ISSN 1941-5923 © Am J Case Rep, 2014; 15: 361-363 DOI: 10.12659/AJCR.890921

Received:2014.04.24Accepted:2014.05.13Published:2014.08.29

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Endosalpingiosis in Conjunction with Ovarian Serous Cystadenoma Mimicking Metastatic Ovarian Malignancy

ABCDEF Study Design A Data Collection B Statistical Analysis C ata Interpretation D Iscript Preparation E Literature Search F Funds Collection G		ABCDEFG 1 ABCDEFG 1 BDE 2 BD 3	Namrata Singhania Neha Janakiraman Douglas Coslett Navid Ahmad	 Department of Family Medicine, Wright Center for Graduate Medical Education, Wilkes Barre, PA, U.S.A. Department of Obstetrics and Gynecology, Wilkes Barre General Hospital, Wilke Barre, PA, U.S.A. Department of Pathology, Wilkes Barre General Hospital, Wilkes Barre, PA, U.S.A 	
	Corresponding Authors: Conflict of interest:		Namrata Singhania, e-mail: namrat09@gmail.com and Neha Janakiraman, e-mail: nehavenkateshj@gmail.com None declared Female, 26 Endosalpingiosis Chronic pelvic pain — Diagnostic laproscopy (conservative management) Obstetrics and Gynecology		
Patient: Final Diagnosis: Symptoms: Medication: Clinical Procedure: Specialty: Objective: Background: Case Report: Case Report: Conclusions: MeSH Keywords: Full-text PDF:		Patient: iagnosis: mptoms: dication: rocedure: specialty:			
		Dbjective: :kground: :e Report: nclusions:	 Challenging differential diagnosis Interesting and unusual case of endosalpingiosis mimicking ovarian malignancy presentation. A 26-year-old GOPO white female presented to our office with chronic pelvic pain. On vaginal examination, a nontender mass in left the adnexal region was palpable. Transvaginal ultrasound showed a left ovarian cyst. Laparoscopy was performed, which revealed diffuse bilateral ovarian excrescences with unusual multiple studdings throughout the peritoneum and abdominal cavity. Due to a suspicion of malignancy, a biopsy specimen was obtained for frozen sectioning. The specimen proved to be consistent with benign papillary serous cystadenofibroma. Gross appearance was still suspicious for malignancy and therefore left paraovarian cystectomy was performed. Additional specimens showed ovarian adenofibroma and endosalpingiosis. The patient's complaint of pelvic pain improved after laparoscopy. Due to diffuse presentation of endosalpingiosis in the peritoneum, serial CT scan of abdomen and pelvis at 6-month intervals was recommended. To our knowledge, this is an unusual case of a young, nulliparous female presenting with diffuse-presentation endosalpingiosis in the abdomen and peritoneum, which on gross examination was suspicious for malignancy. By following a conservative approach and performing serial CT scans, the patient will be clinically monitored. 		
		eywords:	Cystadenofibroma • Endometriosis • Pelvic Pain http://www.amjcaserep.com/abstract/index/idArt/890921		
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Background

Endosalpingiosis (ES), also known as müllerian cyst of uterus [1] or müllerianosis [2] is a rare gynecological disorder of müllerian origin [2,3], which has a pathogenesis similar to endometriosis (EM). This condition was first reported in the literature in 1930 [4]. It is a benign condition characterized by the presence of ectopic epithelium with tubal differentiation, which involves structures of the female genital tract, peritoneum, and sub-peritoneal tissues (uterus, ovaries, fallopian tubes, bladder, colon, omentum, lymph nodes, and skin) [2]. Pathogenesis is believed to be metaplastic transformation of coelomic epithelium into tubal-like epithelium [2,4,5]. Most cases have been reported in postmenopausal women [6-8], but a few cases have been reported in younger patients [9]. Approximately 34% of reported cases have been associated with endometriosis [8]. It is commonly diagnosed incidentally during work-up of other conditions such as infertility, chronic pelvic pain, pelvic mass, menstrual irregularity [10], and chronic back pain [11]. However, most of these symptoms have been reported to be an association with ES rather than being caused by it [8,10]. A potential link between ovarian ES and the development of pelvic serous carcinoma in women who are BRCA mutation carriers has been observed in 1 case report [12]. Malignancy has been reported to occur significantly more in premenopausal women with ES than without it [8].



Figure 1. (A) Left ovary showing excrescences and large ovarian cyst. (B) Right ovary showing excrescences.



Figure 2. Cul-de-sac showing studdings.



Figure 3. Left uterosacral ligament with studdings.

In our patient, ovarian histopathology was positive for serous cystadenoma (OSC), which is a benign ovarian epithelial tumor [13], with peak age incidence at age 40–50 years. It is bilateral in around 15% of cases and can present with pelvic pain, discomfort, or asymptomatic pelvic mass discovered on exam.

Case Report

Our case was a 26-year-old white, nonsmoker, nonalcoholic, nulliparous female with body mass index (BMI) of 40 who presented with a 3-year history of lower pelvic pain and no history of grade B symptoms or family history of gynecological cancers. On physical exam, external genitalia, vagina, and cervix were unremarkable; however, pelvic exam revealed a palpably enlarged left adnexa. Pelvic ultrasound revealed a left ovarian cyst (7×3×4 cm). For further work-up, a laparoscopy was performed. This revealed large ovarian masses (4-5 cm) with multiple surface excrescences (Figure 1A, 1B) and tissue studding throughout the cul-de-sac (Figure 2), and involving the left uterosacral ligament (Figure 3), bilateral pelvic gutters, vesical peritoneum, and right hemidiaphragm, superficial surface of the liver, and posterior serosa of the cervix. The frozen section was positive for nonmalignant papillary cystadenofibromas. However, due to suspicious appearance of the masses, additional biopsies and left paraovarian cystectomy were



Figure 4. Histopathological slide showing benign ciliated epithelium with psammoma body.

performed and sent for histopathology. Histopathological results revealed serous cystadenoma in bilateral ovaries; however, the cul-de-sac and bladder peritoneum biopsies were positive for ES and psammoma bodies (Figure 4). Peritoneal washings were negative for malignant cells. Since implications of the diffuse process were not clear and because it was not clear if it was a precursor of malignancy, the patient was evaluated by a gynecologic oncologist. The patient was advised to have serial CT scans of the abdomen and pelvis every 6 months for the

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first year, followed by annual scans for 5 years. Her follow-up ultrasound 6 months after surgery was within normal limits.

Discussion

Endosalpingiosis is a benign condition caused by ectopic tubal epithelium, similar in pathogenesis to endometriosis, which refers to the presence of ectopic, functional endometrial tissue. ES is usually incidentally diagnosed during work-up for other conditions. It typically does not become malignant but it may present as a large mass or may spread to various organs, including ovaries, uterus, bladder, liver, omentum, and peritoneum [5] and can mimic malignancy [14–16].

Conclusions

Clinicians should be aware of ES, which is benign and does not require extensive surgery. If symptomatic, simple resection of the mass may offer relief.

Conflicts of interest

None.

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