

# Surgical Treatment of Benign Retrorectal Masses in Adults: A Retrospective Study



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## ABSTRACT

**Background:** Tumors that occur in the retrorectal space are called retrorectal tumors. These tumors arise from various tissues. Late symptoms may therefore outweigh diagnosis and treatment. Therefore, the surgery becomes difficult. **Aims and Objectives:** Our study aims to reveal the difficulties in the diagnosis of retrorectal tumors, radiological diagnostic methods, and safe surgical excision surgical techniques. **Materials and Methods:** Twelve patients operated for retrorectal tumor between 2014 and 2020 were included in the retrospective study. Demographic features, clinical findings, diagnostic methods, surgical treatment procedures, evaluations of preoperative/postoperative complications, pathological classifications, length of hospital stay, and duration of surgery were studied. **Results:** Twelve patients [9 females, 3 males; the mean age was  $39.3 \pm 11.8$  (22-56)] due to retrorectal tumors. All lesions were evaluated preoperatively using magnetic resonance imaging (MRI). According to the preoperative examination and radiological findings; Surgical resection was performed with an anterior approach to 3 patients, a posterior approach to 4 patients, and a combined approach to 5 patients. The mean tumor size was  $8 \pm 2.24$  cm. Immature teratoma and epidermoid cyst ( $n = 6$ ) were the most common tumors. We did not have any patients with features of malignancy on final histopathology. **Conclusion:** Although retrorectal tumors are difficult to diagnose, it is important to suspect clinically. A good evaluation of the patients with radiological imaging facilitates the resection of the tumor surgically. Surgery of the patient in centers specialized in these tumors increases surgical success.

**Keywords:** Sacral Region; Surgical Procedure; Radiology; Immature Teratoma; Retro-rectal mass

## INTRODUCTION

Tumors that occur anatomically in the retrorectal space are called retrorectal tumors (RT). This space is limited by mesorectum in the front, presacral fascia at the back, iliac vessels and ureter at the sides, peritoneum at the top, and levator ani muscle at the bottom.<sup>1-3</sup> These tumors can originate from various tissues. They may not be noticed for a long time because their symptoms are not clear. Therefore, it can pass diagnosis and treatment. This delay in diagnosis may cause the tumor to reach large sizes and invade vascular and neurogenic tissues. This causes intraoperative bleeding and neurological complications.<sup>4</sup>

The prevalence of these tumors is unknown, but approximately 1/40,000.<sup>5,6</sup> Computed pelvic tomography, magnetic resonance, and transrectal ultrasound are used

for diagnosis. Surgical removal of these tumors is the most appropriate treatment method. There are some publications in the literature for surgical treatment and histological classification. An algorithm for surgical treatment was published in 2007.<sup>7</sup> Retrorectal tumors were classified as congenital (55-65%), neurogenic (10-12%), osseous (5-11%), inflammatory (5%) and miscellaneous (12-16%) (Table 1).<sup>8</sup>

Prospective and randomized trials are limited as RT is rarely seen clinically. The number of patients is generally limited in case series. There is no consensus on the treatment of RTs. In this study, patients diagnosed retrorectal mass treated retrospectively in our service in the last 6 years were evaluated. Demographic features, clinical findings, diagnostic methods, treatment procedures, pathology results, intraoperative and postoperative complications, and length of hospital stay were recorded. The findings

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were compared with the literature. It is of the opinion that experienced patients will undertake preoperative evaluation, surgical treatment and subsequent follow-up. In this article, we aimed to present our clinical experience to contribute to the literature.

## MATERIALS AND METHODS

### Study design

Patients who underwent surgery for RT at the Dicle University Medical Faculty General Surgery Clinic between January 2014 and January 2020 were included in the retrospective study. This study was approved by the Dicle University Faculty of Medicine Ethics Committee.

