$ sciendo
ПРИЛОЗИ. Одд. за мед. науки, XLIV 3, 2023 МАНУ
CONTRIBUTIONS. Sec. of Med. Sci., XLIV 3, 2023 MASA

10.2478/prilozi-2023-0056 ISSN 1857-9345

PSEUDOCARCINOMATOUS HYPERPLASIA OF TUBAL MUCOSA ASSOCIATED WITH CHRONIC SALPINGITIS – CLINICAL AND PATHOLOGICAL CHALLENGE: A CASE REPORT

Snezhana Bulikj[1], Milica Kalamaras[1], Dzengis Jasar[2], Katerina Kubelka Sabit[2], Gligor Dimitrov[1], Mitko Ivanovski[1], Eva Palchevska[1], Jovana Topencharova[1]

[1] Department of Gynecology and Obstetrics, Acibadem Sistina Hospital, Skopje, RN Macedonia

[2] Histopathology Department, Acibadem Sistina Hospital, Skopje, RN Macedonia

Corresponding author: Snezhana Bulikj, Department of Gynecology and Obstetrics, Acibadem Sistina Hospital, Skopje, Republic of North Macedonia, email: snezana.stojkovska@acibademsistina.mk

ABSTRACT

Pseudocarcinomatous hyperplasia of the tubal mucosa is a rare, reactive response to an underlying inflammatory or neoplastic process. We present a case of pseudocarcinomatous hyperplasia in a 26-year-old woman with clinical symptomatology of pelvic inflammatory disease, and a normal serum Ca 125-level (30 U/ml). The ultrasound finding showed presence of hydrosalpinx characterized with unilateral tubal enlargement in sausage-like shape that arose from the upper lateral margin of the uterus. The young age of the patient, presence of chronic inflammation, epithelial hyperplasia with unremarkable nuclear atypia and mitosis facilitated the right diagnosis. Pseudocarcinomatous hyperplasia can mimic neoplastic processes clinically and pathologically. Differential morphological and clinical features should be considered to ensure accurate diagnosis and proper management.

Keywords: pseudocarcinomatous hyperplasia, chronic salpingitis, fallopian tube, neoplasm

INTRODUCTION

Hydrosalpinx is a common tubal pathology and a form of tubal factor infertility mainly as a result of a chronic inflammatory tubal process. As such, it can be isolated or as a part of complex adnexal pathology like endometriosis, tubal neoplasm, tubal pregnancy, pelvic inflammatory disease (PID), etc. [1].

Pseudocarcinomatous hyperplasia of the fallopian tube as a rare entity that has morphological similarities with tubal neoplasm may lead to misdiagnosis. Thus, this benign condition is a clinical and pathological diagnostic challenge [2].

In the available literature there are less than 20 case reports of tubal epithelial hyperplasia which is often associated with usage of estrogens, some ovarian lesions, chronic salpingitis including the tuberculosis salpingitis [3].

Pseudocarcinomatous hyperplasia is histologically differentiated from adenocarcinoma by several morphological features, such as absence of grossly detectable tumor, unremarkable nuclear atypia, and mitosis with no evidence of invasion of the tubal wall such as in our case.

In this case we consider the serum CA-125 level. Serum CA-125 levels are significantly higher in patients with tubal neoplasms and can help in preoperative differentiation of benign and malignant tubal lesions [4].

CASE REPORT

A 26-year-old nulliparous patient was treated by her gynecologist with antibiotic therapy Doxycycline because of a positive finding of Ureaplasma and Chlamydia. Three months after finishing the antibiotic therapy, the patient was admitted into our hospital with dull pain in the lower parts of the abdomen.

The patient has had low-grade pain in the lower abdomen for the past 2 months. On vaginal examination painful tenderness was present while palpating the left adnexal lodge. The pelvic ultrasound exam showed well defined fluid-filled C -shaped tubular structure that arose from the upper left lateral margins of the uterus. The dilated zone, measuring 3 cm, was in the outer third of the fallopian tube. The tubal fluid with low signal intensity was suggestive of hydrosalpinx with presence of an enhancing intraluminal papillary projections and thick wall of the left tube (Figure 1).

The preoperative level of CA 125 was 30 U/ml and all the other laboratory findings were within normal range.

Laparoscopy was indicated with preoperative diagnosis of hydrosalpinx on the left fallopian tube. The intraoperative finding was a uterus with normal size and morphology. Periadnexal adhesions were present bilaterally. An intrauterine manipulator was used to facilitate vision and to introduce Methylene Blue for the Dye Test. After the injection of Methylene Blue both tubes showed no patency.

