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ABOUT COVER

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The primary aim of *World Journal of Gastrointestinal Surgery* (WJGS, *World J Gastrointest Surg*) is to provide scholars and readers from various fields of gastrointestinal surgery with a platform to publish high-quality basic and clinical research articles and communicate their research findings online.

WJGS mainly publishes articles reporting research results and findings obtained in the field of gastrointestinal surgery and covering a wide range of topics including biliary tract surgical procedures, biliopancreatic diversion, colectomy, esophagectomy, esophagostomy, pancreas transplantation, and pancreatectomy, etc.

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Retrospective Cohort Study

Trans-anal endoscopic microsurgery for non- adenomatous rectal lesions

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Abstract

BACKGROUND

Trans-anal endoscopic microsurgery (TEM) enables a good visualization of the surgical field and is considered the method of choice for excision of adenomas and early T1 rectal cancer. The rectum and retro-rectal space might be the origin of uncommon neoplasms, benign and aggressive, certain require radical trans-abdominal surgery, while others can be treated by a less aggressive approach. In this study we report outcomes in patients undergoing TEM for rare and non-adenomatous rectal and retro-rectal lesions over a period of 11 years.

AIM

To report outcomes in patients undergoing TEM for rare and non-adenomatous rectal and retro-rectal lesions over a period of 11 years.

METHODS

Between January 2008 to December 2019 a retrospective analysis was completed for all patients who underwent TEM for non-adenomatous rectal lesion or retro-rectal mass in our institution. Patients were discharged once diet was well tolerated and no complications were identified. They were evaluated at 3 wk post operatively, then at 3-mo intervals for the first 2 years and every 6 mo depending on the nature of the final pathology. Clinical examination and rectoscopy were performed during each of the follow-up visits.

RESULTS

Out of 198 patients who underwent TEM during the study period, 18 had non-

adenomatous rectal or retro-rectal lesions. Mean age was 47 years. The mean size of the lesions was 2.9 mm, with a mean distance from the anal margin of 7.9 cm. Mean surgical time was 97.8 min. There were no intra-operative neither late post-operative complications. Mean length of stay was 2.5 d. Mean patient follow-up duration was 42 mo.

CONCLUSION

TEM allows for reduced morbidity given its minimally invasive nature. Surgeons should be familiar with the technique but careful patient selection should be considered. It can be used safely for uncommon rectal and selected retro-rectal lesions without compromising outcomes. We believe that it should be reasonably considered as one of the surgical methods when treating rare lesions.

Key Words: Trans-anal endoscopic microsurgery; Rectal lesions; Microsurgery Trans-anal endoscopic microsurgery; Rectal lesions; Microsurgery

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Core Tip: Trans-anal endoscopic microsurgery allows for reduced morbidity given its minimally invasive nature. Surgeons should be familiar with the technique but careful patient selection should be considered. It can be used safely for uncommon rectal and selected retro-rectal lesions without compromising outcomes. We believe that it should be reasonably considered as one of the surgical methods when treating rare lesions.

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INTRODUCTION

Trans-anal endoscopic microsurgery (TEM) is a surgical method which is capable of resecting lesions up to 20 cm[1-3]. It enables for excellent access, visualization of the surgical field and for precise[4] full-thickness excision of rectal lesions. Today, TEM is considered the method of choice for excision of adenomas and early T1 rectal cancer[5]. The rectum, as well as the retro-rectal space, can be the origin of a heterogeneous group of uncommon neoplasms[6] that range from aggressive malignancies to benign lesions that are almost always symptomatic[7-11]. The treatment of this rare and heterogeneous group of lesions varies depending on the location and nature of the lesions; accordingly, certain lesions require radical trans-abdominal surgery, while others can be treated by a less aggressive approach.

Since being introduced in 1984[6], indications for the use of TEM have expanded beyond benign adenomas and early rectal cancer. Today, several studies suggest that TEM can also be an effective method for excision of uncommon lesion of the rectum and retro-rectal space[2,7,8,12,13]. The objective of this study is to report outcomes in patients undergoing TEM for rare and non-adenomatous rectal and retro-rectal lesions over a period of 11 years.

