


## CASE REPORT

# Appendicorectal fistula with a novel solution: First case report and review of the literature

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## Abstract

Appendicorectal fistula can be a cause of chronic abdominal pain, forming years after an occult episode of appendicitis. It can be diagnosed with Colonoscopy and Magnetic Resonance Imaging, and successfully treated surgically with laparoscopic appendicectomy and stapled segmental cuff resection of the rectum.

## KEYWORDS

appendix, circular stapler, colorectal, cuff resection, fistula, rectum

## 1 | INTRODUCTION

Appendiceal fistula is a rare but recognized phenomenon. Due to the uncommon presentation of appendiceal fistula affecting the gastrointestinal tract and the challenges it presents to a surgeon, we discuss the symptoms, preoperative investigations, and our surgical approach to the first reported case of an appendicorectal fistula.

Important pathologies of the appendix include acute appendicitis, appendicorectal diverticulitis, inflammatory bowel disease, and malignancy. Appendicitis is a common emergency presentation, and delayed diagnosis or untreated appendicitis can lead to perforation, abscess formation, peritonitis, and rarely death. A rare but recognized

complication of acute appendicitis is the phenomenon of appendiceal fistula.

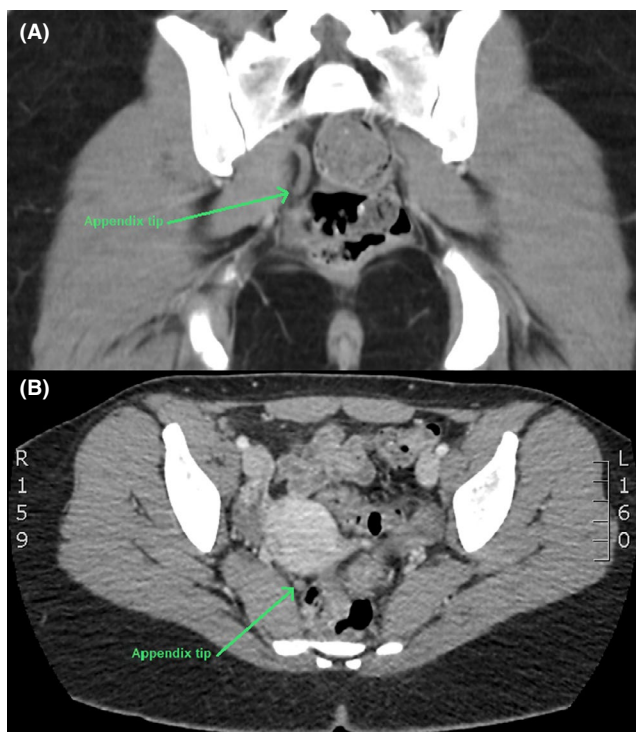
Appendiceal fistula was first described in 1846 when an appendicocutaneous fistula was discovered at autopsy.<sup>1</sup> In 1957, appendiceal fistula was defined as “the primary perforation of the appendix to an adjacent hollow viscus or to the skin.”<sup>2</sup> The hypermobility of the appendix makes it susceptible to adhering to any intra-abdominal organ.<sup>3,4</sup> Various types of appendiceal fistulae have been reported including duodenum<sup>5</sup>; jejunum<sup>6</sup>; Meckel’s diverticulum<sup>7</sup>; ileum<sup>8</sup>; cecum<sup>9</sup>; ascending colon<sup>6</sup>; sigmoid colon<sup>6</sup>; ureter<sup>10</sup>; urinary bladder<sup>11</sup>; tubo-ovarian<sup>12</sup>; uterus<sup>13</sup>; vagina<sup>14,15</sup>; aorta<sup>16</sup>; right iliac artery<sup>17</sup>; and cutaneous<sup>3</sup> (umbilicus,<sup>18</sup> right inguinal hernia,<sup>19</sup> right buttock,<sup>20</sup> right psoas,<sup>21</sup> right loin,<sup>22</sup> trauma<sup>23</sup>).

