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Journal of Clinical Imaging Science

Review Article *Gastrointestinal Imaging*

Presacral tumors: A systematic review of literature

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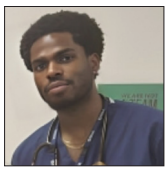

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Received: 15 March 2024
Accepted: 22 April 2024
Published: 29 May 2024

DOI
10.25259/JCIS_27_2024

Quick Response Code:

ABSTRACT

Presacral/Retrorectal tumors (RRT) are rare lesions that comprise a multitude of histological types. Data on surgical management are limited to case reports and small case series. The aim of the study was to provide a comprehensive review of the epidemiology, pathological subtypes, surgical approaches, and clinical outcomes. A PubMed search using terms "retrorectal tumor" and "presacral tumor" was used to identify articles reporting RRT of non-urological, non-gynecologic, and non-metastatic origin. Articles included were between 2015 and 2023. A total of 68 studies were included, comprising 570 patients. About 68.2% of patients were female, and the mean overall age of both sexes was 48.6 years. Based on histopathology, 466 patients (81.8%) had benign lesions, and 104 (18.2%) were malignant. In terms of surgical approach, 191 (33.5%) were treated anteriorly, 240 (42.1%) through a posterior approach, and 66 (11.6%) combined. The mean length of stay was 7.6 days. Patients treated using the posterior approach had a shorter length of stay (5.7 days) compared to the anterior and combined approaches. RRT are rare tumors of congenital nature with prevalence among the female sex. R0 resection is crucial in its management, and minimal access surgery appears to be a safer option in appropriate case selection.

Keywords: Retrorectal tumors, Congenital cystic lesions, Perineal approach, Transabdominal approach, Combined abdominoperineal approach

INTRODUCTION

The anatomical complexity and inherent heterogeneity of retrorectal and presacral space lesions pose significant diagnostic hurdles, rendering their surgical management a subject of ongoing debate within the medical community.[1] The configuration of the retrorectal/presacral space presents significant surgical challenges due to its intricate anatomical architecture. This space is defined by the mesorectum anteriorly, the presacral fascia posteriorly, and the lateral boundaries of the iliac vessels and ureters. While precise epidemiological data regarding these lesions are limited, existing literature suggests an estimated incidence of 1 in 40,000–60,000 hospital admissions. Notably, these neoplasms demonstrate a female predilection and a median age of onset at 45 years.[2,3] The rise in diagnostic accuracy for this pathology can be linked to the combined influence of technological advancements in imaging and the increased use of these technologies in gynecological investigations, where the condition is a common finding. This explains the observed higher prevalence among women in their early reproductive years.[4] Retrorectal tumors (RRT) are characterized by a high prevalence of benignity and asymptomatic presentation. Nevertheless, the significant variability in malignant incidence, ranging from 21% to 50%, necessitates careful clinical assessment and prompt management strategies.[5,6] While the

majority of RRT exhibit benign histology, surgical resection is often advised in light of the non-negligible risk of malignant degeneration (up to 8%) and the potential for suppurative complications, estimated at approximately 30%.[4,7] Surgical management of retrorectal tumors necessitates meticulous selection of the approach to ensure adequate exposure, minimize iatrogenic injury, and optimize clinical outcomes while mitigating complications. Recognizing the rarity of these lesions and the paucity of dedicated literature, RRTs pose a significant diagnostic and therapeutic challenge for contemporary surgeons. Therefore, this systematic review offers a valuable contribution to the existing literature. As such, this study aims to deliver a comprehensive analysis of the epidemiology, pathological subtypes, surgical approaches, and clinical outcomes associated with RRTs.

MATERIAL AND METHODS

To comprehensively elucidate the intricacies of RRT, we conducted a systematic literature review encompassing their epidemiology, pathologic subtypes, diverse surgical approaches, and associated clinical outcomes.

Search strategy

The review was conducted in line with the protocol, in accordance with the Cochrane handbook for systematic reviews of interventions and reported in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines.[8,9] This review was registered in PROSPERO: CRD42024500067.

A PubMed search using the terms "retrorectal tumor," and "presacral tumor" was used to identify the articles reporting of RRT. The search was undertaken on the September 01, 2023, and looked at all articles between March 2015 and September 2023.

Data selection

Initially records were first screened for relevance based on their title and abstract. Two authors (J.O., V.B.) independently screened records for inclusion and were blinded to each other's decisions. Disagreements between individual judgments were resolved by consensus, and if no agreement could be made, a third author (A.M.) was consulted.

Articles were included if they were full-text case reports, case series or review articles describing primary RRT. The target population consisted primarily of adults (≥16 years old). Articles reporting metastatic disease, urological or gynecological origin of tumor were excluded from the study. Articles which were descriptive and non-English were also excluded from the present study. A flowchart of the selection process in the format of the PRISMA is presented [Figure 1].[9]

Data extraction

Two independent reviewers (J.O., V.B.) manually extracted data from full-text articles into a structured database (Microsoft Excel). Extracted fields included primary author, publication year, key study characteristics (sample size, age, gender, and recruitment design), histological type of reported tumors (congenital, neurogenic, osseous, inflammatory, or miscellaneous), surgical approach, operative time, length of stay, resection status for malignancies, overall complication rate, and recurrence status. Disagreements regarding data extraction or categorization were resolved through discussion and consensus. If consensus could not be reached, a senior author (A.M.) adjudicated the case.

