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# Surgical Management of Retrorectal Tumors

## A French Multicentric Experience of 270 Consecutives Cases

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**Objective:** To report the largest multicentric experience on surgical management of retrorectal tumors (RRT).

**Background:** Literature data on RRT is limited. There is no consensus concerning the best surgical approach for the management of RRT.

**Methods:** Patients operated for RRT in 18 academic French centers were retrospectively included (2000–2019).

**Results:** A total of 270 patients were included. Surgery was performed through abdominal ( $n = 72$ , 27%), bottom ( $n = 190$ , 70%), or combined approach ( $n = 8$ , 3%). Abdominal approach was laparoscopic in 53/72 (74%) and bottom approach was Kraske modified procedures in 169/190 (89%) patients. In laparoscopic abdominal group, tumors were more frequently symptomatic (37/53, 70% vs 88/169, 52%,  $P = 0.02$ ), larger [mean diameter =  $60.5 \pm 24$  (range, 13–107) vs  $51 \pm 26$  (20–105) mm,  $P = 0.02$ ] and located above S3 vertebra ( $n = 3/42$ , 7% vs 0%,  $P = 0.001$ ) than those from Kraske modified group. Laparoscopy was associated with a higher risk of postoperative ileus ( $n = 4/53$ , 7.5% vs 0%,  $P = 0.002$ ) and rectal fistula ( $n = 3/53$ , 6% vs 0%,  $P = 0.01$ ) but less wound abscess ( $n = 1/53$ , 2% vs 24/169, 14%,  $P = 0.02$ ) than Kraske modified procedures. RRT was malignant in 8%. After a mean follow up of  $27 \pm 39$  (1–221) months, local recurrence was noted in 8% of the patients. After surgery, chronic pain was observed in 17% of the patients without significant difference between the 2 groups (15/74, 20% vs 3/30, 10%;  $P = 0.3$ ).

**Conclusions:** Both laparoscopic and Kraske modified approaches can be used for surgical treatment of RRT (according to their location and their size), with similar long-term results.

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Retrorectal tumors (RRT) are a heterogeneous group of lesions developed into the retrorectal space. This is a virtual space bounded posteriorly by the presacral fascia, anteriorly by the mesorectum, and laterally by the ureters and iliac vessels. RRT are rare, with an estimated incidence about 1 from 40,000 patients' admissions. They affect preferentially women in the third decade.<sup>1</sup> Regarding pathological features of RRT, several classifications have been reported,<sup>2–5</sup> the most frequently used being based on cell lines, which divide malignant and benign RRT by congenital, neurogenic, osseous, inflammatory, and miscellaneous tumors.<sup>6</sup> Benign RRT are most frequently observed, from 50% to 95% of cases.<sup>1,7–12</sup> Congenital lesions are the most common histological type, mainly because the retrorectal space may contain embryological remnant. Tailgut cyst is the most common lesion encountered from 8% to 59.5% of cases.<sup>1,7–12</sup>

Even if RRT are more frequently benign, surgery is always recommended because the risk of malignant transformation in adenocarcinoma can reach 12% in case of congenital lesions.<sup>13</sup> Moreover, tumor infection responsible of iterative surgeries, which can lead to any misdiagnosis has been reported in 30% of cases, justifying surgical resection, whatever the histological type.<sup>13</sup> Since the first description by Kraske in 1885,<sup>14</sup> several abdominal and perineal approaches have been described for the management of RRT, without any strong evidence for one superior to the others. We have previously reported safety and feasibility of laparoscopy in the management of RRT through 12 patients.<sup>15</sup> Diagnosis and management of RRT have been improved during the past decades thanks to the development of pelvic magnetic resonance imaging (MRI).<sup>16,17</sup> MRI sensitivity for the diagnosis of RRT can reach 100%.<sup>16</sup> Recently, some algorithms for surgical management according to the tumor location and to the involvement of adjacent organs have been proposed.<sup>16</sup> Prediction of malignancy on preoperative examination is one of the cornerstones of RRT management, especially as the prognosis is closely related to pathological characteristics.<sup>8</sup>

Except 1 observational series including 120 patients,<sup>1</sup> current literature on RRT is limited to retrospective, small, non-comparative studies and, there is still no consensus regarding the best surgical

management of RRT. Thus, the aims of our study were to report the results of a large surgical cohort of RRT and to assess postoperative morbidity and long-term results according to the chosen surgical procedure.

## PATIENTS AND METHODS

### Study Population

All the consecutive adult patients surgically managed for RRT from 2000 to 2019 in 18 French academic colorectal surgical departments were retrospectively included. Patients with recurrent RRT, RRT without surgical management, and RRT secondarily referred after outside surgical management were excluded. The collected data, including demographic characteristics, tumor characteristics, perioperative data, pathological, and long-term results were provided by surgeons from each center after institutional approval.

### Surgical Approaches

The anterior surgical approach was considered as an abdominal approach whether performed by laparotomy, laparoscopy, or robotic surgery. The bottom approach included Kraske modified procedures (KMP), perineal or trans-anal resection of RRT.

