

En Bloc Sacrectomy for Retrorectal Tumor: Factors Influencing Immediate Outcome

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Abstract

Background: *En bloc* sacrectomy is the only chance for cure of retrorectal tumor. Total sacrectomy is a complicated, time-consuming and collaborative operation with multiple potential postoperative morbidities and mortality. Well-planned surgery, perioperative care and rehabilitation would be beneficial in surgical outcomes and decrease postoperative complications.

Objective: To define the factors affecting the immediate outcomes of *en bloc* sacrectomy in patients with retrorectal tumor.

Study design: A retrospective cohort at a single academic institute.

Methods: Data were collected retrospectively in all patients who underwent *en bloc* sacrectomy at the Lerdsin Hospital between June 2008 and August 2013. We reviewed demographic data, clinical findings, past medical history, tumor characteristics, tumor invasion, preoperative preparation: iliac artery embolization, ureteric stenting, surgical procedures, intraoperative complication, estimated blood loss, units of blood transfusion, histopathologic findings, postoperative morbidities and mortality, functional outcomes: constipation, urinary retention and gait disturbance. Immediate surgical outcome was defined according to the postoperative complications, hospital length of stay, functional outcomes and gait disturbance. All parameters were analyzed and determined for their significance.

Result: Fifteen patients (6 males and 9 females) who underwent sacrectomy (6 subtotal sacrectomy and 9 total sacrectomy) were studied. The mean age was 53.8 years (range 26-83 years). Of these, 13 had preoperative iliac artery embolization, 6 had ureteric stenting, 8 had anterior approach with bilateral internal iliac vessels ligation, 2 had protective colostomy and 3 had lumbosacral reconstruction. The average operative time was 8.4 hours (4-14 hours). The mean estimated blood loss was 5,020 mL (1,600-10,000 mL) and the average blood transfusion was 17.6 units (range 1-40 units). Pathological diagnosis were chordoma (n=7), malignant peripheral nerve sheath tumor (n=3), chondrosarcoma (n=2), and giant cell tumor, gastrointestinal stromal tumor (GIST) and leiomyosarcoma (1 each). Based on the Clavien-Dindo Grading Systems for postoperative complications, 1 patient was categorized into Class I, 2 in Class II, 4 in Class IIIa, 7 in Class IIIb and 1 in Class V. Eight patients (53%) had major complications and required re-operation and 1 patient died from severe sepsis. Regarding functional outcomes, 14 of 15 patients had postoperative urinary retention and 10 of them (67%) had constipation. The gait disturbance was as follows: 5 patients walked without assistance, 6 patients walked with assistance and 3 patients needed wheelchairs. None had sacral hernia. The mean duration of hospital stay was 77 days (range 24-186 days).

Conclusion: Subtotal sacrectomy was safe and had less complications whereas preoperative iliac artery embolization did not influence intraoperative blood loss. The anterior approach with internal iliac vessel ligation decreased blood loss especially in total sacrectomy and the posterior approach is a safe option for subtotal sacrectomy. The protective colostomy did not affect the incidence of surgical site infection but helped in decreasing its severity. Despite preoperative ureteric stenting, ureteric injury could not be prevented. The closure of sacral defect could be adequately undertaken without prosthesis sheath by gluteal advancement flap or gluteus maximus approximation. There was no difference between total and subtotal sacrectomy in terms of urinary retention and constipation. Gait disturbance is less in subtotal sacrectomy compared to total sacrectomy. Without lumbopelvic reconstruction, total sacrectomy could give favorable intermediate and long term outcome.

Keywords: pelvic surgery, presacral tumor, retrorectal tumor, sacrectomy

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INTRODUCTION

Retrorectal tumors are rare neoplasms locating in the retrorectal (presacral) space. These neoplasms originate from various embryonic remnants and have a large variety in either gross or histological findings. True incidence of retrorectal tumors remains unclear with approximately 1 in 40,000 hospital admissions. Although the majority of retrorectal tumors are benign, malignant neoplasms are observed especially in pediatric population and solid tumor type¹⁻⁶. Because pelvic cavity is a closed narrow space which contains a variety of vital organs, pelvic surgery requires a comprehensive knowledge of pelvic anatomy, expertise of involved surgeons, and a well-organized teamwork to achieve a safe and successful operation.

The primary curative modality for retrorectal tumor is *en bloc* resection which requires multidisciplinary specialists. Operation in this surgical field, however, remains vulnerable to complication and the operative environment is often hostile. In this study, we collected case series and determined the possible factors that affect immediate surgical outcomes and complications of the *en bloc* sacrectomy for retrorectal tumor. The data acquired from this study would facilitate perioperative care and improve morbidities, mortality and quality of life of the patients. Moreover, we may utilize these data to develop standard guideline for the treatment of retrorectal tumors in the future.

