



Excision of Retrorectal Tumors: Comparing Outcome Regarding Surgical Approach

Retrorektal Tümörlerin Eksizyonu: Cerrahi Yaklaşımına Göre Sonuçların Karşılaştırılması

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ABSTRACT

Aim: Tumors of the retrorectal space are very rare, and the clinical characteristics of these tumors, such as their complex anatomy, origin, and surgical techniques used in the treatment are yet to be elucidated. Although several surgical approaches have been identified, the question remains as to which approach provides better results.

Method: A total of 29 patients who underwent surgical excision of retrorectal tumors at Department of General Surgery, İstanbul Cerrahpaşa Faculty of Medicine, were retrospectively evaluated. All characteristics of patients were assessed from their medical files, and patients were informed about the study by phone.

Results: Among the 29 patients, four were male and 25 were female. The mean age was 44.07±16.15 years, and the mean follow-up duration was 7.5±4.4 years. Twenty patients underwent surgery via posterior (perineal) approach, seven via anterior (transabdominal) approach, and two via combined (abdominoperineal) approach. There were three deaths and six tumor recurrences during the follow-up period. The length of hospital stay was significantly higher with the combined approach. Coccygectomy was performed in 11 patients, of which only one had a recurrence, while the remaining five recurrences were in patients without coccygectomy.

Conclusion: The most advantageous surgical approach to retrorectal tumors remains unclear. Various factors (resection success, coccygectomy, tumor features, and type) are thought to affect the outcome. Further studies and structured, systematic reviews may be necessary to identify the role of each factor in the surgical outcome of retrorectal tumors.

Keywords: Retrorectal tumors, transabdominal approach, coccygectomy

ÖZ

Amaç: Retrorektal boşluğun tümörleri çok nadirdir ve bu tümörlerin kompleks anatomileri, kökenleri ve tedavide kullanılan cerrahi teknikler gibi klinik özellikleri henüz açıklığa kavuşturulmamıştır. Cerrahiye yönelik birkaç yaklaşım tanımlanmasına rağmen, hangi yaklaşımın daha iyi sonuçlar sağladığı sorusu halen gündemdedir.

Yöntem: İstanbul Cerrahpaşa Tıp Fakültesi, Genel Cerrahi Anabilim Dalı'nda retrorektal tümörlerin cerrahi eksizyonu yapılan toplam 29 hasta retrospektif olarak incelendi. Hastaların tüm özellikleri tıbbi dosyalarından değerlendirildi ve hastalar telefonla çalışma hakkında bilgilendirildi.

Bulgular: Yirmi dokuz hastanın 4'ü erkek, 25'i kadın; yaş ortalaması 44,07±16,15, ortalama takip süresi 7,5±4,4 idi. Yirmi hasta posterior (perineal), 7'si anterior (transabdominal) ve 2'si kombine (abdominoperineal) yaklaşımla opere edildi. Takip süresi boyunca 3 ölüm ve 6 tümör nüksü saptandı. Hastanede kalış süresi kombine yaklaşımla opere edilen hastalarda anlamlı derecede yüksekti. Koksektomi yapılan 11 hastanın birinde nüks izlenirken, diğer 5 nüks olgusu koksektomi yapılmayan kişilerdendi.

Sonuç: Retrorektal tümörlere en avantajlı cerrahi yaklaşım hala belirsizliğini korumaktadır. Sonuç olarak, çeşitli faktörlerin (rezeksiyon başarısı, koksektomi, tümör özellikleri ve tipi) sonuçları etkilediği düşünülmektedir. Her faktörün retrorektal tümörlerin cerrahi sonuçlarındaki rolünü belirlemek için daha ileri çalışmalar ve sistematik derleme çalışmaları gerekli olabilir.

