The Value of Preoperative Biopsy in the Management of Solid Presacral Tumors

Amit Merchea, M.D.¹ • David W. Larson, M.D., M.B.A.¹ • Martin Hubner, M.D.¹ Doris E. Wenger, M.D.² • Peter S. Rose, M.D.³ • Eric J. Dozois, M.D.¹

1 Division of Colon and Rectal Surgery, Mayo Clinic, Rochester, Minnesota

2 Department of Radiology, Mayo Clinic, Rochester, Minnesota

3 Department of Orthopedic Surgery, Mayo Clinic, Rochester, Minnesota

BACKGROUND: Surgical decision making and the use of neoadjuvant therapy in the management of solid presacral tumors rely greatly on an accurate preoperative diagnosis. The utility of preoperative biopsy has been questioned because of potential complications and the increasing accuracy of modern imaging.

OBJECTIVE: The aim of this study was to analyze biopsy-related morbidity and to compare the accuracy of imaging versus biopsy in making a preoperative diagnosis.

DESIGN: This study is a retrospective review of all patients who underwent biopsy of presacral tumors at Mayo Clinic Rochester between 1990 and 2010. The demographics, pathology, complications of biopsy, and imaging were reviewed. Biopsy results and radiologic findings were matched with the final pathology and analyzed.

SETTINGS: This study was conducted at a tertiary care center.

PATIENTS: Adult patients with solid presacral tumors who underwent preoperative biopsy were evaluated.

MAIN OUTCOME MEASURES: The primary outcomes measured were the biopsy-related complications and the accuracy of preoperative imaging and biopsy in comparison with final pathology.

Correspondence: Eric J. Dozois, M.D., Division of Colon and Rectal Surgery, Mayo Clinic, 200 First St SW, Rochester, MN 55905. E-mail: dozois.eric@mayo.edu

Dis Colon Rectum 2013; 56: 756–760 DOI: 10.1097/DCR.0b013e3182788c77 © The ASCRS 2013 **RESULTS:** Seventy-six biopsies were performed in 73 patients. Fifty-six patients underwent percutaneous biopsies, 14 underwent open biopsies, and 3 underwent both. Biopsy-specific complications included 2 hematomas (1 open, 1 percutaneous). Preoperative biopsy correlated with the postoperative pathologic diagnosis in 63 patients (91%). Of the 6 solid presacral tumors diagnosed incorrectly on biopsy, 1 was falsely reported as benign. Sensitivity, specificity, and positive and negative predictive values of biopsy to detect malignant disease was 96%, 100%, 100%, and 98%. Ten of 35 patients (29%) with a definitive imaging diagnosis were given incorrect diagnoses. Sensitivity, specificity, and positive and negative predictive values of imaging to diagnose malignant disease was 83%, 81%, 83%, and 81%.

LIMITATIONS: This investigation was designed as a retrospective study.

CONCLUSION: Preoperative biopsy of presacral tumors is safe and highly concordant with postoperative pathology in comparison with imaging. Given the significant differences in therapeutic approach for benign versus malignant solid presacral tumors, as well as the current limitations of imaging, a percutaneous preoperative biopsy should be obtained to guide management decisions.

KEY WORDS: Retrorectal tumor; Presacral tumor; Biopsy; Biopsy complications.

The presacral space is a potential site of a group of heterogeneous benign and malignant tumors that often demonstrate indolent growth patterns leading to occult symptoms. These lesions are rare with an estimated incidence of 1/40,000 to 1/60,000.¹⁻³ Given their rarity and diverse pathologic spectrum, patients with these tumors are often a diagnostic and therapeutic challenge.

Financial Disclosures: None reported.

Poster presentation at the meeting of The American Society of Colon and Rectal Surgeons, San Antonio, TX, June 2 to 6, 2012.

