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Submucosal Dermoid Cyst of the Rectum: Report of a Case

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Abstract: Despite the relatively common incidence of sacrococcygeal dermoids, rectal cysts are uncommon. We report the case of a submucosal dermoid cyst occurring in the rectum. A 30-year-old woman visited the Gynecology Department because of pregnancy. A pelvic tumor was accidentally found during the checkup after miscarriage. A barium enema showed an anterior shift of the rectum by the presence of the tumor. Computed tomography and magnetic resonance imaging revealed a tumor located posterior to the rectum occupying almost the entire pelvic cavity, and the tumor was resected. The tumor was located in the submucosal layer of the posterior rectal wall and was well circumscribed. The resected tumor was a cyst entirely covered with a fibrous and firm capsule, which was filled with an amorphous white creamy substance. The histological findings showed the cyst consisting of a keratinizing stratified squamous epithelium with sebaceous gland and hair follicles, which was compatible with benign cystic teratoma. Primary rectal teratoma is very rare and only 36 cases have been reported in the literature worldwide. Furthermore, while the majority of cases were polypoid-shaped dermoid cysts protruding into the rectal lumen, only 3 cases were submucosal dermoid cysts. Therefore, such cases are considered to be extremely rare.

Key Words: dermoid cyst, rectum

Introduction

Although teratomas involving the ovary are frequently found, teratomas which originate in the gastrointestinal tract are uncommon. In contrast, postanal and/or sacrococcygeal dermoids are relatively common. In particular, rectal cysts, which comprise one type of teratoma, are also uncommon and only 36 cases have been reported in the worldwide literature. Among the reported cases of rectal teratoma, the majority tends to be of the pedunculated type, which causes such clinical symptoms as a protrusion of the tumor from the anus. In contrast, submucosal teratomas of the rectum are extremely rare and no particular symptoms appear before they are diagnosed. We describe herein the case of a dermoid cyst which arose in the submucosal layer of the rectum and was histologically proven to be a submucosal dermoid cyst.

Case Report

A 30-year-old woman visited the Gynecology Department of our hospital when she was 5 weeks pregnant in July 1990. She received a periodic ultrasound examination for the fetus at the department, during which time a pelvic tumor was accidentally found by ultrasound and was diagnosed to be an ovarian tumor. The patient was then admitted to the Gynecology Department because she was diagnosed to have an abruption of the placenta (abruptio placentae), and intrauterine fetal death was confirmed. She underwent a resection of the dead fetus on November 19, 1990. She was then referred to the Department of Surgery for a detailed examination of the pelvic tumor on October 5, 1990.

The patient had neither any particular symptoms related to the pelvic tumor nor a history of abdominal pain, constipation, or rectal bleeding. Her blood chemistry showed no abnormality and the serum levels of the carcinoembryonic antigen and CA19-9 were within normal range. An ultrasound examination revealed the tumor to be located in Douglas' pouch and measured 8.9 $\times$ 7.7 $\times$ 6.0 cm in size. The echo level was relatively homogeneous with an association of a high-echoic capsule. An ultrasonography-guided puncture was performed and no cellular component in the cyst fluid

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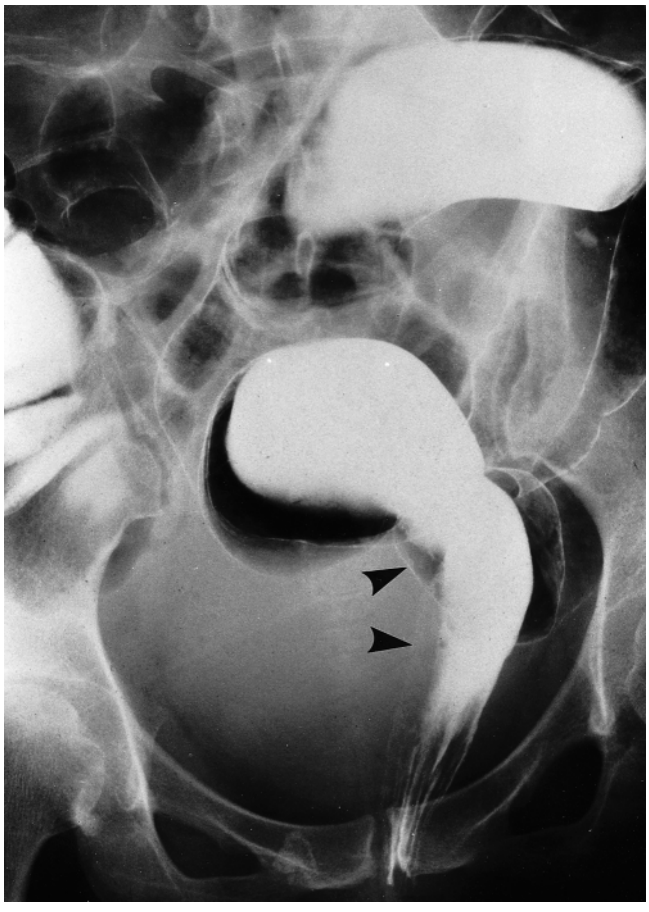

