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Reconstruction of Upper Limb Long Bone Defects with Vascularized Free Fibula in Pediatric Population: A Review of the Functional Outcomes

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ABSTRACT

Objective: The aim of our study was to evaluate the functional outcomes in paediatric patients who underwent upper limb long bone reconstruction with vascularised free fibula.

Study Design: Retrospective descriptive study.

Place and Duration of study: Department of Plastic Surgery, Shifa international Hospital, Islamabad Pakistan from Jun 2015 to Jul 2020.

Methodology: All paediatric patients who underwent upper limb long bone defect reconstruction secondary to sarcoma resection were included in the study. Radiological and functional assessment was done pre and post operatively. Outcomes were assessed in terms of surgical wound dehiscence, flap loss, donor site morbidities and functional outcomes. Functional outcomes were evaluated using Musculoskeletal Tumor Scores for upper limb and pediatric Toronto Extremity Salvage Score for upper limb.

Results: Total of 14 paediatric patients were operated during 5-year period of June 2015-July 2020. Age of the patients ranges from 9 - 15(12.5) years. All the limbs were successfully salvaged with no flap loss and no significant donor site morbidity. Functional latissimus dorsi flap was done in 6 patients to achieve elbow flexion. Overall musculoskeletal tumor society Score was 82% and pediatric toronto exeteremity salvage score mean was 82.43%.

Conclusion: Vascularised free fibula is a reliable and cost-effective option for bridging bone gaps in paediatric patients after sarcoma resection with optimal functional outcomes.

Keywords: Upper Limb Long Bone, Pediatric Toronto Extremity Salvage Score (pTESS), Vascularized Free Fibula.

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INTRODUCTION

Salvaging the upper limb long bone defects in pediatric population poses a challenge to the reconstructive surgeon, be it after tumor resection or other causes. Increased survival in the past few decades secondary to better imaging modalities and multimodal chemotherapy, radiotherapy and immunotherapy has shifted the paradigm of surgical management for sarcoma, from amputation towards salvage with reconstruction of the defect.[1] The primary principle of tumour resection is to achieve oncologically recommended disease free margins histologically,[2] in achieving which the gap between two bony ends could be a challenge to bridge, not just to restore skeletal continuity but achieving optimal function and provide soft tissue coverage for the hardware. This challenge has been revolutionized with the use of vascularized free fibula as the diaphyseal bone replacement.[3] In addition, owing to its long pedicle, predictable vascular anatomy, long and cylindrical shape, fibula has been the gold standard in bridging the long bone gap in tumor resection or trauma. It has potential to hypertrophy and remodel over time in accordance to workload, more so in children and is also able to withstand the effects of radiotherapy.[3-5]

Free vascularized fibula has been in the use for over last 4 decades now since Taylor first described in 1973.[6] In our study we aim to review the functional outcomes after salvage of the upper limb long bone defects secondary to tumour resection in pediatric population. We achieved the soft tissue coverage in some cases using latissimus dorsi pedicled flap, which can also be used as a functional muscle transfer to achieve better outcomes.[2]

Objective was to review the functional outcomes of using vascularized free fibula in peadiatric population for reconstruction of upper limb long bone defects.

METHODOLGY

This was retrospective study of pediatric patients who underwent upper limb reconstruction between 1st

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June 2015 till 30th June 2020. Sample size we calculated using sample size calculator, keeping confidence level 95%, margin of error 5% and population proportion 50%. Total population size was 14 and calculator gave a sample size of 14 under these considerations.

Inclusion criteria: All pediatric patients with upper limb long bone defects of more than 6 cm were included.

Exclusion criteria: Patients with metastatic disease and age over 15 years were excluded.

Study was approved by the Ethical Committee and Institutional Review Board at the hospital vide (IRB# 486-21).

Data was extracted from the electronic medical record. Free fibula was used for reconstruction of the defect in all the patients. All defects were reconstructed by a single surgeon. Radiological and functional assessment was done at 6 and 12 month follow-ups. For each patient age, gender, cause of the defect, location of the tumour, type of tumour, size of the defect and length of fibula harvested, associated soft tissue defect and need for functional muscles transfer and survival were collected. Functional outcomes were assessed using pTESS and MSTS scoring for upper limb. SPSS version 26 was used for the numerical calculation. Mean and standard deviation was calculated for age and median and IQR was calculated for the functional outcome scores. Mean and percentages of the functional outcome scores have also been calculated to ease the comparison with other studies.