Presacral lesions can be solid or cystic, with most cystic lesions being benign.⁴ In contrast, although solid tumors can be both benign and malignant, many are malignant with a wide range of pathologic subtypes, which complicates their management.^{3–8}

An accurate preoperative diagnosis of solid or heterogeneous tumors in the presacral space is crucial, because the clinical management of benign and malignant lesions differs considerably. Surgically, a wide-margin, en-bloc oncologic resection is indicated for most malignant lesions, but a close-margin approach should be attempted for benign lesions to spare function and avoid morbidity. Furthermore, the use of preoperative radiation and/ or chemotherapy may assist in optimizing oncologic outcome in some circumstances, but may be unnecessary for other lesions.

Historically, the diagnosis of these lesions preoperatively has been based on imaging characteristics alone. The use of advanced image-guided preoperative biopsy of these lesions has been described, but many authors question its utility and its role remains controversial. In fact, some consider any presacral tumor deemed amenable to surgical resection a contraindication to biopsy.^{6–11} The use of image-guided biopsy, whether by fine-needle aspiration or core-needle biopsy, for the diagnosis of diseases has become pervasive and its safety has been widely reported.^{12–14} Reports on the utility of biopsy of the presacral region are limited, secondary to the rarity of the disease and because many published reports deem it unnecessary.^{15–19}

The aims of this study were to analyze biopsy-related morbidity and to compare the accuracy of imaging versus biopsy in making a preoperative diagnosis in patients with solid presacral tumors.

MATERIALS AND METHODS

This retrospective cohort study included all patients who underwent a preoperative biopsy for a solid or heterogeneous presacral tumor at our institution between January 1, 1990 and December 31, 2010. After institutional review board approval was received, patients were identified from a prospectively maintained surgical pathology and tumor registry database. All adult patients (age ≥ 18) who underwent preoperative biopsy of a primary presacral tumor were included.

Patient records were reviewed for demographics, preoperative imaging, complications of biopsy, and operative pathology. The radiology reports and the results of preoperative biopsy were compared to assess concordance with postoperative pathology. The ability of preoperative biopsy and imaging to accurately make a diagnosis and, specifically, to differentiate benign versus malignant lesions was assessed. At our institution, both fine-needle aspiration and core-needle biopsy are performed in all patients undergoing image-guided biopsy. In our series, 4 patients elected not to undergo surgical resection. These 4 patients were excluded from the analysis of preoperative biopsy versus postoperative pathology; however, they were included in the analysis of imaging versus pathology with the assumption that the biopsy diagnosis was correct.

Descriptive statistics are reported as a percentage of the total and continuous variables as the median and range. Sensitivity, specificity, and positive and negative predictive values were calculated and reported.

RESULTS

Seventy-three patients (37 female) with presacral tumors underwent 76 biopsies. Median age was 43.7 years (range, 18–72 years). Median BMI was 26.4 (range, 17.7–59.4). Fifty-six patients (77%) underwent percutaneous biopsy, 14 had open biopsy (19%), and 3 patients (4%) had both (Fig. 1).

Biopsies were performed parasacrally or transperineally in 47 patients, transabdominally in 25, and transrectally in 3. Biopsy route was unknown in 1 patient; however, this was a CT-guided percutaneous biopsy (either transabdominal or transsacral). Of the 76 biopsies completed, 2 patients developed complications, a hematoma after an open biopsy and after a percutaneous biopsy. Neither hematoma was of clinical significance.

No MRI or CT imaging was done before biopsy in 4 patients (3 had open biopsies, 1 by ultrasound). Preoperative MRI or CT imaging was indeterminate in 34 patients (49%). Ten of 35 patients (29%) given a definitive diagnosis based on imaging were diagnosed incorrectly in comparison with postoperative pathology; 3 tumors were falsely diagnosed as benign, and 3 were falsely diagnosed as malignant. Of 27 patients who had a CT scan as the only diagnostic test, 4 (15%) had a correct diagnosis, whereas 12/18 (67%) undergoing only MRI had a correct diagnosis. Sensitivity, specificity, and positive and negative predictive values of imaging to diagnose malignant disease was 83%, 81%, 83%, and 81% (Table 1). Of those patients that underwent MRI alone, the sensitivity, specificity, and positive and negative predictive values in terms of predicting malignancy are 50%, 92%, 67%, and 85%.

