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developing countries.





# Outcomes of internal hemipelvectomy for pelvic tumors: a developing country's prospective

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**Introduction:** Previously, external hemipelvectomy was the mainstay of treatment for pelvic tumors. However, with technological advancements, limb salvage procedures such as internal hemipelvectomy have emerged as a viable alternative. However, there is limited literature available on long-term outcomes and complications of internal hemipelvectomy, especially from developing countries. Therefore, the objective of this study was to share our experience of internal hemipelvectomy at a tertiary care center in a developing country.

Materials and methods: A retrospective review was conducted in which all 24 patients undergoing internal hemipelvectomy from January 1, 2005 to December 31, 2015 at our institution were included. Medical record files were reviewed for intraoperative and early and late postoperative complications, and functional outcomes were assessed by contacting each patient on telephone.

Results: Ewing sarcoma was found to be the most common diagnosis, followed by osteosarcoma as the second most common. The mean follow-up period was 18.7 ± 13.9 months. Intraoperatively there were 4 cases of iatrogenic neurovascular injury and 2 cases each of urinary tract injury and dural tear. Four patients developed early wound infections, 7 developed late wound infections, and 2 developed flap necrosis. Three patients developed recurrence, whereas 7 patients developed metastasis postoperatively. The mean survival was calculated to be 28 months and the mean Musculoskeletal Tumor Society score was 19.3 ± 5.2.

Conclusions: Outcomes and prevalence of complications shown in this study are comparable to those in the international literature, which suggests that hemipelvectomy is a viable option in developing countries also. However, more such studies are warranted to validate the findings and to identify the challenges and morbidities associated with hemipelvectomy in Asian and

**Keywords:** Pelvic tumors, Internal hemipelvectomy, Outcomes, Complications

Primary bone sarcomas of the pelvis account for only 5%–10% of all malignant bone tumors, but individual importance is still given to this region because of its anatomic complexity [1–4]. These tumors also tend to present late and therefore pose a major challenge for orthopedic oncologists because of the large tumor size, local extension, and complex anatomy with proximity to major neurovascular structures and intestinal and urinary tracts. Subsequently, these are also associated with less favorable outcomes in terms of prognosis and survival compared with other extremity tumors [2,5–7].

Previously, the mainstay for treatment of malignant pelvic tumors was external hemipelvectomy (hindquarter amputation), but with advancements in surgical techniques and chemotherapy and radiation therapy, limb salvage procedures (internal

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hemipelvectomy) have also emerged as viable modalities <sup>[5]</sup>. This involves resection of the lesion with part or all of the hemipelvis, but preserving the ipsilateral lower extremity. In patients in whom the tumor has not invaded major neurovascular structures, wide resection is possible, and therefore internal hemipelvectomy can be considered without affecting the functionality of the limb. However, in cases of neurovascular invasion and where widemargin excision is difficult, external hemipelvectomy remains the preferred option <sup>[8,9]</sup>.

Whether internal or external, hemipelvectomy is a major surgical procedure and may be associated with significant morbidity and functional impairments including injury to gastrointestinal or genitourinary tract, neurovascular injury, considerable soft tissue defects, blood loss, wound infections, and delayed wound healing <sup>[5,9,10]</sup>. As hemipelvectomy requires special expertise and is not a commonly performed procedure, there is scarcity of literature available on its long-term complications and outcomes, especially from developing countries where there is a lack of resources and training. Therefore, the aim of this study was to share our 10 years' experience of internal hemipelvectomy at a tertiary care center from a developing country's prospective.

#### **Materials and methods**

A retrospective review was conducted between January 1, 2005 and December 31, 2015 in which all patients undergoing internal hemipelvectomy for pelvic tumors were included. All surgeries were performed by a single fellowship-trained orthopedic oncologist at our hospital. The study was approved by the Ethics Review Committee at our institution.

