

# Retrorectal Tumors in Adults: A 10-Year Retrospective Study

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Due to the rarity and large diversity of the primary retrorectal tumors (RTs), the diagnoses are often difficult and they can be misdiagnosed. We present our experience in light of scarce information available on the clinical manifestations of RTs. The retrospective study included 17 patients diagnosed as RTs between January 2004 and January 2014. Demographic characteristics, length of symptoms, clinical findings, diagnostic methods, evaluations on the treatment procedures and postoperative periods, pathology, complications, and length of hospital stay were recorded. A mean of 1.7 of patients were diagnosed with RTs annually in our hospital. Patients comprised 12 females and 5 males. Pain and discomfort were the most common symptoms at presentation. All the lesions were evaluated by using magnetic resonance imaging (MRI) and computed tomography (CT), and all the patients were treated operatively. Based on the preoperative MRI or CT findings, an anterior approach was performed in 7 patients, a posterior approach in 6 patients, and combined approach in 4 patients. Mean size of tumors was 9.2  $\pm$  4.3 cm. Epidermoid cyst (n = 8) was the most common tumor. Except for 1 case of liposarcoma, 16 tumors were confirmed to be of benign nature in histologic examination. Mean length of hospital stay 12.4  $\pm$  6.8 days. Retrorectal tumors are heterogeneous and lead to diagnostic difficulties. A high index of clinical suspicion is needed for diagnosis. Preoperative imaging may be helpful in determining the course of treatment. Total excision of a retrorectal tumor may alleviate pressure symptoms and confirm the diagnosis.

*Key words:* Retrorectal tumors – Epidermoid cyst – Surgical approach – Diagnosis – Outcomes

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**R** etrorectal tumors (RTs) occur in the anatomic region known as the retrorectal space. This space is restrained anteriorly by the mesorectal fascia, posteriorly by the presacral fascia, inferiorly by the levator ani muscle, superiorly by the peritoneal reflection, and laterally by the iliac vessels and ureters.<sup>1-3</sup> RTs constitute a heterogeneous group of both benign and malignant tumors, which can be categorized as congenital, neurogenic, osseous, miscellaneous, or inflammatory.<sup>2-6</sup> The population prevalence of RTs remains unknown; however, the estimated prevalence is one in every 40,000 hospital admissions.<sup>7,8</sup>

RTs generally remain asymptomatic or present with nonspecific symptoms.<sup>9</sup> Therefore, RTs may be overlooked unless a high index of suspicion is present, and the diagnosis is established either incidentally or during exploration for other pathologies. Moreover, RTs present difficulties in treatment although improved techniques are used in perioperative care.<sup>10</sup>

The scarcity in the information available on the clinical manifestations of RTs has led to a hindrance of the development of prospective, randomized clinical studies. Almost all the information available on RTs have been presented by small, single-center studies and due to this scarcity of information, there is no consensus on the ideal management for RT. In this study, we aimed to present our clinical experience to make a contribution to the literature.

## Methods

The retrospective study included 17 patients who were operatively treated for RT at Dicle University Medical School General Surgery Clinic between January 2004 and January 2014. Demographic characteristics, length of symptoms, clinical findings, diagnostic methods, evaluations on the treatment procedures and postoperative periods, pathology, and complications, and length of hospital stay were recorded.

Diagnosis was confirmed by CT or MRI in all the patients. A posterior, anterior or combined approach (anterior or posterior) was preferred depending on the CT and MRI findings.

## Results

A total of 17 patients underwent surgery due to RT. The patients included 5 men and 12 women with a

mean age of 36.4  $\pm$  14.2 years and a mean tumor size of 9.2  $\pm$  4.3 cm.

#### History and physical examination

Two patients (11.8%) were diagnosed incidentally on gynecologic examination. Most of the patients were referred to us from other clinics such as brain surgery, orthopedics, or gynecology, and their most common complaint at presentation was a pain in the pelvic, sacral, lower back or perianal area, followed by difficulty or tenesmus in defecation, palpable perineal lump, lower urinary tract dysfunction, and rectal hemorrhage. Moreover, some patients had more than one complaint at presentation (Table 1). In all the patients, the mean length of symptoms was >1 year.

#### Imaging

MRI scans were obtained in 15 (88.2%), CT in 13 (76.5%), and rectosigmoidoscopy in 10 (58.9%) patients (Fig. 1).

