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Clinical course and prognosis of patients with sacral tumours treated with surgery. A case series

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Abstract

Background: The sacrum is an infrequent localization of tumours. Despite the modern spinal surgical techniques, sacral tumours still are an important challenge for surgeons. This study aimed to describe the clinical course and prognosis of surgically treated patients with sacral tumours.

Material and methods: A retrospective analysis from patients with sacral tumours seen from 2011 to 2017 at the National Cancer Institute, Mexico is presented. Multiple clinical variables were evaluated.

Results: A total of 13 patients were included in the study. The most common primary histologic type was chordoma, followed by chondrosarcoma and giant cell tumours.. The mean follow-up time was 51.4 months, mean progression-free survival was 31.6 months, 54% of the patients had complications related to surgery. At the end of the study, 54% of patients were still alive, with a mean overall survival of 51.3 months.

Conclusion: Sacral tumours are a complex disease with a high surgical complication rate, elevated recurrence, and frequent neurologic and functional sequelae. We present a case series of 13 patients surgically treated with a 54% complication rate.

Abbreviations: ST-Sacral Tumours; QT-Chemotherapy; RT-Radiotherapy; PFS-Progression-free survival; OS-Overall surveillance; FT-Following time; SE-Standard error; ILR-Iliolumbar reconstruction; OS-Overrall NA; WI-Wound infection; FT-Following time; SD-Standard deviation; CV-Coefficient of variation

Introduction

Sacral tumours (ST) are infrequent neoplasms with an incidence from 1–7 % of the spinal tumours [1]; ST may have benign or malignant behaviour, and the treatment is complex due to their elevated morbidity and mortality. Most of the time, the diagnosis is difficult and late by its vague and nonspecific course. Among the malignant ST, chordoma is the most frequent, followed by chondrosarcoma and giant cell tumour [2]. ST with metastatic origin mainly from breast, lung, or prostate tumours [3]. Due to the malignant behaviour of sacral tumours in its majority, it is well known that the natural history of sacral tumours is disease progression in a short time with the involvement of iliac bones, pelvis, abdominal viscera, and femur and all of this diminishes global survival, with a great impact in life quality during this survival period.

To relieve pain, improve nerve function, control local tumours, and improve quality of life for patients, surgery is widely performed, including minimally invasive surgery, palliative surgery, or radical surgery. Due to its poor response to chemotherapy (QT) and radiotherapy (RT), the treatment of choice for ST is total or partial sacral surgical resection [4-6]. However, despite modern spine surgery techniques, ST surgery still is an important challenge associated with high morbidity or severe neurological deficit, so the current challenge lies in the preservation of neurological function. Given the important role of radical resection in survival, reconstruction should be planned to achieve stability [6], without forgetting that quality of life must be one of the most important goals of treatment [7]. An essential aspect of surgical treatment is staging; there are two staging systems widely used for primary tumours of the spine, the Enneking staging system, and Weinstein-Boriani-Biagini classification. Both of them aid the surgeon to make an appropriate surgical decision. On the other hand, there are 3 important considerations in the surgical management of sacral tumours, the preservation and maximization of neurologic function, protection of ventral abdominal and pelvic structures, and lumbopelvic fixation [1]. In unresectable or recurrent tumours, non-surgical strategies have been applied in the management of select ST, like RT, cryoablation [8], or the use of nuclear medicine [9], each one with variable results. To date, surgical excision with wide resection margins offers the best long-term prognosis but it is associated with long-term functional compromise and mechanical instability [10]. Thus, it is important to identify other clinical or surgical characteristics in our patients to improve their outcomes. This study aimed to describe the clinical course and prognosis of surgically treated patients with sacral tumours.

Material and Methods

A retrospective case series study was made from 2011-2017 in the National Cancer Institute, Mexico City, Mexico. Adult patients with a diagnosis of primary or metastatic ST were included, they were subjected to sacral surgery without previous treatment, incomplete expedients were excluded. The following variables were evaluated in our patients: Age, genre, histology of the tumour, clinical characteristics, surgical technique, surgically related complications, progression-free survival (PFS), overall surveillance (OS), and following time (FT). Finally, two representative cases were exposed.

