotation to his sacrum. He had grade 4 strength in forward elevation, external rotation, and internal rotation. The patient had crepitus and shoulder pain with both active and passive range of motion and a positive articular shear test. His neurovascular examination was normal.

Plain radiographs demonstrated Paget's disease involvement of the entirety of the left humerus with deformity and proximal migration of the humeral head (Fig. 1). Preoperative CT scan demonstrated articular irregularities of the humeral head and glenoid (Fig. 2).

Given the patient's new onset of pain, an orthopedic oncology consultation was obtained to rule out sarcomatous transformation. An MRI of the left humerus was obtained which demonstrated no evidence of a malignant tumor and a whole-body bone scan was obtained which demonstrated isolated involvement of the left humerus.

After a discussion of the available nonoperative and operative treatment options, the patient wished to proceed with surgical management in the form of shoulder arthroplasty.

Surgery

After general anesthesia was induced, the patient was positioned in the standard beach chair position. His left upper extremity was prepped and draped in the usual sterile manner. The deltopectoral approach was utilized. The cephalic vein was

preserved and retracted medially. The long head of the biceps tendon was tenodesed in the bicipital groove.

A subscapularis tenotomy was performed. The humeral head was dislocated and noted to be irregular with areas of wear and frank step-off. Evaluation of the rotator cuff demonstrated an irreparable supraspinatus tendon tear and, therefore, RSA was indicated. The humeral head was cut at 30° of retroversion. The humeral canal was entered, reamed, and then broached. Fluoroscopy was available but not utilized in this case. The glenoid was then exposed, prepared, and the metaglene was placed at the inferior aspect of the native glenoid. A central compression screw and three peripheral nonlocking screws were used to obtain fixation. The glenosphere was then impacted onto the metaglene.

Returning to the humerus, the decision was made to proceed with impaction grafting and cementation in accordance with the Ling technique. First, the trial broach was removed and a distal cement restrictor was placed in the medullary canal approximately 2 cm distal to the anticipated humeral stem tip. Next, allograft cancellous bone chips were impacted in the ectatic cavity defects of the humerus. Impaction grafting tamps were used to insert and compress cancellous allograft bone chips around a trial stem, which was intermittently removed and replaced to aid in impaction (Fig. 3). When adequate impaction grafting was confirmed via visual intramedullary inspection, antibiotic-loaded cement was injected into the humeral canal. Fluoroscopy was not required in this case to confirm the adequacy of impaction grafting before cementation but was available had there been any doubt. The final humeral stem, 2 mm smaller in diameter than the trial stem, was then inserted in the appropriate version.

After cement hardening and trialing, the final humeral tray and insert were selected and the joint was reduced. The shoulder was taken through a range of motion and found to be stable without evidence of impingement. The surgical site was thoroughly irrigated, the subscapularis tenotomy was closed with interrupted absorbable suture, and the superficial tissues were closed in standard fashion. The surgical wound was covered with a sterile dressing and the arm was placed into a shoulder immobilizer.

Postoperative course

The patient had an uneventful recovery in the postoperative anesthesia care unit and was admitted to the orthopedic ward. There were no perioperative complications. He was discharged home on postoperative day 1. He began immediate gentle

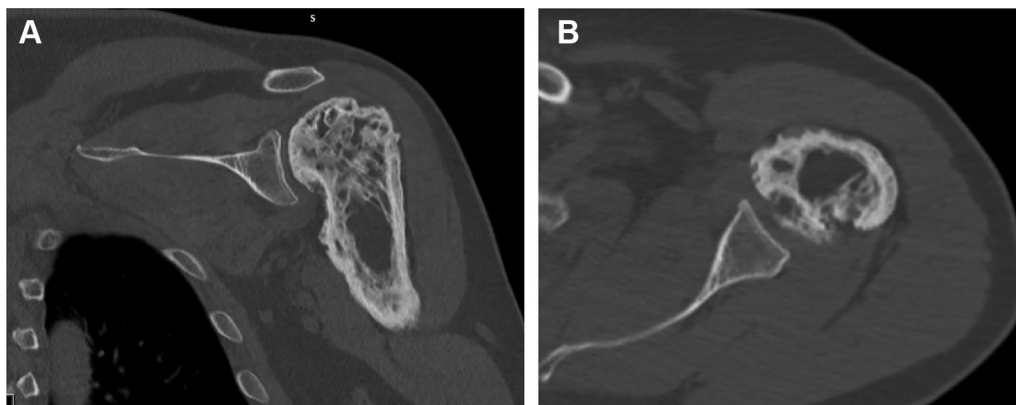


Figure 2 Representative preoperative coronal (A) and axial (B) CT scan images demonstrating Paget's disease isolated to the left humerus as well as bipolar articular irregularities.

