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A simple appendicitis? An anatomical pitfall: A case report

Slawomir Wajman¹, Magdalena Gewartowska², Robert Antoniak³, Marek Stanczyk^{1✉}

¹Department of General, Oncologic and Trauma Surgery, Wolski Hospital, Warsaw, Poland

²Electron Microscopy Research Unit, Mossakowski Medical Research Institute, Polish Academy of Sciences, Warsaw, Poland

³Department of Radiology, Wolski Hospital, Warsaw, Poland

ABSTRACT

Rationale: We present a case of appendicitis with an uncommon course due to rare anatomical location of the appendix in the right retroperitoneal space below the diaphragm and above the liver.

Patient's concern: A 32-year-old, previously healthy male with a history of congenital diaphragmatic hernia repair in childhood, presented with 3 days of mild, colicky, central abdominal pain associated with fever, nausea and vomiting. At presentation, pain was localized to the right lower quadrant.

Diagnosis: Even though diagnosis of appendicitis was clear, we decided to confirm it with computer tomography (CT). CT revealed elevation of the right dome of the diaphragm and perforated appendix located above the liver.

Intervention: Appendectomy was performed *via* right subcostal approach instead of usual incision in the right lower quadrant.

Outcome: Patient recovered well and was discharged on the 5th day after operation.

Lessons: Previous congenital diaphragmatic hernia repair may change the location of the appendix. The appendix at rare locations could lead to an uncommon course of appendicitis. On this very note, surgeons should have a high index of suspicion, and CT may help avoid inadvertent complications.

KEYWORDS: Appendicitis; Acute abdomen; Appendectomy; CT; congenital diaphragmatic hernia repair

1. Introduction

The individual lifetime risk of appendicitis is estimated to be 7 percent for women and 9 percent for men. Symptoms overlap with a number of other conditions making diagnosis a challenge, particularly at the early stage of presentation.

We present a case of appendicitis with an uncommon course due to

rare anatomical location of the appendix in the right retroperitoneal space below the diaphragm and above the liver. Although pathophysiology of appendicitis is well known for a century, controversies continue regarding optimal diagnostic strategy and management. We address these controversies with respect to our case.

2. Case report

This study was approved by the Ethical Committee of Wolski Hospital, and informed consent was obtained from the patient.

A 32-year-old, previously healthy male with a history of congenital diaphragmatic hernia repair in childhood, presented with 3 days of mild, colicky, central abdominal pain associated with fever, nausea, and vomiting. At presentation, pain was localized to the right lower quadrant.

Significance

The rare location of the appendix in the right retroperitoneal space below the diaphragm and above the liver could lead to an uncommon course of appendicitis. We report an unusual case of appendicitis in a 32-year-old male who had a history of congenital diaphragmatic hernia repair. Surgeons should have a high index of suspicion, and CT may help avoid inadvertent complications.

✉To whom correspondence may be addressed. E-mail: stanczyk@poczta.onet.pl

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Physical examination revealed tenderness in the right lower quadrant. White blood cell count (WBC) was $20\,700\text{ WBC/mm}^3$, neutrophilic shift to the left-78%, CRP of 33.5 mg/dL (normal $<0.5\text{ mg/dL}$).

Diagnosis of acute appendicitis was made. Alvarado score was 9 confirming the diagnosis. To hasten operation room booking we performed a confirmatory CT scan.

CT revealed: (1) Elevation of the right dome of the diaphragm with compression of right lung (Figure 1); (2) Right colon and kidney were drawn upward above the liver (Figure 1 and Figure 2);

(3) Dilated and perforated appendix, on the Th9/Th10 level (Figure 3 and Figure 4); (4) Periappendiceal mass with abscess and gas bubbles (Figure 3 and Figure 4).

Abdominal cavity was entered using right subcostal incision. During operation, a massive intraperitoneal adhesion confined abscess cavity to the retroperitoneal space above the liver, below the diaphragm. Thus, appendectomy, debridement, irrigation, and drainage of abscess were performed, and then abdomen was closed primarily. Pathology confirmed gangrenous appendicitis. The patient recovered well and was discharged on the 5th day after operation.