#### Histopathologic examination

Two patients were referred to our clinic after being diagnosed with a benign RT from biopsy. No biopsy was needed for the remaining 15 (88.2%) patients. Final pathology revealed that epidermoid cysts (Fig. 2a and 3a) were the most common tumor (n = 8), followed by schwannoma (n = 2; Fig. 2b and 2c, and 3b), immature teratoma (n = 2), ganglioneuroma (n = 1), hydatid cyst (n = 1), hematoma (n = 1), diverticulitis (n = 1), and liposarcoma (n = 1). The patient with liposarcoma was the only case with a malignant tumor.

#### Surgical approach and follow-up

An anterior approach was performed in 7 patients, a posterior approach (Fig. 3c) in 6 patients, and combined approach in 4 patients. Two patients underwent surgical extirpation with coccygectomy. The patient with liposarcoma underwent total mass excision and developed tumor recurrence at post-operative month 7. In this patient, an *en bloc* resection was performed followed by low anterior resection due to the expansion of the upper half of the rectum. One patient developed intraoperative ureter injury and thus underwent primary anastomosis. Another patient received intraoperative compression and gauze packing due to intraoperative hemorrhage, who then later developed lower

urinary tract dysfunction in the postoperative period. In addition, 1 patient developed wound infection in the postoperative period. The mean length of hospital stay was  $12.4 \pm 6.8$  days and all the patients significantly improved at 1-year follow-up, except for the patient with liposarcoma. This study was approved by the Ethics Committee of Dicle University Medical School.

## Discussion

Retrorectal masses and presacral tumors are rare entities. Due to this rarity, there is a scarcity in the documentation of information about the manifestations of these tumors as well as the diagnostic methods and imaging techniques, surgical procedures, recurrence rates, and overall results. However, knowledge of these masses and expertise in management is essential because it is highly likely that surgeons will have at least 1 patient with a RT during their career.<sup>1,10</sup>

Reported incidence of RTs ranges between 0.9 and 6.3 patients per year.<sup>4,5,11,12</sup> Kwon *et al* found an incidence rate of 1.6 patients per year<sup>13</sup> and Messick *et al* found it as 2.9 patients per year.<sup>1</sup> In this study, we found that 17 patients were detected with retrorectal tumors over 10 years in our hospital, which revealed that 1.7 patients were annually diagnosed with RTs.

Congenital lesions account for the most common retrorectal tumors. These lesions are usually benign and are more commonly seen in females.4-6 However, the importance of gender in the epidemiology of malignant congenital lesions remains unknown. Messick et al reported that these tumors were more common in females (77%) and the prevalence of malignant lesions was higher in females (74%).<sup>1</sup> On the other hand, Jao et al found a higher prevalence of malignant retrorectal tumors in males,<sup>5</sup> whereas Singer et al reported an equal rate between genders.<sup>14</sup> Nevertheless, Duclos et al operatively treated 12 female patients and reported that 10 out of 12 patients had benign lesions.15 In our series, we found that female gender is associated with retrorectal tumors (12/17), and the only patient with a malignant RT was male.

Literature shows that RTs are generally asymptomatic.<sup>1,15–19</sup> In our series, 2 patients were incidentally diagnosed during the gynecologic examination for prolonged and irregular menstrual periods. RTs may present with nonspecific signs and symptoms and they have a great variety of manifestations.<sup>2,6,9,17</sup> This variability is associated with the

location and diameter of the tumor, whether the sacral root has been invaded or not, and the presence or absence of infection.<sup>12,20</sup> Macafee *et al* reported that most of their patients presented with a pain or discomfort in the lower back, over the sacrum or in the perineal area.<sup>21</sup> Similarly, pain or discomfort in the pelvic, sacral, lower back, or perianal area was the most common symptom in our patients.

Most RTs may present with nonspecific signs and symptoms and thus their prompt diagnosis is usually difficult. A high index of suspicion is the first step in the diagnostic process,<sup>10</sup> and CT and MRI remain the gold-standard methods in the diagnosis of RTs. CT can be used to visualize cortical bone destruction, to determine the nature of the lesion (solid or cystic), and also to determine involvement of adjacent organs. MRI is more valuable than CT because it is useful in assessing the planes of resection and spatial relationship to surrounding structures and also in determining the most convenient management and imaging procedures for each patient.<sup>9,10,21–24</sup> In our series, most of our patients had undergone CT in other clinics due to various reasons. Prior to surgery, the patients without MR contraindications were evaluated with MRI, both for diagnosis and treatment.

