The Friend Turned Foe: A Rare Presentation of Bilateral Empyema Secondary to the Perforated Appendix

Abstract
Acute appendicitis is one of the most common surgical emergencies encountered. Although studies have reported a rise in the number of cases over the past decade in Western countries, appendicitis is comparatively lower in Asian countries, mainly due to the prevailing dietary habits. Acute appendicitis can further complicate as either appendicular abscess or an appendicular lump or culminate into peritonitis following gangrene/rupture. Almost one-third of the patients with appendicitis present to the hospital with a ruptured appendix. Management of complicated appendicitis is complex, and the diagnosis itself becomes tricky when it presents unusually. Here, we describe the management of such one rare manifestation in a middle-aged female who had concomitant gangrenous appendicitis and bilateral pyothorax. This case report emphasizes that abdominal pathology can lead to bilateral intrathoracic collection without any preexisting thoracic pathology.

Keywords: Appendicitis, gangrenous appendicitis, pleural empyema, pyothorax

Introduction
Appendicitis is one of the most commonly encountered surgical emergencies in both adult and pediatric groups. The lifetime incidence varies from 7% to 9% with a mortality of <1% in uncomplicated appendicitis.\(^1\)\(^-\)\(^4\) However, in complicated appendicitis, the mortality may range from 4% to 6%.\(^5\)\(^,\)\(^6\) Since 2000, the incidence of appendicitis in Asian countries has been increasing, and a study by Khiria et al. also showed that 25% of the patients with appendicitis were complicated at presentation.\(^5\)\(^,\)\(^6\) There have been published reports on unilateral pleural empyema secondary to acute appendicitis, but the patient discussed herein had simultaneously complicated appendicitis and bilateral pyothorax.

Case Report
A 40-year-old female presented to the emergency department with complaints of lower abdominal pain associated with multiple episodes of nonbilious vomiting, loose stools, occasional breathlessness, and decreased urine output over the past 7 days. The patient had history of fever 3 days back but at the time of presentation, was afebrile.

The general physical examination showed a thin patient and a dry tongue, with no pedal edema or generalized lymphadenopathy. Her clinical examination revealed a pulse rate of 130/min, the blood pressure as 96/80 mmHg, oxygen saturation of 90% on room air, a respiratory rate of 34/min, and a body temperature of 98.4°F. Abdominal examination revealed tenderness in the right iliac fossa with no palpable mass but audible bowel sounds.

Her initial blood tests revealed hemoglobin at 98 g/L, a high total leukocyte count at 32.98 × 10\(^9\)/L, and platelet count at 263.4 × 10\(^9\)/L. The renal function test was deranged with urea at 42.83 mmol/L and creatinine at 159.12 \(\mu\)mol/L, but the electrolyte tests were within normal limits. The liver function profile showed total bilirubin at 12.65 \(\mu\)mol/L, direct bilirubin at 6.15 \(\mu\)mol/L, alanine aminotransferase at 8.2 \(\mu\)kat/L, aspartate aminotransferase at 50.9 \(\mu\)kat/L, alkaline phosphatase at 247.6 U/L, gamma-glutamyl transferase at 25.2 U/L, total protein at 57 g/L, and albumin at 30.8 g/L. The hepatitis panel revealed hepatitis C virus positive status. Bilateral moderate pleural effusion could be seen on a plain chest X-ray. Screening two-dimensional echocardiography showed no abnormality.
The patient was then admitted to the surgical department and underwent a brief resuscitation with intravenous normal saline, pain management, supplemental oxygen, and empirical intravenous antibiotic therapy initiated based on ceftriaxone 1 g every 12 h and metronidazole 500 mg every 8 h. After taking written informed consent from the patient, an ultrasound-guided thoracic paracentesis was performed on both sides that revealed a purulent aspirate. The aspirated pleural fluid was then sent for gram staining, acid-fast bacilli (AFB) staining, culture and antibiotic sensitivity, and biochemical analysis. On analysis of the pleural fluid, the lactate dehydrogenase level was found to be raised at 68,900 U/L. Serum procalcitonin was also elevated. An abdominal ultrasound scan detected intrabdominal collection.

Two hours later, because of bilateral pyothorax and intra-abdominal collection findings, the patient was further evaluated by a contrast-enhanced computed tomography (CT) scan of the torso. The axial view of the lower thorax showed bilateral pleural effusion with multiple subcentimetric ground-glass centrilobular nodules in all lobes of the right lung [Figure 1]. Findings suggestive of acute appendicitis with rupture at its tip and adjacent collection extending in the right posterior pararenal and perihepatic spaces were noted on horizontal sections of the abdomen [Figure 2]. A suspicious communication was also noted in the sagittal plane between the peritoneal and pleural cavities [Figure 3].