MATERIALS AND METHODS

Retrospective analysis was completed for all patients who underwent TEM procedure for non-adenomatous rectal lesion or retro-rectal mass at the Hasharon Hospital, Rabin Medical Center from January 2008 to December 2019.

Data was collected from medical records including patient demographics, tumor location, dimension, preoperative histology if present, and indications for surgery. Additionally, data regarding operative findings, post-operative outcomes and post-operative complications were also collected. The local Institutional review board at Rabin Medical Center approved this retrospective study with a waiver of informed consent.

Prior to surgery, all patients underwent an evaluation protocol for TEM. This protocol included a full colonoscopy with biopsy and rigid proctoscopy to assess the tumor size. Distance from the anal verge and location of the rectal or retro-rectal lesion were identified. Endorectal ultrasound was performed in certain cases. Preparation for surgery in TEM patients included mechanical bowel preparation, preoperatively, and prophylactic antibiotics.

The original Richard Wolf equipment was used to perform the procedure, and the procedure was performed according to the standard technique described by Buess *et al*[1]. Patients were placed in a prone jackknife or lithotomy position depending on the tumor location. When located within the rectal wall the tumor was removed by fullthickness excision with a 1 cm margin. Specimens were pinned and marked for orientation by the surgeon.

Closing methods of large rectal defects were completed by approximation of defect edges by interrupted sutures. Full thickness transverse suturing technique was implemented using absorbable sutures. In the case of peritoneal entry, the

closure of the defect was done in two layers with separate closure of the peritoneum when feasible.

For retro-rectal lesions; a transverse incision of the bulge of the rectal wall was made. The mass was extracted after being dissected within the retro-rectal space from the rectal wall, perirectal and presacral tissues. The rectal opening was closed in the same fashion as described above.

Patients resumed oral intake of liquid and soft diet on post-operative day one; with full diet being subsequently advanced, once tolerated. Pain management included oral dipyrone or paracetamol, with opioids ordered as needed. Patients were discharged once diet was well tolerated and no complications were identified.

Patients were evaluated at 3 wk post operatively. Follow ups were completed at 3-mo intervals for the first 2 years and then every 6 mo depending on the nature of the final pathology. Clinical examination and rectoscopy were performed during each of the followup visits.

RESULTS

Out of 198 patients who underwent TEM during the study period, 18 (9%) had non-adenomatous (non-carcinomas or adenomas) rectal or retro-rectal lesions. Mean age was 47 years (27–81).

The pre-operative diagnoses of these lesions are detailed in Table 1 and include 3 patients with clinical and radiological findings suspicious of gastrointestinal stromal tumor (GI) (GIST), 3 patients with neuroendocrine tumors (NET), 1 patient with leiomyosarcoma, 2 patients with submucosal lipoma, 4 patients with indeterminate pathology and 5 patients with retro-rectal lesions without histological diagnosis.

The mean size of the lesions described was 2.9 mm (1–6 mm), with a mean distance from the anal margin of 7.9 cm (5–13 cm). Regarding the locations of the lesions in the rectal wall; 10 lesions were in the posterior wall, 4 were in the anterior wall, 3 in lateral position, and 1 patient had circular stenosis of rectum. Mean surgical time was 97.8 minutes (50–200). There were no intra-operative complications.

Peritoneal cavity entry occurred during fullthickness excision of lateral rectal wall mass with indeterminate pathology, therefore, an additional laparoscopy was required to visualize the suture line of the rectal wall and to perform an anastomotic leak test. In the postoperative period, 1 patient had urinary retention. Another patient presented with rectal bleeding, which was self-limiting and required no intervention or blood transfusion.

Mean length of stay was 2.5 d (1–4). No late postoperative complications were observed. The final pathologies (detailed in Table 1) of the specimens confirmed the diagnosis. These included tailgut cysts without malignancy, GIST lesions, and indeterminate lesions identified as follows: 1 lesion of endometrioma, a solitary rectal ulcer (SRU), a cloacogenic polyp, and 1 lesions found to be fibrotic tissue within the anastomosis without malignancy. Mean patient follow-up duration was 42 mo (14–80): All patients were disease free, except for the patient with the leiomyosarcoma who died due to lung metastasis at 37 mo following intervention.

