

Complications, secondary interventions and long term morbidity after *en bloc* sacrectomy

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Abstract

Purpose *En bloc* sacrectomy is a demanding surgical procedure to remove tumors from the sacrum. Comprehensive data on readmissions for complications endured months to years after initial discharge are scant. The purpose of this study is to present the long-term complications, readmissions and secondary interventions for patients having undergone *en bloc* sacrectomy.

Methods Patients were included if *en bloc* sacrectomy and follow-up were conducted in the authors institution. Correspondence from all specialties involved in the treatment of patients was retrieved. Predefined parameters were scored and assigned to five distinct phases: diagnostic phase; surgery; postoperative period to 1 year after surgery; second year after surgery until follow-up and last follow-up.

Results Sixteen patients underwent anterior–posterior *en bloc* sacrectomy for a locally aggressive tumor ($n = 2$); malignant tumor ($n = 13$) or solitary metastasis ($n = 1$). The type of resection was low ($n = 1$); middle ($n = 3$); high ($n = 4$); total ($n = 3$) and hemisacrectomy ($n = 5$). The median surgical duration was 12.7 h and median blood lost was 12 l. A total of 73 major complications (average

per patient 5; median 4; range 0–12) were recorded and 73 secondary interventions (average per patient 5; median 5; range 0–11) were performed in the first year postsurgery. From the second year until follow-up complications and secondary interventions markedly decreased. At final follow-up (65–266 months), considerable morbidity was found for the eleven patients still alive.

Conclusions *En bloc* sacrectomy is a procedure with a high rate of major complications, regardless of tumor histology, often necessitating readmissions and secondary interventions. Long-term survival is associated with considerable morbidity and extensive preoperative counseling should be conducted to discuss the risks and outcome of the procedure.

Keywords Sacrectomy · Complications · Long-term outcome · Tumor · Secondary intervention

Introduction

Sacrectomy is a surgical procedure to remove primary tumors and solitary metastases from the sacrum [1]. Benign tumors may be excised intralesionally while malignant tumors are preferably removed completely with a shell of normal tissue (*en bloc*) to reduce the incidence of local recurrence and distant metastasis [2]. Sacral tumors often grow largely unnoticed and are diagnosed only after long-standing pelvic pain and/or loss of neurological function [3]. Radical and wide resections of large pathological processes from the sacrum are usually not feasible without including critically important structures such as the rectum, sacral nerves and internal iliac vessels [4]. Since radical and wide resections carry the risk of sexual dysfunction and irreversible loss of control over lower extremities,

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bladder and rectum, most sacrectomies aim for marginal resection and preservation of function [4]. A recent study found wide and marginal resections lead to similar oncological outcomes for chordomas and chondrosarcomas in adult patients [2]. Regardless of the type of resection, *en bloc* sacrectomy remains a highly demanding surgical procedure fraught with complications but without an entirely predictable oncological outcome [5–7]. Therefore, when contemplating surgery, patients and surgeons need to balance risks/benefits of the procedure based on all available data. Although several studies have reported the surgical details, postoperative complications and oncological results at follow-up, comprehensive data on readmissions for complications endured months to years after initial discharge are scant [4]. In the present study with a median follow-up of 114 months, the complications, readmissions and secondary interventions for a group of sixteen patients having undergone sacrectomy for a variety of malignant or locally aggressive tumors were investigated.

Patients and methods

This study was approved by our Institutional Review Board. The databases from the Departments of Surgical Oncology and Orthopedics were reviewed and patients were included if *en bloc* sacrectomy and follow-up were conducted in the authors institution. The correspondence from all specialties involved in, or consulted during, the treatment of patients from diagnosis to final follow-up (or death) was retrieved by two independent investigators from the central electronic patient record system. Correspondence included reports on diagnostic workup; surgical reports; intensive care unit (ICU) records; clinical discharge reports; notes from consulting in-hospital specialties; scheduled outpatient follow-up reports; notes from outpatient consulting specialties and notes from supporting specialties (e.g. psychiatry; behavioral science; pain consultation). Additional information was obtained from the departments of radiology; nuclear medicine; anesthesiology; microbiology and pathology. Predefined parameters scored from all patient records were subsequently assigned to one of five distinct phases: (1) diagnostic phase; (2) surgery; (3) postoperative period to 1 year after surgery; (4) second year after surgery until follow-up; (5) last follow-up (last information added July 2013). See Table 1 for a listing of the parameters and definitions.