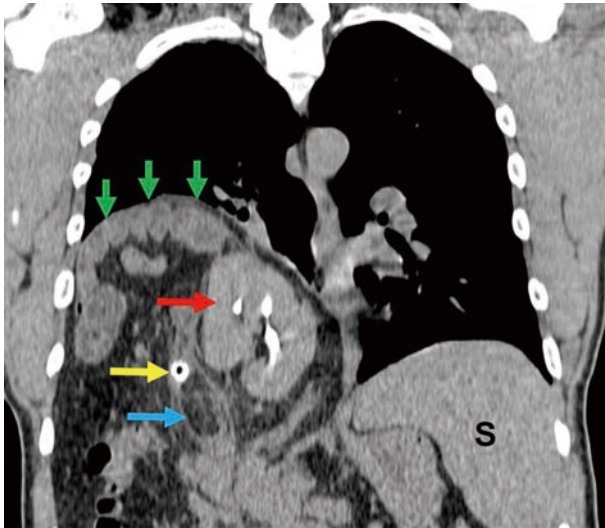


Figure 1. Coronal cut of CT scan of a 32-year-old diagnosed as appendicitis, shows elevation of the right diaphragmatic dome (green arrows), right kidney drawn upward below the diaphragm, backwards and above the liver (red arrow), appendicolith (yellow arrow) and periappendiceal infiltrate in retroperitoneal space (blue arrow). S: Spleen.

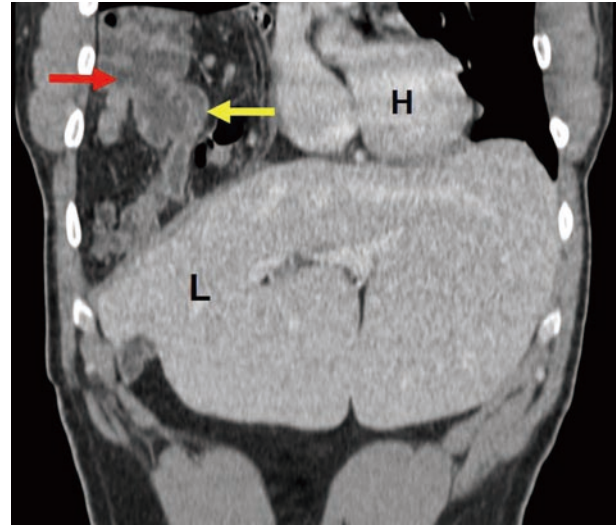


Figure 2. Coronal CT scan shows elevation of the cecum (red arrow) and terminal ileum (yellow arrow) to the right infradiaphragmatic region above the liver. L: Liver; H: Heart.

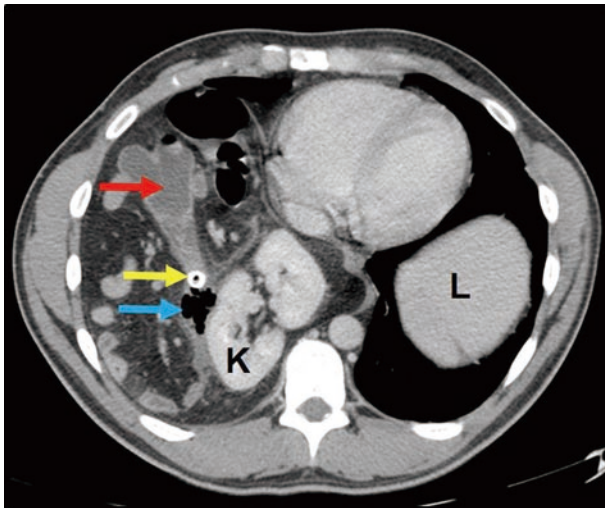


Figure 3. Transverse CT scan demonstrates cecum (red arrow), dilated lumen of appendix with appendicolith (yellow arrow) and secondary to acute appendicitis periappendiceal mass with abscess and gases in the right infradiaphragmatic region (blue arrow). K: Kidney; L: Liver.

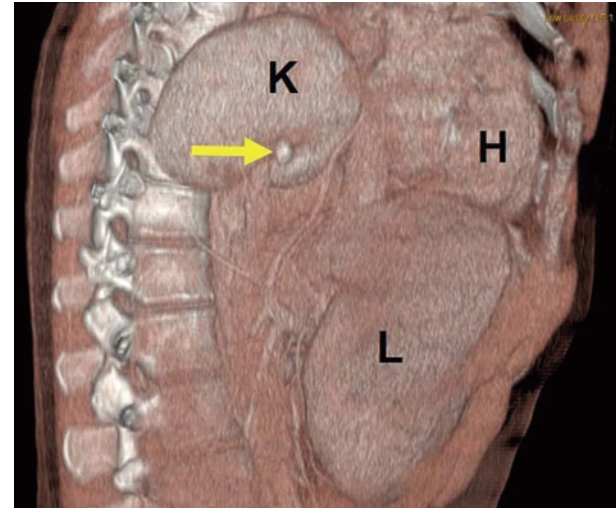


Figure 4. Sagittal CT scan shows appendicolith in the appendicular lumen in the retroperitoneal space on the Th9/Th10 level (yellow arrow), below the kidney and above the liver. K: Kidney; L: Liver; H: Heart.

