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ORIGINAL ARTICLE

Factors affecting operative morbidity and long-term outcomes in patients undergoing surgery for presacral tumours: a multicentric cohort study from the Turkish Collaborative Group for Quality Improvement in Colorectal and Pelvic Surgery

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Abstract

Aim: Data regarding the operative management of presacral tumours present various dilemmas due to their rarity and heterogeneous nature. The aim of this study was to evaluate the management strategy, factors associated with operative morbidity and long-term postoperative outcomes in a large group of patients undergoing surgery for presacral tumours.

Method: This study was designed as a multicentre retrospective cohort study. Records of patients who underwent surgery for presacral tumours at 10 tertiary colorectal centres between 1996 and 2017 were evaluated.

Results: One hundred and twenty seven patients (44 men) with a mean age of 46 years and body mass index of 27 kg/m^2 were included. Fifty eight per cent of the patients had low sacral lesions (below S3). The operative approaches were transabdominal (17%), transsacral (65%) and abdominosacral (17%). The postoperative morbidity was 19%. Thirty per cent of the patients had a malignant tumour. Longer duration of symptoms ($p = 0.001$), higher American Society of Anesthesiologists score ($p = 0.01$), abdominosacral operations ($p = 0.0001$) and presacral tumours located above S3 ($p = 0.004$) were associated with an increased risk of postoperative morbidity. Overall long-term postoperative recurrence and mortality were 6% and 5%, respectively, within a 3-year mean follow-up period in patients with presacral malignant tumours.

Conclusion: Reduced physical condition, omission of symptoms prior to surgery, combined resections and high sacral tumours are the risk factors associated with postoperative complications in patients undergoing surgery for presacral tumours. Meticulous planning of the operation and intensified perioperative care may improve the outcomes in high-risk patients.

KEYWORDS
morbidity, presacral, retrorectal, surgery

INTRODUCTION

Presacral tumours have diverse histological patterns with benign and malignant features. There is limited information about the clinical and pathophysiological characteristics of tumours arising from the presacral area due to their infrequent presentation with occult symptoms [1] While the majority of presacral tumours are benign, their management usually requires surgical treatment, which is highly complex due to the anatomical boundaries of the pelvis and organs therein [2] Surgical treatment of presacral tumours is based on various important factors, including the location of the lesion, findings of the preoperative imaging studies and biopsy results when performed [3]

The majority of data regarding the management strategies and outcomes of patients with presacral tumours exist in limited case series. Operative outcomes in previous studies were only documented without further evaluation, while existing data include the type of tumour, oncological outcomes with regard to pathology results, recurrence and survival [17, 18, 22]. Hence, information regarding risk factors that may worsen the operative results and long-term outcomes following surgical treatment of presacral tumours is very limited. This multicentre study aimed to evaluate the management strategy, factors associated with operative morbidity and long-term postoperative outcomes in a large group of patients undergoing surgery for presacral tumours.

METHODS

The study was carried out in accordance with the Code of Ethics of the World Medical Association (Declaration of Helsinki). After obtaining approval from the local ethical committee, records of patients who underwent surgery for presacral tumours at 10 tertiary colorectal centres between 1996 and 2017 were evaluated. All operations were performed with curative intent. Primary and recurrent locally advanced carcinomas of the rectum and urogenital organs, patients with familial cancers, patients with synchronous or metachronous cancer, metastatic cancer infiltrating the sacrum and primary osseous tumours were excluded. The anatomical characteristics of presacral tumours were defined by Uhlig and Johnson [4].

Patients' demographics, American Society of Anesthesiologists (ASA) score, prior history of pelvic surgery, symptoms, duration of symptoms, tumour location according to the sacral bone, neoadjuvant ± adjuvant treatment, preoperative histological evaluation, operative strategy, length of hospital stay, final pathological diagnosis, and short-term and long-term postoperative outcomes were analysed. The operative morbidity rate was calculated by considering the number of patients who had at least one postoperative complication. Discharge criteria were tolerance to foods without nausea or vomiting, established bowel or stoma function, adequate pain management with oral analgesia and independent walking.

What does this paper add to the literature?