DISCUSSION

TEM is a minimal invasive technique that has been proven to be an alternative for radical surgery aimed at rectal adenomas and early rectal cancer[14–18]. TEM can also, arguably, give the same surgical advantages for non-adenomatous rare lesions, unless these lesions are easily reached by instrumentation[19–23].

Although GIST lesions are the most common mesenchymal-derived neoplasm of the GI tract, rectal GIST lesions remain rare[12]. Surgical resection with negative margins is the recommended treatment for non-metastatic rectal GIST lesions. Given that nodal harvest is not necessary to cure GIST, local excision was proposed for these lesions[13].

TEM is an effective approach for the resection of rectal GIST due to the minimally invasive approach and rate of anal sphincter preservation[14]. Such advantages as shorter operating time, decrease blood loss, enhanced recovery, and low complication rate should be highlighted. Among our 3 GIST patients, we illustrated successful excision without perioperative complications and disease free without recurrence (local or distal) at 6 year follow-up.

NET lesions of the rectum are rare, and often present as asymptomatic incidental findings. Therapeutic recommendations remain controversial. ENETS guideline[15] recommend local treatment for rectal NET between 10 mm and 20 mm, followed by radical surgery, if local resection is incomplete. Therefore, the optimal local treatment procedure should combine highest local R0 rate with lowest possible complication rate. Studies using TEM for treatment of rectal NET lesions achieved very high R0 rate and conclude that TEM is a safe and effective procedure as long as the resection margins are negative[16–18].

In our cohort, the 3 NET patients had free surgical margins. Excisions were completed without complications or need for more invasive surgery. Of note, 1 patient did develop lung metastases at 1 year follow-up.

Rectal lipomas are rare benign lesions and mostly arise from the sub-mucosa. Patients may be asymptomatic or present with rectal bleeding, constipation and/or tenesmus[19]. In our cohort, both lipomas were submucosal. One presented with tenesmus and the other had rectal bleeding prior to diagnosis. Both patients had no recurrence at 5 and 7 years follow-up, respectively.

Retro-rectal or presacral lesions are rare and a homogeneous group of lesions with about two thirds of them being congenital. Most of these are cystic and benign. 10% are of neurogenic origin, 5%–10% are of bone origin and about 15% are from other origins, including metastasis. Surgical approach may be abdominal, posterior or combined. TEM facilitates the excision of the lesion in almost any place retro-rectally[20,21]. There are several series that illustrate use of TEM for excision of retro-rectal/pre-sacral cysts with excellent outcomes and complete excision of the lesions. In our cohort, all 5

Table 1 Study cohort

No	Age (yr)	Preop diagnosis	Final pathology	Size	Distance	Location	Op time	LOS
1	62	GIST	GIST	3	6	Post	65	2
2	58	NET	NET	1	8	Post	60	3
3	28	Indeterminate pathology	Solitary rectal ulcer	3	10	Ant	65	2
4	58	GIST	GIST	1.5	5	Rt post	90	2
5	66	Retro-rectal mass	Tailgut cyst	3	7	Post	110	2
6	54	Retro-rectal mass	Tailgut cyst	4	8	Post	150	3
7	40	NET	NET	1.5	9	Post	100	1
8	45	Retro-rectal mass	Tailgut cyst	3	7	Post	100	3
9	68	Submucosal lesion	Lipoma	6	8	Post	150	3
10	72	Leiomyosarcoma	Leiomyosarcoma	4	6	Ant	95	3
11	48	Retro-rectal mass	Tailgut cyst	2	6	Rt post	100	2
12	80	Indeterminate pathology	Rectal stenosis		7	Circ	50	2
13	54	GIST	GIST	2	11	Ant	50	3
14	25	Indeterminate pathology	Cloacogenic polyp	4	7	Lt	60	2
15	52	Indeterminate pathology	Endometrioma	4	13	Rt	200	4
16	74	Submucosal lesion	Lipoma	3	7	Lt	120	3
17	44	NET	NET	1	10	Rt ant	90	3
18	42	Retro-rectal mass	Tailgut cyst	3	7	Post	105	3

LOS: Mean length of stay; NET: Neuroendocrine tumor; GIST: Gastrointestinal stromal tumor.

retro-rectal lesions were identified as cysts. Complete excision with closure of the rectal defect was performed in all patients. None had abscess or fistula formation.