Reported appendiceal pathologies leading to appendiceal fistula include acute appendicitis, periappendiceal abscess, incomplete appendectomy,<sup>24</sup> malignancy (mucinous adenocarcinoma<sup>25</sup>), goblet cell carcinoid,<sup>26</sup> isolated Crohn's disease,<sup>27</sup> appendiceal diverticulitis,<sup>28</sup> papillovillous adenoma,<sup>29</sup> and neuroma.<sup>30</sup> Reports of adjacent pathologies leading to appendiceal fistula include malignancy (cervical squamous cell carcinoma<sup>31</sup>); sigmoid diverticulitis<sup>32</sup>; Hirschsprung's disease<sup>33</sup>; cystic fibrosis<sup>34</sup>; abdominal aortic aneurysm (primary<sup>16</sup> and secondary to repair<sup>35</sup>); arterial reconstruction<sup>36</sup>; and recent or previous surgery (trauma laparotomy,<sup>23</sup> hysterectomy,<sup>37</sup> transurethral resection of prostate,<sup>38</sup> transurethral resection of bladder tumor with Mitomycin C,<sup>39</sup> right inguinal hernia repair,<sup>19</sup> and right groin hernia repair with propylene plug<sup>40</sup>).

Until now, there was no known report of a fistula occurring between the appendix and rectum. Due to the uncommon presentation of appendiceal fistula affecting the gastrointestinal tract, and the challenges it presents to a surgeon, we discuss the symptoms, preoperative investigations, and our surgical approach to the first reported case of an appendicorectal fistula.

## 2 | CASE REPORT

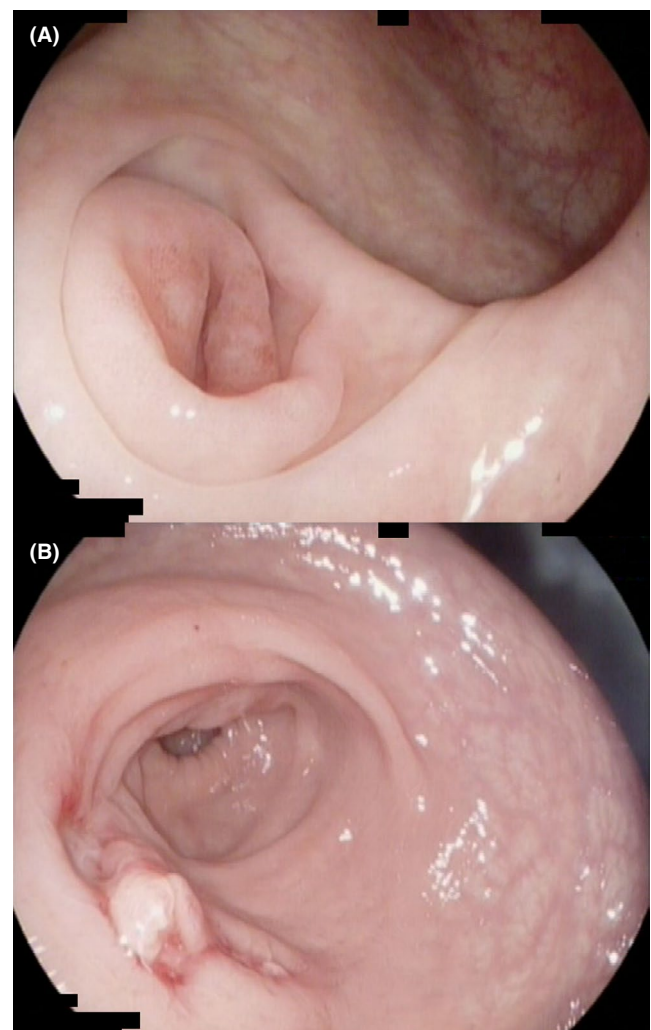
A 23-year-old female experienced an episode of acute abdominal pain that led to a computed tomography (CT) scan of the abdomen. The CT revealed no intra-abdominal pathology



**FIGURE 1** CT Abdomen: (A) Coronal View; (B) Axial View

and her pain resolved spontaneously. However, the CT did demonstrate a long pelvic-oriented appendix with the tip adjacent to the rectum (Figure 1). She went on to experience intermittent pelvic pain every 2-3 months which could last up to thirty minutes at a time. Three years later, she presented to her general practitioner following an episode of severe cramping pelvic pain associated with constipation. The pain subsided after defaecation, but led to large volume diarrhea and the passage of mucous per rectum. She otherwise has no significant past history, no family history of inflammatory bowel disease and no prior colonoscopy.