There is limited information about the clinical and pathophysiological characteristics of tumours in the presacral area. This multicentre study showed that reduced physical condition, omission of symptoms prior to surgery, combined resections and high sacral tumours are the risk factors associated with postoperative complications in patients undergoing surgery for presacral tumours.

Clinical approach

A combination of a CT scan and magnetic resonance imaging (MRI) was used if needed. MRI was used for all patients. MRIs were reviewed by experienced radiologists with operating surgeons in all institutions. Performance of a preoperative biopsy was decided individually by a multidisciplinary tumour board according to clinical and radiological findings of each presacral tumour.

Complete surgical resection is mandatory for presacral tumours, even in asymptomatic patients. The choice of a proper surgical technique depends on the location and tumour size, as well as the relationship to the adjacent organ and/or structures such as intrapelvic viscera, sacrum or pelvic sidewalls. Malignant tumours were resected *en bloc* with adjacent organs to achieve clear resection margins (R0). Upper and mid-sacral tumours that extended above the third sacral vertebra (S3) or attached to internal organs and the pelvic side wall were removed using an abdominal (anterior) (Figure 1) or a combined (abdominosacral) approach (Figure 2), particularly when *en bloc* sacral resection is essential, whereas a perineal (transsacral) approach (Figure 3) was preferred for tumours located below S3 without adherence to any anatomical structure. Open or laparoscopic techniques were preferred depending on the surgeon's discretion and tumour characteristics in our series. The surveillance

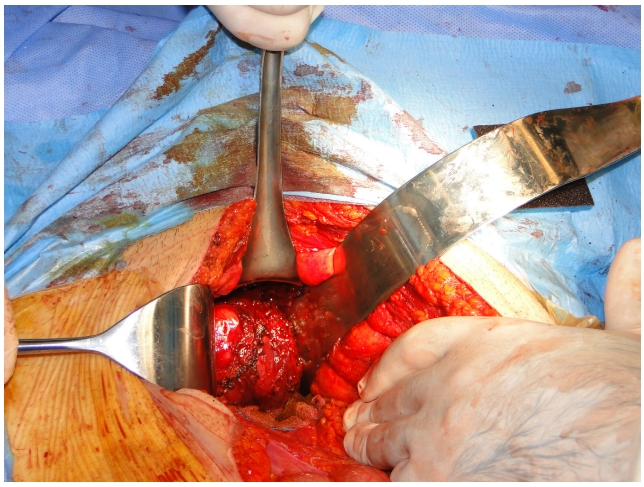

FIGURE 1 A transabdominal approach for schwannoma.

protocol for malignant tumours is colonoscopy once a year and abdominal MRI with thoracic CT every 3 months. If there is a suspicion of tumour recurrence, a positron emission tomography CT scan can be done. Annual abdominal imaging with CT and a colonoscopy are preferred for patients with benign lesions.

Statistical analysis

Categorical variables are given as percentages and numeric variables are given as mean ± SD or median (range) considering the normality. Categorical variables were compared with the chi-square test or Fisher's exact test. Numeric variables were compared with the independent *t*-test or Mann–Whitney *U*-test. After univariate analyses were performed, variables with a *p*-value of less than 0.05 were used in logistic regression to detect factors associated with mortality and morbidity.

