CASE SERIES

Long-term functional outcomes after total scapulectomy with dual suspension reconstruction in children — A case series

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Abstract

Implant reconstruction following scapulectomy in children is a challenging task. Dual suspension reconstruction may offer an alternative but there is a dearth of literature on functional outcomes following this procedure for malignant tumours in children. A retrospective study was conducted at the Aga Khan University Hospital, a tertiary care centre in Karachi, Pakistan. Children with malignant tumours of the scapula who underwent total scapulectomy with dual suspension reconstruction (n=5) between Jan 2009 and June 2015 were included. Mean follow up was 50±13.39 months. There were four boys and one girl having mean age of 11±3.57 years. All patients were Enneking Stage IIB with 4 patients diagnosed as Ewing's Sarcoma and 1 as osteosarcoma. The MSTS scores ranged from 20-25 points, with a median of 23. One patient developed postoperative surgical site infection requiring surgical debridement whereas all patients remained disease-free till last follow up. Our findings suggest that scapulectomy with dual suspension reconstruction achieves satisfactory functional results with low rate of complications.

Keywords: Scapula, Retrospective study, Surgical wound infection, Ewing's sarcoma, Osteosarcoma.

Introduction

The shoulder girdle is a common site for musculoskeletal tumours with Ewing's sarcoma being the most common tumour of the scapula in children. The prevalence of Ewing's Sarcoma is 2 per hundred thousand which makes it as the 2nd most primary tumour after osteosarcoma.[1,2]

Forequarter amputation was considered to be the treatment of choice for shoulder girdle tumours before the 1970's, owing to the fear of local recurrence[3] and the close proximity of these tumours to the neurovascular bundle resulting in poor functional outcomes with local resection. However as amputation is associated with considerable morbidity, the quest for a limb salvage option was not abandoned. Boris Linberg presented the first case series of limb sparing resection for malignant tumours of the upper extremity in 1928.[4] But it was not until after 1970 owing to advances in pre-operative imaging and surgical techniques, that total scapulectomy gained acceptance as the procedure of choice for these tumours where possible. This offered an attractive alternative to amputation in terms of being cosmetically, functionally and emotionally accepted by the patients.[5]

As total scapulectomy involves resection of the glenoid along with the rotator cuff muscles, it has to be accompanied by soft tissue reconstruction to minimize the functional deficit. Rebuilding of shoulder function after total scapulectomy is puzzling. Extracorporeal irradiation and re-implantation is a well-known method of biological reconstruction in orthopaedic oncology; and few reports in the literature describe its specific use in tumours of the scapula.[6,7] Various reconstruction techniques for the gleno-humeral articulation have been reported in literature, ranging from metallic prosthesis to allograft reconstruction to simple proximal humeral suspension.[8]

There is a dearth of literature on functional and oncologic outcomes of scapulectomy for malignant tumours as a whole and specifically for children. Hence we decided to study and report our experience with total scapulectomies in paediatric patients.

Case Series

A single institution retrospective study was conducted at Aga Khan University Hospital, Karachi. This case series included all patients operated between January 2009 and June 2015. Patients with minimum follow up of 2 years were included in the study. Study was started after being approved for exemption from Ethics review committee of Aga Khan Hospital. Patients consent was taken for publishing their data.

US Department of Health and the Food and Drug Administration Criteria for age definition was used to include all patients falling under 18 years of age as

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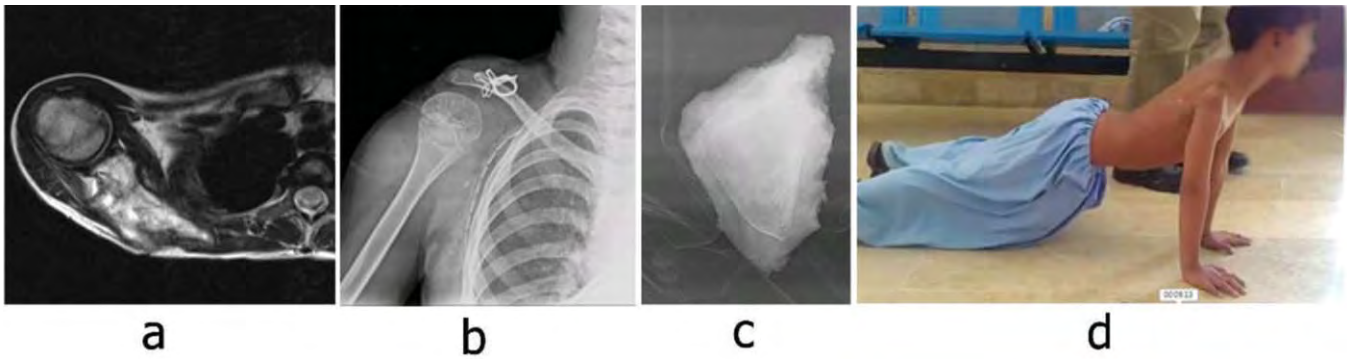

Figure: a) MRI image of patient with Ewing sarcoma before surgery. b) Post-surgery follow up X-ray of patient showing dual suspension. c) Specimen radiograph. d) Patient showing functionality at follow up.

paediatric patients.[9] A total of 23 children with malignant tumours of the scapula were treated at our institution during this time. Of those 5 underwent total scapulectomy and the other patients were excluded as they either had gross neurovascular invasion, or involvement of the chest wall leading to a more radical excision or had been deemed inoperable. Surgical staging was done on the principles of Enneking system[10] with plain radiographs and CT/ MRI scans.

These five patients included four boys and one girl. Their mean age was 11±3.57 years (range: 8-16 years). Four patients had Ewings Sarcoma in their scapulae, whereas one had osteosarcoma. All patients were Enneking Stage IIB and all underwent total scapulectomy with dual suspension Reconstruction. The patients mean follow up was 50±13.39 months (range 33-65 months).