### Data collection

Demographic features, clinical findings, diagnostic methods, treatment procedures, pathology results, intraoperative and postoperative complications, and length of hospital stay were recorded. The diagnosis was confirmed by CT or MRI in all patients. Posterior, anterior, or combined approach (anterior and posterior) was preferred as a surgical technique according to CT and MRI findings.

All patients were seen at the clinic once a month post discharge. An MRI was taken at any clinical signs of recurrence. Patients with recurrence, chronic pain and anal dysfunction were evaluated.

## RESULTS

A total of 12 patients were operated on for RT. The mean age of the patients was  $39.3 \pm 11.8$  years. Three of the patients were male and 9 were female. Mean tumor diameters were  $8 \pm 2.24$  cm.

Most of the patients were referred from other departments such as neurosurgery and orthopedics. In most patients who came to us, the general symptom was perineal pain and tenesmus complaints. Also, patients had complaints of

pelvic, sacral, low back pain, constipation, palpable perineal mass, urinary tract dysfunction, and rectal bleeding at the time of presentation (Table 2). The study was pre-approved by the Institutional Ethical Committee.

All patients underwent an MRI examination preoperatively. Also, 9 (75%) of the patients were evaluated by CT and 5 (41%) patients were evaluated by proctosigmoidoscopy (Table 2).

The patients were not evaluated by biopsy before the operation. Postoperative pathological examination revealed immature teratoma in 4 patients, epidermoid cyst in 2 patients, tailgut cyst in 2 patients, and duplication cyst in one patient. In the remaining three patients, schwannoma and neurofibroma, which are neurogenic lesions, were detected (Table 3)(Figure 1-2).

Anterior approach was applied to 3 patients, a posterior approach to 4 patients(Figure 3-4), and a combined approach was applied to 5 patients. Surgical excision was performed in two patients with coccygectomy. In one patient, a separate tumor in the small intestine was detected together with a teratoma. Segmental small bowel resection was performed in patient due to mass in the small intestine simultaneously, and a diagnosis of neuroendocrine tumor was made pathologically. In tumors that extend to the sacral fascia, are close to the vascular structures, and have the ability to invade, attention should be paid to bleeding. Two patients diagnosed with teratoma had intraoperative bleeding and needed transfusion. Primary suturing was performed in one patient due to rectum injury and a protective loop ileostomy was opened. Ileostomy was closed 3 months later. Also, 1 patient developed wound infection in the postoperative period. In one patient who was operated with a combined approach, an abscess with an intra-abdominal extension of 8\*7 cm was observed in the operating site. The patient was treated with percutaneous drainage and antibiotherapy. The average length of hospital stay was  $16.8 \pm 7.25$  days. The operation times were on

**Table 1: Classification of the retrorectal tumors**

Congenital	Neurogenic	Miscellaneous	Osseous	Inflammatory
1) Developmental cyst	1) Neurofibroma	1) Lipoma/liposarcoma	1) Osteoma	1) Perineal or
a) Dermoid	2) Ependymoma	2) Fibrosarcoma	2) Osteogenic sarcoma	pelvirectal abscess
b) Epidermoid	3) Neurilemoma	3) Leiomyoma/leiomyosarcoma	3) Ewing's tumor	2) Diverticulitis
2) Tailgut cyst	(schwannoma)	4) Hemangioma	4) Chondromyxosarcoma	3) Crohn's disease
3) Teratoma	4) Ganglioneuroma	5) Carcinoid tumors	5) Giant cell tumor	4) Foreign body
4) Teratocarcinoma		6) Hemangioendothelial sarcoma		granuloma
5) Chordoma		7) Extra-abdominal desmoid		5) Infectious
6) Anterior sacral		8) Plasma cell myeloma		granuomas
meningocele		9) Endothelioma		
7) Rectal duplication		10) Pelvic ectopic kidney		
8) Adrenal rest tumor		11) Hydatid cyst		
		12) Hematoma		