Abdominal examination revealed mild lower abdominal tenderness, and rectal examination was normal. Pelvic ultrasound was negative. Proctalgia fugax was suspected. A colonoscopy was performed which demonstrated patchy inflammation around the appendiceal orifice and an upper rectal defect suspicious for a fistula. The defect appearance was similar to an open diverticulum with some mucopurulent exudate at the edges as well as a granulation polyp (Figure 2).



**FIGURE 2** Colonoscopy: (A) Periappendicorectal Inflammation; (B) Upper Rectal Defect



**FIGURE 3** MRI Pelvis: (A) Sagittal View; (B) Axial View

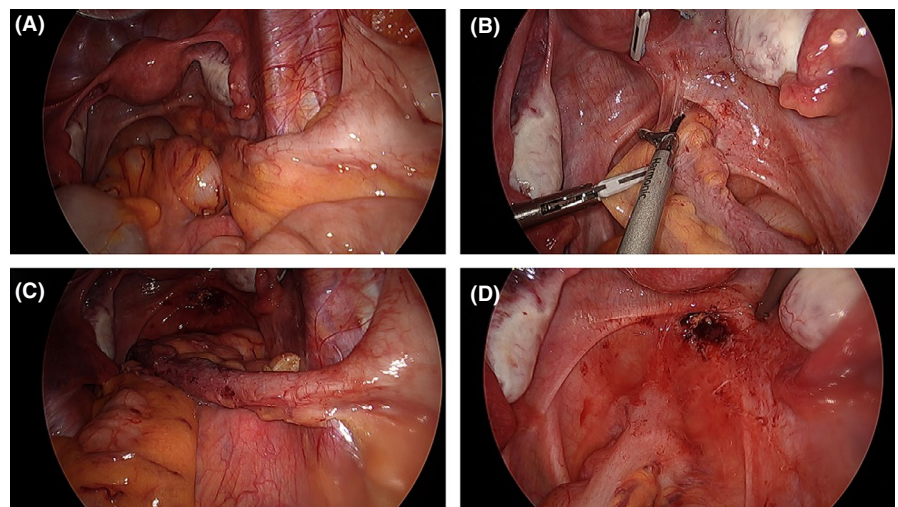
A MRI of the pelvis confirmed a fistulous tract between the tip of a pelvic-oriented appendix and the rectum at about the 12 cm mark (Figure 3).

The patient was referred for a surgical opinion. Together with her surgeon, the patient elected for surgical resection of the appendix and rectum. At the time of the operation, an 80 mm appendix was found extending from the cecum to the anterior rectum and posterior vaginal vault. The tip was dissected sharply from the rectum using an energy device, and a routine appendicectomy was performed. A defect persisted within the rectum and vaginal vault with chronic inflammatory changes around the cut edges (Figure 4). A decision was made to convert to an open Pfannenstiel incision. Small adhesions between the vaginal vault and rectum were taken down. A cuff resection of the affected anterior rectal wall was undertaken using a 29 mm intraluminal circular stapler. The cuff of rectum specimen clearly demonstrated a fistula opening (Figure 5). The staple line was reinforced with 3-0 PDS interrupted sutures, with drain and rectal tubes placed. The rectal tube was removed on day two and the abdominal drain on day four. The patient had an uneventful postoperative course and was discharged on day five. The patient was well upon review at 4 weeks and was discharged from the outpatient clinic.

