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Case report

Rectal submucosal tumor-like lesion originating from intestinal tuberculosis

Toshiyuki Yanagida[1], Masatoshi Oya[1], Naoto Iwase[1], Takashi Okuyama[1], Haruhiko Terada[1], Katsumi Sasaki[1], Shuichi Akao[1], Hiroshi Ishikawa[1], and Hideaki Satoh[2]

Departments of [1]Surgery and [2]Pathology, Koshigaya Hospital, Dokkyo University School of Medicine, 2-1-50 Minami-Koshigaya, Koshigaya, Saitama 343, Japan

Abstract: We present the case of a 55-year-old man who underwent transsacral local excision for a rectal submucosal tumor-like lesion suspected to originate from tuberculosis. The lesion, 2 cm in size, was found incidentally in the posterior wall of the lower rectum during anal fistulectomy. The lesion was apart from the primary crypt of the anal fistula. Barium enema and colonoscopy revealed a protuberant submucosal growth with a shallow depression of the overlying mucosa. Although computed tomography and magnetic resonance imaging showed a well defined round mass within the rectal wall, digital rectal examination suggested extramural origin. Since repeated endoscopic biopsies were negative, we selected the transsacral approach for excisional biopsy to achieve histological diagnosis. The lesion was confined to the rectal wall and the full-thickness rectal wall was excised. Histologically, a foreign-body granuloma with acute inflammation was the main component of the lesion. Caseating granulomas and Langhans' giant cells, consistent with tuberculosis, were also found.

Key words: rectal submucosal tumor-like lesion, anal fistula, tuberculosis, foreign-body granuloma

Introduction

Typical intestinal tuberculosis has become rare in recent years since the advent of effective antituberculous chemotherapy. However, tuberculosis sometimes shows non-specific or atypical clinicopathological features,[1,6] and *Mycobacterium tuberculosis* can grow in the anorectal region in forming an anal fistula.[7,8] Here, we present a case of rectal submucosal tumor-like lesion suspected to have been formed in the course of tuberculosis.

Case report

A 55-year-old man was admitted to our hospital in April 1996 for detailed examination and treatment of a mass lesion in the lower rectum. He had had an anal fistula for 2 years. He also had hemorrhoids, for which rectal suppositories had been given. He was found to have the mass lesion during fistulectomy performed at another hospital. The rectal tumor itself was asymptomatic. He did not have a history of pulmonary tuberculosis or of medical treatment for the condition. He had a 20-year history of diabetes mellitus, for which dietary control had been the main treatment. His family history was unremarkable.

On rectal digital examination, a sessile elastic hard mass, measuring 2 cm in diameter, was palpated just above the dentate line on the right-posterior wall (7 o'clock position) of the lower rectum. The tumor was fixed to the rectal wall. As the operation scar of the fistulectomy was in the left-posterior (4 o'clock) position, the tumor was considered to be unrelated to the anal fistula. Physical examination was otherwise unremarkable. Hematological examination results were within the normal range. Fasting blood sugar was 180 mg/dl and hemoglobin A1c was 7.1%. Glucose tolerance test later disclosed a diabetic pattern. Other serological parameters and tumor markers were all within normal limits. Urinalysis showed a positive reaction for sugar.

Chest and abdominal X-ray films showed no abnormal findings. Barium enema showed a hemispherically elevated lesion on the posterior wall of the lower rectum (Fig. 1). No other abnormalities were detected throughout either the large bowel or the ileocecal

Offprint requests to: T. Yanagida

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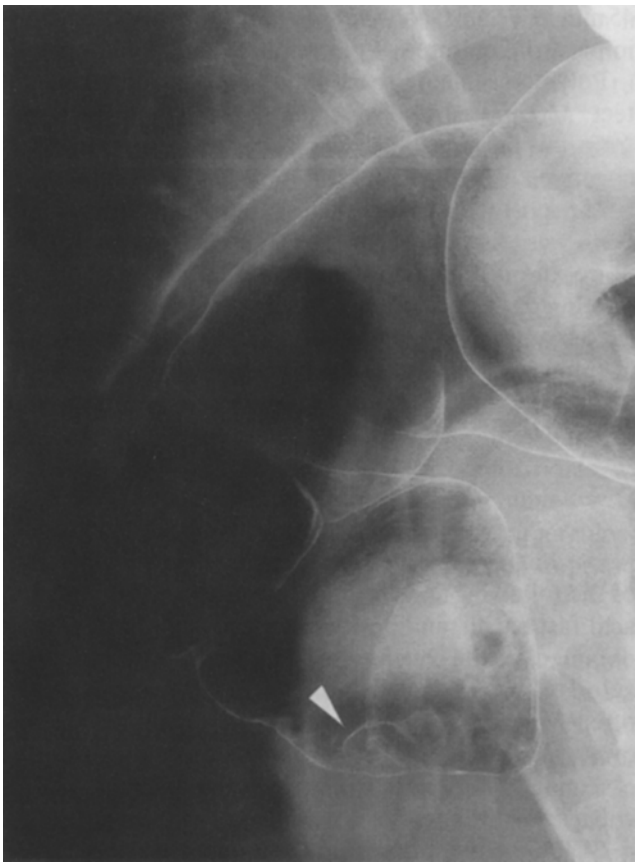