PATIENTS AND METHODS

We retrospectively studied 15 patients who underwent surgery for retrorectal tumor at the Lerdsin Hospital, Department of Medical Services, Ministry of Public Health of Thailand between September 2008 and August 2013. We reviewed demographic data, clinical findings, past medical history, tumor characteristics, tumor invasion, preoperative preparation: iliac artery embolization, ureteric stenting, surgical procedures, intraoperative complication, estimated blood loss, units of blood transfusion, histopathological findings, postoperative morbidities and mortality, functional outcomes: constipation, urinary retention and gait disturbance. We attained patient data only in the admission period by browsing hospital archives.

Statistical analysis

The studied data would be presented in mean or median and range if they were continuous variables. These variables were analyzed by student t test if they had normal distribution and by Chi-squared test or Fisher exact test if they were categorical variables. Mann-Whitney U test was used to test whether two independent samples of observations were drawn from the same or identical distributions, such as tumor size, estimated blood loss and subgroup (subtotal/total sacrectomy) analysis. The overall survival and overall disease-free survival rates were traced using the Kaplan-Meier method. *P* value less than 0.05 was considered statistically significant. Statistical analysis was done using SPSS software.

Preoperative assessment and evaluation

Patients were carefully evaluated following the American Society of Anesthesiologists (ASA) standards. Patients in ASA class I-III were eligible for surgery. Clinical data and radiologic imaging (CT/MRI) of patients were discussed in the manner of team approach involving general surgeon, orthopedist, urologist, plastic surgeon, gynecologist and radiologist. Operative planning and sequence of procedures were summarized in the multidisciplinary preoperative meeting. In the case that surgeon considered preoperative arterial embolization, the procedure would be performed by interventional radiologist. The embolization of the tumor-feeding internal iliac artery was done either unilaterally or bilaterally depending on the angiographic findings 2-3 days prior to surgery.

Surgical technique

En bloc sacrectomy consisted of two parts: anterior approach and posterior approach. In some circumstances such as large tumor, obstructive uropathy and unclear ureteral identification from preoperative imaging, the surgeons might consider ureteric stent placement which would be done by urologist. In anterior approach stage, the patient was in a supine position. After a standard midline laparotomy incision, sigmoid colon and rectum were mobilized from tumor. The rectum must be dissected as distal as possible. In some patients whose rectum tightly adhered to tumor or suspected tumor invasion, the rectum was transected proximal to the adhesion with linear staple, then the proximal end was brought out through abdominal

wall and matured later. The distal end of rectum was left in place and removed altogether with the tumor in posterior approach stage. Both ureters would be freed away from the tumor and also dissected as distal as possible. Both sides of common iliac, external and internal iliac vessels were identified. The internal iliac arteries and veins were ligated and transected at their origin bilaterally, only in the patients who could be safely done. Other soft tissues and structures around the tumor and sacroiliac joints were dissected and cleared out. The abdominal wall was closed as usual.

In posterior approach stage, the patient was in a prone position. A midline incision was made beginning at the level of L3 down to the coccygeal tip 5-10 cm above the anus. If there was a preoperative core needle biopsy tract, the tract and adjacent tissue were removed with the specimen using an elliptical skin incision around the biopsy tract. The incision was extended laterally toward the greater trochanter on both sides

(inverted Y incision). The incision was carried through subcutaneous tissue and lumbosacral fascia. After the fascia was incised, it was elevated off the paraspinous muscles laterally. There was an avascular plane that could be followed to the iliac crest bilaterally. The fascia was elevated off of the iliac crest. Using a subperiosteal technique, the paraspinous muscles were elevated off of the spinous process, the lamina, the facets, and the transverse process of the spine. If a total sacrectomy was planned, this exposure must encompass L3 through L5. If a subtotal sacrectomy was to be performed, then exposure of L5 was sufficient. The paraspinous muscles were then cut transversely at the same level at which the fascia was incised. The gluteal muscles inserting on the lateral aspect of the ilium were elevated, allowing visualization of the sciatic notch. Laminectomy was performed at the level of the most caudal nerve root to be preserved. For the subtotal sacrectomy, S1 laminectomy was made and the S1

