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Retrorectal Tumors: A Comprehensive Literature Review

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Abstract

Importance   Retrorectal tumors are rare lesions that comprise a multitude of histologic types. Reports are limited to small single-institution case series, and recommendations on the ideal surgical approaches are lacking.

Objective   The purpose of the study was to provide a comprehensive review of the epidemiology, pathologic subtypes, surgical approaches, and clinical outcomes of retrorectal tumors.

Evidence Review   We conducted a review of the literature using PubMed and searched the reference lists of published studies.

Results   A total of 341 studies comprising 1708 patients were included. Overall, 68 % of patients were female. The mean age was 44.6 ± 13.7 years. Of all patients, 1194 (70 %) had benign lesions, and 514 patients (30 %) had malignant tumors. Congenital tumors (60.5 %) were the most frequent histologic type. Other pathologic types were neurogenic tumors (14.8 %), osseous tumors (3.1 %), inflammatory tumors (2.6 %), and miscellaneous tumors (19.1 %). Biopsy was performed in 27 % of the patients. Of these patients, incorrect diagnoses occurred in 44 %. An anterior surgical approach (AA) was performed in 299 patients (35 %); a posterior approach (PA) was performed in 443 (52 %), and a combined approach (CA) was performed in 119 patients (14 %). The mean length of stay (LOS) of PA was 7 ± 5 days compared to 8 ± 7 days for AA and 11 ± 7 days for CA ($p < 0.05$). The overall morbidity rate was 13.2 %: 19.3 % associated with anterior approach, 7.2 % associated with posterior approach, and 24.7 % after a combined approach ($p < 0.05$). Overall postoperative recurrence rate was 21.6 %; 6.7 % after an anterior approach, 26.6 % after a posterior approach, and 28.6 % after a combined approach ($p < 0.05$). A minimally invasive approach (MIS) was employed in 83 patients. MIS provided shorter hospital stays than open surgery (4 ± 2 vs. 9 ± 7 days; $p < 0.05$). Differences in complication rate were 19.8 % in MIS and 12.2 % in open surgery and not statistically significant.

Conclusions and Relevance   Retrorectal tumors are most commonly benign in etiology, of a congenital nature, and have a female predominance. Complete surgical resection is the cornerstone of retrorectal tumor management. A minimal access surgery approach, when feasible, appears to be a safe option for the management of retrorectal tumors, with shorter operative time and length of stay.

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Introduction

Retrorectal tumors are rare tumors with a wide range of histology and are found within the pararectal space, which is defined as the potential space between the mesorectum and pelvic wall [1]. The wide range of histological variance may, in fact, be due to the presence of multiple embryologic remnants and miscellaneous tissue types in the presacral and retrorectal spaces [2]. They range from benign cysts to malignant masses that can invade the surrounding pelvic structures [2]. The variety and heterogeneity of retrorectal tumors have resulted in confusion over their classification and management [3–9].

The presentation, diagnosis, and treatment of retrorectal tumors can perplex clinicians. These tumors are generally asymptomatic or present with vague symptoms leading to misdiagnosis [10]. Once a diagnosis is made, surgical resection may be difficult given the anatomical location of the tumor. However, complete surgical resection is recommended in all cases including benign tumors given the potential for development of symptoms and malignancy [2, 11–15]. The surgical approach for these tumors may depend on various factors, such as their size and location. Several surgical approaches have been proposed for resection of retrorectal tumors. The described approaches include the abdominal (anterior), the perineal (posterior), and the abdominoperineal (combined anterior and posterior) approaches [3, 10, 13, 16]. Choosing a suitable approach is key to a successful operation and may help provide optimal exposure, minimize damage, and reduce complications. Laparoscopic approach has been successfully employed in recent years in a few cases.

The rarity of these tumors and the lack of availability of literature pose challenges for surgeons and to date, only small case series exist in literature. Given the variability of pathology, symptoms, and treatment options, we present a comprehensive review of literature describing the etiology of retrorectal tumors, diagnosis paradigm and treatment options for these tumors. This literature review aims to provide the 21st century surgeon a guide to the diagnosis and treatment of these rare lesions.

Methods

A PubMed search using the terms "pararectal tumor," "presacral," and "retrorectal" was utilized to identify the reported cases of retrorectal, non-gynecologic tumors, as well as reviews describing similar cases was conducted. A flowchart of the selection process according to the statement on Preferred Reporting Items for Systematic Reviews and Meta-analyses is presented in Fig. 1. The target population consisted primarily of adults (≥14 years old), with
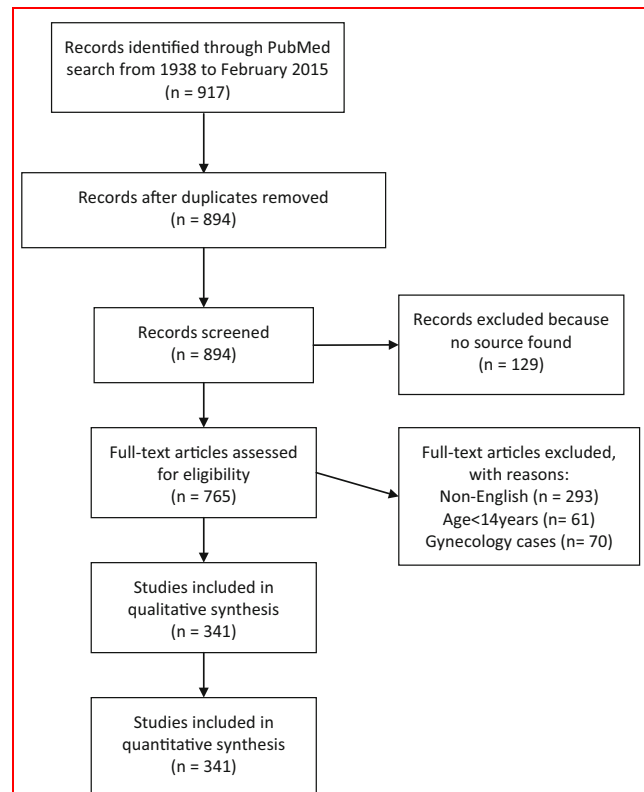
Fig. 1 Flow diagram of the different phases of the systematic review according to the statement on Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA)

histologically confirmed retrorectal tumor diagnoses. Only English-language articles were assessed. Case reports, review articles, and case series were included, irrespective of size. Patients with non-primary tumors and pediatric patients were excluded. Based on available literature [7], pathology was divided into the following five categories: congenital, inflammatory, neurogenic, osseous, and miscellaneous.