Fig. 1. Barium enema examination showed an anterior shift of the rectum, as indicated by the *arrowheads*

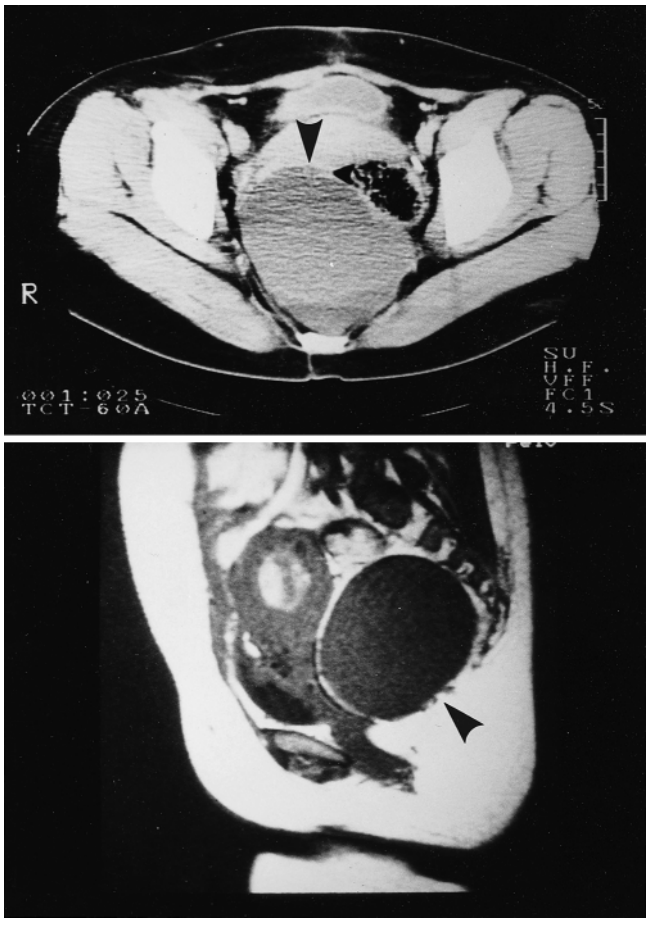

Fig. 2. Computed tomography scan of the pelvic cavity, indicating a huge-sized tumor occupying the entire pelvic space (*top*). Magnetic resonance imaging findings of the lateral view, showing a well-circumscribed tumor occupying the pelvic cavity (*bottom*). The tumor is indicated by the *arrowheads*

was obtained. While a physical examination revealed no tumor in the lower abdomen, an elastic soft tumor with a smooth surface was found in the posterior wall of the rectum by a digital examination per anus. The tumor was located about 5 cm from the anal verge. A barium enema examination showed the rectum to be dislocated anteriorly by the presence of the tumor (Fig. 1). A computed tomography scan revealed a well-circumscribed tumor with a capsule located posterior to the rectum occupying almost the entire pelvic space (Fig. 2, top). A magnetic resonance imaging (MRI) study showed the tumor in the pelvic cavity (Fig. 2, bottom). An angiographic examination showed a posterior shift of the branch of the inferior mesenteric artery by the tumor. However, no vascular encasement was found (Fig. 3).

The operation was performed on April 9, 1991. The tumor was located in the posterior wall of the rectum below the peritoneal reflection. The mesorectum was dissected and the tumor appeared to be located in the submucosal layer under the muscularis propria of the rectum. As the caudal side of the tumor was fibrously attached with the lavator ani muscle, the tumor was carefully detached from the adjacent tissue. After the capsule of the tumor was carefully exposed following the division of the proper muscle layer, and leaving the rectal lumen intact, the whole tumor was resected.