KMP were performed in prone jack-knife position to approach the RRT posteriorly. A vertical median or paramedian incision was performed from the last sacral vertebra to 2 cm from the anus. The anococcygeal ligament was divided to achieve access to the retrorectal space. The coccyx was resected when necessary, and the trans-sacral incision was exceptionally performed. We defined KMP because the original Kraske description<sup>14</sup> was not really performed.

The perineal approach was performed through lithotomy position with a horizontal incision between the anus and the coccyx and we did not consider it as a KMP.

A combined approach was defined by an anterior approach associated with a bottom resection of the RRT.

### Outcomes Measures

Primary outcomes were to report intraoperative complications, postoperative morbidity, and long-term results from the overall RRT cohort.

Secondary outcomes were based on the comparison between the 2 main surgical approaches performed to remove RRT (laparoscopic and KMP) in terms of intraoperative complications, postoperative morbidity, and long-term results.

Intraoperative complications were defined as follows: tumor perforation (effraction of the RRT during surgery), presacral bleeding, rectal, and bladder perforation. Conversion was defined when the surgical approach was changed during the procedure due to operative difficulties, including the change from laparoscopy to open surgery but also the change of surgical approach, from any abdominal approach to any bottom approach (and vice versa).

Postoperative morbidity was defined as any surgical or medical complication occurring during the hospital stay or within 30 days after surgery. Severity of surgical morbidity was classified according to that described by Dindo.<sup>18</sup>

Long-term results included functional results (reported among patients followed more than 6 months) and oncological results.

### Statistical Analysis

Quantitative data were reported as mean and standard deviation, and categorical data were reported as absolute numbers and percentages. Two groups were defined according to the main surgical procedure (laparoscopic and KMP) and were compared.

Quantitative data were analyzed with the Student *t* test. Categorical data were compared using the Pearson  $\chi^2$  test or the

Fisher exact test, as appropriate. All tests were 2-sided, with a level of significance set at  $P < 0.05$ . Statistical analyses were performed using Statistical Package for the Social Sciences (Statistical Package for the Social Sciences Inc., version 24.0, Chicago, IL) software. This study was conducted according to the ethical standards of the committee on human experimentation of our institution and reported according to the Strengthening the Reporting of Observational Studies in Epidemiology guidelines.<sup>19</sup>

## RESULTS

### Patients and Tumor Characteristics

Between 2000 and 2019, 270 patients with RRT were operated in 18 academic colorectal surgical departments: 79% of patients were women ( $n = 213/270$ ) and the mean age was  $46 \pm 15$  (range, 18–77) years.

The diagnosis of RRT was made on clinical symptoms in 56% ( $n = 151/270$ ) patients, mainly represented by pelvic pain (36%,  $n = 90/151$ ), dyschesia (10%,  $n = 15/151$ ), urinary or gynecological disorders (10%,  $n = 15/151$ ), and neurologic symptoms (4%,  $n = 6/151$ ). Regarding preoperative examinations, MRI, computed tomography scan, and endorectal ultrasound were performed in 90%, 46%, and 34% of the patients, respectively. RRT characteristics are reported in Table 1. Mean tumor size was  $57 \pm 29$  mm (range, 6–150). RRT were mainly located below S3 vertebra (79%,  $n = 168/270$ ) (Fig. 1). An involvement of neighboring organs was suspected in 7% ( $n = 20/270$ ) of the patients. Five patients underwent a neoadjuvant treatment (2%), including chemotherapy ( $n = 2$ ), chemoradiotherapy ( $n = 1$ ), hormone therapy ( $n = 1$ ), and radiological embolization ( $n = 1$ ). In laparoscopic group, tumors were more frequently symptomatic (37/53, 70% vs 88/169, 52%,  $P=0.02$ ), larger [mean diameter =  $60.5 \pm 24$  (range, 13–107) vs  $51 \pm 26$  (20–105) mm,  $P=0.02$ ] and located above S3 vertebra ( $n = 3/42$ , 7% vs 0%,  $P = 0.001$ ) than those from KMP group (Table 1).

### Surgical Treatment

Surgery was performed through an abdominal (27%,  $n = 72/270$ ), a bottom (70%,  $n = 190/270$ ), or a combined (3%,  $n = 8/270$ ) approach (Supplementary Fig. 2, <http://links.lww.com/SLA/D325>). Abdominal approach was performed laparoscopically (73.5%,  $n = 53/72$ ), by open (23.5%,  $n = 17/72$ ), or robotic (3%,  $n = 2/72$ ) surgery. The bottom approach included KMP (89%,  $n = 169/190$ ), perineal (8%,  $n = 16/190$ ), and transanal (3%,  $n = 5/190$ ) approaches.

Whatever the surgical approach, the conversion rate was 5% ( $n = 13/270$ ) and included 8 conversions from laparoscopy to open surgery and 5 conversions from laparoscopy to a bottom approach. The mean operative time was  $140 \pm 84$  (range, 30–420) minutes. Intraoperative complications occurred in 46% of patients ( $n = 124/270$ ) and were mainly represented by RRT perforation during the surgery (40%,  $n = 107/270$ ), rectal perforation (4%,  $n = 12/270$ ) and pre-sacral bleeding (4%,  $n = 11/270$ ).