As the patient complained of right-sided abdominal pain and breathlessness at presentation, our differentials included COVID-19 pneumonia, hollow viscus perforation with acute respiratory distress syndrome, disseminated tuberculosis, and non-Hodgkin lymphoma with chylothorax. The possibility of acute appendicitis was added in our differential diagnosis after eliciting tenderness in the right iliac fossa region.

The reverse transcription-polymerase chain reaction (RT-PCR) test, done keeping COVID-19 pneumonia in mind, was found to be negative. An erect chest X-ray showed bilateral pleural effusion but no free air under the right dome of the diaphragm. The pleural fluid analysis was favoring the infective nature of the effusion. Adenosine deaminase level was high at 223.5 U/L, and the triglyceride levels in the pleural fluid were found to be within normal limits. After an ultrasound scan, it was found that the patient had pleural effusion with intra-abdominal collection; then, we narrowed our differentials to disseminated tuberculosis and appendicular perforation.

After passing the necessary information regarding the patient’s condition to the attendants, bilateral intercostal tube drainage (ICD) was accomplished via ultrasound guidance under local anesthesia to evacuate the purulent content. An urgent exploratory laparotomy with appendectomy was performed in the night after ensuring an intensive care unit (ICU) bed availability for postoperative management. The intraoperative finding was a retroperitoneal collection of approximately 100 ml close to the retroceccally positioned gangrenous appendix. The
rest of the small and large bowel was unremarkable for any mass or mesenteric lymphadenopathy, with healthy adnexa. During the surgery, the patient developed hypotension, for which injection noradrenaline 8 mg at 2 ml/h was started as an inotrope. The patient was not extubated at the completion of the procedure due to the prevailing chest condition and ongoing ionotropic support.

Postoperatively, the patient was shifted to ICU on ventilatory support for further management. Her ionotropic support was adjusted as per the mean arterial pressure. Gradually, the noradrenaline support had to be increased to 8 ml/hr with another inotrope: injection vasopressin at 2 units/h over 72 h. The postoperative evolution was satisfactory. The average urine output was maintained throughout, and the kidney function tests were found to be within normal limits with urea at 6.7 mmol/L and creatinine at 97 µmol/L. Because of prolonged ventilation and difficult weaning, the patient underwent tracheostomy on the 7th postoperative day. She was started on total parenteral nutrition (TPN) to take care of her nutrition, and the ionotropic support tapered gradually with cessation on the 10th postoperative day. Her bilateral ICD outputs were monitored and removed after achieving complete lung expansion, as evident on a chest radiograph.

The intraoperative collection showed growth of Klebsiella pneumoniae on culture medium. Her pleural fluid culture also revealed K. pneumoniae species, and antibiotics were adjusted as per the sensitivity pattern. Serotyping of K. pneumoniae was not performed due to the unavailability of that test. Blood culture, collected at presentation to rule out hematogenous spread of infection, did not reveal any organism. Pus evacuated from pleural cavity did not show AFB on Ziehl–Neelsen staining, and the cartridge-based nucleic acid amplification test was negative for tuberculosis. Histopathological examination of the appendix revealed transmural necrosis of the appendicular wall, dense acute inflammatory infiltrates, and bacterial colonies.

On postoperative day 9, right-sided ICD removal was possible, but left-sided ICD could be extruded only on the 11th postoperative day. Her nutrition was taken care of initially by TPN administered through a central line followed by Ryle’s tube feeding and ultimately by oral feeds when decannulation was possible on postoperative day 17. The patient was gradually weaned off the ventilator and shifted out of ICU on the 18th postoperative day. A healing laparotomy wound facilitated suture removal after 2 weeks. The patient was further monitored in the high-dependency unit of the general surgery department for another 48 h and was finally sent home on postoperative day 21. During her follow-up visit in the outpatient department, 1 month from the day of the discharge, the patient had no fresh complaints and could carry out her daily activities without apparent effort.

Discussion

Thoracic empyema is a rare complication of appendicitis and requires a high level of suspicion to diagnose. In a study conducted by Vasquez-Rios et al., it was found that about 19% of appendicular masses develop thoracic empyema following conservative management for the same. Pregnancy can also be a risk factor for empyema formation following acute appendicitis. In the case discussed herein, the patient did not have any features of appendicular mass at the time of presentation, making it difficult to suspect an empyema. Only after a CT of the torso and findings on exploratory laparotomy could we arrive at the final diagnosis. The complication of bilateral empyema secondary to gangrenous appendicitis made our diagnosis and perioperative management challenging, even after early intervention. Postoperatively, the patient was on ventilatory support for a prolonged duration, and the weaning off was difficult; the possible reasons could be delayed presentation and severe sepsis.