Diagnostic workup, surgical technique and follow-up

After completion of the diagnostic workup, the type of tumor was known for all patients; metastatic spread was

excluded by computed tomography and nuclear imaging; MRIs were available (in combination with CT and radiographs) for surgical planning and informed consent was obtained. Except for one patient, all sacrectomies were planned and executed as sequential anterior–posterior procedures by a surgical team consisting of an oncological surgeon, neurological surgeon and orthopedic surgeon, the latter always present if spino-pelvic reconstruction was performed. In two patients, the combined procedure was performed over 2 days (one patient staying in the ICU for 2 days between the procedures; the other patient receiving spino-pelvic reconstruction 2 weeks after resection); the other patients had their surgery completed the same day. The patient not receiving anterior–posterior surgery underwent a lateral approach for a hemisacrectomy. As part of a standardized protocol, intravenous antibiotics were administered at least 1 h prior to incision, repeated every 4 h during surgery and continued for 5 days. Patients were actively warmed during the procedure to maintain body temperature; fluid homeostasis was monitored and depletion of blood components corrected. The anterior approach was used to dissect ureters, great vessels (including tributaries to the tumor) and rectum from the anterior surface of the sacrum. Additionally, the omentum was prepared and moved to the pelvic cavity for closure of the sacral defect during the posterior phase. A vertical rectus abdominis myocutaneous (VRAM) flap was prepared in one case when a very large sacral defect was anticipated [8]. During the posterior approach, the biopsy tract was included in the resection and wide margins were pursued. However, marginal resection planes were accepted when deemed inevitable to preserve neurological function. Neurophysiological monitoring was not available at the time of this series. Reconstruction with pedicle/sacral/iliac screw and rod systems, supplemented with cages if necessary, was performed for total and high sacrectomies and also for hemisacrectomies involving the S1 vertebral body. Postoperatively, almost all patients recovered at the ICU during one or more days before returning to the surgical ward. Air-fluidized beds were used as indicated by the surgical team. Nutritional status was monitored during the entire admission period and supplementation initiated through enteral or parenteral routes when necessary. Dedicated wound care nursing teams assisted in optimizing healing of surgical wounds during the postoperative phase. Analgesics (acetaminophen/NSAIDs/opioids) were administered as required combined with neuropathic analgesics for long-term effect, if necessary. After discharge, patients were scheduled for regular outpatient visits (three visits per year for the first 2–3 years and annual visits thereafter). During these visits, appropriate imaging and laboratory test results were obtained for surgical, oncological and orthopedic follow-up.

Table 1 Parameters extracted from correspondence and notes

Diagnostic phase	Definition
Gender	Male/female
Age	Years
ASIA score	A/B/C/D/E according to American Spinal Injury Association
Urinary function	Intact/impaired
Location/expansion tumor	Sacral level; unilateral/bilateral
Radiotherapy preoperative	Yes/no
Chemotherapy preoperative	Yes/no
Surgery	
Surgical approach	Anterior–posterior/lateral; one stage/two stage
Classification of resection	Low/middle/high/total/hemisacrectomy according to Fournery et al. [4]
Sacroiliac joint resection	No/left/right/bilateral
Nerve root(s) resection	None/L5/S1/S2/S3/S4/S5; left/right/bilateral
Intraoperative tumor spill	Yes/no (as noted by surgeon)
Spino-pelvic reconstruction	None/pedicle screws and rods/pedicle screws and rods and cage
Closure technique	None/omentoplasty/vertical rectus abdominis myocutaneous flap
Colostomy	Yes/no
Suprapubic catheter	Yes/no
Operation duration	Hours
Blood lost	Liters
First year after surgery	
Days in intensive care unit	Total number of days in intensive care unit during first year after surgery
Days in ward	Total number of days in surgical ward during first year after surgery
Margins of resection	Free of tumor/contaminated
Primary culture of wound	As described in medical microbiologist reports
Adjuvant radiotherapy	Yes/no
Adjuvant chemotherapy	Yes/no
Minor complications	Urinary tract infection/any miscellaneous complication not prolonging hospital stay or requiring readmission and/or secondary intervention
Major complications	Superficial wound infection/deep wound infection/delayed wound healing/respiratory infection/central line associated bloodstream Infection/pulmonary embolism/deep venous thrombosis/critical illness polyneuropathy/sacral herniation/delirium/any miscellaneous complications prolonging hospital stay; requiring readmission and/or secondary intervention
Secondary interventions	All interventions requiring hospital admission and (a type of) anesthesia
Second year to follow-up	
Minor complications	See corresponding section in ‘First year after surgery’
Major complications	See corresponding section in ‘First year after surgery’
Secondary interventions	See corresponding section in ‘First year after surgery’
Follow-up (per July 2013)	
Survival	Yes/no
Local recurrence	Yes/no
Distant metastasis	Yes/no
Survival since surgery	Months
Disease-free survival	Months
Asia score	A/B/C/D/E according to American Spinal Injury Association
Mobility	Walk unassisted/walk with crutch or walker/wheelchair bound
Bowel function	Normal/colostomy/incontinence
Urinary function	Normal/self-catherisation/indwelling catheter/incontinence
Erectile function (males)	Normal/impaired
Pain clinic consultation	Yes/no