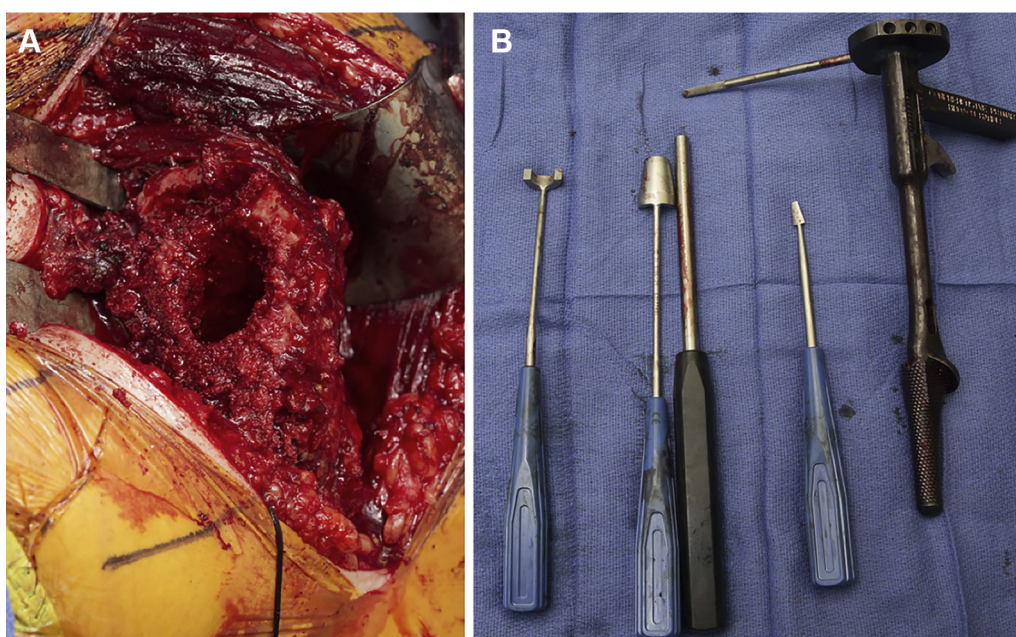


Figure 3 Intraoperative photograph of the proximal humerus after humeral head cut and initial canal preparation demonstrating a large intramedullary cavity (A). Impaction grafting tamps were used to compress cancellous allograft bone chips around a trial stem (B).

pendulum exercises followed by formal physical therapy for strengthening and range of motion at 6 weeks postoperatively. He was able to return to all of his daily activities by 3 months.

At his two-year follow-up appointment, the patient reported that he had no pain at rest or during activity in his left shoulder. He was able to resume comfortable sleep and golf. His active range of motion had increased to 160 degrees of forward elevation, 30 degrees of external rotation, and internal rotation to the hip. His ASES shoulder score was 92 of 100. Radiographs demonstrated an excellent humeral cement mantle with no evidence of progressive loosening or stem subsidence (Fig. 4).

Discussion

Paget's disease is a chronic condition of unknown etiology that results in increased bone resorption, formation, and remodeling that affects older patients of white descent.⁷ The disease may involve arthritic joints and can complicate joint arthroplasty surgery, most commonly THA. The purpose of this case report is to

describe the surgical management of a patient with shoulder arthritis, an irreparable rotator cuff tear, and Paget's disease of the ipsilateral humerus. Using a reverse prosthesis along with the Ling technique, a method of achieving femoral stem fixation in THA via impaction grafting and cementation, the patient was able to obtain an excellent clinical and radiographic outcome at 2 years postoperatively.

The most significant challenge in performing successful joint arthroplasty in patients with Paget's disease is achieving adequate fixation of stemmed components, such as in the femur or humerus.⁴⁻⁶ The ectatic intramedullary deformity of Paget's disease complicates time-zero stem stability. In addition, the inherently abnormal bone remodeling process in patients with Paget's disease prevents normal bony ongrowth with press fit components, resulting in an incompetent bone-implant interface and eventual loosening with stem subsidence.

Many authors have proposed methods to achieve femoral implant stability in patients with Paget's disease undergoing THA due to high rates of loosening. The Ling technique is one of these

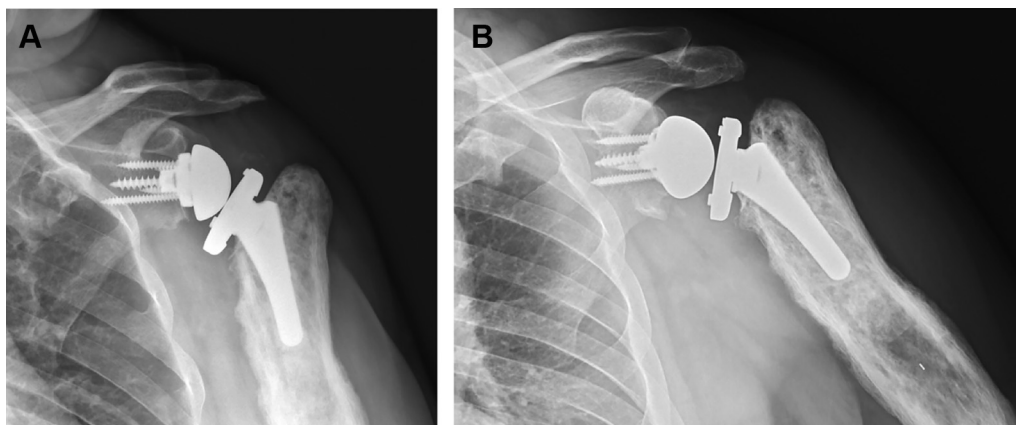


Figure 4 Six-week (A) and two-year (B) postoperative radiographs demonstrate a robust cement/bone graft mantle about the humeral stem with no evidence of loosening or subsidence.