TEM is an effective treatment of anastomotic rectal stenosis. It has been described as an alternative to both abdominal resection of the anastomotic area and endoscopic dilatation[22]. In our cohort we describe a patient with rectal stenosis as a result of a mid-rectal anastomosis following anterior resection. We illustrated the use of TEM as a successful approach for anastomotic resection of the fibrotic ring and re-suturing. Rectal bleeding was noted on the 4th post-operative day, although was self-limiting with no need for blood transfusion. At 3-year follow up, the patient was asymptomatic and reported marked improvement quality of life.

Extra-pelvic endometriosis can affect the rectum and account for 15% of intestinal presentations. Our endometrioma patient had a solitary lesion in the upper rectum without pre-operative pathological diagnosis. TEM was performed with peritoneal entry requiring additional laparoscopy to ensure closure of the intraperitoneal rectum.

SRU syndrome (SRUS) is an uncommon chronic disorder with a wide range of endoscopic findings, clinical presentations and characteristic histopathological features. There is no clear consensus regarding SRUS management, because of its poorly understood pathogenesis and frequent association with various pelvic floor disorders. TEM technique may provide a safe and effective way for full thickness excision of the rectal wall with a 1-cm margin. TEM, as a surgical approach, allows for rectal resection with clear and detail view, specifically for rectal lesions such as SRUS[23].

Rectal leiomyosarcoma is an uncommon malignant tumor, accounting for less than 0.1% of all rectal malignancies[24]. While leiomyosarcoma can occur at any age, it predominantly affects individuals in their 5th and 6th decades of life, with a higher incidence in men.

Surgical intervention remains the primary treatment approach for anorectal leiomyosarcoma. Radical surgical approaches, such as anterior resection or abdominoperineal resection, are preferred over wide local excision in terms of local control. Of note, survival rates do not significantly differ between the two surgical modalities[25]. Neoadjuvant radiotherapy may enhance local control following resection. However, both local and distant recurrence are common and can manifest years after the initial resection. Unfortunately, the prognosis for anorectal leiomyosarcoma is generally poor, with reported 5-year survival rates ranging from 20% to 40%[26].

The histopathological characteristics associated with a poor prognosis include high mitotic activity, intra-tumoral necrosis, and larger tumor size. Moreover, efforts should be made to avoid positive margins during surgical resection, as they independently predict local recurrence[27].

Differential diagnosis that should be considered: fibromatoses, schwannomas, and GIST. An accurate diagnosis is of paramount importance for establishing the treatment modalities. Superficial biopsy specimens may not accurately represent the entire tumor mass, possibly leading to misdiagnosis of leiomyosarcomas. Therefore, full-thickness resection

of the tumor is the best diagnostic modality for precise diagnosis. Additionally, lymph node metastasis is rare in leiomyosarcomas (as reported in previous studies). Thus, lymphadenectomy is not warranted unless preoperative imaging reveals enlarged regional lymph nodes[26,28]. This data provides support for the roll of full-thickness local excision by TEM as a definitive surgical treatment for selected cases.

In our study, the patient who had rectal leiomyosarcoma died 4 years following surgery due to distant lung metastasis disease. No evidence of local recurrence was appreciate during the follow up period.

A cloacogenic polyp of the rectum, also known as a cloacogenic glandular polyp, is a rare type of benign polyp that originates from the cloacal remnants in the rectum. It is considered a developmental anomaly. The cloaca is a common channel during embryonic development that receives waste products from the gastrointestinal, urinary, and reproductive systems before they are expelled from the body[29].

Cloacogenic polyps typically occur in the lower rectum and are composed of glandular tissue resembling the lining of the cloaca. They are usually small in size and asymptomatic, but can occasionally cause rectal bleeding or mucus discharge. Cloacogenic polyps are generally benign, but in rare cases, they may exhibit dysplasia or progress to malignancy. Diagnosis of cloacogenic polyps is made through endoscopic examination, where the polyp's distinct glandular appearance can be visualized. Treatment typically involves complete removal of the polyp through endoscopic or surgical resection[30].