Definitions

Tumors were categorized according to established definitions: Congenital – lesions present at birth (most common); neurogenic – slow-growing tumors arising from peripheral nerves (second most common); osseous – tumors arising from bone, cartilage, or marrow; inflammatory less common, potentially linked to perirectal or abdominal infections; and miscellaneous – encompasses 10–25% of cases, including lipomas, fibromas, hemangiomas, leiomyomas, and liposarcomas.[10]

Data analysis

Given the significant heterogeneity between studies on this topic, a formal narrative synthesis was the most appropriate analytical approach. Statistical analyses primarily relied on descriptive statistics, where findings were presented as rates, means, and ranges. For subgroups defined by histological type (e.g., congenital, neurogenic, osseous, inflammatory, and miscellaneous), survival, malignancy potential, surgical approach, and tumor recurrence rates were expressed as rates and means.

RESULTS

Overview on basic characteristics of included studies

A comprehensive literature search yielded 595 citations encompassing abstracts and full-text articles. Figure 1 illustrates the selection process, ensuring a well-defined and representative sample for subsequent analysis. Rigorous screening eliminated duplicates (26 conference abstracts); irrelevant studies based on selection criteria were excluded (n = 501). Ultimately, 68 studies (570 patients) met the inclusion criteria, focusing on primary RRTs. Table 1 summarizes single case reports, while Table 2 details case series. Females constituted 68.2% of the cohort, with an average age of 48.6 years (range: 18–89).

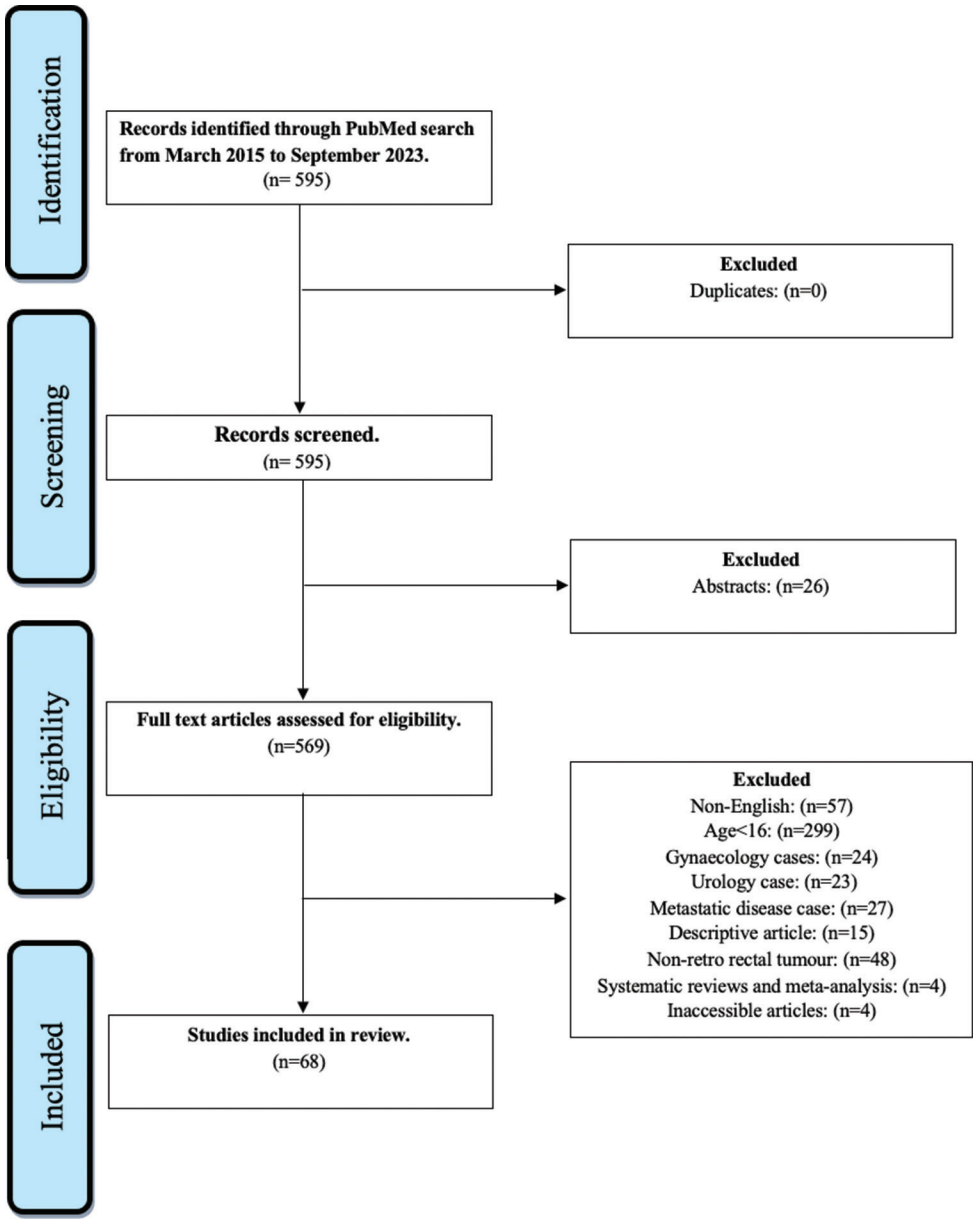

Figure 1: Preferred Reporting Items for Systematic Reviews and Meta-Analyses flow diagram.

The primary tumors in this study were classified according to the Uhlig and Johnson classification system.[11] This system categorizes tumors into five groups: congenital, neurogenic, osseous, inflammatory, and miscellaneous [Table 3]. The majority of tumors in this study were benign (466, or 81.8%). The most common type of benign tumor was congenital (56.8%), followed by neurogenic (15.8%), miscellaneous (8.9%), inflammatory (7.2%), and osseous (0%).

