



Review Article

Clinical manifestations, classification, and surgical management of sacral tumors and the need for personalized approach to sacrectomy

Brian Fiani¹, Juliana Runnels², Alexander Rose², Athanasios Kondilis³, Amelia Wong⁴, Brian L. Musch⁵

¹Department of Neurosurgery, Desert Regional Medical Center, Palm Springs, California, ²School of Medicine, University of New Mexico, Albuquerque, New Mexico, ³College of Osteopathic Medicine, Michigan State University, East Lansing, Michigan, ⁴College of Osteopathic Medicine, Western University of Health Sciences, Pomona, California, ⁵College of Osteopathic Medicine, William Carey University, Hattiesburg, Mississippi, United States.

E-mail: *Brian Fiani - bfiani@outlook.com; Juliana Runnels - jmrunnels@salud.unm.edu; Alexander Rose - alnrose@salud.unm.edu; Athanasios Kondilis - kondilis@msu.edu; Amelia Wong - amelia.wong@westernu.edu; Brian L. Musch - bmusch476901@student.wmcarey.edu



*Corresponding author:

Brian Fiani, D.O.
Department of Neurosurgery,
Desert Regional Medical
Center, Palm Springs,
California, United States.

bfiani@outlook.com

Received : 09 February 2021

Accepted : 07 April 2021

Published : 03 May 2021

DOI:

10.25259/SNI_133_2021

Quick Response Code:



ABSTRACT

Background: Although comprising 7% of all spinal tumors, sacral tumors present with a litany of issues due to their slow growth and difficulty in detection. As a result, sacral tumors can grow unperturbed for years until a patient presents for an incidental workup of an unassociated minor trauma or an offending primary tumor source that has metastasized to the sacrum; in most cases, this includes primary tumors of the breast, prostate, and lung. The goal of this review is to outline the pathophysiology underlying sacral tumors including the various tissues and structures that can be targeted for treatment, along with a discussion of the surgical approach to sacrectomy.

Methods: An extensive review of the published literature was conducted through PubMed database with articles simultaneously containing both search terms “sacral tumors” and “sacrectomy.” No date restrictions were used.

Results: The search yielded 245 related articles. Cross-checking of articles was conducted to exclude of duplicate articles. The articles were screened for their full text and English language availability. We finalized those articles pertaining to the topic.

Conclusion: Once a sacral tumor has reached the point of diagnostic detection, invasive sacrectomy is typically utilized (through an anterior, posterior, or combination approach) to locally isolate and resect the tumor and minimize risk of future tumor growth and additional bone loss. While institutions have varying criteria for surgical approaches, a combination of anterior and posterior approach has traditionally been used in total and high sacrectomies due to the control it provides surgeons toward the rectum and vasculature anterior to the sacrum. A posterior-only approach can be performed for tumors that failed to invade pelvic organs or extend past the lumbosacral junction. Early detection with screenings can help avoid invasive sacrectomy by identifying the onset of tumor formation in the sacrum, particularly for highly metastatic cancers.

Keywords: Bone metastasis, Sacral tumor, Sacrectomy, Sacrum, Spinal tumor

INTRODUCTION

Sacral tumors are rare slow-growing lesions, accounting for less than 7% of all spinal tumors. Many cases remain clinically silent and are incidentally discovered during workup of minor trauma.^[5,12] Symptomatic tumors often mimic common spinal pathologies such as

lumbosacral spondylosis, thereby leading to conservative management of nonspecific low back pain until persistent or progressive symptoms prompt referral for further workup. Moreover, the sacrum may be excluded from initial radiographic studies as these lesions can lie below the sacral 2 (S2) vertebrae or are obscured by bowel gas or stool.^[12,27,36] This combination of nonspecific symptoms, inadequate diagnostic imaging, and clinical rarity contributes to frequent delay in diagnosis and advanced tumor size at time of eventual diagnosis.

The purpose of this review is to examine the pathophysiology of sacral tumors; particularly as a result of clinical manifestations including patient presenting signs and symptoms. The difficulty posed by sacral tumors (and what this review aims to accomplish) from a clinical perspective is to identify unique characteristics of the various sacral tumors instead of the nonspecific and commonly overlapping symptoms presented clinically. This review will focus on the tumors that are primarily localized to the sacrum, however, metastatic lesions to the sacrum will also be described in detail. Further, surgical interventions for the treatment of various sacral tumors will be described (including anterior and posterior approaches) as well as recommended postoperative management to minimize risk of recurrence and other adverse events. The objective is to identify genetic markers, patient presentations, radiographic imaging, histological features, and other highly sensitive and specific tests that may guide the diagnosis, staging, and treatment of sacral tumors regardless of tissue origin or severity.

