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Case Report

Multiple Revised Reverse Shoulder Tumour Arthroplasty: When Going Bigger is Not the Only Solution - A Challenging Case Report

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Abstract

Complications after tumoral shoulder total arthroplasty are common and possibly catastrophic. In this case, the patient had a chronic infection treated with a one-stage revision. Subsequently, instability had to be addressed by components exchange. Loosening of the humeral component led to bone stock loss and auto and allograft bone allowed to create enough support for humeral stem implantation. Radial nerve palsy was also observed, without the need for intervention. The diverse complications presented were addressed with different and bold solutions that proved to be successful, with optimal clinical result at the end.

Keywords: Tumor shoulder arthroplasty; Revision; Bone allograft; Infection; Instability; Loosening; Radial nerve palsy

Introduction

The proximal humerus is the third most prevalent location for primary bone malignancies and frequently the site of metastatic illness [1,2]. Lesions with the need for proximal humeral resection provide a major obstacle, especially when the resection needed involves the rotator cuff tendons. Reverse total shoulder mega-prosthesis have grown to be the main choice for oncologic reconstruction providing functional advantages over previous methods [3,4]. However, complication rates reported in the literature are extremely high averaging at around 28% and possibly as high as 67% [5]. Instability is the most frequent complication, followed by infection (around 20%), implant loosening and nerve palsy [5].

Treatment of complications in this scenario is complex as there are several different variables to consider. The management of the oncologic disease often requires adjuvant treatments such as radiotherapy and/or chemotherapy that greatly influence surgical and medical possibilities. Conversely these patients are often young and if they survive their primary disease, their implant will be at risk for failure for a very long period.

Managing these failures requires a vast knowledge and surgical expertise, especially in infection and loosening situations. Each revision surgery is associated with increasing loss of bone stock and usually increasing the size of the mega-prosthesis is the "easy" way-

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out. However, especially in young active patients, this might not be the ideal solution.

The authors describe a case of a young patient originally treated for proximal humerus osteosarcoma that underwent multiple revision surgeries and presented all the main complications associated to reverse total shoulder mega-prosthesis. A few different strategies were adopted at different time points culminating with complex distal humerus bone stock augmentation.

Case Presentation

This otherwise healthy patient was initially treated in our department's musculoskeletal oncology team for a proximal right humerus osteosarcoma when he was 19 years old. He underwent surgical resection and reconstruction with reverse total shoulder mega prosthesis and also multiple cycles of chemo and radiotherapy. The patient developed an early prosthetic joint infection treated with multiple debridement and antibiotics with implant retention. During follow-up he showed no signs of local or metastatic oncologic disease.

In 2017, at age 37, he was referred to consultation with the multidisciplinary infection team for a persistent infection. He had been living with an active sinus tract for the past seven years with no systemic impact of the infection or obvious loosening (Figure 1) but pain had been increasing recently. Infection was already obvious but a decision to perform fluoroscopic guided peri implant arthrocentesis and biopsies was made to try and know the microorganism(s). Multi sensitive *Staphylococcus Epidermidis* was found and consequently, one-stage revision was performed with a silver-coated modular megaprosthesis using a cemented humeral stem (MUTARS*) (Figure 2). Multiple intraoperative tissue samples were positive for the same microorganism and a 12 weeks long regimen of flucloxacillin and rifampicin was instituted with no further clinical/analytic signs of infection persistence.

At the three weeks' time period the patient presented to the outpatient clinic with a shoulder dislocation that could easily be reduced but also easily recurred and a decision to reoperate was taken. Taking advantage of the prosthesis modularity, a limited proximal humerus revision was performed to increase the length and retroversion. After this procedure the patient presented good

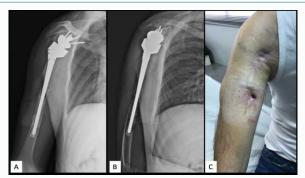


Figure 1: Evaluation of the patient when referred to the multidisciplinary infection team consultation. (A and B). patient's radiographic study; (C). Active sinus tract



Figure 2: (A). Humeral component removed at one-stage revision; (B). Final intra-op aspect after silver-coated modular megaprosthesis and local antibiotic carrier beads in place; (C and D). Radiographic study 1 month post-op, demonstrating prothesis luxation.

evolution, with no further dislocations and being able to perform daily life activities with no significant limitations except minor pain around the middle third of the arm associated with greater efforts (Figure 3).

At the 1.5 years follow-up, pain increased in the elbow and radiologic signs of osteolysis around the cement mantle appeared for the first time. Patient was unwilling to undergo further surgery at this point, but pain gradually increased and subsequent follow revealed progressive osteolysis around the humeral stem and loosening of the stem with very poor bone stock of the distal humerus at the three years' time point. At this point, after extensive discussion exploring different treatment alternatives and associated risks of each one including observation (e.g. increasing pain and high risk of fracture), the patient agreed to a new surgery (Figure 4).

