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Case Report

Appendiceal endometriosis as a cause of ileus in pregnancy

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Summary

Endometriosis is the presence of ectopic endometrial tissue outside the uterine cavity. It is a well-known cause of chronic pelvic pain and infertility in women. Endometriosis of the appendix is rare and may present with acute or chronic abdominal pain. Preoperative diagnosis is difficult. The definitive diagnosis is usually established following histopathological examination of the appendix. The authors report a case of ileus at the ground of adhesions formed due to appendiceal endometriosis clinically presented with recurrent attacks of abdominal pain in the eight month of pregnancy after laparoscopic removal of the ovarian endometriosis.

Key words: Pregnancy; Acute abdomen; Appendix; Endometriosis; Adhesions.

Introduction

Endometriosis, defined as the presence of endometrial glands and stroma outside the uterine cavity, is estimated to affect 4–50% of reproductively-aged women and results in pelvic pain and infertility in up to 50% of these patients. Endometriosis is most commonly found in the gynecologic organs and pelvic peritoneum but may also involve the gastrointestinal system, greater omentum, surgical laparotomy scars, and the mesentery, but is rarely found at distant sites such as the kidney, lungs, skin, and nasal cavity. Gastrointestinal endometriosis is observed in 3–37% of all endometriosis cases, but appendiceal endometriosis accounts for only about 3% of gastrointestinal endometriosis and < 1% of total endometriosis cases. Appendiceal endometriosis may not only cause symptoms of acute and chronic appendicitis but also cyclic and chronic right lower quadrant pain, melena, lower intestinal hemorrhage, coecal intussusception, and intestinal perforation. Definitive diagnosis can be established only by postoperative histological examination.[1]

Case Report

A 29-year-old was admitted in this hospital on February 17, 2015 in her 28th week of pregnancy, due to upper abdominal pain, after surgical and gastroenterological examination in the Emergency room of Clinical Centre of Serbia. Her last menstrual period dated back to August 3, 2014. She had no previous deliveries or abortions. On admission the fetus presented an episode of bradycardia.

Her family history was uneventful. In her personal history the patient had laparoscopic surgery due to endometriotic cyst of the left ovary, accidental lesion of the right ureter, followed by laparotomy for urological treatment. During this procedure the appendix was not recognizable, so the diagnosis autodigestio appendicis endometriotica was established. She was treated with ranitidine, metochlopramide, pantoprazole infusions of fluid. On February 18, 2015, an ultrasound examination showed normal course of pregnancy. Due to abdominal pain the patient was sent to Emergency room for consultation with a surgeon and gastroenterologist. Radiological examination showed distended bowel, up to 45 mm, filled with liquid content, and aperistaltic. In Morison pouch and subhepatic space there was a smaller amount of free fluid. Per os alimentation was suspended and nasogastric suction was inserted and amp. nexium in bolus was administered. The patient returned to this Clinic. Laboratory analyses showed WBC 16.96×10^9/L, CRP 18.5, and D-dimer 3.91.

On February 19, 2015, surgical consultation in the Emergency room was requested again with diagnosis of suboclusion. The patient was admitted to the Emergency room, and after conservative treatment was sent home on February 21, 2015. On the February 27, 2015. gastroscopy was performed because of vomiting, nausea, and diarrhea, the diagnosis was gastritis. In the evening of the same day. she was again admitted in the hospital with severe abdominal pain. Gynecologial and ultrasound examination showed normal course of pregnancy. A day later, the patient experienced nausea vomiting, and abdominal pain, and was administered antibiotics with ranitidine, metochlopramide, pantoprazole, fraxiparin, and i.v. fluid replacement. On March 1, 2015, the authors prescribed artificial fetal lung maturation with dexamethasone. In the next day spontaneous rupture of the fetal membranes occurred, with significant increase of the inflammation parameters, CRP 96, and WBC 9.38×10^9/L, so the authors suspected developing chorioamninitis. The obstetric finding was not suitable for vaginal delivery. Due to consecutive development of fetal asphyxiation, they decided to perform emergency cesarean section due to vital indications for the mother and the baby. At

