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Cas clinique

Appendiceal Endosalpingiosis : A rare cause of atypical acute appendicitis in females.

Endosalpingiose appendiculaire : une cause rare d'appendicite aiguë atypique chez la femme.

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Résumé

Contexte : De nombreuses patientes se présentent avec une suspicion clinique d'appendicite aiguë sans critères cliniques ou biologiques suffisamment évocateurs. Parmi les causes sous-jacentes rares figure l'endosalpingiose appendiculaire (EA).

Observation clinique : Nous rapportons le cas d'une femme de 26 ans présentant des signes atypiques d'appendicite, notamment une douleur de la fosse iliaque droite avec une sensibilité localisée, en l'absence de fièvre ou d'anomalies biologiques. La tomodensitométrie (TDM) a mis en évidence un appendice augmenté de volume, à paroi épaissie, sans inflammation péri-appendiculaire significative. Une appendicectomie par voie laparoscopique a été réalisée ; les constatations peropératoires ont montré un appendice simplement congestif. L'examen histopathologique a permis d'établir le diagnostic d'EA, caractérisée par la présence d'un épithélium cilié de type tubaire à la surface séreuse, sans stroma endométrial associé ni réaction inflammatoire.

Conclusion : Ce cas souligne l'EA comme un diagnostic différentiel inhabituel des formes atypiques d'appendicite aiguë et met en évidence la nécessité de considérer sa possible signification clinique, notamment au regard de l'association suggérée avec certaines malignités gynécologiques. Le diagnostic de certitude repose sur l'examen histopathologique, et la publication de cas supplémentaires est nécessaire afin de mieux préciser sa signification clinique et sa prévalence.

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Mots clés : Appendice, endosalpingiose, appendicite atypique, laparoscopie, appendicectomie, femme.

Abstract

Background: Many patients present with clinical suspicion of acute appendicitis without sufficient supporting clinical or laboratory criteria. One rare underlying cause is appendiceal endosalpingiosis (AE).

Case presentation: We report a 26-year-old woman who presented with atypical features of appendicitis, including right lower quadrant abdominal pain and localized tenderness without fever or laboratory criteria. Computed tomography (CT) scan revealed a thick-walled, enlarged appendix without significant surrounding inflammation. She underwent a laparoscopic appendectomy, and intraoperative findings confirmed just congested appendix. Histopathologic examination established the diagnosis of AE, characterized by ciliated tubal-type epithelium on the serosal surface without associated endometrial stroma or inflammation.

Conclusion: This case highlights AE as an unusual differential diagnosis for atypical appendicitis and underscores the need to consider its potential clinical significance, particularly in view of the proposed association with gynecologic malignancies. Definitive diagnosis relies on histopathological examination, and further reporting is needed to clarify its clinical significance and prevalence.

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Keywords: Appendix, endosalpingiosis, appendicitis, atypical, laparoscopic, appendectomy, female.

Introduction

Emergency laparoscopic appendectomy (LA) is practised based mainly on clinical findings in addition to supportive laboratory and/or imaging modalities. In some cases, typical criteria of acute appendicitis are not fulfilled, but the surgeon has to do LA on suspicion for fear of complications waiting for the histopathologic examination of the appendix. In one study, negative histopathologic inflammatory findings accounted for 14.4% out of 940 participants who underwent appendectomy procedures [1]. Here, we present a rare case of atypical appendicitis proved to be appendiceal endosalpingiosis (AE) in a young woman. Its clinical presentation, imaging, intraoperative findings, and histopathological confirmation are described. **The objective is to raise awareness among the medical community of appendicular endosalpingiosis as a rare cause of atypical appendicitis.**

Case Report

A 26-year-old woman, Para 1+0, Last delivery before one and a half years and stopped lactation before a couple of months is described. **She was midcyclic and did not use contraception.** She presented to the Emergency Department (ER) with a one-day history of right lower quadrant abdominal pain, associated with fatigue, burning micturition, urinary hesitancy, and amenorrhea of two months. Her pain radiated to the lower back and anal region. On examination, she was afebrile with stable vital signs. Abdominal examination revealed generalized lower abdominal tenderness, most marked in the right iliac