Fig. 1. Double-contrast barium enema showed an elevated mass lesion on the posterior wall of the lower rectum (*arrowhead*)

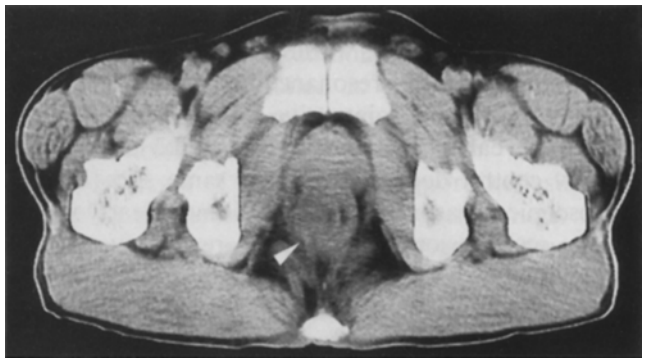

Fig. 2. Computerized tomography revealed a round mass in the right posterior wall of the rectum. The density of the mass was slightly low and its margin was clear (*arrowhead*)

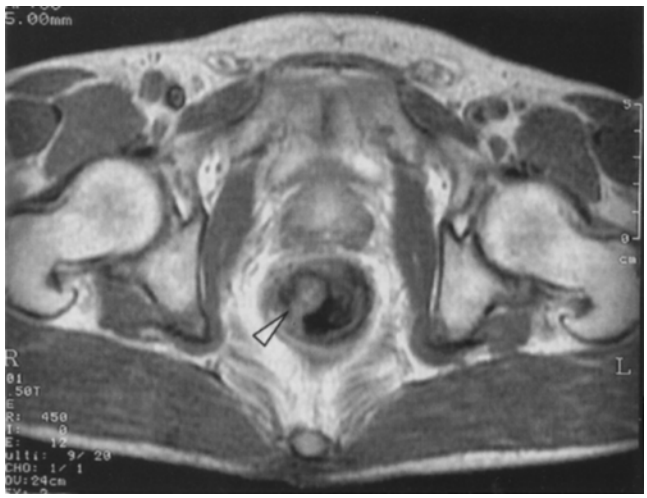

Fig. 3. Magnetic resonance imaging showed a mass with an isointense signal (*arrowhead*)

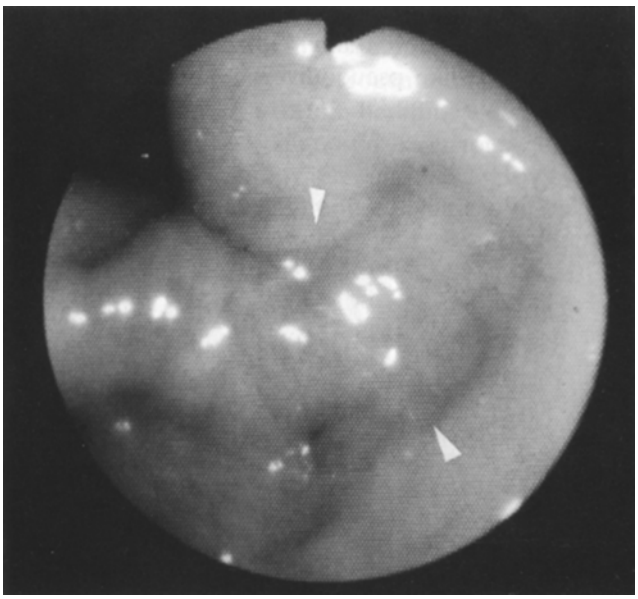

Fig. 4. Colonoscopy revealed a submucosal tumor-like lesion with a shallow central depression in the posterior wall of the rectum (*arrowheads*) (under J-turned view)