SIGNS AND SYMPTOMS

While some sacral tumors are associated with specific demographics or manifestations, they largely exhibit similar signs and symptoms, thus making it difficult or impossible to diagnose the tumor type based on clinical presentation alone [Table 1].^[32] The most common presenting symptom is local pain and tenderness resulting from periosteal stretching from cortical expansion, mass effect, and compression of neighboring structures. This pain typically occurs at night and can be exacerbated by Valsalva maneuvers.^[5,32] Acute onset of pain without trauma or prodromal symptoms can indicate pathological fracture with pain from neural compression.^[5] Clinical progression depends on the tumor's anatomic location within the sacrum and the extent of expansion or infiltration.^[32] Local sacroiliac joint pain can occur with lateral tumor expansion while large lower sacral tumors are often palpated as a mass on rectal examination.^[12,27] Increasing compression of nerve roots can impair reflex arcs and provokes multiradicular sensory deficits to the uni- or bilateral buttocks, posterior thigh, leg, external genitalia, and perineum depending on the level of tumor extension.^[32] Late stage invasion of the gluteus maximus and piriformis

Table 1: Clinical manifestations of sacral tumors.

Diagnosis	Signs and symptoms
General	<ul style="list-style-type: none"> • Local pain/tenderness • Local joint pain • Radiculopathy • Dermatomal sensory deficits – dermatomal distribution • Myotomal motor deficits • Impaired reflex arcs • Bladder and bowel sphincter dysfunction • Sexual dysfunction • Palpable rectal mass
Benign tumors	
Giant cell tumor	<ul style="list-style-type: none"> • Mean duration of pain onset until diagnosis=3–8 months • Local pain over lumbosacral junction or sacrum • Palpable rectal mass
Aneurysmal bone cyst	<ul style="list-style-type: none"> • Mean duration of pain onset until diagnosis=2 years • Lumbosacral radicular pain • Lumbosacral sensorimotor dysfunction
Sacral meningeal cyst	<ul style="list-style-type: none"> • Sacral radicular pain • Sacral paresthesia – especially perineal • Rarely sphincter dysfunction, sensory loss, or motor weakness
Malignant tumors	General systemic symptoms – fever, malaise, anorexia, weight loss
Chordoma	<ul style="list-style-type: none"> • Sharp or dull continuous pain – frequently rectal • Sacral radicular pain • Palpable rectal mass
Lymphoma, multiple myeloma, Ewing's sarcoma, chondrosarcoma, osteosarcoma	<ul style="list-style-type: none"> • Progressive continuous local pain • Lumbosacral radicular pain • Lumbosacral sensorimotor dysfunction • Sphincter dysfunction • Constipation
Sacral ependymoma	<ul style="list-style-type: none"> • Mean duration of pain onset until diagnosis=2–3 years • Lumbosacral radicular pain • Lumbosacral sensorimotor dysfunction • Cauda equina syndrome
Sacral schwannoma, sacral meningioma	<ul style="list-style-type: none"> • Mean duration of pain onset until diagnosis=5 years • Lumbosacral radicular pain • Sacral paresthesia and dysesthesia • Rare motor symptoms or sphincter dysfunction
Sacrococcygeal teratoma	<ul style="list-style-type: none"> • Exophytic mass between anus and coccyx covered by normal skin

(Contd...)

Table 1: (Continued).

Diagnosis	Signs and symptoms
Neuroblastoma, ganglioneuroma, neurofibroma	<ul style="list-style-type: none"> • Dystocia • Motor deficits develop in childhood • Constipation and urinary retention
	<ul style="list-style-type: none"> • Chronic constipation • Urinary obstruction or frequency • Dystocia • Dysmenorrhea • Headache
Anterior sacral meningocele	

muscles can cause motor deficits from impaired hip extension and external rotation.^[27] Anterior expansion into the presacral space with unilateral impingement of S2 or S3 is usually associated with mild-to-moderate bladder, bowel, and sexual dysfunction while bilateral lesions always result in complete dysfunction. Moreover, direct compression of the rectum not only impedes bladder and bowel motility but also uterine function, leading to dystocia. There is no evidence of tumors crossing the presacral fascia to invade the rectum.^[32] Uni- and bilateral lesions to S4 and/or S5 roots are not associated with autonomic dysfunction.^[27]

TYPES OF SACRAL TUMORS

Primary bone tumors can be differentiated by histologic origin. The most widely adopted pathologic classification system for bone tumors is the World Health Organization classification system [Table 2]. This classification system also differentiates benign and malignant bone tumors.^[13]

The benign and malignant pathologies that can present at in the sacral level are further classified as metastatic disease, congenital tumors, primary bone tumors, or primary neurogenic tumors, as outlined in [Table 3].