RESULTS

One hundred and twenty seven patients [44 (35%) men] with a mean age of 46 ± 16 years and a body mass index of 27 ± 4 kg/m^2 were recorded. The symptoms were anal/sacral pain (n = 50, 40%), rectal fullness (n = 28, 22%), weight loss (n = 15, 12%), constipation (n = 15, 12%), incidental (n = 9, 7%), rectal bleeding (n = 6, 5%), rectal numbness (n = 3, 2%) and faecal incontinence (n = 1, 1%) (Table 4). The duration of the symptoms was 12 ± 8 months. Nine (7%) patients had a previous history of sacral surgery. The median ASA score of the patients was 2 (1–3). Seventy three per cent (n = 93) of the patients had low sacral (below S3) lesions. A preoperative biopsy was performed in 27 (21%) patients and 19 (15%) patients were diagnosed with a malignant lesion preoperatively. Five (4%) out of 19 patients with a malignant lesion had neoadjuvant treatment. The operative approaches were transabdominal (n = 22, 17%), transsacral (n = 83, 65%) and abdominosacral (n = 22, 17%). A laparoscopic excision was performed for 7 (6%) patients with benign upper sacral lesions. The remaining operations (94%) were performed with an open approach. The robotic approach was not used in any cases. Twenty six patients had concurrent proctectomy. Among these patients 23 had a temporary and three a permanent stoma. The intraoperative complications were vascular injury (n = 3, 2.4%), ureteral injury (n = 2, 1.6%) and rectal injury (n = 1, 0.8%). The risk of postoperative morbidity was 19% (n = 24) (Table 1). There was no short-term mortality. The mean length of hospital stay was 8 ± 7 days. Thirty per cent (n = 38) of the patients were diagnosed with a malignant lesion based on the final pathological evaluation (Table 2). Two (5.3%) out of 38 patients with malignant tumours had R1 resection. One of those two patients had intraoperative radiotherapy and the other underwent adjuvant radiotherapy. Both received adjuvant chemotherapy. Longer duration of symptoms (p = 0.001), higher ASA score (p = 0.01), abdominosacral operation (p = 0.0001) and tumours located above S3 (p = 0.0004) were associated with an increased risk of postoperative morbidity (Table 3). The mean follow-up time was 37 ± 29 months. Locally recurrent disease developed in 3 (3.4%) out of 89 patients with benign lesions [tailgut cyst (n = 2) and leiomyoma (n = 1)]. Disease-free and overall survival were 94% and 95%, respectively, within the 3-year mean follow-up time of patients with presacral malignant tumours (Figure 4). Among the 38 patients with malignant presacral tumours, 10 (26%) developed recurrences. The mean time to recurrence was 20.9 ± 13 months. Primary diagnoses of patients with recurrences were sarcoma (n = 6), chordoma (n = 3) and a gastrointestinal stromal tumour (n = 1). Four out of 10 patients with recurrences died due to disease-related complications.

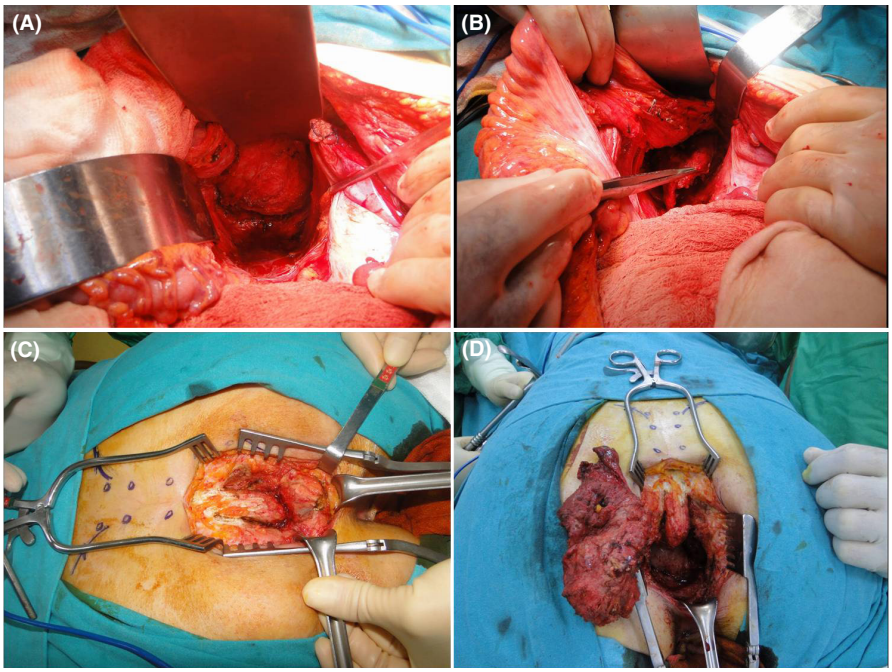

FIGURE 2 A combined approach for a tailgut cyst with benign features. (A) The posterior avascular plan is followed down to the pelvis without disturbing the tumoural lesion. (B) Tumour freed from the intrapelvic surrounding organs without violating their anatomical margins. (C) Access to the tumoral lesion through the rostral part of the sacrum and anal sphincteric complex in the prone position. (D) Complete surgical removal of the tumour at the end of the combined approach.

