

## Perforated endometrial appendicitis in pregnancy

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### Abstract

**Background:** Endometriosis is a common disorder in women of reproductive age. A rare localization is the appendix, which, in most cases, is an incidental finding during appendectomies. The incidence of symptomatic appendiceal endometriosis or endometrial appendicitis might be increased in pregnancy. Moreover, endometrial appendicitis in pregnancy is more likely to present in an advanced stage, given the physiologic changes characterizing the gravid abdomen.

**Materials and Methods:** Description of a case of a pregnant woman presenting to the A&E with acute peritonitis attributable to advanced appendicitis. She underwent emergency laparotomy with appendectomy. The biopsy specimen was examined by the pathology laboratory of the same hospital.

**Results:** The laparotomy revealed perforated appendicitis. The histopathology report described acute endometrial appendicitis. Hippokratia 2012; 16 (2): 181-183

**Key words:** gestational appendicitis, pregnancy appendicitis, endometrial appendicitis, appendiceal endometriosis

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Endometriosis of the gastrointestinal tract has been reported with most common primary location the rectosigmoid. Gastrointestinal endometriosis is quite uncommon and it may cause lower abdominal symptoms such as periodic abdominal pain, obstructive symptoms, tenesmus or constipation in women of reproductive age and more often during pregnancy. The condition should be in the differential diagnosis in female patients with cyclic lower abdominal complaints, especially if they are accompanied with symptoms consistent with abdominal inflammation or obstruction. The treatment is surgical removal of the affected part of the bowel<sup>1</sup>.

While endometriosis is a well-established cause of infertility and pelvic ailments in females of reproductive age, localization of endometrial tissue in the vermiform appendix is scarce. On the other hand, acute appendicitis is a common cause of acute abdomen in pregnancy as it is in most population groups; given the special characteristics of the gravid abdomen, it can commonly present as perforated appendix. Yet, the combination of gangrenous perforated appendicitis in the grounds of appendiceal endometriosis is a very rare entity with a small number of cases described worldwide. One such case is being described below.

### Case Report

Female patient of Finnish origin aged 35, in the 27<sup>th</sup> week of pregnancy, presented to the Emergency Department with history of right lower quadrant pain, anorexia

and fever. Her symptoms had 24 hours duration. The patient denied nausea, vomiting, changes in her bowel habits or vaginal discharge. She had no past obstetric or any history of pelvic inflammatory disease, lower pelvic pain or dyspareunia.

On admission the patient was lying in bed with her legs slightly flexed, surrounding her lower abdomen with her hands. She gave the clinical impression of acute abdomen. Her vitals were compatible with her gravid state, with the exception of a temperature of 38<sup>o</sup> C. The physical exam of the chest was unremarkable. On abdominal palpation there was significant tenderness on the right lower abdominal quadrant. Notably, the point of maximal tenderness was well above the right iliac fossa, located on the right mid-abdomen. She had signs of localized peritonitis, i.e. involuntary spasm of the abdominal wall and rebound tenderness in the right mid-abdomen. The rectal exam was unremarkable. The obstetric evaluation showed satisfactory fetal status and ruled out an obstetric or gynaecological aetiology of the patient's symptoms. The blood results showed leukocytosis with increased neutrophil count.

Based on the physical findings the patient was diagnosed with acute peritonitis secondary to acute appendicitis and was transferred to the operating room for exploratory laparotomy. The surgical approach was through a right paramedian lower abdominal incision. On laparotomy the patient was found to have a friable, thickened, gangrenous vermiform appendix that was perforated at

its base surrounded by plastron formation which had engaged it on the right side of the gravid uterus. The adhesions to the omentum, the uterus and the small bowel loops were easily released, and a formal appendectomy was performed, followed by extensive abdominal cavity washout. No other signs of endometriosis were found during the abdominal exploration. The abdominal wall was closed. The patient tolerated the procedure well. Due to gestation, she was admitted to the Obstetrics and Gynaecology ward for post-operative observation. The fetal status remained satisfactory and the patient's postoperative course was uneventful. The patient was discharged on the third post-operative day.

The histopathology of the vermiform appendix showed suppurative inflammation with loci of endometriosis in the mucosa of the appendiceal lumen.