3. Discussion

Controversy exists regarding the optimal diagnostic strategy for acute appendicitis: clinical history and physical examination alone *vs.* selective imaging in patients with clinical suspicion *vs.* routine imaging of all patients[1,2].

A typical appendicitis scenario consists of periumbilical, colicky pain with vomiting that migrates to the right lower quadrant. Clinical signs or abnormal blood results can be absent in over half of cases. Clinical diagnosis alone may be associated with up to 40% of misdiagnoses and 34% of negative appendectomies[3-5].

In a meta-analysis by Anderson clinical signs and laboratory test results were weak predictors of appendicitis on their own, but they achieved a high predictive value when combined. Clinical signs of peritoneal irritation, history of pain moving into the right lower quadrant and abnormal results of WBC and CRP are most important in making a correct diagnosis[6].

Alvarado constructed a 10-point clinical scoring system based on symptoms, signs, and diagnostic tests to estimate the possibility of appendicitis in patients presenting with suspicious abdominal pain. The system enables risk stratification linking the probability of appendicitis to recommendations regarding discharge, observation, or surgical intervention[7,8].

A meta-analysis by Kabir *et al.* confirmed that the Alvarado scoring system is the most accurately predicts appendicitis in men and can be used as a reasonable starting point in the assessment of suspected cases of appendicitis. However, authors stated that Alvarado score cannot reliably predict appendicitis without further investigations and therefore should not be used alone in further management planning[5].

In our case, the Alvarado score was 9 and the clinical diagnosis of appendicitis was not in doubt. However, a question arises regarding the use of CT in patients with suspected acute appendicitis when clinical picture is convincing. Subsequent clinical management strategy would therefore be surgery.

A periumbilical pain represents referred pain due to visceral innervation of the midgut, and the localized pain is caused by involvement of the parietal peritoneum due to progression of the inflammatory process through appendiceal wall[9].

In our case, periumbilical pain migrated to the right lower quadrant of the abdomen even though the appendix was localized in the right upper quadrant. Intervention based on clinical diagnosis alone would have resulted in traditional incision in the right lower quadrant. In our case such incision even if extended upwards wouldn't provide needed exposure. Results of CT directed us to use right subcostal incision.

Choice of imaging modality remains subject of debate: ultrasound (US) *vs.* CT *vs.* magnetic resonance (MR) alone or in conjunction: US-CT, US-MR.

On one hand, CT is hailed as the gold standard in diagnosing appendicitis with sensitivity and specificity reported between 83% and 98%. It has been shown to decrease negative appendectomy rates to less than 10% (compared to 21.5% in the pre-CT era). On

the other hand, literature reported US to be the most commonly used imaging method to confirm the diagnosis of appendicitis with a sensitivity of 71%-86% and specificity of 81%-97%. Both these modalities have their limitations: US is operator-dependent, and CT produces radiation exposure, contrast related complications and has a high cost. Some authors have recommended use of CT in conjunction with US. If clear signs of appendicitis are present in US, then surgery is performed without need for CT[5,9].

In our case, CT proved to be valuable in avoiding complications and significantly contributed to a good outcome.

Another question relating to indications for an appendectomy is a previous operation for congenital diaphragmatic hernia. Incidental appendectomy is a removal of the normal appendix during an unrelated operation, without evidence of acute appendicitis. It is generally performed to eliminate risk of future appendicitis. Risks and benefits of incidental appendectomy during various operations in children have been debated for over a century. Healy *et al.* reviewed clinical situations that may influence a surgeon's decision to perform an incidental appendectomy. Congenital diaphragmatic hernia is most frequently associated with intestinal non-rotation or malrotation and results in abnormal positioning of the appendix, which can lead to future diagnostic dilemmas[10]. Our case illustrates that incidental appendectomy during the previous operation would have been beneficial.

4. Conclusions

- (1) One should always think of an unusual presentation of a common disease first, rather than search for a rare diagnosis;
- (2) Migration of abdominal pain to the right lower quadrant should always include appendicitis in the differential diagnosis;
- (3) Congenital diaphragmatic hernia repair may change the location of the appendix.
- (4) The rare location of the appendix could lead to an uncommon course of appendicitis. Given this, surgery should have high index of suspicion, and CT may help in avoiding complications.

Conflict of interest statement

The authors report no conflict of interest.

Authors' contributions

S.W.: Clinical management, data acquisition and analysis, manuscript preparation and review; M.G.: Literature search, data analysis, manuscript preparation, editing and review; R.A.: Data acquisition and analysis, manuscript review; M.S.: Clinical management, data analysis, manuscript editing and review.

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