

Original Research Article

Shark bite bone resection in treatment of surface sarcomas

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ABSTRACT

Background: Surface bone sarcomas including parosteal sarcoma as the most common type are considered well differentiated sarcomas with rare intramedullary involvement. They are characterized by low incidence of local recurrence and distant metastasis. As they involve a portion of cortical bone, hemi-cortical resection (shark bite resection) seems to be an appealing option with low morbidity and better function. We aim to evaluate oncological outcome and functional results for a cohort of patients with surfaces sarcomas treated by shark bite resection.

Methods: A prospective study of 25 patients presented with surface malignant bone sarcomas between 2016 and 2022 in our institution. We performed hemicortical resection and biological reconstruction. Follow up period was 2-8 years. Functional outcome was assessed using MSTS scoring system.

Results: The mean MSTS at the end of follow up was 29 (range 27-30). Mean resected bone surface was 9 cm (range 7-14). Mean time for bone union was 8 months (range 6-15 months).

Conclusion: Shark bite resection is a valuable option for treatment of low-grade surface sarcomas. It provides better outcome, native joint preservation and low complication rate.

Keywords: Shark bite, Hemicortical, Resection, Sarcoma, Surface, Parosteal

INTRODUCTION

Most primary malignant bone sarcomas of long bones originate from the metaphyseal region of bone near the joints. This usually necessitates sacrifice of the joint as a part of the wide local resection in limb salvage surgery. These osteoarticular defects require reconstruction with various modalities such as endoprosthetic replacement, allografts or allograft-prosthesis composites. Although endoprosthetic replacement allows immediate weight bearing with adequate long-term results and good survivorship, they are usually complicated with loosening and infection. That is why preservation of the native joint will be the ideal solution for these young patients.^{1,2}

Low grade surface bone sarcomas are quite uncommon entity of tumors that arise from the cortical bone surface. These include parosteal osteosarcoma, adamantinoma and secondary peripheral chondrosarcoma.²

Parosteal osteosarcoma, being the commonest type of surface sarcoma, represents less than 6% of all osteosarcomas. It usually affects distal femur followed by proximal tibia, characterized by low risk of local recurrence, distant metastasis and higher incidence in the third and fourth decade of life and better prognosis than conventional sarcoma.³

Hemicortical resection (also known as shark bite resection) in such type of tumors is a good idea instead of segmental resection maintaining the cortical continuity, bone stock preservation, native joint preservation and without local control compromise.⁴

The objectives were to evaluate the functional result and oncological outcome of shark bite resection of bone of surface bone sarcomas as well as assessment the post-operative complications.

METHODS

Patients

The study has been approved by the ethical committee of our university. Informed consent has been taken from all the patients included in the study regarding treatment and inclusion in any future publication.

This is a retrospective study that included 25 patients presented with surface low grade malignant bone sarcomas in the period between 2016 and 2022 who underwent shark-bite (hemi-cortical) wide tumor excision and biological reconstruction. The study and procedures were performed at El-Hadra University Hospital at Alexandria University.

Inclusion criteria

Inclusion criteria included patients with surface bone sarcomas affecting less than 50% of the cortical circumference and without intramedullary involvement or distant metastasis.

Exclusion criteria

Exclusion criteria patients with advanced disease presenting with intramedullary involvement or affection of more than 50% of the cortical circumference were excluded, patients with distant metastasis or with encroachment on the neurovascular structures were excluded as well.

Tissue biopsy was performed in all cases to confirm pathological diagnosis and grading of the tumor. All patients underwent full staging program using computed tomography (CT) scan of chest, bone scintigraphy and magnetic resonance imaging (MRI) of the affected site. None of the patients had distant metastatic disease or skip lesion.

MRI has confirmed localization and involvement of only a portion of the circumference of long bone without intramedullary involvement which make hemicortical resection possible without compromise of the tumor margin.

Data collection from medical records included age, gender, pathological diagnosis, staging, preoperative neoadjuvant treatment when required.

Operative data included site of involvement, size of defect, and method of reconstruction.

Postoperatively a posterior splint was applied until bony union was revealed radiographically. Partial weight-bearing started when more than 50% of unions of the transverse and longitudinal osteotomies appeared in the radiographic evaluation. All patients underwent follow up sequential imaging, staging (MRI) and CT scan to detect

any local recurrence and confirm bone consolidation and a CT scan of the chest to detect any lung metastasis every four months in the first two years, and every six months thereafter.

Postoperative data included time to the healing of the reconstruction, time to weight bearing, any local or oncological complications encountered.