The data were analyzed with the statistical program SPSS® (SPSS Inc., Chicago, IL, USA) version 17, for Windows®. The t test and chi-square test were used to assess statistical significance and $p < 0.05$ was considered statistically significant.

Results

A total of 341 studies with 1708 patients were identified. The majority of patients were female (68 %). Age ranged from 14 to 91 years, with mean age of 44.6 ± 13.7 years. Table 1 depicts the overall incidence, the incidence of malignancy, and the pathologic diagnosis from large single-institution case series as reported in the literature [3–7, 9, 15–34].

Table 1 Summary of large series (more than 20 cases) of retrorectal tumors

Author	Length of study (years)	Case #	Male/female (%)	Mean age	Malignancy	Classification		Neurogenic	Osseous	Miscellaneous	Surgical procedure (AA/PA/CA)
						Congenital	Inflammatory				
McColl [17]	20	23	NA	NA	10 (43 %)	17 (74 %)	–	2 (8.7 %)	–	4 (17 %)	NA
Whittaker and Pemberton [9]	14	22	36/64	36 (1–73)	11 (50 %)	18 (82 %)	–	–	1 (5 %)	3 (14 %)	0/100/0
Uhlig [7]	30	63	27/73	44.1	26 (42 %)	35 (55 %)	3 (5 %)	6 (10 %)	3 (5 %)	16 (25 %)	NA
Localio et al. [16]	16	20	50/50	45 (16–68)	12 (60 %)	12 (60 %)	–	3 (15 %)	2 (10 %)	3 (15 %)	15/20/65
Cody et al. [6]	28	39	46/54	(0–74)	39 (100 %)	19 (49 %)	–	6 (15.4 %)	4 (10.3 %)	10 (26 %)	30/30/40
Jao et al. [5]	19	120	38/62	43 (0–81)	51 (43 %)	79 (65 %)	–	14 (12 %)	13 (11 %)	14 (12 %)	21/77/2
Stewart et al. [4]	19	20	45/55	43 (0–78)	11 (55 %)	12 (60 %)	–	2 (10 %)	–	6 (30 %)	32/63/5
Vorobyov et al. [18]	18	36	13/23	52 (28–73)	0 (0 %)	–	–	–	–	36 (100 %)	0/29/7
Bohm et al. [3]	12	24	21/79	33 (22–74)	4 (17 %)	20 (83 %)	–	–	–	–	13/62/25
Wang et al. [19]	14	45	44/56	41 (15–76)	22 (48 %)	21 (47 %)	–	4 (9 %)	4 (9 %)	16 (36 %)	56/30/14
Lev-Chelouche et al. [20]	10	42	33/66	41 (21–84)	21 (50 %)	21 (50 %)	–	4 (10 %)	3 (7 %)	14 (33 %)	43/50/7
Glasgow et al. [15]	22	34	38/62	48 (21–80)	7 (21 %)	19 (56 %)	–	6 (18 %)	–	9 (26 %)	41/32/27
Grandjean et al. [21]	15	30	20/80	43 (16–77)	1 (3 %)	30 (100 %)	–	–	–	–	7/86/7
Woodfield et al. [22]	8	27	37/63	38 (21–88)	7 (26 %)	16 (59 %)	–	7 (26 %)	–	4 (15 %)	41/44/15
Pappalardo et al. [23]	14	34	44/56	42 (14–75)	14 (44 %)	20 (63 %)	3 (9 %)	1 (3 %)	3 (9 %)	5 (16 %)	47/16/37
Mathis et al. [24]	23	31	3/28	52 (27–79)	4 (13 %)	31 (100 %)	–	–	–	–	9/20/2
Yang et al. [25]	2	21	6/15	39 (16–74)	7 (33 %)	15 (71 %)	–	–	–	6 (29 %)	NA
Gao et al. [26]	8	39	26/74	40 (14–71)	6 (15 %)	20 (51 %)	3 (8 %)	3 (8 %)	–	13 (33 %)	22/72/6
Li et al. [27]	12	33	61/39	49 (18–71)	4 (12 %)	12 (36 %)	–	15 (45 %)	–	6 (18 %)	30/55/15
Lin et al. [28]	9	62	37/63	41 (15–68)	14 (23 %)	36 (58 %)	–	10 (16 %)	1 (2 %)	15 (24 %)	13/77/106
Bosca et al. [29]	6	30	43/57	47	20 (67 %)	15 (50 %)	3 (10 %)	1 (3.3 %)	2 (6.7 %)	9 (30 %)	15/75/10
Macafee et al. [30]	8	56	34/66	51 (20–88)	21 (38 %)	29 (52 %)	–	12 (21 %)	–	15 (27 %)	36/48/16
Chereau et al. [31]	4	47	13/34	46 (17–85)	9 (19 %)	45 (96 %)	–	–	–	2 (4 %)	1/42/4
Messick et al. [32]	30	109	33/76	48 (18–80)	23 (21 %)	60 (55 %)	–	9 (8 %)	2 (2 %)	16 (15 %)	25/49/10
Sagar et al. [33]	11	76	26/50	48 (19–88)	16 (21 %)	36 (47 %)	–	18 (24 %)	–	6 (8 %)	41/31/4
Simpson et al. [34]	13	26	7/19	38 (16–76)	5 (19 %)	26 (100 %)	–	–	–	–	5/15/6

AA anterior approach, *PA* posterior approach, *CA* combined approach, *NA* not available

Classification

In accordance with the Uhlig and Johnson classification [7], the primary tumors reported in the literature were classified as congenital, neurogenic, osseous, inflammatory, or miscellaneous (Table 2). Of the patients reviewed, 1194 (70 %) had benign tumors, while 514 patients (30 %) had malignant tumors. Malignancy occurred in 44.3 % of the men and 20.5 % of the women ($p < 0.05$). Congenital tumors (60.4 %) were the most frequent histologic type, followed by miscellaneous tumors (19.1 %), neurogenic tumors (14.8 %), osseous tumors (3.1 %), and inflammatory tumors (2.6 %).