Medical record files for all 24 patients included in the study were reviewed. All patients had undergone blood and radiologic work up before surgery. Radiologic investigations included x-ray pelvis and chest and magnetic resonance imaging of pelvis to identify the extent of disease, and only those patients were offered internal hemipelyectomy in whom complete excision of tumor seemed possible. The final tumor size was noted from the final histopathologic report of the specimen and was categorized on the basis of the largest diameter being either  $\geq 20$  cm, between 10 and 20 cm, or < 10 cm<sup>[9]</sup>. All musculoskeletal tumors were staged according to the Enneking system of staging for malignant and benign musculoskeletal tumors. Metastatic pelvic carcinomas from other primary sources were classified as stage IV. Internal hemipelvectomy was further classified according to the Enneking and Dunham classification, and an en bloc resection of the ilium and sacral ala was classified as type IV<sup>[11,12]</sup>. Administration of neoadjuvant and/or adjuvant chemoradiation therapy was dependent on the diagnosis, and the number of cycles given was noted. The percentage response to neoadjuvant therapy in terms of tumor necrosis was also noted from the final histopathology report.

Intraoperative, early postoperative (during the hospital stay), and late postoperative (on follow-up visits) complications were noted and their management was also reviewed. Functional outcome after the surgery was assessed using the Musculoskeletal Tumor Society (MSTS) scoring system. Patients were contacted and interviewed on telephone to calculate the MSTS score. For patients who were deceased before the study, their cause and time duration after the primary surgery for the mortality were obtained from the family. Subsequently, the average survival rate was calculated using Kaplan-Meier analysis. Independent sample t test was used for comparison of means, whereas  $\chi^2$  test was used for categorical variables. A P-value of < 0.05 was considered significant.

#### **Results**

Table 1 describes the characteristics for all included patients. Of a total of 24 patients, 14 (58.3%) were male and 10 (41.7%) female, and their mean age was  $23.6 \pm 13.6$  years. The most common diagnosis for primary pelvic tumor was found to be Ewing sarcoma (50%), whereas second most common diagnosis was osteosarcoma (12.5%). Three patients (12.5%) underwent internal hemipelvectomy for metastases of lung, rectal, and endometrial carcinomas with the intention of palliative treatment and pain relief. A total of 8 patients (33.3%) required reconstruction and flap coverage, and the most common type of flap used was the rectus abdominus flap (75%), whereas the vastus lateralis flap and the anterolateral thigh flap were used in 1 patient each. The mean duration of surgery was calculated to be 356±108 minutes, the mean length of hospital stay was 11.3 ± 6.5 days, whereas the mean follow-up period was  $18.7 \pm 13.9$  months. There were 5 patients who were lost to follow-up.

All but 1 patient suffering from Ewing sarcoma received neoadjuvant chemotherapy with a mean of 7 cycles (range, 4–18 cycles). Of these 11 patients, 2 were also given neoadjuvant radiotherapy. Six patients also required adjuvant chemotherapy with a mean of 5.7 cycles, whereas one of them received concomitant adjuvant radiotherapy also. All 3 patients with

osteosarcoma received neoadjuvant chemotherapy with a mean of 4.3 cycles and none of them required any adjuvant treatment. All patients with other primary carcinomas and metastases to the pelvis received neoadjuvant chemotherapy in accordance with the standard regime for the particular primary carcinomas. Patient 17 also received neoadjuvant radiotherapy, whereas patient 22 required 6 cycles of adjuvant chemotherapy. The only other diagnosis requiring neoadjuvant therapy was spindle cell carcinoma, in which 4 cycles of chemotherapy were administered in addition to radiotherapy.

There were 15 patients (62.5%) with a largest tumor diameter of < 10 cm, 6 patients (25%) with a tumor diameter between 10 and 20 cm, and 3 patients (12.5%) with  $\geq$  20 cm. Seven patients presented with invasion of the neurovascular bundle by the tumor. Two patients had both neural and vascular invasion, out of whom 1 presented with invasion of the obturator nerve and vessels, whereas the other had invasion of the sciatic nerve and terminal branches of the external iliac artery. The most commonly involved nerve was the sciatic nerve (3 patients), followed by the femoral, obturator, and sacral plexus in 1 patient each. The only other patient with vascular involvement had invasion of superior and inferior gluteal vessels along with obturator artery. In each case, the involved nerve was dissected and the involved vessel was divided and ligated. Negative margins were confirmed on frozen sections. For the minimum tumor-free margin, the mean was calculated to be  $0.8 \pm 1.2$  cm, whereas 4 patients still had 1 positive resection margin. The mean intraoperative blood loss was 1900 ± 1600 mL, and subsequently, the mean intraoperative pack cell volume transfusion requirement was  $1200\pm1000$  mL, with a further requirement of  $800\pm700$  mL during hospital stay. In addition, the mean transfusion requirement for fresh frozen plasma and platelets during hospital stay was  $400 \pm 800$  mL and  $100 \pm 200$  mL, respectively.