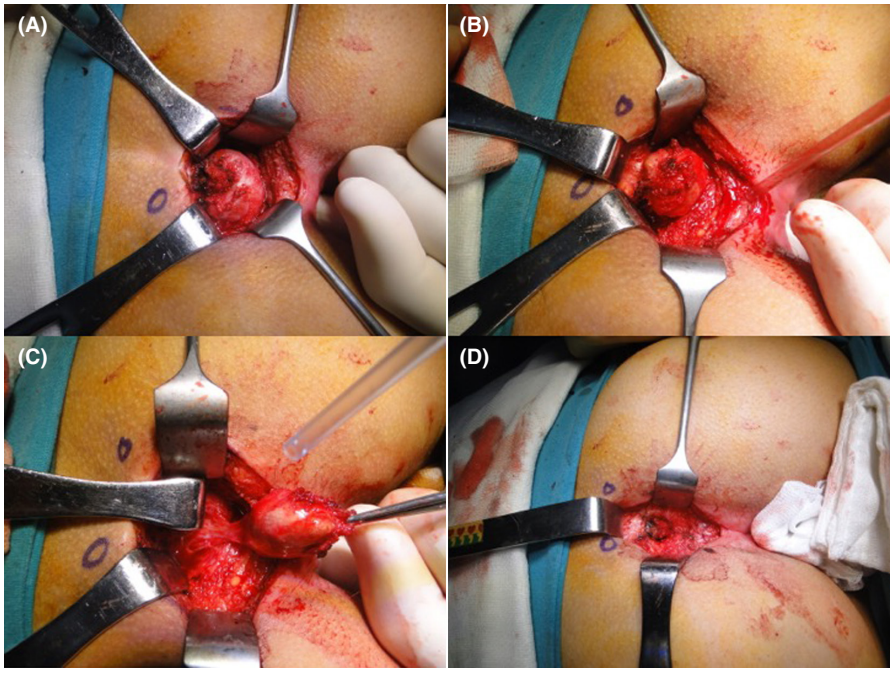

FIGURE 3 A perineal approach for a desmoid cyst. (A)–(C) Dissection of the lesion without damaging its integrity. (D) Surgical field after removal of the desmoid cyst.

TABLE 1 Postoperative complications.

Complication	*n* (%)
Surgical site infections	12 (9.5)
Ileus	10 (7.9)
Urinary retention	7 (5.5)
Deep venous thrombosis	6 (4.7)
Cardiovascular	4 (3.2)
Bleeding	3 (2.4)
Incontinence	2 (1.6)

TABLE 2 Final pathological diagnoses.

Diagnosis	*n* (%)
Malignant	
Sarcoma	13 (10)
Undifferentiated sarcoma	3 (2.4)
Leiomyosarcoma	2 (1.6)
Liposarcoma	2 (1.6)
Malignant peripheral nerve sheath tumour	2 (1.6)
Chondrosarcoma	1 (0.8)
Fibrosarcoma	1 (0.8)
Osteogenic sarcoma	1 (0.8)
Haemangioendothelial sarcoma	1 (0.8)
Chordoma	10 (7.9)
Gastrointestinal stromal tumour	6 (4.7)
Teratocarcinoma	5 (3.9)
Plasma cell myeloma	2 (1.6)
Malignant neoplasm of unknown type	2 (1.6)
Benign	
Tailgut cyst	39 (30.7)
Epidermoid cyst	17 (13.4)
Teratoma	15 (11.8)
Inflammatory	8 (6.3)
Schwannoma	5 (3.9)
Hamartoma	4 (3.1)
Endometrioma	1 (0.8)

DISCUSSION

Our results revealed that patients undergoing abdominosacral resection and patients with high sacral tumours (above S3) are at risk of operative morbidity. Patients undergoing abdominosacral operation have longer incisions than patients undergoing isolated abdominal or perineal surgery. Length of incision is an independent risk factor for the development of surgical site infection [5]. Patients undergoing surgery for upper presacral tumours are at risk of sacral nerve root injury which causes intestinal and urinary dysfunction [6, 7]. The majority of our patients had symptoms for longer than a year prior to diagnosis of their presacral tumour.