Congenital lesions

Congenital tumors were identified in 1033 patients (60.5 %). The mean age was 43.7 ± 13.5 years. A female predominance (74 %) was found in patients with congenital tumors; 767 patients (74 %) had benign tumors, and tailgut cyst was the most prevalent lesion (45 % of benign congenital lesions). The remaining 265 (16 %) had malignant tumors. Chordoma was the most common malignant congenital tumor (64 %). Malignant degeneration occurred in 65 patients and included 60 tailgut cysts, 3 dermoid cysts, 1 epidermoid cyst, and 1 duplication cyst.

Patients with malignant tumors were significantly older than those with benign tumors (53.9 ± 11.5 vs. 40.1 ± 12.2 years; $p < 0.05$). The presence of a congenital malignant tumor was more common in males ($p < 0.05$) (Table 3). Malignant tumors had a higher rate of postoperative complications (21 vs. 7 %; $p < 0.05$). Furthermore, malignant tumors had a higher rate of recurrence than benign tumors (46.2 vs. 11.4 %; $p < 0.05$) (Table 3).

Neurogenic lesions

Neurogenic tumors were identified in 252 patients (14.8 %). The mean age of these patients was 42.8 ± 12.8 years. The majority of neurogenic tumors were benign (89 %, $n = 224$). Neurilemmomas were the most common benign neurogenic tumors (86 %, $n = 160$) (Table 3). Neurofibrosarcoma ($n = 7$) were the most common malignant neurogenic tumors. Malignant tumors had a higher recurrence rate compared to benign neurogenic tumors (42 vs. 6.7 %; $p < 0.05$) (Table 3).

Osseous lesions

Fifty-three patients (3.1 %) had osseous tumors, with a mean age was 37.9 ± 12.7 years. Of these patients, 58 %

Table 2 Classification of retrorectal tumors in our series (1708 cases in 341 papers)

Classification	Case (%)
Congenital	1033 (60.5)
Benign	768 (45.0)
Tailgut cyst	346
Teratoma	155
Epidermoid cyst	136
Dermoid cyst	67
Developmental cyst	24
Duplication cyst	21
Anterior sacral meningocele	16
Indeterminant cyst	1
Adrenal rest tumor	1
A-V malformation	1
Malignant	265 (15.5)
Chordoma	169
Tailgut cyst (malignant degeneration)	60
Malignant teratoma	31
Dermoid cyst (malignant degeneration)	3
Epidermoid cyst (malignant degeneration)	1
Duplication cyst (malignant degeneration)	1
Neurogenic	253 (14.8)
Benign	217 (12.7)
Neurilemmoma	160
Neurofibroma	41
Ganglioneuroma	19
Meningioma	2
Ganglioneurofibroma	1
Neuroblastoma	1
Malignant	28 (1.6)
Neurofibrosarcoma	7
Malignant Neurilemmoma	6
Neuroendocrine carcinoma	4
Neuroblastoma	4
Ependymoma	3
Ganglioneuroblastoma	2
Ependymal cell glioma	1
Neurofibroma, malignant degeneration	1
Osseus	53 (3.1)
Benign	16 (0.9)
Giant cell tumor	10
Aneurysmal bone cyst	2
Osteoma	1
Simple bone cyst	1
Osteochondroma	1
Synovioma	1
Malignant	37 (2.2)
Ewing tumor	16
Chondrosarcoma	13

Table 2 continued

Classification	Case (%)
Osteogenic sarcoma	4
Myeloma	2
Chondromyxosarcoma	1
Osteoblastoma	1
Inflammatory	45 (2.6)
Benign	45 (2.6)
Abscess	31
Granuloma	4
Fibrosis	4
Cyst hydatid	1
Inflammatory cyst	1
Unknown cyst	4
Malignant	0 (0)
Miscellaneous	326 (19.1)
Benign	142 (8.3)
Leiomyoma	50
Fibroma	12
Myelolipoma	12
Hemangiopericytoma	10
Lipoma	9
Angiomyxoma	9
Fibrolipoma	4
Paraganglioma	3
Desmoid tumor	3
Hemangioendothelioma	2
Granular cell tumor	2
Lymphoid hyperplasia (Castleman's ds)	2
Parachordoma	2
Hemangioma	2
Bronchogenic cyst	2
Hemangioblastoma	2
Angioleiomyoma	1
Rectal diverticulum	1
Hibernoma	1
Lymphangioma	1
Retention cyst of anal gland	1
Unknown	10
Malignant	184 (10.8)
Metastatic	24
Carcinoid	19
Gastrointestinal stromal tumor	19
Lymphoma	17
Leiomyosarcoma	16
Liposarcoma	13
Undifferentiated sarcoma	10
Fibrosarcoma	9
Rhabdomyosarcoma	8
Adenocarcinoma	7

Table 2 continued

Classification	Case (%)
Mucinous tumor	4
Primary carcinoma	4
Angiosarcoma	3
Plasma cell myeloma	2
Histiocytoma	2
Squamous cell carcinoma	2
Myelolipoma	2
Reticulum cell sarcoma	2
Myeloliposarcoma	1
IPMN	1
Mesothelioma	1
Hemangioma, malignant degeneration	1
Hemangioendothelial sarcoma	1
Granular cell tumor	1
Hemangiopericytoma	1
Epithelial tumor	1
Myosarcoma	1
Fibromyxoid sarcoma	1
Unknown	11
Total	1708 (100)

were male and 37 patients (70 %) had malignant tumors. Giant cell tumors were the most common benign osseous tumors (63 %), and Ewing tumors were the most common malignant osseous tumor (43 %). Increasing age was associated with more malignant tumors compared to benign osseous lesions (41.6 ± 14.0 vs. 32.2 ± 8.4 years, $p < 0.05$). However, there was no difference in postoperative complications and recurrence.

Inflammatory lesions

Inflammatory tumors were present 2.6 % of reported cases ($n = 45$). All of these tumors were benign: 31 abscesses, 4 granulomas, 4 fibrosis, 1 cyst hydatid, 1 inflammatory cyst, and 4 unknown cysts. The mean age was 44.9 ± 9.8 years. A male predominance (56 %) was found in the patients with an inflammatory lesion (56 vs. 44 %, $p > 0.05$).

