

Resection of multiple rectal carcinoids with transanal endoscopic microsurgery: Case report

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was successfully performed to remove the three lesions with accurate full-thickness resection followed by secured suture closure. The postoperative pathology revealed neuroendocrine tumors G1 (carcinoids) located within the submucosal layer without lymphatic or vessel infiltration. Both the deep and lateral surgical margins were completely free of tumor cells. The patient recovered quickly and uneventfully. No tumor recurrence was observed at the six-month follow-up. For the multiple small rectal carcinoids without muscularis propria or lymphatic invasion, transanal endoscopic microsurgery offers a reliable and efficient alternative approach to traditional laparotomy for select patients, with the added advantages of minimally invasive surgery.

Key words: Colorectal; Gastrointestinal surgery; Oncological surgery; Technical; Minimally invasive surgery

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Core tip: A rare case of multiple small rectal carcinoids being successfully removed using the transanal endoscopic microsurgery technique. On the basis of careful preoperative evaluation and detailed postoperative pathological examination, transanal endoscopic microsurgery offers a safe, reliable and efficient alternative approach to the traditional surgeries for select patients with multiple rectal carcinoids.

Abstract

Multiple rectal carcinoids are rare. Due to the unreliability of endoscopic polypectomy in treating these submucosal lesions, a laparotomy is usually performed. We present a case report on multiple rectal carcinoids with three carcinoid foci < 10 mm in diameter located in the mid-rectum. Preoperative examination showed the lesions to be confined to the submucosal layer with no perirectal nodal involvement. A transanal endoscopic microsurgery

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INTRODUCTION

With the rise in numbers of patients undergoing

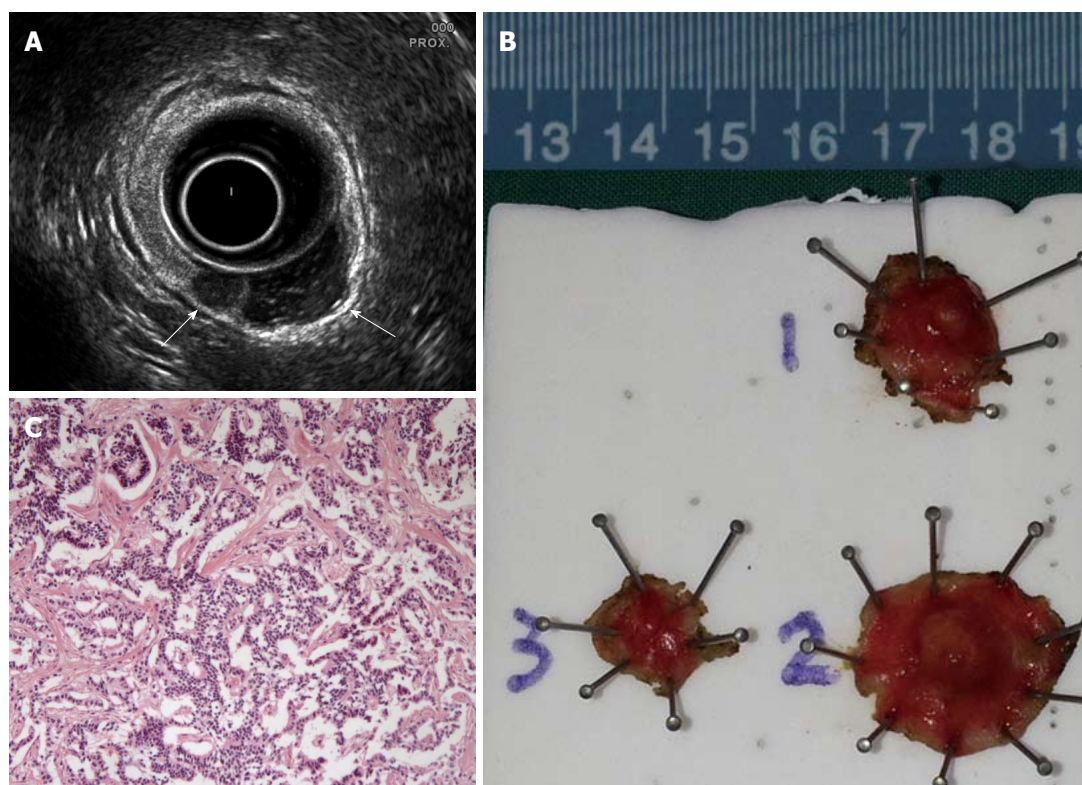


Figure 1 Ultrasound image, surgical specimens and pathological image of the carcinoids. A: Transrectal ultrasound image showing two hypoechoic nodules (arrow), 0.5 cm and 0.72 cm in maximum diameter (lesions 1 and 2, respectively), confined to the submucosal layer of the rectal wall; B: Surgical specimens of the three lesions (two carcinoids, specimen 1 and 2; and one scar site with residual tumor, specimen 3); C: The pathological image revealing neuroendocrine tumor cells within the submucosal layer (hematoxylin and eosin, $\times 100$).

