

Case Report

Tuberculous Appendicitis Presenting with Lower Gastrointestinal Hemorrhage - A Case Report and Review of the Literature

Kittichai Kuntanapreeda MD*

* Department of Surgery, Faculty of Medicine, Thammasat University, Pathumthani

The author presents a young patient presenting with lower gastrointestinal hemorrhage due to tuberculous appendicitis. Though an ileo-cecal region is the most frequent site of involvement of the intestinal tuberculosis, vermiform appendix involvement is rare. Moreover, intestinal tuberculosis is an uncommon cause of lower gastrointestinal hemorrhage. Therefore, bleeding from tuberculous appendicitis is a very rare presentation. In addition, the author demonstrates colonoscopic appearance of the tuberculous appendicitis.

Keywords: Intestinal tuberculosis, Tuberculous appendicitis, Lower gastrointestinal hemorrhage

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Intestinal tuberculosis is still common in developing countries⁽¹⁾. Patients may be either asymptomatic or have chronic abdominal pain, obstructive symptoms, weight loss, diarrhea, and/or abdominal mass⁽²⁾. A few patients might present with acute complications including intestinal obstruction, hemorrhage, and perforation. Intestinal tuberculosis is a rare cause of lower gastrointestinal hemorrhage⁽²⁾. Although an ileo-cecal region is the most frequent site of the intestinal tuberculosis, vermiform appendix involvement is uncommon^(2,3). So bleeding from tuberculous appendicitis is a very rare presentation.

Case Report

A 20-year-old male patient presented with a history of acute bleeding per rectum for 2 days. The appearance of bleeding was dark-red blood mixed with stool. The bleeding was not accompanied by nausea, vomiting, and abdominal pain. There was no history of weight loss, chronic abdominal pain, chronic fever, chronic diarrhea, and contact with a tuberculous patient.

On physical examination, the patient was found to be normotensive, normothermic, and had no tachycardic. General condition was healthy, normosthenic build, and not pale. No peripheral lymphadenopathy.

Correspondence to: Kuntanapreeda K, Department of Surgery, Faculty of Medicine, Thammasat University, Pathumthani 12120, Thailand, E-mail: kittkult@yahoo.com

Respiration had no adventitious sound. Abdominal examination was soft, not tender, and no palpable mass.

Rectal examination and proctoscopy revealed maroon-bloody stool without any evidence of bleeding point.

Hematocrit was 40%. Total leukocyte count was 8,000/mm³ and a differential count showed 61% of PMNs, 20% of lymphocytes and 2% of eosinophils. Serum albumin and globulin were 3.4 and 4.2 gm/dL respectively. Anti-HIV was found non-reactive. Coagulogram was within normal limit. His chest X-ray was unremarkable.

Colonoscopy was performed 8 hours later, after complete bowel preparation. The findings were blood staining along the mucosa from rectum to cecum, but no ileal blood staining. A small congested lobulated-mass protruded from the appendiceal opening with blood clot adherence (Fig. 1). There was no ulcer at the cecum and terminal ileum. No diverticulum, angiodysplasia, nor polyps were noted. Multiple biopsies were performed at the mass.

The bleeding ceased spontaneously, with no further episode of bleeding. Histopathological report revealed acute colitis, and no evidence of granuloma, dysplasia, and malignancy.

Repeated colonoscopy for re-biopsy of the mass was performed 1 month later. Colonoscopic finding still revealed a lobulated-mass larger than the

previous, with congestion protruding from the appendiceal opening (Fig. 2). There were a few new superficial clean-based ulcers on the terminal ileum and another one on the ileo-cecal valve. The intervening mucosa, however, was uninvolved (Fig. 3, 4). No deformity of ileo-cecal valve and cecum were found. Multiple biopsies were performed at the mass and the ulcers.