Miscellaneous lesions

There were 326 patients (19.1 %) who had miscellaneous tumors. Mean age was 50.9 ± 14.4 years old and 62 % of the patients were female. There were no differences in age between benign and malignant groups. Malignant tumors had greater recurrence rates than benign tumors (53.4 vs. 11.5 %; $p < 0.05$). Leiomyoma was the most frequent benign histologic type (35 %), followed by a fibroma

Table 3 Demographic findings and clinical outcomes of retrorectal tumors included in the review

Classification	Case (%)	Male/female (%)	Mean age (range)	Complication (%)	Recurrence (%)
Congenital	1033 (60.5)	26/74	44 (14–80)	10	20.2
Benign	768 (45.0)	17/83*	40 (14–80)*	7*	11.4*
Malignant	265 (15.5)	52/48*	54 (18–76)*	21*	46.2*
Neurogenic	253 (14.8)	39/61	43 (14–79)	24.4	9.6
Benign	224 (13.1)	39/61	43 (14–79)	23.9	6.7*
Malignant	28 (1.6)	42/58	41 (17–72)	14.3	41.7*
Osseus	53 (3.1)	58/42	37.9 (14–73)	28.6	55.6
Benign	16 (1.4)	40/60	32.2 (14–50)*	100	55.6
Malignant	37 (2.3)	71/29	41.6 (15–73)*	23.1	55.6
Inflammatory	45 (2.6)	56/44	45 (24–75)	0	8.3
Benign	45 (2.6)	56/44	45 (24–75)	0	8.3
Malignant	0 (0)	–	–	–	–
Miscellaneous	326 (19.1)	38/62	51 (18–91)	10.3	31.5
Benign	142 (6.9)	33/67	50 (17–91)	9.5	11.5*
Malignant	184 (11.7)	43/57	52 (18–85)	11.5	53.4*
Overall benign	1184 (69.7)	24/76	42 (14–91)*	11.4	11*
Overall malignant	514 (30.3)	50/50	52 (15–85)*	17.5	49*
Total	1708 (100)	32/68	45 (14–91)	13.2	21.6

* Are reported as statistically significant $p < 0.05$

(8.5 %). The most frequent malignant miscellaneous tumors were metastatic tumors (13 %).

Clinical presentation

Of the patients reviewed, 20.6 % were asymptomatic at the diagnosis time. If present, the most common symptoms were lower back pain (14.8 %), abdominal pain (10.0 %), constipation (8.8 %), urinary symptom (5.3 %), pelvic pain (5.0 %), abscess/fistula (4.3 %), asymptomatic mass (4.2 %), leg pain (2.9 %), hip pain (1.4 %), tenesmus (0.8 %), rectal bleeding (0.8 %), dyspareunia (0.3 %), and headache (0.5 %). At the physical examination, 1554 patients (91 %) had a palpable mass in the presacral region. In most cases, other diagnostic evaluations were conducted, including sigmoidoscopy (36.5 %), ultrasonography (39.7 %), computed tomography (CT; 80.7 %), and magnetic resonance imaging (MRI; 69.5 %). Biopsy was performed in 27 % of the patients. Of these patients, incorrect diagnoses occurred in approximately half (44 %).

Surgical approach and outcomes

The operative approaches are listed into Table 4. The posterior approach was performed in 443 patients (51.5 %), anterior approach in 299 patients (34.7 %), and a combined approach in 119 patients (13.8 %). Adjacent organs (sacrum, coccyx, or rectum) were resected in 35 % of patients. The combined resection rate was significantly higher using a combined approach (81 %), compared with that of the anterior or posterior approach (12 vs. 33 %, respectively; $p < 0.05$). The overall mean postoperative hospital stay was 8 ± 6 days, with 8 ± 7 days after an anterior approach; 7 ± 5 days after a posterior approach; and 11 ± 7 days after a combined approach ($p < 0.05$).

The overall postoperative complication rate was 13.2 %. Posterior approach had an associated morbidity of 7.2 %, anterior approach 19.3 % and combined approach 24.7 % ($p < 0.05$). Postoperative complications included post-operative bleeding (28 %), neurogenic bladder (23 %), neurologic complication (18 %), wound infection (15 %), rectal injury (5 %), ureter injury (3 %), leakage (3 %), constipation (3 %) and meningitis (2 %). The overall postoperative recurrence rate was 21.6 %, with 6.7 % after an anterior approach, 26.6 % after a posterior approach, and 28.6 % after a combined approach ($p < 0.05$).

Minimal invasive approach

Laparoscopy was performed in 83 patients (Table 4). Laparoscopy was associated with shorter length of stay compared to open surgery (4 ± 2 vs. 9 ± 7 days; $p < 0.05$). Laparoscopic surgery had lower recurrence rates than open surgery (0 vs. 21.2 %; $p < 0.05$). Complication rate was similar between the two groups (19.8 % laparoscopy vs. 12.2 % open, $p = 0.44$). The operative time was shorter in laparoscopic surgery (148 ± 74 vs. 175 ± 126 min; $p < 0.05$). Six percent of all laparoscopic

cases involved malignant tumors ($n = 5$), including one tailgut cyst with malignant transformation, one GIST, one colloid sarcoma, one Ewing tumor, and one chondrosarcoma [35–38].

Robotic surgery was applied in five benign and two malignant minimally invasive cases (malignant neurofibroma, GIST). No recurrence or operative death was reported [39–41].

Discussion

Retrorectal tumors are heterogeneous group of tumors given that they arise from the pararectal space, which contains multiple embryologic remnants derived from various tissues; retrorectal tumors are rare tumors and thereby no consensus has been made in the literature regarding specific diagnosis, treatment, and surgical approach. To date, despite a multitude of smaller reviews and case reports, the optimal surgical approach is still in question. Our series of 1708 patients is the largest review of these entities ever conducted. We show that these tumors have a female predominance, and are most commonly benign in etiology and of a congenital classification. The most common presentation is that of perianal and low back pain, and these tumors can often be palpated on physical exam. Diagnostic methods such as sigmoidoscopy, ultrasonography, CT and MRI can be used to aid in diagnosis. Malignant lesions are more common in men and are associated with higher complication rates after resection, as well as higher recurrence rates. According to our results, the posterior approach emerges as the preferred method of operation with the lowest morbidity rate. Combined anterior/posterior approach had the highest recurrence and complication rate. However, a minimally invasive method of treatment specifically laparoscopy and robotic-assisted laparoscopy approach is feasible and was associated with significant shorter length of stay and possible lower recurrence rate, although it was mostly employed in cases of benign tumors.