Table 2 Diagnostic phase

No	M/F	Age	Biopsy	Diagnosis	Planned approach
1	F	39	Open	Chondrosarcoma	Lateral
2	M	48	Percutaneous + open	Chondrosarcoma	Anterior–posterior
3	M	62	Percutaneous	Chordoma	Anterior–posterior
4	M	38	Percutaneous	Chordoma	Anterior–posterior
5	M	65	Unknown	Chondrosarcoma	Anterior–posterior
6	M	58	Percutaneous	Chordoma	Anterior–posterior
7	M	74	Unknown	Chordoma	Anterior–posterior
8	F	63	Percutaneous	Chordoma	Anterior–posterior
9	M	48	Open	Malignant peripheral nerve sheath tumor	Anterior–posterior
10	F	38	Open	Hemangioblastoma	Anterior–posterior
11	M	48	Percutaneous	Chordoma	Anterior–posterior
12	M	30	Percutaneous	Chondrosarcoma	Anterior–posterior
13	F	24	Percutaneous + open	Schwannoma	Anterior–posterior
14	M	62	Percutaneous	Chordoma	Anterior–posterior
15	M	52	Percutaneous	Solitary metastasis renal cell carcinoma	Anterior–posterior
16	F	30	Percutaneous	Chondrosarcoma	Anterior–posterior

M male, F female

Results

A total of sixteen patients, eleven males and five females aged between 30 and 74 years (average 49 years; median 48 years) were identified having undergone *en bloc* sacrectomy. Except for one patient (case 1) all were operated using an anterior–posterior approach between December 2001 and September 2009.

Diagnostic phase

The most frequent symptom leading to diagnosis of the sacral tumor was persisting pain in the lower back or pelvis and/or radicular pain (14/16 patients). Three patients presented with mild neurological deficits of the lower extremities (all ASIA D) at admission; one patient had partially lost control of the urethral sphincter; the other thirteen patients were neurologically intact. Six patients were referred to our center after biopsy was performed elsewhere. The histological diagnoses of the sacral tumors were chordoma ($n = 7$); chondrosarcoma ($n = 5$); schwannoma ($n = 1$); malignant peripheral nerve sheath tumor (MPNST) ($n = 1$); hemangioblastoma in Hippel–Lindau disease ($n = 1$) and solitary metastasis of renal cell carcinoma ($n = 1$). Table 2 lists the characteristics, diagnosis and planned surgery for individual patients. No patient received chemotherapy prior to surgery. One patient had received nephrectomy; radiotherapy and re-irradiation of the sacrum; multiple embolizations and surgical debulking of his sacral metastasis of renal cell carcinoma, all performed more than a year before an attempt was made at *en bloc* sacrectomy.

Surgery

Resections were classified according to Fourney et al. [4] as low ($n = 1$); middle ($n = 3$); high ($n = 4$); total ($n = 3$) and hemisacrectomy ($n = 5$). The left sacroiliac joint was resected in six patients, in three patients both sacroiliac joints were included in the specimen and in seven patients the sacroiliac joints could be preserved. Except for one case, two or more sacral roots had to be cut to obtain tumor-free margins. Tumor spill occurred twice; in one case the tumor was accidentally opened just before removal and in another case the *en bloc* specimen fragmented during removal. In the latter case, a re-resection of the contaminated margin was performed and frozen sections created from the newly created margin were shown to be free of tumor histologically. Spino-pelvic reconstruction was performed in four patients with screws and rods only; cages were used in addition to screws and rods in eight patients (see also Fig. 1a–c). The other four patients (with low and middle sacrectomies) did not require reconstruction. Permanent colostomies were created in ten patients; in two other patients the colostomy was scheduled for reversal of 1 year after surgery. Closure was facilitated by omentoplasty in fourteen patients (one had additional VRAM), in one patient the omentum could not be retrieved from the pelvic cavity during the posterior phase and in one patient closure could be achieved without additional wound closing techniques. The median operation duration was 12.7 h (range 5, 1–17, 7 h) and median blood loss was 12 l (range 3–39 l). Table 3 lists the details of surgery.

