



CASE REPORT

Colo-appendico-duodenal fistula: Rare presentation of extrapulmonary tuberculosis

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Abstract

A colo-appendico-duodenal fistula is a rare occurrence that results from extrapulmonary tuberculosis (TB) complications, especially in the endemic region.

KEYWORDS

antitubercular agents, fistula, gastrointestinal tuberculosis, tuberculosis

1 | INTRODUCTION

Colo-appendico-duodenal fistula secondary to tuberculosis (TB) is rarely reported. A 63-year-old man with a previous history of pulmonary TB presented with chronic abdominal pain and diarrhea. Investigations revealed a complex abdominal organ fistula, which was surgically corrected. He completed the anti-TB treatment for a total duration of 1 year.

A fistula is defined as an abnormal connection between two epithelialized surfaces. Fistula formation normally occurs in the abdomen. These may include digestive tract components or adjacent organs, such as kidneys, bladder, uterovaginal tract, and even major vessels.¹ Generally, fistulae involving more than two organs are rarely reported. There is a myriad of etiologies for fistula formation, among those reported include malignancy, previous abdominal

surgery, inflammatory bowel disease, cystic fibrosis, and tuberculosis (TB).²⁻⁵ TB is deemed endemic in Malaysia, with 29,000 new cases annually and an incidence rate of 92 cases per 100,000 population in 2018.⁶ From this, there are a minority of cases that involved extrapulmonary TB, including gastrointestinal TB.⁷⁻⁹ Hence, this report presents a rare case of a colo-appendico-duodenal fistula, which to the extent of the authors' knowledge, is the first case reported in our region.

2 | CASE REPORT

A 63-year-old gentleman presented with central abdominal pain for 2 years associated with loose stools, lethargy, and marked weight loss. He denied fever, night sweats, or respiratory symptoms. He has a previous history of

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pulmonary TB over the past 10 years and completed treatment for 6 months. In addition, he was previously diagnosed to have bilateral renal calculi with moderate renal impairment. The patient had no previous history of abdominal surgery but was cachexic. Examination of the abdomen was unremarkable, and no organomegaly or lymphadenopathy was noted. The systematic examination, including the respiratory system, appeared normal. Blood investigations showed macrocytic anemia with hemoglobin level of 9.6 g/dl (normal range: 11.6–15.1), mean cell volume of 102.6 fl (normal range: 80.6–95.5), folate level of 2.7 ng/ml (normal range: 2–10), and normal vitamin B12 level. Thyroid function and stool samples were normal.

Esophago-gastro-duodenoscopy and colonoscopy revealed fistulous openings in the third part of the duodenum and proximal transverse colon. However, there was no evidence of malignancy or inflammatory bowel disease. Computed tomography (CT) scan showed irregular thickening of the transverse and descending colon, extending proximally up to the hepatic flexure. There was contrast opacification of small bowels adjacent to the transverse colon, but no significant communication could be demonstrated. Multiple subcentimeter hypodensities were also noted in both liver lobes with focal calcification, as well as a granuloma at the left lung base. Overall findings were suggestive of disseminated TB. A barium enema was also done; however, it was unable to demonstrate the fistula configuration.

A joint decision was made to start him on anti-TB treatment empirically for 6 months before surgery, as given by the radiological findings. However, extensive investigations including sputum stains, culture, and polymerase chain reaction were tested negative for TB. Meanwhile, the stool microscopy was unremarkable. An exploratory laparotomy was performed subsequent to completing the intensive phase of anti-TB treatment. Intraoperatively, a colo-appendico-duodenal fistula involved the transverse colon, proximal appendix, and D3 (Figure 1). Passage of the fistula was confirmed with an appendicostomy and passage of a feeding tube into the duodenum and transverse colon. The liver appeared normal, and there were no peritoneal nodules or enlarged mesenteric nodes. Wedge resection of both colonic and duodenal ends of the fistulae was performed with linear cutter staplers, followed by an appendectomy (Figures 2 and 3). The appendix and fistula tract histology revealed no evidence of active TB infection, malignancy or transmural inflammation. TB-polymerase chain reaction for the resected specimen was negative. During the outpatient review, he had a resolution of his symptoms and completed the anti-TB medication in 1 year.