Pain arising from the sacrum and/or anus was the most common symptom associated with presacral tumours in our patients. Vague and nonspecific presentations usually cause delayed diagnosis of presacral tumours [2, 8]. Prolonged duration of symptoms was associated with an increased risk of operative morbidity in our series. Delayed diagnosis of a presacral tumour may also lead to worse long-term outcomes, particularly in cancer patients. The physical condition of patients undergoing surgery for a presacral tumour is another predictor of operative morbidity, as was observed in our series. The relation between a high ASA score and the poor

TABLE 3 Factors associated with postoperative morbidity.

Factors	Morbidity (−) ($n=103$)	Morbidity (+) ($n=24$)	p-value
Age (years)	46 ± 19	46 ± 15	0.88
Sex, male, *n* (%)	32 (31.1)	14 (58.3)	0.08
BMI (kg/m^2)	27 ± 4.62	27 ± 4.15	0.13
Estimated blood loss (mL)	240 (90–750)	305 (140–1100)	0.06
Duration of symptoms (days) (mean)	6,7	13,7	0.001
ASA score 3, *n* (%)	3 (2.9)	4 (16.7)	0.01
Prior pelvic surgery, *n* (%)	6 (5.8)	3 (12.5)	0.25
Tumour above S3, *n* (%)	22 (21.4)	12 (50)	0.004
Combined (abdominosacral) surgery, *n* (%)	9 (8.7)	13 (54.2)	0.0001
LOH (days)	5 ± 3	16 ± 10	0.0001
Malignant tumour, *n* (%)	28 (27.2)	10 (41.7)	0.16

Abbreviations: ASA, American Society of Anesthesiologists; BMI, body mass index; LOH, length of hospital stay.

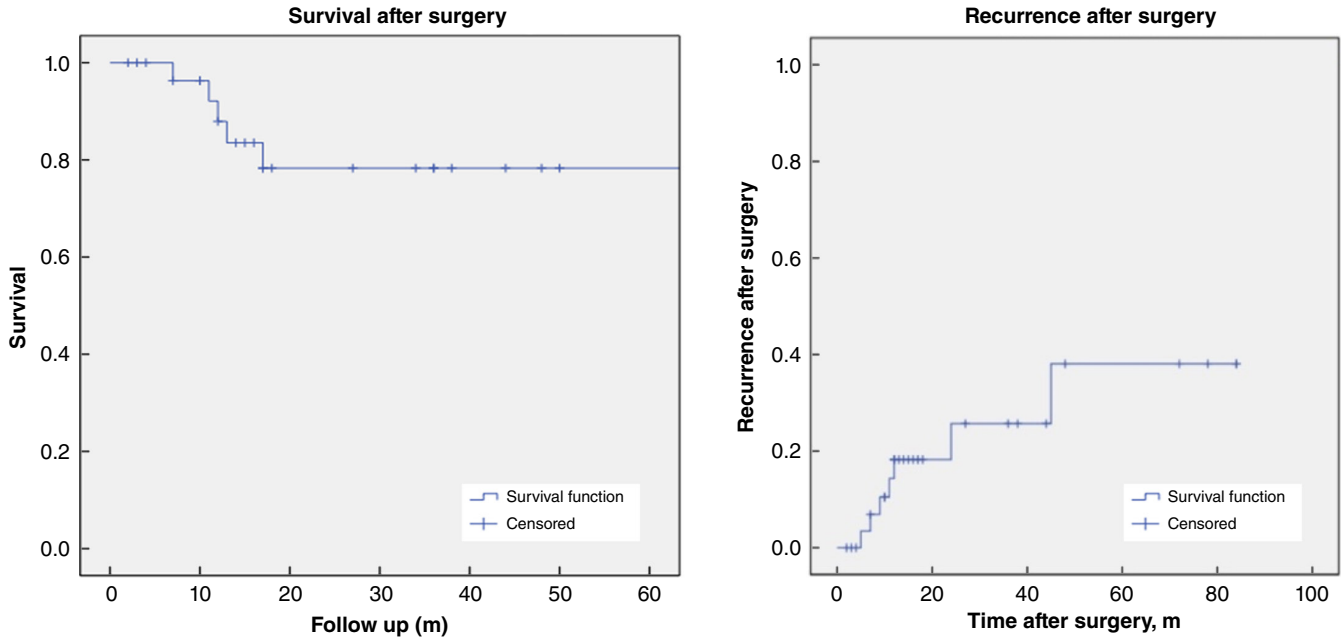

FIGURE 4 Overall survival and recurrence after surgery in patients with a malignant presacral tumour (m, months).