Focusing on the 2 main surgical approaches, operative time was significantly longer during laparoscopic approach than KMP [ $183 \pm 76$  (70–355) vs  $97 \pm 63$  (30–318) minutes,  $P = 0.001$ ]. The conversion rate was significantly higher during laparoscopy in comparison with KMP (23%,  $n = 12/53$ , vs 1%,  $n = 1/169$ ,  $P = 0.001$ ). The perioperative adverse events were similar between the 2 groups (47%,  $n = 25/53$ , vs 47%,  $n = 80/169$ ,  $P = 1$ ; Table 2).

### Postoperative Morbidity

The mean length of stay for the overall population was  $6 \pm 4$  (range, 1–30) days. Overall morbidity rate was 30% ( $n = 81/270$ ; Table 2). Surgical morbidity was mainly represented by wound (11%,

**TABLE 1.** Characteristics of 270 Patients Operated for Retrorectal Tumors

	Overall Population* n = 270	Laparoscopic approach n = 53	Kraske modified approach n = 169	P value
Diagnosis				
Incidental	119 (44) <sup>†</sup>	16 (30)	81 (48)	<b>0.02</b>
Symptomatic	151 (56)	37 (70)	88 (52)	
Pelvic pain	90 (34)	23 (68)	53 (64)	0.8
Obstructed defecation	15 (6)	2 (6)	11 (13)	0.3
Urinary or gynecological disorders	15 (6)	3 (9)	9 (11)	1.00
Neurologic symptoms	6 (2)	1 (3)	1 (1)	0.5
Others	17 (6.5)	5 (15)	9 (11)	0.5
NA	8	3	5	
Preoperative workup				
MRI	242 (90)	47 (92)	150 (93)	0.8
CT scan	123 (45.5)	26 (51)	66 (42)	0.3
Endorectal ultrasound	93 (34)	27 (55)	50 (32)	<b>0.004</b>
Biopsy	36 (13)	12 (23)	15 (10)	<b>0.001</b>
Adjacent organ involvement	20 (7)	4 (8)	8 (6)	0.5
Tumor size (mm)	57 ±29 [6–150] <sup>‡</sup>	60.5 ±24 [13–107]	51 ±26 [22–105]	<b>0.02</b>
Location				<b>0.001</b>
Above S3ab	9 (4)	3 (7)	0	
Above and below S3	36 (17)	11 (26)	14 (11)	
Below S3	168 (79)	28 (67)	116 (89)	
NA	57	11	39	
Malignant suspicion	22 (8)	8 (27)	14 (13)	0.07
Neoadjuvant treatment	5 (2)	0	1 (0.6)	1.00

\*The overall population included retrorectal tumors operated by abdominal laparoscopic approach and Kraske modified procedures, but also abdominal open or robotic, transanal, transperineal and combined approaches.

<sup>†</sup>Number (percentage).

NA indicates not available; MRI: magnetic resonance imaging; CT: computed tomography.

<sup>‡</sup>mean ± standard deviation [range].

n = 29/270) and pelvic (6%, n = 16/270) abscesses. Unplanned reoperation occurred in 5.5% (n = 15/270) of patients, to drain pelvic abscess or hematoma (n = 13), and to perform a diverting colostomy for rectal fistula (n = 2).

The laparoscopic approach was associated with a longer length of stay in comparison with KMP (7 ±4 vs 5 ±4 days, *P* = 0.001). No significant difference was reported in terms of surgical complications, complications severity, unplanned reoperation, or readmission between the 2 groups (Table 2). The laparoscopic approach was significantly associated to a higher risk of rectal fistula [6% (n = 3/53) vs 0% (n = 0/169), *P* = 0.01] and postoperative ileus [7.5% (n = 4/53) vs 0% (n = 0/169), *P* = 0.002], and a

lower risk of wound abscess [2% (n = 1/53) vs 14% (n = 21/169), *P* = 0.02] than KMP (Table 2).

### Pathological Findings

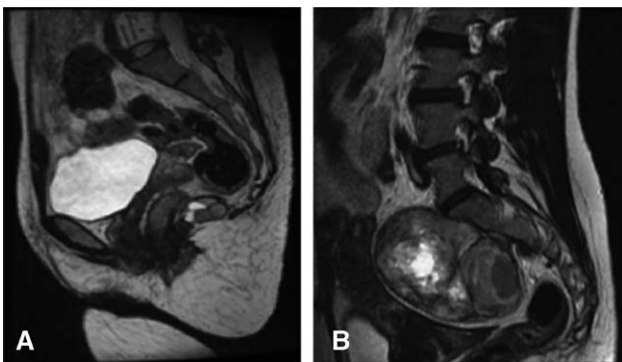
Among the RRT, 92% of them were benign (n = 248/270). Pathological results are summarized in the Supplementary Table, <http://links.lww.com/SLA/D325>. Involved resection margin was reported in 19 patients (7%), with benign (n = 11), or malignant (n = 8) RRT. Among the 8 malignant RRT with R1 resection, 6 were managed by an adjuvant therapy (radiotherapy n = 3, additional surgery n = 2, chemoradiotherapy n = 1). No significant difference was observed between the 2 groups in terms of R1 resection (7.5% vs 4.7%, *P* = 0.5).

