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Case Report GI/GU/Thoracic/Non Vascular Interventions

Recurrent symptomatic abdominopelvic cystic lesions of unknown etiology treated with image-guided sclerotherapy: A case report

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ABSTRACT

This case report highlights the diagnostic approach and treatment for 30 cm bilateral multiloculated pelvic peritoneal cystic masses. The patient had a history of spina bifida with ventriculoperitoneal shunt placement and neurogenic bladder with partial cystectomy and ileal conduit diversion. The cysts were suspected to be peritoneal inclusion cysts. Due to prior failed surgical intervention, the patient was treated with image-guided sclerotherapy.

Keywords: Adnexal mass, Image-guided, Peritoneal inclusion cyst, Sclerotherapy

INTRODUCTION

Enlarging multiloculated pelvic cysts are often misdiagnosed as ovarian malignancies. The differential for pelvic cysts can include ovarian tumor, paratubal cyst, lymphocele, hematoma, urinoma, cerebrospinal fluid pseudocyst (CSFoma), and peritoneal inclusion cyst. Ultrasound with color Doppler along with cancer antigen-125 (CA-125) levels and the use of a risk of ovarian malignancy algorithm can aid in assessing malignancy risk.^[1] Once malignancy risk has been assessed and risk is low, treatment options include observation, percutaneous drainage, image-guided sclerotherapy, and surgical intervention through laparoscopic or laparotomy approaches. Symptomatic cysts require intervention whereas asymptomatic cysts can be treated with expectant observational management. Symptomatic cysts can cause pelvic pain, pelvic fullness, or compressive mass effects including pain radiating to the back, dyspareunia, early satiety, changes in stool habits, or increased urinary frequency/pressure.^[2] Infertility has also been reported.^[1] Pain may be exacerbated if the cyst ruptures and irritates the peritoneum or if it becomes infected.

Ovarian cysts have been treated conservatively with hormonal therapy using combined contraceptive pills and/or drainage. Drainage often requires sonographic guidance or computed tomographic guidance if the cyst is deep within the pelvis. Other surgical treatment options include minimally invasive surgery or laparotomy for cyst excision; however, this is not appropriate for all patients such as those with extensive pelvic surgery history or high risk for anesthesia.^[3]

We present a case of chronic, large, and recurrent abdominopelvic cystic lesions in a medically complex patient with extensive prior surgical history, along with the diagnostic approach,

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treatment, and patient outcomes using a multidisciplinary team.

CASE REPORT

A 28-year-old woman presented with abdominal pain and had a complex medical history including spina bifida with prior ventriculoperitoneal (VP) shunt placement, neurogenic bladder with prior partial cystectomy, Mitrofanoff and ileal conduit diversion, as well prior Malone antegrade colonic enema procedure. Computed tomography (CT) imaging revealed $30 \times 25 \times 20$ -cm multiloculated cystic masses likely originating from ovaries/adnexa, extending from the level of the kidneys to the pelvic floor [Figure 1]. Tubing from the VP shunt was present in the peritoneal cavity.

Prior imaging revealed chronic fluid collections in the abdomen present for at least 10 years, with a significant increase in size in the past 5 years. Differential diagnosis included ovarian neoplasm, CSFoma, urinoma, and peritoneal inclusion cyst. The patient had previously had an exploratory laparotomy surgery for these cystic lesions revealing benign cyst wall; however, due to extensive pelvic adhesions, cyst walls were not completely resected.

On admission, the patient presented with abdominal pain that had been worsening for the previous 2 weeks. She characterized the pain as fullness, with severe sharp pain, causing her to be unable to stand upright or lay down comfortably. The pain had been present for months but was now worse with movement and causing significant interference with her daily function. She noted increasingly more abdominal distention with associated nausea and occasional vomiting. In addition, she was concerned that she was obtaining less volume of urine during her intermittent self-catheterizations.

Workup was initiated with aspiration of 3.5L of tea-colored fluid. Beta-2 transferrin level was obtained to rule out cerebrospinal fluid collection which returned negative. There were no malignant cells on cytology, and CA-125 was normal, making cancer of ovarian origin unlikely. Microbial cultures were also negative.

After the initial aspiration, symptoms improved but worsened several days later. She was having similar symptoms to her initial presentation and was having complaints of feeling bloated, having difficulty sleeping and walking due to the pain. Repeat imaging revealed recurrent large, loculated fluid collections in the lower abdomen and pelvis with new obstruction of the left ureter. Urology was consulted and recommended ureteral stent placement if clinical status and renal function worsened.

Repeat aspiration was performed 4 days later, removing 2 L of clear, slightly tea-colored fluid. However, her symptoms



Figure 1: A 29-year-old female who presented with pelvic pain. Computed tomography without contrast showing large pelvic fluid collections.

and the cystic lesions recurred. There was a high suspicion for bilateral peritoneal inclusion cysts as a diagnosis of exclusion. Gynecologic oncology and general surgery services reviewed prior records and imaging and decided operative intervention to be delayed given extensive surgical history and failed prior attempt at resection. Interventional radiology was consulted for drain placement and evaluation for possible sclerotherapy.

Interventional radiology performed sclerotherapy on 11/16/23 [Figure 2]. Two 10-French pigtail catheters were placed under CT guidance, with 50 cc of serous fluid removed from each side. Absolute ethanol was injected into each abdominopelvic cyst (7 cc on the right and 5 cc on the left). The patient had resolution of her pain and discomfort after the sclerotherapy procedure on post-operative day 1. There was continued output from the drains, so the patient was discharged with the drains in place. Repeat CT imaging showed significant improvement in pelvic cysts size. The drains were removed once the output from the drains had decreased to <10 cc/day.