The second histopathological report revealed focal active colitis, no evidence of granuloma, dysplasia, and malignancy. The patient was informed of the risk

and benefit for appendectomy. Then appendectomy was done through a grid-iron incision. The appendix was found to be enlarged, hard, and thickened, but had no appendiceal mass. The normal mesoappendix was found. Three feet of distal ileum and cecum were normal. No acute and chronic inflammatory changes were found in the pericecal region. A few mesenteric lymph nodes were palpable in 0.5-0.8 cm diameters. Appendectomy was done with wedge cecectomy to ensure that the whole protruded mass was removed.

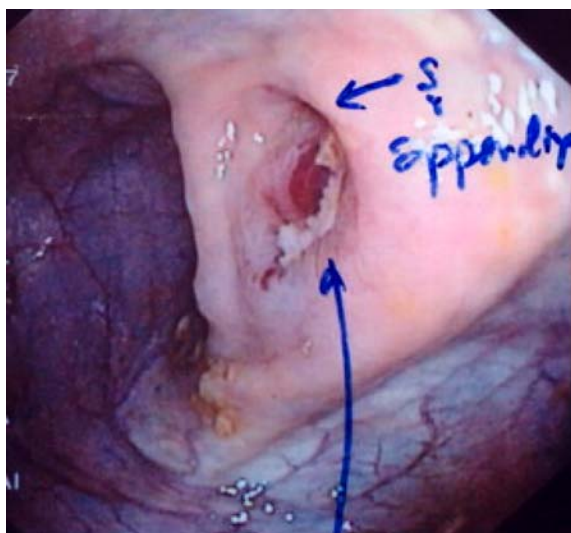


Fig. 1 First colonoscopy: Small protruded mass at appendiceal opening

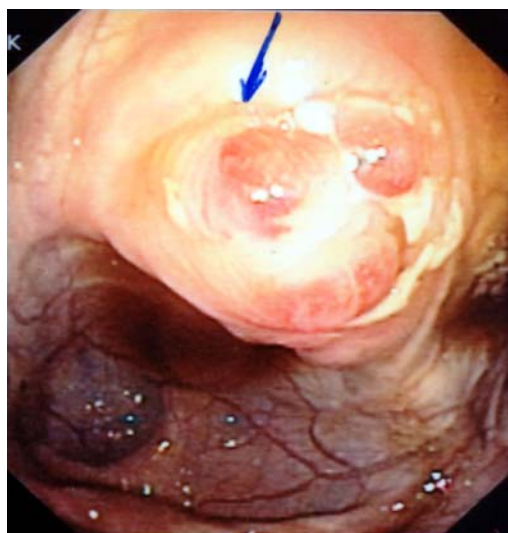


Fig. 2 Second colonoscopy: Larger protruded mass at appendiceal opening

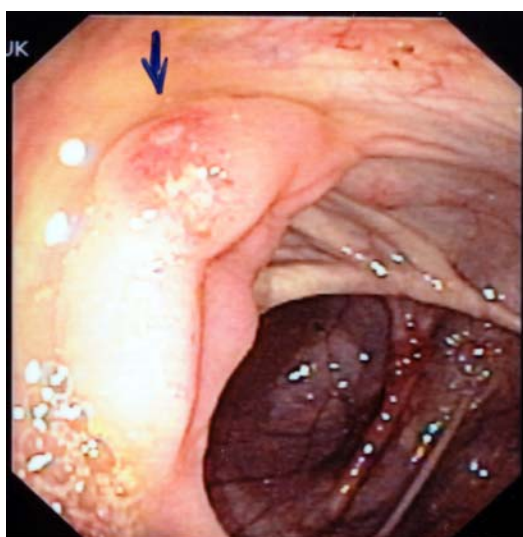


Fig. 3 Superficial ulcer at ileocecal valve

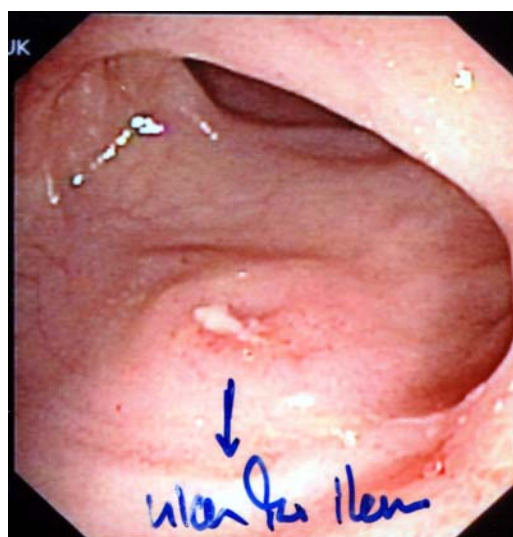


Fig. 4 Superficial ulcer at terminal ileum

Two mesenteric lymph nodes were excised. All specimens were sent for histopathology examination. The patient was discharged 3 days later. Postoperative period was uneventful.

After that, the surgical histopathological report revealed caseating granulomatous inflammation of both appendix (Fig. 5, 6) and mesenteric lymph nodes. Special staining for AFB of both was found positive stain for *Mycobacteria*.

The 3-days of sputum AFB staining was negative for *Mycobacteria*. Definite treatment with anti-tubercular drugs was started.

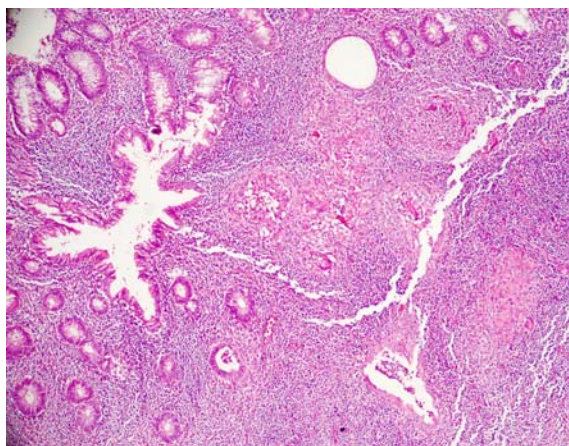