This study identified 324 patients (56.8%) with congenital tumors, of whom 284 (87.7%) harbored benign lesions (primarily tailgut cysts at 44.8%). Malignant tumors were diagnosed in 40 patients (12.3%), exhibiting a statistically significant association with older age (51.4 vs. 43.1 years). Compared to benign counterparts, malignant tumors displayed a markedly higher recurrence rate (24.2% vs. 2%).

Amongst the studied patients, 18.1% (n = 103) presented with neurogenic tumors, characterized by a mean age of 48 years and a striking predominance of benignity (87.4%, n = 90). Schwannomas emerged as the leading benign subtype (9.5%, n = 54), while retention cyst of the anal gland constituted the primary form of malignancy (n = 4). Notably, malignant neurogenic tumors exhibited a significantly higher recurrence rate compared to their benign counterparts (7.1% vs. 3.2%) [Table 4].

Table 1: Summary of case reports of retrorectal tumors

Author	Patient Age	Sex	Primary Lesion / Recurrence	Primary Symptoms	Diagnosed Incidentally?	Diagnosis method	Preoperative Histopathology	Mean tumor diameter on cross sectional imaging (cm)	Lesion extension Above/ Below S3	Operative approach (A/P/C)	Operative method (Open/Lap/ Robotic/Converted- reason for conversion/TAMIS)	Benign/ Malignant	Histopathology	Mean OR (min)	LOS (days)	Complete resection? (Y/N/NR)	Combined resection? (Y/N/ NR)	Overall Complication? (Y/N/ NR)	Recurrence reported in F/U period? (Y/N/ NR)
Kumassah PK, *et al.*[12]	37	F	Primary	Coccydynia CIBH	No	MRI	FNA	NR	Above	C	Open	Mal	Cystic hamartoma, with mucinous adenocarcinoma	NR	6	Y	Y (en bloc with rectum with coloanal anastomosis)	N	N
Iwata E, *et al.*[13]	25	F	Primary	Coccydynia	No	CT	FNA	3.5	Below	P (Kraske)	Open	Mal	Tailgut cyst in which a Grade 2 Neuroendocrine Tumor	NR	NR	Y	N	NR	NR
Cataneo J, *et al.*[14]	34	F	Primary	CIBH	No	MRI	Not performed	NR	Below	A	Robotic	B	Dermoid cyst	NR	1	Y	N	N	N
Fang H, *et al.*[15]	43	M	Primary	Asymptomatic	Yes	MRI	FNA	16.1	Above	A	Open	B	Tailgut cyst	NR	5	Y	N	N	N
Mora-Guzmán I, *et al.*[16]	56	F	Primary	Coccydynia, anal fistula	No	MRI	FNA	4.1	Below	P (Kraske)	Open	B	Tailgut cyst	NR	5	Y	Y (en bloc with the coccyx)	N	N
Jun C, *et al.*[17]	22	F	Primary	Asymptomatic	Yes	MRI	FNA	NR	Above	A	Robotic	B	Schwannoma	NR	3	Y	N	N	NR
Criss CN, *et al.*[18]	19	F	Recurrence	Left buttock and thigh pain	No	MRI	Not performed	8.2	Above	A	Robotic	B	Lipoblastoma	960	4	Y	N	N	NR
Yang BL, *et al.*[19]	36	M	Recurrence	Coccydynia	No	MRI	Known from prior resections	4	Below	P (Kraske)	Open	Mal	Moderately differentiated adenocarcinoma	NR	NR	Y	N	N	N
Cho MH, *et al.*[20]	70	F	Primary	Coccydynia	No	MRI	FNA	3.7	NR	NR	NR	B	Myelolipoma	NR	NR	Y	N	N	NR
Roy SP, *et al.*[21]	29	F	Primary	Coccydynia	No	USS	Not performed	4.6	Above	A	Robotic	B	Tailgut cyst	NR	2	Y	N	N	NR
Kesavan S, *et al.*[22]	51	F	Primary	Intermittent lower abdominal pain	Yes	USS	Not performed	4.7	Above	A	Laparoscopic	B	Epidermoid cyst	NR	2	Y	N	N	N
