Original Article

Indian Journal of Cancer Education and Research
Volume 6 Number 1, January - June 2018
DOI: http://dx.doi.org/10.21088/ijcer.2321.9815.6118.3

Limb Conservation for Tumors of the Proximal Humerus by Endoprosthetic Reconstruction: Our Experience

M. Muralidhar*, M. Srinivasulu*

Author's Affiliation: *Assistant Professor, Department of Surgical Oncology, MNJ Institute of Oncology and Regional Cancer Center, Osmania Medical College, Hyderabad, Telangana 500095, India.

Corresponding Author: M. Muralidhar, Assistant Professor, Department of Surgical Oncology, MNJ Institute of Oncology and Regional Cancer Center, Osmania Medical College, Hyderabad, Telangana 500095, India.
E-mail: mrlidharndu@yahoo.com

Received on 20.10.2017, **Accepted on** 25.11.2017

Abstract

Introduction: Humerus is a common site for malignant tumors. Advances in adjuvant therapies and reconstructive methods provide salvage of the upper limb with improved outcomes. *Materials and Methods:* In our surgical oncology department five patients with tumors of the proximal humerus were operated over the course of two years from 2010 - 2012. *Results:* All patients had different tumor pathology emphasising the wide heterogeneity of sarcomas of bone and soft tissue. Ages ranged from 12yrs to 52yrs, again a broad range. Five patients described above all presented with arm swelling and pain. One patient diagnosed outside as a Giant Cell Tumor by CT scan and FNAC had curettage performed. Both children had high grade sarcomas of the proximal humerus. The 12 year old had a STS of bone and had an intraarticular resection albeit with abductors resected. The 14 year old had an osteosarcoma for which 5 cycles of cisplatin based neoadjuvant chemotherapy had been given. In her case a modified Tickhoff - Linberg resection had to be performed for adequate margin and the prosthesis was suspended statically and dynamically for a stable shoulder. As her abductors had been resected along with the glenoid she had severe restriction of shoulder motion. The 45 year old female underwent a modified Tickhoff - Linberg resection with deltoid sacrifice for a low grade STS due to extensive soft tissue involvement and therefore had severe restriction of shoulder function. We obtained our best results in the two male patients with benign tumors as we were able to spare both the abductors and glenoid and replace the resected humerus with a prosthesis. Both recovered rapidly and had excellent abduction at the shoulder. All had near complete relief of pain and complete function at elbow and wrist. *Conclusions:* Routine endoprosthetic reconstruction for proximal humeral reconstruction is recommended in view of its simplicity of fixation, durability and excellent long term outcome and we reserve autologous fibula only in situations where a prosthesis cannot be used.

Keywords: Humerus; Tickhoff - Linberg Resection; Osteosarcoma.

Introduction

The shoulder girdle is much less frequently involved by tumors than the pelvic girdle. The classical sites of involvement about the shoulder are the proximal humerus and the scapula. The common primary tumors in these areas are Osteosarcomas and Ewings tumors in children and adolescents and Chondrosarcomas in adults. Metastatic tumors too have a propensity to involve the proximal humerus. Common primaries for these metastatic tumors are breast, lung, myeloma, and especially renal cell carcinoma. The axilla is an area of special concern since tumors here can grow to a large size before detection. When soft tissue sarcomas occur in this area they can secondarily involve the bone and bone tumors here can in turn involve the soft tissues. Both situations require similar reconstructive techniques. The magnitude of bone and soft tissue resection and the extant of remaining motor groups dictate the degree of shoulder function achievable [1-6].

The shoulder girdle consists of the scapula, the clavicle (the distal 1/3) and the proximal humerus and surrounding soft tissues. Forequarter amputation was the mainstay of treatment for

shoulder tumors until the middle of the 20th century. Happily nowadays 95% of patients with shoulder girdle sarcomas can be managed with limb sparing surgery such as the Tikhoff-Linberg resection and its modifications in the best centres in the west [7-15].