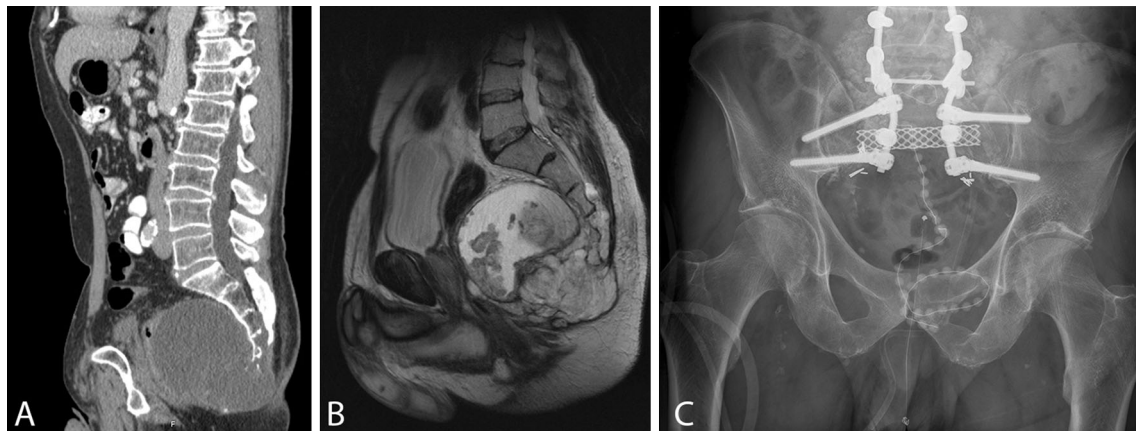


Fig. 1 a–c Radiographical examinations from a 62-year-old male (case 14) with a sacral tumor showing: **a** preoperative midsagittal computed tomography image of a sacral chordoma, **b** preoperative midsagittal slice of a T2-weighted MR image of a sacral chordoma,

c pelvic radiograph 2 weeks after high sacrectomy and reconstruction with pedicle screws, rods and cage (also note the presence of gentamycin beads for local antibiotic treatment after recent debridement for wound infection)

Postoperative period to 1 year after surgery

Fifteen patients recovered on the ICU for a median of 5 days (average 13 days, range 1–58 days) before returning to the ward. Following pathological examination, the margins of the resected specimens were declared free of tumor in eleven cases. The five contaminated specimens included the two cases where tumor spill was noted intraoperatively. Three patients with contaminated resection margins received additional radiotherapy and/or chemotherapy. During the first year after surgery, patients stayed in the hospital for a median of 65 days (average 92 days, range 29–295 days). Note: this number includes the days the patients were readmitted for the treatment of major complications. Minor complications (mainly urinary tract infections) occurred in nine patients; these complications had no repercussion on length of hospital stay, number of readmissions or secondary interventions. A total of 73 major complications occurred resulting in an average of five major complications (median 4; range 0–12) for individual patients in the year following surgery (see also Table 4). The major complications occurring most often were complicated wound healing and wound infection, together affecting 12 patients, although delirium, incisional (sacral) herniation and postoperative hemorrhage were also frequently noted. A total of 73 secondary interventions were performed in the first year after surgery resulting in an average of five secondary interventions (median 5; range 0–11) per patient. The vast majority of secondary interventions ($60/73 = 82\%$) were necessary to treat complicated wound healing and deep infection ($n = 49$); to control postoperative hemorrhage ($n = 7$) or to create tracheostomies in patients with prolonged (≥ 30 days) stay on the ICU ($n = 4$). During this phase, local recurrence (or more likely ‘progression of the initial tumor’) and distant

metastasis developed in one patient 3 months after surgery for a chondrosarcoma complicated by intraoperative tumor spill (case 16); this patient died 8 months later of her disease.

Second year after surgery until follow-up

Minor complications (‘multiple’ urinary tract infections) were reported for only one patient in this period. However, 32 major complications (range 0–7 complications per patient) were reported involving fourteen patients (Table 4). Note that median and average values are not presented here due to ongoing cohort attrition during this phase. Urosepsis ($n = 5$) and pathological fractures of lower extremities ($n = 5$) were the most frequently observed complications followed by insufficiency fractures/pseudarthrosis of the remaining sacral segments ($n = 4$) and hydronephrosis ($n = 3$). A total of 39 secondary interventions, mostly of urological nature, were performed (range 0–15 secondary interventions per patient): 13 interventions were needed for placement or revision of nephrostomy catheters; two for treatment of hydrocele and one for ureter reimplantation. A further six surgical interventions were required for revision of the spino-pelvic reconstruction ($n = 4$) and to treat proximal femur fractures ($n = 2$). No obvious correlations were noticed between the (Fourney) level of resection or type of instrumentation used and complications/secondary interventions. A marked decrease in average number of complications and secondary interventions was observed from the first to tenth year after surgery even when corrected for attrition (see also Fig. 2). During this phase, four patients developed local recurrences followed by distant metastases and all died of their disease after living an average period of 21 months (range 12–27 months) without evidence of disease.

