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Transsacral Approach to Resect a Gastrointestinal Stromal Tumor in the Rectum: Report of Two Cases

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Abstract
Gastrointestinal stromal tumors (GISTs) rarely arise in the rectum. Whereas a local resection with negative margins is generally considered adequate for resectable GISTs, a wide resection is usually indicated for rectal lesions because of the technical impossibility of local resection. We report the cases of two patients who underwent resection of a rectal GIST using a transsacral approach. Both patients had an uneventful postoperative course, and no evidence of recurrence has been identified. The transsacral approach appears to be less invasive and should be considered as the treatment of choice for a rectal GIST.

Key words Gastrointestinal tumor · Rectum · Transsacral approach

Introduction

Gastrointestinal stromal tumors (GISTs) comprise the most common subset of mesenchymal tumors arising from the gastrointestinal tract, with the poorest prognosis.[1] GISTs arise most commonly in the stomach, followed by the small bowel, and occasionally the rectum, esophagus, and colon.[2] Localized GISTs are treated effectively by surgical excision. Although a wedge resection of the stomach or a segmental resection of the intestine is considered adequate, a local resection of primary GISTs in the esophagus, duodenum, and rectum is often technically impossible. Thus, wide resections are usually the treatment of choice for GISTs in these locations.[3] We report two cases of rectal GIST, treated using a less-invasive transsacral approach with good functional results.

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Case 1

A 75-year-old woman presented to our clinic complaining of unusual bowel movements. A pelvic examination revealed a smooth mass between the vagina and rectum. Transvaginal ultrasonography revealed a well-encapsulated solid mass of size 3.6 × 4.1 cm. Core needle biopsy of the mass showed a spindle-shaped tumor, positive for c-kit and CD34, with a mitotic rate of less than 5 per 50 high-power fields. The tumor was located about 5 cm proximal to the anal verge. Although we recommended abdominoperineal resection with a colostomy, the patient refused; hence, we decided to use a transsacral approach for tumor resection. The patient was positioned in the prone jackknife position and a paramedian skin incision was made. We dissected up to the sacrococcygeal joint, then removed the coccyx and incised the fascia of Waldeyer to expose the perirectal area. After mobilizing the rectum circumferentially from the vagina, we performed a segmental resection and an end-to-end anastomosis. Postoperatively, the patient did not experience pain while sitting or dysfunction of the sphincteric mechanism. When last seen, 2 years after her operation, there was no evidence of recurrence, and her bowel movements were normal.

Case 2

A 55-year-old man was referred to our department for the investigation of a rectal mass detected during a routine physical examination. He denied any history of constipation or hematochezia. A similar lesion had been found 10 years earlier, and the results of a rectal biopsy had led to a diagnosis of benign leiomyoma. On rectal examination, a mobile and nontender mass with a smooth surface was palpable in the anterior wall of the rectum. We biopsied the rectal mass and GIST was diagnosed. Magnetic resonance imaging showed a 5.5 ×

5.0 cm mass adjacent to the prostate without evidence of distant metastasis (Fig. 1a, b). We performed a wedge resection of the rectum using a transsacral approach (Fig. 2). There was no sign of invasion to adjacent organs. The resected specimen contained an ovoid, elastic-hard tumor (Fig. 3). Pathological examination yielded results consistent with intermediate-grade GIST with negative margins (Fig. 4a, b). The patient had an uneventful postoperative course and was discharged 12 days after his operation. The anal sphincter and associated innervations were spared and the patient experienced no postoperative incontinence.