Metastasis

Osseous metastasis is one of the most frequent and debilitating manifestations of advanced cancer. Metastatic bone tumors are more common than primary bone tumors.^[10] Nearly half of sacral tumors are metastases.^[7] In adults, 80% of osseous metastasis are caused by primary breast, prostate, or lung cancer.^[31] In pediatric patients, bony metastasis are most commonly associated with rhabdomyosarcoma, neuroblastoma, or clear cell sarcoma of the kidney.^[16] Rare sacral metastasis from rare primary cancers including malignant schwannoma, medulloblastoma, and angiomyolipoma has been reported.^[7] If suspecting metastatic disease to bone from an unknown primary malignancy, workup should include CT of the chest, abdomen, and pelvis.

Table 2: Histologic origin of benign and malignant primary bone tumors.

Histologic origin	Benign	Malignant
Hematopoietic		Myeloma Lymphoma
Chondrogenic	Osteochondroma Chondroma Chondroblastoma Chondromyxoid fibroma	Chondrosarcoma
Osteogenic	Osteoid Osteoma Osteoblastoma	Osteosarcoma
Fibrogenic	Fibroma	Desmoplastic Fibroma Fibrosarcoma
Vascular	Hemangioma Lymphangioma	Hemangiopericytoma Chordoma
Notochord		Ewing's tumor Malignant giant cell tumor Adamantinoma
Unknown	Giant cell tumor	

Bone scans can also be used to evaluate the extent of osseous disease. Osteolytic bone metastasis has a hypointense signal on T1-weighted sequences and an isointense signal on T2-weighted sequences in comparison to bone marrow.^[33]

Congenital tumors

Teratoma

The most common sacral tumors in the pediatric population are sacrococcygeal teratoma.^[24] The lesion may be diagnosed on prenatal ultrasound or in the neonatal period. Teratomas are solid lesions comprised of tissue from each germ cell layer. Over 90% of congenital sacrococcygeal teratomas are benign, but risk of malignant transformation increases with increasing age.^[9]

Hamartoma

Congenital spinal hamartomas are comprised of well-differentiated mesodermal and ectodermal tissue.^[25] This lesion is most commonly associated with neurofibromatosis type 1 or spinal dysraphism. Hamartomas may be overt on physical examination with occult spinal involvement.^[25] Therefore, imaging is warranted. Tissue biopsy is particularly helpful for differentiating hamartomas from more common lesions such as lipomas.

Dermoid cyst

As the neural groove begins to seal between the 3rd and 5th weeks of embryonic life, inclusion of ectodermal elements can form a dermoid cyst.^[28] These midline closure defects

Table 3: Classification of sacral tumors.

Metastasis	Congenital tumors	Primary bone tumors	Primary neurogenic tumors
Breast (adult)	Teratoma	Giant cell tumor	Schwannoma
Prostate (adult)	Hamartoma	Aneurysmal bone cyst	Neurofibroma
Lung (adult)	Dermoid cyst	Chordoma	
Rhabdomyosarcoma (pediatric)	Tarlov cyst	Lymphoma	
Neuroblastoma (pediatric)	Meningocele	Multiple myeloma	
Clear cell sarcoma (pediatric)		Ewing's sarcoma	
Malignant schwannoma		Chondrosarcoma	
Medulloblastoma		Osteosarcoma	
Angiomyolipoma		Chondromyxoid fibroma	

have a predilection for the lumbosacral region when located in the spine. Approximately 20% of lesions are associated with a dural sinus. On CT, dermoid cysts appear as well-defined lesions with minimal attenuation. Notably, calcification or teeth may be observed on CT. On MR, T1-weighted sequences reveal hyperintensity localizing sebaceous gland secretions and lipid metabolites. The solid portions of the lesion are often hyperintense yet heterogeneous.^[28]

Perineural cyst

Also referred to as Tarlov cysts, perineural cysts are caused by meningeal dilations of the spinal nerve root sheath. Perineural cysts involving the sacrum can lead to profound bony erosion, and cases of resultant compression fracture have been reported.^[34]

Meningocele

Anterior meningocele is caused by herniation of the dural sac through a sacral defect. The pathognomonic radiographic finding is the "scimitar sign," which describes a sacrum with a round, concave border devoid of any destruction.^[23]

Primary bone tumors

Chordoma

Chordomas are the most common primary tumor of the sacrum and arise from notochordal tissue. Chordomas, which arise from notochordal remnants in the sacrum, are the most common malignant primary sacral tumor.^[9] They have been reported in neonates but can occur at any age and most commonly present in middle-aged adults. The characteristic appearance of a chordoma on radiograph is midline osseous expansion and lytic destruction with intertumoral calcifications. CT also demonstrates a large soft-tissue mass with an average size of 10 cm.^[33] MRI reveals nonspecific destructive midline lesion with a large soft-tissue mass. T1-weighted images show hypo- to isointensity. High signal intensity is seen on T2-weighted images. Epithelioid cells arranged in cords with vacuolated, foamy cells are present on biopsy.^[8,33]

Aneurysmal bone cyst (ABC)

ABCs are benign tumors that can be locally aggressive. Large lesions can cause mass effect or pathologic fracture. Imaging often reveals an expansile lytic lesion with a thin calcific rim and characteristic multiloculated spaces with fluid levels.^[8] Surgical resection is the most common definitive treatment for ABCs, but radiotherapy, embolization, or sclerotherapy have also been used.