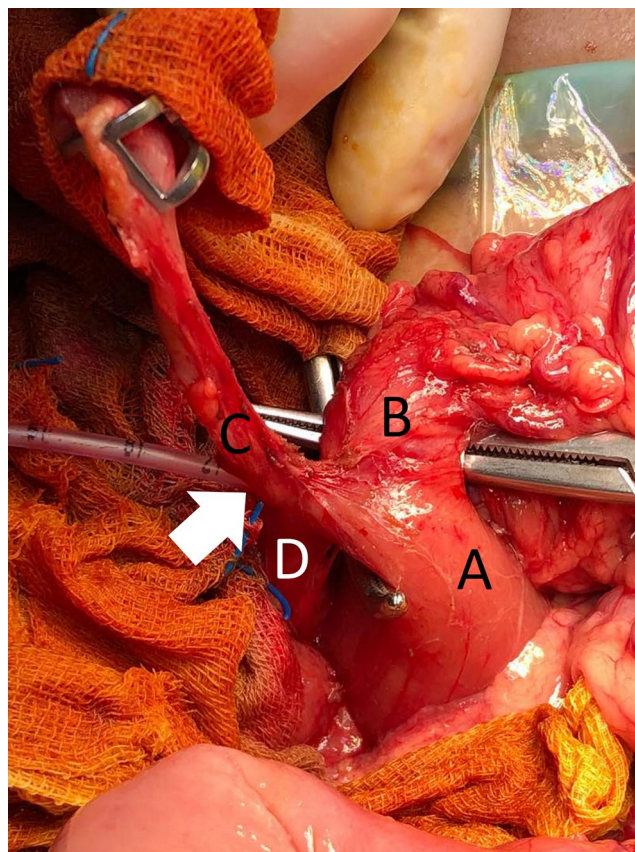


FIGURE 1 Fistulation between the transverse colon (A), duodenum (B), and appendix (C) separated by forceps. Ryle's tube was inserted through the lumen (arrow) of the appendicular base (D), demonstrating patency of the fistula into the transverse colon

3 | DISCUSSION

Duodeno-colonic fistula has been widely reported; however, to the authors' knowledge, this is the first reported case of colo-appendico-duodenal fistula. Appendico-duodenal fistulae are much rarer, and to date, there has only been one case of fistulation between the duodenum and a peri-appendiceal abscess cavity in literature.¹⁰ More commonly, appendico-enteric fistulae tend to occur in post-operative patients involving gastro-jejunostomies, colon, and rectal anastomosis.¹¹ In this case, this research postulated that the underlying history of pulmonary TB was the culprit of the occurrence of gastro-intestinal TB despite a negative preoperative diagnosis. The radiological findings of TB and endoscopy that had ruled out malignancy/inflammatory bowel disease had resulted in the decision-making for empirical anti-TB therapy.

Abdominal TB may manifest as diffuse, white-yellowish peritoneal nodules measuring 3–5 mm. They may also form bowel adhesions, strictures, fistula, ulcers, and pseudopolyps. Traditional acid-fast bacilli stain is only positive in 25% of ascitic fluid, whereas ascitic adenosine deaminase (ADA) is highly sensitive for TB.¹² Definitive

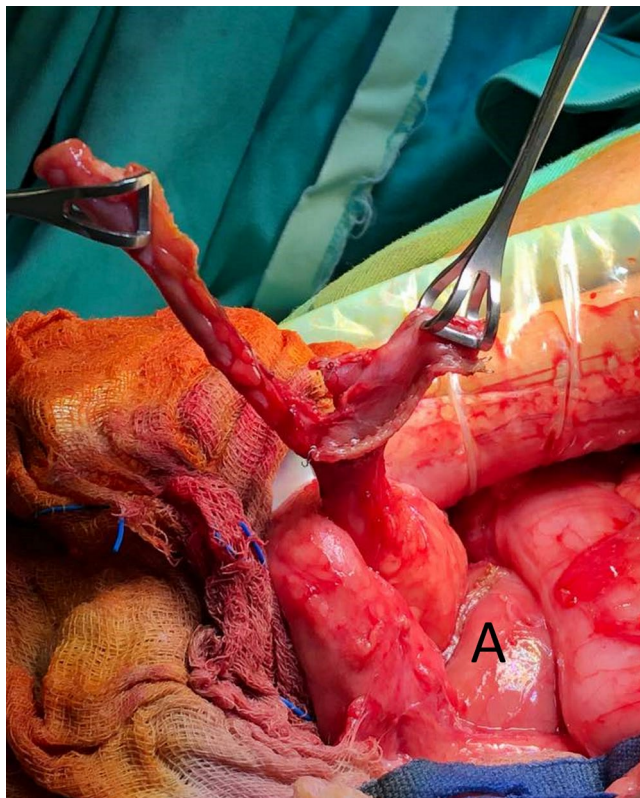


FIGURE 2 Appendix with a stapled colonic (A) and duodenal ends of the fistula

diagnosis can be attained via histology of a resected nodule or polymerase chain reaction.¹² Histopathological studies may show the typical necrotizing granuloma containing macrophages, lymphocytes, Langhans giant cells, and caseous necrosis. However, in general, the sensitivity of the tissue diagnosis is only 80% which might substantiate the rationale of negative tissue or pathologic findings in the extrapulmonary TB patient.¹³ Due to the poor yield of acid-fast bacilli in sputum, it is common for patients to be started on TB treatment empirically in endemic nations such as Malaysia. This may explain the lack of typical operative and histological features, but it is justified by the reduction in surgical complication rates, such as wound dehiscence, anastomotic leak, and enterocutaneous fistula.¹⁴