Prosthetic reconstruction initially was developed for the lower limb bony reconstruction and later for upper limb and shoulder girdle defects. Over a period of time the design has undergone significant refinement and now has been very well standardised. The complication rates of endoprosthetic reconstruction are lower than reconstruction with allograft, composite reconstruction or arthrodesis. Prosthetic survival is reported as 95 - 100% at 10 years. Proximal humeral tumors are technically very challenging .However despite their complexity the majority of these tumors can be resected with present day techniques and amputation avoided. Currently the most commonly used method of reconstruction is endoprosthetic reconstruction. It is used after both intraarticular (Type I) and extraarticular (Type V). After tumor resection and prosthetic placement muscle transfers are used to cover the prosthesis and stabilise the shoulder joint. Type I resection (intraarticular) is avoided for high grade tumors due to high risk of local recurrence. A stable shoulder with normal wrist and elbow function can be achieved in the majority. Here we present a series of cases in our experience.

Materials and Methods

In our surgical oncology department we have five patients with tumors of the proximal humerus operated over the course of two years from 2010 - 2012. They represent the wide spectrum of tumors found in the proximal humerus and also the wide age range in which they are found.

Five consecutive patients were operated by us over a two year period . All procedures were carried out by oncosurgeons, at a single tertiary referral bone tumor center. Following Institutional Board Review approval, data was collected through retrospective analysis of medical records, imaging studies, and questionnaires structured for measurement of patient-derived outcome. There were no wound complications, including superficial and deep infections. All of them had the expected functional recovery of shoulder, elbow and wrist joints. All had a stable shoulder with no pain, subluxation or dislocation. The follow up ranges from two and a half to six years. All are on regular follow up and are disease free at the time of writing. An objective assessment of function was first performed at three months after the procedure and then at 6 monthly intervals.

Results

All patients were referred to us from other hospitals where initial evaluation was done. Interestingly all patients had different tumor pathology emphasising the wide heterogeneity of sarcomas of bone and soft tissue. Ages ranged from 12yrs to 52yrs, again a broad range.

Our five patients all presented with arm swelling and pain. One patient diagnosed outside as a Giant Cell Tumor by CT scan and FNAC had curettage performed, but had a recurrence within two months.

Table 1 and 2 detail the patients age, the type of resection performed and the final outcome achieved as a percentage according to the revised MSTS scoring system after resections involving the upper limb. Both children had high grade sarcomas of the proximal humerus but underwent different types of resections. The 12 year old had a STS of bone and had an intraarticular resection albeit with abductors resected. The ideal reconstruction would have been a prosthesis with a head which could have been articulated with the glenoid and which would have simulated a normal glenohumeral joint Unfortunately the child was thin statured and even the smallest size of the modular prosthesis was too large for her remnant humerus. We were forced to use her fibula for bony reconstruction and we feel that this could have compromised her final outcome of shoulder abduction.

Table 1: Revised musculoskeletal tumor society functional score for upper limb

	Pain	Function	Emotional acceptance	Hand positioning	Manual Dexterity	Lifting ability
Patient 1 (45/F)	4/5	2/5	3/5	3/5	5/5	2/5
Patient 2 (12/F)	5/5	2/5	4/5	3/5	5/5	2/5
Patient 3 (52/M)	5/5	3/5	5/5	4/5	5/5	3/5
Patient 4 (23/M)	5/5	3/5	5/5	4/5	5/5	3/5
Patient 5 (14/F)	4/5	2/5	3/5	3/5	5/5	2/5

Table 2: Type of resection and MSTS Scoring System for upper limb function

45/F	VB	19/30	63%
12/F	IB	21/30	70%
52/M	IA	25/30	83%
23/M	IA	25/30	83%
14/F	VB	19/30	63%

The 14 year old had an osteosarcoma for which 5 cycles of cisplatin based neoadjuvant chemotherapy had been given. In her case a modified Tickhoff - Linberg resection had to be performed for adequate margin and the prosthesis was suspended statically and dynamically for a stable shoulder. As her abductors had been resected along with the glenoid she had severe resstriction of shoulder motion.

The 45 year old lady underwent a modified Tickhoff - Linberg resection for a low grade STS due to extensive soft tissue involvement and therefore had severe restriction of shoulder function.

We obtained our best results in the two male patients with benign tumors as we were able to spare both the abductors (rotator cuff, deltoid and axillary nerve) and glenoid and replace the resected humerus with a prosthesis. Both of these patients were very fit, muscular men and the older patient was a manual labourer. Recovered rapidly and hand excellent abduction at the shoulder. All had near complete relief of pain and complete function at elbow and wrist.