Giant cell tumor

Giant cell tumors predominantly manifest in the extremities, but most often occur at the sacrum when involving the axial skeleton. Although primary giant cell tumors are histologically benign, metastases to the lung have occasionally been reported.^[26] On radiography, GCTs appear radiolucent. CT demonstrates soft-tissue attenuation with sharp margins. GCTs have a variable appearance on MR, often demonstrating heterogeneous, low-intensity signal on both T1- and T2-weighted sequences.^[8] Biopsy will reveal multinucleated, osteoclastic giant cells dispersed throughout spindle cell stroma.^[8] ABCs and GCTs are often managed similarly. Surgical resection is standard.

Lymphoma

Primary lymphoma of the bone is a rare round cell malignancy that can be locally destructive. T1-weighted images show an ill-defined soft-tissue mass.^[33]

Multiple myeloma

Unifocal multiple myeloma manifests as a solitary osseous plasmacytoma. On radiograph or CT, solitary plasmacytoma appears as an expansive lytic mass with peripheral sclerosis. T1-weighted MRI demonstrates low signal intensity, and T2-weighted images will display postcontrast enhancement.^[30]

Ewing's sarcoma

Ewing's sarcoma occurs most frequently in young males. On imaging, Ewing's sarcoma appears as an osteolytic lesion with

soft-tissue component. The lesion commonly appears as a homogenous hypointense signal on T1-weighted images and an isointense signal on T2-weighted MR.

Chondrosarcoma

Chondrosarcomas are associated with a lobular appearance on imaging and pathology commonly demonstrates lobules and chondroid matrix. Characteristic radiographic finding is an osteolytic right sacral mass with a soft-tissue component and intratumoral chondroid-type calcifications.^[21]

Osteosarcoma

Osteosarcomas account for approximately 4% of all sacral primary bone tumors. Sunburst calcifications are characteristic on imaging and spindle cells with osteoid matrix characteristic on biopsy.^[33]

Chondromyxoid fibroma (CMF) of the sacrum

CMFs of the sacrum are a rare benign cartilaginous tumor that histologically is characterized by hypochromic lobules of stellate or spindle-shaped cells. The tumor stains positive for S-100, Sox 9, and Type II collagen.^[29]

Primary neurogenic tumors

Schwannoma

Schwannomas are associated with a characteristic appearance on imaging. MR reveals a large, well-defined heterogeneous mass that may be associated with minor underlying erosion. Cystic formation, hemorrhage, and necrosis may also be apparent. In contrast to neurofibromas, schwannomas are encapsulated.^[8]

Neurofibroma

Neurofibroma originates in nerve fascicles comprised of Schwann cells, fibroblasts, mast cells, and axons. Neurofibromas appear radiolucent and well circumscribed on imaging. Biopsy is characterized by short spindle cells with long, wavy nuclei that stain positive for S100.^[33]

Staging

At present, two systems are available for staging primary malignant bone tumors – the Musculoskeletal Tumor Society System (MSTS) and the American Joint Committee on Cancer (AJCC) system. The staging system adopted by the MSTS was first described by Enneking *et al.* in 1980 and was based on three criteria: extent of tumor, metastasis, and grade.^[11] Each criterion is defined in [Table 4]. The AJCC system is based on the size of the tumor, lymph node involvement, metastasis,

and grade, as summarized in [Table 5].^[1] Notably, the AJCC staging system does not apply to primary lymphoma of the bone or multiple myeloma.