Preoperative investigation (including arthrocentesis and boneimplant interface biopsies were negative for infection) and thus a decision was made to do a partial stem revision. Major difficulty was the lack of appropriate bone stock in the distal humerus. Biologic reconstruction using structural allograft around the distal humeral shaft and impaction grafting using autologous posterior iliac crest autograft was the first step.

In the post op radial nerve palsy was observed, and an expectant attitude adopted, with total resolution after 6 months. From the cultural samples collected intraoperatively, one was positive for *Staphylococcus epidermidis* and another for *Staphylococcus capitis*, so a 12-week treatment with flucloxacillin and rifampicin was prescribed without further clinical or analytic signs of infection. During the first 3 months the patient was kept on brachial suspension and was only



Figure 3: Radiographic evolution after revision due to instability. (A). Post-op; (B). 3 months post-op; (C). 6 months post-op; (D). 1 year post-op; (E). 1,5 years post-op; (F). 2 years post-op; (G-J). 3 years post-op.

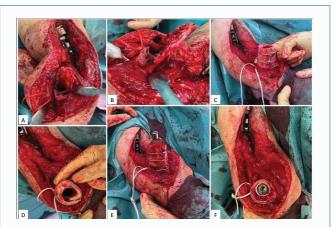


Figure 4: Revision surgery. Through an anterolateral and a medial approach, the revision started by isolating the radial nerve and cubital nerve, respectively. Next, the stem and cement were removed, leaving the proximal components. The remaining humeral bone was debrided and found to be extremely thin and with a lotoff ocal discontinuities (A and B). After copiouslavage, two halves of cortical femoral allo graft bone were attached to the remaining humeral bone with cablecerclage and regularized (C and D). Autograft of bone from both posterior iliac crests was collected and impaction graft performed. This created enough support to implant a cementless stem, and ascrew was added (trough allograft and stem) to giverotational stability (E and F). Lastly the intermediate modular components were assembled granting the correct length and stability to the prothesis.

allowed to do physiotherapy-oriented mobilization of the elbow and shoulder without rotational movements. After this period, there were signs of bone integration, the brachial suspension was removed, and a more intense physiotherapy routine initiated. This allowed having very good mobility at 6-months post op (shoulder: active anterior elevation-80°, abduction-80°, adduction and internal rotation-L4; elbow: flexion-130°, extension- -15°, prono-supination-85° to 85°). The scar developed argyria, a greyish pigmentation due to the silver coat in the tumoral components. With 2 years of follow-up, the allograft bone is totally integrated, without signs of stem loosening; the patient presents no pain complaints and has an active life (Figure 5) (Video 1).

https://www.youtube.com/watch?v=821YOl8Qwzw

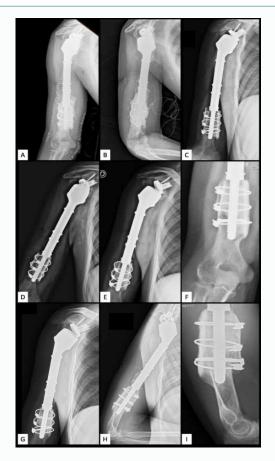


Figure 5: Radiographic evolution after last revision. (A and B). Immediate post-op; (C). 1,5 months post-op; D: 3 months post-op; E and F: 1-year post-op; G-I: 1,5 years post-op.

Discussion

As frequent as complications may be in reverse total shoulder mega-prosthesis, presenting four of the main complications in the same patient is not common. Regarding total shoulder arthroplasty infection, two-stage revision is being considered the gold-standard for the treatment, but one-stage revision shows to be at least as effective, as long as right criteria is adopted [6,7]. In this case, the isolated organism was a multisensitive bacteria, with antibiotic options for systemic and local administration, the patient had no other health problems, and one-stage revision proved to be a good solution.

The instability observed after the revision was related to a slight anteversion and shortage of the humeral component. As a modular stem had been previously used, this allowed correcting it by simply exchanging the modular components to improve rotation and length without removing the implanted stem. Humeral stem loosening is a potentially serious complication due to the poor bone quality secondary to severe bone loss it entails. In this case, the remaining bone stock was not enough to support a new (cemented or uncemented) stem. Mimetizing solutions described for femoral and tibial bone loss, impaction grafting after reinforcing the remaining humerus with allogenic femoral diaphysis bone graft proved to be a solution with excellent results, both clinical and radiologically.

The expectant attitude in the presence of nerve palsy, allowed for the resolution without the need for further interventions, as described for most of the cases in literature [8]. In summary, a profound knowledge of the different solutions and surgical techniques is critical for a successful treatment of each of the various and serious complications of tumour shoulder arthroplasties. One-stage revision arthroplasty is an effective weapon on treating chronic infections, even in the presence of fistula as long as it can be included in the approach and properly debrided. Impaction grafting together with bone allograft is able to create appropriate support for stem revision without the need to "go bigger". An appropriate surgical and medical treatment together with careful rehabilitation was keys to overcome the otherwise catastrophic situation presented.

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