Anahtar Kelimeler: Retrorektal tümörler, transabdominal yaklaşım, koksektomi



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Introduction

The retrorectal space (presacral space) is defined as the gap between the rectum and the sacrum/coccyx. Tumors of the retrorectal space are very rare.¹ In adults, the annual incidence has been reported as 1/40000-60000 with being more frequent in females.² Even though benign-malignant distinctions of these tumors are unclear, many case series in literature have concluded that the majority of tumors show benign features.³ Retrorectal tumors present with nonspecific symptoms, and the majority of cases are initially misdiagnosed despite advanced imaging techniques.⁴ However, after the clinician suspects a retrorectal tumor, magnetic resonance imaging (MRI) results can be instrumental in the differential diagnosis, may reveal important characteristics of the tumor, and determine treatment approach.⁵ Computerized tomography (CT) may also be used to evaluate retrorectal lesions and involvement of surrounding tissue; however, better resolution of soft tissue in MRI may place it a step ahead of CT.⁴ The use of biopsy in retrorectal lesions is disputed due to concerns about biopsy-related complications and the almost-absolute requirement of total resection regardless of the biopsy result.⁶ A consensus on this matter is that definitive diagnosis should be based on the pathological evaluation of the lesion after surgical resection. Thus, surgery and surgical approach can be considered to be the most crucial aspects of the management of retrorectal tumors. When a patient is diagnosed with a retrorectal tumor, the standard approach for treatment is surgical resection.⁷ Various surgical approaches for retrorectal tumors exist, including anterior (trans-abdominal), posterior (perineal), and combined abdominoperineal approaches. Due to the rareness of these tumors, large case series of retrorectal tumors have not been published.⁸ Thus, the clinical characteristics of these tumors, such as their complex anatomy, origin, and surgical techniques used in the treatment are yet to be elucidated. The objective of the study was to determine the outcome and clinical features of retrorectal tumors based on surgery type.

Materials and Methods

Our study was a retrospective study conducted in İstanbul Cerrahpaşa Faculty of Medicine, Department of General Surgery. The study group consisted of 29 patients who underwent surgery for retrorectal tumors between 2001 and 2015. Two patients who refused to participate in the study were excluded from the study. Our pathology department made the diagnoses via histopathological examination and immunohistochemistry analysis of tissue samples obtained during surgery. All patients included in the study had

undergone tumor resection. Surgeries were performed using the anterior, posterior, or combined transabdominal approach. We used the open surgical technique with complete resection of tumors. The appropriate approach was determined after the evaluation of each patient by the multidisciplinary team following a review of tumor characteristics. Ethical approval was obtained from İstanbul Cerrahpaşa Faculty of Medicine, Clinical Research Ethics Committee. The study was performed according to the principles put forth by the Helsinki Declaration and Good Clinical Practice Guidelines. The informed consent of patients was taken by phone or in follow-up examinations. Demographics, complaints, radiological results, pathologic reports, surgical approach, length of hospital stay (LoS), follow-up duration, and presence of recurrence and deaths were obtained from the medical records of patients.

Statistical Analysis

All analyses were performed by using SPSS v20 (IBM, Armonk, NY, USA). The Shapiro-Wilk test was used for the normality test of variables. Continuous variables were given as mean \pm standard deviation for normally distributed data and median (interquartile range=IQR) for non-normally distributed data. Survival at up to 15 years was reported using the Kaplan-Meier cumulative survival plots. Comparisons between groups were made with the Mann-Whitney U and Kruskal Wallis tests. Analysis of categorical variables was made with the chi-square test. Fisher's exact test was used where sample sizes were small. $P \leq 0.05$ values were accepted as statistically significant.

Results

Four of the patients (13.8%) were male, and 25 (86.2%) were female. Their ages ranged from 21 to 77 years, with a mean of 44.07 ± 16.15 years. The mean follow-up period was 7.5 ± 4.4 years. Two cases were diagnosed incidentally, while 13 patients admitted with rectal pain, four with low back pain, two with femoral pain, three with inguinal pain, three with abdominal pain, one with constipation, one with gluteal pain, and one with perianal fistula. Among the study group, seven patients (24.1%) underwent surgery due to tailgut cysts and six (20.7%) due to epidermoid cysts. Eighteen of the cases (62.1%) were cystic, 11 (37.9%) had solid (or heterogeneous) pathology. One tumor was determined to be a malignant adenocarcinoma based on the tailgut cyst. Recurrence occurred in six patients (20.6%). Fourteen patients (48.3%) had rectal pain, and seven (24.1%) had waist-to-groin pain. The majority of patients (72.4%) did not undergo biopsy. Three patients died during the follow-up period; two of these cases died due to causes other than retrorectal tumors. The characteristics of the

patients are given in Table 1. In terms of surgical approach, 20 patients (68.9%) underwent posterior, seven (24.1%) underwent anterior, and two (6.9%) underwent combined abdominoperineal surgery. The posterior approach was used in patients in whom the upper border of the tumor reached the S2 level, but was palpable and mobile during a rectal examination. The LoS of patients who were operated with the combined method was significantly higher than patients who underwent surgery with other approaches. However, no statistically significant difference was found between the anterior and posterior methods in terms of LoS [anterior, median=9 days (IQR=6-12), posterior, median=6 days (IQR=4-9), $p>0.05$]. No significant differences were found between surgical procedures in terms of follow-up period [anterior, median=63.84 months (IQR=39.23-114), posterior, median=86.21 months (IQR=49.95-130.74), combined median=50.78 months (IQR=24.90-76.65), $p>0.05$].