endoscopy screening, the detection rate of early rectal carcinoids has increased notably in recent years^[1-4]. In most cases, rectal carcinoids occur in a singlet; however, multiple rectal carcinoids can occur and the reported incidence rates range from 2% to 4.5%^[5-8]. The consensus is that small rectal carcinoids (≤ 10 mm), without muscularis propria invasion, can be treated curatively using local excision since they rarely metastasize^[4,9,10]. Here, we present a case of multiple rectal carcinoids being successfully treated with local resection by transanal endoscopic microsurgery (TEM).

CASE REPORT

A 47-years-old Chinese male patient, presented with a year-long intermittent hematochezia, was admitted to our tertiary care center in October 2013. The signs of carcinoid syndrome were not observed. One month prior to his admission, the patient underwent a colonoscopy in a local hospital. The examination revealed three sessile nodules on the posterior wall of the rectum, 6 to 8 cm above the anal verge. The nodules, covered with yellowish discolored mucosa, 0.5 to 0.8 cm in diameter, were located within a relatively small area at 1 to 2 cm away from each other. During endoscopy, the smallest nodule was removed via snare polypectomy, and histological examination confirmed the diagnosis of rectal carcinoid. After one and a half months, the

patient attended our clinic for further treatment. A transrectal ultrasound was performed which identified two hypoechoic nodules, 0.5 and 0.72 cm in maximum diameter, confined to the submucosal layer of the rectal wall (Figure 1A). No enlarged perirectal lymph node was observed. Hence, the initial diagnosis of multiple rectal carcinoids was made. Because of the small tumor size (< 10 mm) and the absence of muscularis propria invasion and nodal involvement, local excision was performed using the TEM technique.

Rigid sigmoidoscopy was used to confirm locations of the three foci (including one scar site), followed by TEM under general anesthesia. The patient was placed in the lithotomy position with the lesions placed at the bottom of the operating field. The procedure was performed as previously described by Buess *et al.*^[11], using the Buess original TEM system (Richard Wolf GmbH, Knittlingen, Germany). After marking the resection area with coagulation dots by a needle cautery, ensuring a free margin area of 1 cm, the two submucosal tumors and the scar site of the third lesion were removed one at a time with full-thickness excision. Defects in the rectal wall were irrigated and closed using the running sutures of 3/0 absorbable monofilaments (Figure 2). The operation was completed within 40 min with a proximate blood loss of 10 mL.

No analgesic was required postoperatively. An elementary diet was initiated on the second day, and the patient was