**Table 2: Characteristics of patients**

Case Age (years)	Gender	Preoperative symptoms	Preoperative evaluation	Tumor diameter radiologically (MR)(cm)	Tumor Location (MR)
32	Female	Pelvic pain	MR/CT/RSS	8,5×4	Coccyx inferior
45	Female	Pelvic pain and tenesmus	MR/CT	9×8	Presacral
32	Female	Perineal pain, lump in the gluteal region	MR/CT/RSS	5,4×5,3	Presacral
53	Female	Perineal pain	MR/CT	3,6×3,4	Pre-coccygeal
43	Male	Perineal pain	MR/CT	7×6	Presacral
29	Female	Rectal bleeding and Lower back pain	MR/CT	7×5	Presacral
22	Female	Defecation difficulty, pelvic pain	MR/CT	16×13	Presacral
42	Male	Perineal pain and lower back pain	MR/RSS	56×38	Coccyx inferior
53	Female	Perineal pain	MR/PET	4,4×3,3	Presacral
56	Female	Perineal pain	MR	8×6	Presacral
43	Male	Pelvic and sacral pain	MR/CT/RSS	7×5	Presacral-rectorectal
22	Female	Perineal pain	MR/CT	16×12	Presacral

MR; magnetic resonance, BT; computed tomography, RSS; rectosigmoidoscopy, PET; Positron emulsion tomography.

**Table 3: Findings during and after the operation**

Operative approach	Intraoperative complication	Postoperative complications	Pathology and Tumor diameter(cm)	Operative time (min)	Length of Stay (days)
Posterior	None	None	Immature teratoma (5*4*2)	132	17
Combined	None	None	Schwannoma (10*10*8)	352	20
Anterior	Bleeding	None	Schwannoma (6,5*6*4,5)	222	24
Posterior	None	None	Epidermoid cyst (5,5*4*3,5)	141	7
Combined	None	Perianal fistula (In the 3rd month)	Immature teratoma (7,7*6,5*5,5)	160	13
Combined	Rektum İnjury (repair with suture +loop ileostomy)	Intraabdominal abscess	Duplication cyst (7,5*5,5*5)	180	21
Combined	None	None	Tailgut cyst (12*10*4)	170	23
Posterior	None	None	Epidermoid cyst (7*2*5)	120	8
Anterior	Nerve injury	None	Neurofibroma (7,7*5*3,1)	281	6
Combined	Bleeding	Wound infection	Immature teratoma (7,5*6,5*5,5)	230	27
Anterior	None	None	Immature teratoma (7,7*6,5*5,5)	160	13
Posterior	None	None	Tailgut cyst (12*10*4)	170	23

