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Reverse shoulder arthroplasty after failed megaprosthesis for osteosarcoma of the proximal humerus: A case report and review of literature

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ABSTRACT

The involvement of proximal humerus by Osteosarcoma is quite common, with the survival rates highly dependent on the staging of the tumor. Numerous salvage methods have been described but without any consensus. We present a case of a 17-year-old patient who had a wide excision of the tumor and underwent a mega prosthesis fixation, which underwent implant failure with the revised prosthesis subluxing. Finally, Reverse Shoulder Arthroplasty was done. Ten years follow up shows the success of the above. Salvage surgery in young patients yields good functional and cosmetic results. However, an insight into the complications associated with extended use of the mega prosthesis is needed. Prolonged survival after bone tumors is associated with surgical, medical and psychological challenges.

1. Introduction

Osteosarcomas are a common entity during bone tumor discussions as they are the most common malignant neoplasm and have been linked to a 15–30% five-year survival rate if metastatic. The proximal humerus is a reasonably frequent site for both primary and metastatic malignancies accounting for 15% of osteosarcoma locations.[1] Early management of these neoplasms is essential for prolonged survival. However, due to vague symptoms, late presentation and lack of diagnostic parameters especially in developing countries, operative intervention is usually radical with reconstruction both complex and expensive.[2]

The Shoulder is a complex joint that requires static and dynamic stabilizers to keep the humeral head in place and to allow for a great range of motion and stability.[3] Historically, amputation or disarticulation was considered as the only available surgical method for the above. Chemotherapy use in conjunction with the surgical intervention was only realized in the 1970s.[4] Currently, there is a paradigm shift towards limb salvage surgery with more than 90% of patients with osteosarcoma undergoing salvage procedures in specialized centers mainly due to progression in prosthetics, diagnostic models and surgical techniques.[5]

Limb-sparing is now the preferred method of managing proximal humeral tumors.[6] Numerous options are available after resection of the tumor for proximal humerus.[7] These include Leaving a flail shoulder (usually not recommended), arthrodesis the shoulder using intercalary graft and arthroplasty if glenoid allows.[7–9] The primary purpose of reconstruction is to restore function and limit complications.[10] Proximal humerus reconstructions secondary to resection is challenging because the joint is naturally unstable, requiring both static and dynamic stabilization systems, much of which is affected during resection to achieve a wide margin.[11]

No consensus as to which method should be used for reconstruction is available. Megaprosthesis, however, is favored due to some reasons including predictable functional outcome, early rehabilitation post surgery, intraoperative flexibility in the length of reconstruction and is usually not affected by adjuvant chemotherapy (nonbiologic).[4] However, its most significant challenge has been wear and tear leading to loosening and failure. Reattachment of soft tissues such as tendons to the prosthesis also poses a test to the surgeon. Proximal migration of the prosthesis has been

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reported by some studies secondary to rotator cuff instability.[12] A decline in prosthetic survival rate has been observed from 94.5% to 88.3% at 5 and ten years respectively[12–14]; hence a possible shift to more geometric and functional alternatives is eminent in the future. Reverse Shoulder Arthroplasty (RSA) is gaining popularity around the world and is used for patients who have shoulder instability with the irreparable or absent rotator cuff. RSA is conventionally reserved for elderly and low demand patients, but an improved understanding of its biomechanics leading to better implant designs and surgical techniques mean that its indications have been expanded vastly.[11,15] Hence surgeons are becoming more adventurous in its use especially in salvage procedures and nonconventional patients.

The purpose of this case report is to highlight some of the problems associated with prolonged survival after tumor surgery around the proximal humerus with an addendum into the challenges faced with megaprosthesis use in a nonconventional patient.

2. Case report

We present a case of a 28-year-old lady who first came to the clinic ten years ago (2008) with a diagnosis of osteosarcoma of the left proximal humerus. She had been diagnosed with the above after a biopsy at a peripheral facility. On presentation, she already had high doses of methotrexate neo-adjuvant chemotherapy. Enneking staging system was Grade II. All other parameters were within normal ranges.

She underwent wide resection with reconstruction using an endoprosthesis done (Fig. 1a and b), after three months of adjuvant chemotherapy with High Dose Methotrexate, Cisplatin, and Entracycline.

She presented to us five years later (2013) with pain in the left arm. Radiographic images revealed a bent prosthesis with subluxation (Fig. 2a and b) We attributed this to her level of activity, is a bubbly teenager who made good use of her shoulder. Revision surgery was carried out in 2014 with PET CT scans during this visit showed no progression of the tumor.

She then presented to us three years later (2017) with pain and limited mobility in her left upper arm. Radiographs (Fig. 3a and b) this time revealed subluxed prosthesis with no signs of infection or loosening. A decision to go for an RSA (Fig. 4a and b) was made given her having sufficient abductor strength and glenoid bone. Subsequent follow-ups have been uneventful after that.

