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Local excision of rectal schwannoma using transanal endoscopic microsurgery: A case report

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ABSTRACT

INTRODUCTION: Schwannoma is a neoplasm originating from the neural crest cells (schwann cells) that form nerve sheaths. These tumors are thought to be benign with little risk of malignant transformation. They rarely affect the gastrointestinal tract, and primary rectal involvement is extremely rare. Until 2013, only 11 cases of anorectal schwannoma have been reported. Optimal surgical treatment of rectal schwannoma has not been established.

PRESENTATION OF CASE: We herein describe a 70-year-old woman with a submucosal tumor arising from the posterior wall of the rectum with features mimicking a gastrointestinal stromal tumor. After discussing the operative procedures and obtaining written informed consent, we attempted local excision of the tumor using a transanal endoscopic microsurgery (TEM). The tumor was proved to be S-100 positive schwannoma on immuhistochemical studies. Her postoperative course was uneventful, and there is no evidence of tumor recurrence as of 6 months after surgical excision.

DISCUSSION: An extremely rare rectal schwannoma was successfully treated using a TEM without compromising anorectal function.

CONCLUSION: TEM is a feasible approach for local excision of rectal tumors with low risk of malignancy.

1. Introduction

Schwannoma is a nerve sheath tumor which frequently occurs in the intracranial acoustic nerve and spinal nerves, while involvement of the gastrointestinal tract is uncommon.[1–4] Although gastrointestinal shwannoma is commonly detected as a submucosal tumor (SMT), definitive preoperative diagnosis is often difficult. Although radical resection is required for malignant schwannoma presenting with a huge tumor, radical resection with a wide surgical margin for rectal schwannoma should be avoided if possible, because there have been no report on malignant rectal schwannomas of the rectum in the English literature.[5] We herein report a rare case of rectal schwannoma which was successfully treated by transanal endoscopic microsurgery (TEM) and review the literature.

2. Case report

A 70 year-old female presented with a SMT of the rectum. She had no complaints, and the tumor was discovered incidentally as a SMT in the posterolateral wall of the rectum, 10 cm from the anal verge (Fig. 1A) when she underwent colonoscopy for positive fecal occult blood tests on a periodic health checkup. Endoscopic ultrasound (EUS) showed an 18 mm × 18 mm hypoechoic mass (Fig. 1B), and EUS-guided biopsy was performed for immunohistochemical analysis, which revealed weakly positive staining for CD117. Therefore, the tentative pathological diagnosis was gastrointestinal stromal tumor (GIST). On magnetic resonance imaging (MRI), the mass was diffusely hypointense on T1-weighted and rather hyperintense on T2-weighted images (Fig. 2). The serum levels of carcinoembryonic antigen (CEA) and CA19-9 were within normal ranges.

After discussing the operative procedures and obtaining written informed consent, we attempted local excision of the tumor using transanal endoscopic microsurgery (TEM) (Fig. 3A). Macroscopic examination of the resected specimen confirmed a solid tumor with a size of 28 mm × 23 mm × 20 mm. The cut surface was uniformly elastic, soft and exhibited bright appearance (Fig. 3B). The resection margin was free of the tumor. The histopathological examination

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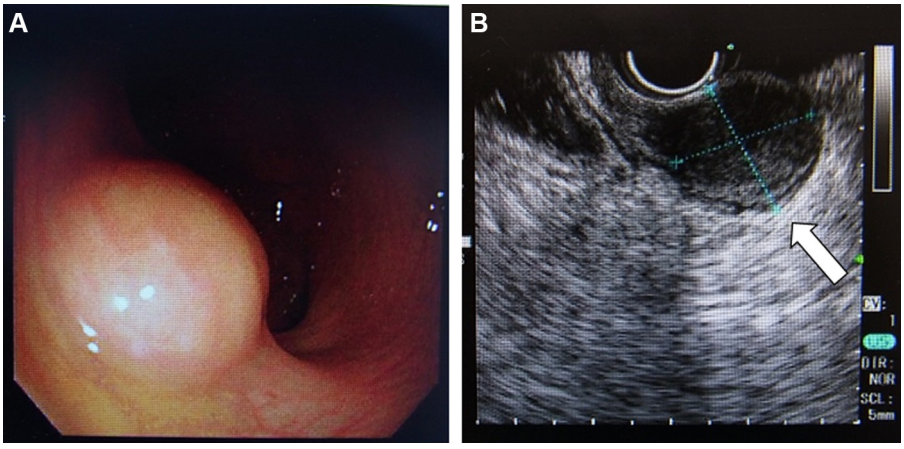

Fig. 1. (A) Colonoscopy disclosed a SMT in the rectum. (B) Transrectal US demonstrated a hypoechoic mass 18 mm × 18 mm in diameter originating from the submucosal layer (arrow) in the rectum.

