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Original Article

The Rarely Diagnosed Retrorectal Tumor: Experience of a Single Center

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ABSTRACT

Objective: Retrorectal tumors (presacral, precoccyxgeal) are rare, and remain a significant diagnostic and therapeutic challenge. The aim of this study was to describe the surgical experience of 1 hospital with retrorectal tumors.

Methods: Twelve patients admitted to the Dr. Lutfi Kirdar Training and Research Hospital/ Istanbul Department of General Surgery between January 2012 and December 2016 were included in our study. Medical records, radiology results, pathology reports, and surgical techniques were analyzed retrospectively.

Results: Of the 12 patients evaluated, 10 were female and 2 were male. The mean age was 45 years (range: 31-65 years). Eight patients had masses of a congenital origin, 3 patients had masses of neurogenic origin, and 1 had an angiomyxomatous tumor. Both an anterior and posterior surgical approach were used.

Conclusion: Complete removal of the tumor and preservation of neurological function remain major aspects of successful treatment. To achieve the best results, a multidisciplinary approach and correct planning are important.

INTRODUCTION

Retrorectal tumors (presacral, precoccyxgeal) are rare, and remain a diagnostic and therapeutic challenge.[1] The retrorectal space, also referred to as the presacral space, is surrounded posteriorly by the presacral fascia, anteriorly by the fascia propria of the rectum, and laterally by the iliac vessels and ureters.[2] Anatomically, the space is divided by the rectosacral fascia into inferior and superior portions. The floor of the retrorectal space is formed by the fusion of the presacral parietal fascia and the rectal visceral fascia, and lies above the levator ani muscle at the level of the anorectal junction. Retrorectal tumors are classified as congenital, neurogenic, inflammatory, osseus, or miscellaneous, according to an extensive literature search[3–7] (Table 1).

The aim of this study was to describe the surgical experience of 1 institution with retrorectal masses. Cases treated between January 2012 and December 2016 were retrospectively analyzed.

MATERIAL AND METHODS

Twelve patients admitted to the Kartal Dr. Lutfi Kirdar Training and Research Hospital/Istanbul Department of General Surgery between January 2012 and December 2016 were included in the study. Medical records, radiology results, pathology reports, and surgical technique were analyzed retrospectively.

RESULTS

Of the 12 patients evaluated, 10 were female, and 2 were male. The mean age was 45 years (range: 31–65 years). Eight patients had masses of a congenital origin (1 teratoma, 2 infected cysts, 1 keratinous cyst, 2 epidermal cysts, 1 dermoid cyst, 1 tailgut cyst), 3 patients had masses of neurogenic origin (1 benign spindle cell tumor, 2 myxopapillary ependimomas), and 1 patient had an angiomyxomatous tumor (Table 2). One of the myxopapillary ependimomas metastasized to the right kidney in the

Table 1. Types of retrorectal tumor

Congenital (55–65%)	Neurogenic (10–12%)	Osseous (5–11%)	Inflammatory (5%)	Miscellaneous (12–16%)
Developmental cyst	Ganglioneuroma	Chondromyxosarcoma	Foreign body granuloma	Leiomyoma/leiomyosarcoma
• Dermoid		Giant cell tumor	Infectious granuloma	Hemangioma
• Epidermoid				Carcinoid tumor
Tailgut cyst				Plasma cell myeloma
Teratoma				Hemangioendothelial sarcoma
Teratocarcinoma				Endothelioma
Chordoma				Pelvic ectopic kidney
Adrenal rest tumor				Extra-abdominal desmoid
Anterior sacral meningocele				
Rectal duplication				

Table 2. Pathological type of retrorectal tumor in 12 patients

	Pathology	Number
	Teratoma	1
	Infected cyst	2
Congenital (8 patients)	Keratineous cyst	1
	Epidermal cyst	2
	Tailgut cyst	1
	Dermoid cyst	1
Neurogenic (3 patients)	Spindle cell tumor	1
	Myxopapillary ependymoma	2
Other (1 patient)	Angiomyxoma	1
Total		12

fourth year of follow-up and was resected with a right lumbar incision.