The resected specimen consisted of a 10.0×8.0 cm cyst that was entirely circumscribed with a fibrous and relatively firm capsule (Fig. 4, left). The cyst was filled with an amorphous white creamy substance, which was compatible with a "sebaceous" nature (Fig. 4, right). The lining of the cyst wall consisted of a keratinizing stratified squamous epithelium associated with the sebaceous gland and hair follicles (Fig. 5). These findings were compatible with a benign cystic teratoma (dermoid cyst). The patient had an uneventful postoperative course and was discharged from the hospital 3 weeks after the operation.

Table 1. Clinical findings of the three previously reported cases and the present case of primary submucosal dermoid cyst of the rectum

Author Ref.	Year	Age	Sex	Initial symptoms	Tumor location
Bland-Sutton[1]	1922	39	F	Protrusion of the tumor	Rectum
Russell[5]	1974	77	F	Bloody stool	Posterior wall
Aldridge et al.[2]	1983	51	M	Bloody stool Protrusion of the tumor	Left-sided posterior wall
Present case	2000	30	F	Free	Posterior wall

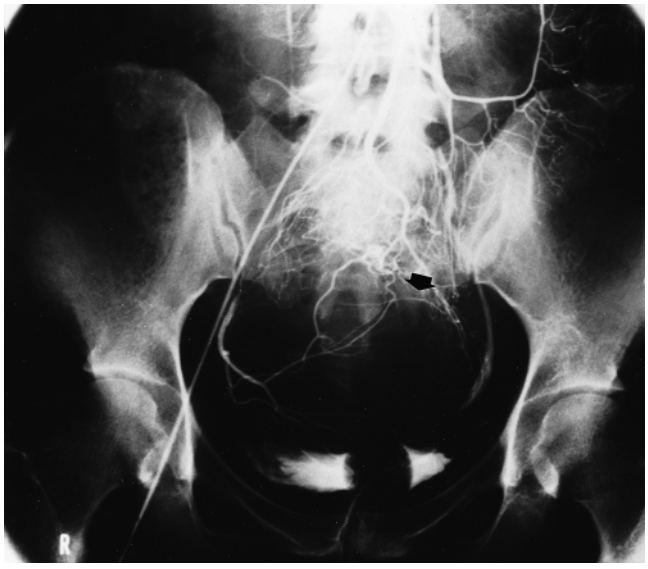

Fig. 3. Angiography findings, indicating a shift in the branches from an inferior mesenteric artery and superior rectal artery by the presence of the tumor, as indicated by the *arrow*. No feeding vessels to the tumor were identified

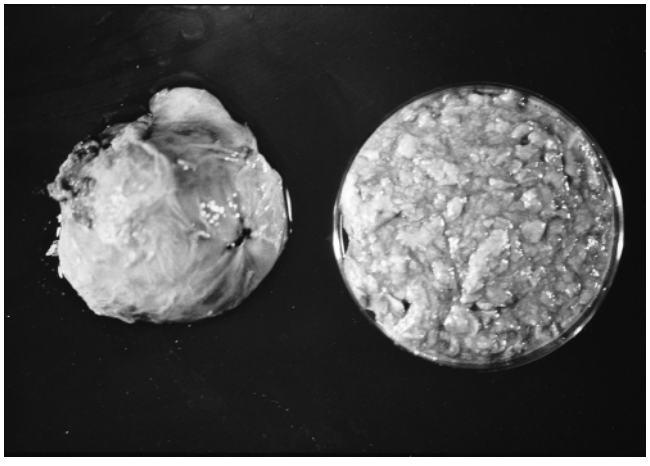

Fig. 4. Resected specimen (*left*) and cyst content (*right*). The cyst was lined with a fibrous capsule filled with amorphous materials

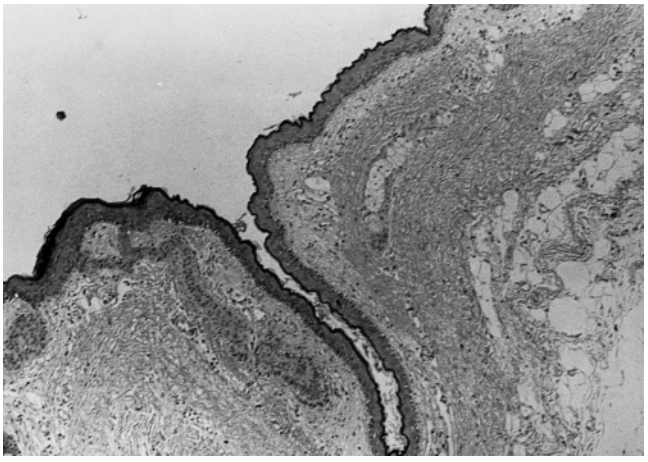

Fig. 5. Histological findings of the resected specimen, showing a keratinizing stratified squamous epithelium associated with the sebaceous gland and hair follicles, which were compatible with a benign cystic teratoma