Functional status included assessment using the MSTs scoring system.⁵

Statistical analysis of the data

Data was fed to the computer and analyzed using IBM statistical package for the social sciences (SPSS) software package version 20.0. (Armonk, NY: IBM Corp). Qualitative data were described using number and percentage. The Shapiro-Wilk test was used to verify the normality of distribution. Quantitative data were described using range (minimum and maximum), mean, and standard deviation, median and interquartile range (IQR). Significance of the results obtained was judged at the 5% level. The tests used were Chi-square test, Fisher's Exact or Monte Carlo correction, Student t-test, Mann Whitney test, analysis of variance (ANOVA) with repeated measures (periods) and F-test (ANOVA) (groups).

Surgical technique

The operation performed under appropriate anesthesia and a pneumatic tourniquet was used on the proximal thigh. The biopsy site was resected in all cases and taken in the incision line to be totally removed, Identification of the neurovascular bundle then ligation of small branches from bundle to the tumor segment was done. Level of bone osteotomy and safety bony margin >1 cm was planned from pre-operative MRI. Osteotomy is carefully performed to avoid fracture of the cortex or violation of the tumor margin. The fixation augmentation with a titanium plate and screws was used with or without recycled pasteurised autograft and \pm non vascularized fibula or soft tissue reconstruction. The gross soft tissues tumour was thoroughly removed from the specimen, and then treated in saline, preheated at 70°C for 60 minutes, and then re-implanted again using fixation with a titanium plate and screws (Figures 1-3).⁵



Figure 1: Preoperative X-ray.



Figure 2: Preoperative MRI.

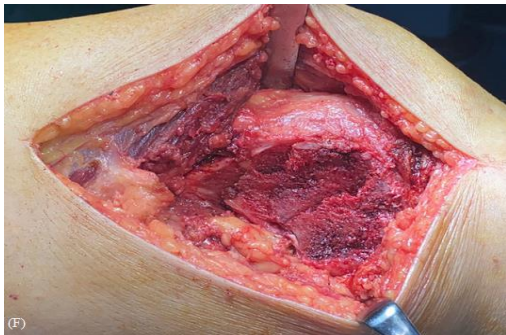


Figure 3: Intraoperative photo.

RESULTS

Patients' ages ranged from a minimum of 19 years old to a maximum of 45 years old with a mean age of 29.20 ± 14.36 years old. Fourteen of the cases were females and eleven were males (Table 1).

Table 1: Distribution of the studied group regarding demographic data (n=25).

Characteristics	N	%	P value
Age (years)			
16-20	1	4	0.736
21-30	10	40	
31-40	12	48	
41-50	2	8	
Sex			
Male	11	44	0.342
Female	14	56	
Site			
Femur	13	52	0.204
Tibia	12	48	

Femur was involved in 13 patients while tibia was involved in 12 patients.

According to pathological diagnosis, parosteal osteosarcoma was involved in 12 patients, chondrosarcoma in 7, and adamantinoma in 6 patients.

Mean resected bony surface was 9 cm (range 7-14 cm).

In our study, wide hemicortical resection of the lesion with tumor free margins was achieved in all patients. None of the patients received neoadjuvant chemotherapy. None of the patients were lost during the follow-up. The mean MSTS functional score for all patients at the end of the last follow up was 28 (range 21-30) (Table 2). The average postoperative follow-up was months 59 ± 16.94 months (range, 30 to 110 months).

Table 2: Distribution of the studied patients regarding the functional score (n=25).

Total MSTS score	N	%	P value
Unsatisfactory	0	0.0	0.641
Poor (<12)	0	0.0	
Fair (12-17)	0	0.0	
Satisfactory	25	100.0	
Good (18-23)	3	10.0	0.336
Excellent (24-30)	22	90.0	
Min-max	21.0-30.0		
Mean±SD	28.22±1.16		

Figures 4-6 shows preoperative X-ray, preoperative MRI and postoperative X-ray. Mean bony union was 8 months (6-12 m).



Figure 4: Preoperative X-ray

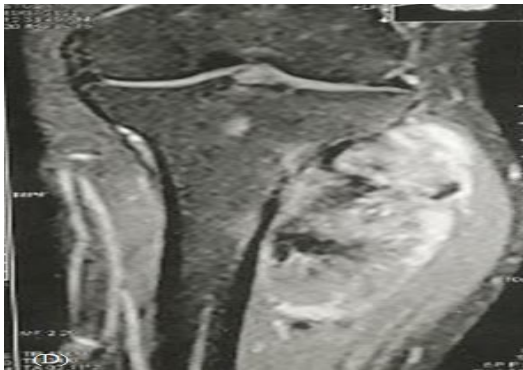


Figure 5: Preoperative MRI.

Only one patient developed early superficial infection in tibial sarcoma that treated with debridement. One patient developed local recurrence after five years in distal femur

parosteal osteosarcoma and managed by wide margin resection and tumor prosthesis.

