CASE REPORT

Rectal mucocoele in the anterior wall of the rectum

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SUMMARY

An 84-year-old man presented in 2009 with a sensation of discomfort in his anus, combined with difficulty in urination. He had previously undergone a haemorrhoidectomy in 1964. After examination, he was diagnosed with a rectal mucosal cyst and followed up for observation. In 2015, he presented to our hospital complaining that the cyst was prolapsing from his anus. CT revealed a 48×41 mm cystic mass in the anterior wall of the rectum. Tumour extirpation, via a transanal route, was performed. The postoperative pathological diagnosis confirmed a rectal mucocoele. Rectal mucocoeles are extremely rare, with no prior report of a mucocoele in the anterior wall of the rectum. In this case, we believe the mucocoele developed from an invagination of the mucous membrane or obstruction of the anal gland during suturing during the previous haemorrhoid surgery.

BACKGROUND

Mucocoeles are commonly associated with the appendix, gall bladder and cranial sinus,¹ and are extremely rare in the rectum. Rectal mucocoeles are presumed to develop secondary to invasion of the mucous membrane of the rectum by an intervention or obstruction of the anal gland. In this report, we describe a case of a rectal mucocoele that developed in the anterior wall of the rectum after a haemorrhoidectomy.

CASE PRESENTATION

An 84-year-old man presented to our hospital in 2015 with a mass prolapsing from his anus.

Previous medical history

The patient had undergone a haemorrhoidectomy in 1964. Based on our rectal examination, we speculated that the haemorrhoidectomy had been performed using Whitehead's procedure.

The patient first presented to our hospital in 2004 for treatment of an anal fissure and sphincter insufficiency. The symptom was considered to have resulted from haemorrhoidectomy, and conservative treatment was carried out. Because the patient recovered with conservative treatment, image examination was not enforced.

The patient was diagnosed with ascending colonic diverticulitis by CT imaging in 2006. At that time, a rectal mucocoele was not pointed out definitely.

History of present illness

In May 2009, he reported a sense of discomfort in his anus and difficulty urinating. Physical rectal palpation revealed an elastic soft mass in the

anterior wall of the rectum, with a normal mucous membrane. He underwent further examination for a suspected rectal submucosal tumour. CT imaging revealed a lower rectal submucosal cyst (figure 1A). A lower rectal endoscopic examination and MRI revealed that the interior of the cyst was uniform, with a watery consistency and absence of nodule formation. As per our hospital guidelines, the patient was followed up for observation. In January 2015, the cyst began prolapsing from the anus.

Family history

The family history was unremarkable.

INVESTIGATIONS**Current disease**

At the time of admission, the patient was 159.9 cm tall, with a weight of 54.0 kg. Digital rectal and proctoscopy examination confirmed a soft and elastic mass in the anterior wall of the rectum, with no abnormality of the surrounding mucosa.

Blood test findings

All blood tests were normal, including tumour marker values.

Lower digestive tract endoscopy examination

A bulging lesion was identified on the anterior wall of the rectum, 1.5 cm from the dentate line, with no abnormal findings of the mucous membrane (figure 2).

Transanal ultrasonic endoscopy

A cystic mass, 52.4×31.0 mm in size and with a clear boundary, was observed between the mucosal and submucosal layers. The interior of the cyst was homogeneous, with no nodule formation. No invasion of the mass into the muscularis propria layer was observed (figure 3).

Contrast CT examination

On CT imaging, a 48×41 mm large cystic mass was identified on the dorsal side of the prostate and anterior wall of the rectum, with no obvious solid components observed and no abnormal findings in the thoraco-abdominal organs (figure 1B).

TREATMENT**Surgical findings**

Although there was no evidence of malignancy in the rectal submucosal cyst, considering the effect of the cyst on the patient's quality of life, we proceeded with surgical treatment. With the patient placed in a prone Jack knife position, we incised

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Figure 1 (A–B) Contrast-enhanced CT. A cystic mass was identified on the dorsal side of the prostate and anterior wall of the rectum, with no obvious solid components observed. The mass size of (A) was 40×35 mm and (B) was 48×41 mm.

the rectal mucosa, via a transanal route, preserving the perianal muscle, and extirpated the cystic mass ([figure 4](#)).

OUTCOME AND FOLLOW-UP

Resected specimen

The cyst was 5×7 cm in size, with a relatively strong film and a mucosal interior ([figure 5](#)).