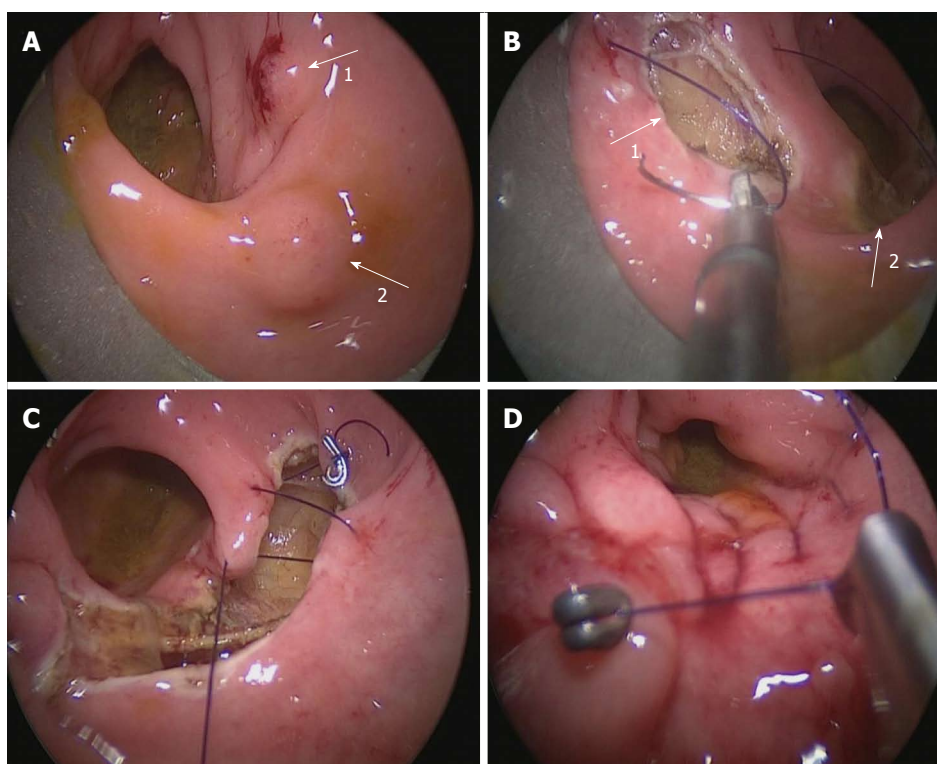


Figure 2 Details of the resection of multiple carcinoids by transanal endoscopic microsurgery. A: Rectoscopic view of the two rectal carcinoids (lesions 1 and 2, see arrows 1 and 2), lesion 3 is not included in this view; B: After removing two carcinoids (lesions 1 and 2, see arrow 2) and one scar site (lesion 3, see arrow 1) with full-thickness excision, defects in the rectal wall (arrows 1 and 2) were closed using running sutures; C: Lesion 1 and 2 defects were adjacent and taken as a whole during the suturing; D: Completion of the suturing of the defect of lesions 1 and 2.

discharged two days after the surgery with an uneventful recovery. At the six-month follow-up, the patient was well with no carcinoid recurrence.

The postoperative pathology revealed two neuroendocrine tumors of 5 mm and 7 mm in diameter (specimen 1 and 2). In specimen 3, the residual neuroendocrine tumor cells were identified in the submucosal layer (Figure 1B and C). All three tumors were located within the submucosal layer with no lymphatic or vessel infiltration. Both the deep and lateral surgical margins were completely tumor-free in all specimens. Tumor cells showed a low cell proliferation index (Ki-67 < 2%). Immunohistochemistry study showed that tumor cells were positive for CD56 (NK-1) and Syn, and negative for CgA and p53 staining. The final diagnosis was multiple rectal neuroendocrine tumors G1 (carcinoids).

DISCUSSION

Multiple carcinoid tumors develop rarely in the rectum, with the incidence around 2% to 4.5%^[5-8]. Sporadic reports of such cases can be retrieved from the literature written in the English language. Table 1 shows a summary of six multiple rectal carcinoids cases from five reports written in the English language. Among these six cases, four were extremely rare cases containing numerous endocrine cell micronests in the rectal wall^[12-14], necessitating the radical resection with lymph node dissection. However, due to the small sample size (four out of six cases) it is not appropriate to conclude that multiple

rectal carcinoids are often accompanied with numerous endocrine cell micronests.

Small rectal carcinoids (≤ 10 mm) without muscularis propria invasion can be curatively treated using local excision, due to the low metastasis rate^[4,9,10]. Endoscopic polypectomy has been widely used for treatment of rectal carcinoids. However, most carcinoid tumors arise from the deep portion of the epithelial glands, penetrating the muscularis mucosa into the submucosal layer where the carcinoids form a nodular lesion^[9]. Hence, the intrinsic limitations of the conventional endoscopic polypectomy result in a high chance of incomplete resection^[9,15]. In the case of our patient, the postoperative pathologic result showed that lesion 3 had not been completely removed by endoscopic polypectomy. More advanced endoscopic resection methods, including endoscopic mucosal resection with cap, endoscopic submucosal resection with band ligation, and endoscopic submucosal dissection, or surgical local excision can be considered as more appropriate treatment methods^[9].

Since its introduction by Buess *et al.*^[16] in 1983, TEM has emerged as an effective minimally invasive surgery for local resection of rectal lesions. With an average distance from the anal verge of about 7-8 cm^[2,9], most small rectal carcinoids without metastasis are ideal candidates for local resection under TEM. This technique enables full-thickness excision and ensures accurate resection with sufficient margins by applying the delicate instruments under the superior visualization. In addition, TEM allows suturing

Table 1 Summary of cases of multiple rectal carcinoids reported in the English literature