Fig. 5 Histopathology of appendiceal cross section: Show caseating granulomatous inflammation (central) (H&E)

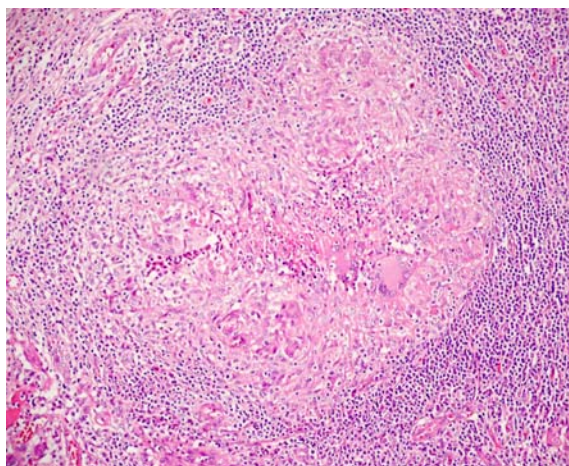


Fig. 6 Magnified view of caseating granulomatous inflammation (H&E)

Discussion

Tuberculosis can affect any tissue and organ in the body. Tuberculosis occurs in extra-pulmonary sites in 10-15% of non-HIV patient and up to 70% of those infected with HIV^(2,4). The extra-pulmonary tuberculosis is composed of tuberculosis of the bones and joints (30%), urinary system (24%), lymph nodes (13%), sexual organs (8%), cerebrospinal meninges (4%), and alimentary system (3%)⁽⁵⁾. Intestinal tuberculosis is the sixth most frequent form of extra-pulmonary tuberculosis⁽²⁾. Only 15-20% of patients with intestinal tuberculosis have concomitant active pulmonary tuberculosis^(2,6). A few patients with intestinal tuberculosis present with acute complications including intestinal obstruction, hemorrhage, and perforation. The ileo-cecal region is the most common site of intestinal tuberculosis, possibly because of the increased physiological stasis, increased rate of fluid and electrolyte absorption, minimal digestive activity, and abundance of lymphoid tissue at this site^(2,7). Although the ileo-cecal region is the most frequent site, such a region is involved in approximately 85% of intestinal tuberculosis, but vermiform appendix involvement is very rare^(2,5). The Shah RC and et al series of 20 specimens of appendix obtained from right hemicolectomies done for ileo-cecal tuberculosis, did not show any evidence of appendiceal involvement⁽⁸⁾.