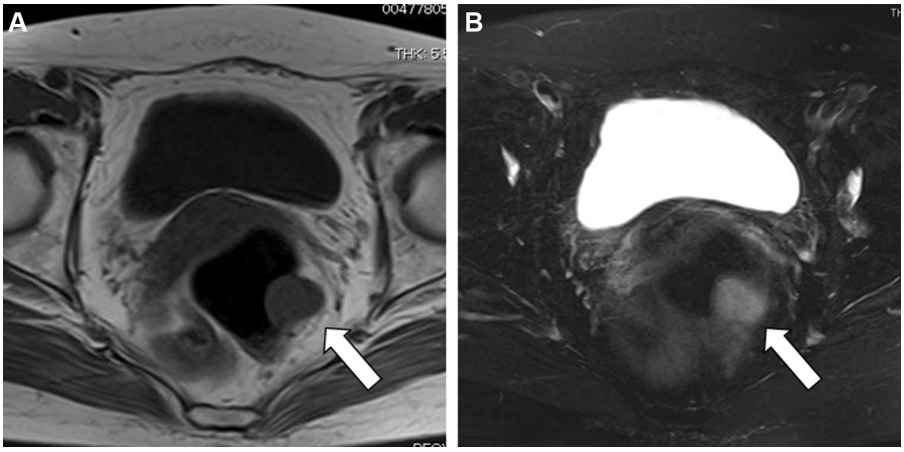

Fig. 2. Magnetic resonance imaging. (A) Transverse T1-weighted image demonstrated a homogeneous mass without signal intensity (arrow). (B) Transverse T2-weighted image demonstrated a heterogeneous mass with high signal intensity (arrow).

of the tumor revealed a well-encapsulated tumor surrounded by lymphoid tissue, which was located mainly in the submucosa of the rectum. It was composed of clusters and palisading of spindle-shaped vesicular-nucleated cells. Mild nuclear pleomorphism and mitotic activity of 1/10 HPF were noted. Immunohistochemical staining revealed that the tumor cells were positive S-100 and negative for CD34 and CD117 (Fig. 4). The tumor was therefore diagnosed as rectal schwannoma rather than GIST. The patient had no

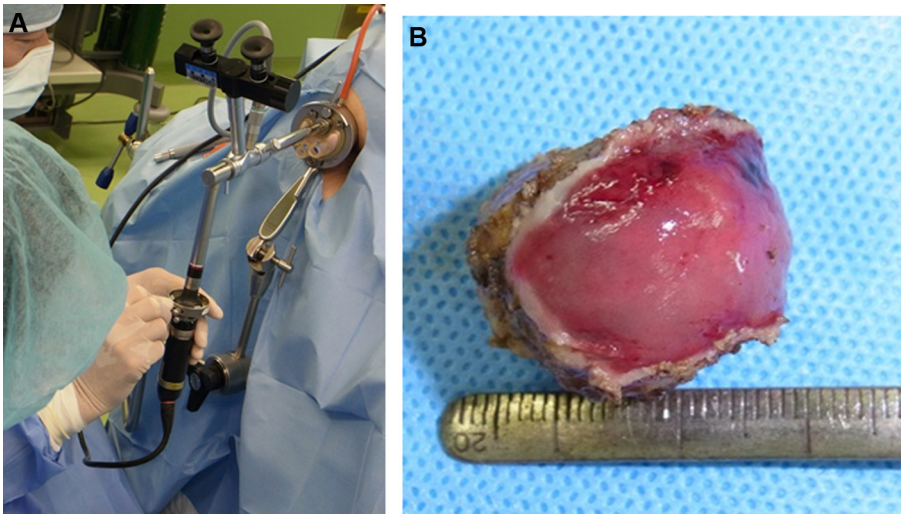

Fig. 3. (A) An intraoperative photograph of transanal endoscopic microsurgery (TEM). (B) Macroscopic view of the resected rectal SMT.