Source	Sex	Age, yr	n	Size of carcinoids, mm	Surgical approach	Presence of micronests	Lymph node metastasis
Kato <i>et al</i> ^[12]	M	61	52	1-6	NA	Yes	NA
Maruyama <i>et al</i> ^[13]	M	52	5	4-10	AR	Yes	No
Okamoto <i>et al</i> ^[19]	M	54	4	< 6	ESMR-L	No	NA
Haraguchi <i>et al</i> ^[20]	M	69	30	< 10	APR	No	No
Sasou <i>et al</i> ^[14]	M	51	7	< 8	APR	Yes	Yes
	M	58	3	< 7	AR	Yes	Yes

APR: Abdominoperineal resection; AR: Anterior resection; ESMR-L: Endoscopic submucosal resection with band ligation; M: Male; NA: Not available.

of the rectal wall defects after tumor resection, thus securing sufficient excision without worrying about bowel perforation. In comparison with endoscopic resection methods, including advanced techniques of endoscopic mucosal resection with cap and endoscopic submucosal resection with band ligation, TEM enables a much larger extent of resection, ensuring more satisfactory oncological results for lesions with malignant potential.

In our case, three rectal carcinoids laid adjacent to each other within a relatively small area. It only took a small adjustment of the rectoscope to accomplish excision of all three lesions without changing the patient's position. After removal of the tumors, adjacent defects were regarded as a whole and sutured together. The operation was, therefore, performed conveniently and efficiently using the TEM technique. However, for cases of multiple carcinoids containing numerous micronests of tumor cells in the rectum, an extremely rare condition^[13,14], anterior resection or even abdominoperineal resection should be performed due to the inability of the naked eye to detect diffusely scattered tumor foci. For our patient, postoperative pathology excluded such a condition.

Although the majority of the small rectal carcinoids can be radically removed by local excision, some authors have reported that rectal carcinoids no larger than 10 mm may still spread to perirectal lymph nodes, with reported incidence rates of 7% to 9.7%^[10,17]. Before making a local excision by TEM technique, we routinely perform the transrectal ultrasound to assess the depth of tumor invasion and evaluate the status of perirectal lymph nodes. Prior to surgery, a careful examination with rigid sigmoidoscopy is necessary to determine the exact location and orientation of the lesion, which is important for planning of the patient's positioning during the surgery, allowing the lesion to sit at the 6-o'clock position and to facilitate the TEM operation.

Careful postoperative histological examination of specimens was required to evaluate the tumor size, the depth of invasion, the margin status, and the presence of risk factors of metastasis^[2,10,18]. Tumors with invasion to muscularis propria, angiolymphatic invasion presence, or the increased indices of cell proliferation (such as Ki-67) should be treated with a salvage radical resection, or close follow-up should be performed.

To our knowledge, this is the first reported case of multiple rectal carcinoids being managed by TEM. In this case, high standard local tumor resection of multiple

lesions was performed efficiently with minimal operative trauma; the patient recovered smoothly and quickly, further indicating the advantages of the TEM technique. Although this report presents only one case with a short follow-up period, we strongly believe that repeated TEM procedures for treating multiple rectal carcinoids will provide surgeons with invaluable experience, insight and knowledge of the disease and the best methods for treating this condition.

COMMENTS

Case characteristics

The patient was admitted with a year-long intermittent hematochezia.

Clinical diagnosis

The clinical diagnosis of multiple rectal carcinoids was made according to the result from colonoscopy that revealed three small submucosal nodules covered with yellowish-white mucosa on the rectal wall.

Differential diagnosis

The small nodules on the rectal wall should be differentiated from the rectal adenomas, inflammatory polyps, gastrointestinal stromal tumors, lipomas, hamartomas, and rare diseases such as juvenile polyps, ganglioneuromatosis, etc.

Laboratory diagnosis

Laboratory tests revealed no abnormal results.

Imaging diagnosis

The transrectal ultrasound showed small hypoechoic nodules being confined to the submucosal layer of the rectal wall.

Pathological diagnosis

Postoperative pathology revealed neuroendocrine tumors with a low cell proliferation index (Ki-67 < 2%).

Treatment

The three lesions were surgically removed through local excisions using the transanal endoscopic microsurgery technique (TEM).

Related reports

Five cases of multiple rectal carcinoids reported in articles written in the English language were reviewed and summarized.

Experiences and lessons

For multiple small rectal carcinoids without muscularis propria or lymphatic invasion, TEM offers a reliable and efficient alternative approach to traditional laparotomy for select patients, with advantages of minimally invasive surgery. Careful preoperative evaluation and detailed postoperative pathological examination are mandatory to guarantee the radical removal of the tumor cells.

Peer-review

Most reviewers found this topic interesting. The manuscript was praised for being well-written and structured. Reviewers also recommended a summary of the previous reports on multiple rectal carcinoids.

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