This is the first case report of tuberculous appendicitis that presented, rarely, with lower gastrointestinal hemorrhage. Common causes of lower gastrointestinal hemorrhage are perianal diseases, angiodysplasia, diverticulum, colorectal cancer, and inflammatory bowel diseases. Intestinal tuberculosis is a rare cause of hemorrhage, overall about 4%^(2,9,10). It has been suggested that colonic tuberculosis increases the capillary vascularity, and small arteries undergo obliterative endarteritis that make bleeding uncommon^(2,9). So bleeding from tuberculous appendicitis is a very rare presentation. Colonoscopy is widely considered as the most useful diagnostic tool for lower gastrointestinal hemorrhage, because of its direct visualization and retrieval of tissue for investigation.

In the presented case, the first colonoscopic finding found only one significant lesion. Congested lobulate-mass protruded from appendiceal opening was the source of bleeding although no active or oozing bleeding were noted, but the presence of adherence blood clot could make the author confident. However, differentiation, the mass between neoplasm and other disease is very difficult. Colonoscopy is an excellent tool to diagnose colonic and terminal ileal diseases⁽²⁾.

In this case, the two colonoscopic biopsies did not reveal a definitive diagnosis. It has been reported that in ranges between 40-80% of the patients, colonoscopic biopsy specimens enable a histological diagnosis of colonic tuberculosis^(4,7,9,11,12). A combination of histology and culture of biopsy can establish the diagnosis in over 60% of cases^(1,2), but the culture of a biopsy specimen is time-consuming. Factors which influence the diagnostic yield of colonoscopic biopsies include the number of biopsies taken, age of the lesions, because in early lesions the characteristic granulomas with caseation may not have developed, and superficial biopsies may miss the submucosa in which characteristic histologic features are present^(1,7). These limitations, then, colonoscopic biopsy specimens do not reveal definitive diagnosis; finally, surgical removal is indicated.

Tuberculous involvement of the appendix in all appendectomies performed is rare, occurring in only about 0.1 to 3%, with an incidence of 1.5 to 30% among patients who are known cases of tuberculosis⁽¹³⁾.

Tuberculosis of the appendix is divided into primary or secondary form. The secondary form, that is well known, may be involved from either ileo-cecal tuberculosis or tuberculosis elsewhere in the body^(11,13,14). The primary form, so-called "isolated" form, has no detectable focus of infection anywhere else in the body, and is extremely rare with an incidence of 0.1 to 0.6%^(3,8,13). The absence of tuberculosis elsewhere in the body after thorough investigations or other pathological foci at laparotomy can conclude the diagnosis of primary tuberculous appendicitis^(11,13-15).

The exact mechanism of the appendiceal involvement remains unclear in either primary or secondary form. The modes of the secondary form infection are hematogenous, retrograde lymphatic spreading, secondary involvement from neighboring tuberculosis, and by ingestion of infected sputum^(1-3,11,13,14). The mode of the primary form infection is considered ingestion of contaminated foods^(3,13). The rarity of primary tuberculous appendicitis has been explained by the minimal contact of the luminal mucosa of the appendix with the intestinal contents^(8,13,14).

Pathologically, the gross appearance may vary from normal to thick walled appendix, very large appendix, or a mass⁽¹⁶⁻¹⁸⁾. The disease within any part of gastrointestinal tract and appendix itself has two main forms^(1,16,18,19). The first form is ulcerative, that may bleed, perforate, or form fistulas. Tuberculous ulcer may be single or multiple. The ulcer is relatively superficial, usually does not penetrate beyond the

muscularis, usually transversely oriented, and the intervening mucosa is usually not involved^(1,2,20). The second form is hyperplastic, which may cause obstruction or present as a mass. The classic histologic finding is a caseating granuloma including central necrosis with a peripheral area of lymphocytes, plasma cells, and Langhans giant cells.