Patients with cystic pathology had higher mean survival than those with solid pathologies ($p=0.036$). The 5 year survival rate of patients with solid pathology was 77.9%. We were able to obtain the radiological results of 27 cases in our study. Radiologically determined diameters of the tumors were compared with diameters determined after pathology. Median radiological diameter was 6.3 cm (IQR=5-12.75), while median pathological diameter was 7.2 cm (IQR=3.5-12.5); the difference was not significant ($p>0.05$). There were no surgical complications in any of the patients. Six patients had a recurrence. One patient (a 68-year-old female) died 60 months after the surgery. She had a diagnosis of squamous cell cancer with perineural invasion. Additionally, two patients died during the follow-up period due to causes unrelated to retrorectal tumor, one at 63 months, and the other at 128 months. The characteristics of patients are summarized in Table 1.

Discussion

Although many studies have presented findings for retrorectal tumors and their treatment, the majority of these studies have a low number of cases. Thus, determining the most appropriate surgical approach to retrorectal tumors is still a controversial matter. We aimed to evaluate and compare the surgical methods utilized for the treatment of retrorectal tumors at our center. Studies have shown that retrorectal tumors are mostly benign. However, it is important to keep in mind that cystic lesions with solid walls and heterogeneous components have a higher chance of malignancy.^{9,10,11} In our study, 79.3% of the cases were benign. It was found that one of the malignant cases occurred based on tailgut cysts. The most frequent symptom in this study was a pain in the rectal region and lower back. Among those with benign masses, 82.6% had pain, while all (100%) patients with malignant mass had pain. However,

the difference was not statistically significant, presumably due to the low number of malignant cases. In previous case series studies, 86-88% of patients with malignancies were reported to have pain, while in benign cases, this value was 22-39%, which is much lower than our result.⁴ In a review, the most common symptom was also reported as pain in the rectal area, which was often associated with infection and malignancy.¹² Surgical resection is the best therapeutic choice for retrorectal tumors, even in asymptomatic patients. Many lesions are considered to contain malignant elements; thus, they may have the potential for growth or transformation to malignancy, or they may cause complications such as infection.¹³ Depending on the pathology, radiological results, and the location of the retrorectal lesion, three different surgical approaches can be utilized: posterior, anterior, and combined. In general, lesions above S4 are operated with the anterior or combined approach, and lesions below S4 are operated through the posterior approach. However, higher lesions that are palpable and mobile during the rectal examination may also be operated with the posterior approach,³ which provides relatively better access to the caudal component of the mass and also ensures excellent results with minimal morbidity. Furthermore, in a study including 1708 patients, Baek et al.¹² reported that the posterior approach was the preferred method of retrorectal tumor surgery and provided the lowest morbidity rate. Therefore, at our center, the posterior approach was chosen in the majority of cases (68.9%), as long as the mass was palpable and mobile during the rectal examination.

Resection of the coccyx during surgery is also a controversial matter. Removal of the coccyx improves surgical exposure of the tumor site, and some authors claim that the coccyx may contain tissue remnants that lead to cystic formations (and cause recurrence).^{14,15} However, whether the coccyx truly causes recurrence remains unknown, and studies have shown that recurrence mostly occurs in malignant tumors and cases where total resection could not be performed.^{8,16} The majority of recent studies suggest that the coccyx should not be resected unless the lesion is directly attached to the coccyx.^{16,17} In the current study, coccygectomy was performed on 11 (37.9%) patients, and only one (9.1%) of these patients developed recurrence. A total of five recurrences were found in the remaining 18 patients (27.7%). Our results are in agreement with contemporary literature, which reports that the rate of local recurrence is around 25-56% in cases with incomplete resection of the tumor and without coccygectomy.^{15,18} Although this finding may encourage the idea that coccygectomy should be performed in retrorectal tumors, further studies in which patient groups are adjusted for various factors (surgical approach, tumor features, age, comorbidities, resection success) are needed to conclude.