The left tube was fixed to the ovary with dense and vascular adhesions. Its distal ampulla diameter was around 3 cm (as measured previously on ultrasound). After the salpingectomy and cutting the tube wall, serous fluid was found in the lumen along with absence of folds on the mucosa. According to the classification of distal tubal occlusion (American Fertility Society) we decided to perform left salpingectomy.

The right fallopian tube presented with fimbriae phimosis (agglutination of the fimbriae) coexisting with periadnexal adhesions. Fimbrioplasty was performed with anatomical and functional repair of the infundibulum. The surgical repair was achieved with fine atraumatic forceps. We took account of the patency, wall, and tubal mucosa before our decision to do surgical repair.

The macroscopic finding revealed a left fallopian tube measuring 6.5 cm in length and 0.7-1.3 cm in diameter. In addition, 2 other smaller segments were part of the surgical specimen measuring 0.8 and 1.5 cm in the greatest diameter, macroscopically with features of tubal fimbriae. The dilated zone measuring 3 cm was in the outer third of the fallopian tube.

Microscopically, mucosal epithelial hyperplasia was identified, with papillary growth and focal fusion of edematous plicae. The epithelium showed nuclear crowding with unremarkable stratification and abundance of inflammatory cells in the background. Cytological atypia and mitoses were unremarkable. A few cells in the subepithelial portion of the tubal plicae were arranged in small nests with pseudo glandular arrangement but with benign features.

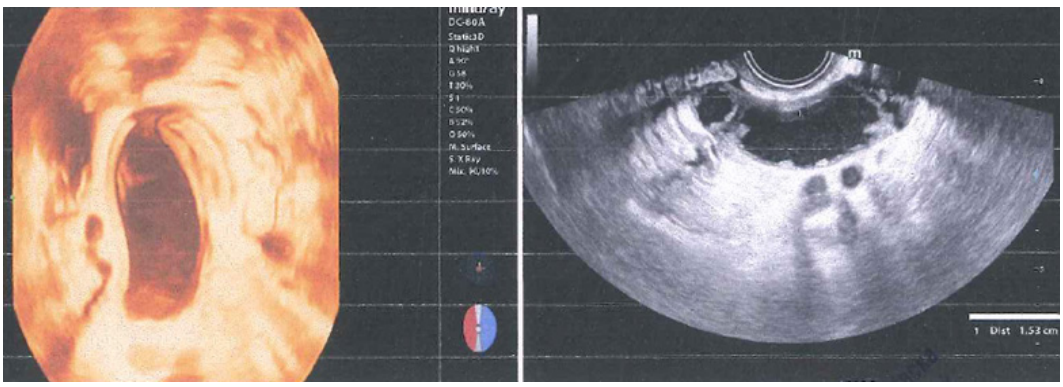


Figure 1. *Transvaginal ultrasonography easily detects the presence of left hydrosalpinx, appears as a sausage-shaped, with well-defined walls and papillary projections into the tubal lumen (arrows).*

The final histopathological diagnosis was pseudocarcinomatous hyperplasia of tubal mucosa associated with chronic salpingitis on the left tuba (Figure 2, 3 and 4).

Two control check-ups were done postoperatively after 6, 12 and 18 months. The patient was with laboratory findings within the normal range and in a good condition. Ultrasonography examination after 6 -, 12- and 18-month post-surgery showed no pathological findings in the adnexal lodges. The microbiological analysis was negative. The serum levels of CA-125 were within the normal range 6-, 12- and 18 months post-surgery.