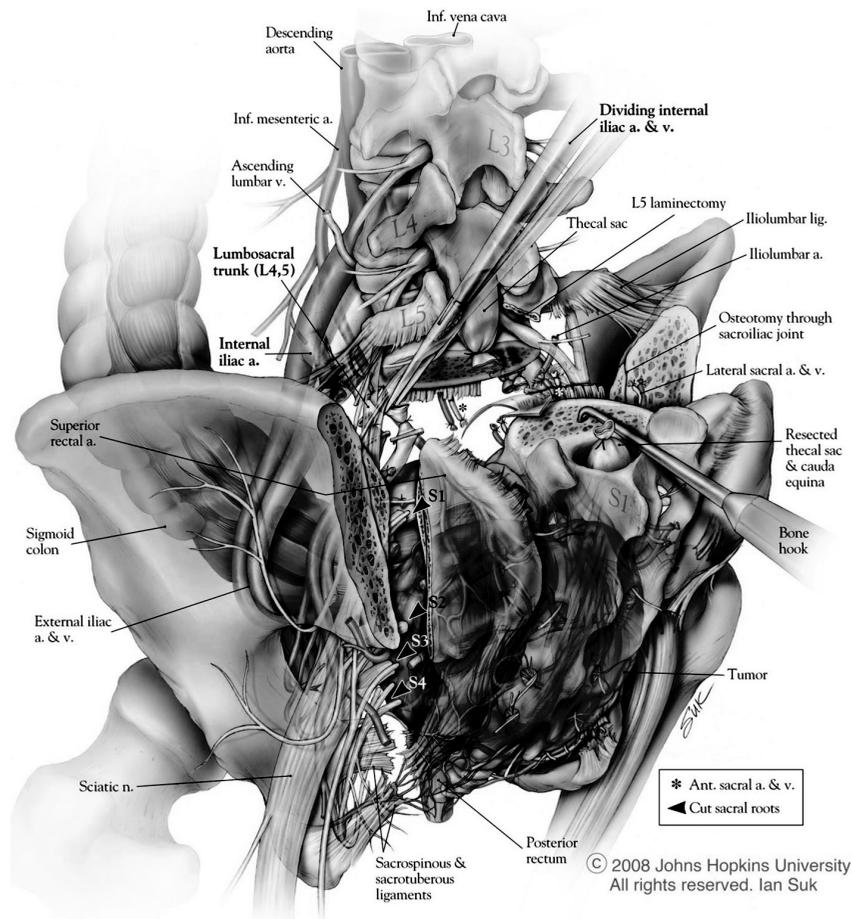


Figure 1 The summary illustration of total sacrectomy from posterior aspect. (Printed with permission. ©2008 The Johns Hopkins University Neurosurgery-Ian Suk) (From Ian suk. Dissecting a complex neurosurgical illustration: step-by-step development. World Neurosurg. 76, 6:497-507, 2011.)

roots were carefully identified and preserved. Dural sac was divided and ligated at the S1-2 level, and then the sacral body was exposed for a safe osteotomy at the S1-2 junction. Posterior osteotomy was continued on the ala of sacrum and the sacroiliac joints, midline osteotomy was performed, connecting the lateral osteotomies through the vertebral body, and subsequently the sacrum could be mobilized. S2-4 nerves were separated from sciatic nerves, piriformis muscles and ligamentous structures (sacrospinous, sacrotuberous, sacrospinous ligament) were divided. The internal iliac arteries and veins were ligated and transected if they were not previously performed in anterior approach stage. In the same way, these processes were done with other branches of the vessels such as iliolumbar vessels and lateral sacral vessels. The mesorectum was carefully dissected off the capsule of tumor, in cases where rectoproctectomy was performed, distal rectum and anus were removed together with the tumor, then *en bloc* tumor resection was accomplished. For total sacrectomy, an L5-S1 laminectomy was performed and the dural sac was ligated at L5-S1 with preservation of the L5 roots. An L5-S1 discectomy was performed followed by a posterior osteotomy at the sacroiliac joints for total sacrectomy. The S1-4 nerves were then separated from the sciatic nerves, the following steps were the same as previously described in subtotal sacrectomy. Figure 1 demonstrates summary of the total sacrectomy operation.

Lumbopelvic reconstruction was done in three patients by Galveston reconstruction system, however, two patients had postoperative pelvic infection that required instrument removal. Thereafter, the surgeons did not consider lumbopelvic reconstruction in the rest of total sacrectomy cases. Sacral defect closure was done by approximation of the gluteus maximus. In the cases with large sacral defect with inadequate remained viable muscles, reconstruction has been done with gluteal advancement flap. Subsequently, the skin flap was closed, self-suctioning drains were placed. In the patients who underwent rectoproctectomy or Hartmann's procedure concurrent with *en bloc* sacrectomy, colostomy would be matured after the patients were repositioned back to supine position.

RESULTS

Summary of patients' data was shown in Table 1.

Fifteen patients with retrorectal tumors underwent *en bloc* sacrectomy from September 2008 to August 2013. They were 6 male (40%) and 9 female (60%) with a mean age at presentation of 53.8 years (range 26-83 years). All of them were ASA functional classification I-III. Pathological diagnosis included chordoma (n=7), malignant peripheral nerve sheath tumor (n=3), chondrosarcoma (n=2) and 1 each of giant cell tumor, GIST and leiomyosarcoma. The mean tumor volume was 1,755 mL (average 12 × 12 × 12 cm) and 5 of them (30%) had sacral bone invasion.