Discussion

The proximal humerus is the third most common site for primary bone tumors and also a common site for metastatic disease [1]. Since the 1970s the management of primary bone tumors has changed from amputations to limb conservation, with replacement of resected bone and muscle reconstruction, due to advances in chemotherapy, surgical techniquies and imaging. Studies have shown that limb conservation results in survival rates similar to amputation at the cost of increased local recurrences [2,3,4]. For patients, limb salvage is a phenomenal improvement in their quality of life, emotionally and socially. In addition, salvage of the upper limb results in much superior function than after an amputation [5]. The best method of reconstruction after proximal humerus resection has long been controversial, in part due to the literature having relatively few studies with significant number of enrolled patients, making reliable conclusions difficult. The options are autografts (fibular autografts or autoclaved humerus), osteoarticular allografts, allograft - prosthetic composites, the clavicula pro humero procedure or finally a endoprosthesis. (5-11 see D kumar et al.). Some authors have criticised the endoprosthesis as being little more than a spacer (12-14 see D kumar et al.). However it must be noted that it is the technically simplest form of reconstruction for the proximal humerus. In addition endoprostheses are readily available.

The major difficulties in resection of the proximal humerus are a) obtaining an adequate margin of normal tissue around the tumor because of the proximity of the neurovascular bundle to the bone and b)restoring functional movement of the shoulder joint, particularly when the deltoid and rotator cuff have to be sacrificed. D. Kumar et al. [1] studied 100 patients who had undergone endoprosthetic replacement of the proximal humerus .They assessed survival of patients, salvaged limbs and prostheses. Function was determined in 47 survivors of whom 30 were assessed using the MSTS scoring system and 38 using the TESS questionnaire. Median age of the patients was 34 years and median follow up of surviving patients was 9 years. The mean MSTS score was 79% and the mean TESS score was 72%.The length of resected bone influenced the functional outcome. Abduction of the shoulder was 45 degrees in most patients. Overall survival was 42% at 10 years and of the limb 93%. Using mechanical failure of the prosthesis as the end point the prosthesis survival was 86.5% at 20years. They concluded that endoprosthetic replacement of the proximal humerus is a predictable procedure providing reasonable function of the arm with a low rate of complications at long term follow up.

Mayilvahanan [16] from India used custom made megaprostheses in 57 patients with aggressive benign and malignant tumors of the proximal humerus. The pathology was most commonly osteosarcoma, followed by GCT and chondrosarcoma. They achieved extraarticular and wide resection margins in all primary malignant tumors and narrow margins in benign and metastatic tumors. 6 patients died of disease, 4 developed local recurrences and 43 were disease free at an average follow up of 5.5 years (range 2-14.5years). 5 patients required revision replacements. The most common complications were proximal subluxation and aseptic loosening. They assessed functional outcome as satisfactory in 78% of cases. One informative study by Michiel A J van de

Sande et al. [17] compared the outcome, complications and survival of 38 consecutive proximal humeral reconstructions using allograft- prosthetic composite (n=10), osteoarticular allograft (n=13) and a modular prosthesis (n=14) after a transarticular resection. The mean follow up was 10 years. 28 patients survived their disease (10 died). The endoprosthetic group had the smallest complication rate of 21%(n=1), compared to 40% (n=4) in the allograft - prosthetic composite and 62% (n=8) in the osteoarticular allograft group. Only one revision was performed in the endoprosthetic group for a case of shoulder instability. Infection after revision (n=3), pseudoarthrosis (n=2), fracture of allograft (n=3) and shoulder instability (n=4) were the major complications of allografts in general. Kaplan - Meier analysis showed a significantly better implant survival for the endoprosthetic group (p=0.002). At the final follow up the MSTS scores were an average of 72% for the allograft - prosthetic composite (n=7, median follow up 17 years), 76% for the osteoarticular allograft (n=3, 19 years) and 77% for the endoprosthetic reconstruction (n=10, 5 years). An endoprosthetic reconstruction after transarticular proximal humerus resection resulted in the lowest complication rate, higher implant survival and comparable functional results when compared to allograft- prosthetic composite and osteoarticular allograft use. Another systematic review of outcome after reconstruction of the proximal humerus for tumor by Jeun Jeunis MD et al. [18] tried to address the best reconstructive option after proximal humerus resection. They searched the literature from the Medline, Embase and the Cochrane library databases and selected studies reporting results in English, Dutch or German and with follow ups of 80% or more of the patients at a minimum of 2 years were included. They selected 29 studies with a total of 693 patients. They recognised significant limitations in the study methodology due to in homogeneity of the selected studies but however concluded that allograft-prosthetic composites and prostheses seem to have similar functional outcome and survival rates and both seem to avoid fractures that are observed with osteoarticular grafts. They finally stated that further collaboration using RCTs is required to establish the superiority of any particular treatment.