Sigmoidoscopy can be used to determine transmural tumor penetration.<sup>9</sup> Proctoscopy may not detect the small-scale lesions, but it can visualize the extraluminal compression in large lesions.<sup>19</sup> In our study, sigmoidoscopy was performed in 10 patients who had large tumor with extraluminal compression. Transmural mucosal penetration was not detected in none of them.

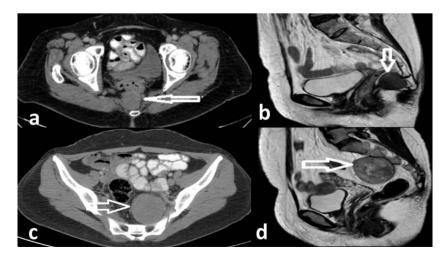
The importance of preoperative biopsy in the treatment of RTs remains controversial. Bullard Dunn maintained that biopsy is safe in all approaches except for the transrectal approach.25 Messick *et al* proposed that biopsy can be performed regardless of the risk of tumor seeding.<sup>1</sup> Ghosh *et al* argued that preoperative local biopsy is normally unnecessary because the decision of surgery is rarely affected by biopsy findings.<sup>26</sup> Wolpert et al and Verazin et al suggested that biopsy should be preferred only when the resection of the lesion is difficult and the histodiagnosis is needed for planning additional treatments such as chemotherapy or radiation therapy.<sup>9,27</sup> Biopsies can cause a number of serious complications including fatal septic complications, perforation, bleeding, and fistulas. Moreover, tumor seeding is another serious risk which may arise from biopsy.<sup>14,28</sup> In our series, 2

Case	Age	Gender	Preoperative symptoms	Preoperative evaluation	Mass size (cm)
1	17	F	Pelvic pain	MRI	$15 \times 8$
2	38	F	Defecation difficulty or tenesmus	MRI/CT	8  imes 6
3	39	М	Occasional rectal bleeding	CT	$10 \times 7$
4	46	М	Defecation difficulty, pelvic pain	MRI/CT	6  imes 7
5	72	М	Pain radiating to the back and legs while defecation	MRI/CT	$4 \times 5$
6	20	F	Perineal pain, palpable perineal lump	MRI	$5 \times 3$
7	30	F	Perineal pain, lump in the gluteal region	MRI	$5 \times 6$
8	54	F	Perineal pain	MRI/CT	$4 \times 5$
9	33	F	Pelvic, lower back pain	MRI/CT	$11 \times 11$
10	45	F	İncidental vaginal bleeding	MRI/CT	$9 \times 8$
11	29	F	Pelvic, lower back pain	MRI/CT	20  imes 7
12	17	F	İncidental pelvic pain, irregular menstrual cycles	MRI	8  imes 6
13	35	F	Lower back pain	MRI/CT	13  imes 9
14	27	F	Lower back pain	MRI/CT	$4 \times 5$
15	48	F	Lower back pain	MRI/CT	4  imes 7
16	27	М	Pelvic and sacral pain	СТ	9  imes 8
17	41	М	Pelvic pain, difficulty in defecation or tenesmus, urinary tract dysfunction	MRI/CT	15  imes 9

*Table 1 Characteristics of patients* 

patients had undergone biopsy before presenting to our clinic. We performed no biopsies for any patient, considering that the biopsy results would not affect our decision to operate and performing a biopsy might lead to serious complications. In addition, biopsy was contraindicated in 1 of our patients due to hydatid disease (HD) in areas with HD endemics.

Retrorectal tumors are usually divided into 5 categories including congenital, neurogenic, inflammatory, osseous, or miscellaneous.<sup>2,4–6,29</sup> The classification of the tumors is summarized in Table 2. Congenital tumors account for almost two-thirds of all RTs and they can be cystic or solid tumors. Developmental cysts, which may be caused by all 3 embryonic germ layers (endoderm, mesoderm, and ectoderm), constitute the largest group of congenital retrorectal lesions. These cysts are usually benign and more common in females.<sup>9</sup> In our study, congenital epidermoid cyst (8/17) was the most common type of RTs and most of them were found in females. Teratomas are true neoplasms that include elements of all 3 germ layers. For this reason, they include epithelium of the digestive, respiratory, and nervous systems. The nature of these lesions can be either solid or cystic, but most of them can have both of these components. Moreover, they can have the potential for malignant degeneration.<sup>9,10,30</sup> In our study, teratomas were detected only in 2 patients.



**Fig. 1** (a) Axial contrast-enhanced CT and (b) sagittal T2 MRI scans showing presacral epidermoid cyst (arrow); (c) Axial contrast-enhanced CT and (d) sagittal T2 MRI scans showing presacral schwannoma (arrow).

