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World J Clin Cases 2020 November 26; 8(22): 5496-5834



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WJCC mainly publishes articles reporting research results and findings obtained in the field of clinical medicine and covering a wide range of topics, including case control studies, retrospective cohort studies, retrospective studies, clinical trials studies, observational studies, prospective studies, randomized controlled trials, randomized clinical trials, systematic reviews, meta-analysis, and case reports.

INDEXING/ABSTRACTING

The *WJCC* is now indexed in Science Citation Index Expanded (also known as SciSearch®), Journal Citation Reports/Science Edition, PubMed, and PubMed Central. The 2020 Edition of Journal Citation Reports® cites the 2019 impact factor (IF) for *WJCC* as 1.013; IF without journal self cites: 0.991; Ranking: 120 among 165 journals in medicine, general and internal; and Quartile category: Q3.

RESPONSIBLE EDITORS FOR THIS ISSUE

Production Editor: *Ji-Hong Liu*; Production Department Director: *Xiang Li*; Editorial Office Director: *Jin-Lai Wang*.

NAME OF JOURNAL

World Journal of Clinical Cases

ISSN

ISSN 2307-8960 (online)

LAUNCH DATE

April 16, 2013

FREQUENCY

Semimonthly

EDITORS-IN-CHIEF

Dennis A Bloomfield, Sandro Vento, Bao-Gan Peng

EDITORIAL BOARD MEMBERS

<https://www.wjnet.com/2307-8960/editorialboard.htm>

PUBLICATION DATE

November 26, 2020

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INSTRUCTIONS TO AUTHORS

<https://www.wjnet.com/bpg/gerinfo/204>

GUIDELINES FOR ETHICS DOCUMENTS

<https://www.wjnet.com/bpg/GerInfo/287>

GUIDELINES FOR NON-NATIVE SPEAKERS OF ENGLISH

<https://www.wjnet.com/bpg/gerinfo/240>

PUBLICATION ETHICS

<https://www.wjnet.com/bpg/GerInfo/288>

PUBLICATION MISCONDUCT

<https://www.wjnet.com/bpg/gerinfo/208>

ARTICLE PROCESSING CHARGE

<https://www.wjnet.com/bpg/gerinfo/242>

STEPS FOR SUBMITTING MANUSCRIPTS

<https://www.wjnet.com/bpg/GerInfo/239>

ONLINE SUBMISSION

<https://www.f6publishing.com>

Rectoseminal vesicle fistula after radical surgery for rectal cancer: Four case reports and a literature review

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Author contributions: Xia ZX designed and drafted the manuscript, collaborated in the patient's perioperative care and performed pathological analysis; Zhang H and Cong JC performed the operations, reviewed and revised the manuscript; all authors read and approved the final manuscript.

Informed consent statement:

Written informed consents were obtained from the patients for publication of this report and any accompanying images.

Conflict-of-interest statement: The authors declare that they have no competing interests.

CARE Checklist (2016) statement:

The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

Open-Access: This article is an open-access article that was selected by an in-house editor and fully peer-reviewed by external reviewers. It is distributed in accordance with the Creative Commons Attribution

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Abstract

BACKGROUND

A rectoseminal vesicle fistula (RSVF) is a rare complication after anterior or low anterior proctectomy for rectal cancer mainly due to anastomotic leakage (AL). Limited literature documenting this rare complication is available. We report four such cases and review the literature to investigate the etiology, clinical manifestations, and the diagnostic and treatment methods of RSVF in order to provide greater insight into this disorder.

CASE SUMMARY

Four cases of RSVF were presented and summarized, and a further 12 cases selected from the literature were discussed. The main clinical symptoms in these patients were pneumaturia, fever, scrotal swelling and pain, anal pain, orchitis, diarrhea, dysuria, epididymitis and fecaluria. Imaging methods such as pelvic X-ray, computed tomography (CT), sinus radiography, barium enema and other techniques confirmed the diagnosis. CT was the imaging modality of choice. In cases presenting with reduced levels of AL, minimal surrounding inflammation, and controlled infection, the RSVF was conservatively treated by urethral catheterization, antibiotics administration and parenteral nutrition. In cases of severe RSVF, incision and drainage of the abscess or fistula and urinary or fecal diversion surgery successfully resolved the fistula.

CONCLUSION

This study provides an extensive analysis of RSVF, and outlines, summarizes and examines the causes, clinical manifestations, diagnostic procedures and treatment options, in order to prevent misdiagnosis and treatment errors.

Key Words: Case report; Seminal vesicles; Proctectomy; Anastomotic leakage; X-ray computed; Tomography

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Manuscript source: Unsolicited manuscript

Specialty type: Surgery

Country/Territory of origin: China

Peer-review report's scientific quality classification

Grade A (Excellent): 0
Grade B (Very good): 0
Grade C (Good): C
Grade D (Fair): 0
Grade E (Poor): 0

Received: May 22, 2020

Peer-review started: May 22, 2020

First decision: September 14, 2020

Revised: September 16, 2020

Accepted: September 25, 2020

Article in press: September 25, 2020

Published online: November 26, 2020

P-Reviewer: De la Plaza Llamas R

S-Editor: Zhang H

L-Editor: MedE-Ma JY

P-Editor: Ma YJ



Core Tip: A rectoseminal vesicle fistula (RSVF) is a rare complication after anterior or low anterior proctectomy for rectal cancer mainly due to anastomotic leakage. Four cases of RSVF were presented and summarized, and additional 12 cases selected from the literature were reviewed and discussed. This study provides an extensive analysis of RSVF, and outlines, summarizes and examines the causes, clinical manifestations, diagnostic procedures and treatment options, in order to prevent misdiagnosis and treatment errors.