SURGICAL APPROACHES

Optimal surgical technique typically prefers wide surgical margins because it prevents incomplete resection that can lead to local regrowth. A wide tumor resection includes a continuous encasement of healthy tissue around the tumor.^[37] Conversely, if the dissection occurred along the pseudocapsule, a marginal definition was given.^[14] Benign and/or low-grade tumors within the spinal canal or within dorsal elements of the sacrum can be approached with a sacral laminectomy.^[3] Tumors strictly confined within the posterior compartments can also be performed with a sacral laminectomy, all others will require a different surgical approach. To achieve a sacral laminectomy, patients are placed prone on a flat, open table and a midline incision is made. Dissection of the dorsal musculature off the sacrum occurs, along with sacrifice of the dorsal rami. Within the L5-S1 level, the supraspinous and interspinous ligaments are resected. This area allows for an appropriate starting point for the sacral laminectomy.^[3]

While institutions have varying criteria for surgical approaches, a combination of anterior and posterior

Table 4: Enneking staging system for primary malignant tumors of the bone.^[11]

Enneking staging system			
Stage IA	T1	M0	G1
Stage IB	T2	M0	G1
Stage IIA	T1	M0	G2
Stage IIB	T2	M0	G2
Stage III	T1 or T2	M1	G1 or G2

T1 designates an intracompartmental tumor. T2 designates an extracompartmental tumor. M0 signifies no regional or distant metastasis. M1: Regional or distant metastasis. G1: Low grade, G2: High grade

Table 5: The 8th Edition AJCC staging system for primary malignant tumors of bone.^[1]

AJCC anatomic stage/prognostic groups				
Stage IA	T1	N0	M0	G1,2 Low grade, GX
Stage IB	T2	N0	M0	G1,2 Low grade, GX
	T3	N0	M0	G1,2 Low grade, GX
Stage IIA	T1	N0	M0	G3,4 High grade
Stage IIB	T2	N0	M0	G3,4 High grade
Stage III	T3	N0	M0	G3,4
Stage IVA	Any T	N0	M1a	Any G
Stage IVB	Any T	N1	Any M	Any G
	Any T	Any N	M1b	Any G

approach has traditionally been used in total and high sacrectomies due to the control it provides surgeons toward the rectum and vasculature anterior to the sacrum [Table 6].^[2,6,35,39] The anterior approach provides the surgeon opportunity to dissect the rectum and internal iliac vessels away from the anterior surface of the sacrum. Operatively, a longitudinal midline incision is made 5 cm above the umbilicus down to the lower abdomen.^[22] Both ureters are identified and cleared off from the tumor using a transperitoneal approach. Ligation of the internal iliac arteries, veins, and middle sacral vessels also occurs. The rectum is mobilized off the tumor and then either the L5-S1 or L4-L5 disc is exposed and partially removed.^[22] Gauze was used as landmarks for the posterior osteotomies and isolated the tumor from the rectum, ureters, and vessels by packing the gauze anterior to the tumor. Performance of

the posterior approach then allows for tumor removal and additional spine/pelvic reconstruction.^[37]

Institutions have used a posterior-only approach in middle, low, and distal sacrectomies, as an anterior-posterior approach shows itself to be implausible.^[6] Posterior-only approach can be performed for tumors that failed to invade pelvic organs or extend past the lumbosacral junction.^[4] However, tumor invasion into the presacral fascia, rectum, and iliac vessels excludes the use of the posterior-only approach.^[6] The technique involves a midline posterior approach coupled with bilateral iliac osteotomies and midline osteotomy or discectomy and transperineal dissection, allowing delivery of the *en bloc* sacral specimen. The dissection carries down lateral to the sacrum to release the presacral fascia, the sacrotuberous ligaments, the sacrospinous ligaments, and the piriformis.^[22]

Table 6: Summarization of literature publishing surgical approaches, inclusion/exclusion criteria, and results.

Author	Surgical approach	Inclusion/exclusion criteria	Results
Clarke <i>et al.</i> ^[6]	Posterior-only Approach	Exclusion criteria <ul style="list-style-type: none"> • Tumor invasion into the rectum requiring rectal diversion • Tumor extension caudally above the L5/S1 disc space • Involvement of the iliac vessels Inclusion criteria <ul style="list-style-type: none"> • Including high and total sacrectomy 	Surgical margins were marginal in 34 cases out of 36 of the cases. There was a correlation with functional outcomes with extent of sacrectomy and roots sacrificed
Zang <i>et al.</i> ^[39]	Posterior-only approach	Not specified	With a sample size of 10 patients, follow-up occurred for a mean of 22 months. Adequate margins were seen in eight patients and two patients saw recurrence. Two patients died from disease progression and a 5-year overall survival rate by Kaplan–Meier analysis was 70%
Fuchs <i>et al.</i> ^[17]	Anterior-posterior or posterior-only approach	Inclusion for anterior-posterior <ul style="list-style-type: none"> • All lesions that extended above S2 Inclusion for posterior only <ul style="list-style-type: none"> • All lesions below S3 For lesions extending to S2 or S3, 17 had a posterior-only approach while 17 had an anterior-posterior approach	Survival rate was significantly higher in approaches in which wide margin was achieved. Although surgical approach did not influence survival ($P=0.138$), an anterior-posterior approach led to more successful wide surgical margins
Angelini and Ruggieri ^[2]	(Modified Osaka) Posterior-only approach	Criteria for the selection of posterior-only approach <ul style="list-style-type: none"> • Proximal to S1 not indicated – anterior-posterior approach preferred • S1 or S2 level – central lesion with no or minor involvement of sacroiliac joints. Minimal pelvic invasion • S3 level or below – always perform posterior only 	Performed resection on 13 patients, with 9 proximal resections and 4 distal resections. Wide margins were achieved in 10 patients. Nine patients were disease free at a mean follow-up of 35.5 months
Sahakitrungruang <i>et al.</i> ^[35]	Anterior-posterior approach	Not specified	Two total sacrectomies, one extended total sacrectomy, and five subtotal S1 sacrectomies were performed. Wide margins were achieved in all patients, and no patients developed recurrence from the primary tumor at 4 years mean follow-up