### Long Term Follow-up

The mean follow-up after surgery was 27 ±39 (range 1–221) months. A recurrence was reported in 8% of the patients (n = 20/241). The recurrence was locoregional (n = 18), metastatic (n = 1), and combined (n = 1). With regards to the pathological type, 15 and 5 recurrences occurred among 219 benign (7%), and 22 malignant (23%) RRT, respectively (*P* = 0.025).

Among the patients with recurrences, 10 were reoperated, 2 received chemotherapy, and 1 was treated by radiotherapy (Table 3). There was no significant difference in terms of recurrence according to the surgical approach whether for benign RRT (12%, n = 5/43 in laparoscopic group vs 3.5%, n = 5/135 in KMP group, *P* = 0.06), or malignant RRT (60%, n = 3/5 vs 10%, n = 1/10, *P* = 0.08, respectively).

The long-term functional results were reported after a follow-up longer than 6 months, in 116 patients (Table 3). A chronic pain was observed in 20/116 patients (17%). Although not significant, a lower rate of chronic pain was noted after laparoscopy (10%, n = 3/30) than



**FIGURE 1.** Magnetic resonance imaging illustrating a very low retrorectal tumor located below the third sacral vertebra (A) and a voluminous retrorectal tumor located above the third sacral vertebra (B).

**TABLE 2.** Perioperative Data of 270 Patients Operated for Retrorectal Tumor

	Overall Population* n = 270	Laparoscopic approach n = 53	Kraske modified approach n = 169	P value
Operative time (min)	140 ±84 [30–420] <sup>(a)</sup>	183 ±76 [70–355]	97 ±63 [30–318]	<b>0.001</b>
Conversion <sup>†</sup>	13 (5) <sup>(b)</sup>	12 (23)	1 (1)	<b>0.001</b>
Adjacent organ resection	5 (2)	1 (2)	1 (1)	0.4
Coccyx resection	101 (37)	0	97 (57)	–
Perioperative complication <sup>‡</sup>	124 (46)	25 (47)	80 (47)	1
Tumor perforation	107 (40)	20 (38)	71 (42)	0.6
Rectal perforation	12 (4)	3 (6)	8 (5)	0.7
Presacral bleeding	11 (4)	3 (6)	4 (2)	0.4
Bladder perforation	1 (0.4)	1 (2)	0	0.2
Length of stay (d)	6 ±4	7 ±4	5 ±4	<b>0.001</b>
Overall morbidity	81 (30)	15 (28)	51 (30)	0.7
Medical morbidity	15 (5.5)	5 (9)	6 (3.5)	0.1
Acute urinary retention	7 (2.5)	1 (2)	5 (3)	1
Urinary infection	6 (2)	4 (7.5)	2 (1)	<b>0.03</b>
Others <sup>(c)</sup>	3 (1)	2 (4)	0	0.06
Surgical morbidity	72 (27)	13 (24.5)	47 (28)	0.6
Wound abscess	29 (11)	1 (2)	24 (14)	<b>0.02</b>
Pelvic abscess	16 (6)	3 (6)	12 (7)	1
Pain requiring morphine	15 (5.5)	2 (4)	10 (6)	0.7
Bleeding	11 (4)	2 (4)	8 (5)	1
Ileus	5 (2)	4 (7.5)	0	<b>0.002</b>
Rectal fistula	3 (1)	3 (6)	0	<b>0.01</b>
Others <sup>(d)</sup>	3 (1)	1 (2)	0	0.2
Severity of surgical morbidity				1
Dindo I/II	56 (21)	10 (19)	36 (21)	
Dindo III/IV	16 (6)	3 (6)	11 (6.5)	
Unplanned reoperation	15 (5.5)	3 (6)	10 (6)	1
Radiological drainage	1 (0.4)	0	1 (1)	1
Unplanned readmission	27 (10)	5 (9)	19 (11)	0.8

\*The overall population included retrorectal tumors operated by abdominal laparoscopic approach and Kraske modified procedures, but also abdominal open or robotic, transanal, transperineal and combined approaches.

<sup>†</sup>Conversion was defined when the surgical approach was changed during the procedure due to operative difficulties, whether from laparoscopy to open surgery or from an anterior to a posterior approach and conversely.

<sup>‡</sup>Some patients underwent several complications. (a) mean ±standard deviation [range]; (b) number (percentage); (c) include fecaloma (n = 1), headache (n = 1) and acute renal deficiency (n = 1); (d) include neurologic complication (n = 2), and gas embolism (n = 1).

P < 0.05 was considered as significant (in bold).