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Table 1. Sacrectomy cases. The cases are exposed in chronological order. ILR: Iliolumbar reconstruction; PFS: Progression free survival; OS: Overall NA (Does not apply); WI: wound infection

Patient	Diagnoses	Genre	Age	First symptom	Enneking	Surgery	Bleeding (ml)	Surgery time (Hr)	Surgery related complications	Kamofsky presurgery (%)	Kamofsky postsurgery (%)	Status	PFS	OS	FT
1	Chordoma	Female	59	Axial pain	IIB	Partial Sacrectomy	1700	6	CSF Leak, WI	90	70	Alive	31	91	94
2	Chondrosarcoma	Male	32	Axial pain	IIB	Partial Sacrectomy	2000	6	Postsurgical gluteus hematoma	80	90	Alive	77	77	80
3	Chordoma	Male	59	Axial pain+ Radicular pain	IIB	Total Sacrectomy	1500	8	NA	80	80	Death	14	29	33
4	Solitary fibrous tumour	Female	26	Constipation	IIB	Total Sacrectomy	2000	7	Hypovolemic shock grade III	90	0	Death	0	0	2
5	Desmoplastic medulloblastoma	Male	18	Radicular pain	IIB	Partial Sacrectomy	1600	5	NA	80	70	Death	36	42	47
6	Ewing sarcoma	Male	31	Axial pain+ Radicular pain	IIB	Partial Sacrectomy	2000	4	WI	90	100	Death	18	34	50
7	Chordoma	Male	53	Axial pain	IIB	Total Sacrectomy	6000	2.3	Hypovolemic shock grade IV	90	90	Alive	70	72	74
8	Chordoma	Female	31	Constipation	IIB	Total Sacrectomy	2300	5	Hypovolemic shock grade, Death	90	0	Death	0	0	5
9	Giant cell tumour	Female	21	Radicular pain	IB	Partial Sacrectomy	3000	4	Hypovolemic shock grade III	90	100	Alive	108	108	110
10	Chordoma	Male	32	Axial pain	IIB	Partial Sacrectomy	4000	4	NA	100	90	Alive	8	17	20
11	Chordoma	Male	59	Axial pain+ Radicular pain	IIB	Partial Sacrectomy	1500	4	NA	90	100	Death	13	63	69
12	Follicular thyroid carcinoma	Female	61	Radicular pain	III	Partial Sacrectomy	500	4.3	NA	70	90	Alive	22	66	70
13	Chondrosarcoma	Male	30	Radicular pain	IIB	Total Sacrectomy	1700	2	Postsurgical intestinal perforation	90	50	Alive	12	12	14
MEAN			39.3				2292.3	5		86.9	71.54		31.5	43,3	47.83
SD			16.1				1386.5	1.67		7.51	34.84		34.7	34.06	33.85
CV			0.41				0.60	0.33		0.09	0.49		1.10	0.79	0.71

Surgical Methods

Total sacrectomy can be performed by anterior or posterior approaches. The anterior procedure is an intraperitoneal approach used to expose the anterior surface to ligate the main tumour vessels, and to perform an anterior partial sacrectomy. After this, the rectus abdominis myocutaneous flap, based on the inferior epigastric vessel, is prepared, and a posterior sacrectomy is performed, dividing all sacral nerve roots in the thecal sac. After complete en bloc extirpation of the sacrum with the tumour, spinopelvic reconstruction and closure with a myocutaneous flap are performed. Spinopelvic reconstruction is undertaken using a modified Galveston technique or dual iliac screw fixation combined with posterior lumbar segmental fixation. These provide a long lever arm within the ilium to counteract the forces exerted by the lumbar spine. In the posterior approach, a spinolaminar and sacrococcygeal dissection is performed, the total or partial sacrectomy is achieved, and finally, iliac screw fixation combined with posterior lumbar segmental fixation is performed. The posterior approach is preferred by our group because complete resections can be yielded with fewer complications like bleeding, vascular injury, intestinal perforation, and postoperative recovery is better than the anterior approach.

Statistical analysis

Descriptive analysis was performed with means SE (standard error) or percentage as appropriate in each case.

Results

A total of 13 patients were included in the study, of which 11 were primary tumours and 2 metastases. 8 cases were male, 5 female, and the mean age was 39.3 years. The most frequent tumour was chordoma (46%), followed by chondrosarcoma, fibrous solitary tumour, Ewing sarcoma, and giant cell tumour. The metastatic tumours were desmoplastic medulloblastoma and follicular thyroid carcinoma. 53% of the patients presented as first related symptom axial lumbosacral pain, two patients (15%) presented with new-onset constipation as the first symptom, in all the patients' total resection of the tumour was achieved partial sacrectomy was performed in the 61.5% of the population, total sacrectomy in 38.5% of patients, and 15.3 % an iliolumbar fixation was required. Mean bleeding was 2292 mL (500-6000 mL). 84.6% of cases presented IIb stage according to the Enneking staging system and the presurgery Karnofsky scale was 90% or more in 61.5% of the patients. Post-Surgery Karnofsky at 6 months was 90% or more in 53.8% of the cases. The postsurgery ECOG score was the same or lesser than pre-surgery (except for the deceased patients). All surgeries were performed by posterior approach. Partial sacrectomy was performed in 61.5% of the population, and an iliolumbar fixation was conducted in 15.3%. The mean surgery time was 4.73 hours.