Of 15 patients, 6 underwent subtotal sacrectomy and 9 underwent total sacrectomy while 13 of them had preoperative iliac artery embolization. Ureteric stent placements were also done in six patients. For anterior approach: eight patients underwent bilateral internal iliac vessels ligation and two patients underwent protective colostomy. Among the total sacrectomy group, lumbosacral reconstruction was done in 3 patients and hemipelvectomy was done in 1 patient due to tumor extension who later had severe sepsis and died after 71 days of admission. The mean operative time, estimated blood loss and units of blood transfusion were 8.4 hours (range 4-14 hours), 5,020 mL (range 1,600-10,000 mL), and 17.6 units (range 1-40 units), respectively. The details of studied data are listed in Table 1.

According to Clavien-Dindo Grading System for the Classification of Surgical Complications (Table 2)⁷, patients were classified as 1 in class I, 1 in class II, 4 in class IIIa, 7 in class IIIb, 0 in class IV and 1 in class V. As mentioned above, there were 8 patients (53%) with severe complications (Class IIIb, IV, V) who needed re-operations or died. Common complications included 11 wound infections (73%) 7 of which required re-operations for wound debridement, 1 intra-abdominal collection (6.7%) which required re-exploration and drainage, 2 ureteric injuries (13.4%) which required percutaneous nephrostomy, 2 rectal injuries (13.4%) which required re-operation for Hartmann's procedure. There were three patients who underwent a combined total sacrectomy and lumbopelvic reconstruction, two of them experienced postoperative wound infection with instrument failure. Both of them needed re-operation for instrument removal. None of patients in this study had sacral hernia during admission or after the 2-4 year follow-up.

Table 1 Summary data of the Patients' demographic, tumor factors, preoperative factors, operative factors, complications, functional outcomes and length of stay

Patient	Demographic data		Tumor Factors		Preoperative Factors		Operative Factors: Technique				Operative Factors: Others			Complications					Functional outcomes			Others			
	Age	Gender	ASA	Tumor pathology	Tumor size (cm)	Bone invasion	Embolization	Ureteric stenig	Operation type	Internal iliac vessels ligation	Colostomy	Cystostomy	Operative time (hrs)	EBL (cc)	TBT (units)	Wound infection	Collection	Sacral hernia	Others	No. of re-operation	Clavien-Dindo score		Constipation	Urine retention	Gait disturbance
1	59	M	2	Chondrosarcoma	13x20x24	+	+	+	subtotal sacrectomy	-	-	-	10	1,600	11	+	-	-	8 (8 wound debridement)	IIlb	+	+	walk, no assistance	90	
2	32	F	1	Chondrosarcoma	7x11x10	+	+	+	total sacrectomy +LPR	+	-	-	12	6,000	40	+	+	rectal injury, instrumental failure	IIlb	-	-	+	walk, with assistance	107	
3	54	F	1	Giant cell tumor	12x25x11	-	+	-	total sacrectomy	-	+	-	8	15,000	30	-	+	-	1 (1 instrument removal, 7 debridement)	IIlb	-	+	wheel chair	110	
4	56	F	1	Chordoma	7x6x7	-	+	-	subtotal sacrectomy	-	-	5	300	1	-	-	-	-	-	I	+	+	walk, no assistance	24	
5	58	F	2	MPNST	12x13x14	+	+	+	total sacrectomy +LPR	+	-	14	8,000	28	+	+	-	ureter injury, DVT, instrumental failure	IIlb	+	+	+	wheel chair	186	
6	60	M	1	Chordoma	13x12x16	-	+	+	total sacrectomy +LPR	+	-	6	4,000	14	-	-	-	-	-	II	-	-	+	walk, with assistance	35
7	26	F	1	GIST	4x3x2	-	+	-	subtotal sacrectomy	+	-	4	3,800	12	+	-	-	-	-	IIla	+	+	walk, no assistance	33	

Table 1 Summary data of the Patients' demographic, tumor factors, preoperative factors, operative factors, complications, functional outcomes and length of stay