Histopathological findings

The mass was diagnosed as a unilocular cystic lesion, with the majority of the wall formed of mucous columnar epithelium, with a component of laminated stratified squamous epithelium. The interior of the mass was filled with a large amount of mucus and a collection of histiocytes. There was no evidence of invasion into other tissues ([figure 6](#)).

Epithelial immunostaining revealed the following: CK7 (+/-), CK20 (+) and CDX-2 (+). Because of the partial CK7 positivity and the observed transition to squamous epithelium, among other findings, a tumour derived from the anal gland epithelium was considered. In the absence of clear malignant findings, we considered that the transition to squamous epithelium as a tumorous change equivalent to a linear tumour. As such, we diagnosed the mass as a mucocoele that had formed in the pararectal region.

Figure 2 Lower digestive tract endoscope. A bulging lesion was identified on the anterior wall of the rectum, 1.5 cm from the dentate line, with no abnormal findings of the mucous membrane.

Figure 3 Transanal ultrasonographic endoscope. A cystic mass, 52.4×31.0 mm in size and with a clear boundary, was observed between the mucosal and submucosal layers. The interior of the cyst was homogeneous, with no nodule formation. No invasion of the mass into the muscularis propria layer was observed.

Figure 4 Surgical findings. With the patient placed in a prone Jack knife position, we incised the rectal mucosa, via a transanal route, preserving the perianal muscle, and extirpated the cystic mass.

Postoperative course

The patient's postoperative course was good, with no impairment in anal function. The patient was discharged on postoperative day 13. Currently, 3 years after surgery, the patient continues to be followed up for observation, with no recurrence of the relapse to date. But an anal fissure and sphincter insufficiency persist.

DISCUSSION

Mucocele are commonly associated with the appendix, gall-bladder, and cranial sinus¹; occurrence of such cysts in the rectum is extremely rare. A PubMed search, using “rectum” and “mucocele” as key words, identified only seven cases other than our own^{1–7}; these are summarised in [table 1](#).

All cases of a rectal mucocele were locally treated, with surgical intervention or drainage. The causes of mucoceles in previously reported cases were as follows: three cases, in addition to our case, developed after a previous haemorrhoidectomy; two cases from an imperforate anus; a rectal stump after surgery for a spinal cord injury³; Crohn's disease⁶; and ulcerative colitis.¹ The posterior wall of the rectum was the most frequent site of occurrence, with our case being the first reported case of a mucocele located in the anterior wall of the rectum. Mucoceles may be more frequent in the posterior than anterior wall of the rectum because the posterior rectal wall is covered by rough tissue, called the rectal lumen. In addition, the presacral space, a region bounded by the rectum on the ventral side, the sacrum on the dorsal side, with the levator ani muscle forming the caudal side and the peritoneal flippers the cephalic side, is a common

Figure 5 Resected specimen. The cyst was 5×7 cm in size, with a relatively strong film and a mucosal interior.

Figure 6 Histopathological findings. The mass was diagnosed as a unilocular cystic lesion, with the majority of the wall formed of mucous columnar epithelium, with a component of laminated stratified squamous epithelium. The interior of the mass was filled with a large amount of mucus and a collection of histiocytes. There was no evidence of invasion into other tissues. A mass derived from the anal gland epithelium was considered, due to partial positivity of CK7 and its transition to squamous epithelium, among other findings. It was diagnosed as a mucous cyst formed in the pararectal region.

site for the development of various congenital masses due to the presence of embryonic remains.⁸

The symptomology of rectal mucoceles includes tenesmus, abdominal pain and ileus obstruction, symptoms which overlap with symptoms of an abdominal tumour. Treatment of reported cases of rectal mucoceles included tumour extirpation (three cases), residual rectal resection (one case) and drainage (four cases). Of the four cases treated using drainage only, recurrence was identified in two cases, and repeated drainage was required in another case.³ In our case, we proceeded with cyst extirpation due to the increasing size of the cyst, concerns of recurrence, if treated using drainage only, and the need for pathological diagnosis.