Several authors [19] believe that a proximal humeral endoprosthesis following an intraarticular, deltoid and axillllary nerve sparing resection provides a durable reconstruction with a relatively low complication rate. Although it provides a stable platform for elbow & hand function, actual shoulder function is limited. P. Raiss et al. [20] have studied replacement of proximal humerus in 39 patients using a MUTARS endoprosthesis .Survival rate of the limb was 90% at 11.5 years. The mean MSTS score was 19. Patients with a complete rotator cuff repair showed a better range of motion compared to partial or no repair. There were no signs of implant loosening on post op X-rays. At 11.5 Years implant survival without revision was 70%.

Kasab M et al. [21] performed 29 shoulder reconstruction for proximal tumors of the humerus for neoplasms and followed up for 7 years. Tumors were 20 chondrosarcomas, 5 osteosarcomass, 2 ewings sarcomas and one malignant hemangiopericytoma. In 17 patients abductors were preserved and in 12 patients they were resected.15 had massive humerus prostheses, 3 had composite allograft prosthesis and 3 had inverted prosthesis placed. Scapulo-humeral arthrodesis was performed in 3. Mean follow up was 7 years, the mean MSTS functional score was 88% for inverted prosthesis, 76% for composite allograft prosthesis, 72.6% for massive prosthesis and 75% for scapulohumeral arthrodesis .The most frequent complications were glenohumeral instability in 11 patients. 65% patients were alive and disease free after a mean of 83.5 months. He advocated the following algorithm for proximal humeral resection. In intraarticular resections if rotator cuff and/or deltoid (or axillary nerve) are removed there are 2 options. Scapulohumeral arthrodesis or humeral prosthesis, for patients who do not want a complex therapy with a long post operative period. If rotator cuff and/or deltoid are preserved, reconstruction with a allograft composite (normal/inverted) prosthesis with reconstruction of rotator cuff was ideal. He personally preferred the inverted allograft composite prosthesis. If the deltoid is preserved but not the rotator cuff, allograft composite inverted prosthesis is preferred.

Janos kiss MD et al. [22] performed 91 shoulder resections for tumors in 90 patients. Mean age was 34 years in patients with primary tumors and 61 years in those with metastases. 56 were proximal humeral resection with Tikhoff-Linberg procedures. Prosthesis implantation was done in 41 cases. Autologous fibula was used in 19 cases and massive homologous bone grafting was in 4 patients. 37 patients were reviewed, with a mean follow up of 4.7 years (range 1-20 years). The best results were achieved after humeral resection reconstructed with fibula, when the rotator cuff was preserved. After humeral resection with implantation of endoprosthesis function of the shoulder remained moderate because the rotator cuff was damaged. Overall satisfaction was generally good after all types of shoulder resection as a result of pain relief, preserved hand function and improvement of psychological states. The patients could compensate

extremely well by using the preserved joints and the contralateralupper limb, and he concluded that therefore patient satisfaction does not depend on shoulder function alone. In recent reports endoprosthetic reconstruction of the proximal humerus has been considered a reliable method. However, instability of the shoulder joint caused by loss of the rotator cuff and deltoid muscle function is often seen after such an endoprosthetic reconstruction.

Taketsugu Fujibuchi et al. [23] have described endoprosthesis suspension with polypropylene monofilament knitted mesh. Here the endoprosthesis is suspended from the bone structure and was used in 9 patients. Subluxation of shoulder joint sometimes occurs in the traditional suspension method because of elongation of attached soft tissue. Here as the attachment is to bone there is no leeway for subluxation to occur. In the above 9 patients there was no subluxation, mean shoulder flexion was 35degrees and 65degrees and abduction was 40degrees and 40degrees for soft tissue attachment groups and bone attachment groups respectively .