Incidence

The incidence of retrorectal tumors in the general population is unknown. In medical literature, various reports have indicated the potential incidence of retrorectal tumors. At the Mayo Clinic, Whittaker et al. collected 22 cases from 1922 to 1936 and found the incidence to be only 1 out of approximately 40,000 registrations [9]. Jao et al. reported 120 cases of retrorectal tumors during a 19-year period, suggesting that approximately 6.3 patients would be diagnosed with retrorectal tumors on a yearly basis [5]. However, reports are generally from referral centers and thus do not represent the true prevalence of these tumors; therefore the above reported statistics may be much higher than that of the general population [2]. Uhlig and Johnson reported 63 adult cases over 30 years and demonstrated that, on average, 2 patients per year were diagnosed with retrorectal tumors in a standard major metropolitan area [7]. Our review demonstrates that retrorectal tumors are rare entities and in fact, Hobson et al. reported that a surgeon practicing outside the setting of a major referral center can expect to see, on average, at least 1 patient with a presacral tumor during the course of a typical career [2].

Pathological classification

As these tumors tend to have heterogeneity with respect to their histology, various classification systems have been proposed to categorize them [8]. In our analysis, we adhered to the classification proposed in 1975 by Uhlig and Johnson, distinguishing among congenital, inflammatory, neurogenic, osseous, and miscellaneous tumors [7].

The other important consideration for categorizing a retrorectal mass is its malignant nature [42]. Overall,

Table 4 Operative outcomes of retrorectal tumors according to surgical approach

	Case #	Mean OR (min)	LOS (days)	Complete resection (%)	Combined resection (%)	Complication rate (%)	Recurrence rate (%)
Approach							
Anterior	299	177 (±87)	8 (±7)	93.4	12.3*	19.3*	6.7*
Posterior	443	149 (±114)	7 (±5)	91.6	33.0*	7.2*	26.6*
Combined	119	255 (±171)*	11 (±7)*	87.6	80.5*	24.7*	28.6*
Operation method							
Open	1064	175 (±126)*	9 (±7)*	91.7	36.5*	12.2	21.2*
Laparoscopic	83	148 (±74)*	4 (±2)*	98.4	6.8*	19.8	0*

OR operative time, *LOS* length of hospital stay, ± standard deviation, *NA* not available

* Are reported as statistically significant $p < 0.05$

21–50 % of presacral tumors are malignant or contain areas of malignant change [5, 7, 15, 20]. The most common malignant lesion is the chordoma. Usually, malignant lesions are solid with signs of local invasion and bone destruction. Lev-Chelouche et al. divided retrorectal tumors into 4 groups according to the lesions' pathology: benign congenital, malignant congenital, benign acquired, and malignant acquired [20]. In the medical literature we reviewed, 1194 (70 %) had benign tumors, and 514 patients (30 %) had malignant tumors.

Retrorectal congenital tumors are more common in women (74 %), while the other disease groups had a similar distribution between the two genders. It is noteworthy, however, that malignant tumors occur more frequently in men [3, 5, 10]. Stewart et al. reported 20 cases of presacral tumors over 20 years and claimed malignancy occurred in 50 % of adults and was more common in men (62 %) than in women (38 %) [4]. In our review, 32 % of the patients were male, and malignancy occurred in 44.2 % of the men and 20.5 % of the women. A malignant diagnosis increased the risk of recurrences in congenital (46.2 vs. 11.4 %; $p < 0.05$), neurogenic (42 vs. 6.7 %; $p < 0.05$), and miscellaneous tumors (53.4 vs. 11.5 %; $p < 0.05$). Patients with malignant congenital tumors had also a higher risk to develop postoperative complications (21 vs. 7 %; $p < 0.05$). In an attempt to guide the therapeutic approach, it has been noted that solid tumors have a higher probability of malignancy than cystic lesions; however, cystic masses may still develop into malignant lesions [5, 15].

Diagnosis

Due to their retroperitoneal location, retrorectal tumors often grow to a significant size so to become symptomatic due to their mass effect on surrounding organs. Lower back pain and abdominal pain are undoubtedly the most common symptoms, and are more frequently associated with infectious or malignant etiology [4, 5]. Glasgow et al. reported that pain is present in 71 % of patients with malignancies versus 22 % of patients with benign lesions [15]. Retrorectal tumors can frequently present with infection, and patients whose tumors are infected might also experience pain. An infection within the tumor may present as an abscess, a draining sinus, or a fistula tract. Therefore, these tumors may be initially misdiagnosed as fistulas, perirectal abscesses, or pilonidal diseases. Singer et al. reported that patients underwent an average of 4.1 surgical procedures before the correct diagnosis of a retrorectal lesion was made [10]. Thus, it is important to have a high index of suspicion of a retrorectal tumor for the successful diagnosis and treatment of this condition [7, 15].

Since symptoms associated with retrorectal tumors may be vague and non-specific, the most important aspect for the diagnostic process is an accurate physical assessment, which facilitates the identification of an appropriate surgical method. A careful rectal examination is essential to ascertain the diagnosis in >90 % of the patients [2, 5]. Most lesions are soft, compressible, and easily missed if the physician does not maintain a high index of suspicion [2, 5]. Although small lesions may not be detected by sigmoidoscopy, flexible sigmoidoscopy can determine the involvement of the rectal mucosa and confirm the level of proximal extension of the tumor [2]. Transrectal ultrasonography (TRUS), combined with proctoscopy, has a sensitivity of 100 % and provides information on the size, consistency of the mass, and evidence of local invasion [15].