Operative approach	Intraoperative complication	Pathology	Postoperative complications	Length of stay (days)
AA	No	Epidermoid cyst	No	10
CA	No	Epidermoid cyst	No	7
CA	Bleeding	Hematoma	lower urinary tract dysfunction	15
AA	no	İmmature teratoma	no	20
PA	Rectal injury (primary suture)	Diverticulitis	No	29
PA	No	Epidermoid cyst	No	7
PA	No	Teratoma	No	13
PA	No	Epidermoid cyst	No	7
AA	No	Schwannoma	No	24
CA (coccygectomy)	No	Schwannoma	Wound infection, difficulty in defecation	13
AA	No	Ganglioneuroma	No	10
AA	No	Epidermoid cyst	No	10
CA	No	Epidermoid cyst	No	13
PA (coccygectomy)	No	Epidermoid cyst	No	5
PA	No	Epidermoid cyst	No	4
AA	No	Hydatid cysts	No	7
AA	Ureter injury (primary	Liposarcoma	Difficulty in defecation,	16
	anastamosis)	-	recurrence (second operation)	

Table 1 Extended

Neurogenic tumors, which account for 10% to 12% of RTs, are the second most frequently seen RTs following congenital lesions. These tumors normally originate from peripheral nerves and 85% of them are benign.<sup>9,20,31</sup> In our series, 3 patients had neurogenic tumors, with 2 of them presenting with schwannoma and 1 of them with ganglioneuroma.

Miscellaneous tumors account for 12% to 16% of RTs.<sup>2,9,32</sup> In our series, only 1 patient was present with hematoma, hydatid cysts, and liposarcoma, who was also the only patient with a malignant lesion.

Inflammatory tumors, which constitute 5% of all RTs, generally arise from foreign substances such as

barium and suture material or result from a retrorectal or abdominal infection such as pelvic sepsis, Crohn's disease, and perforated diverticulitis.<sup>21,24,25</sup> In our study, only 1 patient had an inflammatory tumor, which was caused by diverticulitis.

RTs should be totally resected even if they are asymptomatic because they can cause infection and they have the potential for malignant degeneration. The surgical approach should be planned depending on the preoperative MRI findings including the size and extent of the lesion, involvement of adjacent vital structures, whether the patient previously had extirpation of the primary tumor, and on

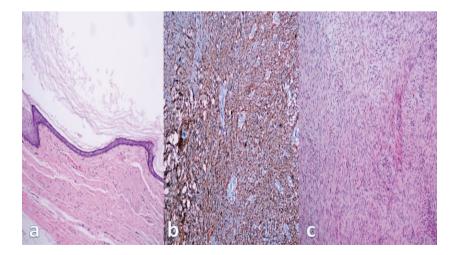


Fig. 2 (a) Cyst wall construction lined by squamous epithelium and keratinous material in the lumen (H&E,  $\times$ 100); (b) S-100 immunoreactivity in the tumor (schwannoma) (immunoperoxidase A,  $\times$ 100); (c) A tumoral structure of schwannoma composed of fusiform cells, which formed bundles crossing each other (H&E,  $\times$ 100). Fig. 3 (a) Macroscopic appearances of epidermoid cyst excised and coccygectomy. (b) Macroscopic appearances of schwannoma excised. (c) Posterior approach for retrorectal tumor.

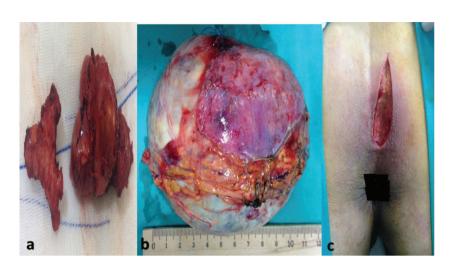
the expertise of the surgeon in pelvic or postsacral anatomy.<sup>1,9,21,24,33</sup> The anterior approach allows the surgeon to control the proximal vascular structures and to mobilize the rectum and other internal organs. An anterior or combined approach is usually performed for the lesions above the level of S3, whereas the posterior approach is preferred for the resection of small lesions below the level of S3. In other words the superior limit can be reached by digital rectal examination.<sup>8,14,16</sup> However, the anterior approach should be avoided if the sacrum is involved. If the adjacent organs are involved, an en bloc resection should be performed, and a combined approach can be preferred for large RTs,

which could not be removed either by an anterior or posterior approach alone.<sup>8,16</sup> In our study, an anterior approach was performed in 7 patients, a posterior approach in 6 patients, and combined approach in 4 patients.