A sacrifice of nerve roots with functional impairment presents itself in both partial and complete sacrectomies, but this sacrifice is necessary to achieve proper local control.^[14] Quality of surgical margins has been described as the main prognostic factor of local recurrence.^[37] When a sacral tumor resection with a minimum of 50% of the sacroiliac joint is affected, reconstruction becomes mandatory. Recent techniques into reconstruction following sacrectomy include the modified Galveston reconstruction, triangular frame reconstruction, sacral rod reconstruction, four-rod reconstruction, and bilateral fibular flaps reconstruction.^[20,22,40] In addition, soft-tissue reconstruction has been used due to the large sacrectomy defect. Systematic review has shown the elevated complications of spinopelvic reconstruction and only 24–44% of patients ambulating independently after reconstruction.^[22] In an iliosacral resection that occurs medial to or through or lateral but close to (less than 3 cm from posterior iliac spine) the sacroiliac joint, reconstruction may not be needed.^[22] Consideration of a total sacrectomy without spinopelvic reconstruction in patients with this resection has been shown a reasonable alternative to reconstruction surgery.^[22]

POSTOPERATIVE MANAGEMENT

The most common complaint after sacrectomy is sacral pain. Average duration of pain is 8 months with 15% reported risk of neuropathic pain and complex regional pain syndrome.^[15,19] In addition, as sacral nerve root sacrifice is common in sacrectomy, restoration of neurologic function is a critical component of successful postoperative management. This is achieved through a multimodal approach which can include opioid and nonopioid medications, patient-controlled anesthesia, early initiation of in-bed resistance training, and progressive mobilization toward sitting, standing, and ambulation as tolerated.^[22] Tilt tables are effective in initiating early mobilization to minimize motor deficits as well as for preventing early postoperative orthostatic hypotension that arises secondary to prolonged supine immobilization and activity restrictions.^[19] A case study by Guo and Yadav demonstrated improved pain control and earlier mobilization with use of lumbar-sacral corset external orthosis by decreasing lumbar-sacral load and motion during patient transfers and rehabilitation exercises.^[19]

Surgical site infection and wound dehiscence are also common complications after sacrectomy. Enteral feeding is conventionally associated with improved postoperative nutritional and immunologic status. However, operative disruption sacral nerve roots leading to bladder and bowel dysfunction increases the risk of infection as fecal leak can contaminate surgical wounds and provide a nidus for infection. The most common bacterial pathogens implicated are *Enterococcus* (23%) and *Escherichia coli* (20%). To abate this risk, Gao et al. endorsed

early postoperative fasting and total parenteral nutrition while others may elect to place an ostomy.^[18,19,22] As a result, it may be pertinent to provide education on ostomy care and use of external catheters or intermittent catheterization for neurogenic bladder management. Other important considerations include patient and family education on home care management and counseling services for coping with functional losses, as well as referral for chronic pain management, sexual dysfunction, and physical or occupational rehabilitation.^[38]

CONCLUSION

Sacral tumors continue to pose a challenge in the field of spinal surgery, as their slow growth and relative clinical silence over prolonged periods promote the onset of debilitating symptoms once clinically manifested. In this review, sacral tumors were identified based on unique clinical presentations and markers of diagnosis. Surgical approaches for the resection of sacral tumors were described as well as ideal postoperative management to mitigate long-term sequelae and tumor recurrence.

To quickly identify the onset of tumor formation in the sacrum, it is imperative patients engage in regular screenings for highly metastatic cancers including those of the lung, prostate, and breast which are commonly found to metastasize to bone. Because metastasis to bone accounts for almost half of all sacral tumor cases, regular screening allows the physician ample opportunity to utilize diagnostic imaging to investigate an oncological etiology of a patient's localized chief complaint, particularly from individuals with a prior history of any of the aforementioned primary cancerous lesions. Research and development into genetic markers of individual tumors would aid in rapid detection that can be missed by diagnostic imaging in the early stages of tumor formation. Unfortunately, markers for sacral tumors are rare and in many cases nonspecific. Until primary tumor markers of high sensitivity and specificity are shown to be clinically viable, imaging will remain the most effective diagnostic tool currently available. As such, caution must be placed with chief complaints of low back pain or radiculopathy and should include sacral tumors in the differential diagnosis with appropriate follow-up.

Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Amin MB, Greene FL, Edge SB, Compton CC, Gershenwald JE, Brookland RK, *et al.* The eighth edition AJCC cancer staging manual: Continuing to build a bridge from a population-based to a more personalized approach to cancer staging. *CA Cancer J Clin* 2017;67:93-9.
2. Angelini A, Ruggieri P. A new surgical technique (modified Osaka technique) of sacral resection by posterior-only approach: Description and preliminary results. *Spine (Phila Pa 1976)* 2013;38:E185-92.
3. Benzel EC, Steinmetz MP. *Benzel's Spine Surgery: Techniques, Complication Avoidance, and Management*. 4th ed. Philadelphia, PA: Elsevier; 2017.
4. Bydon M, De la Garza-Ramos R, Gokaslan ZL. Editorial: Total sacrectomy for malignant sacral tumors via a posterior-only approach. *J Neurosurg Spine* 2015;22:561-2.
5. Ciftdemir M, Kaya M, Selcuk E, Yalniz E. Tumors of the spine. *World J Orthop* 2016;7:109-16.
6. Clarke MJ, Dasenbrock H, Bydon A, Sciubba DM, McGirt MJ, Hsieh PC, *et al.* Posterior-only approach for en bloc sacrectomy: Clinical outcomes in 36 consecutive patients. *Neurosurgery* 2012;71:357-64; discussion 364.
7. David OI, Lupaşcu-Ursulescu CV, Lupaşcu CD, Sandu AM, Strâmbu VD, Cristian DA, *et al.* Histopathological diagnosis and its correlations with anatomoclinical features, surgical approach and postoperative prognosis in sacral tumors. *Rom J Morphol Embryol* 2017;58:393-408.
8. Deutsch H, Mummaneni PV, Haid RW, Rodts GE, Ondra SL. Benign sacral tumors. *Neurosurg Focus* 2003;15:E14.
9. Diel J, Ortiz O, Losada RA, Price DB, Hayt MW, Katz DS. The sacrum: Pathologic spectrum, multimodality imaging, and subspecialty approach. *Radiographics* 2001;21:83-104.
10. Ebraheim NA, Thomas BJ, Fu FH, Muller B, Vyas D, Niesen M, *et al.* Orthopedic surgery. In: Brunicaardi FC, Andersen DK, Billiar TR, Dunn DL, Kao LS, Hunter JG, *et al.*, editors. *Schwartz's Principles of Surgery*. 11th ed. New York: McGraw-Hill Education; 2019.
11. Enneking WF, Spanier SS, Goodman MA. A system for the surgical staging of musculoskeletal sarcoma. *Clin Orthop Relat Res* 1980;153:106-20.
12. Feldenzer JA, McGauley JL, McGillicuddy JE. Sacral and presacral tumors: Problems in diagnosis and management. *Neurosurgery* 1989;25:884-91.
13. Fletcher CD, World Health Organization. *WHO Classification of Tumours of Soft Tissue and Bone*. Lyon: IARC Press; 2013.
14. Fournay DR, Rhines LD, Hentschel SJ, Skibber JM, Wolinsky JP, Weber KL, *et al.* En bloc resection of primary sacral tumors: Classification of surgical approaches and outcome. *J Neurosurg Spine* 2005;3:111-22.
15. Francis GJ, Ngo-Huang A, Rhines LD, Bruera E. The challenges of providing rehabilitation for patients undergoing sacrectomy: Two case reports. *Eur J Phys Rehabil Med* 2019;55:526-9.
16. Fritchie KJ, John I. Soft tissue and bone pathology. In: Reisner HM, editor. *Pathology: A Modern Case Study*. 2nd ed. New York: McGraw-Hill Education; 2020.
17. Fuchs B, Dickey ID, Yaszemski MJ, Inwards CY, Sim FH. Operative management of sacral chordoma. *J Bone Joint Surg Am* 2005;87:2211-6.
18. Gao S, Zheng Y, Liu X, Tian Z, Zhao Y. Effect of early fasting and total parenteral nutrition support on the healing of incision and nutritional status in patients after sacrectomy. *Orthop Traumatol Surg Res* 2018;104:539-44.
19. Guo Y, Yadav R. Improving function after total sacrectomy by using a lumbar-sacral corset. *Am J Phys Med Rehabil* 2002;81:72-6.
20. Hugate RR, Dickey ID, Phimolsarnti R, Yaszemski MJ, Sim FH. Mechanical effects of partial sacrectomy: When is reconstruction necessary? *Clin Orthop Relat Res* 2006;450:82-8.
