



Pneumobilia Associated with Acute Appendicitis: A Case Report

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Abstract

Acute appendicitis is a frequent cause of acute abdomen. Since its first description in 1886, many clinical signs of appendicitis, with varying specificities and sensitivities, have been introduced. In recent years, ultrasound- and Computer Tomogram (CT) -scans have made confirmation of clinical suspicion more reliable. Yet, even an entity such as acute appendicitis may present as an unexpected diagnostic challenge. We report a case of acute appendicitis in a young, otherwise healthy adult with atypical clinical presentation and sonographically detected, unexplained pneumobilia.

Keywords: Pneumobilia, Appendicitis, Emergency medicine

Case Presentation

A thirty-year-old man presented to our emergency department with fever (39.9°C) and orthostatic hypotension with lypothimia since the day before. He reported a single episode of epigastric pain and alimentary vomiting, three days before onset of fever. Pain and vomiting had promptly resolved with paracetamol and scopolamine. The patient also noticed his faces were more fluid than usual, without increased bowel movement frequency. The patient denied consumption of illicit drugs, foreign travel or close contact with wild animals in recent months. He reported no medical disorders; he did not take any medication regularly.

The patient was awake, alert and fully oriented, he felt tired and dizzy. Physical examination was unremarkable. He had no abdominal discomfort, pain or tenderness. Mc-Burney, Murphy, Rosving, Blumberg and psoas signs were all negative. Digital rectal examination and trans-rectal palpation of the prostate and Douglas pouch were negative.

Systolic blood pressure was 90 mmHg and heart frequency 100 beats per minute (sinus tachycardia at electrocardiography).

Blood tests are shown in Table 1 and were normal except for moderate lymphocytopenia, mild thrombocytopenia, borderline increase of renal retention values, without loss of renal function. Inflammatory markers were raised. Bilirubin was increased, without concomitant increase in any other cholestatic parameters or hepatic enzymes.

Focused abdominal and cardiac ultrasound scans were performed. Cardiac ultrasound showed signs of hypovolemia with hyperdynamic left ventricular function (visually assessed ejection fraction ca. 70%, reference range 55% to 65%), and a normal vena cava diameter (1.3 cm) with complete collapse upon inspiration.

Abdominal ultrasound showed a small fluid collection in the Douglas pouch (Figure 1b) pneumobilia in the left liver lobe (Figure 1c), without any focal or diffuse changes in the liver parenchyma. Gallbladder, biliary ducts, and portal system were normal (Figure 1a). No signs of pneumoperitoneum, ileus, or bowel dilation were observed. Both kidneys appeared normal, showed no focal parenchymatous changes or hydronephrosis. The appendix was not visible.

A diagnosis of severe sepsis was made. Intravenous fluids and empirical intra-venous broad-spectrum antibiotics (piperacillin-tazobactam) were initiated. Blood pressure and heart frequency promptly normalized. An abdominal focus was suspected: either a sonographically unrecognized gangrenous cholangitis or a bilio-enteric fistula. A CT-Scan of the abdomen was obtained.

CT images showed: an enlarged inflamed appendix (length 15 cm), ascending retroceally and reaching into the epigastric region (Figure 2c), localized peritonitis with signs of paralytic ileus and free air along the ascending mesocolon (Figure 2a), as well as localized pneumobilia of the left liver

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Table 1: Vital parameters and blood tests.

Variable	Reference Range	At admission	At discharge
Blood pressure Systolic/diastolic (mmHg)	120/80	90/60	120/80
Heart rate	80-100	100	85
Glucose (mg/dl)	74-106	195	92
Creatinine (mg/dl)	0.8-1.3	1.3	1
eGFR nach MDRD* (ml/min)	>60	>60	>60
Sodium (mmol/L)	136-146	133	140
Potassium (mmol/L)	3.5-5.1	3.2	4.3
Calcium (mmol/L)	2.2-2.7	2.4	2.1
Aspartate aminotransferase (U/L)	<50	28	45
Alanine aminotransferase (U/L)	<50	25	50
Lactate dehydrogenase (U/L)	<247	280	196
Gamma-glutamyltransferase (U/L)	<55	51	201
Lipase (U/L)	<67	5	39
Bilirubin (mg/dl)	0.3-1-2	3.1	1.6
C-reactive Protein (mg/dl)	<0.5	28	13
hs Procalcitonin (ng/ml)	<0.5	35.2	14.5
White-cell count (per nl)	10-Apr	2.8	10.1
Hematocrit (%)	37-53	43	36
Hemoglobin (g/dl)	14-18	15.7	12.8
Platelet count (per nl)	150-400	115	162

*Glomerular filtration rate estimated using the MDRD-Formula [8]

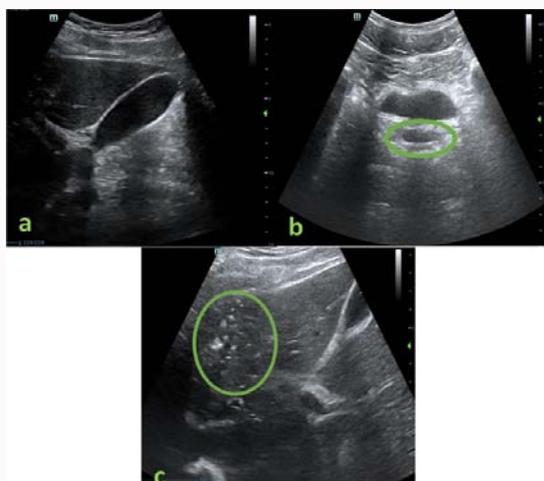


Figure 1: Salient ultrasound findings at admission. (A) Normal sized gall bladder without thickened or swollen walls, nor evidences of cholelithiasis. (B) Small fluid collection in Douglas' pouch (oval). (C) Focal, milliary hyperechoic enhancement of the left liver lobe consistent with pneumobilia (circle).

lobe (Figure 2b). There were no signs of bilio-enteric or bilio-renal fistulae. No radiologic signs of intra-abdominal infection (other than pneumobilia, which without further signs remained unexplained) were noticed.