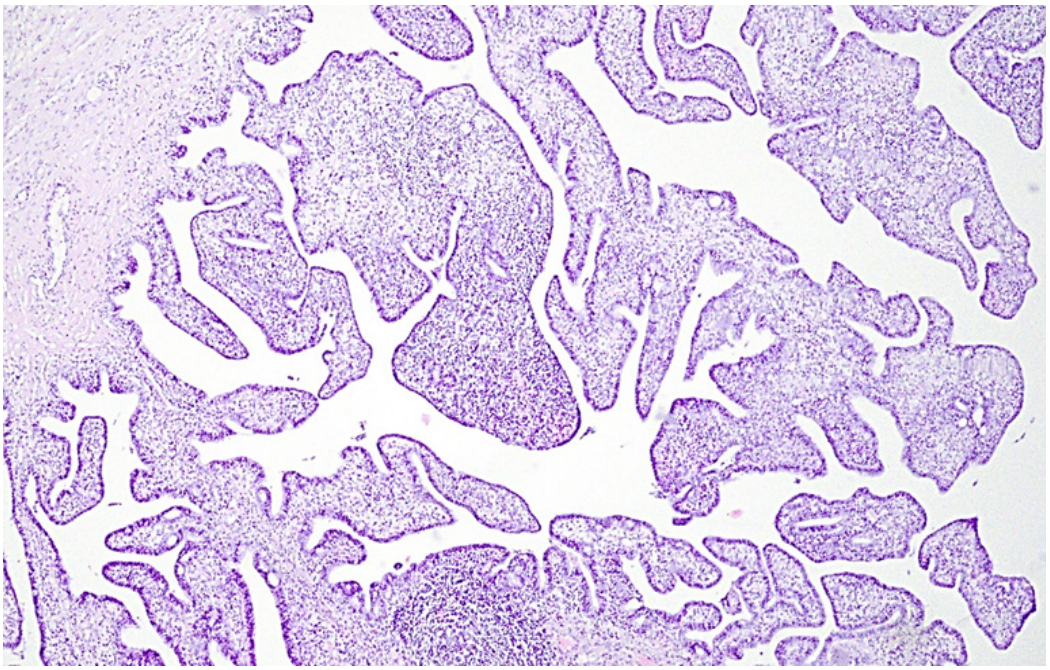


Figure 2. *Papillary growth and focal fusion of edematous plicae with inflammatory cells in the submucosal space (H&E, x4)*

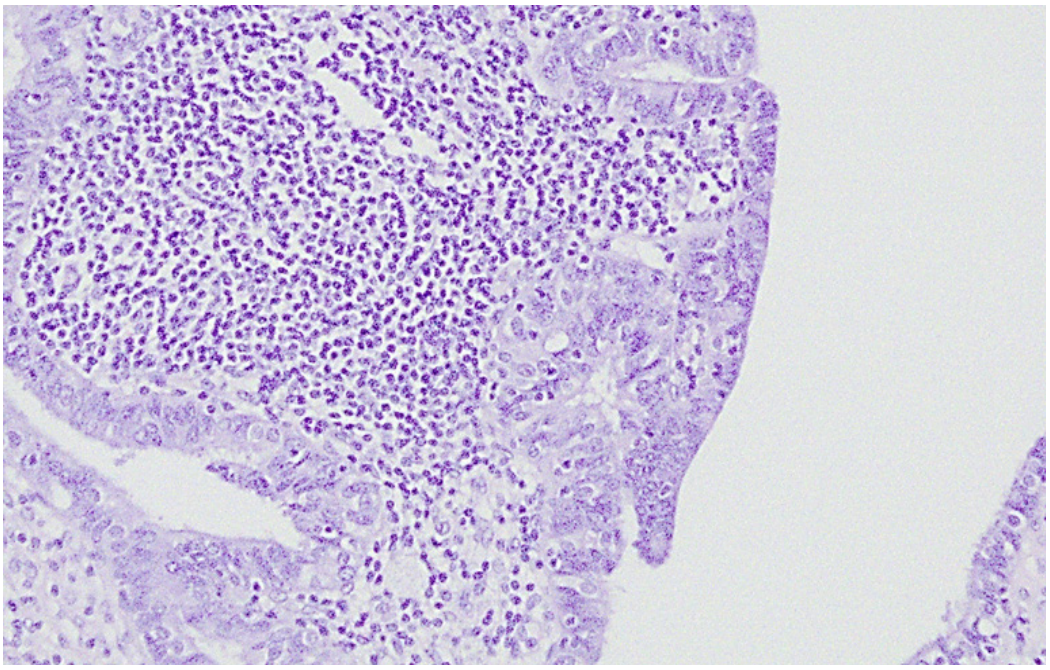


Figure 3. *Nuclear crowding of the epithelium with unremarkable stratification and abundance of inflammatory cells in the background. (H&E, x20)*