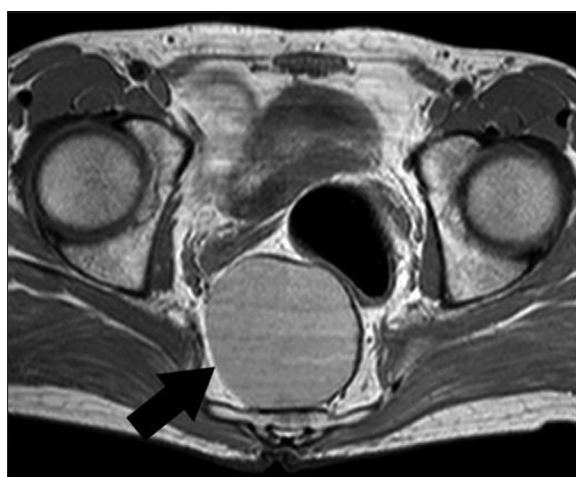


Figure 1: Axial MRI scans showing presacral immature teratoma

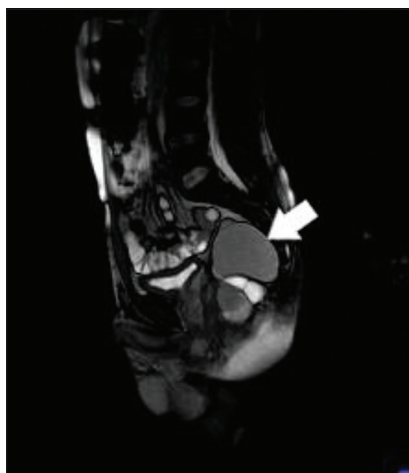
average  $193 \pm 67.6$  minutes (Table 3). No recurrence or anal dysfunction was observed during the follow-up of the patients. However, 3 patients had pain in the perianal region that lasted for about 2 months.

### DISCUSSION

Retrorectal masses and presacral tumors are rare. Due to their rarity, information about the clinical, diagnostic, and imaging techniques, surgical procedures, recurrence rates, and general outcomes of these tumors are not sufficient.<sup>1,4</sup>

The incidence of reported RTs ranges from 0.9 to 6.3 per year.<sup>9-11</sup> In this study, we detected 12 patients with retrorectal tumors for 6 years in our hospital.





**Figure 2:** Sagittal MRI scans showing presacral immature teratoma

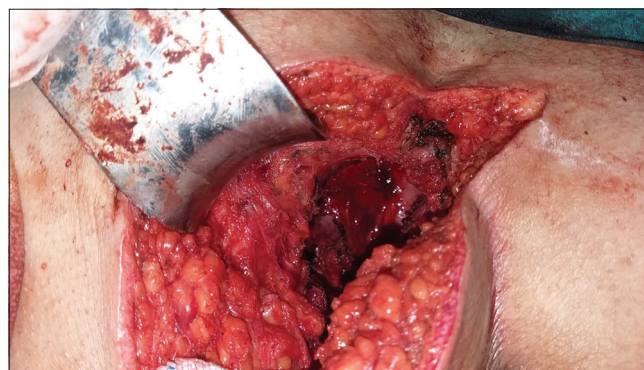


**Figure 3:** Posterior approach for retrorectal tumor

Congenital types are the most common of these tumors with histologically different types. Congenital types are benign and more common in women.<sup>10-12</sup> However, epidemiologically, the importance of gender is unknown in malignant congenital RT Patients.<sup>1</sup> In our series, it was observed that female sex was more common with retrorectal tumors (9/12). No malign tumor was detected in our patients.

RT patients were generally asymptomatic, but they may occur with nonspecific signs and symptoms.<sup>2,12,13</sup> The location of the tumor varies with its diameter, presence of invasion, and infection status.<sup>14</sup> Symptomatic patients may appear as pain in the perineal region, chronic constipation, rectal or urinary incontinence, or sexual dysfunction. Some tumors cause perianal discharge. Therefore, it can be confused with perianal fistulas.<sup>2,15</sup> Our patients complained mostly of pain in the perianal and sacral region.

Since the symptoms of the patients are not specific, it is necessary to suspect the diagnosis first. CT and MR are the gold standard for diagnosis. With CT, we can distinguish whether the mass is solid or cystic, but MR gives more valuable findings in these patients.<sup>4</sup> In our series, 9 of our



**Figure 4:** Postoperative lodge view

patients received MR + CT, 2 patients received MR, and one patient received MR + Scintigraphy. Scintigraphy was added to the diagnostic tests to investigate the presence of metastasis in the patient who had undergone surgery for bladder adenocarcinoma 1 year ago. No finding in favor of metastasis was found. MR was used to determine both the diagnosis and the treatment protocol to evaluate the mass before surgery.

The importance of preoperative biopsy in the treatment of RT is controversial. Bullard Dunn argued that biopsy is safe outside of the transrectal approach.<sup>15</sup> Messick et al., suggested that we can do the biopsy to all patients.<sup>1</sup> Ghosh et al., argued that preoperative biopsy is unnecessary, as the decision of surgery is not affected by biopsy.<sup>16</sup> Biopsy should not be performed especially in cystic lesions. It can usually be diagnosed radiologically. However, chemotherapy and radiotherapy may be required in patients with osteogenic sarcoma and lymphoma.<sup>17</sup> We did not do biopsy in any patient, considering that biopsy results would not affect our surgical decision and cause serious complications.