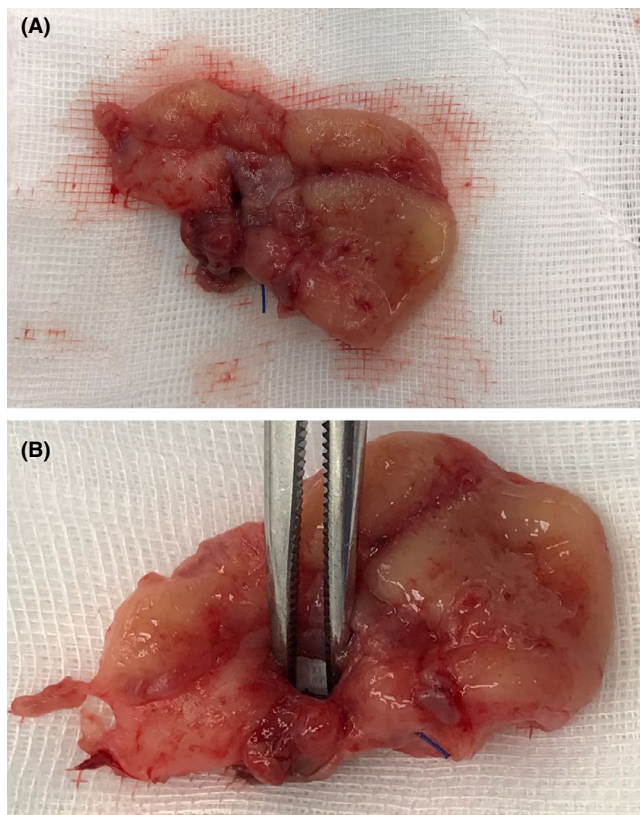
Histological examination of the appendix demonstrated features of mild acute on chronic appendicitis with crypt architectural distortion and numerous non-necrotising granulomata and lymphoid aggregates present throughout the appendiceal wall. Granulation tissue was adherent to the serosal surface of the distal tip of the appendix and the rectal wall showed perforation with granulation tissue adherent to the deep margin extending through the wall to the mucosal surface, consistent with fistula formation.

### 3 | DISCUSSION

A review of the literature demonstrates that a common presentation of “spontaneous” appendiceal fistula is chronic abdominal pain measured in years. Appendiceal fistula formation



**FIGURE 4** Laparoscopy: (A) Appendicorectal Fistula; (B) Dissection; (C) Separation; (D) Rectal Defect



**FIGURE 5** Specimen: (A) Cuff of Rectum; (B) Fistula Opening

should therefore be considered in patients with chronic abdominal pain and a suspected chronic inflammatory process: a distant yet memorable episode of pain may reflect an episode of acute appendicitis. Other risk factors for appendiceal fistula include persistent or recurrent urinary tract infection; previous abdominal surgery; a chronic draining cutaneous sinus or an abscess requiring repeat incision; and drainage. There is also a notable population in patients with cystic fibrosis.<sup>34,41-44</sup>

Optimal surgical treatment of appendiceal fistula will of course be determined by the underlying pathology and structures involved. In our case, the investigation of a suspected case of proctalgia fugax led to the discovery of an appendicorectal fistula by colonoscopy and MRI, which may have been the sequelae of an episode of acute appendicitis on a long pelvic-oriented appendix years earlier. As there was no evidence of malignancy, a local resection of the appendix and rectum was deemed appropriate. The closure of the inevitable defect in the rectum was achieved by novel stapled segmental cuff resection which owes its origins to and is a recognized treatment of endometriosis.<sup>45,46</sup>

## 4 | CONCLUSIONS

This is the first reported case in the published literature of an appendicorectal fistula following what appears to be an acute

on chronic episode of acute appendicitis. Appendiceal fistula is a rare phenomenon that is important to diagnose and can be challenging to treat. Awareness of the types of appendiceal fistula and the various surgical options assists a general surgeon to ensure positive outcomes. Stapled segmental cuff resection of a rectal defect is a novel method of closure and treatment of appendicorectal fistula.

## CONFLICT OF INTEREST

None declared.

## AUTHOR CONTRIBUTIONS

SM: Primary author, reporting and literature searches. JK: Critical revision, final approval. PG: Critical revision, final approval. BM: Critical revision. NL: Critical revision.