Surgery complications were present in 53.8% of patients, two patients (15.3%) died during the first week after surgery from bleeding, more precisely due to hypovolemic shock in the Intensive Care Unit, and three more patients presented bleeding with hypovolemic shock without long-term complications. The most common post-surgery complication was hypovolemic shock with 30.7%. 7.6% of patients presented cerebrospinal fluid leak, and 15.38% wound or soft tissue infection (WI) (Table 1). The mean follow-up time was 51.4 months, mean progression-free survival was 31.6 months.

Representative cases

- Case 1 (Case 7 in table 1) In 2007 a 53-year-old male without relevant medical history, presented severe lumbosacral pain without irradiation, MRI was carried out. A tumor involving the inferior half of the sacrum was observed. A partial sacrectomy was performed, and the patient evolved without neurological deficit or complications. 70 months later, it was observed to recur with left sacroiliac joint involvement (Figure 1). A total sacrectomy with Galveston technique reconstruction was performed (Figure 1). The patient exhibited complications after surgery such as hypovolemic shock and an enterocutaneous fistula, which was resolved by colostomy. Two months after surgery, S2-S4 hypoesthesia and loss of sphincter´s control were evident. To date, there is no evidence of tumour recurrence.

- Case 2 (Case 5 in table 1) A male of 18 years old with a history of desmoplastic medulloblastoma in the fourth ventricle and subtotal surgical resection (QT, and RT 36

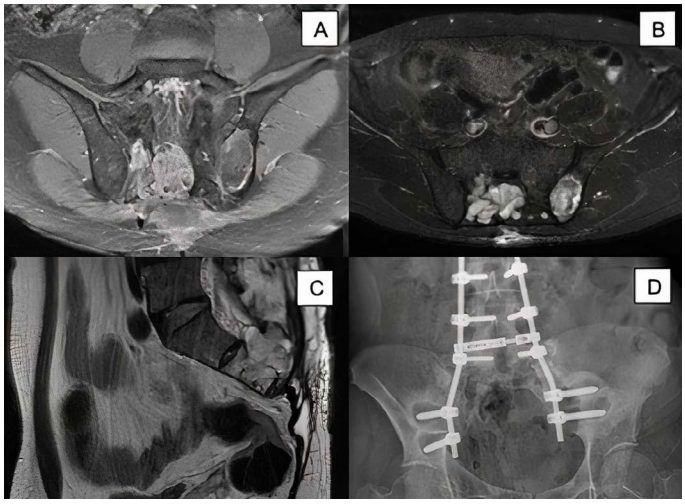

Figure 1. Representative case 1. A, B, C: MRI of the sacrum in diverse sequences showing a solid tumour in the inferior half of the sacrum and left sacroiliac joint enhanced with gadolinium administration. D: Postsurgical pelvic anteroposterior radiography. It is evident the absence of sacrum and coccyx, Iliolumbar reconstruction with the Galveston technique can be appreciated

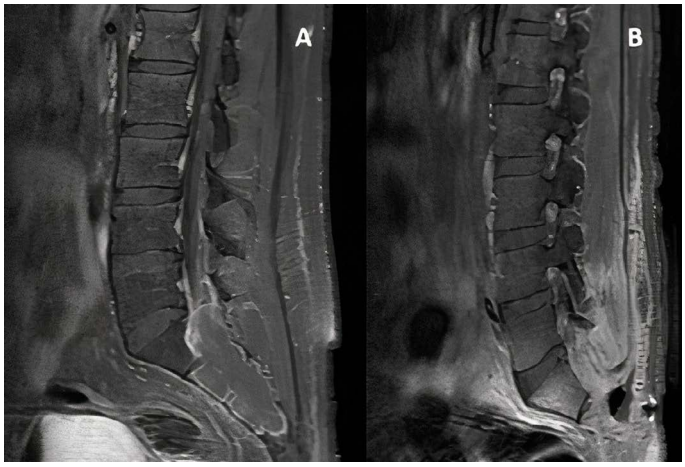

Figure 2. Representative case 2. A: Presurgery MRI showing a solid tumour in sacrum and coccyx enhanced by gadolinium administration. B: Postsurgical MRI showing no evidence of residual tumour

+ 20 Gy), was followed up. Two years after brain surgery, he started with paraesthesia, pain in the right L5 territory, saddle hypoesthesia, and constipation. Lumbosacral MRI was carried out and a solid tumour in the inferior sacrum was evident, fractionated RT 30 Gy to sacrum was done. Progression of the disease at sacrum occurred one year later. A partial sacrectomy was performed (Figure 2), no complications were observed, progression was evident three years later. Six months later the patient died due to systemic dissemination of the disease.