In the surgical intervention of RTs, the coccyx can be transected in order to allow adequate exposure for surgical exploration. Coccygectomy is reported to be useful both for enhancing surgical exposure and for reducing the recurrence risk which is associated with the totipotential cellular remnants of the coccyx.<sup>7,33,35</sup> On the other hand, Messick et al suggested that coccygectomy is not necessary and it increases the mortality rate, and this view is favored by the findings

Cor ellaneous  $(55^{\circ})$ to 16%) Dev liposarcoma D Ε Tail oma Tera na/ osarcoma Tera oma Cho tumors Ant pendothelial n Rec lominal d Adı ell myeloma ioma opic kidney cyst а





Congenital (55% to 65%)	Neurogenic (10% to 12%)	Osseous (5% to 11%)	Inflammatory (5%)	Miscel (12%
Developmental cyst Dermoid Epidermoid	Neurofibroma	Osteoma	Perineal or pelvirectal abscess	Lipoma/lip
Tailgut cyst	Ependymoma	Osteogenic sarcoma	Diverticulitis	Fibrosarcor
Teratoma	Neurilemoma (schwannoma)	Ewing's tumor	Crohn's disease	Leiomyoma leiomyos
Teratocarcinoma	Ganglioneuroma	Chondromyxosarcoma	Foreign body granuloma	Hemangio
Chordoma		Giant cell tumor	Infectious granulomas	Carcinoid (
Anterior sacral meningocele				Hemangioe sarcoma
Rectal duplication				Extra-abdo desmoid
Adrenal rest tumor				Plasma cel
				Endothelio
				Pelvic ecto
				Hydatid cy
				Hematoma

of other studies.<sup>1,7,35,36</sup> We suggest that the coccyx should be preserved unless an *en bloc* resection is required for the tumors densely adhered to the coccyx. We performed coccygectomy in 2 female patients due to the presence of a schwannoma.

Intraoperative hemorrhage, a serious complication of retrorectal surgeries, may occur when the major vessels are impaired or the presacral venous plexus is lacerated. Therefore, the surgeon should be extremely careful to avoid injury to the vessels and the presacral venous plexus. Lin *et al* reported that intraoperative hemorrhage occurred in 2 of their patients due to the injury to the presacral venous plexus, and the hemorrhage was stopped by using Xu's hemostasis procedure.<sup>19</sup> In our study, intraoperative hemorrhage occurred only in 1 patient and the hemorrhage was controlled by intraoperative gauze packing.

In the postoperative period, wound infection was detected in 1 of our patients (5.9%). Jao et al reported a similar rate (10%) for postoperative infections.<sup>5</sup> Recurrence occurred only in the patient with a malignant RT, and the patient was reoperated on and still being followed up in our clinic. Two patients had difficulty with defecation and 1 patient developed lower urinary tract dysfunction.

With the improvements in laparoscopic surgery, laparoscopic resections of RTs have been reported by several studies.<sup>12,15,37,38</sup> The studies demonstrated that the laparoscopic approach had major benefits of smaller wound, less postoperative pain, and it facilitated excellent visualization of the deep structures in the retrorectal space, which prevents vessel and nerve injuries. Kye et al performed laparoscopic surgery in 3 patients and concluded that laparoscopic approach may be a meaningful method in abdominal approach for the resection of benign retrorectal tumors.<sup>12</sup> Duclos et al reported that laparoscopic resection of RTs is a reliable, feasible, useful approach, and it allows complete excision of tumors located in the retrorectal space with low morbidity.<sup>15</sup>

Transanal endoscopic microsurgery (TEM) is a minimally invasive technique. It has been employed in the excision of benign RTs by Serra Aracil *et al*<sup>32</sup> and Zoller *et al*.<sup>39</sup> They reported no significant complications and complete excision of the cysts.

## Conclusion

Retrorectal tumors are rare and lead to diagnostic difficulties. They usually present with nonspecific symptoms or may be diagnosed incidentally during

exploration for other pathologies. A high index of clinical suspicion is needed for diagnosis. MRI and CT aid in the detection of retrorectal tumors and provide help in the determination of surgical approach. Biopsy is usually not required. Total excision of the tumor with negative resection margins is advised for relieving pressure symptoms and also for establishing a definitive diagnosis. Patients with retrorectal tumors should be treated in a major tertiary hospital by surgeons who have advanced knowledge and experience in pelvic surgery.

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