Repeat CT imaging performed 3 weeks later showed the absence of previous fluid collections and a simple-appearing 4-cm right adnexal cyst [Figure 3]. She has followed up with gynecologic oncology and has had intermittent pelvic pain complaints. Her continued complaints of pelvic pain are suspected to be multifactorial in nature given her complex

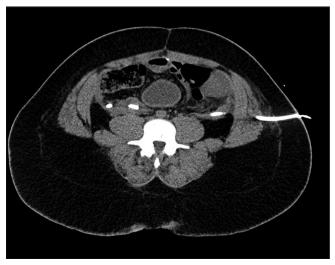


Figure 2: A 29-year-old female with suspected peritoneal inclusion cysts undergoing computed tomography (CT)-guided drainage by interventional radiology. CT during interventional radiologic sclerotherapy showing penetration of inclusion cyst.



Figure 3: A 29-year-old female status post-interventional radiologic (IR) drainage of peritoneal inclusion cysts. Computed tomography post-sclerotherapy showing reduction in cysts size.

history. Since her sclerotherapy, she has been treated for urinary tract infections and treated with pelvic floor physical therapy. Close surveillance with serial pelvic imaging has confirmed near complete resolution of abdominopelvic masses with the overall improvement of initial pain symptoms. This case describes a unique presentation of chronic pelvic pain and recurrent symptomatic abdominopelvic cystic lesions suspected to be bilateral peritoneal inclusion cysts which were successfully treated with image-guided sclerotherapy in the setting of a complex medical history.

DISCUSSION

This case report described the successful treatment of complex abdominal pelvic lesions suspected to be peritoneal inclusion cysts with image-guided drainage and ethanol sclerotherapy. Peritoneal inclusion cysts are benign, multiloculated cysts, containing fluid trapped between intraperitoneal adhesions. Pathologically, these are pseudocysts lined with hyperplastic, proliferating mesothelial cells within granulation tissue walls.^[4] Risk factors for peritoneal inclusion cysts include inflammation, endometriosis, prior abdominal surgery, trauma of the abdomen, radiation history, or inflammatory bowel disease.^[2,5] Due to trauma, inflammation, or surgical history, the peritoneum may have a decreased ability to resorb fluid. Fluids can then become trapped and produce a complex cystic mass. On imaging, these cysts can be mistaken for ovarian neoplasms which are why a malignancy risk assessment is recommended. Peritoneal inclusion cysts are classically described with a "spider in the web" pattern. These cysts are multiseptated, fluid-containing, are often adjacent to pelvic structures, and/or adherent to the ovary but do not involve the ovarian parenchyma. On CT, these cystic masses may have regular or irregular borders, which contain fluid or material. Magnetic resonance imaging (MRI) typically reveals low T1 signals and high T2 signals consistent with serous fluid.^[5] Fluid cytology is typically non-specific which aids to misdiagnosis as well.^[1]

Peritoneal inclusion cysts can be treated with expectant management, hormonal therapy, or image-guided aspiration. However, there is a high recurrence rate, up to 50%, for peritoneal inclusion cysts and often surgical excision is required.^[2] At present, medical and surgical options for treatment are utilized based on patient presentation, symptoms, and goals of care. For the patient mentioned in this case, the surgical excision attempt had been aborted due to inability for complete resection due to adhesions. With rapidly enlarging and recurrent bilateral pelvic cysts, a multidisciplinary team approach was utilized, and the patient underwent image-guided sclerotherapy.

Image-guided sclerotherapy has been used to treat ovarian endometriomas, reduce recurrence, and maintain fertility as compared to laparoscopy or laparotomy interventions.^[6,7] Sclerotherapy has also been used by interventional radiologists for other intra-abdominal and pelvic cystic lesions. A study of 29 patient's evaluated longterm outcomes for peritoneal inclusion cysts treated by sclerotherapy with only three recurrences noted, suggesting this to be a safe and effective treatment option. Image-guided sclerotherapy is performed after image-guided drainage. Most often, the drainage catheter is left in place for repeated sclerotherapy sessions, and to assess reduction in daily output from the cyst.^[8] Many sclerosing agents have been used, including doxycycline, betadine, and absolute ethanol. During a sclerotherapy session, contrast is injected into the cyst under fluoroscopy to exclude spillage and estimate the cyst volume.^[3,6,8] Roughly half the residual volume of the cyst is used as the volume of sclerosant. Sclerosant is left in situ with the drainage catheter capped for at least 1 h before uncapping the drain and aspirating the contents. Alcohol sclerotherapy works by causing protein denaturation, cytotoxic damage, and fibrosis. The goal of the procedure is to allow alcohol to contact the entire cyst wall without extravasation. The alcohol is aspirated to reduce pain and complications and the catheter is removed. Sclerotherapy is typically performed as an outpatient procedure and may involve conscious sedation.

CONCLUSION

Peritoneal inclusion cysts should be considered in the differential diagnosis for pelvic pain and large multicystic pelvic masses. Physicians should be familiar with characteristic imaging findings and risk factors for these benign cysts. Care is tailored to patient symptoms, goals, and appropriateness for surgery. For poor surgical candidates and those with recurrent cysts, such as the patient described in this case, sclerotherapy is an innovative, reasonable, and effective option. This case demonstrates a successful treatment of symptomatic peritoneal inclusion cysts by image-guided percutaneous drainage and sclerotherapy. After excluding other differential diagnoses and assessing malignancy risk to be low, physicians should be aware that for poor surgical candidates, sclerotherapy is an option for peritoneal inclusion cyst treatment.

Ethical approval

The Institutional Review Board approval is not required.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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