All the sarcoma cases were discussed in multidisciplinary team meeting pre-operatively. Wide local excision was done by the orthopedic team under frozen section control. Defect was analyzed and free fibula was harvested based on peroneal vessels by the standard procedure. According to the defect dimensions and reconstructive requirement, pedicled latissimus dorsi flap was harvested from ipsilateral side using standard technique. After flap dissection heparin was given according to weight of the patient and pedicle divided and handed over to orthopedic team. Inset of the fibula was done using dynamic compression plates spanning the entire length of the flap and secured distally and proximally with screws. After the osseus flap inset, anastomosis was done and latissimus dorsi flap was inset accordingly (as functional muscle or soft tissue coverage of the hardware).

Figure-1 summarizes the journey of a 14year old male patient, diagnosed case of chondroblastic osteosarcoma of the left proximal humerus. He underwent neoadjuvant chemotherapy. His MRI showed an expansile lesion with soft tissue compeont involving the metaphysis of the left humerus. His wide local excision was done by orthopedic team and created a bony defect of 17cm along with excision of rotator cuff muscles. Bony defect was bridged using a vascularized free fibula and latissimus dorsi was used for soft tissue coverage of the hardware. His 2-year follow up X-ray showed good bony union and good function of 90-degree abduction was achieved. Overhead abduction was not possible due to shoulder joint arthrodesis.

Figure-1: A 14-year old male patient, diagnosed with left proximal humerus osteosarcoma, A: Pre-operative, B: MRI scan showing tumor in proximal humerus. C: Specimen after excision. D: Inset of vascularized free fibula. E: Soft tissue reconstruction using pedicled Latissimus Dorsi flap. F: Post-operative X-ray showing good bony union. G,H: Function after 2 months and 1 year respectiverly

Figure-2 shows a 12 years old male patient diagnosed with left proximal humerus Ewing Sarcoma. He underwent 10 cycles of neoadjuvant chemotherapy. His scans showed a large lesion involving the proximal humerus and extending beyond the bone into the soft tissue. His wide local excision was performed by the orthopedic team and a defect of 16cm was created along with the soft tissue surrounding it. His neurovascular bundle was spared. His bony reconstruction was done using vascularized free fibula and latissimus dorsi muscle was used to provide hardware coverage and to compensate for the functional loss. His shoulder was arthrodhesed. Patient achieved good functional outcome of 90-degree abduction as evident in 6 month and 5 year follow up.