Tarchouli M, *et al.*[23]	45	M	Primary	Intermittent lower abdominal pain	No	MRI	Not performed	7	Below	P (Kraske)	Open	Mal	Low-grade leiomyosarcoma	NR	2	Y	N	N	N
Bouzid A, *et al.*[24]	22	F	Primary	Intermittent lower abdominal pain	No	USS	Not performed	8.5	Above	A	Laparoscopic converted to open due to suspicion of S1 nerve involvement	B	Ganglioneuroma	NR	7	N	N	N	N
Gutierrez O, *et al.*[25]	37	F	Primary	Acute pelvic pain	Yes	MRI	Not performed	2.9	Below	P (York-Mason)	Open	B	Cystic hamartoma	NR	NR	Y	N	Y	NR
Tan GHC, *et al.*[26]	76	M	Primary	Acute pelvic pain	Yes	CT	Not performed	15	Above	A	Open	B	Angiomyxoma/liposarcoma	415	43	Y	N	Y (SSI required surgical drainage)	N
Seydafkan S, *et al.*[27]	52	F	Recurrent	Asymptomatic	Yes	USS	Known from prior resections	5.3	Below	P (Kraske)	Open	B	Tailgut cyst	NR	NR	Y	N	NR	NR
Carchman E, Gorgun E[28]	76	M	Primary	Obstructive uropathy	No	CT	FNA	8	Above	A	Robotic	B	Low-grade fibromyxoid sarcoma fibrosarcoma	NR	3	Y	N	N	NR
Alvarez-Sarrado E, *et al.*[29]	49	F	Primary	Asymptomatic	Yes	CT	Not performed	13.5	Above	A	Open	B	Epidermoid cyst	NR	60	Y	N	Y (Decompensation of epilepsy)	NR
Şahin S *et al.*[30]	55	F	Primary	Asymptomatic	Yes	MRI	Not performed	17.3	Below	P (Kraske)	Open	Mal	Tailgut cyst (malignant degeneration)	NR	NR	Y	N	NR	NR
Pizzuti V *et al.*[31]	41	M	Primary	Coccydynia	No	MRI	Not performed	4	Below	P (Modified Kraske)	Open	B	Schwannoma	NR	4	Y	N	N	NR
Patel A *et al.*[32]	54	F	Recurrence	Coccydynia	No	MRI	Known from prior resections	9.7	Below	C	Laparoscopic also Kraske	B	Tailgut cyst	160	4	Y	NR	N	N
Perungo T *et al.*[33]	23	F	Primary	Coccydynia	No	CT	Not performed	4.6	Below	P (Kraske)	Open	B	Epidermoid cyst.	NR	NR	Y	N	N	N
Inada R *et al.*[34]	67	F	Primary	Asymptomatic	Yes	CT	Not performed	NR	Below	A	Laparoscopic	B	Tailgut cyst	199	NR	Y	Y (en block with descending colon)	N	N
Huang M, *et al.*[35]	34	M	Primary	Asymptomatic	Yes	USS	FNA	11.3	Above	A	Open	Mal	Schwannoma	NR	7	Y	N	N	N
Kawamura J, *et al.*[36]	61	F	Primary	Asymptomatic	Yes	CT	FNA	NR	Below	P	TAMIS	Mal	Extra-nodal marginal-zone lymphoma of mucosa associated lymphoid tissue (MALT)	100	7	Y	N	N	NR
Zhu XL[37]	51	M	Primary	Abdominal pain	No	CT	Not performed	9.3	Above	A	Open	Mal	primary alveolar rhabdomyosarcoma	NR	NR	Y	N	N	N
Tobias-Machado M[38]	60	M	Primary	Abdominal pain	No	CT	Not performed	NR	Above	A	Laparoscopic	B	Schwannoma	150	2	Y	N	N	NR
Alshahri J[39]	74	M	Primary	Back pain	No	CT	Not performed	11.2	Above	A	Open	Mal	Synchronous Chondroma of sacrum and moderately differentiated adenocarcinoma	NR	NR	Y	Y (en bock with sigmoid colon with end-to-end colorectal anastomosis as well as low sacrectomy with VRAM flap)	Y (SSI requiring IR drainage as well as postoperative PE)	N
Kearney D[40]	58	F	Primary	Chronic abscess in sacral area	Yes	MRI	FNA	9.3	Below	P (Kraske)	Open	B	Tailgut cyst	NR	5	Y	N	N	N