Patient	Demographic data		Tumor Factors		Preoperative Factors		Operative Factors: Technique				Operative Factors: Others			Complications						Functional outcomes			Others		
	Age	Gender	ASA	Tumor pathology	Tumor size (cm)	Bone invasion	Embolization	Ureteric stenig	Operation type	Internal iliac vessels ligation	Colostomy	Cystostomy	Operative time (hrs)	EBL (cc)	TBT (units)	Wound infection	Collection	Sacral hernia	Others	No. of re-operation	Clavien-Dindo score	Constipation		Urine retention	Gait disturbance
8	76	M	3	Chordoma	17x15x11	-	-	+	subtotal sacrectomy	-	-	8	4,000	12	+	-	-	-	-	-	IIla	+	+	walk, with assistance	74
9	62	F	2	Chordoma	12x10x10	-	-	-	subtotal sacrectomy	-	-	4	1,500	6	+	-	-	-	-	-	IIla	+	-	walk, no assistance	34
10	36	M	1	MPNST	12x7x6	+	+	-	subtotal sacrectomy	-	-	4	2,600	7	-	-	-	-	-	-	I	+	+	walk, no assistance	37
11	42	F	2	MPNSt	6x6x10	+	+	+	Total sacrectomy + lt. hemipelvec-tomy	-	-	12	5,500	13	+	-	-	sepsis, dead	4 (4) wound debridement	V	+	-	walk, no assistance	71	
12	60	F	2	Sarcoma	12x7x10	-	+	-	total sacrectomy	+	-	9	15,500	29	+	-	-	-	-	-	IIla	-	+	walk, with assistance	80
13	31	M	1	Chordoma	17x15x14	-	+	-	total sacrectomy	-	-	12	11,000	21	+	+	-	ureter injury	3 (2) wound debridement	IIlb	+	+	walk, with assistance	82	
14	83	F	3	Chordoma	8x6x10	-	+	+	total sacrectomy	+	-	6	10,000	25	+	-	-	-	2 (2) wound debridement	IIlb	+	+	whell chair	99	
15	72	M	3	Chordoma	12x13x10	-	+	-	total sacrectomy	+	-	12	4,000	15	+	-	-	rectal injury foot drop	8 (8) wound debridement	IIlb	-	+	walk, with assistance	97	

ASA = American Society of Anesthesiologists Physical Status Classification System; EBL = Estimated Blood Loss (intraoperation); TBT = Total Blood Transfusion; LOS = Length of Stay

Table 2 Clavien-Dindo Grading System for the Classification of Surgical Complications

Grades	Definition
Grade I:	Any deviation from the normal postoperative course without the need for pharmacological treatment or surgical, endoscopic and radiological interventions. Allowed therapeutic regimens are: drugs as antiemetics, antipyretics, analgetics, diuretics and electrolytes and physiotherapy. This grade also includes wound infections opened at the bedside.
Grade II:	Requiring pharmacological treatment with drugs other than such allowed for grade I complications. Blood transfusions and total parenteral nutrition are also included.
Grade III:	Requiring surgical, endoscopic or radiological intervention
Grade III-a:	Intervention not under general anesthesia
Grade III-b:	Intervention under general anesthesia
Grade IV:	Life-threatening complication (including CNS complications: brain haemorrhage, ischaemic stroke, subarachnoid bleeding, but excluding transient ischaemic attacks) requiring IC/ICU management.
Grade IV-a:	Single organ dysfunction (including dialysis)
Grade IV-b:	Multi-organ dysfunction
Grade V:	Death of a patient
Suffix 'd':	If the patients suffers from a complication at the time of discharge, the suffix 'd' (for 'disability') is added to the respective grade of complication. This label indicates the need for a follow-up to fully evaluate the complication.

Daniel D, Nicolas D, Pierre-Alain C: Classification of Surgical Complications: A New Proposal With Evaluation in a Cohort of 6336 Patients and Results of a Survey. Ann Surg. Aug 2004; 240(2): 205-213.

Table 3 Patient status in October 2014

Patient	Follow-up duration (years)	Type of operation	Functional outcomes			Tumor recurrence	Sacral hernia	Remarks
			Defecation	Urination	Gait			
1	loss F/U since 2011 (3-yr. F/U)	subtotal sacrectomy	moderate difficulty/ laxative use	straining	walk, no assistance	Yes (at 2009)	No	re-operation (wide excision at 2009)
2	4	total sacrectomy + LPR	required enema/ evacuation	on urinary catheter	walk, no assistance	No	No	
3	4	total sacrectomy	on colostomy	on urinary catheter	wheel chair, standable 1-2 min.	No	No	
4	4	subtotal sacrectomy	moderate difficulty/ laxative use	straining	walk, no assistance	No	No	
5	4	total sacrectomy + LPR	required enema/ evacuation	on PCN	walk, with assistance	No	No	
6	4	total sacrectomy + LPR	moderate difficulty/ laxative use	self catheterization	walk, no assistance	No	No	
7	2	subtotal sacrectomy	required enema/ evacuation	straining	walk, no assistance	No	No	
8	3	subtotal sacrectomy	required enema/ evacuation	on urinary catheter	walk, with assistance	No	No	
9	3	subtotal sacrectomy	normal defecation	normal urination	walk, no assistance	No	No	
10	3	subtotal sacrectomy	required enema/ evacuation	self catheterization	walk, no assistance	No	No	
11	-	total sacrectomy + lt. hemipelvectomy	-	-	-	-	-	Death
12	2	total sacrectomy	on colostomy	self catheterization	walk, with assistance	No	No	
13	2	total sacrectomy	required enema/ evacuation	self catheterization	walk, with assistance	No	No	
14	2	total sacrectomy	required enema/ evacuation	self catheterization	wheel chair	No	No	
15	1	total sacrectomy	on colostomy	self catheterization	walk, with assistance	No	No	