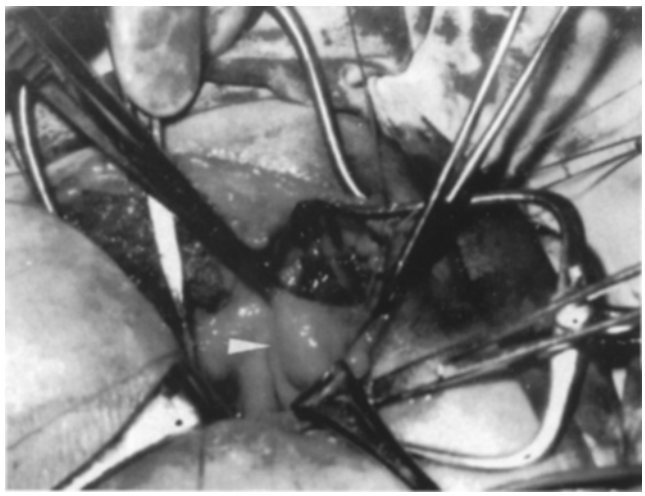

Fig. 5. Full-thickness rectal wall including the tumor was exicsed by transsacral local excision. The lesion is viewed from the lumen of the rectum (*arrowhead*)

region. Computerized tomography (CT) revealed a well-defined round mass within the posterior rectal wall (Fig. 2). On magnetic resonance imaging (MRI) study, the tumor had an isointensive signal (Fig. 3). Colonoscopy revealed a submucosal tumor-like lesion with a shallow central depression at the same site (Fig. 4). Endoscopic ultrasonophy (EUS) did not clearly visualize the mass lesion. Only normal rectal mucosa was obtained with repeated endoscopic biopsy.

As we could not exclude the possibility of malignant submucosal tumor, we performed transsacral local excision of the tumor, on June 17. With the patient under general anesthesia, we made an oblique skin incision along the right side of the sacrum, and approached the posterior wall of the rectum after division of the levator muscle. The tumor was confined to the rectal wall, and the external anal sphincter was intact. We excised a portion of full-thickness rectal wall that included the tumor and closed the defect with single-layer interrupted sutures (Fig. 5). We then approximated the levator muscle and closed the skin with a suction drain at the retrorectal space.

Macroscopically, the cut surface of the tumor was almost homogeneous, yellow-white in color, elastic in consistency, and clearly demarcated.

Histological examination of the resected specimen revealed caseating granulomas, Langhans's giant cells, and peripheral collars of inflammatory cells (Fig. 6a). An oily-drop appearance and scattered phagocytosis were observed in the submucosal layer (Fig. 6b). However, no evident foreign body was identified in the phagocytic cells (Fig. 6c).

From these findings, the tumor was suspected to be an inflammatory granuloma due to tuberculosis. Postoperatively, purified protein derivative (PPD) skin test gave an induration of 10 × 8 mm and erythema of 50 × 45 mm in 48 h. *Mycobacterium tuberculosis* was negative in bacterial cultures of gastric juice and stool.

Polymerase chain reaction (PCR), carried out to detect *Mycobacterium tuberculosis* in DNA extracted from the paraffin block of the resected specimen was negative.

The postoperative course was uneventful. The patient was discharged on the 17th postoperative day. Since the PPD test was strongly positive, isoniazid (200 mg/day) and rifampicin (450 mg/day) were given for 6 months, as suggested by a thoracic internist at our hospital. No evidence of recurrence of the tumor has been observed for 6 months' course.

Discussion

According to the criteria of Paustian et al.[1] at least one of the following specific findings: caseating granulomas, acid-fast bacilli, and positive bacterial cultures, must be obtained for the conclusive diagnosis of intestinal tuberculosis. The absence of reliable evidence, therefore, often makes a diagnosis of tuberculosis difficult. However, the resected specimen of our patient showed caseating granulomas, from which the diagnosis of *Mycobacterium* tuberculosis was made.

PCR examination of DNA extracted from the resected specimen was negative. The DNA may have been damaged because the specimen had been fixed with formalin before PCR.[9] If tuberculosis is suspected preoperatively, a fresh specimen obtained during operation should be used for PCR.