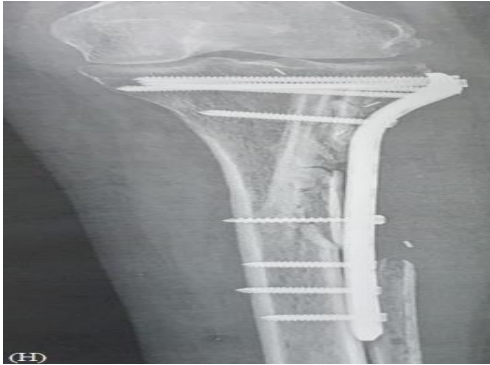


Figure 6: Postoperative X-ray.

DISCUSSION

Low grade surface sarcomas include parosteal osteosarcoma, surface chondrosarcoma and adamantinoma. They are well differentiated low grade tumors with low incidence of intramedullary involvement and/or distant metastasis. The 10-year survival rate is high more than 90%.^{2,5}

Wide local resection of these tumors is curative in most cases with low incidence of local recurrence. Traditionally, wide local resection usually includes the adjacent joint. Reconstruction is followed by either endoprosthetic replacement or osteoarticular allografts.⁶

Endoprosthetic replacement has its drawbacks in the form of high risk of infection, loosening, breakage and the need for revisions given the long-term survival of patients. Osteoarticular allografts are also blamed for high risk of infection, fractures and unpredictable healing.⁷

Hemi-cortical resection (shark bite resection) of these low-grade tumors is an appealing procedure because it allows preservation of the major bone stock, preservation of the native joint with possible better function. And more importantly, allows curative wide resection of these tumors because of the limited affection to the cortex and absence of intramedullary invasion. This technique was first described by Campanacci et al in 1982. Since then, hemicortical resection has become a well-known therapeutic option for low grade surface sarcomas.^{4,8}

Hemicortical resection has shown optimum oncological and functional result with different modes of reconstruction of the bone defect using polymethyl methacrylate (PMMA) or biological materials such as fibular autografts, allografts or pasteurized/irradiated host bones, with or without prophylactic fixation.⁹

The main complications of hemicortical resection include recurrence, fractures and infections.

Local recurrence can be prevented with first, meticulous preoperative planning using advanced imaging to confirm that the tumor is limited to less than 30% of the cortical circumference and to confirm absence of any intramedullary invasion. In our study only one patient with distal femur parosteal osteosarcoma had local recurrence after 60 months and the patient was treated by wide resection and endoprosthetic replacement.

Fracture is a potential complication that may happen in the perioperative period because the corners of osteotomy act as points of high stress. This complication can be avoided by using neutralization plate the bridge the osteotomy. We didn't encounter any case of fracture because we used plating in all patients except one patient with distal femur surface chondrosarcoma limited to a very small portion of the metaphysis which was resected with a very small cortical bone and the remaining distal femur was strong enough that it didn't require fixation.

Infection is a potential complication because of the demanding nature of the procedure. In our series, we had one case of infection that resolved with debridement.

There is paucity in the literature regarding this technique, because of rarity of these tumours and the high selectivity for the patient who are deemed suitable for this kind of limited resection. Our study conforms with results obtained in most of the previous reports.

Our study had minimal complications and good function. The mean MSTS functional score for all patients at the end of the last follow up was 28 (range 21-30).

Funovics et al found that the oncological and functional outcomes following prosthetic replacement and biological reconstruction were similar in terms of the incidence of local recurrence and metastasis, but the incidence of revision was significantly higher in patients with a prosthetic replacement.¹⁰

Deijkers et al reported 22 patients treated by allograft hemicortical reconstruction with promising oncological and functional outcome. All patients had excellent MSTS score, low complication and full graft incorporation within thirty months postoperative. The overall incidence of fracture of the host bone is 27.3%.¹¹

Liu et al reported 13 patients diagnosed with PAO and managed with wide excision and reconstruction with pasteurized hemicortical autograft and internal fixation with mean bony union within 11 months. Average follow up period was around 100 months with mean MSTS 88%. He reported low complication rate with one case of perioperative fracture that required internal fixation.¹²

Agarwal et al studied 10 patients who underwent hemicortical excision of the distal femur and reconstruction with autograft or allograft, the functional

outcome was satisfying with an incidence of fracture of 10% and there was no local recurrence.⁴

Limitations

The limitation in the study included small number of patients and selection bias because only patients with less than 25% involvement of the circumference are considered ideal for this type of resection.

CONCLUSION

Limb salvage surgeries are complex surgeries which need multidisciplinary teamwork. Tumor endoprosthesis is the best option for limb reconstruction following tumor excision sacrificing the joint allowing early mobilization and local recurrence with high rate of revision (infection, loosening). Shark bite (hemicortical) resection offers better outcome, native joint preservation and less complication (revision, infection) rate of prosthesis. The local recurrence and metastatic rate was similar between the biological reconstruction and the mega tumor prosthesis. The results of our study favours hemicortical resection and biological reconstruction and internal fixation to minimize the risk of nonunion or fracture postoperatively.

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Conflict of interest: None declared

Ethical approval: The study was approved by the Institutional Ethics Committee

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