(Contd)

Table 1: *(Continued).*

Author	Patient Age	Sex	Primary Lesion / Recurrence	Primary Symptoms	Diagnosed Incidentally?	Diagnosis method	Preoperative Histopathology	Mean tumor diameter on cross sectional imaging (cm)	Lesion extension Above/ Below S3	Operative approach (A/P/C)	Operative method (Open/Lap/ Robotic/Converted- reason for conversion/TAMIS)	Benign/ Malignant	Histopathology	Mean OR (min)	LOS (days)	Complete resection? (Y/N/NR)	Combined resection? (Y/N/ NR)	Overall Complication? (Y/N/ NR)	Recurrence reported in F/U period? (Y/N/NR)
Mahajan UV[41]	62	F	Primary	Back pain radiating to the whole lower limb	No	MRI	FNA	3.1	Above	P	Open	B	Perineural	NR	7	Y	Y (A sacral laminectomy)	Y	N
Li W[42]	33	M	Primary	Asymptomatic	Yes	MRI	Not performed	6.4	Below	P (Kraske)	Open	B	Tailgut cyst	NR	10	Y	Y (en block with coccyx)	N	NR
Naf F[43]	59	F	Primary	Coccydynia	Yes	MRI	Not performed	4.7	Below	A	Laparoscopic	B	Teratoma	NR	10	Y	N	NR	NR
Singh A, *et al.*[44]	63	M	Primary	CIBH	No	MRI	FNA	5.3	Below	NR	NR	Mal	Well-differentiated neuroendocrine tumor (Grade I)	NR	NR	Y	N	NR	N
Tokuyama S[45]	31	M	Primary	Pain in the thigh	Yes	CT	Not performed	3.4	Above	A	Laparoscopic	B	Tailgut cyst	132	6	Y	N	N	N
Borsuk DJ[46]	31	F	Recurrence	F/U post previous resection	No	MRI	Not performed	8	Below	A	Robotic	B	Epidermoid cyst	NR	1	Y	N	N	N
Schleinstein HP[47]	94	M	Primary	CIBH	No	CT	Not performed	10	Below	A	Open	B	Schwannoma	NR	7	Y	N	Y (Major bleeding during surgery required 4 units of RBC)	NR
Colombo F[48]	46	M	Recurrence	Second opinion regarding his recurrent sacral epidermoid tumor	No	MRI	Not performed	NR	Below	P	Open	Mal		NR	NR	N	N	N	Y (local recurrence diagnosed during surveillance at 6 moths)
Benjamin B[49]	29	F	Primary	Acute lower abdominal pain	Yes	MRI	Not performed	6	Below	A	Open	B	Castleman's	NR	NR	Y	Y (en block with rectosigmoid with end-to-end colorectal anastomosis)	N	N
Carvalho BJ[50]	41	M	Primary	Acute lower abdominal pain	No	CT	Not performed	3.9	Above	A	Laparoscopic	B	Schwannoma	260	1	Y	N	N	N
Turati L[51]	75	M	Primary	Coccydynia	No	MRI	Not performed	14.5	Below	A	Open	Mal	Leiomyosarcoma	NR	35	Y	Y (Total pelvic exenteration)	Y (chylous leak, conservatively treated with an alipidic diet)	Y (distant disease diagnosed during surveillance at 6 months)
Rakici SY, *et al.*[52]	55	F	Primary	Inability to walk that emerged in the last 1 year	No	MRI	Incision biopsy	1.25	Above	P	Open	B	Ganglioneuromas	NR	NR	Y	N	Y (unable to urinate post operative day 2)	Y (local disease recurrence diagnosed during surveillance in 1 year)
Tsarkov PV[53]	52	F	Primary	Acute lower abdominal pain	No	MRI	Not performed	6	Above	A	Laparoscopic	B	Tailgut cyst	120	3	Y	N	N	N
Zhao XR[54]	44	F	Primary	Coccydynia	No	CT	FNA	16	Above	A	Open	Mal	Cystic hamartoma	NR	NR	N (Adherent to surrounding structures, so unresectable)	N	N	NR
Maemoto R[55]	70	F	Primary	Follow up post rectal cancer surgical treatment	Yes	CT	Not performed	2.7	Above	A	Open	B	Epidermoid cyst.	NR	NR	Y	Y (en bloc with colonic involved mesentery preserving vasculature)	N	N
Santos AJ[56]	68	F	Primary	Lower abdominal pain	Yes	MRI	Not performed	6.3	Above	A	Open	B	Schwannoma	NR	3	Y	N	N	N
Bhadarge PS[57]	65	F	Primary	Lower abdominal pain	No	MRI	Not performed	NR	Above	A	Open	Mal	Primitive neuroectodermal tumors	NR	NR	Y	N	N	NR
Emohare O[58]	39	M	Primary	Chronic low-back pain	Yes	MRI	Not performed	8	Above	A	Laparoscopic	B	Schwannoma	249	4	Y	N	N	N
Brackzynski AK[59]	71	F	Primary	Severe bacterial meningitis	Yes	MRI	FNA	NR	Above	A	Open	B	Anterior sacral meningocele	NR	NR	Y	N	N	N
Rege S[60]	40	F	Primary	Multiple miscarriages	Yes	USS	Not performed	10	Below	P	Open	B	Fibro collagenous cyst wall lined by stratified squamous epithelium	NR	NR	Y	Y (Sacral laminectomy)	N	N
Lorusso D *et al*[61]	47	F	Primary	Anal fissure	Yes	MRI	FNA	7	Above	A	Open	Mal	Mucinous adenocarcinoma with osseous metaplasia	NR	NR	Y	N	N	NR
Andrade P[62]	60	F	Primary	Chronic low-back pain	Yes	MRI	FNA	4	Below	P	Open	Mal	Squamous cell carcinoma	NR	NR	Y	Y en block with coccyx	N	N
Total		51 (33F18M)	45 Primary, 6 Recurrent	42 symptomatic 9 asymptomatic	23 Incidental	30 MRI 15 CT 6 USS	31 Not performed. 16 FNA 3 Known from prior resections. 1 Incision Biopsy		26 Above 24 Below 1 NR	30 A 19 P 2 C 2 NR	32 Open 9 Laparoscopic 6 Robotic 2 Converted 1 TAMIS 1 NR	35 Benign 16 Malignancies				48 complete 3 incompletes	39 Uncombined 11 Combined 1 NR	7 Complications 5 NR	19NR 3 Recurrences

Key: F – Female, M – Male, NR – Not reported, Y – Yes, N – No, A – Anterior, P – Posterior, C – Combined, CT – Computed Tomography, MRI – Magnetic Resonance Imaging, TAMIS – Transanal Minimally Invasive Surgery, FNA – Fine Needle Aspiration, Mal – Malignant, B – Benign, F/U – Follow Up, USS: Ultrasound scan, CIBH: Change in bowel habit, MALT: Mucosa associated lymphoid tissue, LOS: Length of stay, OR: Operative time, SSI: Surgical site infection, IR: Interventional radiology, PE: Pulmonary embolism

Table 2. Summary of case series of retrorectal tumors.