An interesting study by Wittig, James C MD [24] analyzed long term outcome associated with limb spearing surgery for osteosarcoma of the proximal humorous. Out of 23 patients with osteosarcoma of the proximal humerus there was one IIA lesion,18 stage IIB lesions and four stage III lesions. Twenty two patients underwent extraarticular resection that included the deltoid and rotator cuff and one patient had an intraarticular resection sparing the shoulder abduction muscles. In all patients, the proximal humorous was reconstructed by a cemented endoprothessis that was stabilized by a static technique (Dacron tapes) and dynamically by muscle transfer. At 10 year median follow up 65% (15/23 patients) were alive without evidence of disease .There were no local recurrences. The prothesis survival was 100%. The MSTS upper limb score ranged from 80-90% All shoulders were stable & pain free. Elbow & hand function was preserved in all cases. The most common complication was a transient neuropraxia (n=8). They concluded that en bloc exraarticular resection and endoprothestic reconstruction was a safe and reliable procedure for high grade extracompartmental osteosarcoma of the proximal humerus.

Another study evaluating allograft–prosthesis composite reconstruction of proximal humerus by Ayesha Abdeen MD et al. [25] used an allograft-prosthesis composite to reconstruct the defect following tumor resection in 36 consecutive patients at their institution over a sixteen year period. Reconstruction was performed at the time of resection in 30 cases and for failure of a previous reconstruction in 5 cases and for local recurrence in 1 patient .Mean duration of follow up was five years. One patient had a glenohumeral dislocation. There were 3 failures due to progressive prosthesis loosening that necessitated removal of the prosthesis. Survival of the prosthesis (estimated) with revision as end point was 88% at 10 years. Four patients required an additional bone grafting procedure to treat a delayed union of the osteosynthesis site. Deltoid resection(partial or complete)resulted in a reduced postoperative range of motion but had no effect on MSTS scores. Extra articular resections were associated with lower MSTS scores. All patients had either mild or no pain and normal hand function at final follow up. They concluded that the allograft–prosthesis composite is a durable procedure associated with an acceptable complication rate. Deltoid preservation & intraarticular resection are associated with a greater range of shoulder motion and a superior functional outcome respectively.

Conclusion

A critical and detailed analysis of the literature leads one to conclude that over the last few decades in spite of many patients being successfully treated for tumors of the proximal humerus and in spite of numerus descriptive studies there is a dearth of high quality evidence to point to regarding reconstruction. The main conclusions seem to be that osteoarticular grafts should be abandoned due to numerous complications and need for revisions and that there is no clear consensus regarding which of the remaining two methods of reconstruction should be routinely used. We support the current trend of using an endoprosthesis routinely for proximal humeral reconstruction in view of its simplicity of fixation, durability and excellent long term outcome and reserve autologous fibula (plain or vascularised) in situations where a prosthesis cannot be used. Future directions need to be in the design of better prostheses and conduction of well powered and well designed RCTs by large institutions in collaboration to establish clear evidence based guidelines.

References

1. D. kumar, R.J. Grimer, A. Abudu, S.R. Carter, R.M. Tillman. Endoprosthetic replacement of the proximal humerus _ The Bone & Joint Journal. J Bone Joint Surg [Br] 2003;85-B:717-22.