A diagnosis of acute perforated appendicitis was made and the patient underwent urgent laparoscopic appendectomy. Laparoscopy did not find any fistulae between the appendix and the biliary system.

At surgery, a retrocolic abscess, compatible with the ultrasound findings (Figure 1b), as well as several adhesences between the

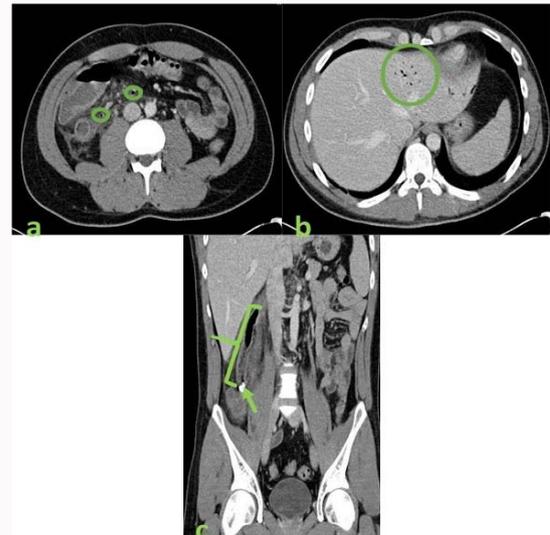


Figure 2: Salient CT-Scan findings at admission: (A) Free air in the abdomen (small circles). (B) Pneumobilia in the left liver lobe (circle). (C) Swollen, inflamed, ca. 15 cm long appendix ascending cranially and medially towards the liver containing an appendicolith (arrow) consistent with an acute perforated appendicitis.

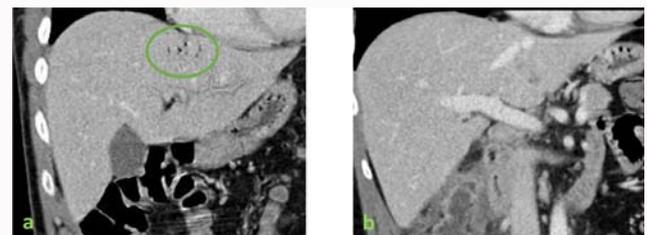


Figure 3: CT-Scan at admission (A) and at 15-days follow-up (B), showing complete resolution of the pneumobilia (circle) after appendectomy.

small intestine and the abdominal wall were observed and surgically detached. Blood and abscess-fluid cultures were positive for *Escherichia coli*.

The patient was discharged home after four days of intravenous antibiotics. Oral antibiotic treatment (ciprofloxacin, antiobigram based), was continued for ten further days. At short-term follow-up, no signs of pneumobilia were seen at CT-Scan (Figure 3), and abnormal blood tests had normalized (Table 1).

Discussion

Acute appendicitis is the most common surgical emergency worldwide [1-5]. Although a well-known and mostly straightforward clinical entity, some patients with acute appendicitis may pose a diagnostic challenge, especially in the emergency setting [6].

The retro-coecal position of the appendix presumably precluded sonographic visualization. In such cases where clinical signs are non-specific and sonography is negative or yields confusing findings, CT-scan may prove diagnostic. We describe a young adult with an atypical and non-specific presentation of appendicitis, probably due the retro-coecal position of the appendix. To our knowledge, no case of pneumobilia associated with acute perforated appendicitis has so far been described.

Lubin J published in 2009 the clinical case of an elderly patient

with necrotizing appendicitis evolving to generalized enteritis/colitis, which resulted in portomesenteric air (but no pneumobilia) most probably as a result of extensive *Pneumatosis intestinalis* [7].

The pathophysiologic mechanism in our case remains obscure, but since both pneumobilia and hyperbilirubinemia promptly resolved after surgery, we postulate that the pneumobilia was associated with acute appendicitis.

Our first clinical hypothesis was that a fistula had formed between the perforated appendix, whose tip reached the epigastrium, and the left biliary tract. The connection would have allowed passage of intestinal air into the biliary tract, impairing biliary flow and causing indirect hyperbilirubinemia. However, this hypothesis could not be confirmed, neither by imaging nor during laparoscopic surgery.

A second hypothesis, most plausible in our case, is that increased pressure in the small intestine (resulting from paralytic ileus) caused passage of air through an intermittently insufficient papilla of Vater into the biliary tract. With restored intestinal motility after surgery, intestinal pressure dropped and Vater's papilla regained its normal function, leading to disappearance of pneumobilia and hyperbilirubinemia. Descriptions of retrograde passage of air into morphologically normal biliary tracts have been reported in the literature [9,10].

Conclusion

Acute appendicitis in a young adult is often a straightforward, easy to recognize and treat condition. Yet, at times, clinical presentation can be confusing, and ultrasound imaging unreliable. This may delay diagnosis with possible unfavorable consequences for the patient.

This case report is to our knowledge the first description of acute appendicitis correlated with pneumobilia and may suggest adding acute perforating appendicitis to the differential diagnosis of otherwise unexplained pneumobilia.

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