### Complications

Overall complications associated with hemipelvectomy are summarized in Figure 1. In 2 cases, there was an injury to the femoral nerve, whereas in 1 case, the sciatic nerve was partially cut accidentally and each of these nerves were repaired. One patient had injury to the urinary bladder and 1 patient suffered injury to the prostatic urethra due to close proximity with the tumor. Both injuries were managed with the help of a urologist. Immediate postoperative bleeding was noted in 2 patients from the internal iliac artery and required angioembolization. Two of the early wound infections were managed with antibiotics, whereas 2 required surgical debridement. Late wound infections were noted at a mean of  $30 \pm 16$  days after surgery, out of which 2 patients had superficial infections and were managed by antibiotics, whereas the rest required a mean of 1.6 (range, 1-3) surgical debridements. Two patients with flap necrosis presented at the 21st and 34th postoperative days with necrotic rectus abdominus flaps and both were managed with a second vastus lateralis flap.

# Oncologic outcome

Local recurrence was noted in 3 patients (12.5%) at a mean of 4 months (range, 1–6 mo) postoperatively. No significant correlation was found between recurrence and diagnosis, tumor size, or the administration of adjuvant therapy as shown in **Table 2**. The mean percentage response to neoadjuvant therapy was found

Table 1

Characteristics of th	e study	population.
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Patient No.	Age (y)	Sex	Diagnosis	Stage	Largest Tumor Diameter (cm)	Type of Hemipelvectomy
1	17	Male	Spindle cell carcinoma	IIB	10–20	
2	25	Female	Chondrosarcoma	IIB	10–20	I/II
3	16	Female	Ewing sarcoma	IIB	< 10	I/II
4	16	Male	Ewing sarcoma	IIB	< 10	II/III
5	14	Female	Ewing sarcoma	IIB	< 10	1/11/111
6	32	Male	Chondrosarcoma	IIB	10–20	III
7	30	Female	Giant cell tumor	III	10–20	1
8	21	Male	Osteosarcoma	IIB	< 10	II
9	8	Male	Ewing sarcoma	IIB	≥20	I/II/IV
10	47	Male	Giant cell tumor	III	≥20	I/II
11	16	Male	Osteosarcoma	IIB	10–20	1/11/111
12	13	Male	Ewing sarcoma	IIB	< 10	III
13	5	Male	Ewing sarcoma	IIB	< 10	I/II
14	15	Male	Ewing sarcoma	IIB	< 10	1
15	14	Male	Ewing sarcoma	IIB	< 10	1
16	22	Female	Ewing sarcoma	III	< 10	I/II
17	55	Female	Lung carcinoma	IV	< 10	1/11/111
18	12	Female	Ewing sarcoma	IIB	< 10	1
19	14	Female	Ewing sarcoma	IIB	< 10	I/IV
20	40	Male	Osteosarcoma	IIB	≥20	1/11/111
21	33	Male	Rectal carcinoma	IV	< 10	I/IV
22	42	Female	Endometrial carcinoma	IV	< 10	11/111
23	17	Male	Ewing sarcoma	IIB	10–20	I/II/IV
24	45	Female	Hydatid disease	III	< 10	II/III

to be significantly lower in patients with recurrence compared with those without recurrence (P = 0.04). Both cases of Ewing sarcoma recurrence were managed with chemotherapy, but the patients died 2 and 10 months after recurrence, whereas the patient with giant cell tumor was lost to follow-up.

A total of 7 patients (29.2%) developed metastatic disease at a mean of  $18.8\pm20$  months after surgery and the most common site for metastasis was the lungs in 5 patients (71.4%). Other sites involved were the brain (1 patient) and the knee joint (1 patient).