Author	Number of CS included	Mean Age	F (*n*)	M (*n*)	Primary symptoms	Diagnosed incidentally	Asymptomatic	Diagnosis method	Ratio F:M in %	B (*n*)	Mal (*n*)	Mean tumor diameter on cross-sectional imaging (cm)	Underwent Surgery	Operative approach (A.P.C)	Operative method (open/lap/roboticconverted/both)	Mean OR (min)	LOS (days)	Complete resection? (Y/N/NR)	Combined resection? (Y/N/NR)	Overall Complication? (Y/N/NR)	Recurrence? (Y/N/NR)
L. Hopper, *et al.*[63]	69	50	39	30	Pain (abdominal, buttocks, flank, sacrococcygeal, rectal), bowel, neurological (sciatica, altered sensation) & urinary symptoms, and presence of a mass.	19	3	CT and MRI performed 28/69 (41%) and 34/69 (49%), respectively	57:43:00	40	29	NR	27	6.15.6	Open	NR	NR	NR	Y (6/69)	NR	NR
Yin J, *et al.*[64]	7	44	6	1	NR	NR	NR	MRI 100%	86:14:00	6	1	NR	All	5.0.2	Robotic	84	5.7	Y (1/7)	Y (2/7)	N	N
Leclerc A, *et al.*[65]	6	52	3	3	Constipation, dysuria, radicular or lower back pain	0	0	All had CT and MRI	50:50:00	6	0	NR	All	A	Open	240	6	Y (1/6)	NR	N	N
Isla A, *et al.*[66]	19	51	11	8	Lower back pain with lower limb radiation, paresthesia,	6	6	All had CT/MRI, or both	58:42:00	18	1	NR	All	6.9.4	Open	NR	NR	Y14/19	NR	1 lumbar spinal stenosis, 1 Impairment of external popliteal sciatic nerve	Y 3/19
Kilic A, *et al.*[67]	16	41	10	6	Pelvic, sacral, lower back and perianal pain, discomfort, changes in bowel habits, difficulty in defecation, and tenesmus	0	0	MRI 100%	63.5:37.5	13	3	NR	13	5.8.1	NR	NR	NR	NR	NR	Y (2/14)	Y (1/14)
Rompen IF, *et al.*[68]	5	47	5	0	NR	5	4	MRI 100%	100:0	0	5	NR	All	A	Robotic (da Vinci)	235	5.6	NR	Y (2/5)	NR	NR
Manabe T, *et al.*[69]	3	46	1	2	NR	3	3	CT and MRI were performed	33:67	3	0	NR	All	A	Laparoscopic	265	7.7	Y	N	N	N
Ramalingam K, *et al.*[70]	4	41	4	0	Back pain, constipation, CIBH	1	0	3MRI, 1 CT	100:0	4	0	NR	All	P (Kraske)	Open	NR	NR	NR	NR	N	NR
Wang B, *et al.*[71]	10	43	9	1	Pain (Abdominal, leg, lower back, perineal, sacral), CIBH,	0	0	7CT, 9 MRI, 4 USS	90:10:00	10	0	NR	6	2.4.0	5 Open, 1 Laparoscopic	NR	NR	NR	N	Y 3/10	NR
Oguz A, *et al.*[72]	17	36	12	5	Tenesmus, palpable perineal lump, lower urinary tract dysfunction, and rectal hemorrhage	2	0	15 MRI, 16 CT	71:29:00	16	1	NR	All	7.6.4	NR	NR	12.4	NR	NR	Y (4/17)	Y (1/17)
Carpelan-Holmström M, *et al.*[73]	52	43	40	12	Lower abdominal pain	30	30	MRI 100%	77:23:00	48	4	NR	All	7.44.1	NR	118	3	Y (51/52)	N	Y (11/52)	14/52 (1 mal)
Dwarkasing RS, *et al.*[74]	28	NA	22	6	Nonspecific pelvic pain, obstructed defecation	NR	NR	MRI 100%	79:21:00	23	5	NR	NR	NR	NR	NR	NR	NR	NR	NR	Y (2/28)
Maddah G *et al.*[75]	50	42	26	24	CIBH	NR	NR	NR	52:48:00	0	50	NR	All	22.34.30	NR	NR	NR	NR	NR	NR	NR
Buchs NC, *et al.*[76]	62	44	50	12	Pain, tenesmus, constipation, incontinence	13	13	45 MRI, 20 CT, 8 ERUS	81:19:00	49	13	NR	All	55.7.0	NR	NR	NR	Y (56/62)	NR	NR	9
Xu XM[77]	8	34	6	2	Sacrococcygeal pain, urinary retention, constipation	1	1	CT and MRI were performed for all	75:25:00	7	1	NR	All	1.6.1	Open	NR	NR	Y	NR	N	Y (1 Mal)
Huisman JF[78]	20	64	6	14	NR	NR	NR	NR	30:70	20	0	NR	All	P	Laparoscopic	NR	NR	Y	NR	NR	NR
Gould LE, *et al.*[79]	143	46	106	37	Pain, CIBH, rectal bleeding	NR	0	76 MRI	74:26:00	125	18	NR	107	24.64.15	78 (Open) 10 (Laparoscopic)	NR	NR	NR	Y (24)	Y (104/143)	Y (5)
Total	519		356	163		80	60			388	131		406 28NR	161.221.64	87 Open 6 NR 13 Lap 2 Robotic			126 complete	34 reported combined resections	125 reported complications	36 reported recurrences

CS: Case series, F: Female, M: Male, n: Number, Y: Yes, N: No, NR: Not reported, CIBH: Change in bowel habit, CT: Computed Tomography, MRI: Magnetic Resonance Imaging, USS: Ultrasound scan, ERUS: Endoscopic Rectal Ultrasound, B: Benign, Mal: Malignant, A: Anterior, P: Posterior, C: Combined, Lap: Laparoscopic, OR: Operative time, LOS: Length of stay

Osseous tumors comprised a mere 1.4% (n = 8) of all cases, characterized by a strikingly young mean age of 39.8 years and exclusive malignancy. Interestingly, chondrosarcomas constituted half of these malignant lesions. When compared to other tumor types, osseous tumors emerged as the youngest subgroup within the analyzed population.

This study identified a subset of 41 patients (7.2%) harboring inflammatory tumors, exclusively benign in nature. The spectrum encompassed abscesses (28 cases), diverticulitis (1 case), fibrosis (3 cases), cyst hydatid (3 cases), inflammatory cysts (2 cases), and unclassified cysts (3 cases). Notably, this group exhibited a higher mean age compared to other tumor categories, at 47.6 years.

Among the analyzed tumors, a noteworthy 16.5% (n = 94) were classified as miscellaneous, encompassing a diverse spectrum of lesions. Patients with miscellaneous tumors exhibited a higher mean age compared to other groups, at 51.3 years. A concerning trend emerged within this category: malignant miscellaneous tumors displayed significantly higher recurrence and complication rates compared to their benign counterparts (14% vs. 0% and 4.7% vs. 0.7%, respectively). Malignant composition – gastrointestinal stromal tumors took the lead among malignant miscellaneous tumors, with nine cases identified. Undifferentiated sarcomas followed closely behind, accounting for five cases. Further investigation into the specific subtypes and clinical characteristics of these aggressive miscellaneous tumors is warranted.