In 33 patients, a benign result on preoperative biopsy guided the surgical team toward a conservative, nerveand function-sparing operative approach. Fifteen patients underwent neoadjuvant therapy; 10 of these patients had either a benign or indeterminate diagnosis based on imaging alone, but biopsy demonstrated a malignant process. Of the remaining 5 patients, a malignant diagnosis on imaging was confirmed on biopsy.

The surgical approach to resection included anterior-only (n = 33, 45%), combined anterior and posterior (n = 21, 29%), and posterior-only (15, 21%). Four patients

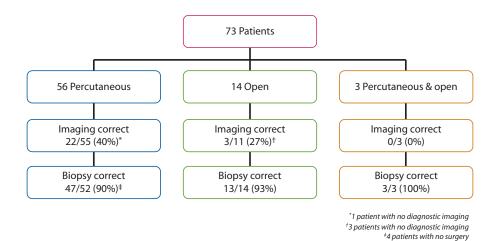


FIGURE 1. Biopsy methods and results compared with imaging.

(5%) elected not to undergo surgery. Median resected tumor size was 9.4 cm (range, 3.4-24 cm). The most common final pathologic diagnosis was schwannoma (n = 24, 33%), followed by sarcoma (n = 22, 30%) (Table 2).

Diagnosis based on preoperative biopsy correlated with the postoperative pathologic diagnosis in 63/69 patients (91%) who underwent surgical resection. Of the 6 tumors diagnosed incorrectly on biopsy, 1 was falsely benign (neurofibroma on biopsy; neurofibrosarcoma on final surgical pathology). The remaining 5 tumors were diagnosed incorrectly; however, the histopathologic biopsy diagnoses were still correct in terms of identifying benign versus malignant tissue. Sensitivity, specificity, and positive and negative predictive values of biopsy to detect malignant disease was 96%, 100%, 100%, and 98% (Table 1).

DISCUSSION

This review of preoperative biopsy of solid presacral tumors at our institution demonstrated that: (1) biopsy of these lesions is safe, (2) a significant percentage of patients (49%) could not be given a definitive diagnosis based on imaging alone and, of these, 35% were malignant tumors, (3) a greater percentage of patients undergoing MRI had a correct diagnosis in comparison with CT (67% vs 15%), and (4) there was a high concordance of preoperative biopsy with postoperative pathology (91%) in comparison with imaging alone.

TABLE 1.	Biopsy versus imaging in differentiating malignant and	
benign disease		

	Biopsy, %	Imaging, %
Sensitivity	96	83
Specificity	100	81
PPV	100	83
NPV	98	81

PPV = positive predictive value; NPV = negative predictive value.

The utility of preoperative biopsy of solid or heterogeneous lesions of the presacral space has been debated and is controversial. Recent publications claim no role for preoperative biopsy in the management of these lesions, stating that most information needed to make a diagnosis, and to plan surgery, can be obtained from advanced imaging such as MRI.^{3,8-11} Moreover, other authors have stated concerns about safety and tumor seeding, although these conclusions are based on anecdotal experience and referencing historical data.^{17,20} Of the imaging modalities currently available, MRI appears to have an advantage over CT because of superior soft-tissue contrast resolution that provides improved delineation of the anatomic extent of the tumor and superior tissue characterization.²¹ Furthermore, MRI may have improved ability over CT to differentiate benign versus malignant tumors, as demonstrated by the higher accuracy to discriminate these in our study. However, despite the increased correlation of MRI over CT with postoperative pathology, one-third of tumors in our series were still incorrectly diagnosed on imaging alone.

Selection of imaging modality can be individualized, because some tumors will benefit from either CT and/or MRI imaging. For presacral tumors with secondary sacral involvement, MRI demonstrates the extent of marrow infiltration and the relationship to exiting and traversing

TABLE 2. Final tumor pathology	
Final pathologic diagnosis	Number of patients (total = 73)
Schwannoma	24
Sarcoma	22
Neurofibroma	7
Tailgut cyst	6
Chordoma	5
Ependymoma	4
Ganglioneuroma	1
Unknown (no surgery)	4

neurologic structures better than CT alone. In our experience, when evaluating primary or metastatic tumors of the bone with an associated presacral soft tissue mass, CT provides an advantage over MRI in differentiating between benign and malignant lesions in the bone.