Mucosal cells that secrete mucus are the primary cells involved in the formation of mucoceles. Appendiceal mucoceles are a well-known cause, with mucoceles resulting from the accumulation of mucus in the lumen of the appendix, leading to occlusion of the appendix.² Regarding the pathogenesis of rectal mucoceles, lesions are assumed to form by a mechanism resembling that of the formation of reserved cyst, secondary to invasion of the mucous membrane by a previous treatment or an obstruction of the anal gland. In the case of the rectal mucocele treated using Hartmann's procedure in ([table 1](#)),³ the mucocele was thought to be associated with an abnormal contraction of the anal sphincter, due to the spinal injury, and development of an infection. The pathological findings in our case indicated that the mucocele likely developed from blockage of the anal gland or invagination of the mucosa at the time of suturing during the previously performed haemorrhoidectomy. In previous reports, the period between the causative treatment and mucocele treatment ranged between 2 months and 27 years, with an average period of 80 months. In our case, this period was much longer, at 51 years. The slow growth of the mucocele, up to 8 mm over a 6-year period, is the reason for this protracted timeline of 51 years in our case ([figure 1](#)). With this protracted cyst development, the amount of mucus within the mucocele increased slowly and, therefore, could be accommodated by the tissues or contained the slight occluded portion of the cystic lesion. It has been reported that the size of a mucocele may vary over time and that mucocele-related symptoms may be intermittent due to the possibility of partial drainage of the mucus inside the rectal lumen.⁷ In our case, the patient had not noticed any discharge of liquid from his anus. The size of rectal mucous cysts has been

Table 1 Reported cases of rectal mucocoeles

Case	Primary disease	Operation	Period	Location (o'clock)	Size (cm)	Therapy	Ref
1	High imperforate anus	Pull-through surgery	nm	nm	nm	Drainage	2
2	Spinal trauma	Hartmann's procedure	2 months	6	9	Drainage	3
3	Hirschsprung's disease	Pull-through surgery	2 years	nm	nm	Extirpation	4
4	Haemorrhoid	Haemorrhoidectomy	4 years	6 and 10	nm	Extirpation	5
5	Crohn's disease	Total colectomy	5 years	6	15	Drainage	6
6	Ulcerative colitis	Subtotal colectomy	27 years	6	nm	Proctectomy	1
7	Haemorrhoid	Haemorrhoidectomy	28 months	3	6	Drainage	7
8	Haemorrhoid	Haemorrhoidectomy	51 years	0	7	Extirpation	Our case

nm, not mentioned.

reported to range between 7 and 15 cm, with a volume of fluid, among cases treated with drainage, ranging between 500 and 3000 mL.

The differential diagnoses of a rectal submucosal mass includes benign (mucocoele, vascular mucocoele, vascular myofibroblastoma, neurofibroma, leiomyoma and a Bartholinian cyst) and malignant (mucinous liposarcoma and malignant fibrous histiocytoma) conditions. Each of these diseases differs slightly with regard to the site of occurrence and clinical symptoms, with MRI being useful for differential diagnosis⁹ and transanal ultrasound necessary to confirm the diagnosis of a mucocoele.⁷ The salient findings of a mucocoele on imaging include a uniform internal cystic mass, with high T2 signal intensity and absence of internal nodules (mucous lake). A tailgut cyst is a cystic tumour that occurs around the rectum, which forms from the remains of the hindgut and is typically recognised in the early foetal period in the anterior sacral space (on the dorsal side of the rectum). As a tailgut cyst is a cystic tumour and considering that the location of the mass in our case, on the anterior wall of the rectum, was not consistent with that of a tailgut cyst,⁸ we excluded a tailgut cyst as a potential diagnosis. With advances in rectal cancer surgery, it is now possible to preserve anal function in more than 80% of cases of rectal cancer, with patients' desire to preserve sphincter function increasing.¹⁰ In addition, the use of intersphincteric resection (ISR) has been expanded by combining this approach with preoperative chemoradiotherapy and other techniques.¹¹ Considering the mechanism underlying the development of

rectal mucocoeles, partial ISR may be a triggering factor and, therefore, the occurrence of rectal mucocoeles may increase as a function of an increased use of ISR in the future.

Contributors These authors saw this case in the department of surgery, and equally contributed for preparation of this manuscript: DI and TA confirmed this case as a rectal mucocoele with a physician; DI and TA operated on the patient and wrote the case report; DI obtained consent from the patient; SI is the assistant director of the hospital, and HY is the director of the hospital; SI and HY rolled the instructor of the operation and instructed of the overall description of this manuscript; all the authors critically appraised the manuscript and contributed to make necessary changes in the article.

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Learning points

- ▶ Rectal mucocoeles are extremely rare.
- ▶ The diagnosis of submucosal tumours of the rectum should be carefully considered among patients with a history of surgery around the anus.
- ▶ Rectal mucocoeles probably result for a blockage of the anal gland or invagination of the mucosa by a previous treatment.
- ▶ The occurrence of rectal mucous cysts may increase as a function of the increasing use of intersphincteric resection in the future.

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