**TABLE 3.** Long-term Results After Resection of Retrorectal Tumor in 270 Patients

	Overall Population* N = 270	Laparoscopic Approach N = 53	Kraske Modified Approach N = 169	P value
Follow up (mo) <sup>†</sup>	27 ±39 [1–221] <sup>(a)</sup>	37 ±42 [1–130]	24 ±36 [1–133]	0.06
Recurrence of benign RRT	15/219 (7) <sup>(b)</sup>	5/43 (12)	5/135 (4)	0.06
NA	29	5	24	
Recurrence management <sup>‡</sup>				1
No treatment	7 (47)	2 (40)	3 (60)	
Surgery	8 (53)	3 (60)	2 (40)	
Recurrence of malignant RRT	5/22 (23)	3/5 (60)	1/10 (10)	0.08
Recurrence management <sup>‡</sup>				0.1
Surgery	2 (40)	0	1 (100)	
Chemotherapy	2 (40)	2 (67)	0	
Radiotherapy	1 (20)	1 (33)	0	
Functional results				
Available > 6 months	N = 116	N = 30	N = 74	
Chronic pain	20 (17)	3 (10)	15 (20)	0.3
Urinary dysfunction	6 (5)	4 (13)	0	<b>0.006</b>
Dyschesia	4 (3)	3 (10)	0	<b>0.02</b>
Chronic wound healing complications	4 (3)	0	3 (4)	0.6
Sexual dysfunction	3 (3)	1 (3)	1 (1)	0.5
Faecal incontinence	2 (2)	1 (3)	1 (1)	0.5
Incisional hernia	2 (2)	2 (7)	0	0.08

\*The overall population included retrorectal tumors operated by abdominal laparoscopic approach and Kraske modified procedures, but also abdominal open or robotic, transanal, transperineal and combined approaches.

<sup>†</sup>Follow-up was reported for benign and malignant RRT.

<sup>‡</sup>% was estimated from the number of patients with recurrence.

(a) mean ±standard deviation [range]; (b) number (percentage); P < 0.05 was considered as significant (in bold).

after KMP (20%,  $n = 15/74$ ,  $P = 0.3$ ). Urinary dysfunction and dyschesia were more frequently observed after laparoscopy than after KMP ( $n = 4/30$  vs  $0$ ,  $n = 0/74$ ,  $P = 0.006$ , and 10%,  $n = 3/30$  vs 0%,  $n = 0/74$   $P = 0.02$ , respectively).

## DISCUSSION

We report here the largest multicentric experience on surgery for RRT in 270 patients. In this series, the bottom approach was most frequently performed (70%), and mainly represented by KMP (89%). The abdominal approach was mainly represented by the laparoscopy (74%). Our study suggested that laparoscopy and KMP were not firmly

performed for the same kind of RRT. Laparoscopy was more frequently performed for symptomatic, larger, and RRT located above S3 vertebra than KMP. We did not report any significant difference between the 2 surgical groups in terms of morbidity, severity of surgical morbidity, reintervention, and readmission. Moreover, no difference in terms of recurrence either for benign and malignant RRT have been reported according to the surgical approach.

The previous largest series of RRT was reported in 1985 with 120 patients: 102 patients were operated on through posterior ( $n = 79$ ), abdominal ( $n = 21$ ), and combined ( $n = 2$ ) approaches.<sup>1</sup> Since then, the laparoscopic resection of RRT has been proposed.<sup>20</sup> Indeed,

**TABLE 4.** Literature Review About Surgical Management of Retrorectal Tumor

	Total	Surgical Approaches	Postoperative Morbidity	Follow-up <sup>(b)</sup>	Recurrence
Jao et al 1985 <sup>1</sup>	102	Posterior 79 Abdominal 21 Combined 2	NA	9.6 [3–23] yr	15% benign 70% malignant
Glasgow et al 2005 <sup>23</sup>	34	Posterior 11 Abdominal 14 Combined 9	5 (15) <sup>(a)</sup>	22 mo	0 benign 100% malignant
Woodfield et al 2008 <sup>16</sup>	27	Posterior 12 Abdominal 11 Combined 4	4 (15)	49 [2–72] mo 26 [10–61] mo	5% benign 29% malignant
Mathis et al 2010 <sup>10</sup>	31	Posterior 20 Abdominal 9 Combined 2	8 (26)	2 [1–22.6] yr	3%
Dozois et al 2011 <sup>24</sup>	37	NA	21 (57)	5 yr	62%
Bosca et al 2012 <sup>25</sup>	20	Posterior 15 Abdominal 3 Combined 2	8 (40)	33 ±30 mo 29 ±17 mo	45%
Macafee et al 2012 <sup>26</sup>	56	Posterior 20 Abdominal 27 Combined 9	19 (34)	46 [6–90] mo	0 benign 12% malignant
Duclos et al 2012 <sup>15</sup>	12	Abdominal 12	2 (17)	34 [12–79] mo	0
Messick et al 2013 <sup>9</sup>	84	Posterior 50 Abdominal 25 Combined 9	NA	8.3 [0.2–225] mo	11% benign 30% malignant
Lev-Chelouche et al 2013 <sup>12</sup>	42	Posterior 21 Abdominal 18 Combined 3	15 (36)	NA	0 benign 57% malignant
Chéreau et al 2013 <sup>8</sup>	47	Posterior 42 Abdominal/combined 5	4 (8.5)	71 [2–168] mo	3% benign 22% malignant
Sagar et al 2014 <sup>27</sup>	76	Posterior 31 Abdominal 41 Combined 4	NA	NA	NA
Patsouras et al 2015 <sup>28</sup>	17	Posterior 17	5 (29)	13 [3–36] mo	6%
Hopper et al 2015 <sup>29</sup>	24	Posterior 15 Abdominal 3 Combined 6	NA	20 [5–66] mo	NA
Sakr et al 2019 <sup>30</sup>	24	Posterior 11 Abdominal 10 Combined 3	10 (42)	12 [1–66] mo	0
Carpelan-Holmstrom et al 2020 <sup>22</sup>	52	Posterior 44 Abdominal 7 Combined 1	11 (21)	2 yr	27%
Current series 2021	270	Posterior 190 Abdominal 72 Combined 8	81 (30)	27 ±39 mo	6% benign 23% malignant
Total	955	Posterior 578 Abdominal 273 Combined 62	193 (29)	–	7% benign * 46% malignant * 19% overall*