fossa. Laboratory investigations showed normal complete blood picture (CBC) and C-reactive protein (CRP) level. The white cell count was $3.01 \times 10^9/L$, neutrophils $2.5 \times 10^9/L$, and C-reactive protein (CRP) level was 1.1 mg/L. Renal function and coagulation profiles were unremarkable. Routine pregnancy test was negative. Ultrasonography of the abdomen and pelvis was unremarkable. Due to the disproportionate severity of pain despite being afebrile, having normal laboratory results, and an unremarkable ultrasound, a CT scan was performed. A contrast-enhanced CT scan revealed a 09 mm appendicular thickness with thickened and enhancing walls, submucosal edema, and no surrounding fat stranding or signs of perforation (Figure 01). Mild pelvic free fluid was observed but no significant intraperitoneal gases were seen. The patient was counselled to be subjected to laparoscopic intervention and signed a consent for it. After obtaining Institutional Review Board approval (IRB: PAT0223840) and insurance authorization, the patient was evaluated by the anesthesia team and scheduled for an emergency laparoscopy. Under general anesthesia, standard triple laparoscopic approach was used. The appendix was retrocecal and thickened with a fecalith (figure 02-A). Additionally, a small ruptured right ovarian corpus luteum cyst was noted, for which the gynecologist (AD) recommended no intervention.

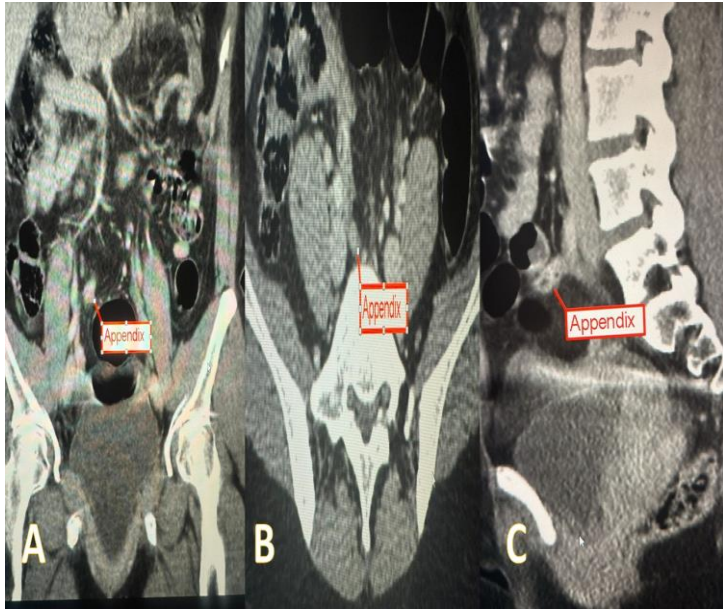


Figure 01: A,B,C: Axial, coronal and Sagittal CT scans with IV contrast showing thickened and enhanced appendicular walls with submucosal edema yet with lack of surrounding fat strands (In THE sagittal view the appendix appear as a tubular structure with lobulation mimicking a tumor).

[Axial (a) and coronal (b) contrast-enhanced CT images of the abdomen and pelvis, showing an enlarged, thick-walled appendix consistent with acute appendicitis (arrows).]

Laparoscopic appendectomy procedure was done smoothly (figure 02 B). Due to the diagnosis of a ruptured corpus luteum cyst, a pelvic suction drain was inserted. Upon completion, peritoneal lavage was performed, and an intraperitoneal drain was placed. The drain was removed the following morning, and the patient was discharged after an uneventful postoperative course, receiving a short course of broad-spectrum antibiotics. Histopathological examination of the appendix grossly revealed an appendix measuring 8x0.7 cm in length and width respectively and showed surface congestion

(evident in figure 02 A as well). It was surrounded by fat and showing surface congestion. Microscopically, it showed cystically dilated glands lined by ciliated tubal epithelium on the serosal surface, with no evidence of endometrial stroma or inflammation (Figure 02 C-D). There was no epithelial dysplasia or neoplasia. The peri-appendiceal fat demonstrated congestion and chronic inflammatory infiltrates.