Most of the patients were complaining of defecation problems, a palpable presacral mass, and vague symptoms diagnosed incidentally. History, physical examination, and laboratory data revealed no abnormality. The majority of the cases were diagnosed with pelvic magnetic resonance imaging (MRI) (Fig. 1a, b).

The largest of these retrorectal masses was 18x13x5.5 cm in size and was a deeply located, aggressive angiomyxoma. Two previously described surgical techniques were used.[8] For the anterior surgical approach, the patient was placed in the supine position and the internal iliac artery and vein were dissected on both sides. The external and middle sacral arteries and veins were ligated. For the posterior surgical approach, the patient was positioned in the jack-knife position with cranial and caudal flexion of the operation table, and an incision was made from the sacrococcygeal area inferiorly to the midline. Two patients were operated on using the anterior approach, and 10 patients were operated on using the posterior approach. There were no significant postoperative complications; 2 patients showed signs of a surgical site infection, which was treated successfully. There was no mortality. The median hospitalization time was 2 days (range: 1–4 days).

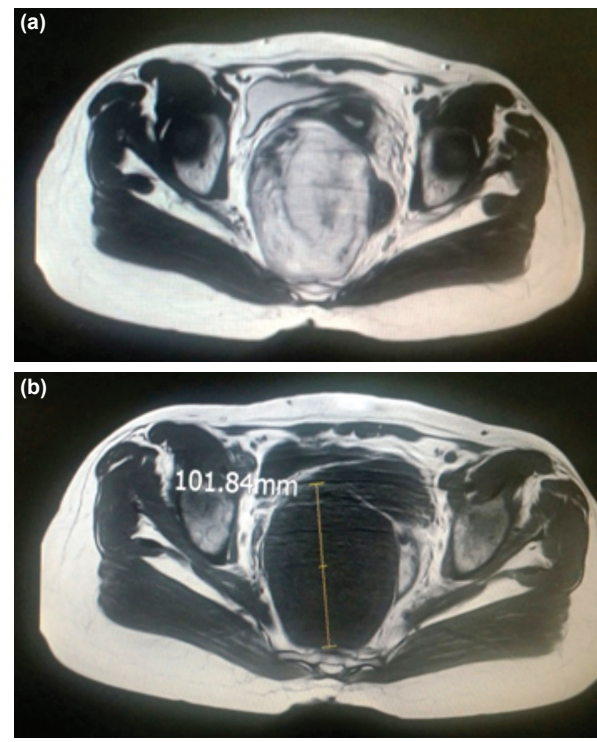

Figure 1. (a, b) Pelvic magnetic resonance images of retrorectal tumors.

The majority of the cases were followed up for approximately 20 months (range: 9–58 months). Only 1 patient, whose pathology was myxopapillary ependimoma, was reoperated on 3 years later due to a local recurrence.

DISCUSSION

The rarity of retrorectal tumors has contributed to the difficulties associated with successful diagnosis and treatment. In this series, 12 patients were treated within a short period of time. We believe being a referral center led to a large number of these cases.

In general, the physical examination plays a major role in the diagnosis of these masses. The majority of our patients revealed physical signs. We were able to palpate some of the masses with a digital rectal examination. Some patients were complaining of defecation problems and rectal fullness. We believe retrorectal masses should be taken into consideration in the differential diagnosis of mechanical intestinal obstructions.

Although some researchers believe that plain abdominal X-rays may be utilized for diagnosis, they did not reveal any sign of the masses in our series. Some authors have reported using computed tomography (CT) scans to detect these tumors. They reported that CT scans could differentiate cystic and solid components. MRI is able to illuminate any kind of invasion to adjacent tissues and nerve involvement.[9,10] We used pelvic MRI studies for the diagnosis in all cases. We believe that MRI is the best modality for diagnosis. Our preoperative strategy was planned according to MRI studies and the individual patient.