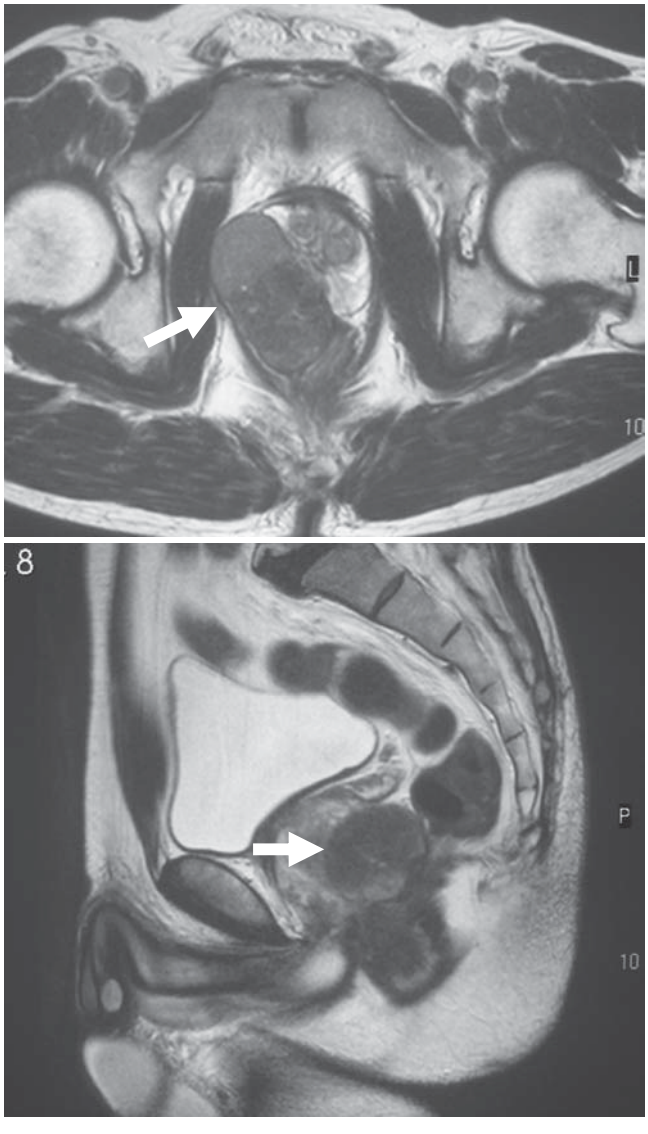

Fig. 1. Pelvic T2-weighted imaging shows a rectal mass and its anatomic relationships to the adjacent structures (case 2). **a** Axial view; **b** sagittal view

Discussion

The clinicopathological profiles of GISTs of the rectum are more obscure than those of GISTs of the stomach or small intestine.[1] As GIST is a recently defined molecular and pathological entity, its management at initial diagnosis and the treatment of local and advanced disease have not yet been standardized. For localized, resectable GISTs, surgery is the preferred treatment, and neither radiotherapy nor conventional chemotherapy has proved efficient as adjuvant therapy.[4] Moreover, no consensus has been reached among experts regarding the need for preoperative diagnosis by core-needle biopsies, taken either by endoscopic ultrasound or percutaneously.[3] However, consideration of biopsy should be based on the extent of the disease and suspicion of a given histological subtype.[5] The rarity of rectal GIST makes the assessment of the precise extent of surgical resection difficult. According to our review of the literature, most patients with rectal GIST have undergone more extensive procedures such as abdominoperineal or low-anterior resection.[6] However, complete surgical resection with en-bloc (R_0) resection of the tumor and the surrounding normal tissue is the current treatment of choice for GISTs, followed by surveillance for metastasis and recurrence.[2] There is no evidence that procedures more extensive than the removal of all gross neoplasm prolong survival or delay recurrence. A complete resection of a primary GIST is associated with a 5-year survival rate of 48%–65%.[7]

Several minimally invasive treatment options are available for small favorable lesions in the rectum.[8] Endoscopic snaring or excision is useful for small polyps at any level in the rectum, whereas transanal approaches have been developed recently to resect rectal tumors. This approach is less invasive, but the resection of tumors distant from the anal verge is difficult.[4] A transsphincteric approach is usually indicated for lesions in the lower rectum that are too large for transanal resection. A posterior transsacral approach for disease located in the rectum was well described by Kraske,[9] and a modified version has been used for benign rectal problems and selected rectal cancers.[10] Transsacral surgical approaches to the rectum are still avoided by most surgeons because these procedures are considered to be associated with an increased risk of complication such as fecal fistula, anal dysfunction, and poor perineal wound healing.[8] However, most studies that reported a high incidence of complications were conducted before the era of proper bowel preparation, prophylactic antibiotics, and adequate drainage techniques.[11,12] Neither of our patients suffered complications after surgery and both were discharged within 2 weeks. Furthermore, anastomotic leakage after a posterior approach will cause only localized problems,