RTs are divided into 5 groups with a classification made in 1949. This classification is summarized in Table 1. Congenital ones can be cystic or solid and makeup about 60% of RTs. They are more common in women and are generally benign.<sup>4</sup> The majority of our patients (9/12) were in this group. Teratomas are true neoplasms that contain the elements of the 3 germ layers. Therefore, they contain the epithelium of the gastrointestinal, respiratory, and nervous systems. Although they are generally benign, they may have the potential for malignant degeneration.<sup>4,18</sup> In our study, teratoma (Figure 5) was detected in 4 patients and an epidermoid cyst in 2 patients. The second frequency is neurogenic tumors. These tumors are caused by peripheral nerves and 85% of them are benign. In our series, 3 patients had neurogenic tumors, 2 of them had schwannoma and 1 had neurofibroma. There were no less common miscellaneous tumors, inflammatory tumors, and osseous tumors in our patients.



**Figure 5:** Immature teratoma macroscopic view

Although RTs are asymptomatic, they can cause infection and undergo malignant degeneration. Therefore, it must be fully resected surgically. The surgical approach should be planned based on the size and extent of the lesion, preoperative MRI findings, based on the surgeon's pelvic or postrectal anatomy specialization.<sup>1,17,19</sup> The anterior approach benefits the surgeon in terms of protecting intra-abdominal organs and vascular structures. It is more advantageous with anterior or combined approaches when dealing with masses that have reached a large diameter (> 5 cm) and are above the S3 level. The posterior approach is suitable for patients who are below the S3 level and whose upper limit can be reached by digital rectal examination.<sup>6,20-22</sup> In our study, surgery was performed with anterior to 3 patients, posterior to 4 patients, and a combined approach to 5 patients.

While performing RT surgery, there may be intraoperative and postoperative complications. Serious bleeding can occur when large vessels or the sacral vein plexus are injured. Since two of our patients had bleeding during the operation, transfusion was needed. When bleeding occurs, while compressing is sometimes sufficient, severe bleeding that may require iliac artery ligation may occur. In our patients, first compression was applied to the bleeding in the sacral region, then simple ligation was made in the sacral region and the bleeding was stopped. Wound infection developed in one of our patients postoperatively. The patient showed improvement after antibiotic therapy.

With the development of laparoscopic surgery, laparoscopic resections were also on the agenda. In 2014, Duclos J et al., published a series of 12 cases. All patients underwent R0 resection and one patient was reported as malignant, and no recurrence was observed in their follow-up.<sup>23</sup> Studies have shown that the laparoscopic approach has important benefits, such as smaller wounds, less postoperative pain, and facilitates excellent visualization of deep structures in the retrorectal space, which prevents vascular and nerve injuries.<sup>24,25</sup>

## CONCLUSION

Retrorectal tumors are rare and have diagnostic difficulties. They usually occur with nonspecific symptoms or can be diagnosed when examining other pathologies. Doubt is essential for diagnosis. MR and CT assist in demonstrating the location of the tumor and preparing the surgical plan. In order to get a good surgical result, the joint decision of the branches of interest is important when treating these tumors. To reduce the risk of recurrence, surgery should be performed by hands experienced in pelvic anatomy and pelvic surgery.

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**Authors Contribution:**

**HB**-Concept and design of the study; prepared first draft of manuscript, Statistically analysed and interpreted, preparation of manuscript; **ÖB**- Interpreted the results; reviewed the literature and manuscript preparation; Concept, coordination, review of literature and manuscript preparation; **AO**- Statistically analysed and interpreted, preparation of manuscript and revision of the manuscript

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