Indication for surgery included a malignant tumour localised in the scapula without local invasion into the axilla or chest wall nor any distant metastases. The surgical technique used for total scapulectomy on our paediatric patients was based on Malawer's description of shoulder girdle resections and total scapulectomy (intra-articular scapular resection, type III)[6] which was employed in all cases. Two limbs of the utilitarian incision are utilised with the anterior incision being used to mobilize the axillary vessels and nerves and the posterior incision for exposure of the scapula, rhomboids, latissimus dorsi, and teres muscles. All muscles are transected from the bone starting from the lowest point inferiorly including the rotator cuff. Soft tissue reconstruction for shoulder stability is done using dual-suspension technique with Dacron tape from the clavicle for static support and reattaching the biceps and triceps muscles through drill holes. Tenodesing the deltoid to the pectoralis major and trapezius muscles further enhances the stability.[6] Radiographs and follow up pictures of one of the cases is presented in Figure (a, b, c, d).

Patients were provided a sling postoperatively, and motion was restricted until the incision healed. The sutures were removed at about 2 weeks after surgery. Active motion of the elbow and hand was initiated to preserve strength and range of motion and to help minimize oedema. At about 2 weeks, the sling was removed for passive shoulder range of motion (ROM) and pronation and supination at the wrist. Passive ROM of the shoulder (flexion, abduction, and external and internal rotation and pendulum exercise) with the help of a family member or physical therapist was encouraged. Active ROM was started after 6 weeks. All patients were followed up as a routine in the clinic by the chief surgeon at 1 month, 3 months and every 3 months for at least 2 years then annually thereafter.

Musculoskeletal Tumour Society (MSTS score) was calculated by the operating surgeon as a routine during follow up clinic visits to assess the functional outcome.

Table-1: Patient demographics, diagnoses, functional outcome and complications.

S.No	Age	Sex	Tumour	Follow Up (Months)	Complications	MSTS Score	Oncologic Outcome
1	8	Male	Ewing's Sarcoma	52	Wound Infection	20	Disease free
2	8	Male	Ewing's Sarcoma	65	None	22	Disease free
3	11	Male	Ewing's Sarcoma	33	None	25	Disease free
4	12	Female	Ewing's Sarcoma	60	None	23	Disease free
5	16	Male	Osteosarcoma	40	None	23	Disease free

This was subsequently extracted from the patients' records. Similarly complications and their subsequent management was also extracted from medical records. Descriptive analysis was performed on IBM SPSS analytical software, version 20.

All children had an acceptable functional outcome with restrictions mainly in overhead abduction and lifting ability. Median MSTS score was 23 (Range 20-25). One (20%) patient developed post-operative infection which required surgical debridement and a course of antibiotics after which it resolved. There was no local recurrence nor any distant metastases in any of the patients. All patients were followed up till December 2019. Table shows over all patient demographics, diagnoses, functional outcome and complications.

Discussion

There is a dearth of literature on scapulectomies due to the rare occurrence of malignant tumours at this site. Most studies available do not segregate adult and paediatric patients while reporting survival and functional outcomes. As the paediatric population is different with regards to the aetiology of tumours and remaining growth potential their outcomes need to be studied separately. Very few studies have addressed the paediatric population separately in terms of reconstruction options and functional outcomes.

Our study focussed on the long term follow up of children < 18 years so that a better assessment of survival and functional outcomes of this population may be made. Our mean follow up was 50±13.39 months which corresponded to skeletal maturity in most of our patients. Schmalzl J, et al. reported that shoulder function in their patients improved more at 5 to 8 years of follow up and the patients were disease free as well.[1] Although scapula is an uncommon site for Ewing's sarcoma, 80% of our cases had Ewing's sarcoma.[11] Multiple reconstructions techniques after total scapulectomy have been described in literature ranging from no reconstruction, soft tissue reconstruction to prosthetic reconstruction. No reconstruction leads to poor shoulder function and is not a recommended option. Total Scapulectomies in paediatric patients poses a unique challenge in soft tissue reconstruction owing to the remaining growth potential in this population. A prosthetic reconstruction is demanding because the remaining growth of the patient and estimated early wear of the implant limit its utility and survival.

Reconstruction techniques successfully used in paediatric patients include humeral suspension or irradiated bone/ allograft. Ahmed M. et al. used irradiated bone and reported the mean MSTS scores as 87% and 8 out of 10 patients in his study were able to achieve shoulder elevation above 90 degrees in scapular and forward plane. Over the follow up time, all patients function had improved. In his study complications were also reported, wound gaping occurred in 2 patients, 3 had dislocation of acromioclavicular joint while 50% patients had resorption of scapular graft.[7]

In our study we used humeral suspension with the mean MSTS scores as 23 and all children had good functional outcomes with more improvement seen in our long term follow up. In earlier months of post-surgery, patients had only restriction issues of overhead abduction. Out of our 5 patients, only 1 had complication of wound infection requiring wound debridement and antibiotic treatment. All of our patients were disease free till the year 2019 follow up. None of them reported for distant Metastasis.

We acknowledge limitations of this retrospective study. First, the number of patients is small as with many orthopaedic oncology studies and heterogeneity of the patient population with no control group making any comparison difficult. But our follow up time was sufficient to report long term outcomes with regards to disease recurrence, complications and functional outcomes.

Conclusion

Our findings suggest that scapulectomy with dual suspension reconstruction achieves satisfactory functional results with low rate of complications. Multicentre studies need to endorse these long term findings in comparison with other reconstruction options.

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Conflict of Interest: None.

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