Generally, various differential diagnoses must be contemplated in this case, as they can be classified as either medical or surgical etiologies. Hyperthyroidism, bacterial infection, and parasitism can be considered but were excluded after the biochemical investigations. Surgical conditions, namely, malignancy and inflammatory bowel disease, can also be reckoned; however, the intraluminal causes were then sought with negative endoscopic assessment. The patient in this report presented symptoms which are consistent with most reported cases of the duodeno-colonic fistula, which included chronic

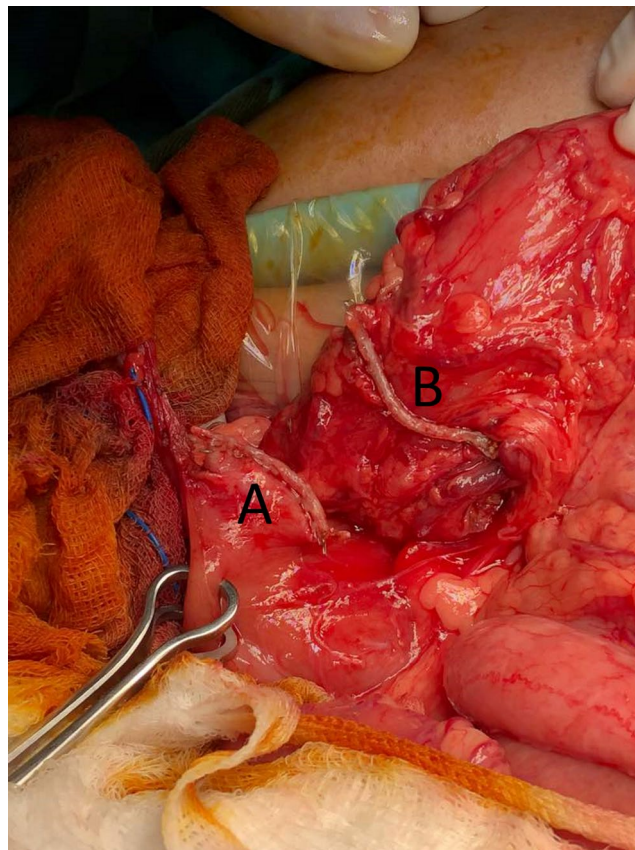


FIGURE 3 Stapled colonic (A) and duodenal (B) ends of the fistula post-appendectomy

diarrhea, abdominal pain, and malnutrition.¹⁵ Diarrhea and abdominal pain have been attributed to the colonization of the small bowel by colonic flora. This factor has also been suggested to cause macrocytic anemia. Other possible mechanisms include colonic irritation due to the passage of duodenal content via the fistula and lipid malabsorption. On the contrary, feculent vomiting is rare due to the presence of pyloric sphincter.

In this work, the patient was treated with colonic wedge resection and duodenal fistula sites with primary closure using surgical staplers with good effect. Meanwhile, other authors tend to perform colectomies even for benign causes. Nevertheless, these are necessary for patients with obstructing lesions or when malignancy is suspected. The duodenal component is usually excised with a partial duodenectomy, followed by either primary repair or a serosal patch from the jejunum. However, a pancreatoduodenectomy is indicated in larger fistulae or malignancies, especially in primary duodenal or pancreaticobiliary tumors. The use of parenteral nutrition is also important in nutritional rehabilitation in malnourished patients who do not require emergent surgery. Most importantly, the initiation and continuation of anti-TB medication remain the cornerstone of treatment in any extrapulmonary TB.³ During follow-up, patients need to be reviewed on the resolution

of the gastrointestinal symptoms and the completion of anti-TB medications within a period of at least 9 months to 1 year.

4 | CONCLUSION

A colo-appendico-duodenal fistula is a rare form of the gastrointestinal fistula, which can be considered in patients presenting with chronic loose stools, lethargy, and weight loss. TB can be the culprit despite negative laboratory findings, especially among ex-pulmonary TB patients. Laparotomy, resection, and primary anastomosis of the fistula can be applied once the anti-TB medication has been initiated.

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CONFLICT OF INTEREST

None declared.

AUTHOR CONTRIBUTION

DEYG and AOP involved in drafting the manuscript and provided the literature review. RS provided the expert opinion. FH involved in revising the manuscript. All authors approved the final version of the manuscript and agree to be accountable for all aspects of the work ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

CONSENT

Verbal and written informed consent was obtained from the patient to participate in this report.

DATA AVAILABILITY STATEMENT

The data used and/or analyzed during the current report are available from the corresponding author on reasonable request.

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