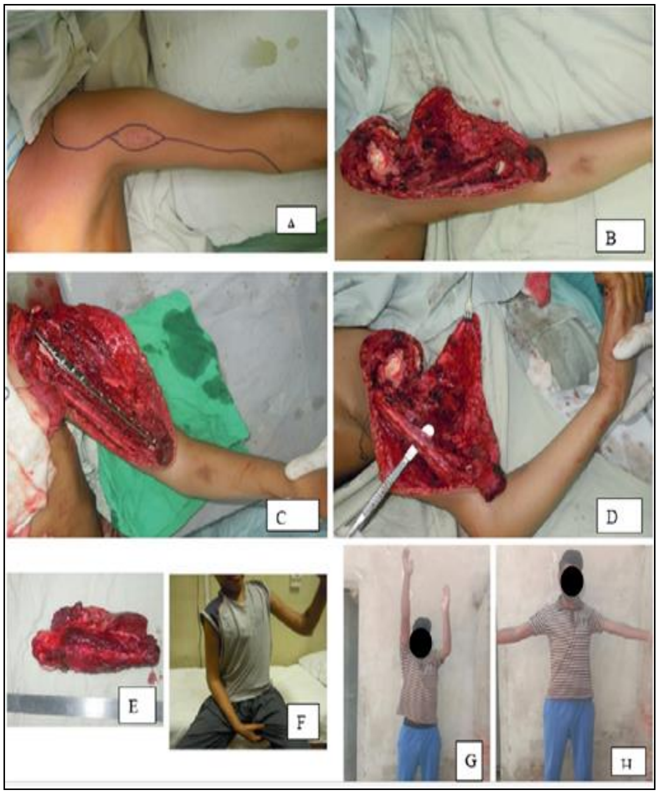

Figure-2: A 12-year old male patient diagnosed with left proximal humerus Ewing Sarcoma, A: Pre-operative marking including site of biopsy in excision. B: Defect after excision of tumor along with head of humerus. C: Inset of vascularized free fibula. D: Intact neurovascular bundle and Latissimus Dorsi flap for soft tissue reconstruction. E: Tumor specimen. F: Functional outcome 6 months post reconstruction. G,H: functional outcome 5 years post reconstruction

Figure-3 shows a 15-year old female patient diagnosed with synovial sarcoma of left forearm. Pre operative MRI scan showed the involvement of distal radius and ulna along with extensor and flexor compartments. Radial nerve and artery were also involved. Her wide local excision created a bony defect of 10cm along with involved muscles and radial artery and nerve. Bony reconstruction of radius was done using vascularised free fibula and pedicled latissimus dorsi was used to regain lost flexor compartment (flexor digitorum profundus and flexor pollicis longus). Defect of ulna was bridged with IM nail. Post operative functional outcomes showed opposition, abduction and adduction of fingers along with good achievement of finger and thumb flexion.

Figure-3: A 15-year old female patient diagnosed with Ewing sarcoma of left forearm, A: Pre-operative MRI scan. B: Swelling on the forearm. C: Per-operative specimen involving left forearm sarcoma along with segments of radius, ulna and muscles. D: Defect post excision. E: Inset of vascularized free fibula. F: Closure after pedicled functional myocutanous Latissimus Dorsi flap inset. G,H,I: Post operative functional outcomes showing opposition, abduction and adduction of fingers

RESULTS

Upper limb long bone reconstruction was done in total 14 patients with the age of patients ranging from 9-15 years (12.5±1.9 years). Male to female ratio was 4:3. Summary of the etiology of the defect is provided in Table-I. Osteosarcoma was the most common etiology. All of the patients had immediate reconstruction using vascularized free fibula. Average length of the defect was 13.5±4 cm. Latissimus dorsi was used as functional muscle or for soft tissue coverage in 6 patients. Tendon transfers were done in 2 patients.

Table-I: Summary of the Etiology (n=14)

Region	Osteosarcoma	Chondrosarcoma	Ewing sarcoma	Synovial sarcoma
Radioulna	2	0	0	1
Humerus	5	3	2	1
Wrist	0	0	0	0

Table-II summarizes the functional outcomes using MSTS scoring. Median of the MSTS was 24.5 with IQR of 2. Mean score was 82%.

Table-II: Summary of MST Scoring

Patients	Score
1	28
2	24
3	24
4	25
5	25
6	24
7	23
8	25
9	26
10	24
11	21
12	26
13	27
14	24
Median(IQR)	24.5(2)

Table-III summarizes the scoring based on Toronto extremity salvage score for pediatric patients in upper limb. Median of the pTESS score was 115.5 with IQR of 13.25. Mean score 82.48%.

Table-III: Summary of pTESS Scores

Patients	Score
1	124
2	105
3	118
4	117
5	111
6	104
7	97
8	116
9	115
10	104
11	96
12	116
13	120
14	116
Median (IQR)	115.5 (13.25)

One patient (14%) had donor site seroma and 1 patient (14%) had wound dehiscence at the recipient site. There was no microvascular failure in any of the patients. None of our patients had any neurological injury, fracture at the donor or recipient site and bony union was achieved in all patients. Four patients (28.6%) died from metastatic disease in the following years and 1(7.1%) had an accidental death.

DISCUSSION

The survival of patients with malignant bone tumors has drastically improved at 5 years, in the last 30 years, from 10 to 20% to 60% presently, due to the advancement in chemotherapy.[7] Before the development of effective chemo therapeutic agents, patients developed metastasis even after gaining surgical control of the disease. Neoadjuvant tumor specific regime chemotherapy has revolutionized the survival outcomes. All patients in our study had received preoperative chemotherapy.