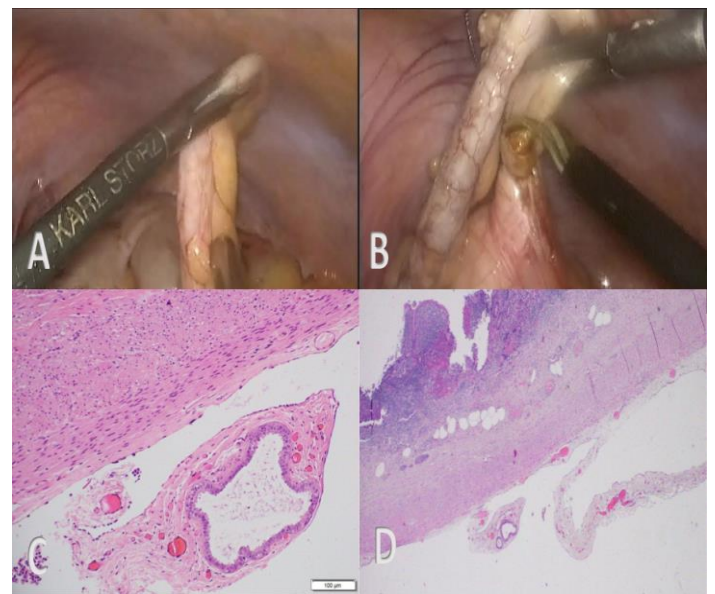


Figure 02. A. Laparoscopic appearance of the appendix. B. Laparoscopic appendectomy. C and D. Histological sections of the appendiceal wall showing cystically dilated glands lined by ciliated tubal epithelium on the serosal surface. No endometrial stroma identified. (H&E staining).

[A: Laparoscopic appearance of the appendix with congested surface. B. Completely excised appendix. C,D: Appendiceal wall was showing mild cystically dilated glands in the serosal surface line by ciliated tubal epithelium, no endometrial stoma was identified (H & E, x10).]

Discussion

This case illustrates an uncommon clinical scenario of right lower quadrant abdominal pain mimicking acute appendicitis. Initial investigations, including CBC, CRP, and ultrasonography, were not suggestive of acute appendicitis. A potentially misleading finding was the presence of free fluid in the pouch of Douglas, along with a small right ovarian cyst, which raised suspicion of a ruptured corpus luteum cyst. However, the laparoscopic appearance of minimal volume of fluid and the small size of the ruptured cyst made a gynecologic cause for the acute abdomen unlikely. Moreover, both pelvic ultrasonography and CT failed to identify the ruptured corpus luteum cyst, which was diagnosed only at diagnostic laparoscopy. This can be explained by the small size of the corpus luteum cyst, the minimal amount of free fluid in the pouch of Douglas, and, more importantly, the CT findings of a thickened appendix that diverted diagnostic attention.

This highlights the definitive role of laparoscopy and its superiority in detecting subtle lesions in equivocal clinical settings. A decision was made to proceed with LA to establish a definitive diagnosis and excise the thickened appendix observed on CT despite the incomplete picture of appendicitis. Based on a review of the literature, AE is an exceptionally rare histopathologic finding.

The first documented case was reported in 1990 [2]. In a large database review of over 58,000 LA from 1998 to 2013, only 20 cases of AE were histopathologically diagnosed [3]. Generally, endosalpingiosis is a rare histopathological condition characterized by benign ectopic tubal-type

ciliated glandular epithelium that resembles normal endosalpinx tissue. Histologically, it differs from endometriosis in that it contains ciliated epithelium without endometrial stroma and lacks an associated inflammatory response characteristic of endometriosis [4]. It has been associated with gynecologic malignancies, particularly serous borderline tumors and, less commonly, cystadenocarcinomas [5].

The exact etiology of endosalpingiosis including AE remains unclear. The most widely accepted theory is coelomic metaplasia, wherein mesothelial cells undergo transformation into Müllerian-type epithelium. An alternative theory suggests endometrial cells or their precursors are transported via various routes (transtubal, hematogenous, lymphatic, or direct implantation) and become established in ectopic locations [6]. Unfortunately, it is a histopathologic diagnosis without any blood screening test. Clinical presentation varies, often mimicking other conditions. Endosalpingiosis may present in the reproductive age or adolescent females. However, it has been reported in a postmenopausal woman with appendiceal thickening (11 mm on CT) suggestive of appendicitis, but without inflammatory signs [7] as seen in this case. The diagnosis of appendicular endosalpingiosis should not be lightly considered as there are some case reports of perforated appendicitis with concurrent endosalpingiosis [8].

Moreover, long-term gynaecological follow-up for the patient, given the potential association of endosalpingiosis with malignancies is recommended. It reached

40% of cases in a retrospective case-control study of 967 patients [9].

Being a single case with poor literature on endosalpingiosis is a clear limitation of this publication.

Conclusion

Despite being very rare without laboratory screening test, AE should be put as a differential diagnosis of any case of acute right lower abdominal pain without typical criteria of appendicitis. CT scan is highly valuable in cases with uncertain diagnosis. Implementation and collaboration with gynaecologists would save genital organs from unneeded surgical interventions.

More publications on AE would help estimate its real prevalence and would help its inclusion in the management plan of cases of acute right-sided lower abdominal pain.

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Conflict of Interest

All authors declare no conflicts of interest.

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