LPR = lumbopelvic reconstruction

Postoperative functional outcomes were also reviewed. Ten patients experienced constipation (67% of all patients, 90% of patients without colostomy) and 14 of 15 patients had urinary retention. Gait disturbances were noted as follows: 5 patients (33%) could walk without assistance, 6 patients (40%) could walk with assistance, 3 patients (20%) needed wheelchairs and 1 died (7%). The average hospital stay was 77 days (range 24-186 days).

Although it is beyond the scope of this study, in October 2014, 13 patients (87%) had the follow-up period lasting 2-4 years, 1 was lost to follow-up and 1 died after the operation. After analyzing the data with Kaplan-Meier method, the overall survival rate was 92.9% and overall disease free survival rate was 93.3%. A local recurrence was discovered in a single patient one year after surgery. The functional outcomes by the time of follow-up were listed in Table 3. Three patients (21%) had colostomy, 1 patient had normal defecation, 3 patients (21%) had moderate difficult defecation and required laxative, 6 patients (42%) required enema or evacuation. Urinary dysfunctions were also noted, 4 patients (28%) retained urinary catheter, 1 patient (7%) had normal urination, 3 patients (21%) had straining, and 6 patients (42%) performed self-urinary catheterization. Patients' gait disturbances were recorded, 2 patients (14%) used wheelchairs, 7 patients (49%) could ambulate without assistance and 5 patients (35%) could ambulate with assistance.

Reviewing record of the patient who lost follow-up, he regularly came in the first 3 years (2008-2010), however tumor recurrence was detected in one year after the operation. He underwent the re-operation of wide excision to remove the recurrent tumor. After the second operation, he continued another 2 years of follow-up until he lost contact in 2011. However, during his follow-up, he experienced moderate difficult defecation and required laxative, straining of urination and ambulation without assistance.

DISCUSSION

Various studies reported that *en bloc* sacrectomy is the only chance for cure of retrorectal tumors⁸⁻¹⁰. Our study showed that the intermediate and long term outcomes are favorable after 2-4 year follow-up (Table 3) with the overall survival rate of 92.9% and overall disease-free survival rate of 93.3%. Most of patients

Table 4 Factors influencing Clavien-Dindo Grading System for the Classification of Surgical Complications

	Clavien-Dindo Score (≤ IIIa)	Clavien-Dindo Score (≥ IIIb)	P-Value
Demographic data			
Age (year) : N (x̄, SD.)	7 (53.71, 16.987)	8 (53.88, 18.373)	0.986
Gender : N (%)			1.000
Male	3 (42.9)	3 (37.5)	
Female	4 (57.1)	5 (62.5)	
ASA : N (%)			1.000
I	4 (57.1)	3 (37.5)	
II	2 (28.6)	3 (37.5)	
III	1 (14.3)	2 (25.0)	
Tumor Factors			
Tumor Patholog : N (%)			0.576
Chondrosarcoma	-	2 (25.0)	
Giant cell tumor	-	1 (12.5)	
Chordoma	4 (57.1)	3 (37.5)	
MPNST	1 (14.3)	2 (25.0)	
GIST	1 (14.3)	0 (0.0)	
Leiomyosarcoma	1 (14.3)	0 (0.0)	
Tumor Size (cm ³) : N (Median)	7 (840)	8 (1,873)	0.247
Sacral bone invasion : N (%)			0.282
No	6 (85.7)	4 (50.0)	
Yes	1 (14.3)	4 (50.0)	
Preoperative Factors			
Embolization			0.200
No	2 (28.6)	0 (0.0)	
Yes	5 (71.4)	8 (100.0)	
Ureteric stenting			0.608
No	5 (71.4)	4 (50.0)	
Yes	2 (28.6)	4 (50.0)	
Operation Factors : Technique			
Operation : N (%)			0.041*
Subtotal sacrectomy	5 (71.4)	1 (12.5)	
Total sacrectomy	2 (28.6)	7 (87.5)	
IIV ligation : N (%)			0.619
No	4 (57.1)	3 (37.5)	
Yes	3 (42.9)	5 (62.5)	
Colostomy : N (%)			1.000
No	6 (85.7)	7 (87.5)	
Yes	1 (14.3)	1 (12.5)	
Cystostomy : N (%)			-
No	7 (100.0)	8 (100.0)	
Yes	-	-	

could resume their activities of daily life.