The operative approaches are listed in Table 5. The posterior approach was performed in 240 patients (42.1%), anterior approach in 191 patients (33.5%), and a combined approach in 66 patients (11.6%). The mean post-operative hospital stay was 8.4 days after an anterior approach was performed; 5.7 days after a posterior approach; and 6.6 days after a combined approach.

The overall post-operative complication rate was 20.2% and included surgical site infection, neurological complications (lower limb weakness, paresthesia), hematoma, post-operative bleeding, and change in bowel habit. The overall post-operative recurrence rate was 6.9%.

Thirty-one patients underwent a minimally invasive procedure [Table 5], an approach which had lower recurrence and complication rates when compared to open surgery (0 vs. 12 % and 10 vs. 32.1%, respectively) and was associated with shorter length of stay (4.2 vs. 13.1 days). The operative time was also shorter in minimally invasive surgery (242.8 vs. 327.5 min).

DISCUSSION

RRT management presents a unique dilemma due to their rarity and diverse histology. While open surgery dominates existing literature, minimally invasive techniques are gaining traction. However, robust data scarcity, limited to case reports and small series, impedes definitive conclusions on patient selection, perioperative complications, and oncological outcomes for minimally invasive approaches. Our systematic review reveals female predominance, benign etiology, and congenital classification as common features. Malignant lesions, more prevalent in males, exhibit higher complication and recurrence rates post-resection. Posterior approach emerges as the minimally invasive method of choice with minimal morbidity. Laparoscopy and robotic-assisted laparoscopy, though primarily used in benign cases, show promise with shorter hospital stays and potentially lower recurrence rates. Further research with robust data is crucial to solidify the role of minimally invasive techniques in retrorectal tumor management, particularly for malignant cases.

Radiological presentations of RRT range from asymptomatic and incidental findings to symptomatic manifestations, often associated with infectious or malignant processes. Barraqué *et al*. report 50% asymptomatic cases in a series of 53, highlighting the potential for subclinical presentation.[80] Symptomatic cases typically present with abdominal and lower back pain, possibly indicating underlying infection or malignancy.[81] Other reported symptoms include rectal fullness and sciatic pain.[82]

The diagnosis of RRTs presents a significant challenge due to their complex anatomical location and diverse presentations. A multimodal approach is essential for accurate diagnosis. While physical examination, often aided by proctoscopy, has limited sensitivity, with palpable lesions detectable in only approximately 35% of cases, it can be useful in excluding lower rectal involvement.[82] Proctoscopy and flexible sigmoidoscopy offer restricted visualization but can be employed to detect mucosal involvement. In cases with a draining sinus, fistulography may be utilized.[83] Magnetic resonance imaging has emerged as the gold standard imaging modality due to its exquisite soft-tissue resolution, enabling accurate assessment of tumor size, location, extent, and relationship to adjacent structures, thereby facilitating surgical planning.[84] Computed tomography (CT) scanning serves as a complementary tool, providing valuable information regarding RRT consistency (cystic vs. solid) and the degree of invasion into surrounding tissues.[81] In addition, CT scan features can offer clues to the underlying pathology, with homogeneous lesions suggesting benignity and heterogeneous lesions indicating a higher likelihood of malignancy.[80] In select cases, transrectal ultrasound can be beneficial, particularly in differentiating solid from cystic lesions and evaluating the tumor's proximity to the rectum.[85]

The definitive role of biopsy in diagnosing RRTs remains a subject of debate. While biopsy is generally avoided for cystic

Table 3: Classification of retro-rectal tumors in our series (570 cases in 68 papers)

Classification	Case	(% from overall total)
Congenital	324	56.8
Benign	284	49.8
Tailgut Cyst	145	25.4
Teratoma	33	5.8
Epidermoid Cyst	36	6.3
Dermoid Cyst	25	4.4
Developmental Cyst	1	
Duplication Cyst	38	6.7
Anterior Sacral Meningocele	4	
Indeterminant Cyst	2	
Malignant	40	7
Chordoma	20	3.5
Primitive neuroectodermal	2	
Tailgut cyst (malignant degeneration)	6	
Malignant Teratoma	3	
Germ Cell Tumor	1	
Developmental Cyst (malignant transformation)	5	
Dermoid Cyst (malignant degeneration)	2	
Duplication Cyst (malignant degeneration)	1	
Neurogenic	103	18.1
Benign	90	15.8
Neurofibroma	5	
Ganglioneuroma	5	
Paraganglioma	2	
Schwannoma	54	9.5
Neuroblastoma	3	
Perineural cyst (Tarlov)	21	3.7
Malignant	13	2.3
Neurofibrosarcoma	2	
Neuroendocrine Carcinoma	3	
Neuroblastoma	1	
Ependymoma	2	
Ganglioneuroblastoma	1	
Malignant peripheral nerve sheath tumor	4	
Osseus	8	1.4
Benign	0	0
Malignant	8	1.4
Ewing Tumor	2	
Chondrosarcoma	4	
Synovial Sarcoma	1	
Malignant Giant Cell Tumor	1	
Inflammatory	41	7.2
Benign	41	7.2
Abscess	28	4.9
Fibrosis	3	
Cyst hydatid	3	
Inflammatory Cyst	2	
Diverticulitis	1	
Fistula	1	
Unknown Cyst	3	
Malignant	0	0
Miscellaneous	94	16.5
Benign	51	8.9
Leiomyoma	6	
Fibroma	4	
Myelomeningocele	2	
Myelolipoma	3	
Solitary fibrous tumor (SFT)	4	
Hemangiopericytoma	1	
Oleogranuloma	1	
Lipoma	1	
Angiomyxoma	4	
Neuroenteric Cyst	1	
Angiomyofibroblastoma	1	
Fibrolipoma	1	
Peritoneal serous cystadenoma	1	
Desmoid tumor	1	
Lymphoid hyperplasia (Castleman's disease)	1	
Lipoblastoma	1	
Bronchogenic cyst	1	
Angiomyolipoma	1	
Sclerosing Epithelioid Fibrosarcoma	1	
Hematoma	1	
Angiofibroma	1	
Lymphocele	1	
Retention cyst of anal gland	4	
Vascular	1	
Normal tissue	1	
Unknown	6	
Malignant	43	7.5
Gastrointestinal stromal tumor	9	
Lymphoma	4	
Leiomyosarcoma	4	
Liposarcoma	3	
Undifferentiated sarcoma	7	
Rhabdomyosarcoma	1	
Adenocarcinoma	5	
Mucinous Tumor	1	
Squamous cell carcinoma	1	
Spindle cell tumor	1	
Desmoid type fibromatosis	1	
Plasmacytoma	2	
Intra-osseus ganglion cyst	1	
Unknown	3	
Total	570	100