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12.20 a boy 41 cm long, weighing 1,730 grams, with Apgar score of 3/5 was delivered. Intraoperative finding showed that the size of the uterus corresponded to the gestational age. Broad, cobweb-like adhesions of small intestine with posterior uterine wall were found, also with thick and dense interintestinal adhesions involving ileocoecal region and convolute of the small intestine leading to mechanical ileus. At the ileocoecal region, tumor presenting as periappendicular infiltration was noticed. Abdominal surgeon was called for consult. Periappendicular biopsy was performed and ex tempore analysis was demanded. After receiving the result (inflamed decidua) it was decided to remove the appendix and periappendicular tissue in toto and send it to pathohistological examination, which revealed endometriosis of the appendix. The abdominal wall was closed. Postoperative course was normal, the patient was treated with antibiotics and anticoagulant therapy, and peristalsis was restored.

Discussion

Endometriosis is defined as the presence of endometrial tissue outside the uterus, which induces a chronic inflammatory reaction, scaring, and formation of adhesions that may distort a woman's pelvic anatomy [2]. It is usually associated with dysmenorrhea, chronic pelvic pain, and infertility. The causes of infertility remain uncertain, but laparoscopic ablation of endometriosis in combination with medical treatment improves fertility [3]. Most peritoneal implants appear as subtle, non-pigmented lesions.

Appendiceal localization of endometriosis is rare, with a prevalence of 2.8% in patients with endometriosis. In patients with right lower quadrant or pelvic pain, the appendix should be inspected for endometriosis and evidence of non-gynecologic disease. [4] About half of appendiceal endometriosis involves the body and half involves the tip of the appendix. In the present patient the entire appendix was involved. Current treatment options include medical, surgical, or a combination of these approaches.

The surgical removal of endometriotic implants in minimal-mild severity endometriosis was shown to improve fertility and all visible implants should be excised. In more severe stages of endometriosis, a surgical approach that normalizes pelvic anatomic distortion and provides adhesiolysis can enhance fertility [5]. A definitive diagnosis of endometriosis is often made by laparoscopy or laparotomy with a biopsy and is particularly useful in patients with intestinal implants. Patients with appendiceal endometriosis may experience chronic pelvic pain with significant decrease in the quality of life. Long term symptoms' resolution was noticed following appendectomy [6]. Involvement of the appendix may present as appendicitis, mucocoele of appendix, or appendicular mass that may mimic a neoplasm. Perforation of the appendix may occur especially during the first two trimesters of pregnancy. The present patient experienced problems in pregnancy at the end of the second trimester. Women with endometriosis describe the pain associated with adhesions as "stabbing, sharp, pulling, sickening, intense and nauseating", whereas adjectives most commonly used to describe active endometriosis pain itself are "burning, pinching, dull, heavy, and miserable". [7]

The type of pain in the present patient suggested acute abdomen. Chronic irritation of the pelvic tissues from endometriosis may initiate occurrence of the adhesions.

Local inflammation is a key factor in adhesion formation. Adhesions vary in appearances from thin, flimsy, and transparent to thick, dense, and opaque, sometimes to such an extent forming a "frozen" or "fixed" pelvis. [8] Adhesions' formation is a common postoperative complication in women with endometriosis, especially in those with repeated surgeries [9]. The intestinal symptoms are related to a certain degree of bowel obstruction. When the obstruction is severe, the patient will present with nausea, distention, and vomiting. Radiological confirmation of the obstruction is necessary so treatment may be performed, but during pregnancy, surgeons and radiologists are not willing to request radiography. Because bowel may be intimately involved with the adhesive process, the patient has to be aware that the treatment may require bowel surgery through conventional laparotomy incision. Adhesions that led to the ileus in the present patient were thick and dense involving ileocoecal region with unrecognizable appendix. Appendectomy may be considered as part of the surgical treatment. Incidental appendectomy during surgical treatment of pelvic endometriosis is controversial. The decision for an elective appendectomy should be dictated by a meticulous inspection including the length, diameter, color, and relationship of the appendix with the surrounding tissues. Before any surgical treatment for ovarian endometriosis, the surgeon should preoperatively inform the patient regarding the fact that appendiceal pathology including endometriosis is found frequently, regardless of concurrent symptoms or gross finding of the appendix. Furthermore, the surgeon should consider the possibility of appendiceal pathology during operation. [10] During the present patient's first surgery due to ovarian endometriosis, the altered appearance of the appendix was noted, but it was considered to be condition known as autodigestio appendicis endometriotica, without histological confirmation. Nowadays, surgery during pregnancy is considered to be safe and effective. Laparoscopy has become an acceptable alternative to standard laparotomy and should be considered when surgeons with appropriate skills and experience are available. Laparoscopic approach in the third trimester of pregnancy is difficult, due to the size of the uterus. Important to be mentioned is that the surgeons are often refrained regarding operating on pregnant woman. Open surgery is sometimes necessary and even life-saving procedure, like in cases of appendicitis. Physiological changes during pregnancy often make the clinical presentation indistinct. In most cases a multidisciplinary approach is essential to optimize outcomes for both the mother and the fetus [11].