3. Discussion

Osteosarcoma of the proximal humerus poses a challenge for both the surgeon and the patient, equally. The late detection of these tumors coupled with few operable cases makes it a controversial topic with varied opinions amongst various schools of thought. Patient survival is dependent on the tumor grade, whether metastasis has occurred and how the tumor responds to chemotherapy. However, this can be interrupted by inadequate resection by the surgeon.[3,4] Reconstruction following resection has only picked up in the past decade with the forequarter amputation been

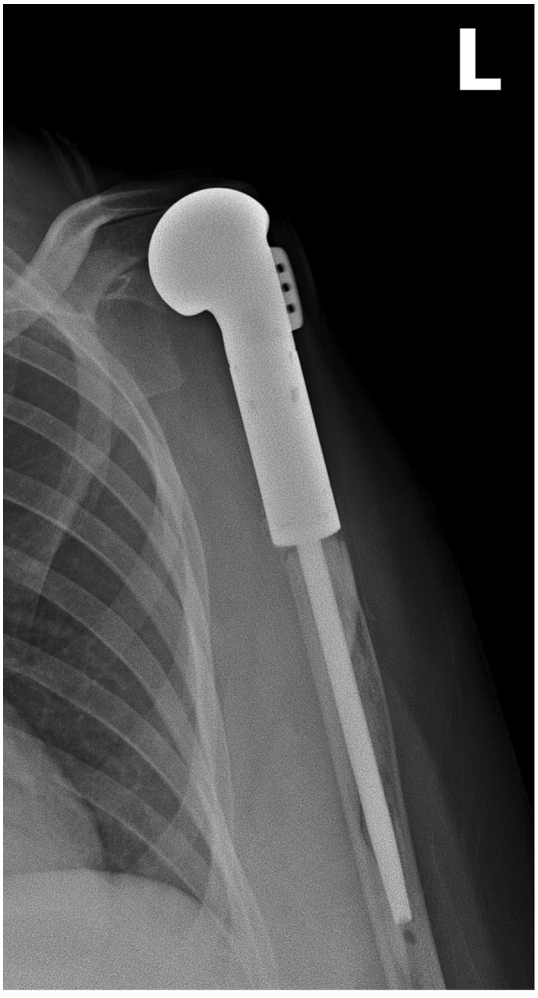

Fig. 1a. AP view of mega-prosthesis post wide excision.

Fig. 1b. Lateral view of mega-prosthesis post wide excision.

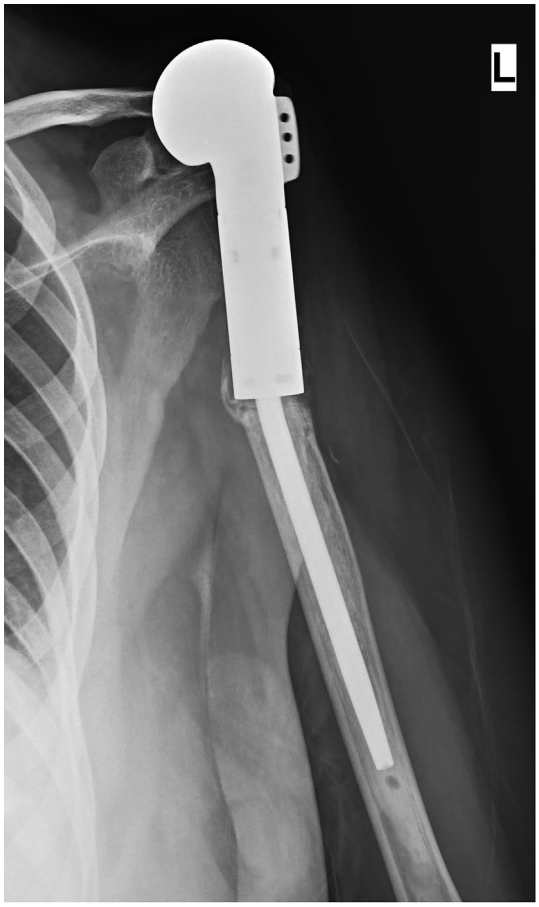

Fig. 2a. AP view of bent mega-prosthesis with subluxation.