Three clinical presentations of tuberculous appendicitis entity have been described as follows: an acute onset, a chronic, and incidental type^(2,3,13,19). The chronic type is the commonest presentation, which presents with recurrent vague pain or a mass in the right lower quadrant, vomiting, and diarrhea^(3,13). The acute type, often presents as acute appendicitis, occurs due to severe pyogenic infection that is superimposed on the tuberculous appendicitis⁽¹³⁾. Acute bleeding is a rare presentation, as in the presented case. No previous reports about tuberculous appendicitis cause of lower gastrointestinal hemorrhage were found. The incidental type is discovered incidentally in which the organ is unchanged in gross appearance.

In all three clinical presentations, the symptoms and signs, and imaging diagnosis studies are usually non-specific. There is no pathognomonic clinical feature. Pre-operative diagnosis does not alter the management of the acute presentation, as clinical features are similar to acute appendicitis because that must have an appendectomy^(13,16). If tuberculous appendicitis is established in acute presentation as bleeding with spontaneous cessation, should be treated with anti-tubercular therapy. Inversely, in case of either massive bleeding or diagnosis is not established, appendectomy should get rid of appendiceal bleeding. The diagnosis is usually made after histopathological examination of the appendectomy specimen^(3,13).

Because tuberculosis is a systemic disease with localized manifestations, anti-tubercular therapy must be started in all patients either post-appendectomy or non-appendectomy patients, if the pathologic specimen reveals tuberculosis to prevent complications such as sinus or fistula formation^(13,14,16,21). The regimen is the same as for pulmonary tuberculosis at least 6 months including initial 2 months of isoniazid, rifampicin, pyrazinamide, and ethambutol^(1,2). However many physicians extend the treatment duration from 9 to 18 months⁽²⁾.

Conclusion

Because of spreading of HIV/AIDS, tuberculosis is still common in developing and developed countries. Even in endemic areas in which tuberculosis

is common, pre-operative diagnosis of tuberculous appendicitis is very difficult, especially in cases without any manifestation of pulmonary tuberculosis. The author suggests that bleeding from tuberculous appendicitis should be treated with anti-tubercular therapy, except it massive bleeding or diagnosis is not established, surgical treatment is the recommendation. The prevention of misdiagnosis, complications, dissemination, and contiguity, all surgically removed appendix specimens should be histopathologically examined, whether or not the specimens are macroscopically normal.

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รายงานผู้ป่วยและรวบรวมรายงานผู้ป่วยทั้งหมดเรื่องวัณโรคไส้ติ่งอักเสบที่มาด้วยเลือดออกจาก
ทางเดินอาหารส่วนล่าง

กิตติชัย กุลธนปรีดา

ผู้นิพนธ์นำเสนอผู้ป่วยอายุน้อยที่มาด้วยภาวะเลือดออกของทางเดินอาหารส่วนล่าง อันมีสาเหตุจาก
วัณโรคไส้ติ่ง แม้ว่าตำแหน่ง ileo-cecal เป็นที่ที่พบวัณโรคลำไส้มากที่สุด แต่เกี่ยวข้องกับไส้ติ่งเป็นกรณีหายาก
ยิ่งกว่านั้นวัณโรคลำไส้ที่เป็นเหตุให้เกิดภาวะเลือดออกของทางเดินอาหารส่วนล่างพบได้ไม่บ่อย ดังนั้น ภาวะเลือดออก
จากวัณโรคไส้ติ่งเป็นอาการแสดงที่หาได้ยากมาก ผู้ป่วยรายนี้อาจเป็นรายงานผู้ป่วยรายแรก นอกจากนี้ผู้นิพนธ์
ยังสาธิตลักษณะของวัณโรคไส้ติ่งที่เห็นผ่านกล้องส่องตรวจลำไส้ใหญ่