Moving onto imaging techniques, CT and MRI are widely used to confirm the diagnosis of a retrorectal tumor for preoperative surgical planning [2]. A CT scan of the pelvis can identify small tumors, distinguish a cystic lesion from a solid lesion, and reveal sacral involvement or invasion of adjacent structures. [2, 5]. MRI is particularly useful in delineating soft tissue planes and evaluating the presence or absence of bony invasion and nerve involvement. Nonetheless, Glasgow et al. reported the accuracy of an MRI and a CT scan for a specific histologic retrorectal tumor type was only 28 and 18 %, respectively [15]. They advocated that it is not advisable to avoid resection based solely on noninvasive studies [15].

Biopsy of a retrorectal mass is controversial, because it can lead to contamination or tumor spread [2, 15]. Jao et al. reported their findings for patients who had biopsies prior to tumor resection, and they advocated that preoperative biopsies may elicit tumor spread, abscesses, fecal fistulas, or meningitis, so biopsies should not be performed if the tumors are potentially resectable [2, 5, 43, 44]. A biopsy should be performed only if the lesion appears to be unresectable and if a tissue diagnosis is required to guide adjuvant therapy. In our series, 27 % of the patient of the patients underwent biopsy to confirm the diagnosis. Of these patients, incorrect diagnoses occurred in 44 %. An accurate pathological diagnosis with a preoperative biopsy is often very difficult and the biopsy may lead to a misdiagnosis. Also, the histological type of retrorectal tumor does not influence the choice of surgical approach. Therefore, a biopsy prior to an operation is usually not recommended.

Treatment paradigm and surgical approaches

Once the diagnosis is established, surgical resection is the best therapeutic option, even in asymptomatic patients, as many lesions may contain malignant elements, possess the potential for growth or malignancy, or cause complications, such as infection [2]. Complete surgical resection is

the cornerstone in the management of retrorectal tumors. En bloc excision permits confirmation of the diagnosis and eliminates the risk of complications such as infection, compression, potential for recurrence, and malignant degeneration [5, 12, 20].

The narrow space of the pelvis and the anatomical complexity of the retrorectal region can be challenging with respect to the surgical treatment of these tumors. The extent of surgery is determined by the characteristics of the tumor. Benign retrorectal tumors require complete gross resection, whereas malignant tumors will require radical resection, including en bloc resection of adjacent organs [42].

In certain circumstances where there is significant adjacent organ invasion, or severe adhesion to normal organ, combined resection including the normal adjacent organs may be necessary. Invasion of the rectum requires a rectal resection, and sacrococcygeal invasion requires coccygectomy or sacrectomy. In these complicated cases, a multidisciplinary team consisting of a radiologist, colorectal, urological, plastic, orthopedic, and neuro-surgeons may be necessary.

The surgical treatment of retrorectal tumors may be associated with an increased risk of complications such as presacral bleeding and rectal and nerve injuries. Jao et al. reported that the rate of postoperative complication after resection of a retrorectal tumor was 45 % [5]. In our literature review, postoperative complications included neurogenic bladder (15 %), wound infection (11 %), dysesthesia (7 %), fecal incontinence (7 %), massive bleeding (4 %), retrorectal abscess (3 %), and fecal fistula (1 %) [5]. Glasgow et al. reported 7 major postoperative complications in their 34 cases of retrorectal tumors, which included the following: transected ureter ($n = 1$), extensive soft-tissue infection ($n = 1$), reoperation for bleeding ($n = 2$), and impotency ($n = 2$) [15]. They mentioned that complications tended to occur more frequently in patients with malignant tumors; however, the choice of operative approach did not influence the occurrence of a postoperative complication [15].

Three major surgical approaches have been proposed for the resection of retrorectal tumors: the anterior (abdominal) approach, the posterior (perineal, trans-sacral, and parasacrococcygeal) approach, and the combined anterior and posterior pelvic approach. The appropriate technique depends on the lesion's location, size, as well as the tumor's relationship to the adjacent structures [20].

Anterior approach

Surgical management of tumors above the S3 level is best managed with an anterior (open or laparoscopic) or combined approach. Also, lesions which are potentially malignant are amenable to this approach, as it provides direct visualization of the pelvic side walls and pelvic viscera. The anterior approach has traditionally been the preferred method for tumors with the inferior most portion extending above the S4 level and if the sacrum is not involved [2, 42, 45]. A particular advantage of the anterior approach is that it allows the surgeon to have excellent exposure to major pelvic structures, such as the iliac vessels and ureters [3].

Posterior approach

In contrast to the anterior approach, the posterior approach is preferred for small, benign tumors that do not extend above the S4 level. When the superior border of the tumor is palpable and mobile on distal examination, the posterior approach should be considered. Also, this approach is preferred in cases of nerve involvement, given the improved visualization of nerves via a posterior approach [2]. However, the major disadvantages of the posterior approach are the risk of major intraoperative pelvic hemorrhage and potential injury to the lateral pelvic nerves [12]. A larger tumor, or a tumor in an intermediate location, may require a combined approach [2, 3]. Because of the potential for progression of these tumors deep into the pelvis and as identification of the planes between the tumor and the surrounding tissues becomes more difficult, patients may have to be repositioned for the perineal phase of the procedure.

Combined approach

The benefits of the combined approach include improved visualization of vital structures such as the ureters, vessels, pelvic nerves, and rectum by means of the anterior approach as well as enhanced exposure of the nerve roots provided by the posterior approach. If large tumors or infected masses involve the rectum or presacral fascia, the tumors often obscure normal surgical planes and may cause significant adhesions to the adjacent tissue. A combined approach is suggested in these situations [2, 3, 28].

Minimally invasive approach

The location of these tumors pose a challenge given that working in a narrow, deep space can be difficult and may increase the risk of damage to the pelvic vessels and nerves. In recent years, minimally invasive surgical techniques have been increasingly utilized to excise retrorectal tumors (Table 5).