We did not perform any biopsies and this was consistent with the current literature. Biopsies can cause fatal septic complications and should be performed only when the lesion appears to be unresectable and a tissue diagnosis is required to guide adjuvant therapy.[11,12]

Complete resection of the tumor, preservation of neurological and sphincteric function, and a low mortality rate remain major factors for successful surgery. Surgical treatment is based on the size of the tumor, the caudal extent, involvement of visceral structures and the sacrum, and features of malignancy on imaging studies. For masses above the S3 level, an abdominal approach is recommended. In our series, only 2 cases were above the S3 level and our surgical choice was transabdominal resection. Tumors below the S3 level can be resected posteriorly, and result in faster recovery with less pain.[13] We applied this technique in 10 of our patients. Visceral or sacral involvement requires an abdominal or combined approach irrespective of the tumor level. Alternative operative methods include transrectal[14] and transvaginal[15] approaches. Some authors have preferred to use laparoscopy[16,17] or transanal endoscopic microsurgery[18] for retrorectal lesions, but these were small series. Future studies are required.

There are some reports related to surgical complications, such as wound infections and hemorrhage.[4] We experienced 2 surgical site infections with no instance of mortality. To prevent major complications, we believe that this type of surgery should be performed by experienced surgeons at referral centers.

Conclusion

Complete removal of the tumor and preservation of neurological function remain the major aspects of successful treatment. A multidisciplinary approach and careful planning should be employed to achieve the best results.

Ethics Committee Approval

Approval has been obtained from University of Kartal Dr. Lütfi Kırdar Training and Research Hospital Ethics Committee.

Informed Consent

The study design was retrospective observational study.

Peer-review

Internally peer-reviewed.

Authorship Contributions

Concept: Ö.A.; Design: Ö.A., S.K.; Data Collection: Ö.A., S.K.; Analysis and/or interpretation: Ö.A., S.K., Y.E.A; Literature search: Ö.A., S.K.; Writing: Ö.A.; Critical review: Ö.A., S.K.

Conflict of Interest

None declared.

REFERENCES

1. Heidenreich A, Decoud J, Lembrande RO, Rapela RO.Retrorectal Tumors and Tumor-Like Lesions: Diagnosis and Treatment. Dig Surg 1990;7:201–6. [CrossRef]
2. García-Armengol J, García-Botello S, Martinez-Soriano F, Roig JV, Lledó S. Review of the anatomic concepts in relation to the retrorectal space and endopelvic fascia: Waldeyer's fascia and the rectosacral fascia. Colorectal Dis 2008;10:298–302. [CrossRef]
3. Uhlig BE, Johnson RL. Presacral tumors and cysts in adults. Dis Colon Rectum 1975;18:581–9. [CrossRef]
4. Jao SW, Beart RW Jr, Spencer RJ, Reiman HM, Ilstrup DM. Retrorectal tumors. Mayo Clinic experience, 1960-1979. Dis Colon Rectum 1985;28:644–52. [CrossRef]
5. Glasgow SC, Birnbaum EH, Lowney JK, Fleshman JW, Kodner IJ, Mutch DG, et al. Retrorectal tumors: a diagnostic and therapeutic challenge. Dis Colon Rectum 2005;48:1581–7. [CrossRef]
6. Stewart RJ, Humphreys WG, Parks TG. The presentation and management of presacral tumours. Br J Surg 1986;73:153–5. [CrossRef]
7. Strupas K, Poskus E, Ambrazevicius M. Retrorectal tumours: literature review and vilnius university hospital "santariskiu klinikos" experience of 14 cases. Eur J Med Res 2011;16:231–6. [CrossRef]
8. Maddah G, Abdollahi A, Etemadrezaie H, Ganjeifar B, Gohari B,

Abdollahi M, et al. Problems in Diagnosis and Treatment of Retrorectal Tumors: Our Experience in 50 Patients. Acta Med Iran 2016;54:644–50.