TABLE 4 Prediagnostic symptoms.

Symptom	*n* (%)
Anal/sacral pain	50 (40%)
Rectal fullness	28 (22%)
Weight loss	15 (12%)
Constipation	15 (12%)
Incidental	9 (7%)
Rectal bleeding	6 (5%)
Rectal numbness	3 (2%)
Faecal incontinence	1 (1%)

postoperative outcome has been confirmed in large groups of surgical patients [9]. The risks and severity of complications increased mainly in patients in ASA classes III and IV. Surgical site infections, ileus and urinary retention were the leading postoperative complications in our series. According to our data, a longer duration of symptoms is associated with an increased risk of postoperative morbidity. The omission of symptoms by patients seems to be the major factor delaying proper treatment.

Surgical treatment of presacral tumours should be performed at tertiary care referral centres by multidisciplinary teams experienced in complex abdominopelvic surgery for reasons of a wide variety of difficulties related to disease, a hostile intrapelvic environment and complicated surgical technique [10]. The type of surgery is planned based on the location of the presacral tumour. The anterior, pure abdominal approach can be performed when the tumour is located above the level of S3 with an open or advanced laparoscopic technique. This approach provides excellent exposure to the iliac vessels and ureters and direct access to the pelvic sidewalls and pelvic viscera. The posterior, pure perineal,

approach is ideal for small, purely cystic and benign tumours that do not extend above S3. When the superior pole of the tumour is palpable and mobile on digital rectal examination, the posterior approach should be considered. A large bulky tumour, densely adhered to the adjacent viscera, or a tumour in an intermediate location must be resected via a combined approach. The combined approach (abdominoperineal) permits vascular control and provides good exposure for the protection of vital structures and radical resection of large presacral tumours with malignant features. With the potential for extension of these tumours deep into the pelvis the identification of surgical planes between the tumour and the surrounding tissues may become more difficult, particularly if neoadjuvant therapy was administered, and patients may have to be repositioned. The surgeon can wear a double glove on his or her nondominant hand, place the index finger in the anal canal and lower rectum and express the lesion into the wound by applying forward pressure with the index finger to prevent injury to the rectal wall during the dissection. However, partial resection of the sacrum, nerve sacrifice and even loss of the rectum may be needed to achieve complete resection of a presacral tumour.

We were able to use laparoscopy for a limited number of patients in our series. Although it is doable, the oncological safety and recovery benefits of laparoscopic resection for complex and malignant presacral tumours are inconclusive [11, 12]. While minimally invasive surgery potentially offers various advantages, including a shorter hospital stay and faster recovery compared with open surgery in patients with a virgin abdomen, a history of previous abdominal surgery and the potential characteristics of complex presacral tumours may reduce the expected benefits of minimally invasive surgery.

Currently, imaging modalities including MRI provide us with nearly accurate information about the nature of the presacral tumour [13, 14]. Presacral tumours with invasive features including heterogeneous and solid wall appearance should be biopsied [3]. Diagnosis of malignant tumours may indicate neoadjuvant treatment to increase the ultimate success of treatment and prepare patients mentally and physically for radical surgery. A complete surgical excision can be considered as the index approach for patients with a clearly resectable tumour to eliminate the complications of biopsy such as tumour seeding, haemorrhage and subsequent secondary infection [15]. Biopsies of presacral tumours should never be done transrectally, transabdominally or transvaginally. Covered Tru-Cut needles should be used to reduce the risk of tumoral seeding. Chordomas and sarcomas, which are the most common malignancy of the presacral area, are usually unresponsive to radiotherapy [16, 17]. However, some experts recommend the use of preoperative biopsy of presacral tumours for differentiating benign versus malignant solid presacral tumours due to the current limitations of imaging [3]. Two large series from the Cleveland Clinic and the Mayo Clinic reported no recurrences identified at the biopsy site [3, 18]. Preoperative biopsy may also prevent some patients from having unnecessary surgery for presacral tumours that can be treated primarily with chemotherapy alone, such as myeloma and lymphoma. Our biopsy preference has evolved in the last 20 years. Around a fifth of our patients had a preoperative biopsy. Our interactive experience among the members of our collaborative group was to perform a biopsy selectively by evaluating every case individually according to the MRI findings, resectability of the tumour and the decision of the multidisciplinary tumour board. We only had two patients with positive surgical margins. One of these patients had intraoperative radiotherapy (IORT). Current data regarding the role of IORT for advanced presacral tumours are scarce. IORT has been used for dose escalation in patients with retroperitoneal sarcoma or a reirradiation modality for recurrences [19].