Citation: Xia ZX, Cong JC, Zhang H. Rectoseminal vesicle fistula after radical surgery for rectal cancer: Four case reports and a literature review. *World J Clin Cases* 2020; 8(22): 5645-5656

URL: <https://www.wjgnet.com/2307-8960/full/v8/i22/5645.htm>

DOI: <https://dx.doi.org/10.12998/wjcc.v8.i22.5645>

INTRODUCTION

According to a report from the United States, the incidence of colorectal cancer (CRC) ranks third of all malignant tumors^[1]. Rectoseminal vesicle fistula (RSVF) is a rare complication after invasive surgery, which is different from urethral fistulas such as rectal ejaculatory duct fistula^[2], rectovesical fistula^[3,4], colovesical fistula^[5-7], prostatic-rectal fistula^[8] and coloseminal vesicle fistula^[9,10]. There have been 10 relevant articles which reported a total of 12 cases of RSVF worldwide since 1989^[11-20]. Among the various etiologies of RSVF such as prostatectomy^[14], and transrectal seminal vesicle (SV) puncture drainage^[16], the most common etiology is delayed emergence of anastomotic leakage (AL) following anterior resection (AR) or low anterior resection (LAR) for rectal surgery^[11-13,15,17,19,20]. This study details four cases of RSVF from two surgical teams at the CRC Surgery Department between January 2009 and January 2019.

This study was reviewed and approved by the Ethics Committee of Shengjing Hospital of China Medical University (2017PS301K). Written informed consent was obtained from each patient. All patients were treated without chemotherapy or radiotherapy. Due to underlying diseases and economic reasons, patients 2 and 3 refused preoperative neoadjuvant therapy and protective ileostomy, and relevant agreements were signed. The patients were followed-up until May 2020 by clinical visit or by telephone. The follow-up period ranged from 8 to 88 mo after the first operation.

We conducted a comprehensive review of the literature from PubMed, EMBASE, Ovid and Springer published in English before May 2020. The following key words were used to identify articles: Rectoseminal vesicle fistula, recto-seminal vesicle fistula, rectal seminal vesicle fistula, and seminal vesicle rectal fistula. We also reviewed the bibliographies of relevant articles to identify additional studies. The inclusion criteria were as follows: (1) Patients with rectal SV fistula; and (2) Acceptance of open or laparoscopic rectal cancer radical surgery. The exclusion criteria were as follows: (1) Patients with colonic SV fistula; and (2) No detailed case information. The flow chart of the screened cases is shown in [Figure 1](#).

Patients in two surgical treatment groups underwent a total of approximately 8000 CRC radical operations, and the incidence of AL was 2%-3% within ten years. This study detailed four cases of RSVF with an incidence of 2.02% secondary to AL following AR or LAR for rectal cancer between July 2013 and January 2019. The clinical data of 12 reviewed cases are summarized in [Table 1](#) (No. 1-12), while the therapeutic and prognostic information of the additional four cases are shown in [Table 1](#) (No.13-16).

CASE PRESENTATION

Chief complaints

Case 1: A 64-year-old man developed symptoms of fever, with a maximum temperature of 38.5°C and stool frequency of 7-8 /d on postoperative day (POD) 5. Fecal residue due to AL was observed in the intrapelvic drainage tube.

Table 1 General data of 16 patients with rectoseminal vesicle fistula

No.	Ref. or case	Patient age (yr)	Etiology	Symptoms	Diagnostic methods	Initial treatment		Re-treatment	
						Methods	Effects	Methods	Effects
1	Goldman <i>et al</i> ^[11]	76	LAR, AL, diarrhea associated with antibiotic	Pneumaturia, orchitis, urinary tract infection, diarrhea	Sinus radiography, barium enema	Catheterization, anti-inflammatory, sinus incision and drainage	Improved		
2	Kollmorgen <i>et al</i> ^[12]	32	LAR, pelvic abscess	Fever, dysuresia	Suspicious symptoms	APR (fecal diversion), incision and drainage of subcutaneous abscess	Failure	Anti-inflammatory	Cured
3	Carlin <i>et al</i> ^[13]	64	AR	Fever	CT + rectal contrast	Abscess drainage	Failure	APR (fecal diversion)	Cured
4	Calder ^[14]	NS	Prostatectomy	NS	Barium enema	NS	NS		
5	Roupret <i>et al</i> ^[15]	NS	Rectal cancer invaded seminal vesicle, LAR, pelvic abscess	Epididymitis, orchitis	CT	Anti-inflammatory, proctectomy, abscess drainage	Cured		
6	Hammad ^[16]	NS	Transrectal seminal vesicle puncture drainage	Fever, anal pain	NS	Perianal drainage	Cured		
7	Sýkora <i>et al</i> ^[17]	66	Laparoscopic AR	Pneumaturia, fever, epididymitis, scrotal swelling and pain	CT	Catheterization, anti-inflammatory, suprapubic cystostomy (urinary diversion)	Failure	Anti-inflammatory	Cured
8	Izumi ^[18]	74	NS	Pneumaturia, fever, orchitis,, scrotal swelling	Cystography	Bilateral scrotal drainage	Cured		
9	Nakajima <i>et al</i> ^[19]	73	LAR, AL	Pneumaturia, fever, orchitis	CT, sinus radiography	Catheterization, anti-inflammatory	Cured		
10	Nakajima <i>et al</i> ^[19]	76	LAR, water sac was not aspirated when removing ureter	Pneumaturia, orchitis,, scrotal swelling	CT, barium enema	Catheterization, anti-inflammatory	Failure	Fecal diversion	Cured
11	Nakajima <i>et al</i> ^[19]	49	LAR, AL	Fever, fecaluria	Ejaculatory duct radiography	Catheterization, anti-inflammatory, drainage of pelvic abscess,	Failure	Urinary + fecal diversion	Improved
12	Kitazawa <i>et al</i> ^[20]	53	LAR, AL, diarrhea associated with antibiotic	Pneumaturia, fever, diarrhea, cystitis	CT	Anti-inflammatory, parenteral nutrition	Cured		
13	Case 1	64	LAR, AL	Fever, diarrhea, dysuresia, pneumaturia, epididymitis, scrotal swelling, orchitis, anal abscess	CT	Catheterization, anti-inflammatory, parenteral nutrition drainage of pelvic abscess, incision and drainage of epididymal abscess	Failure	Fecal diversion, incision and draining perianal abscess	Cured
14	Case 2	69	Laparoscopic AR, AL	Pneumaturia, scrotal swelling, hematochezia, anal pain	CT, MRI, Urethral retrograde urography	Fecal diversion, Catheterization, anti-inflammatory	Cured		
15	Case 3	74	Laparoscopic LAR, AL	Fever, abdominal pain, scrotal swelling, pneumaturia, fecaluria, dysuresia	CT	Catheterization, anti-inflammatory	Cured		