Discussion

Partial or total sacrectomy is the procedure of choice to treat most sacral tumours. However, it's a surgery with a high risk of complications and mortality, besides a local elevated recurrence rate [1,2,11,12]. The most frequent ST in our case series was chordoma, which correlates with previous studies reporting this one as the most frequent [13-15]. On the other hand, chondrosarcoma is one of the most common malignant tumours arising from the sacrum. Although chondrosarcoma is a low to intermediate-grade malignancy, it is invasive and has a high rate of local tumour recurrence [16]. In our study, just one patient had chondrosarcoma with a PFS of 79 months after a total sacrectomy. Giant cell tumours were found just in one case, although are reported as one of the most common benign sacral tumours and as highly recurrent in many studies [11,17,18]. Ewing sarcoma is not common but is diagnosed frequently at a younger age (in the first two decades). Our hospital provides care only for adult patients, and the youngest was 18 years old. He was treated with partial sacrectomy and showed recurrence 18 months later. On the other hand, we found one patient with PFS of 22 months and OS of 66 months derived from follicular thyroid metastasis carcinoma, which is rare by the few cases of this nature reported in the literature with sacral lesions [18]. Pan, *et al*. reported that factors like an age > 60 years, distant metastases, and nonsurgical therapy were independent risk factors for survival reduction [19]. Locoregional recurrence affects > 50% of patients [20,21], and in dedifferentiated chordomas, characteristics such as tumour diameter >10 cm, marginal resection, and sacroiliac joint infiltration may be associated with increased risk of local recurrence and mortality. In this study, our patients were <60 years, predominantly men, with good presurgical Karnofsky, without identified metastases and all were treated surgically.

Patients who developed local recurrence had a PFS between 14 and 70 months, which is common on this type of tumour [22]. An important surgical goal in sacrectomy is to preserve the sacral nerves as much as possible, without compromising clear surgical margins, besides it is known that an adequate wide margin can improve the survival rate [10,23,24]. In parallel, maintaining the sphincter function gives satisfactory results from the patient's perspective, it has been reported S3 nerve root is crucial to maintain the neurophysiology of bowel, bladder, and sexual function, and conservation of at least one S2 during surgery might benefit the patients [25,26]. Preoperative conditions of the bladder, bowel, motor functions, level of sacral tumour involvement, and corresponding level of sacrectomy predict their long-term functionality after surgery [27]. Higher resections have more pain and loss of physical function in comparison to patients with lower resections [26]. Two of the patients died due to hypovolemic shock. This is the greatest risk of the surgery, because of the high vascularity density of the region, composed by the iliac arteries and veins, presacral venous plexus, and arteries. A limitation of the posterior-only approach is inadequate visualization of these many vessels resulting in a possible unrecognized injury, an aggregate to this problem is the loss of normal anatomy and neovascularization of the tumour. Among the literature, the recommendation is previous arterial embolization of the tumour to prevent bleeding, due to the fact that it is an expensive procedure many of the patients refused to perform it. The main predictors in mortality and bleeding are the size and invasion of the tumour, the type of resection, and if it was treated previously with arterial embolization. On the other hand, there are few publications studying mortality and bleeding, the publications that report these adverse events had similar incidence rates (3.7-17.6%) [28-30]. As for the complexity and morbidity of this surgery, it is of most value to a highly trained multidisciplinary team to achieve the best outcome for these patients.

In our case series, case number 1 exposed above in representative cases, after total sacrectomy presented alterations in sensation, strength in lower limbs, and sphincters. Iliolumbar reconstruction was performed in two cases because they underwent type 2 resections and the instability of the spine was highly probable; according to the literature patients with type I resection do not require reconstruction [31]. None of our patients were reoperated for reconstruction. In most cases, in our study, the first ST-related symptom was axial lumbosacral pain, which is nonspecific and of late diagnosis. After surgery patients manifested bleeding frequently, even though it was lower than other mean bleeding volumes reported in the literature [27], however, it caused the death of two patients. WI was a common post-surgery problem, but it did not interfere with the hospital's discharge. The PFS and OS were similar to previously described in the literature in each case depending on the tumour type [20].

Conclusion

ST is a challenging disease for the surgeon, most of the time they are recurrent, and the patient courses with neurological or mechanic sequelae. The vague symptoms and clinical course make the early diagnosis difficult. The PFS and OS depend on the biology of the tumor, and iliolumbar reconstruction is required in few cases with generally good mechanical results. More experience is needed to improve the outcomes of patients with ST.

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