Table 1

Preoperative, intraoperative, and postoperative considerations for patients with Paget’s disease undergoing shoulder arthroplasty.

Preoperative	Intraoperative	Postoperative
Paget’s sarcoma	Geometric deformity	Stem subsidence
Cardiac clearance	Humeral canal preparation	Periprosthetic fracture
Anticipated increased bleeding	Humeral implant fixation (use of the Ling technique)	Heterotopic ossification
	Intraoperative fracture (e.g., during impaction grafting)	

described techniques that utilize impaction grafting of allograft cancellous bone combined with cementation using a smooth, tapered, collarless stem.^{6,8} Implant retrieval histology studies after use of the Ling technique have demonstrated that allograft chips are largely replaced over time by viable cortical bone and, further, that the interface between cement and tissue ultimately resembles that of a primary cemented arthroplasty.¹⁰ Subsidence of the femoral stem still occurs in the Ling technique at rates of 20%–40%, but rates of implant revision surgery are lower than with other techniques.^{4,5} It has been shown that THA stem subsidence after the Ling technique is typically of the stem within the cement mantle, not of the stem/cement complex within bone, and that this stem within cement subsidence is due to the normal “cold flow” phenomenon of smooth, collarless THA implants.^{5,6} Therefore, the subsidence seen in the Ling technique after THA may not be clinically relevant and may not translate to shoulder arthroplasty.

Borrowing from the THA literature, we applied the Ling technique to this patient with Paget’s disease to achieve lasting stability of the humeral stem. We had considered the possibility that the increased constraint of the reverse prosthesis, as compared with an anatomic prosthesis, would confer greater strain to the humeral stem and therefore compromise its stability. Fortunately, in this patient, this appears not to have been the case.

One other case of shoulder arthroplasty in a patient with Paget’s disease has been described.¹³ In this report, an anatomic TSA was performed in a 76-year-old male. Humeral stem fixation was achieved via cementation without the use of cancellous impaction grafting as described by Ling. This patient also had an excellent clinical outcome at two years and no evidence of humeral subsidence or loosening. Despite a not-insignificant incidence of Paget’s disease, particularly in the older population, there appears to be no other reports of shoulder arthroplasty performed in patients with Paget’s disease of the humerus. This is likely a reflection of the lower frequency of shoulder arthroplasty, as compared with hip and knee arthroplasty, as well as a lower incidence of humeral involvement in Paget’s disease, as compared with femoral involvement.

Although we believe that humeral stem stability is the critical aspect of successful shoulder arthroplasty in a patient with Paget’s disease, there are other variables that the surgeon should consider before proceeding with shoulder arthroplasty in these patients (Table 1). Preoperatively, patients with Paget’s disease should be screened for cardiac abnormalities and prepared for an elevated anticipated blood loss, which can be substantial due to the hypervascular nature of Paget’s bone. Some authors recommend having blood products on standby and to use cell saver during surgery.^{7,8,13} In addition, although Paget’s Sarcoma is rare, it is relatively more common in the proximal humerus and should be evaluated as the potential cause of new onset or worsening shoulder pain in these patients.^{2,3,12} Intraoperatively, the humeral canal preparation can be complicated by intramedullary cortical bone and require the use of fluoroscopy, burrs, and cannulated reamers. Owing to thinned cortices, these patients may be at greater risk of intraoperative fracture, particularly with impaction grafting, which can be mitigated with the use of prophylactic cerclage tapes or wires as needed. Postoperatively, these patients are predisposed to periprosthetic fracture due to their deposition of disorganized bone and weakened architecture.¹ Finally, heterotopic ossification is common after THA in patients with Paget’s disease, although this has not yet been reported in shoulder arthroplasty.⁷ We do not recommend the use of routine heterotopic ossification prophylaxis.

Conclusion

We report a patient with shoulder arthritis, an irreparable rotator cuff tear, and humeral Paget’s disease who underwent successful RSA via the Ling technique with excellent clinical and radiographic outcomes at 2 years. The Ling technique has been used successfully in the lower extremity for patients with Paget’s disease of the femur and provided stable humeral component fixation in our patient. Based on this case, we believe that the Ling technique can be successfully combined with RSA in patients with

Paget's disease of the humerus. Longer-term follow-up and more patient outcomes are needed to fully evaluate the efficacy of RSA via the Ling technique in patients with Paget's disease of the humerus.

Conflicts of interest

The 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.

Funding

No external source of funding was used.

Patient consent

Obtained.

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