16	Case 4	49	LAR, AL	Diarrhea, anal pain, pneumaturia	CT	Catheterization, anti-inflammatory	Failure	Fecal diversion, anastomotic stenosis	Cured
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LAR: Low anterior resection; AL: Anastomotic leakage; AR: Anterior resection; NS: Not stated; CT: Computed tomography; MRI: Magnetic resonance imaging.

Case 2: A 69-year-old man developed symptoms of pneumaturia, left scrotal swelling, hematochezia and anal pain on postoperative month (POM) 5 which gradually became worse.

Case 3: Abdominal swelling and pain, fever and abnormal characteristics of the intrapelvic drainage tube with an average of 15-30 mL feces appeared on POD 3 in a 74-year-old man. His abdominal pain intensified and new symptoms of left scrotal swelling and urine turbidity emerged on POD 4.

Case 4: Urinary retention and diarrhea with 10–20 stools/d developed on POD 17 and POD 20, respectively, in a 49-year-old man. Physical examination revealed slight anastomotic stenosis, right anterior wall AL approximately 2 cm from the anal verge, local tissue hardness and positive tenderness.

History of present illness

Chest, abdominal and head X-rays and computed tomography (CT) scans showed no distant metastases. Open LAR was performed in case 1 and case 4, while laparoscopic AR was carried out in the other two cases with additional ileostomy in case 3. The patients, irrespective of open or laparoscopic proctectomy, all underwent end-to-end anastomosis using a stapler.

History of past illness

Case 3 had coronary heart disease for ten years. The remaining cases had no history of past illness.

Personal and family history

None of the patients had a history of drug allergies or genetic diseases.

Physical examination

The average tumor diameter in these four patients was 4.5 cm, and the distance from the anal margin ranged from 4 cm to 8 cm. With the exception of case 2 with right wall tumor, the tumor was located in the anterior wall in the other three patients.

Laboratory examinations

AL in case 1 was locally restricted, and culture for blood bacteria and fungi was negative. All patients had different degrees of elevated white blood cells.

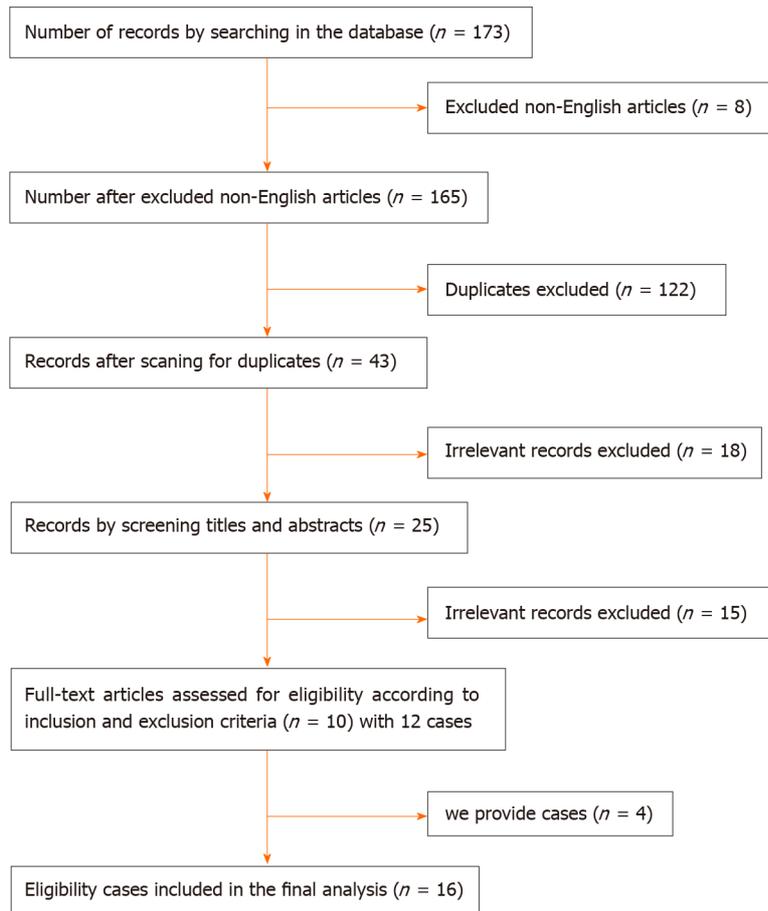


Figure 1 Flow chart of screening process.

