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Case Report

Diffuse abdominal and pelvic endosalpingiosis: A case report[☆]

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ABSTRACT

Endosalpingiosis is defined as the ectopic location of benign ciliated tubal epithelium outside of the fallopian tubes. It is a rare entity that was previously regarded as an incidental finding on pathology, and is becoming more prevalent within the medical literature. Diagnosis is made based on histologic sampling. There are no specific radiological features but commonly reported findings include numerous cystic and solid masses scattered throughout the pelvis. Common ectopic locations seen on imaging include the serosa of the uterus, fallopian tubes, ovaries and the pelvic cul-de-sac. Less common locations include the bladder wall, omentum, bowel serosa, and skin. We present the clinical presentation of a patient with histologically proven endosalpingiosis. Atypical imaging findings and correlative histology are also reviewed.

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Introduction

Endosalpingiosis is characterized by the ectopic presence of glandular tissue consisting of benign ciliated tubal epithelium outside of the fallopian tubes [1]. Endosalpingiosis is one of a spectrum of pathologies that involve the secondary mullerian system; the others include endometriosis and endocervicosis [2]. Common ectopic locations include the uterine serosa, fallopian tubes, ovaries and pouch of douglas. Less

common locations include the omentum, small bowel serosa, and skin. Commonly reported imaging findings are of well circumscribed pelvic masses with variable cystic and solid components with varying enhancement patterns [3].

This paper presents the case of a 47-year-old gravida 2 para 2 woman who presented with abdominal pain and distention. The imaging demonstrated diffuse abdominal and pelvic involvement by endosalpingiosis, a finding commonly reported, but less commonly depicted on diagnostic imaging.

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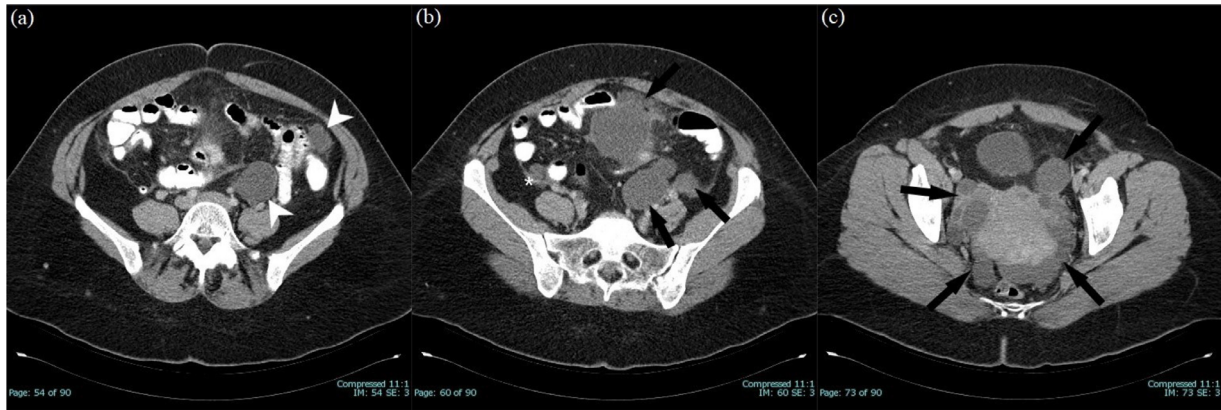


Fig. 1 – Computed axial tomography images, axial views (a-c) shown above. A cystic lesion is seen in the lower abdominal peritoneum and in the greater omentum on the left (white arrowhead)(a). Cystic lesions are also noted outlining the uterine serosa and bilateral adnexa with impression on the left ureter (black arrow) (b,c). A solitary cystic lesion is seen contacting the appendiceal tip (asterisk) (b).



Fig. 2 – Computed axial tomography images, coronal views (d-f) shown above. A large cystic lesion within the pelvis demonstrating thin rim enhancement and mild surrounding inflammatory changes (black arrowhead) (d). Coronal views show numerous cystic lesions abutting small and large bowel loops, uterine serosa, and bilateral adnexa (white arrow) (e,f).

Case Report

A 47-year-old (G2P2) premenopausal female presented to her primary care physician with complaints of lower abdominal pain and distention. A CT scan of the abdomen and pelvis with IV contrast was obtained (Figs. 1, 2). CT images demonstrated extensive cystic lesions which were seen intimately related to the uterus and bilateral adnexa. There was also cystic involvement of the greater omentum, multiple small bowel loops, pelvic cul-de-sac, appendix, and bilateral adnexa. One of the cystic lesions within the pelvis demonstrated thin mural enhancement and surrounding fat infiltration. The imaging differential diagnosis included ovarian malignancy, atypical pelvic inflammatory disease, and endometriosis.

Subsequent follow up with OB/GYN elicited a more detailed history, with the patient describing the sudden onset of pain and bloating. The pertinent medical history included obesity and diverticulosis. Surgical history included a prior ce-

sarean delivery. There was no reported history of pelvic inflammatory disease or abdominal infection. The patient denied changes in bladder or bowel habits, abnormal uterine bleeding, rectal bleeding, hematuria, recent weight changes or fevers/chills. Physical exam revealed mild abdominal tenderness to palpation as well as a midline mass that was palpated just superior to the pubic symphysis. Pelvic examination demonstrated no cervical motion tenderness and nonpalpable ovaries. Blood work demonstrated a minimally elevated CA-125 of 58 U/mL (normal ≤ 35), and normal CA 19-9 and CEA levels.