Fine-needle aspiration and core-needle biopsy techniques have been widely used and are established approaches in the diagnosis of soft-tissue and bony lesions.12,13 Previous reports have examined the utility of image-guided biopsy in diagnosing lesions at all levels of the spine; however, similar reports focusing on the presacral region are rare.^{15,19} One concern frequently reported when performing percutaneous biopsy is the possibility of needle-tract seeding; however, little evidence exists regarding the actual prevalence of this.²² As we have reported, it has been our practice to include the biopsy tract in the resected specimen in malignant cases.⁵ As such, biopsy tracts are placed in a near-midline approach to minimize gluteal contamination and facilitate resection. Needle insertion site may be marked with methylene blue to allow later identification. For tumors with bony involvement, direct midline biopsy is avoided to prevent contamination of the epidural compartment of the sacrum.

We believe there are 2 central issues when considering the role of preoperative biopsy in the management of solid presacral tumors: one is safety, and the other is how the results of the biopsy will impact management decisions regarding both the type of surgical resection and the role of neoadjuvant chemoradiation. In our current practice, we do not biopsy purely cystic lesions and we never biopsy lesions transrectally or transvaginally. Three patients in this series did undergo transrectal biopsy. These patients were biopsied before evaluation by a surgeon specializing in these tumors. We believe that performing biopsies in this manner significantly risks infection, can disrupt tissue planes, and increase the risk of biopsy-related fistulas, all of which have the potential to complicate subsequent resection.

For the evaluation of solid or heterogenous cystic lesions, our algorithm includes a parasacral or transperineal CT-guided biopsy and MRI. Because most nonchordoma malignant presacral tumors are sarcoma variants, it is our practice to use preoperative radiotherapy and, in selected cases, intraoperative radiation therapy for large, locally advanced high-grade tumors.⁵ Because we would not give neoadjuvant therapy without a tissue diagnosis, a biopsy is mandatory. Moreover, because all malignant presacral tumors get a wide-margin resection that often includes sacrectomy, we would not subject patients to urinary and sexual dysfunction, or other potentially morbid outcomes, without certainty that the tumor is malignant before the operation. In addition, because many benign, solid lesions are neurogenic in origin, we use a nerve-sparing approach that includes leaving the tumor pseudocapsule intact.^{5,23} For benign tumors, preservation of function is our

primary goal over complete tumor resection. We would not want to mistakenly use this approach in a malignant lesion, compromising the oncologic outcome.

Limitations of our study include its retrospective analysis and the inherent bias of a single-institution study. Moreover, the radiologic analysis was not standardized and included multiple radiologists with varying levels of subspecialty expertise. The ability to accurately predict the histologic diagnosis solely on imaging would likely be improved if interpretation is done by a radiologist with subspecialty expertise in musculoskeletal imaging. Finally, this review spans a 20-year period, improvements in imaging resolution and techniques have occurred that have led to improvements in the diagnostic capability of CT and MRI.

CONCLUSION

Preoperative biopsy of presacral tumors is safe and has a higher concordance with postoperative pathology in comparison with imaging alone. Interpretation of imaging by a radiologist with subspecialty expertise in complex pelvic tumors may increase diagnostic accuracy without biopsy. Given the current limitations of imaging to make a definitive diagnosis, percutaneous biopsy of solid or heterogeneous presacral tumors should be obtained preoperatively to facilitate decision making for the use of neoadjuvant therapies and for optimizing surgical planning.