Focusing on immediate outcomes, significant factors for a successful *en bloc* sacrectomy in retrorectal tumors were: preoperative preparation, a comprehensive knowledge of sacral anatomy and biomechanics of lumbosacropelvic region, surgical technique and prophylactic procedures. As mentioned earlier, in

Table 5 Factors influencing estimated blood loss (EBL)

	EBL (≤ 2,000 cc.)	EBL (≥ 2,000 cc.)	P-Value
Tumor Pathology : N (%)			0.862
Chondrosarcoma	1 (33.3)	1 (8.3)	
Giant cell tumor	-	1(8.3)	
Chordoma	2 (66.7)	5 (41.7)	
MPNST	-	3 (25.0)	
GIST	-	1 (8.3)	
Leiomyosarcoma	-	1 (8.3)	
Tumor Size (cm3) : N (Median)	3 (1,200)	12 (1,200)	0.885
Sacral bone invasion : N (%)			1.000
No	2 (66.7)	8 (66.7)	
Yes	1 (33.3)	4 (33.3)	
Preoperative embolization : N (%)			0.371
No	1 (33.3)	1 (8.3)	
Yes	2 (66.7)	11 (91.7)	

Table 6 Number of patients and estimated blood loss in subtotal/total sacrectomy group, separate to non ligation and ligation of Internal Iliac Vessels group

	Non Ligate	Ligate	P-Value
Subtotal sacrectomy: N (Median EBL cc.)	4 (1,550)	2 (3,900)	0.133
Total sacrectomy: N (Median EBL cc.)	3 (15,000)	6 (5,500)	0.024*

order to provide a comprehensive surgical care in *en bloc* sacrectomy, a multidisciplinary team composing of general surgeon, orthopedist, urologist, plastic surgeon, nursing staff, physiotherapist, and psychologist should be involved to achieve an effective outcome.

Sacral resection is a challenging procedure and at the same time is also a worrisome operation for the involved surgeons because of 1) the complexity of anatomy and surgical technique, which is compulsory to perform both anterior and posterior approach; 2) the multiple potential complications including massive bleeding, associated organ injuries and sacral hernia; 3) degree of functional impairment; more or less, patients will experience anorectal and urogenital dysfunction which might need permanent colostomy or cystostomy; and 4) gait disturbance and remained lumbosacropelvic stability. When surgeons are aware of factors affecting surgical outcomes, then the possible complications could be prevented. It would be enormously beneficial for surgical performance, effectiveness and patient safety.

Among demographic data, tumor factors, preoperative factors and operative technique factors, the statistical analysis showed that the only single factor associated with severe complications leading to re-operation and death (Clavien-Dindo classification IIIb, IV, V) was operation types (subtotal/total sacrectomy, $p=0.041$), as listed in Table 4. Of 6 patients with subtotal sacrectomy, only 1 patient (16.7%) had severe complication while 7 of 9 patients (77.8%) who underwent total sacrectomy had severe complications. We concluded that subtotal sacrectomy was significantly safe and caused less severe complication comparing to total sacrectomy.

We evaluated the effects of tumor factors and preoperative embolization on intraoperative blood loss. Massive blood loss was defined as 50% of blood volume loss within 3 hours or a rate of blood loss more than 150 cc/min¹¹. We found that there was no factor significantly affecting intraoperative bleeding as shown in Table 5. However, when subgroup analysis was done affecting among operation types, the anterior approach with internal iliac vessels ligation in subtotal sacrectomy did not reduce intraoperative bleeding ($p=0.133$). On the other hand, for total sacrectomy, anterior approach with internal iliac vessels ligation significantly reduced the intraoperative bleeding ($p=0.024$) as shown in Table 6. Therefore, subtotal sacrectomy may not require anterior approach for internal iliac vessels ligation which is consistent with the report of various authors that subtotal sacrectomy can be safely performed with posterior approach alone^{12,13} while anterior approach with internal iliac vessels ligation should be conducted in all total sacrectomy to reduce intraoperative blood loss¹⁴⁻¹⁶.

When determining the correlation of protective colostomy and wound infection, wound infection could not be prevented by protective colostomy ($p=0.506$) listed in Table 7. However, two patients with protective colostomy had less severe postoperative wound infection that required no further re-operation for surgical debridement. We proposed that the small sample size may not allow us to see the significant statistical difference.