Imaging examinations

Case 1: Pelvic CT revealed RSVF secondary to AL following LAR, air bubbles were seen to enter the ejaculatory duct opening of the urethral prostate caruncle due to direct infection, and the bladder along the posterior urethra and the scrotum along the deferent duct showed retrograde infection on POD 21 (Figure 2A-E). Urinary system color Doppler ultrasound diagnosed a right epididymis tail abscess, epididymitis and scrotal edema (Figure 2F).

Case 2: Colonoscopy revealed local inflammatory edema and leakage of anastomoses, while cystoscopy showed no obvious abnormalities. However, CT demonstrated a pelvic pus cavity and a left scrotal abscess secondary to AL after proctectomy (Figure 3A-D). Magnetic resonance imaging (MRI) diagnosed a sinus from a pelvic encapsulated abscess to the SV associated with rectal AL (Figure 3E-G). Urethral retrograde urography demonstrated contrast agent retrogradely entering the ejaculatory duct from the ejaculatory duct opening of the urethral prostate caruncle (Figure 3H and I).

Case 3: Preoperative MRI in case 3 revealed penetration of the rectal front wall by the large tumor including the full thickness of the rectal layers and invasion of Denonvilliers' fascia (DF) at the level of the SV (Figure 4A and B). The left swollen and infected scrotum was revealed by postoperative CT (Figure 4C). Transabdominal sinus radiography identified rectal AL but not urethral leakage (Figure 4D). RSVF was identified by CT when contrast agent was seen to retrogradely enter the ductus deferens around the entrance to the epididymis (Figure 4E) and air bubbles in the SV and bladder *via* the sinus secondary to AL (Figure 4F).

Case 4: Pelvic CT scan, sinus radiography, cystoscopy and transurethral retrograde angiography showed no obvious sinus; however, rectovesical fistula was excluded. Pelvic enhanced CT revealed a fistula between the right SV and the rectal anastomotic site, and air bubbles located within the right SV, which confirmed RSVF secondary to

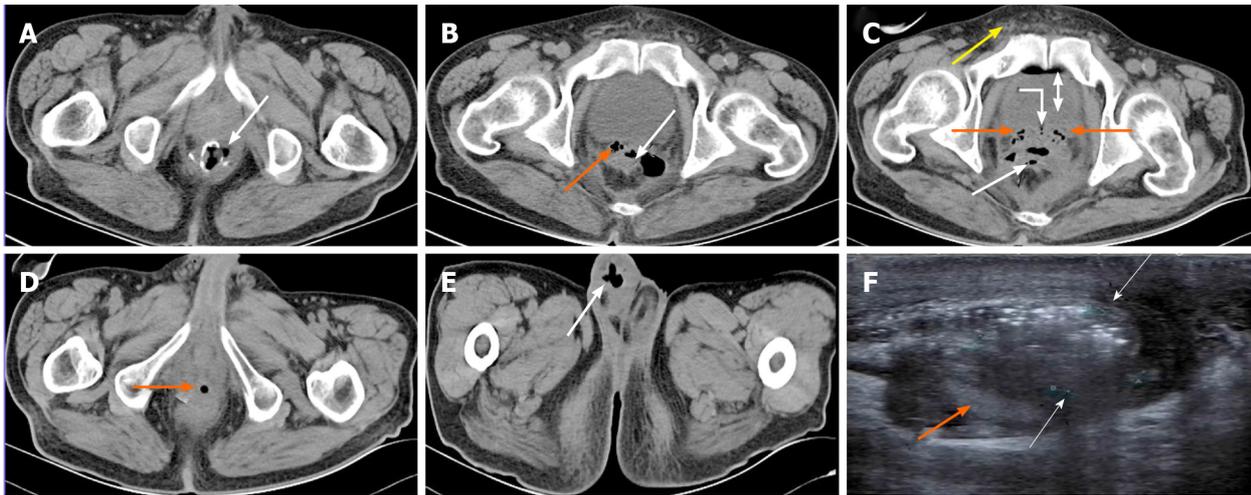


Figure 2 Imaging findings of case 1. A-C: X-ray computed tomography (CT) demonstrated edematous rectal wall and double seminal vesicles (SV), incomplete anastomosis with an obvious rectal anastomotic leakage (AL) (white arrows). A large pelvic cavity around the AL formed a sinus from the rectum to SV, and air bubbles entered bilateral SVs (orange arrows), and ampulla of deferent duct (curved arrow). Some bubbles present in the bladder (double arrow) and a few in the right deferent duct within the spermatic cord (yellow arrow); D: CT displayed air bubbles (orange arrow) located within ejaculatory duct opening of the urethral prostate; E: Bubbles (white arrow) from the right sperm duct retrogradely entered the right edematous and infected scrotum; F: Urinary system color Doppler ultrasound showed a right epididymis tail 2.2 cm x 1.8 cm x 1.5 cm enclosed abscess and another small abscess in the right edematous scrotum with a blurred boundary, a number of strong gas echoes and peripheral rich blood (white arrows) around the testis and epididymis (orange arrow).

AL (Figure 5A and B).

FINAL DIAGNOSIS

Case 1: RSVF, AL, right epididymis tail abscess. Case 2: RSVF, AL, left scrotal abscess. Case 3: RSVF, AL. Case 4: RSVF, AL.