lesions due to their high likelihood of benignity and potential for infection, image-guided tissue acquisition remains crucial for definitive diagnosis and treatment planning.[1,83] This is particularly true for specific tumor types such as Ewing sarcoma, osteogenic sarcoma, neurofibrosarcomas, and desmoid tumors, which benefit from neoadjuvant therapy.[86,87]

Table 4: Demographic findings and clinical outcomes of retrorectal tumors included in the review.

Classification	Case (%)	Mean age (range)	Complication (%)	Recurrence (%)
Congenital	324 (56.8)	44.8 (21-74)	2	6.5
Benign	284 (49.8)	43.1 (21-71)	2.1	2.4
Malignant	40 (7.0)	51.4 (25-74)	1.5	24.2
Neurogenic	103 (18.1)	48 (22-94)	6.4	3.6
Benign	90 (15.8)	48 (22-94)	7.1	3.2
Malignant	13 (2.3)	47	0	7.1
Osseus	8 (1.4)	39.8 (16-58)	25	NR
Benign	0	-	-	-
Malignant	8 (1.4)	39.8 (16-58)	25	NR
Inflammatory	41 (7.2)	47.6	2.4	7.3
Benign	41 (7.2)	47.6	2.4	7.3
Malignant	0	-	-	-
Miscellaneous	94 (16.5)	51.3 (19-76)	1.6	3.2
Benign	51 (8.9)	49.3 (19-76)	0.7	0
Malignant	43 (7.5)	53.6 (35-75)	4.7	14
Overall benign	466 (81.8)			
Overall malignant	104 (18.2)			
Total	570 (100)			

Table 5: Operative outcomes of retrorectal tumors according to surgical approach.

Approach	Case #	Mean OR (min)*	Mean LOS (days)*				
Anterior	191	293.2	8.4				
Posterior	240	100	5.7				
Combined	66	160	6.6				
Operation method	**Case #**	**Mean OR (min)***	**Mean LOS (days)***	**Complete resection (%)**	**Combined resection (%)**	**Complication rate (%)**	**Recurrence rate (%)**
Open	119	327.5	13.1	82.8	9.5	32.1	12
Minimally invasive	31	242.8	4.2	85.1	20	10	0

*Many cases were "Not Reported" – the mean values only consider the cases with reported values, LOS: Length of stay, OR: Operative time, #means number i.e., Number of cases

Precise surgical planning and meticulous execution are paramount, as highlighted by Balci *et al.* The extent of tumor invasion dictates resection levels, with abdominoperineal or sacral resection necessary in severe cases.[80] The efficacy of neoadjuvant therapy remains under debate, with conflicting results concerning its impact on surgical difficulty and overall outcome.[88] In specific cases, adjuvant radiotherapy or palliative chemotherapy can be considered.[80]

The limitations of this study were that many of the articles were confined to small case series and case studies which also meant that adequately comparing certain criteria such as surgical outcomes based on approach, for instance, was difficult owing to the lack of this information in many of the reviewed articles.

CONCLUSION

RRTs are rare tumors and most commonly congenital in nature with a predominance among the female sex. It is recommended that surgical management with R0 resection is crucial in the management of these tumors, and the minimal access surgery approach appears to be a safer option in appropriate case selection, having an association with shorter length of stay, lower recurrence rates, and shorter operative time.

Author's contributions

Jeffrey Otote (J.O.), Valentin Butnari (V.B.), Praveen Ravichandran (P.R.), Ahmer Mansuri (A.M.), Mehnaz Ahmed (M.A.), Olivia Pestrin (O.P.), Nirooshun Rajendran (N.R.), Sandeep Kaul (S.K.). Conceptualization, S.K. and V.B.; methodology, J.O.; software, V.B.; validation, A.M., N.R. and S.K; formal analysis, V.B. and J.O.; investigation, J.O. and P.R.; resources, P.R. , O.P. and M.A. .; data curation, J.O., V.B., and P.R.; writing—original draft preparation, J.O.; writing—review and editing, V.B. , J.O. P.R; visualization, J.O., V.B., P.R, M.A. and O.P.; supervision, A.M. , N.R. and S.K.; project administration, V.B., A.M. , N.R. and S.K. All authors have read and agreed to the pub-lished version of the manuscript.

Ethical approval

The Institutional Review Board approval is not required.

Declaration of patient consent

Patient's consent is not required as there are no patients in this study.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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How to cite this article: Otote J, Butnari V, Ravichandran P, Mansuri A, Ahmed M, Pestrin O, *et al*. Presacral tumors: A systematic review of literature. J Clin Imaging Sci. 2024;14:17. doi: 10.25259/JCIS_27_2024