In our review, we report 83 cases of laparoscopic excisions or combined laparoscopic and perineal surgical approaches for retrorectal tumors [27, 35–41, 46–79]. These reports demonstrated the safety of laparoscopy for

Table 5 Demographic data and clinical outcomes of minimally invasive approach for retrorectal tumors

Author	Case #	Age	Sex	Diagnosis	Symptom	Procedure type	Tumor location	Mass size (cm)	OR (min)	LOS (day)	Complication
Sharpe et al. [46]	1	29	F	Dermoid cyst	Pelvic pain	Lap	Right pararectal	5	225	1	0
Melvin [47]	1	36	F	Schwannoma	Abdominal pain	Lap	Presacral	2.5	NA	NA	0
Salameh et al. [48]	1	49	F	Duplication cyst	Rectal pain	Lap	Right pararectal	6	NA	3	0
Kohler et al. [49]	1	15	F	Ganglioneurofibroma	None	Lap	Presacral	10	385	11	0
Bax et al. [50]	1	17	F	Teratoma	NA	Combined	Sacrococcygeal	NA	NA	NA	NA
Konstantinidis et al. [51]	2	40	F	Schwannoma	Pelvic pain	Lap	Left pararectal	4	90	3	0
		46	F	Schwannoma	Pelvic pain	Lap	Left pararectal	6	110	3	0
Sriganeshan and Alexis [52]	1	36	F	Tailgut cyst	None	Lap	Presacral	6	NA	3	0
Yang et al. [53]	1	67	M	Schwannoma	Constipation	Lap	Presacral	5.1	NA	4	0
Sharma et al. [54]	1	35	F	Dermoid	Rectal pain	Lap	Presacral	10	NA	NA	0
Palanivelu et al. [55]	1	60	F	Epidermoid	Pain, swelling in perineum	Combined	Presacral perineal extension	16	174	4	0
Gunkova et al. [56]	2	43	F	Retrorectal cyst	Abdominal pain	Lap	Presacral	9.5	75	6	0
		28	F	Retrorectal cyst	Anal pain	Lap	Presacral	9.3	90	5	0
Cho et al. [57]	1	27	F	Teratoma	Lower back pain	Lap	Presacral	10	NA	3	0
Chen et al. [58]	1	15	F	Teratoma	Pelvic pain	Lap	Presacral	10	185	5	0
Al-Khattabi et al. [59]	1	58	F	Epidermoid	Pelvic pain, constipation	Combined	Left pararectal	9	NA	NA	0
Champney et al. [60]	1	32	F	Ganglioneuroma	Lower back pain	Lap	Presacral	5.7	NA	1	0
La Rosa et al. [35]	1	73	F	Tailgut cyst	Abdominal pain	Lap	Presacral	3.7	NA	6	0
Lu and Tseng [61]	1	24	F	Tailgut cyst	Constipation	Lap	Presacral	9	NA	3	0
Rao et al. [62]	1	30	F	Schwannoma	Pelvic pain	Lap	Retrorectal	9	116	2	0
Takahashi et al. [36]	1	54	M	GIST	Pain with defecation, rectal bleed	Lap	Presacral, precoccygeal	4.6	NA	NA	0
Witherspoon et al. [63]	1	37	F	Schwannoma	Abdominal pain	Lap	Retrorectal	10	NA	3	0
Asuquo et al. [64]	1	74	F	Myelolipoma	None	Lap	Presacral	3.5	NA	NA	0
Kye et al. [65]	4	45	F	Fibroma	Abdominal pain	Lap	NA	5.0	NA	NA	0
		59	M	Schwannoma	Abdominal pain	Lap	NA	4.5	NA	NA	0
		20	F	Teratoma	Abdominal pain	Lap	NA	7.0	NA	NA	0
		57	F	Epidermoid cyst	None	Combined	NA	6.0	NA	NA	0
Leppard et al. [66]	1	56	F	Tailgut cyst	None	Lap	Presacral	5.6	NA	2	0
Lim et al. [67]	1	36	F	Tailgut cyst	None	Lap	Presacral, precoccygeal	4, 2.5	90	3	0

Table 5 continued

Author	Case #	Age	Sex	Diagnosis	Symptom	Procedure type	Tumor location	Mass size (cm)	OR (min)	LOS (day)	Complication
Marinello et al. [68]	4	38	F	Teratoma	Abdominal pain	Lap	Presacral	11	210	NA	0
		33	F	Fibrous tumor	Abdominal pain	Lap	Presacral	7.5	180	NA	0
		35	M	Schwannoma	Lower back pain	Combined	Presacral	10	250	NA	Wound infection
		63	M	Schwannoma	Lower back pain	Lap	Presacral	6.5	200	NA	Urinary retention
Tsutsui et al. [69]	1	27	F	Teratoma	NA	Lap	Presacral	6	NA	7	0
Paul et al. [70]	1	38	F	Neurofibroma	Pelvic pain	Lap	Right pararectal	6.3	60	1	0
Morelli et al. [39]	1	17	F	Dermoid cyst	Mass in buttock	Totally RO	Anterior pararectal	17	180	3	0
Jones and Khosa [71]	1	15	F	Dermoid cyst	Abdominal pain	Lap	Left pararectal	7.5	NA	NA	0
Li et al. [27]	1	35	F	Epidermoid oocyst	Perianal pain	Lap	Presacral	2.5	NA	NA	0
Nedelcu et al. [72]	9	35 (19–48)	2F 7M	4 schwannoma 1 ganglioneuroma 2 tailgut cyst 1 anterior meningocele 1 paraganglioma	4 no symptoms 5 pain (rectal pain, nonspecific pain)	Lap	Presacral	6.8 (3–11.5)	NA	4.7 (4–8)	Pelvic hematoma 4 neuro sx (parasthesia, sciatic pain), retrograde ejaculation
Szyllo and Lesnik [73]	1	26	F	Teratoma	Abdominal pain	Lap	Presacral	4	90	7	0
Duclos et al. [37]	12	61	F	Schwannoma	None	Lap	Presacral	5	130	NA	Bleeding
		63	F	Teratoma	None	Lap	Presacral	12	180	NA	0
		55	F	Schwannoma	Abdominal pain	Lap	Presacral	5	70	NA	0
		46	F	Colloid sarcoma	None	Lap	Precoccygeal	5	140	NA	0
		67	F	Tailgut cyst	None	Lap	Precoccygeal	2	150	NA	Rectal injury (stoma)
		22	F	Epidermoid cyst	None	Lap	Precoccygeal	6	90	NA	None
		30	F	Ewing sarcoma	None	Lap	Presacral	3	180	NA	Technical difficulty
		64	F	Teratoma	Constipation	Lap	Precoccygeal	7	80	NA	0
		28	F	Tailgut cyst	None	Lap	Precoccygeal	5	100	NA	UTI
		89	F	Lipoma	Abdominal pain	Lap	Presacral	6	215	NA	UTI
		55	F	Tailgut cyst	None	Lap	Precoccygeal	5	215	NA	0
		40	F	Tailgut cyst	None	Lap	Presacral	6.5	180	NA	0
Fong et al. [74]	10	37	F	Schwannoma	NA	Lap	NA	10.5	NA	NA	0
		31	F	Schwannoma	NA	Lap	NA	9	NA	NA	0
		44	F	Schwannoma	NA	Lap	NA	15	NA	NA	0
		34	F	Ganglioneuroma	NA	Lap	NA	8.5	NA	NA	Wound infx