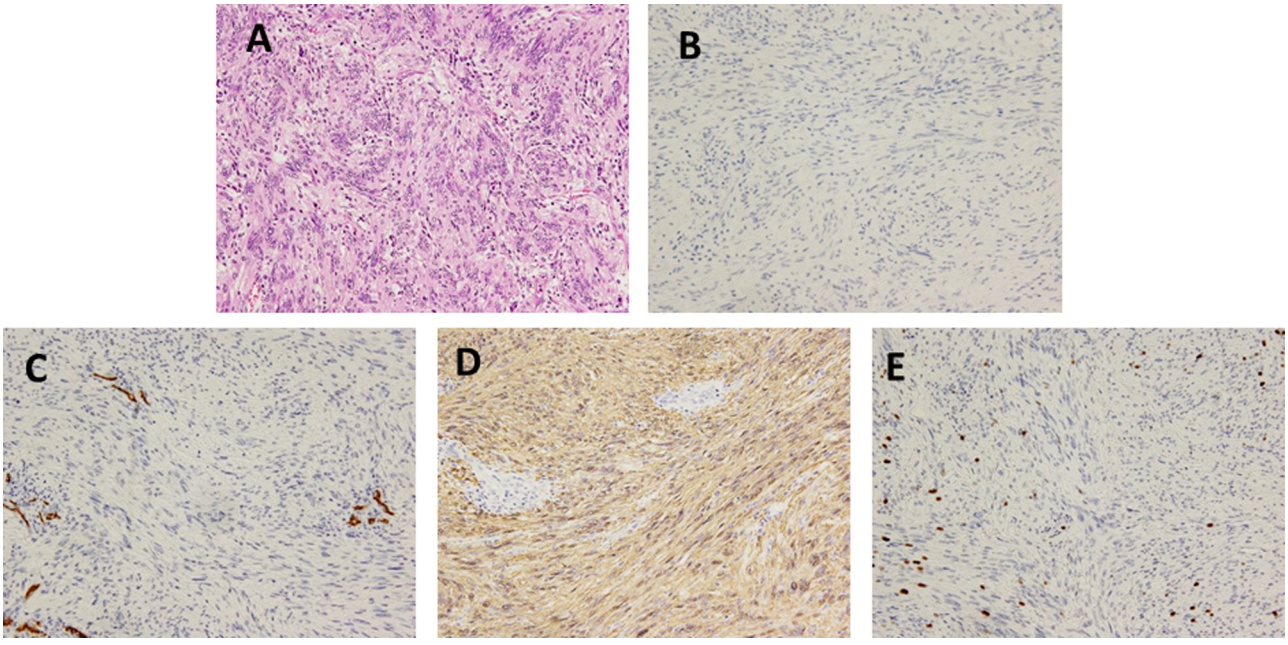

Fig. 4. Microscopic images. (A) Clusters and palisading of spindle-shaped vesicular-nucleated tumor cells are seen (H–E stain, ×200). (B) and (C) Negative immunoreactivity in the tumor cells for CD117 (B, ×200) and CD34 (C, ×200). (D) Immunohistochemistry indicated strong staining for S-100 (×200). (E) MIB-1 index was 3% (×200).

complications and was discharged 5 days after surgery, and shows no evidence of tumor recurrence as of 6 months after surgery.

3. Discussion

Schwannoma of the gastrointestinal tract is uncommon and occurs most frequently in the stomach, while the colon and rectum are rarely affected.[1–5] Colonic schwannoma was first reported by Lamy et al.[6] A number of colonic schwannoma cases have been reported since then, while only 11 cases of anorectal schwannoma to our knowledge have been reported in the English literature.[5] Although rectal schwannoma usually grow very slowly without presenting symptoms, rectal bleeding, colonic obstruction, defecation disorders and pain have been reported,[7,8] for which preoperative diagnosis is often difficult, and result in radical resection based on a suspicion of malignancy.[7] In the current case, although preoperative biopsy of the SMT suggested a GIST, insufficient amount of sample from biopsy may account for false positive results for CD117. Complete excision with a negative microscopic surgical margin is the treatment of choice for schwannoma of the gastrointestinal tract, since their response to chemotherapy and radiotherapy is uncertain.[8,9] Incomplete resection of schwannoma led to unacceptable tumor recurrence rates of 30% locally and 2% in distant organs, meaning that the risk of malignant transformation should not be ignored.[4,10,11]

TEM was first reported by Buess as a new minimally invasive and anal function-preserving technique.[12] This procedure is primarily used for local excision of selected benign and low-grade malignant tumor of the lower, middle and upper rectum via the anus.[13] TEM allows a full-thickness resection with wide margins and an intact capsule in tumors up to 5–20 cm from the anal verge[13] while unsuitable for lesions close to the anal verge. Currently, TEM is used for surgical resection of SMTs such as GIST in the rectum.[15]

TEM has several benefits over more invasive anterior resection, including postoperative complications, morbidity rate and length of stay.[12–15] Appropriate use of TEM for benign rectal tumors and rectal cancer limited to the mucosa under supervision of experienced surgeons would benefit the patient.

4. Conclusion

Although extremely rare, schwannoma needs to be included in the differential diagnosis of rectal SMT. Local excision using a TEM is a feasible surgical approach for benign rectal tumors such as small GIST and schwannoma.

Conflict of interest

None.

Funding

None.

Ethical approval

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in Chief of this journal on request.

Author contributions

Toshiaki Suzuki contributed to data collections and wrote the manuscript.

Takenori Hada, Katsuhito Suwa and Tomoyoshi Okamoto participated in diagnosis and treatment of the patient.

Tetsuji Fujita and Katsuhiko Yanaga assisted manuscript preparation.

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