Discussion

Dermoid cysts of the rectum are generally considered to be due to the faulty inclusion of the ectoderm during embryonic development. They do not usually appear until adulthood and are more frequently seen in females than in males.[1] There has been only one case of a male with rectal dermoid reported.[2] Dermoids of the rectal wall occur either as cysts or solid tumors.[3,4] Teratomas originally arising in the gastrointestinal tract are uncommon. A rectal cyst, one type of teratoma, is also uncommon, and only 36 cases have been reported in literature worldwide. Among the 36 cases previously reported, only 3 cases were presented as a submucosal cyst of the rectum,[1,2,5] and the clinical findings are summarized in Table 1. The other reported cases were polypoid-shaped tumors associated with the stalk occasionally protruding into the rectal lumen. The present case is the fourth case of a submucosal dermoid cyst of the rectum found in the literature, as far as we know, which is extremely rare.

While our patient had no specific symptoms related to the tumor, the most common symptom of a rectal der-

moid cyst is generally the protrusion of hair and/or the tumor itself through the anus occasionally on defecation or at childbirth.[6] Whereas these symptoms were considered to appear only for cases with pedunculated-type tumors, no particular symptoms except those caused by the compression of the rectum were noted for the cases with submucosal-type tumors such as intestinal obstruction or constipation. The lack of clinical symptoms sometimes necessitates more sophisticated diagnostic procedures such as transrectal ultrasound and MRI imaging of the pelvis.[7]

While the submucosal cyst seen in our case originated from the rectal wall, it is difficult in some cases to determine whether the tumor is a primary rectal teratoma or a teratoma of an adjacent organ or tissues that has eroded through the rectal wall. Teratomas involving the ovaries are frequently found, and their direct extension and growth originating in the ovaries has been reported. For example, cases with benign cystic teratoma of an ovary that ruptured into the rectum have been reported.[2,8–10] Peterson et al.[11] collected 1007 cases of benign cystic teratoma of the ovary, and in only 13 cases did rupture occur either into intraperitoneal cavity or into adjacent organs. Ruptures occur into the intraperitoneal space in the majority of cases, but they rarely rupture into the adjacent viscus. Although these cases with a rupture presented some lesions in other organs into which the benign cystic teratoma ruptured, it is occasionally difficult to determine whether the teratoma of the rectum is primary or secondary. Fried and Stone[12] demonstrated that the presence of a well-defined pedicle that connects the tumor and the rectal wall indicates rectal origin. In the present case, because the dermoid cyst was located in the submucosal layer and the muscularis propria was intact, the cyst was determined to be a primary lesion in the rectum.

Because there has been no report of a malignant teratoma of the rectum, a simple complete resection has usually been indicated for treatment. Minimally invasive surgery including endoscopic polypectomy or tumor resection is also indicated for the cases with a well-developed stalk and with no evidence of malignancy of the tumor. In fact, most polypoid types of rectal teratoma were successfully resected by endoscopic manipulation.[6,13] Either an abdominal approach or a transsacral and/or transsphincteric approach could be alternative methods for the resection. Recently, the transsphincteric approach, transsacral approach, and transanal approach have been more frequently indicated for small rectal lesions below the peritoneal reflection, since early-stage rectal lesions are now more frequently found than in the past. Kanemitsu et al.[14] carried out transsphincteric and transsacral approaches in 30 cases with benign and malignant rectal lesions which were inappropriate for either a radical operation and/or recurrent rectal cancer. Rectal benign lesions are particularly good indications for these approaches, only when the preoperative diagnosis as a benign rectal lesion is made. In the present case, since the tumor was large enough to occupy the entire pelvic cavity, the abdominal approach was thus the only choice for a complete resection. A transsphincteric and/or transsacral approach or endoscopic resection should, however, be considered in cases with a small-sized submucosal dermoid cyst.

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