

CLINICAL CASE

UNCOMMON PRESENTATION OF RECURRENT INFLAMMATORY PELVIC DISEASE: MUCOCYSTIC DEGENERATION OF THE FALLOPIAN TUBE**S. Păun^{1,2}, Camelia Alexandroaia², Romina-Marina Sima^{2,3}, Irina Iurieț³, Liana Pleș^{2,3}**¹The Emergency Hospital of Bucharest, Bucharest, Romania²The University of Medicine and Pharmacy „Carol Davila”, Bucharest, Romania³„St. John” Hospital, „Bucur” Maternity, Bucharest, Romania

Corresponding author: Romina-Marina Sima

Phone no. 0040741071243

E-mail: romina.sima@yahoo.es

Abstract

Pelvic inflammatory disease (PID) is an acute and sometimes subclinical infection of the upper genital tract in women, involving any of the uterus, fallopian tubes and ovaries. The condition commonly can involve also the other pelvic organs mainly the peritoneum. It results in oophoritis, salpingitis, tubo-ovarian abscess, peritonitis or perihepatitis. A 26-year-old woman was admitted in our clinic for lower abdominal pain, fever and vaginal discharge. The ultrasound aspect was suggestive for PID with tubal involvement (salpingitis). The lab finding proved an important inflammatory syndrome (high C protein level, elevated white blood cells count). We decided to perform exploratory laparoscopy for PID. Intraoperative findings consisted of: both fallopian tubes were enlarged, with congestive vascularization and adhesion process to the surrounding tissue resulting in bilateral tubo-ovarian abscess. Due to further patient wish to conceive we decided to perform conservative treatment: lavage and drainage of the abscess. The patient outcome was favorable with the discharge in the 3rd postoperative day. The patient returns in our unit after 6 months following the operation for similar symptomatology. The clinical and ultrasound examination established the diagnosis of recurrent PID bilateral sactosalpinx. We decided to perform laparoscopic surgery, with bilateral salpingectomy. The patient had no gynecological complain one year later. The histopathological examination depicted an unusual degeneration of the fallopian tube of mucocystic aspect. We presented a case with clinical and imaging scan that imposed the diagnosis of Pelvic Inflammatory Disease. The particularity of this case is the pathological exam that proved the mucocystic degeneration of the fallopian tube. It represents an extremely rare condition.

Keywords: *pelvic inflammatory disease, mucocystic degeneration, uterus, fallopian tubes***Introduction**

Pelvic inflammatory disease (PID) is an acute infection of the upper genital tract in women, involving the uterus, fallopian tubes, and ovaries that can evolve in a large spectrum of clinical manifestation ranging from very

noisy to subclinical type. Due to the modality of continuous inflammation, the condition commonly affects all pelvic organs causing oophoritis, salpingitis, tubo-ovarian abscess, peritonitis or perihepatitis. Most of PID cases are produced by sexually transmitted pathogens or in some cases by bacterial vaginosis-

associated pathogens. In a small percentage (almost 15%) of acute PID cases are not sexually transmitted and they are associated with enteric germs (such as *Escherichia coli*, *Campylobacter* spp, *Bacteroides fragilis* and Group B streptococci) or respiratory pathogens (e.g. *Streptococcus pneumoniae*, *Haemophilus influenzae*, Group A streptococci, and *Staphylococcus aureus*) that colonize the lower genital tract[1].

The most important sequels of PID is infertility caused by tubal obstruction and /or adhesions at the fallopian tubes and ovaries. Rarely the condition heals by itself and the tubes evolved in hydric pouches - hydrosalpinx. The histological findings at the mucosa of hydrosalpinx show usually and chronic inflammatory pattern with no ciliated cells.

Case presentation

A 26-year-old woman was admitted in our clinic for lower abdominal pain, fever and vaginal discharge. The symptoms appeared 24 hours earlier.

Her obstetrical history consists in two vaginal deliveries and no abortion. Her last menstrual period was 14 days prior to hospital admission. Her past medical record registered appendectomy and no other medical or surgical events.