\*Calculated with available data.

(a) number (percentage); (b) median [range] or mean ±standard deviation; NA: Not available.

we have previously reported the results of laparoscopic approach in 12 consecutive patients with RRT. Perioperative adverse events were observed in 3 of them, including exposure difficulties, rectal injury and bleeding leading to a conversion into open surgery in 2 cases.<sup>15</sup> A recent systematic review of 34 studies, including 82 patients with RRT who underwent minimally invasive surgery (73 laparoscopy and 9 robotic surgery), reported a conversion rate of 5.5%, a postoperative morbidity of 15.7% and a median length of stay of 4 days, confirming the feasibility and the safety of these procedures for RRT.<sup>21</sup> In our study, we reported that laparoscopy was most frequently performed for symptomatic, large RRT and those located above S3 vertebra than KMP. This may explain the worse intraoperative outcomes observed with laparoscopy in comparison with KMP, such as higher conversion rate or longer operative time. Indeed, even if the abdominal procedures were performed by an open approach, Chéreau et al reported longer operative time with the abdominal approach, due to the necessity to do a rectal en bloc resection.<sup>8</sup> Likewise, Mathis et al have reported that the abdominal or combined approach was performed when RRT was located above S3 vertebra, or if malignancy or any contact with neighboring structures were suspected. For these reasons, postoperative complications (54.5% vs 10%,  $P = 0.007$ ) and hospital stay (6.9 vs 2.6 days,  $P = 0.002$ ) were significantly worse after abdominal or combined approach than after posterior approach.<sup>10</sup>

We have also observed that laparoscopic approach was significantly associated with higher rates of postoperative ileus (7.5% vs 0%,  $P = 0.002$ ) and rectal fistula (6% vs 0%,  $P = 0.01$ ) but lower rate of wound abscess (2% vs 14%,  $P = 0.02$ ) than KMP. The length of stay was significantly longer after laparoscopy than after KMP ( $7 \pm 4$  vs  $5 \pm 4$  days,  $P = 0.001$ ), but no difference was observed in terms of severity of morbidity, unplanned reoperation, or readmission rate. These postoperative results may be explained by the different preoperative RRT characteristics between the 2 surgical approaches. Whatever the surgical approach, we observed postoperative morbidity in 30% of patients, which is in accordance with others (Table 4).

We observed that recurrence occurred in 8% of patients after a mean follow up of  $27 \pm 39$  (range, 1–221) months. Recurrence occurred most frequently in the case of malignant RRT when compared with benign RRT, in accordance with others (Table 4), but only 1 independent predictive factor has been identified. Indeed, Carpelan-Holmström et al have reported recurrence in 27% of 52 RRT operated on, and identified the multilobular characteristic as an independent predictive factor.<sup>22</sup> In our series, the recurrence rate was not statistically different between the 2 main surgical approaches.

The last issue in the management of RRT is long-term functional complication. Indeed, we have noted an occurrence of chronic pain, urinary dysfunction and dyschesia in 17%, 5%, and 3% of patients with functional results available after 6 postoperative months. Even if this difference was not significant, we have observed that chronic pain occurred in 10% after laparoscopy versus 20% after KMP ( $P = 0.3$ ). Likewise, Chéreau et al have reported 9.5% of chronic pain after posterior resection and no fecal incontinence, after a median follow-up of 71 (2–168) months.<sup>8</sup> Mathis et al reported 2 pelvic floor dysfunctions improved with biofeedback and 1 sexual dysfunction among 31 RRT operated on.<sup>10</sup> Glasgow et al reported 2 ventral hernia and 2 sexual impotencies among 34 RRT operated on, without significant difference between surgical approaches.<sup>23</sup>

Finally, our current series is mainly limited by its retrospective nature, leading to some missing data. Yet, the rarity of RRT does not allow any prospective series, and the largest size of our series provides an important experience in the management of RRT. Another limit is the short follow-up, due to the higher rate of benign RRT. The follow-up should be longer, since the recurrence may occur after 7 years, as we have observed.