Table 3 Surgical details

No	Sacrectomy type	Resected SI	Resected nerve root	Spill	Reconstruction	Duration	BL
1	Hemi S1–S2	Left	S1–S2 left	Yes	No	5,1	NA
2	High	Left	S3–S4 left	No	Screws/rods	9,8	14
3	Middle	No	None	No	No	8,4	3
4	Middle	No	S3–S5 bilateral	No	No	15,6	12
5	Total	Left	S1–S3 left, S3 right	No	Screws/rods/cage	12,3	13
6	Middle	No	S3–S5 bilateral	No	No	13,6	14
7	High	No	S2–S5 bilateral	No	Screws/rods/cage	13,0	10
8	High	Bilateral	S1–S5 bilateral	No	Screws/rods/cage	13,0	8
9	Hemi S1–S2	Left	S1–S2 left	No	Screws/rods	9,1	20
10	Hemi S1–S2	No	S1–S2 left	No	Screws/rods	9,4	18
11	Low	No	S4–S5 bilateral	No	No	16,0	9
12	Total	Bilateral	S1–S5 bilateral	No	Screws/rods/cage	11,6	6
13	Hemi S1–S3	Left	S3–S5 left	No	Screws/rods/cage	11,9	9
14	High	No	S3–S5 bilateral	No	Screws/rods/cage	14	21
15	Total	Bilateral	S1–S5 bilateral	No	Screws/rods/cage	16	39
16	Hemi S1–S5	Left	L5–S5 left	Yes	Screws/rods/cage	17,7	7

Resected SI resection of sacroiliac joint, *Spill* intraoperative tumor spill, *BL* blood loss, *NA* not available

Last follow-up

At the last follow-up, eleven of the sixteen patients were alive; eight patients had no evidence of disease (median follow-up 114 months, range 65–266 months) and three patients survived with distant metastases (median follow-up 97 months, range 94–107 months). The results below are for these eleven patients. Six patients were able to walk without walking aids (despite ASIA D neurological status in three); two patients walked with crutches (both ASIA D) and three patients were wheelchair bound (two patients with ASIA C and one patient with ASIA E, the latter having undergone Girdlestone resection arthroplasty after pathological destruction of his proximal femur). Except for one patient who presented with ASIA D in the diagnostic phase (and remained ASIA D after surgery) all other patients alive had been neurologically intact before surgery. Two patients had normal urinary function; the other nine patients relied on indwelling catheters ($n = 6$) or were incontinent ($n = 3$). Five patients had normal bowel function (two after reversal of colostomy); five patients had a permanent colostomy and one patient was incontinent for feces. Six out of eight male patients experienced permanent erectile dysfunction; in one patient (with unilateral S3–S4 nerve root transection) erectile function was normal and in one patient (with bilateral transection of the S3 nerve root) it was not reported. Five patients frequented the outpatient pain consultation clinic while six patients regulated their use of analgesics independently. The average number of major complications for patients alive at follow-up was seven (median 7; range 1–19); the average number of secondary interventions was also seven (median 3; range 0–26). See also Table 5 for a listing of the outcome at last

follow-up. Analysis of eight important dichotomous domains of outcome (free of disease: yes/no; neurological deterioration: yes/no; urinary function: normal/impaired; bowel function: normal/impaired; sexual function: normal/impaired; pain: yes/no; major complications: yes/no) showed that patients had, on average, abnormalities in 4, 7 domains (median 5; range 2–7) at follow-up. No patient could, therefore, be considered burden free.

Discussion

In this study the complications, secondary interventions and long-term morbidity were investigated for a group of sixteen patients who underwent *en bloc* sacrectomy. The main finding was that, regardless of tumor histology, major complications were endured by all patients and almost all patients were subjected to two or more secondary interventions. The morbidity of patients alive at a minimum of 5 years follow-up was substantial. No patient achieved the status of being free of disease without significant loss of function in the absence of pain. Based on our results, it seems that the surgical intervention ‘*en bloc* sacrectomy’ represents an even more demanding procedure, from the standpoint of patients, than previously thought.