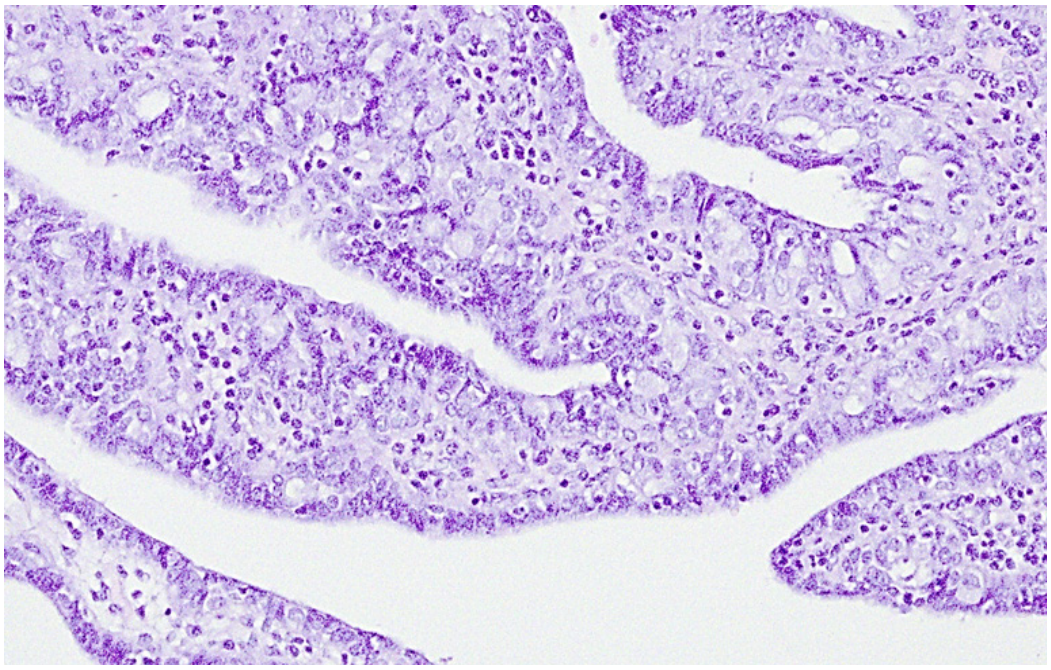


Figure 4. *Cytologic atypia and mitoses were unremarkable although the microscopic view mimics neoplastic tissue (H&E, x20)*

DISSCUSION

Pseudocarcinomatous hyperplasia is a rare entity that shows pathohistological signs of florid epithelial hyperplasia with atypical features.

In the available literature there are less than 20 case reports of patients aged 17-40. Almost half of them are associated with chronic salpingitis, while others are associated with pyosalpinx, tubo-ovarian abscess or hydrosalpinx [1]. Gupta et al. associated this entity with tuberculous salpingitis mimicking tubal adenocarcinoma [3]. A variety of benign conditions can be misinterpreted as malign neoplasms, both clinically and pathologically. At the very beginning, because of scarce information in the literature, the initial cases lead to radical hysterectomy because of the wrong diagnosis of carcinoma.

In our case, the finding was associated with chronic salpingitis. Some characteristics ease the differential diagnosis of pseudocarcinomatous hyperplasia and tubal carcinoma (Figure 3). Primary, the age of the patients. Usually, patients with pseudocarcinomatous hyperplasia are young. Secondly, there is no clearly visible tumorous formation involving the ovary. And as third, the patients with pseudocarcinomatous hyperplasia show clinical and laboratory signs of PID, while the patients with tubal carcinoma do not [5].

However, sometimes radiological examination can lead to exaggeration of differentiating chronic salpingitis from tubal tuberculosis and tubal carcinoma [5]. Tubal carcinoma is manifested with dilated tube that can resemble a hydrosalpinx or tubo-ovarian abscess. On the other hand, understanding of the pathogenesis and clinical manifestations of hydrosalpinx may aid for proper diagnosis of complex adnexal masses at MR imaging enabling avoidance of unnecessary procedures [6].

Serum CA 125 levels preoperatively help distinguish benign from malignant adnexal masses, as serum CA 125 levels are much higher in patients with tubal carcinoma. [3]. Despite this, the preoperative CA 125 level has a low specificity needed to diagnose the presence of malignancy [4, 7].

In the presence of hydrosalpinx the outcomes of reconstructive surgery performed by laparoscopy are not encouraging in terms of pregnancy rate [8, 9]. Therefore, in our case we decided to do salpingectomy to the left fallopian tube. On the other hand, the right fallopian tube presented only with fimbriae phimosis, and we performed fimbrioplasty. The intrauterine pregnancy rate after laparoscopic fimbrioplasty varies between 40-48% [10].

Pseudocarcinomatous hyperplasia histologically is different from adenocarcinoma. Some

morphological characteristics, such as absence of macroscopic finding of a tumor, presence of severe chronic inflammation, absence of severe epithelial proliferation, mild nuclear atypia, absence of mitotic figures and no evidence of invasion within other layers, can help differentiate pseudocarcinomatous hyperplasia from tubal carcinoma. The absence of mitotic figures is an important criterion for pathological diagnosis of this condition. In our case mitotic figures were unremarkable.

