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Case Report

A case report on peritoneal inclusion cyst with entrapped ovary and an endometriotic cyst causing diagnostic dilemma

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ABSTRACT

Peritoneal inclusion cysts (PIC) occur most frequently when abdominal adhesions attach to an ovary. These adhesions are commonly seen in patients with a history of abdominal surgery. Physiologic fluid following ovulation that would normally be absorbed by the peritoneum gets entrapped within these adhesions. Women most frequently complain of back pain or pelvic pain when peritoneal inclusion cysts are present. We present a patient with prior surgical history who presented with suprapubic pain and was found to have a complex cyst on imaging. Gynecologic oncology was consulted due to its complex imaging appearance and large size and was ultimately resected despite negative tumor markers. We present our case with pathologic correlation of imaging findings to emphasize this benign entity with no malignant potential. A confident imaging diagnosis in correlation with laboratory markers can help prevent aggressive surgical management.

Keywords: Peritoneal inclusion cyst, Peritoneal pseudocyst, Peritoneal inflammatory cysts, Entrapped ovarian cyst

INTRODUCTION

Peritoneal inclusions cysts (PIC), also known as peritoneal pseudocysts, entrapped ovarian cysts, or peritoneal inflammatory cysts, are formed when there are abdominal adhesions to an active ovary.[1-4] During the reproductive years, the ovaries are the primary source of peritoneal fluid. This fluid is normally absorbed by the peritoneum. However, circumstances such as inflammation, infection, or surgery can potentially decrease the absorptive capacity of the peritoneum and cause exudative fluid from the ovary post ovulation to accumulate between the ovary and adhesions.[2,3]

PIC is benign reactive mesothelial proliferations that can vary widely in size. They are typically found in the third or fourth decade of life. However, there have been studies reporting PIC in adolescent females, most with prior history of surgery which was usually in infancy for congenital genitourinary anomalies, necrotizing enterocolitis, etc.[5,6] Typical symptoms include acute or chronic abdominal and/or pelvic pain, and back pain. However, they can also present with tenesmus, urinary incontinence or frequency, anorexia, and dysfunctional uterine bleeding.[4] PIC are nearly exclusively seen in patients with a history of abdominal or pelvic surgery, usually between 6 months up to 20 years after the surgery.[2] Literature mentions PIC after laparoscopic surgery is rare compared to abdominal hysterectomy and for the same reason, laparoscopic resection is preferred over laparotomy when considering surgical resection of PIC.[7] PIC can also be seen with chronic inflammatory processes

including endometriosis, inflammatory bowel disease, and pelvic inflammatory disease.[4,7,8] We present a patient in the reproductive age group with an extensive history of abdominal surgeries, myriad of typical and atypical clinical symptoms, and negative tumor markers.

CASE REPORT

A 48-year-old multiparous female with a past surgical history of multiple abdominal surgeries including open ovarian cyst removal, open appendectomy, and cholecystectomy, presented to her gynecologist with abdominal pain, early satiety, bloating, and menorrhagia. On physical exam, she had diffused suprapubic pain with mild rebound tenderness and fullness, but no discrete mass was palpable. Her initial laboratory tests including a CBC, BMP, urinalysis were within normal limits and a negative pregnancy test. Her subsequent post-imaging workup with tumor marker levels (CA 19-9, CA 125, and CEA) were also negative.

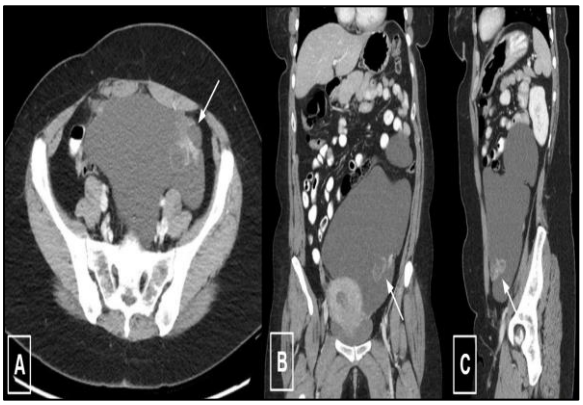

Figure 1: CT abdomen and pelvis: Axial (A), coronal (B) and sagittal (C) CT images of the abdomen and pelvis demonstrating 20x6x20 cm large, irregular shaped, abdominopelvic cystic lesion with heterogeneous appearing left ovary (white arrow) containing follicles and mildly enhancing irregular ovarian tissue. The mass displaces the non-dilated small bowel loops to the right abdomen without compression.