TEM is a safe and effective technique for removing such polyps that are not amenable for endoscopic resection. However, regular follow-up is recommended to monitor for recurrence or any potential malignant transformation, although the risk of malignancy is considered low.

In our study we had 1 patient with this type of pathology who was treated by TEM. No recurrence was observed during the follow up period.

CONCLUSION

It can be argued that TEM allows for reduced morbidity given its minimally invasive nature. Surgeons should be familiar with the technique but careful patient selection should be considered. TEM can be used safely for uncommon rectal and selected retro-rectal lesions without compromising outcomes. We believe that TEM should be reasonably considered as one of the surgical methods when treating rare lesions.

Limitations

The limitation of this study is the low power, given small sample size.. Although TEM as a surgical modality, has a potential benefit, no solid conclusion can be made from this study alone. Additional research is necessary, with more extensive and diverse sample size, to expand upon the findings of this study.

ARTICLE HIGHLIGHTS

Research background

Trans-anal endoscopic microsurgery (TEM) is a surgical method which is capable of resecting lesions up to 20 cm. It enables for excellent access, visualization of the surgical field and for precise full-thickness excision of rectal lesions.

The rectum, as well as the retro-rectal space, can be the origin of a heterogeneous group of uncommon neoplasms that range from aggressive malignancies to benign lesions that are almost always symptomatic. The treatment of this rare and heterogeneous group of lesions varies depending on the location and nature of the lesions; accordingly, certain lesions require radical trans-abdominal surgery, while others can be treated by a less aggressive approach.

Research motivation

Several studies suggest that TEM can also be an effective method for excision of uncommon lesion of the rectum and retro-rectal space.

Research objectives

The objective of this study is to report outcomes in patients undergoing TEM for rare and non-adenomatous rectal and retro-rectal lesions over a period of 11 years.

Research methods

Retrospective analysis was completed for all patients who underwent TEM procedure for non-adenomatous rectal lesion or retro-rectal mass from January 2008 to December 2019. The original Richard Wolf equipment was used to perform the procedure, and the procedure was performed according to the standard technique described specimens were pinned and marked for orientation by the surgeon. Patients resumed oral intake of liquid and soft diet on post-operative day one; with full diet being subsequently advanced, once tolerated. Patients were discharged once diet was well tolerated and no complications were identified.

Patients were evaluated at 3 wk post operatively. Follow ups were completed at 3 mo intervals for the first 2 years and then every 6 mo depending on the nature of the final pathology. Clinical examination and rectoscopy were performed

during each of the followup visits.

Research results

Out of 198 patients who underwent TEM during the study period, 18 (9%) had non-adenomatous (non-carcinomas or adenomas) rectal or retro-rectal lesions. Mean age was 47 years (27–81). The mean size of the lesions described was 2.9 mm (1–6 mm), with a mean distance from the anal margin of 7.9 cm (5–13 cm). Regarding the locations of the lesions in the rectal wall; 10 lesions were in the posterior wall, 4 were in the anterior wall, 3 in lateral position, and 1 patient had circular stenosis of rectum. Mean surgical time was 97.8 min (50–200). There were no intra-operative complications. Mean length of stay was 2.5 d (1–4). No late postoperative complications were observed. The final pathologies (detailed in Table 1) of the specimens confirmed the diagnosis. Mean patient follow-up duration was 42 mo (14–80). All patients were disease free, except for the patient with the leiomyosarcoma who died due to lung metastasis at 37 mo following intervention.

Research conclusions

It can be argued that TEM allows for reduced morbidity given its minimally invasive nature. Surgeons should be familiar with the technique but careful patient selection should be considered. TEM can be used safely for uncommon rectal and selected retro-rectal lesions without compromising outcomes. We believe that TEM should be reasonably considered as one of the surgical methods when treating rare lesions.

Research perspectives

Additional research is necessary, with more extensive and diverse sample size, to expand upon the findings of this study.

FOOTNOTES

Author contributions: Shilo Yaacobi D contributed to methodology, original draft preparation, and manuscript review and editing; Bekhor EY contributed to investigation, statistics, and manuscript review and editing; Khalifa M contributed to original draft preparation and manuscript review and editing; Sandler TE contributed to investigation and statistics; Issa N contributed to project administration, methodology, original draft preparation, and manuscript review and editing.

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