Altered clinical presentation of the mechanical ileus in

pregnancy in the present patient caused the delay in surgical treatment. Eventually, worsening of patients condition would make the surgery necessary. The fact that suspected chorioamnionitis and development of fetal asphyxiation prompted the authors to perform the cesarean section which also enabled them to perform exploration of the abdominal cavity and diagnose the cause of acute abdomen.

Conclusion

Endometriosis of the appendix can mimic appendicitis and sometimes present as acute abdomen. It should always be considered in the differential diagnosis in young women with non-specific recurrent lower abdominal pain. Diagnosis is particularly difficult to establish in pregnancy because physiological changes in pregnancy often make the clinical presentation indistinct. The history of infertility and previous ovarian surgeries due to endometriosis are the most important factors of adhesion formation as a result of chronic inflammatory reaction.

References

[1] Al Oulaqi N.S., Hefny A.F, Joshi S, Salim K., Abu-Zidan F.M.: "Endometriosis of the appendix". *Afr. Health Sci*., 2008, *8*, 196.
[2] Kennedy S, Bergqvist A, Chapron C, D'Hooghe T, Dunselman G, Saridogan E, *et al.*: "ESHRE guideline on the diagnosis and management of endometriosis". *Hum Reprod.,* 2005, *20*, 2698.
[3] Ozkan S., Murk W., Arici A.: "Endometriosis and infertility: epidemiology and evidence-based treatments". *Ann. N. Y. Acad. Sci*., 2008, *1127*, 92.
[4] Gustofson RL, Kim N, Liu S, Stratton P.: "Endometriosis and the appendix: a case series and comprehensive review of the literature". *Fertil Steril*., 2006, *86*, 298.
[5] Adamson G.D., Baker V.L.: "Subfertility: causes, treatment and outcome". *Best Pract. Res. Clin. Obstet. Gynaecol*., 2003, *17*, 169.
[6] Barrier B.F., Frazier S.R., Brennaman L.M., Taylor J.C., Ramshaw B.J.: "Catamenial appendicitis". *Obstet. Gynecol*., 2008, *111*, 558.
[7] Hummelshoj L.: "Endometriosis: a special problem with adhesions". *Adhesions News and Views*, 2002, *1*, 25.
[8] Wie H.J., Lee J.H., Kyung M.S., Jung U.S., Choi J.S.: "Is incidental appendectomy necessary in women with ovarian endometrioma?" *Aust. N. J. Z. Obstet. Gynaecol*., 2008, *48*, 107.
[9] Bulletti C., Coccia M.E., Battistoni S., Borini A.: "Endometriosis and infertility". *J. Assist. Reprod. Genet*., 2010, *27*, 441.
[10] Duffy D.M., diZerega G.S.: "Adhesion controversies: pelvic pain as a cause of adhesions, crystalloids in preventing them". *J. Reprod. Med*.,1996, *41*, 19.
[11] Kizer N.T., Powell M.A.: "Surgery in the pregnant patient". *Clin. Obstet. Gynecol*., 2011, *54*, 633.

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