Table 3 shows different factors associated with the development of metastases of primary pelvic tumors, none of which were found to be significant.

# Survival and functional outcome

The mean survival was calculated to be 28 months. The Kaplan-Meier analysis in **Graph 1** shows a 2-year survival rate of 50%. There was no significant association between survival and other

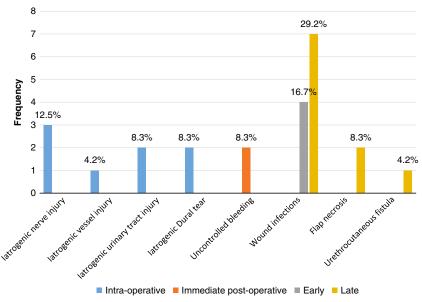


Figure 1. Frequency of different complications noted.

Table 2
Characteristics with reference to the development of local recurrence (n = 24).

	Local Recurrence [n (%)]			
Characteristics	Yes	No	P	
Diagnosis			0.65	
Ewing sarcoma	2 (16.7)	10 (83.3)		
Osteosarcoma	0	3 (100)		
Giant cell tumor	1 (50.0)	1 (50.0)		
Spindle cell carcinoma	0	1 (100)		
Chondrosarcoma	0	2 (100)		
Hydatid disease	0	1 (100)		
Metastatic disease	0	3 (100)		
Tumor size (cm)			0.358	
< 10	2 (13.3)	13 (86.7)		
10–20	0	6 (100)		
≥20	1 (33.3)	2 (66.7)		
Adjuvant therapy			0.19	
Yes	0	8 (100)		
No	3 (18.8)	13 (81.2)		

characteristics including the presence of metastases (P = 0.25), local recurrence (P = 0.35), tumor size (P = 0.21), and requirement of flap reconstruction (P = 0.77). The mean MSTS score for the surviving patients was  $19.3 \pm 5.2$ . Five of the 14 patients were walking without support, whereas 1 patient was bedridden. The rest were using either canes or crutches for ambulation. It was found that patients who needed flap coverage had a significantly lower mean MSTS as compared with those patients who did not (P = 0.03).

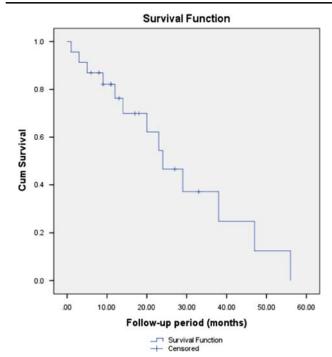
## **Discussion**

Internal hemipelvectomy is performed for all pelvic tumors including primary bone and soft tissue sarcomas and secondary metastatic tumors, with the intention of either local tumor control or palliative care and pain control. The most common malignant pelvic tumor is reported to be chondrosarcoma, followed by

Table 3
Characteristics with reference to the development of metastasis in cases of primary pelvic tumors (n = 21).

	Metastases [n (%)]		
Characteristics	Yes	No	P
Diagnosis			0.76
Ewing sarcoma	5 (41.7)	7 (58.3)	
Osteosarcoma	1 (33.3)	2 (66.7)	
Giant cell tumor	1 (50.0)	1 (50.0)	
Spindle cell carcinoma	0	1 (100)	
Chondrosarcoma	0	2 (100)	
Hydatid disease	0	1 (100)	
Tumor size (cm)			0.33
<10	4 (33.3)	8 (66.7)	
10–20	1 (16.7)	5 (83.3)	
≥20	2 (66.7)	1 (33.3)	
Chemoradiotherapy*			1.00
Yes	5 (33.3)	10 (66.7)	
No	2 (33.3)	4 (66.7)	

<sup>\*</sup>Adjuvant or neoadjuvant therapy.



Graph 1. Kaplan-Meier analysis for survival.