A CT of the abdomen and pelvis (Figure 1A-C) showed a large volume left the abdominopelvic peritoneal fluid collection with loculations, thin septations, and irregular soft tissue associated with this collection. An MRI of the pelvis with and without contrast was obtained (Figure 2A-D) which demonstrated an irregular T1 hypointense, T2 hyperintense loculated fluid collection with non-enhancing thin septations primarily arising from the left hemipelvis and extending superiorly into the abdomen. The lesion measured 20×6×20 cm. The left ovary was seen along the left anterior margin of this complex fluid collection. The ovary had T2 hyperintense follicles and also T1 hyperintense, T2 hypointense hemorrhagic blood products within a few of these follicles. These findings were consistent with a mildly complex peritoneal inclusion cyst with an entrapped left ovary. Due to the complex nature of the cyst, a referral was made to gynecologic oncology. Other findings on imaging included mildly thickened endometrium that measured 1.3 cm and a few nabothian cysts in the cervix.

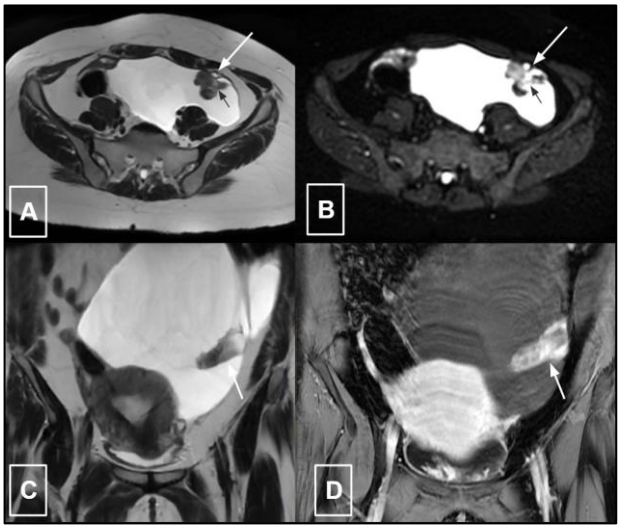

Figure 2: MRI of the pelvis: A- Axial T2-weighted image, B- Axial diffusion-weighted image (DWI), C- Coronal T2-weighted image, D- Coronal T1 post-contrast image with fat suppression. Large T1 hypointense, T2 hyperintense irregular-shaped cyst with entrapped normal-sized left ovary (white arrow) with altered morphology along the left anterior margin of this cyst (A, B, C). Small areas of intrinsic T1 hyperintensity (not shown here) with T2 shading which show areas of restricted diffusion representing hemorrhagic blood products from endometriosis (black arrow) (A, B, C). Enhancement was noted in the normal ovarian tissue (D).

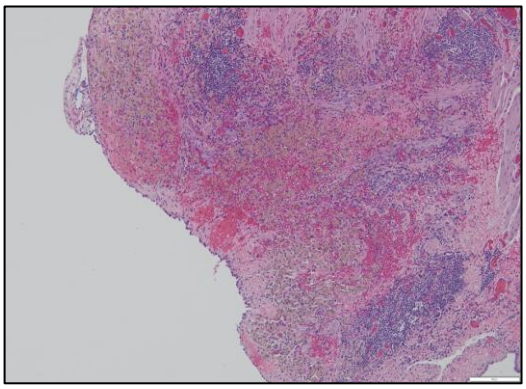

Figure 3: The histology image shows a fallopian tube with shortened, fused plicae, and a chronic inflammatory infiltrates consisting of mature lymphocytes and pigment-laden macrophages, consistent with chronic salpingitis.

Gynecologic Oncology physicians recommended diagnostic laparoscopy, exploratory laparotomy, left

salpingo-oophorectomy, dilation and curettage given the complex appearance of the cystic mass despite negative tumor markers.

Her surgery was complicated due to extensive abdominal adhesions and was converted to an open laparotomy. 2 liters of straw-colored peritoneal fluid drained from the cyst. Pathology findings of the left fallopian tube and ovary showed endometriosis and chronic salpingitis. There were no signs of malignancy on the pathology examination (Figure 3, 4).

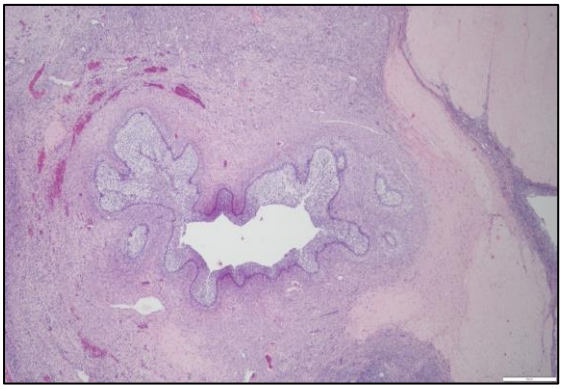

Figure 4: Histology image shows an ovary with a corpus luteum cyst, along with corpus albicans. Both findings are consistent with normal physiologic change.