REFERENCES

- 1. Whittaker LD, Pemberton JD. Tumors ventral to the sacrum. *Ann Surg.* 1938;107:96–106.
- 2. Spencer RJ, Jackman RJ. Surgical management of precoccygeal cysts. *Surg Gynecol Obstet*. 1962;115:449–452.
- Jao SW, Beart RW Jr, Spencer RJ, Reiman HM, Ilstrup DM. Retrorectal tumors. Mayo Clinic experience, 1960–1979. *Dis Colon Rectum*. 1985;28:644–652.
- Mathis KL, Dozois EJ, Grewal MS, Metzger P, Larson DW, Devine RM. Malignant risk and surgical outcomes of presacral tailgut cysts. *Br J Surg.* 2010;97:575–579.
- Dozois EJ, Jacofsky DJ, Billings BJ, et al. Surgical approach and oncologic outcomes following multidisciplinary management of retrorectal sarcomas. *Ann Surg Oncol.* 2011;18:983–988.
- Boscà A, Pous S, Artés MJ, Gómez F, Granero Castro P, García-Granero E. Tumours of the retrorectal space: management and outcome of a heterogeneous group of diseases. *Colorectal Dis.* 2012;14:1418–1423.
- Glasgow SC, Birnbaum EH, Lowney JK, et al. Retrorectal tumors: a diagnostic and therapeutic challenge. *Dis Colon Rectum.* 2005;48:1581–1587.
- Macafee DA, Sagar PM, El-Khoury T, Hyland R. Retrorectal tumours: optimization of surgical approach and outcome. *Colorectal Dis.* 2012;14:1411–1417.
- Böhm B, Milsom JW, Fazio VW, Lavery IC, Church JM, Oakley JR. Our approach to the management of congenital presacral tumors in adults. *Int J Colorectal Dis*, 1993;8:134–138.

- Lev-Chelouche D, Gutman M, Goldman G, et al. Presacral tumors: a practical classification and treatment of a unique and heterogeneous group of diseases. *Surgery*. 2003;133:473–478.
- Hobson KG, Ghaemmaghami V, Roe JP, Goodnight JE, Khatri VP. Tumors of the retrorectal space. *Dis Colon Rectum*. 2005;48: 1964–1974.
- 12. Sápi Z, Antal I, Pápai Z, et al. Diagnosis of soft tissue tumors by fine-needle aspiration with combined cytopathology and ancillary techniques. *Diagn Cytopathol*. 2002;26:232–242.
- Bommer KK, Ramzy I, Mody D. Fine-needle aspiration biopsy in the diagnosis and management of bone lesions: a study of 450 cases. *Cancer*. 1997;81:148–156.
- Welker JA, Henshaw RM, Jelinek J, Shmookler BM, Malawer MM. The percutaneous needle biopsy is safe and recommended in the diagnosis of musculoskeletal masses. *Cancer*. 2000;89:2677–2686.
- Bauer CA, Thompson RC, Blout ER. The active centers of Streptomyces griseus protease 3 and alpha-chymotrypsin: enzyme-substrate interactions remote from the scissile bond. *Biochemistry*. 1976;15:1291–1295.
- Schwartz HS, Spengler DM. Needle tract recurrences after closed biopsy for sarcoma: three cases and review of the literature. *Ann Surg Oncol.* 1997;4:228–236.

- 17. Verazin G, Rosen L, Khubchandani IT, Sheets JA, Stasik JJ, Riether R. Retrorectal tumor: is biopsy risky? *South Med J*. 1986;79:1437–1439.
- Woodfield JC, Chalmers AG, Phillips N, Sagar PM. Algorithms for the surgical management of retrorectal tumours. *Br J Surg.* 2008;95:214–221.
- Syed R, Bishop JA, Ali SZ. Sacral and presacral lesions: cytopathologic analysis and clinical correlates. *Diagn Cytopathol.* 2012;40:7–13.
- Cody HS 3rd, Marcove RC, Quan SH. Malignant retrorectal tumors: 28 years' experience at Memorial Sloan-Kettering Cancer Center. *Dis Colon Rectum*. 1981;24:501–506.
- Yang BL, Gu YF, Shao WJ, et al. Retrorectal tumors in adults: magnetic resonance imaging findings. *World J Gastroenterol*. 2010;16:5822–5829.
- 22. Robertson EG, Baxter G. Tumour seeding following percutaneous needle biopsy: the real story! *Clin Radiol.* 2011;66: 1007–1014.
- 23. Dozois EJ, Wall JC, Spinner RJ, et al. Neurogenic tumors of the pelvis: clinicopathologic features and surgical outcomes using a multidisciplinary team. *Ann Surg Oncol.* 2009;16: 1010–1016.