In a recent prospective study by Street and coauthors, adverse events (defined as any untoward occurrence in a patient; major and minor, medical and surgical) were demonstrated to be grossly underreported in 942 consecutive patients undergoing major spine surgery [9]. Out of the five postoperative adverse events with the highest incidence reported in their study (electrolyte imbalance; medication related; constipation/ileus; nausea; urinary tract

Table 4 Number of days admitted; major complications and secondary interventions

No.	Adm	Major complications 1st year	SI	Major complications 2nd year to FU	SI
1	0 + 68		0	Pseudarthrosis bone graft; incisional herniation	0
2	3 + 102	Superficial wound infection; deep wound infection; complicated wound healing	8	Persisting fistula sacrum	1
3	1 + 135	Postoperative hemorrhage; superficial wound infection; deep wound infection; complicated wound healing; recto-cutaneous fistula	10	Urosepsis; psychosis	1
4	2 + 29	Superficial wound infection; deep wound infection; complicated wound healing; incisional herniation	1	Seroma; multiple urinary tract infections; insufficiency fracture sacrum	5
5	18 + 35	Respiratory infection; pulmonary embolism; delirium; meningitis	1	Pathological fracture proximal femur; hydrocele	2
6	58 + 99	Postoperative hemorrhage; blindness OD; hemiparalysis vocal cord; superficial wound infection; deep wound infection; respiratory infection; CLABSI; pulmonary embolism; delirium; critical illness polyneuropathy; incisional herniation; phlebitis	6	Periarticular ossification hips; cardiotoxicity after chemotherapy; insufficiency fracture sacrum; urosepsis (2x); pathological fracture proximal femur; hydronephrosis	5
7	30 + 154	Superficial wound infection; deep wound infection; complicated wound healing; respiratory infection; delirium; incisional herniation	11		0
8	5 + 85	Superficial wound infection; deep wound infection; complicated wound healing; delirium; incisional herniation	6	Pathological fracture proximal femur	0
9	18 + 44	Postoperative hemorrhage; deep wound infection; CLABSI; deep venous thrombosis; delirium; critical illness polyneuropathy	2	Pathological fracture tibial plateau; hydrocele	1
10	3 + 26	Complicated wound healing	0		0
11	6 + 27	Superficial wound infection; deep wound infection; complicated wound healing; incisional herniation	0	Recurrence incisional herniation; isolated lung metastasis (2x)	3
12	2 + 60	Incisional herniation	5	Progressive neurological deficit	0
13	1 + 38	Insufficiency fracture sacrum	1	Pseudarthrosis pelvis	1
14	7 + 65	Superficial wound infection; deep wound infection; CLABSI; delirium	6	Hydronephrosis; herniation colostomy	5
15	50 + 245	Superficial wound infection; deep wound infection; complicated wound healing; respiratory infection; CLABSI; delirium; critical illness polyneuropathy; meningitis; acute respiratory distress syndrome; construct failure; retained gauze	11	Recurrence construct failure (2x); urosepsis (2x); hydronephrosis	15
16	4 + 46	Superficial wound infection; deep wound infection; complicated wound healing; persisting CSF leakage; infection port-a-cath; pathological fracture femur condyle	5	Patient died in first year after surgery	

Adm number of days admitted in the first year after surgery (days on intensive care unit + days in surgical ward), *SI* secondary intervention, *FU* follow-up, *OD* oculus dexter (right eye), *CLABSI* central line associated bloodstream infection, *CSF* cerebrospinal fluid

infection, in that order) only urinary tract infections were noted in our cohort and subsequently recorded as ‘minor complications’. It should be noted that their series did not comprise patients undergoing sacrectomies. Considering the characteristics of *en bloc* sacrectomy surgery (massive transfusion; large wound bed; intra-abdominal access; high-dose opioid administration; admission to ICU) the absence of the other four adverse events during admission of our patients seems unlikely. As our study is

retrospective, it is possible that some adverse events have been regarded as clinically inconsequential and, as a result, were not reported in medical correspondence [9].

Most clinical articles reporting adverse events or complications are retrospective in nature and comparison of data may be hampered by dissimilarities in reporting. For instance, some authors describe the extent of resection performed rather than using a staging system such as proposed by Fourny et al. [4]. In our series, we did not

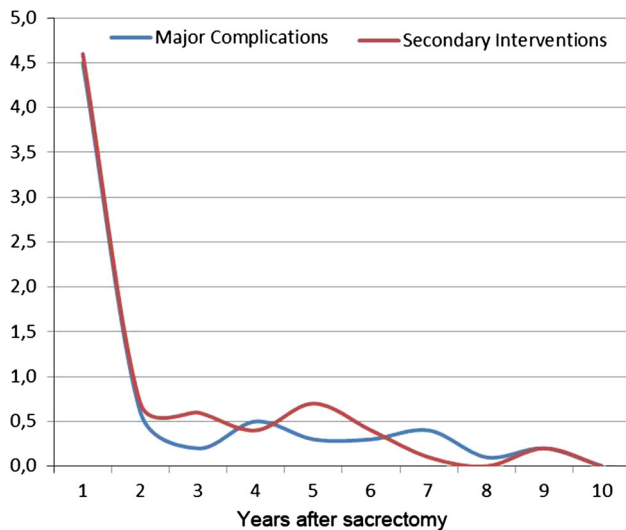


Fig. 2 Graphical representation of the mean number of major complications and secondary interventions per patient in the years following surgery (corrected for patient attrition during follow-up)