21. Kemp WL, Burns DK, Brown TG. *Pathology of the Bones and Joints. Pathology: The Big Picture*. Ch. 19. New York: The McGraw-Hill Companies; 2008.
22. Kiatischevi P, Piyaskulkaew C, Kunakornsawat S, Sukunthanak B. What are the functional outcomes after total sacrectomy without spinopelvic reconstruction? *Clin Orthop Relat Res* 2017;475:643-55.
23. Kwaan MR, Stewart DB Sr., Dunn KB. Colon, rectum, and anus. In: Brunicaardi FC, Andersen DK, Billiar TR, Dunn DL, Kao LS, Hunter JG, *et al.*, editors. *Schwartz's Principles of Surgery*. 11th ed. New York: McGraw-Hill Education; 2019.
24. Lam CH, Nagib MG. Nonteratomatous tumors in the pediatric sacral region. *Spine (Phila Pa 1976)* 2002;27:E284-7.
25. Malelak EB, Lauren C, Argie D, Nugraheni T. Congenital midline spinal hamartoma in a 5-month-old infant. *World Neurosurg* 2020;145:142-7.
26. Manaster BJ, Graham T. Imaging of sacral tumors. *Neurosurg Focus* 2003;15:E2.
27. Mavrogenis AF, Patapis P, Kostopanagiotou G, Papagelopoulos PJ. Tumors of the sacrum. *Orthopedics* 2009;32:342-56.
28. Mhatre P, Hudgins PA, Hunter S. Dermoid cyst in the lumbosacral region: Radiographic findings. *AJR Am J Roentgenol* 2000;174:874-5.
29. Minasian T, Claus C, Hariri OR, Piao Z, Quadri SA, Yuhan R, *et al.* Chondromyxoid fibroma of the sacrum: A case report and literature review. *Surg Neurol Int* 2016;7 Suppl 13:S370-4.
30. Patel N, Maturen KE, Kaza RK, Gandikota G, Al-Hawary MM, Wasnik AP. Imaging of presacral masses-a multidisciplinary approach. *Br J Radiol* 2016;89:20150698.
31. Patel SR. Soft tissue and bone sarcomas and bone metastases. In: Jameson JL, Fauci AS, Kasper DL, Hauser SL, Longo DL, Loscalzo J, editors. *Harrison's Principles of Internal Medicine*. 20th ed. New York: McGraw-Hill Education; 2018.
32. Payer M. Neurological manifestation of sacral tumors. *Neurosurg Focus* 2003;15:E1.
33. Peh WC, Koh WL, Kwek JW, Htoo MM, Tan PH. Imaging of painful solitary lesions of the sacrum. *Australas Radiol* 2007;51:507-15.
34. Puffer RC, Gates MJ, Copeland W 3rd, Krauss WE, Fogelson J. Tarlov cyst causing sacral insufficiency fracture. *Oper Neurosurg (Hagerstown)* 2017;13:E4-7.
35. Sahakitrungruang C, Chantra K, Dusitanond N, Atittharnsakul P, Rojanasakul A. Sacrectomy for primary sacral tumors. *Dis Colon Rectum* 2009;52:913-8.
36. Stephens M, Gunasekaran A, Elswick C, Laryea JA, Pait TG, Kazemi N. Neurosurgical management of sacral tumors: Review of the literature and operative nuances. *World*

- Neurosurg 2018;116:362-9.
37. Varga PP, Szövérfi Z, Lazary A. Surgical treatment of primary malignant tumors of the sacrum. *Neurol Res* 2014;36:577-87.
 38. Wang Y, Liang W, Qu S, Zhang Y, Du Z, Ji T, *et al.* Assessment of patient experiences following total sacrectomy for primary malignant sacral tumors: A qualitative study. *World J Orthop* 2019;120:1497-504.
 39. Zang J, Guo W, Yang R, Tang X, Li D. Is total en bloc sacrectomy using a posterior-only approach feasible and safe for patients with malignant sacral tumors? *J Neurosurg Spine* 2015;22:563-70.
 40. Zhu R, Cheng LM, Yu Y, Zander T, Chen B, Rohlmann A. Comparison of four reconstruction methods after total sacrectomy: A finite element study. *Clin Biomech (Bristol, Avon)* 2012;27:771-6.

How to cite this article: Fiani B, Runnels J, Rose A, Kondilis A, Wong A, Musch BL. Clinical manifestations, classification, and surgical management of sacral tumors and the need for personalized approach to sacrectomy. *Surg Neurol Int* 2021;12:209.