DISCUSSION

PIC is caused by an excess of peritoneal fluid inside previously existing abdominal adhesions, which is often exudative in nature.1 PIC typically occurs in women with an incidence rate of 82% in the ratio of 4-5:1 female to male.[7,8] Peritoneal inclusion cysts have been shown to have estrogen and progesterone receptors on the cyst walls.[4] This fluid has also been seen to fluctuate with steroid hormones, and with the menstrual cycle in women: during the follicular phase with an abrupt increase during ovulation. Peritoneal inclusion cysts have been shown to decrease in size after menopause and with the usage of combined oral contraceptives.

Pathologically, PIC is pseudocysts that are non-neoplastic mesothelial proliferation within inflamed fibrous granulation tissue walls. In long-standing cysts, the cyst wall can get epithelialized and the epithelial cells can produce fluid resulting in enlarging cysts.[7] The cysts can contain serous, mucinous, or gelatinous fluid which is the reason for the variable appearance of cyst contents on imaging.[3,5]

PICs are mostly not palpable on the clinical examination like in our patient and hence imaging helps in the evaluation of these patients who usually present with abdominal or pelvic pain.[1,5] Ten percent of small-sized PIC are incidentally detected. Since they present with intermittent abdominal pain and typically in a patient with prior abdominal or pelvic surgery, mechanical bowel obstruction should be ruled out as a cause of pain. Though PIC is from the adhesion of the peritoneum to the ovary, there is generally no adhesion with bowel loops in these cases. The pain reported in these patients is thought to result from torsion of the ovarian stalk within the PIC. Imaging evaluation is usually performed with ultrasound and CT. MRI is used as a problem-solving tool in complex appearing cysts. The white paper of the ACR incidental findings committee mentions no further evaluation is required in an incidental PIC.[10] The PIC has an irregular shape due to pseudocyst lacking wall and conforms to shape from the adjacent structures.[5,8] Ultrasound demonstrates anechoic cysts with thin septations <5 mm and in case of hemorrhagic or proteinaceous contents, they can appear as echogenic cysts with septations. The ovary entrapped in the cyst between adhesions can have a “spider in a web” appearance however the ovaries are not usually clearly delineated on ultrasound like in an MRI.[2,3,5,8] Doppler examination will demonstrate a low-pressure flow pattern with resistive index (RI) of 0.50- 0.60 in the septal wall.[2] CT similarly depicts fluid density mass with irregular borders with attenuation varying depending on the fluid contents.[3,5,8] On MRI, the lesion with serous content demonstrates T1 hypointensity and T2 hyperintensity; and the one with hemorrhagic content demonstrates T1 hyperintensity with T2 intermediate or hypointensity.[8]

The differential diagnosis for this condition includes paraovarian cyst, hydrosalpinx or pyosalpinx, lymphocele, mesenteric or omental cysts, pseudomyxoma peritonei, malignant mesothelioma, and adenomatoid tumors.[2,3,4] Paraovarian cysts should be separate with a normal ipsilateral ovary separated from the cyst.[2] When the cyst collects loculated fluid adjacent to the uterus, mesothelial tissue can project into the lumen, creating a cogwheel appearance with adhesions contained inside the fluid that can mimic hydrosalpinx. Patients with pyosalpinx have cysts with echogenic fluid and constitutional symptoms such as fever and leukocytosis. Lymphoceles are usually thin-walled, unilocular simple fluid collections seen following lymphadenectomy surgery and are seen along the course of pelvic vessels.[9] Lack of smooth muscle component in the walls of cyst favors PIC over cystic mesothelioma.[2] Adenomatoid tumors have soft tissue components along with cystic lesions which differentiate them from PIC. Malignant mesothelioma is a peritoneal tumor likely from asbestos exposure with solid components and infiltrative features which are not seen with PIC.[10]

The treatment options available to patients with PIC include observation with imaging follow-up, pain medications, hormonal therapy to suppress ovulation (thereby decreasing fluid from ovulation being trapped between adhesions), image-guided sclerotherapy to disrupt the internal septae of the cysts. This has been shown to decrease the size by >50% and complete resolution in

>50% of patients. Cystic fluid aspiration through the endovaginal approach can be used in select cases.[3] Laparoscopic resection of a cyst or surgical lysis of adhesions can be attempted in patients who do not improve with conservative management and those who have infertility from an entrapped ovary.[2,3]

CONCLUSION

PIC is a benign pathology, almost exclusively occurring in premenopausal women with risk factors of prior history of surgery, trauma, or infection. The clue to the diagnosis in complex PIC lies in negative serum tumor markers for ovarian malignancy. It has no malignant potential however; recurrence is seen in 30-50% of patients following surgical resection of PIC. Hence, conservative treatment should be attempted first and surgery (laparoscopy preferred over laparotomy) resorted only in patients with failure of medical management or those with infertility problems.

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