Table 1. The characteristics of patients

No	Gender	Age	Pre-operative period			Operation			Post-operative period				
			Complaints	MRI	CT	Bx	Approach	RoC	Area (cm ²)	Pathology	LoS	Rec.	D
1	F	77	Cons.	+, c	-		P	-	10.8	Tailgut cyst	1	-	-
2	F	68	RP	+	-	+	P	-	3.5	Squamous cell ca	16	+	+
3	F	66	Low back pain	+	-	+	P	+	99	Schwannoma	6	-	-
4	F	65	RP	+, c	+, c	-	P	-	132	Dermoid cyst	6	+	-
5	F	61	Rectal pain	-	+, c, 2	-	A	+	72	Tailgut cyst	16	-	-
6	F	61	Femoral pain	-	+	-	A	-	99	Solitary fibrous tumor	12	+	+
7	F	59	Rectal pain	+, 2	-	-	P	-	5.5	Retrorectal cyst	10	-	-
8	F	54	Incidental	+, c	-	-	P	-	123.5	Epidermoid cyst	24	-	-
9	F	50	Low back pain	+, c	+	+	A	-	32.4	Epidermoid cyst	6	-	-
10	M	49	Rectal pain	+	-	-	P	+	16	GIST	2	-	+
11	M	48	Incidental	+	-	-	A	-	117	Schwannoma	8	-	-
12	F	43	Inguinal pain	-		-	A	-	63	Mature cystic teratoma	6	-	-
13	F	41	Rectal pain	+, c	+, c	-	P	+	33	Tailgut cyst	3	-	-
14	F	37	Abdominal pain	+		-	P	-	182	GIST	4	+	-
15	M	36	Abdominal pain	+	+	-	A	-	48.7	Schwannoma	9	-	-
16	F	35	Inguinal pain	-	+, c	-	P	-	130	Tailgut cyst	4	-	-
17	F	35	Rectal pain	+, c	-	-	P	-	8.7	Hindgut malformation	6	-	-
18	F	34	Rectal pain	NA	-	-	P	-	11.3	Epidermoid cyst	11	-	-
19	M	32	Low back pain	-	-	+	P	+	4.5	Epidermoid cyst	4	-	-
20	F	31	Rectal pain	+	-	-	P	+	5.1	Mature cystic teratoma	10	+	-
21	F	30	Low back pain	+, c	-	-	P	+	19.3	Epidermoid cyst	6	-	-
22	F	26	Inguinal pain	+, c	-	-	P	+	4.5	Epidermoid cyst	3	-	-
23	F	24	Rectal pain	+, c	+	-	P	+	13	Tailgut cyst	2	-	-
24	F	23	Rectal pain	+, c	-	+	P	+	13	Cystic hamartoma	7	-	-
25	F	22	Perianal fistula	+	-	+	cap	-	40	Hindgut malformation	34	+	-
26	F	62	Femoral pain	NA	-	-	P	-	12	Tailgut cyst	6	-	-
27	F	31	Rectal pain	-	-	+	A	-	6	Neuroectodermal tumor	10	-	-
28	F	57	Gluteal pain	-	-	+	P	-	262	Adenocarcinoma	7	-	-
29	F	21	Rectal pain	+, c	-	-	cap	+	46.8	Tailgut cyst	26	-	-

NA: Not applicable, M: Male, F: Female, MRI: Magnetic resonance imaging, CT: Computed tomography, LoS: Length of hospital stay, GIST: Gastrointestinal stromal tumors, RP: Rectal pain, Cons: Constipation, RoC: Resection of coccyx, Rec: Recurrence, Cap: Combined approach

In the literature, the recurrence rate of malignant tumors is reported to be around 30-75%; this rate is significantly lower (0-11.1%) in benign lesions.^{5,19,20} In the current study, recurrence was observed in 50% of malignant cases and 13% of benign cases [with a total of six (20.6%) recurrences]. In studies where complete resections were reported, the rates of recurrence were reported as 0% at 10-year follow-up in one study⁶, and 6.2% at 5-year follow-up in another.²

A study by Hjermsstad and Helwig²¹ reported a recurrence rate of 11.1% during a 11-year follow-up of patients with tailgut cysts, and Gao et al.⁸ reported 17.9% recurrence in their series of patients with presacral lesions, which are closer to our findings. In addition to the malignancy of the initial tumor, complete excision of the lesion and any involved entity seems to be the most important factor in the prevention of recurrence.

The limitations of this study include its retrospective design that may introduce assessment bias. However, the design was unavoidable, given the rarity of these tumors. Another limitation is the fact that radiological results of two patients could not be obtained, which may reduce the feasibility of comparisons. The long follow-up duration of patients and the relatively high number of patients (although from a single center) are strengths of the study.

Conclusion

Retrorectal tumors are mostly benign. However, total resection is the only viable treatment course; thus, evaluation of surgical outcome in terms of approach is an important topic. The posterior approach was the most preferred surgical method with relatively shorter LoS. The rarity of retrorectal tumors leads to a limited number of cases, especially in single-centered studies. Therefore, multicenter and prospectively designed studies may be useful for a better understanding of the characteristics of retrorectal tumors and may contribute to a better surgical approach.

Ethics

Ethics Committee Approval: Ethical approval was obtained from İstanbul Cerrahpaşa Faculty of Medicine, Clinical Research Ethics Committee. The study was performed according to the principles put forth by the Helsinki Declaration and Good Clinical Practice Guidelines.

Informed Consent: Retrospective study.

Peer-review: External and internal peer-reviewed.

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