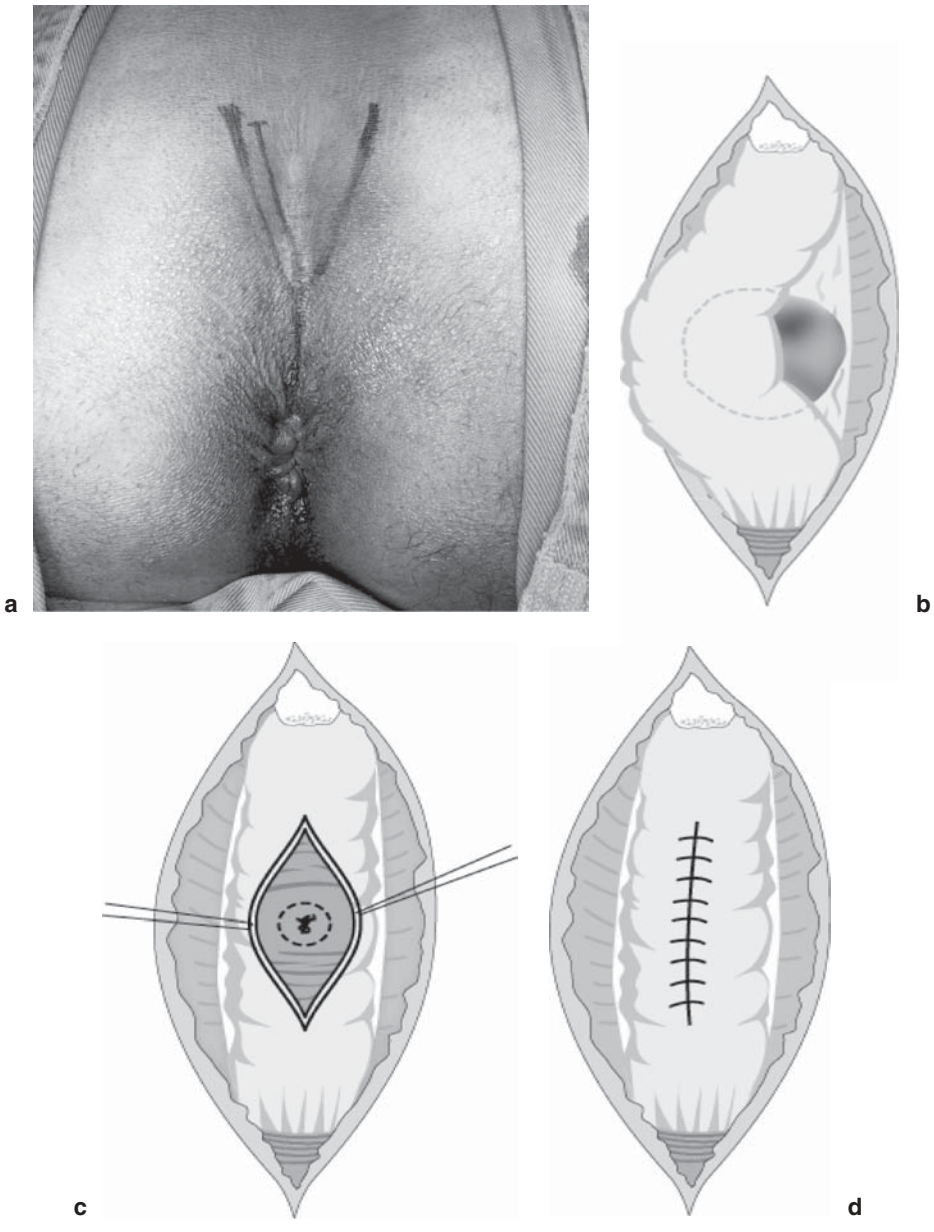

Fig. 2. Schematic description of the technique. **a** The patient is positioned prone on the table and the buttocks are taped to the side of the table. A skin incision is made just lateral to the midline, extending to the anal verge. **b** The coccyx is excised with electrocautery. After incision of Waldeyer's fascia, circumferential mobilization of the rectum is performed. **c** After opening the posterior wall of the rectum, full-thickness excision of the lesion is performed with a normal margin. **d** The rectum is closed in a longitudinal fashion with Albert–Lembert sutures

Fig. 3. The gross specimen from case 2 was ovoid and measured $5.5 \times 3\,\text{cm}$

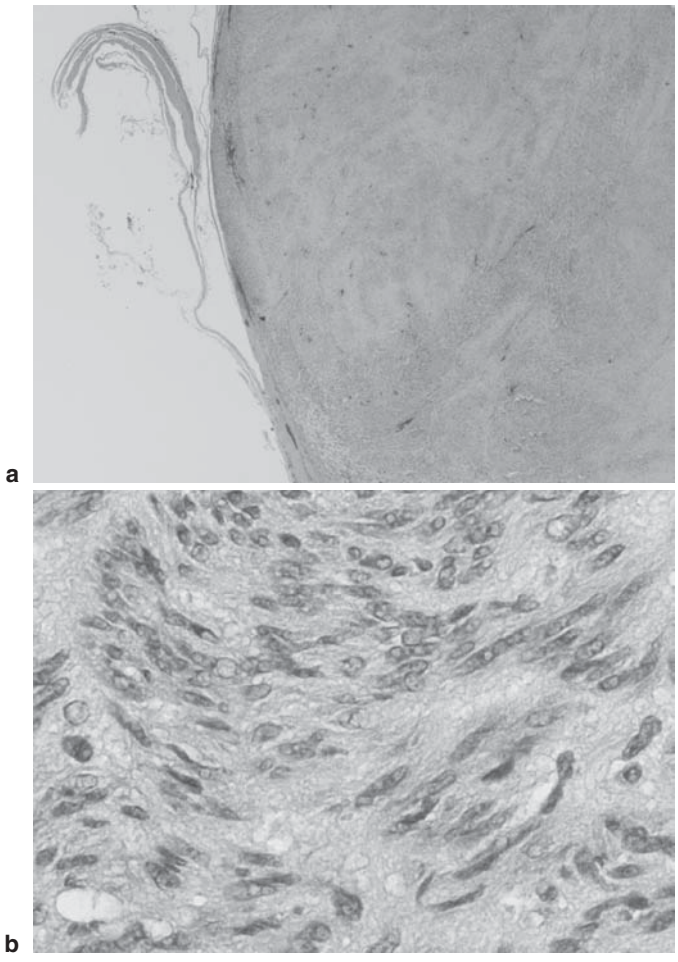

Fig. 4. a, b Histopathological examination of the tumor from case 2 revealed the proliferation of spindle-shaped cells with a negative surgical margin

limited to the extraperitoneal cavity, without creating generalized peritonitis.

In conclusion, these two case reports show the safety and feasibility of a transsacral approach to resect a rectal GIST.

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