CONCLUSION

As a conclusion, pseudocarcinomatous hyperplasia of the fallopian tube is a benign, reactive response to the primary inflammatory or neoplastic process. The pathohistological diagnosis depends mostly on the morphological characteristics that help differentiate pseudocarcinomatous hyperplasia from tubal carcinoma.

Because of the similar morphological, histopathological, and clinical characteristics, we ought to be careful with the diagnosis to provide the right diagnosis and proper management for the condition for the patient.

REFERENCES

1. Cheung AN, Young RH, Scully RE. Pseudocarcinomatous hyperplasia of the fallopian tube associated with salpingitis. A report of 14 cases. Am J Surg Pathol. 1994; 18:1125–30.
2. Limaiem F, Chelly I, Mekni A, et al. Challenging diagnosis: florid epithelial hyperplasia versus adenocarcinoma of the fallopian tube. Pathology. 2010; 42:99–101.
3. Gupta S, Singh P, Bala J, et al. Pseudocarcinomatous hyperplasia of the fallopian tubes which was associated with female genital tract tuberculosis, histologically mimicking tubal adenocarcinoma: a diagnostic challenge. J Clin Diagn Res. 2012; 6:1419–21.
4. Suh DS, Moon SH, Kim SC, et al. Significant simultaneous changes in serum CA19-9 and CA125 due to prolonged torsion of mature cystic teratoma of the ovary. World J Surg Oncol. 2014; 12:353.
5. Rawal G, Zaheer S, Yadav AK et al. Pseudocarcinomatous Hyperplasia of the Fallopian Tube Mimicking a Tubal Neoplasm: A Rare Entity.J Midlife Health. 2018; 9(2): 104–105.
6. Lee NK, Choi KU, Han GJ, et al. Pseudocarcinomatous hyperplasia of the fallopian tube mimicking tubal cancer: A radiological and pathological diagnostic challenge. J Ovarian Res. 2016; 9:79.
7. Ma Y, Duan W. Clinical and survival analysis of 36 cases of primary fallopian tube carcinoma. World J Surg Oncol. 2014; 12:311.
8. Mencaglia L, Minelli L, Wattiez M. Manual of Gynecological Laparoscopic Surgery. Endo-Press® Tuttlingen. Germany. 2008; 257:155.
9. Kim MY, Rha SE, Oh SN, et al. MR Imaging findings of hydrosalpinx: a comprehensive review. Radiographics. 2009; 29:495–507.
10. Mencaglia L, Minelli L, Wattiez M. Manual of Gynecological Laparoscopic Surgery. Endo-Press® Tuttlingen. Germany. 2008; 257: 110-111.

Резиме

ПСЕВДОКАРЦИНОМАТОЗНА ХИПЕРПЛАЗИЈА НА ТУБАРНА МУКОЗА ПОВРЗАНА СО ХРОНИЧЕН САЛПИНГИТИС – КЛИНИЧКИ И ПАТОЛОШКИ ПРЕДИЗВИК: ПРИКАЗ НА СЛУЧАЈ

Снежана Булиќ[1], Милица Каламарас[1], Џенгис Јасар[2], Катерина Кубелка Сабит[2], Глигор Димитров[1], Митко Ивановски[1], Ева Палчевска[1], Јована Топенчарова[1]

[1] Оддел за гинекологија и акушерство, болница „Аџибадем Систина“, Скопје, РС Македонија

[2] Хистопатолошки оддел, болница „Аџибадем Систина“, Скопје, РС Македонија

Псевдокарциноматозната хиперплазија на тубарната слузница е редок ентитет, кој се јавува како реактивен одговор на основниот воспалителен или неопластичен процес. Прикажуваме случај на псевдокарциноматозна хиперплазија кај 26-годишна жена со клиничка симптоматологија на пелвична инфламаторна болест и ниво на CA 125 во серум во референтни вредности (30 U/ml). Ултразвучниот наод покажа едностранo кобасичесто проширување на Фалопиевата туба, кое се наоѓаше на горната латерална маргина на матката. Младата возраст на пациентката, присуството на хронично воспаление, епителната хиперплазија со незабележителна нуклеарна атипија и митоза го олеснуваат поставувањето на точната дијагноза. Сепак, псевдокарциноматозна хиперплазија на туба може клинички и патолошки да имитира неопластичен процес. Затоа е потребно да се земат предвид диференцијалните морфолошки и клинички карактеристики меѓу двата ентитета за да се постави точна дијагноза и правилен третман.

Клучни зборови: псевдокарциноматозна хиперплазија, хроничен салпингитис, фалопиева туба, неоплазма