9. Rosenthal DI, Scott JA, Mankin HJ, Wismer GL, Brady TJ. Sacrococcygeal chordoma: magnetic resonance imaging and computed tomography. AJR Am J Roentgenol 1985;145:143–7. [CrossRef]
10. Bullard KM, Rothenberger DA. Colon, rectum, and anus. In: Brunicardi FC, Andersen DK, Billiar TR, Dunn DL, Hunter JG, Matthew JB, et al., editors. Schwartz's Principles of Surgery. 8th ed. New York: McGraw Hill Medical; 2005. p. 1095.
11. Hobson KG, Ghaemmaghami V, Roe JP, Goodnight JE, Khatri VP. Tumors of the retrorectal space. Dis Colon Rectum 2005;48:1964–74.
12. Verazin G, Rosen L, Khubchandani IT, Sheets JA, Stasik JJ, Riether R. Retrorectal tumor: is biopsy risky? South Med J 1986;79:1437–9.
13. Abel ME, Nelson R, Prasad ML, Pearl RK, Orsay CP, Abcarian H. Parasacrococcygeal approach for the resection of retrorectal developmental cysts. Dis Colon Rectum 1985;28:855–8. [CrossRef]
14. Pidala MJ, Eisenstat TE, Rubin RJ, Salvati EP. Presacral cysts: transrectal excision in select patients. Am Surg 1999;65:112–5.
15. Aslan E. Transvaginal excision of a retrorectal tumor presenting as rectocele. Int Urogynecol J Pelvic Floor Dysfunct 2008;19:1715–7.
16. Gunkova P, Martinek L, Dostalik J, Gunka I, Vavra P, Mazur M. Laparoscopic approach to retrorectal cyst. World J Gastroenterol 2008;14:6581–3. [CrossRef]
17. Chen Y, Xu H, Li Y, Li J, Wang D, Yuan J, et al. Laparoscopic resection of presacral teratomas. J Minim Invasive Gynecol 2008;15:649–51. [CrossRef]
18. Zoller S, Joos A, Dinter D, Back W, Horisberger K, Post S, et al. Retrorectal tumors: excision by transanal endoscopic microsurgery. Rev Esp Enferm Dig 2007;99:547–50. [CrossRef]

Nadir Tanı Alan Retrorektal Tümörler: Tek Merkezli Kliniğin Tecrübesi

Amaç: Retrorektal tümörler (presakral, prekoksigeal) nadir karşılaşılmasından dolayı tanı ve tedavisi zordur. Bu çalışmanın amacı hastanemizdeki retrorektal tümörlere cerrahi deneyimimizi irdelemek.

Gereç ve Yöntem: Çalışmamızda Ocak 2012 ve Aralık 2016 tarihleri arasında S.B. Üniversitesi Kartal Dr. Lütfi Kırdar Eğitim ve Araştırma Hastanesi/İstanbul Genel Cerrahi Kliniği'ne başvuran 12 hasta değerlendirildi. Radyolojik bulgular, patoloji raporları ve cerrahi teknik geriye dönük olarak tıbbi kayıtlardan analiz edildi.

Bulgular: Toplam olarak 12 hasta değerlendirildi; 10 hasta kadın, iki hasta erkekti, ortalama yaş 45.2 (dağılım, 31-65 yaş) idi. Sekiz hastanın kitlesi doğuştan orijinli iken üç hastanın kitlesi nörojenik orijinli ve bir hastanınki anjiyomiksomatöz kaynaklı tümördü. Anteriyor ve posteriyor olmak üzere iki cerrahi teknik uygulandı.

Sonuç: Tümörün total cerrahi rezeksiyonu ve nörolojik fonksiyonların korunması başarılı tedavinin asıl amacıdır. En iyi sonuca ulaşmak için multidisipliner yaklaşım ve doğru planlama önemlidir.

Anahtar Sözcükler: Cerrahi; retrorektal; tümör.