In conclusion, diagnosis, and management of RRT are challenging, because of the rarity and the heterogeneous histological features. The 2 main approaches represented by laparoscopy and KMP have their own postoperative morbidity, but they are not performed for the same type of lesions. The laparoscopic approach could be preferred for large RRT, located above the S3 vertebra or those with rectal involvement for which a rectal en bloc resection is required. On the opposite KMP seem the best option for low and small RRT.

## REFERENCES

- Jao SW, Beart RW, Spencer RJ, et al. Retrorectal tumors. Mayo Clinic experience, 1960-1979. *Dis Colon Rectum*. 1985;28:644–652.
- Middeldorf K. Zur Kenntnis der angeborenen Sacralgeschwulste. *Virchow's Arch Pathol Anat*. 1885;101:37–44.
- Lovedaly SB, Dockerty MB. Extragenital pelvic tumors in women. *Am J Obstet Gynecol*. 1949;58:215–234.
- Malafosse M, Gallot D, Douvin D, et al. Peri-ano-rectal cysts and tumours of vestigial origin in the adult. *J Chir*. 1977;113:351–360.
- Barthod F, Herve? de Sigalony JP, Parc R. Les formations kystiques vestigiales re?trorectales de l'adulte. A? propos de trois cas. *Ann Chir*. 1996;50:906–912.
- Uhlig BE, Johnson RL. Presacral tumors and cysts in adults. *Dis Colon Rectum*. 1975;18:581–596.
- Hobson KG, Ghaemmaghami V, Roe JP, et al. Tumors of the retrorectal space. *Dis Colon Rectum*. 2005;48:1964–1974.
- Chéreau N, Lefevre JH, Meurette G, et al. Surgical resection of retrorectal tumours in adults: long-term results in 47 patients. *Colorectal Dis*. 2013;15:e476–e482.
- Messick CA, Hull T, Rosselli G, et al. Lesions originating within the retrorectal space: a diverse group requiring individualized evaluation and surgery. *J Gastrointest Surg*. 2013;17:2143–2152.
- Mathis KL, Dozois EJ, Grewal MS, et al. Malignant risk and surgical outcomes of presacral tailgut cysts. *Br J Surg*. 2010;97:575–579.
- Baek SK, Hwang GS, Vinci A, et al. Retrorectal tumors: a comprehensive literature review. *World J Surg*. 2016;40:2001–2015.
- Lev-Chelouche D, Gutman M, Goldman G, et al. Presacral tumors: a practical classification and treatment of a unique and heterogenous group of diseases. *Surgery*. 2003;133:473–478.
- Chêne G, Voitellier M. Tératome bénin mature pré-sacré et formations kystiques vestigiales rétro-rectales chez l'adulte. *J Chir*. 2006;143:310–314.
- Kraske P. Zur extirpation hochsitzender mastdarm-krebse. *Verhandl Deutsch Gellesch Chir*. 1885;14:464–474.
- Duclos J, Maggiori L, Zappa M, et al. Laparoscopic resection of retrorectal tumors: a feasibility study in 12 consecutive patients. *Surg Endosc*. 2014;28:1223–1229.
- Woodfield JC, Chalmers AG, Phillips N, et al. Algorithms for the surgical management of retrorectal tumours. *Br J Surg*. 2008;95:214–221.
- Yang B-L, Gu Y-F, Shao W-J, et al. Retrorectal tumors in adults: magnetic resonance imaging findings. *World J Gastroenterol*. 2010;16:5822–5829.
- Dindo D, Demartines N, Clavien P-A, et al. Classification of surgical complications: a new proposal with evaluation in a cohort of 6336 patients and results of a survey. *Ann Surg*. 2004;240:205–213.
- von Elm E, Altman DG, Egger M, et al. The strengthening the reporting of observational studies in epidemiology (STROBE) statement: guidelines for reporting observational studies. *Lancet*. 2007;370:1453–1457.
- Sharpe LA, Van Oppen DJ. Laparoscopic removal of a benign pelvic retroperitoneal dermoid cyst. *J Am Assoc Gynecol Laparosc*. 1995;2:223–226.
- Mullaney TG, Lightner AL, Johnston M, et al. A systematic review of minimally invasive surgery for retrorectal tumors. *Tech Coloproctol*. 2018;22:255–263.
- Carpelan-Holmström M, Koskenvuo L, Haapamäki C, et al. Clinical management of 52 consecutive retro-rectal tumours treated at a tertiary referral centre. *Colorectal Dis*. 2020;22:1279–1285.
- Glasgow SC, Birnbaum EH, Lowney JK, et al. Retrorectal tumors: a diagnostic and therapeutic challenge. *Dis Colon Rectum*. 2005;48:1581–1587.
- Dozois EJ, Jacofsky DJ, Billings BJ, et al. Surgical approach and oncologic outcomes following multidisciplinary management of retrorectal sarcomas. *Ann Surg Oncol*. 2011;18:983–988.
- Boscà A, Pous S, Artès MJ, et al. Tumours of the retrorectal space: management and outcome of a heterogeneous group of diseases. *Colorectal Dis*. 2011;14:1418–1423.