The clinical examination revealed an abundant purulent vaginal discharge, the cervix had reddish appearance, the uterus had a normal volume but was sensitive when mobilized. Both adnexal zones were also occupied by tumoral masses and exhibited intense pain in examination.

The ultrasound examination exposed the normal size uterus and right latero-uterine tumor a 2/3 cm, anechoic with thin walls and septae, and left latero-uterine a 4/5 tumor with the same ultrasound appearance. The ultrasound aspect was suggestive for PID with tubal and ovarian involvement (tubo-ovarian abscess). The lab finding proved that the inflammatory syndrome was present. (high protein C level, elevated white blood cells count). The first approach consisted in large spectrum antibiotherapy, anti-inflammatory and pain killer drugs. The initial symptoms reduced but

the inflammatory syndrome persisted following the lab charts.

We decided to perform exploratory laparoscopy for PID. The open entry technique was used (Hasson). We found that both fallopian tubes were enlarged, with congestive vascularization and adhesion process to the surrounding tissue. The diagnosis was bilateral tubo-ovarian abscess. Due to further patient wish to conceive we decided to perform conservative surgical treatment. The fallopian content was evacuated and washed after taking bacterial sample. The fluid was yellow-cream suggestive for bacterial involvement (Figures 1-4). The patient outcome was favorable with the discharge in the 3rd postoperative day. The bacterial culture from the fallopian fluid was negative, maybe because of the preoperative antibiotherapy.



Figure 1 – Laparoscopic aspect



Figure 2 – Laparoscopic aspect

The patient returned in the hospital 6 months following the initial surgery for similar symptomatology. The clinical and ultrasound examination imposed the diagnosis of recurrent PID with bilateral hydrosalpinx. We decided to perform laparoscopic surgery, with bilateral salpingectomy. Her outcome was without any complication. The patient had no gynecological complain one year later.



Figure 3 - Laparoscopic aspect

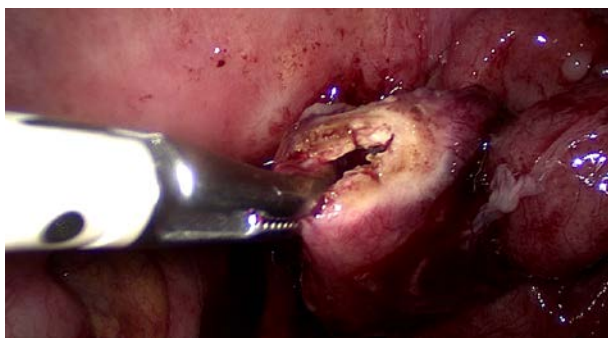


Figure 4 – Laparoscopic aspect

The histopathological examination of the removed fallopian tubes was a surprise depicting an unusual mucocystic degeneration of the fallopian tube wall.

Discussions

PID consists in a spectrum of infection and there is no unique gold standard diagnosis. The most important practical approach remains the clinical diagnosis. Several expert guidelines have tried to establish the clinical standard diagnosis of PID[2,3]. A report estimated that a cohort of 100,000 females that were diagnosed with PID between 20 and 24 years of age may have 18,600 cases of chronic pelvic pain; 16,800 cases of infertility; and 8550 ectopic pregnancies[4].

The differential diagnosis includes cervicitis, appendicitis, endometriosis, urinary tract infection, and adnexal tumors. PID may result in tubo-ovarian abscess and may cause Fitz-Hugh–Curtis syndrome (perihepatitis)[5]. A rare and life-threatening complication is the acute rupture of tubo-ovarian abscess and it may result in peritonitis and necessitate urgent surgical intervention[6,7,8]. It is considered that mediated variation in immune response have an essential role in development of PID[9].

Variants in the genes that control toll-like receptors (TLRs), have been associated with of *C trachomatis* infection in PID[10].

Treatment is applied for patients with this presumptive clinical diagnosis of PID, even if findings are minimal, because long-term complications are more common if treatment is delayed[11]. Den Hartog et al identified a contributing role of 5 single-nucleotide polymorphisms (SNPs) in some genes encoding recognition receptors in local tubal cells and macrophages. The presence of SNPs is correlated with increased tubal pathology identified using laparoscopy[12].