An initial CT scan report, done outside at a local hospital, did not report any pleural fluid collection, whereas a repeat tomography done at our institute revealed collections in both pleural and abdominal cavities, suggesting the empyema formation secondary to the intra-abdominal pathology. Microbiological analysis of the empyema and the intra-abdominal collection also showed the same species of bacteria, hence, confirming the possible hypothesis of the spread. The pleural fluid analysis demonstrated K. pneumoniae species with sensitivity to amikacin, gentamicin, imipenem, doripenem, and chloramphenicol. Previous studies had reported the isolation of Escherichia coli, Bacteroides, and K. pneumoniae as the common organisms from the pleural fluid.

Studies have suggested that whenever an intrathoracic collection is detected, without prior respiratory pathology, always an intra-abdominal source of infection should be suspected. The intra-abdominal collection following an episode of acute appendicitis might get collected in the dependent areas, especially in the Morrison’s pouch if the patient is bedridden; this can eventually irritate and erode the diaphragm paving the way for collection into the pleural cavity. A pressure gradient between the abdominal and the thoracic cavities also favors the transport of an abdominal collection into the thoracic cavity. Dietrich et al. have suggested that bacterial translocation and spread through lymphatics into the pleural cavity may result in empyema formation following an appendicitis attack. A retroperitoneal collection following an appendicular abscess can gain access to the thoracic cavity via the retrocrural space.

Pleuroperitoneal connection has been reported in patients of chronic kidney disease undergoing ambulatory peritoneal dialysis, but our patient gave no such history and had normal serum creatinine and urine output at the time of presentation.
Thoracic empyema can be associated with pneumonia, history of any previous pneumothorax, tuberculosis, trauma, surgery in the past, and abdominal abscess. Our patient had no chest trauma in the past and pneumothorax in the chest radiograph. The pressure gradient between the abdominal and the thoracic cavities and bacterial spread through lymphatics into the pleural cavity may have resulted in the empyema formation after appendicular perforation.

**Informed consent**

Written informed consent was obtained from the patient for her anonymized information to be published in this article.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initials will not be published and due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

**Acknowledgments**

Both Deepak Rajput and Subramanian Chezhian share co-first authorship. CT images were provided, courtesy of the department of Radio-diagnosis, AIIMS Rishikesh. We are also grateful to all those involved in the patient’s care during her ICU stay.

**Financial support and sponsorship**

Nil.

**Conflicts of interest**

There are no conflicts of interest.

**References**

1. Dixon F, Singh A. Acute appendicitis. Surgery (Oxford) 2020;38:310-7.
2. Naveen K, Sareesh Nn, Satheesha Bn, Murlimanju Bv, S Suhani, Hosapattana Mamatha, *et al.* Appendicitis and appendectomy: A retrospective survey in South Indian population. J Surg Acad 2013;3:10-3.
3. Tierney J, Melvin WV, May AK, Bonatti H. Non-operative management of acute appendicitis in a patient with ulcerative colitis. Surg Infect Case Rep 2016;1:161-4.
4. Ferris M, Quan S, Kaplan BS, Molodecky N, Ball CG, Chernoff GW, *et al.* The global incidence of appendicitis: A systematic review of population-based studies. Ann Surg 2017;266:237-41.
5. Khiria LS, Ardhnari R, Mohan N, Kumar P, Nambiar R. Laparoscopic appendicectomy for complicated appendicitis: Is it safe and justified?: A retrospective analysis. Surg Laparosc Endosc Percutan Tech 2011;21:142-5.
6. Vasquez-Rios G, Calixto-Aguilar L, Pajuelo R, Alarcon W. Thoracic empyema as a rare complication of an appendicular mass: A case study and review of the literature. Case Rep Pediatr 2018;2018:9640397.
7. Tokat AO, Karasu S, Barlas AM, Akgiin YA. Thoracic empyema due to bacterial translocation in acute appendicitis. Tuberk Toraks 2013;61:54-6.
8. Dietrich A, Nicolas M, Iniesta J, Smith DE. Empyema and lung abscess as complication of a perforated appendicitis in a pregnant woman. Int J Surg Case Rep 2012;3:622-4.
9. Lai HW, Loong CC, Tai LC, Wu CW, Lui WY. Incidence and odds ratio of appendicitis as first manifestation of colon cancer: A retrospective analysis of 1873 patients. J Gastroenterol Hepatol 2006;21:1693-6.
10. Yaxley J, Twomey K. Peritoneal dialysis complicated by pleuropertoneal communication and hydrothorax. Ochsner J 2017;17:124-7.
11. Herline A, Burton EM, Hatley R. Thoracic empyema in a patient with acute appendicitis: A rare association. J Pediatr Surg 1994;29:1623-5.