Fig. 2b. Lateral view of bent mega-prosthesis with subluxation.

preferred by the most surgeons before that.[16] Reconstruction of the shoulder, after wide resection of the tumor, is challenging. Malawer et al.[16] reported the use of an endoprosthesis after resection of proximal humerus tumor in 1985 with favorable results in his 20-year follow-up. Wittig et al. showed 100% prosthetic survival rate though his sample size was a limiting factor.[3,16]

Shoulder instability has been reported in cases after tumor resection and reconstruction. However, it is an uncommon occurrence as described by Bickels et al. observed that the shoulder is not a weight-bearing joint and allows for even distribution of forces during mobility.[3] Wittigs et al.[16] reported no case of instability, in his study, while Wang et al. reported 40% cases of instability following reconstruction using Allograft-Prosthesis Composite, endoprosthesis and osteoarticular grafts.[11,17] Kassab et al. noted 37.9% instability in their cases[18] with Gautam et al. point out that most bone defects in the proximal humerus are secondary to neoplastic lesions which frequently occur in the skeletally immature that may end up requiring a revision later on.[19] Dubina et al. (2017) in meta-analysis showed that up to 30% of megaprosthesis presented with mechanical failure, while only 4% were infected.[10] However, Gkavardina et al. have reported a decline in mechanical complications involved with mega-prosthesis mainly due to the establishment of more robust and modern modular designs and

Fig. 3a. AP view of post revision mega-prosthesis.

Fig. 3b. Lateral view post revision mega-prosthesis.

Fig. 4a. Radiographs post RSA (AP View).

Fig. 4b. Radiographs post RSA (Oblique View).

improved attachment methods of soft tissues to the prosthesis.[20] The choice of reconstruction is a major determinant in post-operative instability. Our case presented to us after five years with a history of bending and subluxation. In keeping with Dubina et al., we presume that this was perhaps because our patient was skeletally immature at the time of the first surgery and an active patient. Blacksin et al. contribute to this by indicating that less soft tissue support at the surgical resection site coupled with larger constrained implants add to the above .[21]

The goal of the surgery is to provide a stable joint and a fulcrum for elbow flexion. Prolonged survival after tumor resection puts a strain on the above goal. To achieve wide and tumor-free margins, an adequate resection is needed which may compromise the vital tissues such as the rotator cuff, capsule and deltoid muscles. Since most neoplasms exist in the skeletally immature individuals, prosthetic stability poses a challenge. In our case, we attribute the superior subluxation of the prosthesis after the revision, to the imbalances between soft tissues and the implants and the deficient rotator cuff. Cannon et al. reported a 29% incidence of proximal humeral migration with a direct association between migration and the length of the following up.[22] Mayilvahanan et al. reported a 10% complication rate of proximal migration with all of their patients being managed conservatively.[12] Older studies have even shown a

higher rate of migration with 76% by Bos et al.[22] and 56% by Ross et al..[22] It suggests that proximal migration is inevitable in the long run in these cases.

Grammont was the first person to develop the RSA, in 1970 and found it as a solution for rotator cuff arthropathy and failed hemiarthroplasties.[23] Over the years it has evolved further and has now found its role extended beyond those indicated by Grammont[24] with authors like Bonnevialle et al. showing promising results in his study on the use of RSA primarily after resection of malignant tumors of the proximal humerus.[25] It is mainly due to its semiconstrained design and a mediatized center of rotation that allows it to deal effectively with soft tissue and bone defects[26] which provides a stable fulcrum and an increasing deltoid lever arm.[25] Its use in revision surgery for failed hemi prosthesis had increased due to the above reasons, before which revision was mostly planned to improve component position and soft tissue balance. Results were unsatisfactory with Sanchez-Sotelo et al. illustrating 27% success following revision surgeries.[27] Merolla et al. showed a satisfactory outcome in his study of the conversion of failed hemiarthroplasty to RSA with only 7% of those in their study requiring revision.[26] It has been backed up by other authors who reported good clinical outcomes in the revision setting.[26] However, Dewilde et al., and Boileau et al. and a variety of other authors have raised the concern of the RSA in revision setting, but it is important to note that these authors had varying clinical and anatomical parameters hence a correct perception cannot be derived.[26]

To our best knowledge, there is a paucity of data about RSA being used as revision surgery for failed megaprosthesis following osteogenic sarcoma. Maclean et al. had carried out a retrospective study in their tumor center based in the United Kingdom and found only eight cases from a pool of 35000 cases with bone tumors with none having a diagnosis of Osteosarcoma.[2] Bonnevialle et al. found only ten patients in their five-year study with only one with a diagnosis of parosteal osteosarcoma.[25] The reason for the rareness of the use of these prostheses was perhaps due to lesser survivorship and patient follow up of those with osteogenic sarcoma. It devoid us of essential data relating to such problems, if any, encountered with prolonged survivorship of these cases. The age of the patient is also of note here with RSA, as it is mostly recommended for the elderly, vis-à-vis in our case, where the patient was in her early twenties when the procedure was done.

In conclusion, various challenges exist with mega prosthetic use after resection of proximal humerus osteosarcoma with less predictability due to a few reported cases of the above. RSA is a viable option for failed hemiarthroplasty following resection of osteosarcomas. Early detection of osteosarcomas and adequate resection with a good understanding of the reverse shoulder biomechanics is the key to its prolonged survivorship.

Disclaimer

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