TREATMENT

Case 1: The patient was treated with lavage *via* an intrapelvic drainage tube, parenteral nutrition, anti-inflammatory and anti-diarrheal therapy as conservative treatment. Due to gradual development of dysuria, pneumaturia, testicular pain, scrotal swelling and intermittent fever with the highest temperature of 39°C, urethral catheterization with a saline flush was performed on POD 17. The most severe urinary tract symptoms appeared on POD 33 to 44. The patient continued with urethral catheterization and underwent surgery for incision and drainage of the right epididymis abscess and colostomy or ileostomy for fecal diversion on POD 45. The patient began to drink a small amount of water and gradually increased the amount of water to increase urine volume and reduce the risk of urinary tract infection. Pneumaturia and frequent defecation gradually improved to 2 times/d and 4-5 stools/d, respectively, on POD 68. The previous conservative treatment was continued and partial enteral nutrition was increased. The fistula had disappeared by POD 120. The catheter was removed, oral feeding was conducted, and intestinal traffic continuity was restored. A chemotherapy regimen which included oxaliplatin and fluorouracil was given twice and terminated due to side effects. The patient experienced repeated perianal discharge at POM 10 and a perianal abscess was diagnosed by pelvic CT. Stool frequency gradually increased with a maximum frequency of 30/d. Conservative treatment was continued for a further 10 mo and the patient still experienced serious symptoms. Finally, in view of the risk of AL and stenosis after stoma, the patient requested resection of the rectal anastomosis (Hartmann operation), a permanent colostomy for defecation and incision and drainage of the perianal abscess were performed to replace the temporary lateral ileostomy or colostomy. The leakage was successfully healed at POD 10 after the second operation. Chemotherapy was administered a further 3 times.

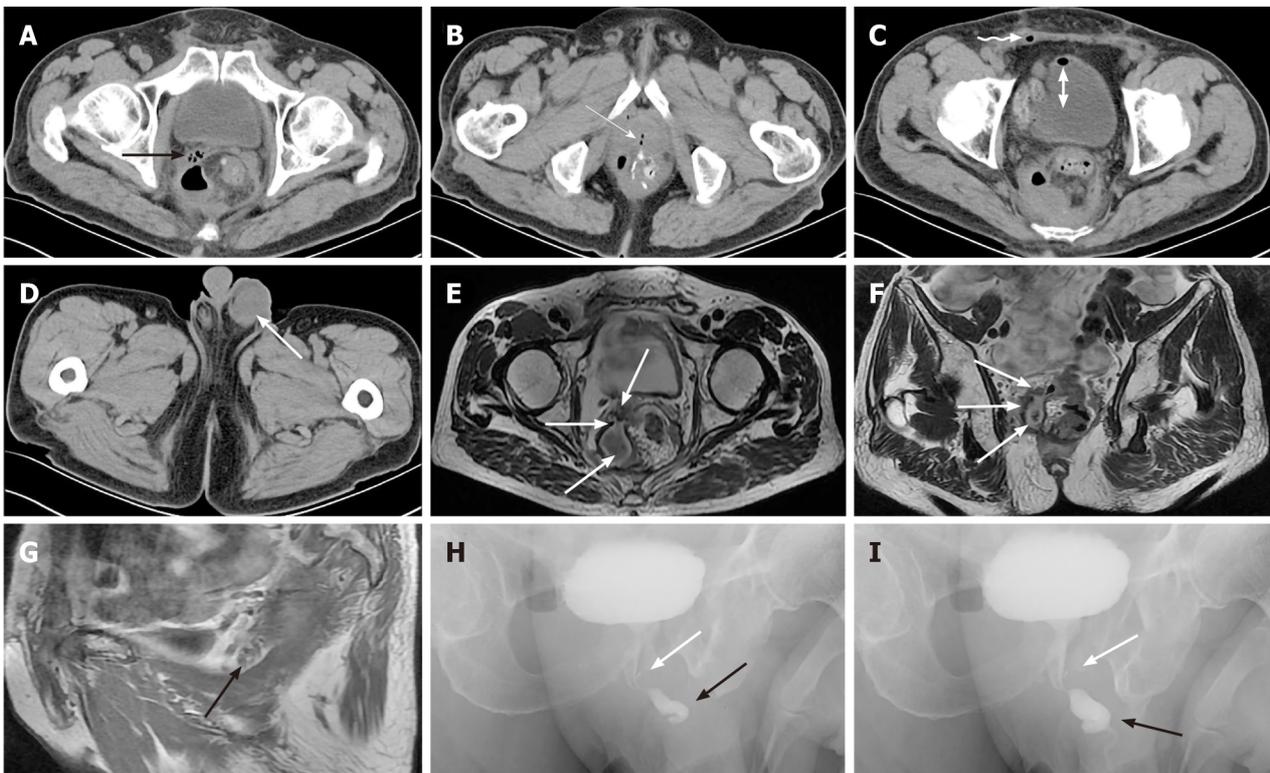


Figure 3 Imaging findings of case 2. A-C: Computed tomography demonstrates an encapsulated cavity filled with air bubbles, pus and effusion with an air-water level connecting with the right seminal vesicles (SV) (black arrow) and ejaculatory duct opening of the urethral prostate caruncle (white arrow) after anastomotic leakage (AL). Some bubbles in the right deferent duct within the spermatic cord (curved arrow) and some in the bladder (double arrow); D: Left edematous and infective scrotum (white arrow); E, F: Cross-section and coronal plane magnetic resonance imaging (MRI) displayed a sinus between encapsulated pus cavity and right edematous SV (white arrows) secondary to AL; G: Sagittal plane MRI demonstrated some bubbles in the right edematous SV (black arrow) between bladder and rectum; H, I: Urethral retrograde radiography showed that the bladder was well filled, the anterior urethra (black arrows) was normal, the posterior urethra was slightly narrow, and the ejaculatory duct was filled with a small amount of contrast agent (white arrows).

Case 2: Anti-inflammatory therapy with broad-spectrum antibiotics and urethral catheterization gradually improved anal pain and pneumaturia, and AL and RSVF resolved on POM 10. CT revealed right SV and left deferent duct stones without symptoms secondary to RSVF (Supplementary Figure 1).