Ewing sarcoma and osteosarcoma [1,5,13]. In comparison, most of our patient's had Ewing sarcoma and only 8% of the patients had chondrosarcoma. An epidemiological study conducted in Pakistan in 2010 showed a similar prevalence of different bone sarcomas as shown in the international literature, but it did not address site-specific prevalence, and therefore the possibility of a different prevalence pattern for pelvic tumors in Pakistan compared with the western world cannot be excluded [14]. The mean follow-up period was comparable to that of other studies, but it was noted that around 21% of the patients were lost to follow-up [7,15]. In our setting, most of the patients belong to lower socioeconomic classes and also come from distant places, and hence financial constraints and inadequate transportation could be the possible causes of loss to follow-up.

The most common complication noted in our study was wound infections, which also corresponds to previously available literature [1,2,15]. In terms of intraoperative complications, injuries to the urinary tract have also been reported previously and are attributed to the complex pelvic anatomy and close proximity of all internal viscera [9]. We also reported 3 cases of iatrogenic neurovascular injury, which were managed immediately without any subsequent complications. It is important to note that in all 3 cases, the tumor size was > 10 cm, which indicates that maneuvering around the major structures without damage and complete tumor excision becomes more difficult with an increased tumor size. The 2 cases of dural tear and cerebrospinal fluid leakage were managed immediately with dural repair, followed by placement of lumbar drain. Donati et al<sup>[16]</sup> in their study for the treatment of pelvic osteosarcoma also reported a case of dural tear and a case of persistent cerebrospinal fluid leakage out of 14 internal hemipelvectomy patients.

There were 3 cases of local recurrence, 2 of Ewing sarcoma, and 1 case of giant cell tumor. Similar recurrence rates have also

been reported previously <sup>[2,15,17]</sup>. It is however interesting to note that all 3 patients had high-grade aggressive lesions, and recurrence occurred despite clear resection margins in each case. Even though there was no significant association between recurrence and receiving adjuvant therapy, none of the 3 patients had received adjuvant chemoradiation. This might suggest that patients with high-grade lesions should receive adjuvant therapy irrespective of resection margins. Previously, Pant et al<sup>[18]</sup> in 2000 also reported the recurrence of a high-grade pleomorphic osteosarcoma after clear margins, which further supports the suggestion. Recurrence of Ewing sarcoma could also be attributed to the significantly reduced response to neoadjuvant therapy in both cases. There is still a lack of literature on evaluating the importance of response to neoadjuvant treatment in preventing recurrence of pelvic tumors after hemipelvectomy.

The overall distant metastasis rate after hemipelvectomy has been shown to range from 13.3% to 28% in international literature, in comparison with 29.2% reported in our study [8,17]. It is important to note that all of the tumors in this study were high grade, stage IIB and above for malignant lesions, and stage III for benign lesions, which could be a possible reason for the higher rate of metastasis. In addition, this also shows that all patients had delayed presentation, which can again be attributed to the transportation and financial constraints, and therefore it is important to improve the overall health care infrastructure for effective intervention and referrals in developing countries such as Pakistan. However, the mean MSTS score is still comparable to that of previous studies, and a significantly lower score in patients requiring flap coverage highlights the increased morbidity associated with reconstruction [8,15].

One of the major limitations of this study is its retrospective design, and therefore the dependence on patient record files for follow-up data. Another limitation is the small sample size and a short mean follow-up period. More such studies are required from the developing world with long-term follow-ups to identify the complications and long-term outcomes of hemipelvectomy. An important finding was the reduced response rate of neoadjuvant therapy in both cases of Ewing sarcoma recurrence. This might suggest a need to revisit the chemoradiation therapy protocol, but more data from multiple centers are required to reach a conclusion.

# **Conclusions**

Hemipelvectomy is the mainstay of treatment for pelvic tumors. The type of hemipelvectomy is dependent on various factors including the extent of disease and patient preference. The similar outcomes and prevalence of complications shown in this study compared with international literature suggests that hemipelvectomy is a viable option in developing countries also. Some important findings pointed out in the study were the different tumor prevalence pattern and reduced response to neoadjuvant therapy in Ewing sarcoma leading to recurrence. However, as there is a considerable lack of literature from developing and Asian countries, more such studies are warranted to validate

these findings and to identify the challenges and morbidities associated with hemipelvectomy in these regions.

#### **Conflicts of interest**

The authors declare that they have no financial conflict of interest with regard to the content of this report.

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