Findings at this point favored inflammatory disease or endometriosis and the patient was suggested to undergo robot-assisted laparoscopy. During surgical evaluation, numerous large cystic masses were identified that were densely adherent to small bowel and mesentery. Other smaller cystic masses were adherent to the omentum, left pelvic sidewall, pelvic cul-de-sac and the appendix. Small bowel loops were dilated proximal to the adherent lesions consistent with a partial

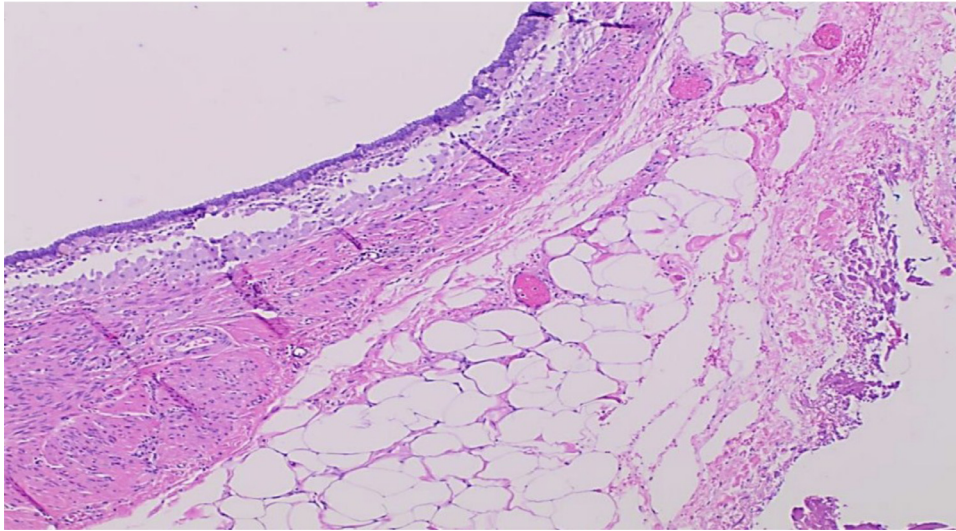


Fig. 3 – Left pelvic sidewall cyst with a single layer of ciliated glandular epithelium surrounded by macrophages and a cuff of metaplastic smooth muscle (H & E, x100).

small bowel obstruction. The cystic masses contained clear serous fluid. Intraoperative frozen sections were obtained and showed benign appearing serous cystic masses with fibromuscular components and some atypia. Due to the diffuse nature of disease a robotic assisted total laparoscopic hysterectomy, bilateral salpingectomy, left oophorectomy, right ovarian cystectomy, partial omentectomy, appendiceal cystectomy, and diffuse lysis of adhesions was performed. Patient recovered uneventfully following the procedure.

Final pathology demonstrated benign endosalpingiotic and endometriotic cysts with smooth muscle metaplasia identified within the left ovary, left pelvic sidewall, the peri-appendiceal region, omentum, and the mesentery (Fig. 3). The uterus demonstrated multiple leiomyomata and patchy adenomyosis. There were no findings of neoplasia. Findings were compatible with endosalpingiosis, endometriosis, uterine leiomyoma, and adenomyosis.

Discussion

During embryogenesis, the paired müllerian (paramesonephric) ducts partially fuse in order to form the primitive fallopian tubes, uterus, cervix and proximal third of the vagina. These structures are referred to as the primary müllerian system. It is hypothesized that vestigial remnants of this system remain and are termed the secondary müllerian system [8].

Numerous conditions of the secondary müllerian system exist, the most commonly known being endometriosis, with lesser known entities including endocervicosis and endosalpingiosis. Although these disease processes can occur independently, they commonly occur together. In a retrospective analysis of 110 cases of histologically proven endosalpingiosis, Prentice et al. demonstrated 72 (65.5%) patients with isolated endosalpingiosis and 38 (34.5%) patients with coexisting

endometriosis. They also demonstrated an increase in concurrent malignancy. While endometrial adenocarcinoma was the most common, associations with cervical and serous ovarian cancers were also shown. A study by Esselen et al also demonstrated a statistically significant association of endosalpingiosis with endometriosis, uterine malignancy and ovarian malignancy.

Common locations for endosalpingiosis include the serosa of the uterus, fallopian tubes, ovaries and the pelvic cul-de-sac. Less common locations include the bladder wall, omentum, bowel serosa, and skin [3]. There have been few reports of endosalpingiosis involving the appendix [7], as well as full thickness infiltrative involvement of the cervix [5]. In our case, the patient demonstrated involvement in all the commonly reported locations with lesions also noted abutting the small bowel, left ureter, appendiceal tip, and within the omentum leading to a partial small bowel obstruction.

Imaging findings of endosalpingiosis are relatively well reported within the literature. Ultrasound findings typically demonstrate expansile hyperechoic or anechoic masses within the pelvis. CT findings commonly demonstrate well circumscribed masses within the pelvis that demonstrate variable cystic and soft tissue components. When there is involvement of the peritoneum, CT may demonstrate numerous calcified granular nodules within the pelvic peritoneum without ascites [3]. Few cases with associated MR findings have been reported, however findings in these cases showed numerous cystic lesions within the pelvis demonstrating hyperintense T2 and iso-hypointense T1 signal [4,6]. Enhancement of internal septations was noted in one case [4].

Imaging findings in this case demonstrated purely cystic lesions scattered throughout the abdominal and pelvic peritoneum with involvement of the appendiceal tip, small bowel loops, omentum, uterus, fallopian tubes, and bilateral ovaries. To the best of our knowledge, such widespread involvement of the abdomen and pelvis by endosalpingiosis has not been reported in the imaging literature. It is important that the di-

agnostic imaging community consider endosalpingiosis and its spectrum of pathology when encountering a case as the one presented. Future work could aim at identifying imaging findings specific to endosalpingiosis.

Patient Consent

Appropriate informed consent was obtained by the patient prior to publication of the information contained within this article.

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