Case 3: Lavage *via* an intrapelvic drainage tube with 250/500 mL normal saline flush and negative pressure, anti-inflammation with a broad-spectrum antibiotic (metronidazole) once/d and enteral nutrition, were administered to improve symptoms, while moxifloxacin was added once/d for unimproved urinary tract symptoms, *e.g.* left scrotal edema and pneumaturia which emerged on POD 12 and fecaluria on POD 14, respectively. Enteral nutrition was replaced by normal eating and urethral catheterization was removed when symptoms of dysuria, pneumaturia, and fecaluria disappeared, scrotal edema was alleviated, and pelvic drainage decreased to approximately 5 mL/d without obvious abscess and feces on POD 20. After 3 d, dysuria and urinary infection reappeared without fever, and a continuous daily moxifloxacin flush *via* the intrapelvic drainage tube was administered with obvious effect, which continued until POD 30. Healing of the fistula was confirmed and stoma closure was eventually performed on POM 7.

Case 4: Urethral catheterization, anti-inflammatory and anti-diarrheal drugs were given but had no effect following gradually aggravated anal pain and pneumaturia. As this conservative treatment did not improve the patient's condition, the patient finally underwent ileal protective stoma with fecal diversion and anastomotic stenosis dilatation on POD 74 with a gradual improvement in symptoms. Stoma closure was performed on POM 8.

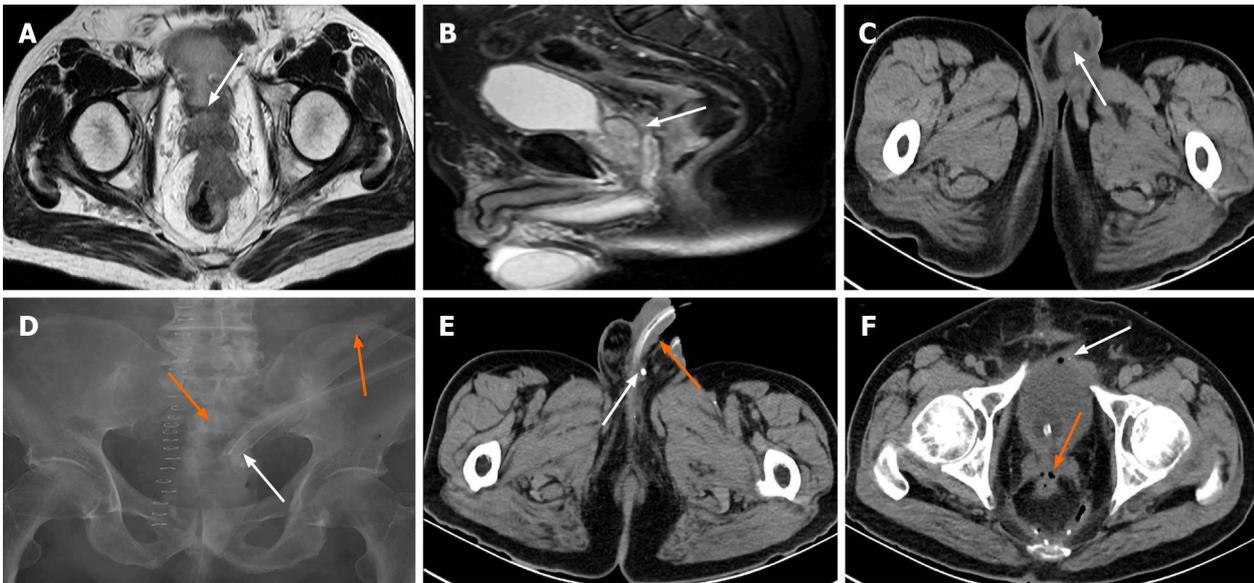


Figure 4 Imaging findings of case 3. A, B: Cross-section and sagittal plane magnetic resonance imaging (MRI) showed that the bladder was filled well, coating of the seminal vesicles (SV) were complete, the huge rectal tumor invaded Denonvilliers' fascia (white arrows) at the level of the SVs; C: The left swollen and infective scrotum (white arrow) by computed tomography (CT); D: Transabdominal sinus radiography showed contrast agent entering the rectum and proximal colon (orange arrows) through intrapelvic drainage tube (white arrow); E: Contrast agent residue (white arrow) in the ductus deferens beside the entrance to the epididymis but not urethra (orange arrow) by CT; F: A few effusion and bubbles in the pelvic cavity, some bubbles entering the SV (orange arrow) and bladder (white arrow) via the sinus from rectal anastomotic leakage.

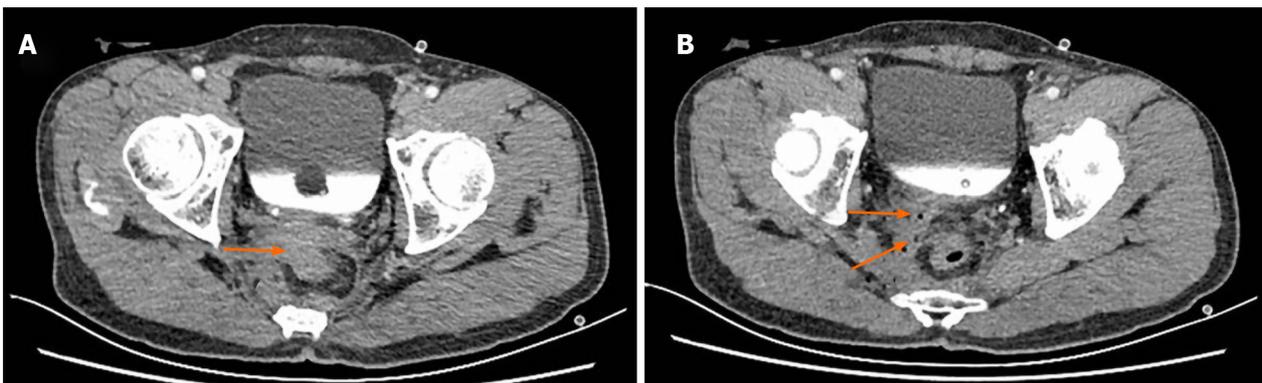


Figure 5 Imaging findings of case 4. A, B: A sinus from the rectal anastomotic site to the right seminal vesicles (SV) secondary to anastomotic leakage and air bubbles squeezed in the right SV (orange arrows) by enhanced computed tomography.