### Discussion

While endometriosis is a common disorder in women of reproductive age, appendiceal endometriosis accounts only for a small fraction of pelvic endometriotic lesions<sup>2</sup>. The localization of endometriosis in the vermiform appendix is extremely uncommon. The cases reported in the literature are scarce<sup>2-6</sup>. In 1999, Ortiz-Hidalgo C et al<sup>7</sup> reported endometriosis of the vermiform appendix frequency as low as 1% of all cases of pelvic endometriosis. A more recent case series published by Gustofson RL et al provides an estimate of the prevalence of appendiceal localization of the disease in patients with chronic pelvic pain (n:133, 13 of whom with previous appendectomy and 109 with history of right lower quadrant pain) and possible endometriosis who underwent laparoscopy<sup>4</sup>. According to this study, the prevalence of appendiceal endometriosis in patients with biopsy-proven endometriosis (n: 97) or with right lower quadrant pain (n:109) was 4.1% and 3.7% respectively. Notably, this was higher than the 2.8 % prevalence confirmed by literature review this research team had performed and much higher than its prevalence in all patient population (0.4%).

If appendiceal localization of ectopic endometrial tissue is a rare finding, the appendiceal inflammation with appendiceal endometriosis is even more uncommon, with only a handful of cases recorded in literature. And even less are known to result in gangrenous appendicitis, such as the case presented above<sup>2</sup>. Notably, the patient described in the present review has been the second recorded case of gangrenous appendicitis in the presence of vermiform appendix endometriosis in a patient who is pregnant to the authors' best knowledge. The other case was described by Perez et al in 2007<sup>8</sup>.

Given the limited volume of available data, it is difficult to establish a correlation between the presence of endometriosis loci in the vermiform appendix and the pathogenesis of appendicitis. Using as a model the pathophysiological mechanisms involved in the context of appendicitis in a patient with a carcinoid tumor or appendiceal adenocarcinoma or even in the patients with appendicitis and appendiceal lymphoid hyperplasia or ap-

pendicolithiasis, we may assume that the pathogenesis of appendicitis in the grounds of appendiceal endometriosis shares the same common final pathway, i.e. appendiceal lumen obliteration, increase of appendiceal luminal pressure inducing mucosal ischaemic changes which herald the progression to appendicitis.

Appendiceal endometriosis is usually asymptomatic or causes vague lower abdominal symptoms. It is quite likely that patients with endometriosis of the appendix are misdiagnosed for appendicitis during pregnancy in a mechanism similar to the catamenial appendicitis phenomenon, as described by Barrier et al. in 2008<sup>9</sup>. Considering the close anatomical relation of the female reproductive system with the appendix and the marked overlapping of the clinical presentation of their pathological conditions, a low threshold for gynaecological input should be sought early in the management of such patients, even in the operating room, as it might had been prudent in the case discussed.

In most cases appendiceal endometriosis is an incidental finding during histopathology examination of the vermiform appendix after appendectomy performed for assumed acute appendicitis. The appendectomy performed is sufficient for the treatment of the condition and there is no need for re-operation.

Endometriosis is a common disorder in women of reproductive age, which may cause lower gastrointestinal symptoms if it involves the lower gastrointestinal tract. A rare localization of endometrial tissue is in the vermiform appendix, with an incidence among patients with known endometriosis ranging between 1- 4.1 %. In most cases, it is an incidental finding during appendectomies for assumed or actual appendicitis. The incidence of symptomatic appendiceal endometriosis or complicated appendiceal endometriosis, i.e. appendicitis, might be increased in hyper-progesteronic states, as it is pregnancy. It may present itself in a delayed state, which is quite common in gravidae operated for appendicitis.

### Summary

It may be concluded that there is positive correlation between appendiceal endometriosis and appendicitis, which may cause an increased incidence of acute appendicitis during pregnancy with appendiceal endometriosis as the triggering factor. Appendiceal endometriosis during gestation may result in increased incidence of appendectomies for wrongly diagnosed appendicitis. Given the limited amount of documented cases there is not sufficient evidence to support the assumption that endometrial appendicitis is aggravated during pregnancy, even though it cannot be excluded that such correlation does exist.

### Conflict of Interest:

The corresponding author verifies on behalf of all authors that there is no conflict of interest

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