The preoperative ureteric stenting for prevention of ureteric injury was also reviewed. Preoperative ureteric stenting did not significantly decrease the rate of ureter injury, and could not prevent ureter injury, listed in Table 8. We suggest that because of the unfavourable

Table 7 Effect of colostomy on severity of wound infection

	Not severe	Severe	P-Value
Colostomy : N (%)			0.506
Not done	7 (77.8)	5 (100.0)	
Done	2 (22.2)	-	

Not severe = no need re-operation (Clavien-Dindo classification I, II, IIIa);
Severe = need re-operation (Clavien-Dindo classification IIIb, IV, V)

Table 8 Effect of ureteric stenting on incidence of ureter injury

	Ureter injury (No)	Ureter injury (Yes)	P-Value
Ureteric stenting			1.000
Not done	8 (61.5)	1 (50.0)	
Done	5 (38.5)	1 (50.0)	

miliar posterior approach position, surgeons should pay more attention in order to clearly identify ureters and conduct the operation cautiously.

In the subgroup analysis of operation types and functional outcomes, listed in Table 9, 10 patients experienced constipation, 6 were in subtotal sacrectomy group, 4 were in total sacrectomy group, and there was no significant difference between the groups ($p=0.182$). Nearly all of patients (14 from 15) had postoperative urinary retention. We concluded that urinary retention or constipation could occur regardless of operation types. In contrast, gait disturbance was associated with operative procedure. Patients undergoing subtotal sacrectomy tended to have less gait disturbance; 5 patients could walk without assistance and 1 patient could walk with assistance while all patients with total sacrectomy need external support (5 of 6 patients) or wheelchairs (3 of 3 patients) ($p=0.003$). Our findings supported the theory of subtotal sacrectomy in which patients would not lose the lumbopelvic ring stability, thus lumbopelvic reconstruction was not necessary. However, after the 2-4 year follow-up, our data showed that 75% of patients (6 of 8 patients) undergoing total sacrectomy whether received lumbopelvic reconstruction or not, was able to ambulate either with or without assistance. Our findings were consistent with the previous studies that total sacrectomy without lumbopelvic reconstruction could give favorable outcomes¹⁷⁻¹⁹. Wuisman et al.¹⁷ concluded that there was no significant difference in functional outcomes among two groups of lumbopelvic reconstruction

Table 9 Effect of operation type on functional outcomes

	Subtotal sacrectomy	Total Sacrectomy	P-Value
Constipation : N (%)			0.182
No	-	2 (40.0)	
Yes	6 (100.0)	4 (60.0)	
Urine retention : N (%)			0.429
No	1 (16.7)	-	
Yes	5 (83.3)	8 (100.0)	
Gait disturbance : N (%)			0.003*
Walk, no assistance	5 (83.3)	-	
Walk, with assistance	1 (16.7)	5 (62.5)	
Wheel chair	-	3 (37.5)	

performing or not performing. The explanation came from the evidence that after completely healed, ileolumbar muscle and ligament would form biologic sling and stabilize the spine which adequately function as weight-bearing joint for individual gait. There was one patient in our study who underwent total sacrectomy with successful lumbopelvic reconstruction, and had an excellent gait function outcome. The patient could walk without any assistance.

There was no report of sacral hernia among the studied group either during admission or after 2-4 years of follow-up. Regardless of postoperative wound infection and re-operative debridement (7 out of 11 patients), we proposed that sacral defect closure following sacrectomy could be adequately done by gluteus maximus approximation or gluteal advancement flap. Prosthesis sheath placement which might increase the rate of wound infection was not routinely necessary for all patients²⁰.

CONCLUSION

En bloc sacrectomy is a complex procedure which is the only chance of cure for retrorectal tumor. The operation requires multidisciplinary team of surgeons and other medical personnel. Even though it is an operation with various possible complications, the intermediate and long term outcomes are favorable, most of patients could resume their activities of daily life. The operation of choice depends on tumor size and location, when free margin resection is obtainable, subtotal sacrectomy is preferred. And when subtotal sacrectomy is a procedure of choice, posterior approach alone can be safely performed. On the other hand, for

all total sacrectomy, anterior approach should be performed for intraoperative internal iliac vessels ligation to decrease intraoperative blood loss. Although protective colostomy does not decrease the incidence of surgical wound infection but the severity tends to have less virulent. The evidence showed that preoperative ureteric stenting could not prevent ureteric injury. In addition, there were no difference in postoperative urinary-bowel function between patients with subtotal sacrectomy and total sacrectomy while patients underwent subtotal sacrectomy had less gait disturbance compared to total sacrectomy. We also suggest that in total sacrectomy, instrument placement for lumbopelvic reconstruction is not always necessary, because most of patients without lumbopelvic reconstruction had favorable outcomes after the intermediate to long term follow-up. Lastly, sacral defect after sacrectomy could be closed with gluteus maximus approximation or gluteal advance flap without prosthesis sheath placing.

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