Williamson et al reported a case of microcystic endometrioid carcinoma of the fallopian tube that simulated an adnexal tumour. They considered that it may be a tumor with wolffian origin[13]. In our case the immunohistochemistry was negative for carcinoma.

The new WHO classification describes the most recent theories on the origin, pathogenesis, and prognosis of ovarian cancer stypes. The tubal origin of hereditary and some non-hereditary high-grade serous cancers is mentioned in contrast to the theory of mesothelial origin of tumors. The category of seromucinous tumors are considered a new entity[14].

Conclusions

We presented a case with clinical and imaging scan that imposed the diagnosis of Pelvic Inflammatory Disease. The particularity of this case is the pathological exam that proved the mucocystic degeneration of the fallopian tube. It represents an extremely rare condition.

References

- [1] Brunham RC, Gottlieb SL, Paavonen J. Pelvic inflammatory disease. *N Engl J Med* 2015; 372:2039.
- [2] Ross J, Judlin P, Jensen J, International Union against sexually transmitted infections. 2012 European guideline for the management of pelvic inflammatory disease. *Int J STD AIDS* 2014; 25:1.
- [3] Workowski KA, Bolan GA, Centers for Disease Control and Prevention. Sexually transmitted

- diseases treatment guidelines, 2015. *MMWR Recomm Rep* 2015; 64:1.
- [4]Yeh JM, Hook EW 3rd, Goldie SJ. A refined estimate of the average lifetime cost of pelvic inflammatory disease. *Sex Transm Dis* 2003; 30:369.
- [5]Wiesenfeld HC, Hillier SL, Meyn LA, Amortegui AJ, Sweet RL. Subclinical pelvic inflammatory disease and infertility. *Obstet Gynecol.* 2012 Jul. 120(1):37-43
- [6]Rivlin ME, Hunt JA. Ruptured tuboovarian abscess. Is hysterectomy necessary?. *Obstet Gynecol.* 1977 Nov. 50 (5):518-22.
- [7]Laohaburanakit P, Treevijitsilp P, Tantawichian T, Bunyavejchevin S. Ruptured tuboovarian abscess in late pregnancy. A case report. *J Reprod Med.* 1999 Jun. 44 (6):551-5.
- [8]Powers K, Lazarou G, Greston WM, Mikhail M. Rupture of a tuboovarian abscess into the anterior abdominal wall: a case report. *J Reprod Med.* 2007 Mar. 52 (3):235-7
- [9]Paavonen J. Chlamydia trachomatis infections of the female genital tract: state of the art. *Ann Med.* 2012 Feb. 44(1):18-28.
- [10]Taylor BD, Darville T, Ferrell RE, Kammerer CM, Ness RB, Haggerty CL. Variants in toll-like receptor 1 and 4 genes are associated with Chlamydia trachomatis among women with pelvic inflammatory disease. *J Infect Dis.* 2012 Feb 15. 205(4):603-9.
- [11]Soper DE. Pelvic inflammatory disease. *Obstet Gynecol* 2010; 116:419.
- [12]den Hartog JE, Ouburg S, Land JA, et al. Do host genetic traits in the bacterial sensing system play a role in the development of Chlamydia trachomatis-associated tubal pathology in subfertile women?. *BMC Infect Dis.* Jul 21 2006. 6:122.
- [13]Williamson JM, Armour A. Microcystic endometrioid carcinoma of the fallopian tube simulating an adnexal tumour of probable wolffian origin. *Histopathology.* 1993 Dec;23(6):578-80.
- [14]Meinhold-Heerlein I, Fotopoulou C, Harter P, Kurzeder C, Mustea A, Wimberger P, Hauptmann S, Sehouli J. The new WHO classification of ovarian, fallopian tube, and primary peritoneal cancer and its clinical implications. *Arch Gynecol Obstet.* 2016 Apr;293(4):695-700. doi: 10.1007/s00404-016-4035-8. Epub 2016 Feb 19. Erratum in: *Arch Gynecol Obstet.* 2016 Jun;293(6):1367.