As survival benefit increased, so did the focus on functionality of reconstruction. Limb salvage surgery became the mainstay treatment. Current options for limb salvage include, cortical allografts, endo-prostheses, distraction osteogenesis, and vascularized fibular graft.[8-12] In our experience microvascular transfer of fibula for long bone reconstruction has been ideal. There was no flap loss reported in our patients. This compares well with flap loss rates up to 15% in a similar study.[13] Fibula is an ideal long bone for bony reconstruction due to its reliable pedicle and relative ease of harvest.[14,15] The rate of bony union is high and functional outcome is also satisfactory. We measured functional outcome in terms of pTESS and MST Scoring performas for upper limb. Limb salvage was achieved in 14 patients with vascularized fibula, all with a minimum follow up of 1 year. In our study, we managed to salvage limb in all the patients, which is high when compared to a study done by Campanacci *et al.*,[25]

A wide majority of patients went on to achieve excellent functional recovery as judged by the scoring systems which corresponds with functional outcome scores identified in our review of the literature.[17-20] All patients were able to perform daily activities, with the large majority reporting pain-free movement without the use of assistive devices. Overall mean MST score was 80.71% for our study. Five patients who underwent excision of glenohumeral joint, for them MSTS were lower and they had restriction in shoulder abduction. These when compared with another study,[21] follow the same pattern of limited mobility after humeral head reconstructions.

The range of MSTS for all patients was from 73.3% - 93.3%. This compares favourably with another study in which average scores were 77 - 100% for use of vascularized epiphyseal transfer with massive bone allograft in children for proximal humeral reconstruction.[22] Outcomes using Capanna technique for bony reconstruction also follows the same pattern for MSTS (67-100%).

In patients (85.7%) with extensive tumors, which were involving the anterior muscular compartment, we employed latissimus dorsi muscle flap for either soft tissue bulk or functional transfer for elbow flexion in conjunction with fibular graft. Complication rate for these cases were also low. All patients with preserved shoulder joint achieved acceptable shoulder abduction of about 60-80%. Patients easily carried out daily activities.

Along with limb salvage and good functional outcome, there were few minor complications as well. One patient (14%) had donor site seroma and 1 patient (14%) had wound dehiscence at the recipient site, both these were managed in outdoor clinic basis. There was no microvascular failure in any of the patients. McCullough *et al.*,[21] reports his complication rate to be 44.8%, however these were appointed to be late in nature with one patient having neurological injury. None of our patients had any neurological injury, pertaining to dissection around the peroneal nerves. Four patients (28.6%) died from metastatic disease in the following years and 1(7.1%) had an accidental death. This is far lower complication rate than other studies.

Postoperative fracture in graft is the most common complication, as determined by another study.[23] However, no fracture ensued in our patients in the postoperative period. Union of graft occurred in all patients. Erol *et al.*,[24] showed in his study that defect size and length of harvested graft had no impact on union rate (94.4%). These radiological results indicate that when the transferred fibular autograft retains its vascularity, bone union will occur within 12 months, followed by graft hypertrophy, which will proceed up to 24 months.

We attribute our low complication rate due to limited study size. pTESS scores could not be compared due to lack of studies which have utilized this score. However, we found this tool to be useful to assess the ability of patient to achieve function enough to perform daily activities based on a scale of ease to perform those activities.

CONCLUSION

Vascularized fibula has stood the test of time for reconstruction of long bone reconstruction. The functional outcome have been optimum and complication rates low. It has been shown to hypertrophy with time and heals well after stress fractures. In our series we aimed to show the upper limb reconstruction after oncological defects in children using vascularised fibula and LD flap. This has become a gold standard technique for our department. These results are also supported by the previous series showing the effectiveness of biological reconstruction in bone sarcomas.

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Authors' Contribution

Following authors have made substantial contributions to the manuscript as under:

MG & MR: Data acquisition, data analysis, critical review, approval of the final version to be published.

SUR & FS: Study design, data interpretation, drafting the manuscript, critical review, approval of the final version to be published.

SA & AK: Conception, data acquisition, drafting the manuscript, approval of the final version to be published.

Authors agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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