Intestinal tuberculosis usually presents with mucosal ulceration, typically occurring as skip lesions that tend to develop circumferentially. Deformity of bowel wall, such as clover-like pseudodiverticulosis, follows ulcer

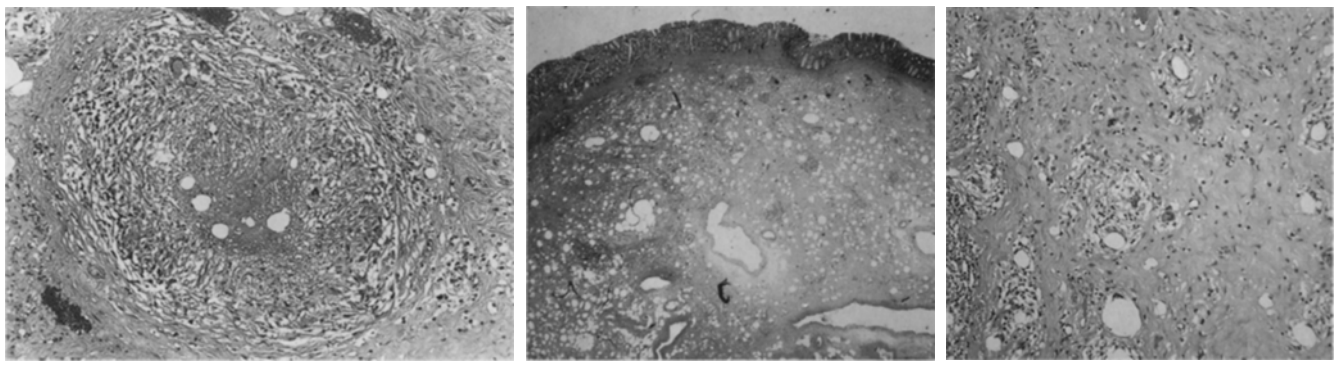

Fig. 6a–c. Histological examination. **a** Caseating granulomas and Langhans' giant cells were demonstrated. H&E × 200. **b** Low-power view of oily-drop appearance of the submucosal layer. H&E × 10. **c** Higher-power view of the oily-drop appearance with scattered phagocytosis. No foreign body was detected in the phagocytic cells. H&E, × 200

healing. The macroscopic appearance of the lesion in our patient is was so different from the usual presentation of intestinal tuberculosis that the possibility of tuberculosis was not borne in mind preoperatively.

The mechanism by which the tumor was formed in our patient is speculative. The tubercle may have damaged the surrounding tissue, leading to necrosis and granulomatous reaction. If this was the case, the foreign-body granuloma would be the result of an autoimmune inflammatory reaction. However, since the tubercle usually grows non-expansively, damage to the surrounding tissue seems unlikely. Moreover, the oily-drop appearance of the granuloma cannot be explained by this process. Considering the patient's longstanding use of suppositories for hemorrhoids, we believe that oily from the suppositories may have been the foreign body. If this was the case, the tuberculosis infection would have been only the first step in granuloma formation, followed by a secondary foreign-body reaction to the oily suppository.

Submucosal tumors of the rectum are uncommon.[7] Tumors of smooth muscle and lymphoid tissue, vascular hamartomas, lipomatous and neurogenic tumors, and other miscellaneous neoplastic tumors arise as nonepithelial tumors of the rectum. Each tumor has clinical and morphological characteristics which enable preoperative differential diagnosis.[7] Tumor of smooth muscle, leiomyoma in particular, is the commonest nonepithelial tumor of the rectum, and is usually red. Tumors of lymphoid tissue usually present in the lower third of the rectum, and are mostly sessile and rarely ulcerated. Vascular hamartomas, particularly lymphangiomas, are soft and their cut surfaces show cystic spaces from which lymph may exude. Lipomatous tumors, which rarely arise within the rectum, are soft and show a positive cushion sign. Neurogenic tumors are commonly associated with certain hereditary diseases.

Preoperatively, we diagnosed leiomyoma, based on the findings of endoscopy, CT, and MRI. However, we were not able to make a preoperative histological diagnosis and we did not consider the possibility of granuloma. We thought that we should perform an excisional biopsy of the full-thickness of the rectal wall including the tumor.[10] We chose transsacral excision as the surgical procedure to achieve this aim, as the transsacral approach gives a good view of the posterior wall of the lower rectum as well as its surrounding tissue. We admit that transsacral local excision would not have been performed for this patient if we had preoperatively diagnosed inflammatory granuloma. Although repeated endoscopic biopsies failed to clarify the pathology, needle biopsy or transanal local excision may have been a better alternative.

Tuberculoma of the lung is a tumor-like lesion consisting of granuloma enclosed by collagenous tissue. The tumor in our patient may have been a tuberculoma of the rectum. In the Japanese literature,[11] we found a report of a patient who had a submucosal tumor-like lesion, suggestive of tuberculoma, in the transverse colon. However, we found no reports of similar lesions in the rectum in our search of Japanese and English language literatures. The lesion in our patient was unique, so that we did not include tuberculosis in the preoperative differential diagnosis.

In conclusion, the tumor presented in this report allows us to recognize a unique clinical course of intestinal tuberculosis.

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