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## REFERENCES

1. Volz A. *Die durch Kothsteine bedingte Durchbohrung des Wurmfortsatzes, die häufig verkannte Ursache einer gefährlichen Peritonitis, und deren Behandlung mit Opium*. Carlsruhe, Germany: CF Müller; 1846.
2. Kjellman T. Appendiceal fistulae and calculi: review of the literature and a report of three cases. *Acta Chir Scand*. 1957;113:123-139.
3. Fievé G, Bour P. Aortoappendicular fistula after aortoiliac prosthetic replacement. *Ann Vasc Surg*. 1989;3:187-189.
4. Matsuura K, Takahara Y, Sudo Y, Ishida K, Nakajima N. A case of fistula of the right iliac aneurism to the appendix. *Ann Vasc Surg*. 2002;16:768-770.
5. Pohlman AG. The appendix forming a duodenal-cecal canal. *JAMA*. 1908;50:961.
6. Walker LG Jr, Rhame DW, Smith RB III. Enteric and cutaneous appendiceal fistulae. *Arch Surg*. 1969;99:585-588.
7. Gile JF, MacCarty WC Jr. Calcified concretions within a Meckel's diverticulum. *Radiology*. 1943;41:494-504.
8. Courty A. Fistule iliaque après appendicite méconnue datant de 17 ans. *Monstpellier Chir*. 1954;6:410-411.
9. Thomas SF. Appendiceal coproliths: their surgical importance. *Radiology*. 1947;49:39-49.
10. Golimbu M, Morales P, Becker MH. Ureteroappendiceal fistula in child. *Urology*. 1974;3:370-372.
11. Keen WW. A case of appendicitis in which the appendix became permanently soldered to the bladder like a third ureter, producing a urinary faecal fistula. *Trans Am Surg Assoc*. 1898;16:243.
12. Hoffer FA, Ablow RC, Gryboski JD, Seashore JH. Primary appendicitis with an appendico-tuboovarian fistula. *Am J Roentgenol*. 1982;138(4):742-743.
13. Sarkar DK. Appendico-uterine fistula: case report. *Va Med*. 1985;112(1):47-48.