OUTCOME AND FOLLOW-UP

The first three cases had moderately differentiated adenocarcinoma, and the fourth case had well differentiated adenocarcinoma. TNM stages were T2N0M0, T3N0M0, T3N2M0 and T2N0M0, respectively.

Case 1: Follow-up was continued until the 50th month after the first operation and no recurrence or metastasis was observed on whole body positron emission tomography .

Case 2: The patient is still alive without local recurrence 75 mo after the first operation.

Case 3: The patient is still alive without recurrence or metastasis 8 mo after surgery.

Case 4: The patient is still alive without local recurrence or distant metastasis 88 mo after the first operation.

In the analysis of the 12 cases from the literature and the present 4 new cases, RSVF developed after proctectomy (12/16), with 75% following LAR and 25% following AR, after resection of the prostate (1/16), after an invasive operation (1/16), for direct tumor invasion (1/16) and for an unknown cause (1/16). The tumors including 13 well/moderately differentiated adenocarcinoma and a giant polyp with severe

atypical hyperplasia and 2 unclear cases, were 4–11 cm from the anal verge. The shortest onset time was POD 3, most of which occurred within one month, while the longest onset time was POM 6. The main clinical manifestations included fever (10/16), pneumaturia (10/16), scrotal swelling and pain (6/16), anal pain (5/16), orchitis (5/16), diarrhea (4/16), dysuria (3/16), epididymitis (2/16), and fecaluria (2/16). Occasionally, urinary tract infection, abdominal pain, hematuria, abnormal sperm, and cystitis were observed. Most RSVFs in these 16 cases were identified by CT including enhanced CT and CT with rectal contrast imaging (10/16), barium enema (3/16), sinus radiography (2/16), and occasionally by cystoscopy, urethral retrograde urography, MRI, ejaculatory duct radiography, and due to clinical symptoms and experience. All imaging examinations had different diagnostic significance, but that of CT, was highest. In total, 4 of the 16 patients were cured by conservative therapy, 4 cases improved or were healed by conservative therapy plus abscess or fistula incision and drainage, 9 cases were cured by colostomy or ileostomy after failed treatment (a temporary protective ileostomy was performed during the first operation in case 3), 2 cases with severe infection underwent urinary and fecal diversions combined with long-term postoperative anti-infection therapy, and in 1 case treatment was unclear.

DISCUSSION

With advancement in surgical techniques after removal of the rectum and in surgical instruments, the rate of anal preservation has improved, and visualization of the local anatomic structures during surgery has been refined. However, the rate of urinary system complications, which are still difficult to eradicate during proctectomy, is approximately 1.5%–3.0%^[21,22]. It is essential to identify and diagnose RSVF as a urethral-related injury in patients who still have temporary urethral symptoms such as scrotal swelling, epididymitis, and orchitis after rectal surgery.

The etiology of RSVF in the four cases presented in this study was analyzed. First, RSVF mainly occurs due to AL. Fecal material accompanied by infectious substances contaminate and damage the tissues surrounding the anastomotic fistula, which gradually penetrate the adjacent exposed SV to form a fistula. Second, during radical proctectomy with total mesorectal excision (TME) as the gold standard^[23,24], DF or anterior rectal fascia, which is the natural barrier for the forward invasion of rectal tumors and the anterior boundary for TME in AR or LAR^[25], is separated between the anterior wall of the rectum and the peritoneum at the bottom of the posterior bladder wall, down to the demarcation line between the prostate, the SV and the rectum, and even to the pelvic floor and levator ani muscle level. The circumferential margin (CRM) is also a major predictor of survival after surgery or neoadjuvant therapy for rectal cancer^[26]. The tumor in our case 3 was located in the anterior wall of the rectum, and it was necessary to ensure a sufficient and safe CRM, which may result in the SV membrane being thinner and even damaged. Inflammation around the fistula more easily affects the SVs with no protection of DF to form the RSVF^[11]. One case with a large rectal adenocarcinoma invading the right SV was treated with bilateral SV resection; however, it is difficult to precisely ligate the bottom of the SV. Postoperative AL leads to local abscess invading the opening of the ejaculatory duct^[19]. Third, long-term and repeated urethral catheterization increases the risk of urinary tract infection due to chronic compression of the ejaculatory tube opening of the urethral prostate and causes local inflammation and ischemia. Finally inflammation retrogradely affects the SVs causing vesiculitis and a fistula. In addition, antibiotic-related intractable diarrhea^[8] inducing AL, particularly in the anastomotic stoma only 2–3 cm from the anal verge, is another etiology of RSVF.

The onset time is mainly determined by the size of the AL, the progress of inflammation and the time of infection invading the SV. In case 1 and case 3, we observed the number and severity of symptoms due to early-onset and obvious AL with inflammation invading the urethra (anterograde direction) and deferent duct (retrograde direction), in contrast to case 2 and case 4 who had relatively few and slight symptoms owing to occult and late-onset AL. The clinical symptoms can be divided into 3 parts. First, the main symptoms are fever, pneumaturia, fecaluria, and dysuria, as a result of a small amount of gas entering the SV along the narrow sinus and discharging into the urethra *via* the ejaculatory duct and its opening in the urethral prostatic caruncle. If the fistula is obvious, part of the fecal residues enter the sinus causing fecaluria. Inflammation stimulates the bladder causing edema and dysuria. Second, the gases, feces, infective necrotic materials and semen can reverse into the deferent duct and epididymis causing orchitis, epididymitis^[9], scrotal pain,

and even an abscess^[13]. Third, the main symptoms are diarrhea and anal pain, related to low anastomosis and AL.