26. Macafee DAL, Sagar PM, El-Khoury T, et al. Retrorectal tumours: optimization of surgical approach and outcome. *Colorectal Dis.* 2012;14:1411–1417.
27. Sagar AJ, Koshy A, Hyland R, et al. Preoperative assessment of retrorectal tumours. *Br J Surg.* 2014;101:573–577.
28. Pastouras D, Pawa N, Osmani H, et al. Management of tailgut cysts in a tertiary referral centre: a 10-year experience. *Colorectal Dis.* 2015;17:724–729.
29. Hopper L, Eglinton TW, Wakeman C, et al. Progress in the management of retrorectal tumours. *Colorectal Dis.* 2016;18:410–417.
30. Sakr A, Kim HS, Han YD, et al. Single-center experience of 24 cases of tailgut cyst. *Ann Coloproctol.* 2019;35:268–274.

## DISCUSSANTS

### Frederic Ris (Geneva, Switzerland)

I would like to thank the ESA for the privilege of being the first discussant of this paper, and the authors for this interesting study. This study describes a comparison between 2 main surgical approaches (laparoscopy and a Kraske modified approach) for retro-rectal tumors. Such a large number of patients has never been previously reported, and this clearly adds value to the current literature on this topic.

My main comments are the following: first, 240 patients across 18 centers represents 15 patients per center over a period of 20 years. Could the author please comment on the need for centralization, especially regarding the high level of perforation (40%)? Are the results different according to the case load of the different centers?

Second, did you observe a different pain rate in the Kraske modified approach with the removal or the preservation of the coccyx?

Finally, the rate of rectal fistula in the abdominal approach is high with a similar rate of rectal perforation during the surgery. How can you explain this? Was it lower in open surgery than in laparoscopic surgery? Did you find differences in the location or size of the tumors that could be used to prevent these injuries in the future?

### Response From Diane Mege (Clichy, France)

Thank you, Dr. Ris, for your interesting questions. With regards to your first question, I agree with you about the necessity of centralization. Yet, I have 3 points to discuss. The first 1 is that the impact of tumor perforation in retro-rectal tumors is probably not the same as in rectal cancer because the majority of retro-rectal tumors are benign. The second point is that, in some cases of large tailgut cysts, the opening of the tumor or the perforation of the tumor is sometimes necessary to mobilize the tumor. The last point to stress is that I have looked at the impact of the volume of centers on tumor perforation, and when we look at a cut-off of 10 patients per center, we have less tumor perforation in low-volume centers than in high-volume ones. When we look at a cut-off of 20 patients per center, we have the same rate of tumor perforation between the low- and high-volume centers. So, I am not really sure that tumor perforation is due to the volume of the centers.

Regarding coccyx resection, among patients with a KMP, 97 patients had a coccyx resection, and 72 patients did not have coccyx

resection. As I mentioned during the presentation, long-term results were considered only for patients with available functional results at least 6 months post-surgery. So, when we look at patients with long-term results, we have 43 patients with coccyx resection and 31 patients without coccyx resection. The rate of chronic pain in these 2 groups is obviously the same, about 20%. In fact, coccyx resection is not associated with chronic pain. However, perhaps, we need to look at more patients to find a difference.

Your last question is about rectal fistula and perforation during the abdominal approach. In fact, the rate of rectal fistula and perforation is the same because the same patients were involved. With regards to open surgery, rectal perforation occurred less than with the laparoscopic approach. In open surgery, rectal perforation was observed in 1 patient. Unfortunately, however, we only have 17 patients with open surgery, so it is probably not sufficient to draw accurate conclusions regarding this. Regarding the location and size of the tumors, we have observed no significant difference between patients with and those without rectal perforation with the laparoscopic approach. Probably, the main explanation for rectal perforation is the learning curve, and perhaps, surgery should necessitate many rectal digitalization to protect the rectum. Perhaps, we also need to open the tumor in the case of a tailgut cyst to help mobilize the tumor without any rectal perforation.

### Eva Angenete (Göteborg, Sweden)

Thank you for a nice presentation. Do you ever defer surgery in patients with small tailgut cysts? If yes, what is your criteria for no surgery and how do you perform your follow-up?

### Response From Diane Mege (Clichy, France)

Thank you for your question. In fact, we have only included patients who were operated on. However, I think that there is a real place for nonoperative management. Two weeks ago, at the ASCRS congress, a team presented research on the nonoperative management of retro-rectal tumors. Criteria to indicate only a follow-up were the size (under 3 cm); radiological aspects, such as a homogenous tumor or cystic tumor; and the absence of symptoms. In these cases, I think that we can simply follow patients, and maybe, perform a MRI every 1 to 2 years.

### Dieter Hahnloser (Lausanne, Switzerland)

How many of the tailgut cysts had malignant signs on preoperative imaging? In such cases, which approach (transabdominal or Kraske) would you recommend?

### Response From Diane Mege (Clichy, France)

This is a very interesting question. We had 4 malignant tailgut cysts in 114 patients in the definitive pathological analysis, but I do not have the data regarding preoperative characteristics. If we have a malignant tumor, my recommendation would be a surgical approach. The surgical approach depends on the location of the tumor. If the tumor is above the S3, I think that we can perform a laparoscopic or robotic abdominal approach. If it is very low, then we prefer a posterior approach.