Table 5 continued

Author	Case #	Age	Sex	Diagnosis	Symptom	Procedure type	Tumor location	Mass size (cm)	OR (min)	LOS (day)	Complication
		55	F	Ganglioneuroma	NA	Lap	NA	7.5	NA	NA	0
		46	M	Schwannoma	NA	Lap	NA	4.5	NA	0	
		23	F	Epidermoid cyst	NA	Lap	NA	5	NA	0	
		46	M	Hemangioma	NA	Lap	NA	5.5	NA	0	
		79	F	Schwannoma	NA	Lap	NA	10	NA	0	Wound infx
	58	M	Schwannoma		NA	Lap	NA	6	0	0	0
Hove et al. [75]	1	NA	NA	Schwannoma	None	SPA	Presacral	5.9	75	3	0
Imboden et al. [76]	1	44	F	Epidermoid cyst	Pelvic pressure	Lap	Presacral	7	90	NA	0
Kassir and Kaczmarek [77]	1	55	M	Dermoid cyst	Constipation	Lap	Presacral	5	NA	NA	0
Kneist et al. [78]	2	45	F	Schwannoma	NA	Lap	NA	NA	NA	NA	NA
		42	F	Teratoma	NA	Lap	NA	NA	NA	NA	NA
Oh et al. [40]	5	45	M	Schwannoma	Numbness	Totally RO	Presacral	>10	240	9	Numbness
		42	M	Meningioma	Rapid growth	Totally RO	Presacral	>10	120	4	0
		42	F	Neurofibroma	Rapid growth	Totally RO	Presacral	>10	300	8	Neuropathic pain
		26	M	Schwannoma	None	Totally RO	Presacral	>10	150	4	0
		29	F	Schwannoma	Pelvic pain	Totally RO	Presacral	>10	280	10	0
Ploumidis et al. [41]	1	17	F	GIST	None	Totally RO	Anterior pararectal	17	NA	3	0
Possover et al. [38]	1	33	F	Chondrosarcoma	Back pain	CA-AS	Right pararectal	NA	360	NA	0
Zhou et al. [79]	8	75	M	Schwannoma	None	Lap	NA	8	120	5	0
		63	F	Schwannoma	Abdominal pain	Lap	NA	6.3	95	4	0
		47	F	Epidermoid cyst	None	Lap	NA	11.8	180	3	0
		29	F	Epidermoid cyst	None	Lap	NA	7	85	5	0
		23	M	Teratoma	None	Lap	NA	10	150	7	0
		15	F	Teratoma	None	Lap	NA	9	150	5	0
		27	F	Epidermoid cyst	None	Lap	Right pararectal	9.3	115	5	0
		43	F	Leiomyoma	None	Lap	NA	9.4	80	3	0

M male, *F* female, *LOS* length of stay, *OR* operation time, *RO* robotic approach, *Lap* laparoscopic approach, *SPA* single-site access, *Combined* combined transabdominal laparoscopic and perineal approach, *NA* not available

treating retrorectal tumors. Laparoscopy offers the advantage of enhanced visualization of pelvic structures and facilitates precise dissection of the tumor from adjacent structures. There are no absolute indications to choose a laparoscopic access over laparotomy, and it still remains unclear whether open surgery or laparoscopy provides the best long-term results. In our review, there was no operative mortality or recurrences of tumor during the follow-up period (mean of 30 months); however, it is noteworthy that only 6 % of laparoscopic cases were for malignant cases (one tailgut cyst with malignant transformation, one GIST, one sarcoma, one Ewing tumor, one chondrosarcoma) [35–38]. Based on that total number of laparoscopic cases, we can conclude that laparoscopy is a safe and feasible method for retrorectal tumors; however, long-term studies with more are needed in order to define an advantage for both benign and malignant cases.

In the context of minimally invasive surgery, robotic surgery has several potential advantages compared to laparoscopic surgery, particularly within the narrow confines of the pelvis (three-dimensional view, superior dexterity, multi-articulated instruments). To date, to the best of our knowledge, only seven robotic cases have been described [39–41], and only in two cases the robotic technique was applied to malignant tumors (one malignant neurofibroma, one GIST). Based on previous studies on robotic resection of rectal tumors [80, 81], and the absence of recurrence or operative death in the robotic cases listed in our review [40, 41], we can hypothesize a similar outcome for patients who underwent robotic and laparoscopic retrorectal tumors resection. However, larger robotic retrorectal case-series are needed in order to draw a definitive conclusion.

Limitations

The limitations of this study include lack of large-scale studies. Because of the rarity of these tumors, the majority of published studies were case reports or case series. A small number of reviews have been described. Despite the limitation of this review, this is the largest and most comprehensive review in the literature and the first to compare surgical outcomes to date.

Conclusion

The most common retrorectal tumors are benign congenital tumors. Diagnosis of these lesions is accomplished via a thorough physical examination in conjunction with radiological imaging, ultrasonography, and ultrasound endoscopy. Biopsy, in most instances, can be avoided since it leads to an inaccurate diagnosis and may cause tumor seeding and other complications. The recommended treatment for retrorectal tumors is complete surgical excision given their malignant potential, their propensity to cause local complications, and their potential for recurrence. Minimal invasive surgery appears to be associated with shorter length of stay, shorter operative time, and possible lower recurrence rates. Robotic approach appears to be a safe and feasible option for the management of retrorectal tumors but conclusion is difficult due to the small number of cases available thus far.

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