In a 3-year mean follow-up, we observed 3.4% and 26% recurrences after the removal of presacral tumours with benign and malignant characteristics, respectively. In previous reports, the risk of recurrence was reported as around 25% for malignant lesions and around 7% for benign lesions within 5 years after curative surgery [13]. We observed recurrences within 2 years after surgery. Patients with presacral sarcoma, chordoma, tailgut cyst, leiomyoma and gastrointestinal stromal tumours were at risk of recurrence in our series. The 5-year risk of recurrence was previously reported as 61% and 40% for patients undergoing radical surgery for presacral sarcomas and sacral chordomas, respectively [17, 20]. In our series, one out of six patients with an extra-gastrointestinal stromal tumour developed recurrence after surgery, which is the mainstay treatment [21] Rare types of presacral tumours such as extra-gastrointestinal stromal tumours should be considered and managed properly.

Our study has some limitations due to its retrospective, nonrandomized and multicentric nature; however, it is one of the largest series to evaluate short- and long-term outcomes in patients undergoing surgery for presacral tumours.

CONCLUSION

In conclusion, poor physical condition, omission of symptoms prior to surgery, combined resections and high sacral tumours are the risk factors associated with postoperative complications in patients undergoing surgery for presacral tumours. Meticulous planning of the operation and intensified perioperative care may improve the outcomes in high-risk patients.

AUTHOR CONTRIBUTIONS

Erman Aytac: Conceptualization; formal analysis; investigation; project administration; writing – original draft; resources; visualization; supervision; validation; data curation; methodology. **Melik Kagan Aktas:** Conceptualization; investigation; formal analysis; project administration; writing – original draft; resources; visualization; supervision; data curation; methodology; validation. **Tahsin Colak:** Conceptualization; data curation; formal analysis; investigation; methodology; project administration; resources. **Sezai Demirbas:** Conceptualization; investigation; methodology; formal analysis; project administration; data curation; resources. **Cihangir Akyol:** Conceptualization; investigation; methodology; formal analysis; project administration; data curation; resources. **Mustafa Oncel:** Conceptualization; investigation; methodology;

formal analysis; project administration; data curation; resources. **Ersin Ozturk:** Conceptualization; investigation; methodology; formal analysis; project administration; data curation; resources. **Funda Obuz:** Conceptualization; investigation; methodology; formal analysis; project administration; data curation; resources. **Basar Ucaroglu:** Conceptualization; investigation; methodology; formal analysis; project administration; data curation; resources. **Sezai Leventoglu:** Conceptualization; investigation; methodology; formal analysis; project administration; data curation; resources. **Ismail Hakki Ozerhan:** Conceptualization; investigation; methodology; project administration; formal analysis; data curation; resources. **Bilgi Baca:** Conceptualization; investigation; methodology; formal analysis; project administration; data curation; resources. **Utku Ozgen:** Conceptualization; investigation; methodology; formal analysis; project administration; data curation; resources. **Mustafa Haksal:** Conceptualization; investigation; methodology; formal analysis; project administration; data curation; resources. **Volkan Tumay:** Conceptualization; investigation; methodology; formal analysis; project administration; data curation; resources. **Ethem Geçim:** Conceptualization; investigation; methodology; formal analysis; project administration; data curation; resources. **Dursun Bugra:** Conceptualization; investigation; methodology; formal analysis; project administration; data curation; resources.

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No funding has been used.

CONFLICT OF INTEREST STATEMENT

The authors have no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ETHICS STATEMENT

Approved.

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