observe postoperative deaths; however, the abundance of complications has been impressive. The major complications with the highest incidence were complicated wound healing and deep infection, affecting 12 patients (75 %). Factors previously shown to contribute to surgical site infections after spine surgery include large blood loss (>2 l); transfusion of red blood cells/serum/autologous blood; prolonged operation duration (>5 h); sacral involvement; surgery for tumor resection; presence of instrumentation; posterior only or anterior–posterior approach; intra-abdominal procedure; bowel reconstruction and number of people scrubbed in [10–16]. Almost without

exception, all of these factors will be experienced by patients undergoing anterior–posterior *en bloc* sacrectomy [17]. Nonetheless, the incidence of wound related complications in our series (75 %) compares unfavorably with previously published studies reporting rates of 5/29 = 17 %; 18/46 = 39 %; 23/52 = 44 %; 9/20 = 45 %; 19/42 = 45 %; 8/16 = 50 % [4, 6, 16, 18, 19]. A major factor contributing to the occurrence of (early onset) complications may have been the amount of blood lost during surgery (our series: mean 13, 5 l, range 3–39 l). Hulen et al. [6] reported a mean volume of 5 l (range 1, 5–8 l) of blood lost after anterior–posterior sacrectomy and Ruggieri et al. [7] reported a mean blood volume of 4 l (range 2–22 l) lost after surgical resection of sacral chordomas (66 % of these procedures were anterior–posterior procedures). The large variation (range) of blood loss in our study and others, suggest a multitude of factors to contribute including duration of surgery; extent of osteotomy; tumor vascularization; intraoperative coagulation and attention to hemostasis [4]. However, also the median blood loss (12 l) from our series does seem higher than previously published. Another factor contributing to wound infections may have been the number of surgeons from various specialties (general/neurological/orthopaedic/plastic) scrubbing in during the procedure [11, 16]. *En bloc* sacrectomy will typically require involvement of a number of disciplines and the associated increase in traffic in the operating theater has shown to negatively affect infection rates. Reducing the number of door openings and implementation of smart strategies for storage and handling of instruments/implants in the operating theater may help prevent surgical site infections [20, 21]. To further prevent wound healing complications, generally good results have

Table 5 Outcome at last follow-up

	No	MC	SI	ASIA	Mobility	Urinary function	Bowel function	ED	Pain	FU	Status
	1	2	0	D	Crutch	Incontinent	Incontinent	NA	No	266	NED
	2	4	9	E	Unassisted	Incontinent	Normal	No	No	139	NED
	3	7	11	E	Unassisted	Normal	Normal	Yes	No	125	NED
	4	7	6	D	Unassisted	Suprapubic cath	Colostomy	Yes	Yes	117	NED
	5	6	3	D	Crutch	Suprapubic cath	Colostomy	Unkn.	Yes	110	NED
	6	19	11	E	Wheelchair	CIC	Colostomy	Yes	Yes	107	AWD
	7	6	11	D	Crutch	Suprapubic cath	Colostomy	Unkn.	Yes	36	DOD
	8	6	6	D	Crutch	Suprapubic cath	Colostomy	NA	No	67	DOD
	9	8	3	C	Wheelchair	CIC	Normal	Yes	Yes	98	NED
	10	1	0	D	Unassisted	Normal	Normal	NA	Yes	97	AWD
	11	7	3	E	Unassisted	CIC	Colostomy	Yes	No	94	AWD
	12	2	5	D	Unassisted	CIC	Colostomy	Unkn.	Yes	18	DOD
	13	2	2	D	Unassisted	CIC	Normal	NA	Yes	84	NED
	14	6	11	E	Unassisted	Suprapubic cath	Colostomy	Unkn.	Yes	36	DOD
	15	16	26	C	Wheelchair	Incontinent	Colostomy	Yes	Yes	65	NED
	16	6	5	D	Wheelchair	CIC	Colostomy	NA	Yes	11	DOD

The cells with gray shading represent patients succumbing to their disease before reaching last follow-up (July 2013)

MC total number of major complications, SI total number of secondary interventions, ASIA american spinal injury classification, cath catheter, CIC clean intermittent (self-)catheterization, ED erectile dysfunction, NA not applicable, unkn. unknown, FU follow-up, NED no evidence of disease, AWD alive with disease, DOD died of disease

been obtained using VRAM flaps to cover sacral defects [22–24]. As VRAM flap reconstruction was a relatively new technique at the time the current series was performed, omentoplasty was relied on to facilitate haemostasis; help sealing of the dural sac after root resection and assist in closure of the wound [8, 25].