14. Kneece SM, Wackym PA, Dudley BS, Sawyers JL, Gray GF Jr. Appendicovaginal fistula and primary appendiceal cystadenocarcinoma. *South Med J*. 1987;80(7):914-916.
15. Chatra PS, Vinayaka US. Appendicovaginal fistula – a rare complication of appendicitis. *Internet J Radiol*. 2012;14(10). <https://print.ispub.com/api/0/ispub-article/13909>
16. Monaghan K, Cogbill TH. Primary aortoappendiceal fistula: case report and review of the literature. *J Vasc Surg*. 2002;35(6):1284-1286.
17. Tyson RR, Maier WP, DiPietrantonio S. Iliaco-appendiceal fistula following dacron aortic graft. *Am Surg*. 1969;35(4):241-243.
18. Collins DC. A study of 50 000 specimens of the human vermiform appendix. *Surg Gynecol Obstet*. 1955;101:437-445.
19. Koak Y, Jeddy TA, Giddings AE. Appendico-cutaneous fistula. *J R Soc Med*. 1999;92(12):639-640.
20. Hedner J, Jansson R, Lindberg B. Appendico-cutaneous fistula. A case report. *Acta Chir Scand*. 1978;144(2):123-124.
21. Singh MP, Samaiya A, Sainia TK, Saxena A. Spontaneous appendico-cutaneous fistula: an unusual presentation of retroperitoneal appendicular perforation associated with psoas abscess. *J Surg Case Rep*. 2016;2016(6):rjw112.
22. Deodhar SD, Jammihal HJ, Patel NP. Appendicular abscess presenting as a sinus in the right loin (a case report). *J Postgrad Med*. 1982;28:41A-42A.
23. Faulkner G, Pettit SH. Post-traumatic appendicular–cutaneous fistula: a case report. *Ann R Coll Surg Engl*. 2010;92(1):e6-e7.
24. Hyett A. Appendicocutaneous fistula: a hazard of incomplete appendectomy. *ANZ J Surg*. 1995;65(2):144-145.
25. Dalton DP, Dalkin BL, Sener SF, Pappas PS, Blum MD. Enterovesical fistula secondary to mucinous adenocarcinoma of appendix. *J Urol*. 1987;138(3):617-618.
26. Di Paola M, Stockwell WS. Appendico-vesical fistula due to an appendix abscess with associated goblet cell carcinoid of the appendix. *Br J Urol*. 1976;48(6):436.
27. Morita H, Murakami H, Kawai S, Kobori O. Appendical fistulae formation as a complication of primary Crohn's disease prior to surgical management: report of a case. *Surg Today*. 1996;26(5):340-344.
28. Kawamura YJ, Sugamata Y, Yoshino K, et al. Appendico-ileo-vesical fistula. *J Gastroenterol*. 1998;33(6):868-871.
29. Timmermans LG, Casselman J, Defloor E. Association of an appendicovesical fistula and an appendiceal adenoma. Case report. *Acta Chir Belg*. 1992;92(1):60-62.
30. Lund PG, Krogh J. Appendicovesical fistula associated with neuroma of the appendix. *Urol Int*. 1988;43(6):362-363.
31. Monjero-Ares I, Gegúndez-Gómez C, Rielo-Arias FJ, et al. Iliac-appendiceal fistula. An unusual cause of gastrointestinal bleeding. *Rev Esp Enferm Dig*. 2012;104(7):387-388.
32. Van Hillo M, Fazio VW, Lavery IC. Sigmoidoappendiceal fistula—an unusual complication of diverticulitis. Report of a case. *Dis Colon Rectum*. 1984;27(9):618-620.
33. Abubakar AM, Pindiga UH, Chinda JY, Nggada HA. Appendicovesical fistula associated with Hirschsprung's disease. *Pediatr Surg Int*. 2006;22(7):617-618.
34. Cakmak MA, Aaronson I. Appendicovesical fistula in a girl with cystic fibrosis. *J Pediatr Surg*. 1997;32(12):1793-1794.
35. Chiche L, Pitre J, Sarfati PO. In situ repair of a secondary aortoappendiceal fistula with a rifampin-bonded Dacron graft. *Ann Vasc Surg*. 1999;13:225-228.
36. Risberg B, Kewenter J. Arterio-appendiceal fistula after arterial reconstruction with synthetic graft. *Acta Chir Scand*. 1979;145(2):121-123.
37. Brenner AS, Baldin A, Molteni R, et al. Appendico-cutaneous fistula following hysterectomy: first case report. *J Coloproctol (Rio J)*. 2017;37(2):157-159.
38. Doherty AP, Whitfield HN. A case of appendico-vesical fistula. *J R Soc Med*. 1992;85(12):757.
39. Feng TS, Rusnack S, Kaminski J, Fleshner P. Appendicovesical fistula following transurethral resection of bladder tumor with mitomycin C. *Urology*. 2016;89:e1-e2.
40. Wijers O, Conijn A, Wiese H, Sjer M. Appendico-cutaneous fistula 20 years after groin hernia repair with a polypropylene plug. *BMJ Case Rep*. 2013;6:2013.
41. Ziparo V, Bocchetti T, Balducci G, et al. An appendiceal-vesical fistula in a patient with cystic fibrosis. *G Chir*. 1998;19(10):387-389.
42. Schier F. Appendico-ileal fistula in cystic fibrosis. *J Pediatr Surg*. 2000;35(4):634-635.
43. Rogers TN, Joseph C, Bowley DM, Pitcher GJ. Appendico-cutaneous fistula in cystic fibrosis. *Pediatr Surg Int*. 2004;20(2):151-152.
44. Morris-Stiff GJ, Islam KA. Appendico-colic fistula complicating appendicitis in cystic fibrosis. *BMJ Case Rep*. 2010;7:2010.
45. Gordon SJ, Maher PJ, Woods R. Use of the CEEA stapler to avoid ultra-low segmental resection of a full-thickness rectal endometriotic nodule. *J Am Assoc Gynecol Laparosc*. 2001;8(2):312-316.
46. Woods RJ, Heriot AG, Chen FC. Anterior rectal wall excision for endometriosis using the circular stapler. *ANZ J Surg*. 2003;73(8):647-648.

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