Each patient has specific requirements due to their individual differences; hence, the diagnostic methods vary. When combined with clinical symptoms and the characteristics of intrapelvic drainage tubes, pelvic CT scan and contrast-enhanced CT had a high diagnostic rate as they clearly showed the fluid and air bubbles around the AL, and identified the sinus between the fistula of rectal anastomosis and infected SV. Of course, MRI can be performed due to its advantages in displaying complicated leakages^[27]. Case 2 experienced symptoms of hematochezia; therefore, we used urethral retrograde urography to detect the small amount of contrast agent retrogradely entering the opening of the ejaculatory duct in the urethral prostate, after CT scanning indicated bubbles in the ejaculatory duct and deferent duct. The rectal fistula draining the sinus and port are usually straighter and wider than that of the sperm duct and ejaculatory duct; thus, transrectal venography (*e.g.*, barium enema and sinus radiography) is usually easier than transurethral (*e.g.*, urethral retrograde urography and cystoscopy). There was a right epididymal abscess and obvious infection in the testes, scrotum and epididymis in case 1. With the rapid development of ultrasound and endoscopy, it is worthwhile investigating transanal endoscopic ultrasonography (TEUS), which can successfully diagnose RSVF by scanning various layers of the rectum with an ultrasound probe. Coulier *et al*^[21] reported that a SV-colonic fistula secondary to a sigmoid diverticulum resulted in emphysematous epididymitis, which was diagnosed by TEUS. The most noteworthy technique may be SV adenoscopy, according to reports^[28,29], transurethral SV adenoscopy has been used in the diagnosis and treatment of SV diseases (such as SV stone, deposition, inflammation, tumor, ejaculatory duct cyst, hemospermia *etc.*). Its application has been confirmed, but it is also expected to become a definite diagnostic and treatment method for RSVF. As multiple similar cases are diagnosed, sometimes with symptoms, we can diagnose the disease clinically.

The treatment of RSVF is divided into conservative treatment and surgical treatment. Conservative treatments include anti-inflammatory therapy with sensitive antibiotics, total parenteral nutrition, lavage *via* the intrapelvic drainage tube to reduce fecal contamination, urethral catheterization in order to decrease irritation and infection in the urinary system, and symptomatic treatment such as anti-diarrheal therapy. In case 2 who had an undiagnosed postoperative AL, this was manifested by various RSVF-related symptoms on POM 5. Due to minor AL, localized inflammation and unobstructed drainage, the patient recovered after conservative treatment. Elderly patients with severe primary disease and unable to tolerate surgical trauma, young patients who have fertility requirements, and those who do not accept colostomy or SV resection can attempt conservative treatment and uncomplicated incisional drainage. A complex sinus, deep location of the abscess, poor drainage, and severe local or systemic infection are the main causes of treatment failure.

In patients with serious AL, extensive infection and obvious urethral symptoms, conservative treatment alone is inadequate and suitable operations are required. Surgical treatments include sinus or abscess drainage, urinary diversion (*e.g.* suprapubic cystostomy, cutaneous ureterostomy *etc.*) and fecal diversion (*e.g.* colostomy, ileostomy *etc.*). If drainage *via* the intrapelvic tube is obstructed, the pus cavity is superficial and located near the skin, subcutaneous or sinus incision and drainage can be performed by the perianal or perineal route and are partly effective^[11,15,16]. In addition, scrotal drainage^[18], or epididymal abscess drainage due to retrograde deferent duct infection as in case 1 can be performed, respectively. Urine/fecal diversion seems to be necessary when drainage fails to relieve symptoms or cure RSVF with a success rate of 75%. After failed operations, some patients still require treatment with long-term anti-infection^[12,16]. Sýkora *et al*^[17] reported that some cases requiring urinary diversion can reduce intra-vesicle pressure and accelerate healing with suprapubic cystostomy. Although we have not attempted urinary diversion, our experience is to maintain the patency of the urinary tract until symptoms improved significantly following urethral catheterization. If the symptoms appear again, urethral catheterization is performed again. The incidence of AL after colorectal surgery varies greatly from 4% to 29.5%^[30-33]. Whether enterostomy can prevent AL has always been a controversial hot topic^[34,35]. However, fecal diversion can effectively prevent feces from continuing to aggravate the infection around the anastomosis, and the diet should be restored as soon as possible to enhance immunity and reduce the release of inflammatory factors^[36]. The limitations of this study are that due to the small number of cases, it is not possible to provide a therapeutic regimen for this complication. Therefore, individualized treatment of RSVF is necessary.

CONCLUSION

The incidence of RSVF is much higher than reported as not enough is known about the disease. Four cases of RSVF that occurred as a complication due to AL after proctectomy along with a review of the literature of similar cases are discussed. This helped us to analyze the disease process and postulate various possible etiologies which will prompt further investigation and open new avenues for research to eventually enable the necessary preventative measures to be taken. Moreover, discussion and analysis of the treatment methods used will alert readers to the efficacy of the various imaging modalities and the conservative and surgical techniques used and in turn open up global discussion on this complication of AL after proctectomy. Thus, by analyzing this lesser-known complication and previous treatment experience, we have highlighted the diagnostic methods used for RSVF, to prompt effective and reasonable treatment to avoid misdiagnosis and mistreatment.

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