Recent publications have shown a posterior-only approach to be feasible for *en bloc* sacrectomy provided the rectum and iliac vessels are spared from tumor invasion and the tumor does not extend beyond the lumbosacral junction [26, 27]. The two main advantages of this approach over combined anterior–posterior procedures are shorter surgical duration and reduced blood loss; factors strongly associated with the incidence of surgical site infection. Lastly, sacral tumors are rare and come in a variety of shapes, size, location, invasion of surrounding structures and biological behavior [1, 28]. As a consequence, gaining ample experience with (and avoiding the pitfalls of) resection of these tumors is a tedious undertaking even for academic quaternary referral centers and a prolonged learning curve must be anticipated [29].

A remarkable difference could be observed for the type of major complication occurring during the first year and years thereafter. In the first year most major complications were, unsurprisingly, directly related to the surgical procedure and postoperative phase. The complication most often observed within 1 or 2 days of surgery was, for example, postoperative hemorrhage ($n = 3$) while complications observed days to weeks after surgery were predominantly related to infection ($n = 33$). However, from the second year onward, complications associated with the functional effects of sacrectomy began to surface. The (partial) loss of urinary function resulting from transection of S3 root(s) probably led to the multiple cases of urinary tract infections, urosepsis and occurrences of hydronephrosis [30, 31]. As a consequence, the majority of secondary interventions ($16/39 = 41\%$) in the later phase was performed by urologists. Pathological fractures of the lower extremities occurred probably as a result from disuse osteoporosis due to impaired mobility and/or due to post-irradiation effects [32–34]. Sacral insufficiency fractures/pseudarthrosis ($n = 4$) and implant failures ($n = 2$) also started to emerge in the second year after surgery and onwards. Based on these results, we suggest the type of complications after *en bloc* sacrectomy to slowly shift from spinal, oncological and plastics to urological and orthopedic during the course of follow-up. Although a relation between tumor histology and clinical outcome may very well exist, the current number of patients in this series precluded in-depth analysis of such a relationship and clear outliers based on histology were not observed. Similarly, a relationship may exist between sacrectomy type (total, hemisacrectomy, high, middle, low) and clinical outcome

in terms of complications and secondary interventions. Patients undergoing hemisacrectomy had the lowest number of complications (on average four complications versus six complications for the ‘high’ and ‘low’ groups and eleven for the ‘middle’ group) and lowest number of secondary interventions (two secondary interventions for the hemisacrectomy group versus nine secondary interventions for the ‘middle’ and ‘high’ groups). These differences were, due to large variance of parameters, however, not statistically significant.

One patient (case 15 with solitary renal cell carcinoma metastasis and previous radiotherapy, multiple embolizations and debulking surgery) stood out due to a very unfavorable starting point before surgery with respect to obtaining tumor-free margins and avoiding wound healing complications. Only after thorough patient counseling, it was decided to perform *en bloc* sacrectomy. This patient had the largest amount of blood lost during surgery (39 l); the longest hospital admission period (295 days in the first year postsurgery); most secondary interventions both in the first year (11 interventions) and thereafter until follow-up (15 interventions). This case serves to illustrate the importance of (multidisciplinary) planning of a definitive treatment strategy before initiation of any intervention in patients with sacral tumors.

To our best knowledge, studies presenting patient reported outcome measures after *en bloc* sacrectomy do not exist. A qualitative assessment of patient experiences following sacrectomy was published in 2010 by Davidge and coworkers [35]. After interviewing twelve patients (six males, six females between 32 and 82 years of age) the study concluded that sacrectomy was a life-changing event for patients and their families. Although all underwent preoperative consultation, patients felt underinformed (or, more accurately, they felt overwhelmed) before undergoing sacrectomy especially with regards to the long-term consequences [35]. Despite the impact sacrectomy had on their lives, all participants were unanimous in expressing their gratitude at still being alive. Appreciating the results from the above work and recognizing many aspects, we feel that the outcome of the present study further underlines the need for extensive preoperative counseling, preferably in the presence of relatives/friends, to reach an informed and deliberate decision to proceed with (or decline) this type of surgery. Although surgical resection is the standard treatment for most sacral tumors, advanced irradiation techniques including intensity modulated radiation therapy (IMRT), robotic radiation systems (Cyberknife[®]) and proton-beam/carbon-ion therapy, are increasingly being used as viable alternatives to surgical resection when the latter treatment cannot be performed without gross loss of function or unacceptable perioperative morbidity/mortality [36].

In conclusion, *en bloc* sacrectomy is a seriously demanding surgical procedure with a high rate of major complications often necessitating readmissions and secondary interventions. Long-term survival is associated with considerable morbidity and extensive preoperative counseling should be conducted to discuss and contemplate many risks and not entirely predictable outcome of the procedure.

Conflict of interest None.

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