2. Malawer MM, Chou LB. Prosthetic survival and clinical results with use of large-segment replacements in the treatment of high grade bone sarcomas. J Bone Joint Surg [Am] 1995;77-A:1154-66.
3. Simon MA, Aschliman MA, Thomas N, Mankin HJ. Limb-salvage treatment versus amputation for osteosarcoma of distal end of femur. J Bone Joint Surg[Am] 1986;68-A:1331-7.
4. Springfield DS, Schimdt R, Graham-Pole J, Marcus RB Jr, Spanier SS, Enneking WF. Surgical Treatment of Osteosarcoma. J Bone Joint Surg (Am) 1988; 70A: 1124-30.
5. Simon MA. Limb salvage for osteosarcoma. J Bone Joint Surg [Am]1988;70-A:307-10.
6. Ross AC, Wilson JN, Scales JT. Endoprosthetic replacement of the proximal humerus.J Bone Joint Surg [Br]1987;69-A:656-61.
7. Olsson E, Andersson D, Brostrom LA, Wallenstein R, Nilsonne U. Shoulder function after prosthetic replacement of proximal humerus. Ann Chir Gynaecol 1990;79:157-60.
8. Kochialkowski Wallace WA, Harvey L. Fate of frozen intercalary allograft at one year after implantation with adjuvant chemotherapy treatment:a case report .Clin Orthop 1991;272:146-51.
9. Gebhardt MC, Roth YF, Mankin HJ. Osteoarticular allografts for reconstruction in the proximal part of the humerus after excision of a musculoskeletal tumour. J Bone Joint Surg[Am]1990;72-A:334-45.
10. Mankin HJ, Gebhardt MC, Jennings LC, Springfield DS, Tomford WW. Long term results of allograft replacement in the management of bone tumours.Clin Orthop 1996;324:86-97.
11. Rodl RW, Gosheger G, Gebert C,et al. Reconstruction of the proximal humerus after wide resection of tumours. J Bone Joint Surg[Br]2002;84-B:1004-8.
12. Jensen KL, Johnston JO. Proximal humeral reconstruction after excision of a primary sarcoma. Clin Orthop 1995;311:164-75.
13. Damron TA, Rock MG, O'Connor MI , et al. Functional laboratory assessment after oncologic shoulder joint resections. Clin Orthop 1998;348:124-34.
14. Frassica JF, Sim FH, Chao EY. Primary malignant bone tumours of the shoulder girdle surgical technique of resction and reconstruction. Am Surg 1987;53:264-69.
15. Fuhrmann RA,Roth A,Venbrocks RA. Salvage of the upper extremity in cases of tumorous destruction of proximal humerus.J Cancer Res Clin Oncol 2000;126:337-44.
16. Mayilvahanan,N.,Paraskumar,A.Sivaseelam,A.et al. Custom mega-prosthetic replacement for proximal humeral tumours. International Orthopedics 2006;30-3:158-62.
17. Van de Sande,M.A.J.,Dijkstra,P.D.S. &Taminiau,A.H.M. International Orthopaedics(SICOT)(2011)35:1375.
18. Teun Tenis, Sjoerd P.F T Nota, Francis J Hornicek, Joseph H Schwab, Santiago A. Lozano-Calderon. Clin Orthop Relat Res 2014;472:2245-53.
19. Christopher P. Cannon, Giovanni U. Paraliticci, Patrick P. Lin, Valerae O. Lewis, Alan W Yasco. Functional outcome following endoprosthetic reconstruction of the proximal humerusJournal of Shoulder and Elbow Surgery 2009;18(5):705-10.
20. Raiss P, Kinkel S, Sauter U, Bruckner T, Lehner B. Replacement of the proximal humerus with MUTARS tumor endoprostheses Eur J Surg Oncol. 2010 Apr;36(4):371-7.
21. Kassab M1, Dumaine V, Babinet A, Ouaknine M, Tomeno B, Anract P. Twenty nine shoulder reconstructions after resection of the proximal humerus for neoplasm with mean 7-year follow-up. Rev Chir Orthop Reparatrice Appar Mot. 2005 Feb;91(1):15-23.
22. Kiss J, Sztrinkai G, Antal I, Kiss J, Szendroi M. Functional results and quality of life after shoulder girdle resections in musculoskeletal tumors.J Shoulder Elbow Surg. 2007 May-Jun;16(3):273-9.
23. Fujibuchi T, Matsumoto S, Shimoji T, Ae K, Tanizawa T, Gokita T, Hayakawa K.J. New endoprosthesis suspension method with polypropylene monofilament knitted mesh after resection of bone tumors in proximal humerus. Shoulder Elbow Surg. 2015 Jun;24(6):882-8.
24. Wittig JC, Bickels J, Kellar-Graney KL, Kim FH, Malawer MM. Osteosarcoma of the proximal humerus: long-term results with limb-sparing surgery. Clin Orthop Relat Res. 2002 Apr;(397):156-76.
25. Ayesha Abdeen, MD. Allograft-Prosthesis Composite Reconstruction of the Proximal Part of the Humerus. Functional Outcome and Survivorship. Bone Joint Surg Am, 2009 Oct 01;91(10):240615.