

Subserosal adenomyotic cysts and peritoneal inclusion cysts – Unusual differential diagnoses of multicystic pelvic masses: A review of two cases

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ABSTRACT

Background: Multiloculated pelvic cysts are commonly misdiagnosed as ovarian tumors or malignancies. We report 2 patients diagnosed with subserosal adenomyotic cysts and peritoneal inclusion cysts, mimicking multiloculated pelvic tumors. We discuss their clinical presentation, investigations, operation findings, and histopathology, present a literature review.

Cases: Case 1 was a 44-year-old patient with abnormal uterine bleeding. Imaging showed an enlarging multiloculated cystic structure over the right uterine wall. She underwent a diagnostic laparoscopy and right salpingo-oophorectomy. Intra-operatively, she was found to have multiple subserosal uterine cysts, diagnosed as adenomyotic cysts on histology.

Case 2 was a 50-year-old patient with history of laparoscopic cystectomy done 20 years ago. She was incidentally found to have a multiloculated cystic lesion in the pelvis. The lesion was located midline, anterior and superior to the uterus and bladder. She underwent a total abdominal hysterectomy, bilateral salpingo-oophorectomy, and bladder peritonectomy. Intra-operatively, multiple cystic lesions were noted over the anterior and fundus of uterus, bladder peritoneum, and pelvic side walls. The condition was confirmed to be peritoneal inclusion cysts on histology.

Conclusion: Subserosal adenomyotic cysts are a rare presentation of adenomyosis. They typically occur in premenopausal women. Treatment is usually by hormonal medications or surgical excision.

Many patients with peritoneal inclusion cysts have a history of peritoneal insults. Surgical excision is the most commonly described management as they often mimic malignancy. Both conditions are unusual presentations of multiloculated pelvic masses. A high recurrence rate is found, hence long-term follow-up with imaging is essential.

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1. Introduction

Large multiloculated pelvic cysts are easily misdiagnosed as ovarian tumors or malignancies. We report two patients presenting with similar finding of suspicious multiloculated pelvic cysts. One patient was diagnosed with adenomyotic cysts, and the other patient was diagnosed with peritoneal inclusion cysts.

Adenomyotic cysts and peritoneal inclusion cysts are uncommon conditions. Although adenomyosis is a common gynecologic condition, cystic adenomyosis is uncommon. Clinical symptoms are often nonspecific and may include dysmenorrhea, chronic pelvic pain and dysfunctional uterine bleeding. Literature is limited to few case reports and small cases series [1–7].

Peritoneal inclusion cysts are a rare entity, with fewer than 150 reported cases [8]. The pathophysiology of peritoneal inclusion cysts is thought to be reactive in origin [9]. Due to the rarity of this condition, it is often misdiagnosed as ovarian tumour or malignancy [10,11].

Surgery is useful for definitive histological diagnosis; however, patients have up to 50% risk of recurrence after surgery [12].

In our case report, we discuss the presentations, investigations, and histopathological findings of the two patients; we also present a brief literature review on these two rare conditions that may mimic malignancy.

2. Case 1

A 44-year-old nulliparous woman with a history of laparoscopic ovarian cystectomy for a benign cyst presented with abnormal uterine bleeding (AUB) and symptomatic anemia. Ultrasound imaging of the pelvis showed a 8.9 cm mass in the right adnexa containing cystic lesions measuring up to 3.3 cm, suggestive of an enlarged right ovary. A 1.6 cm endometrial polyp was also detected. She underwent a hysteroscopy, dilatation and curettage. A levonorgestrel-releasing intrauterine system (LNG-IUS) was inserted for treatment of AUB. Histology of the endometrial curetting showed non-atypical endometrial hyperplasia. Subsequent endometrial biopsies performed three months and nine months later showed resolution of the hyperplasia.

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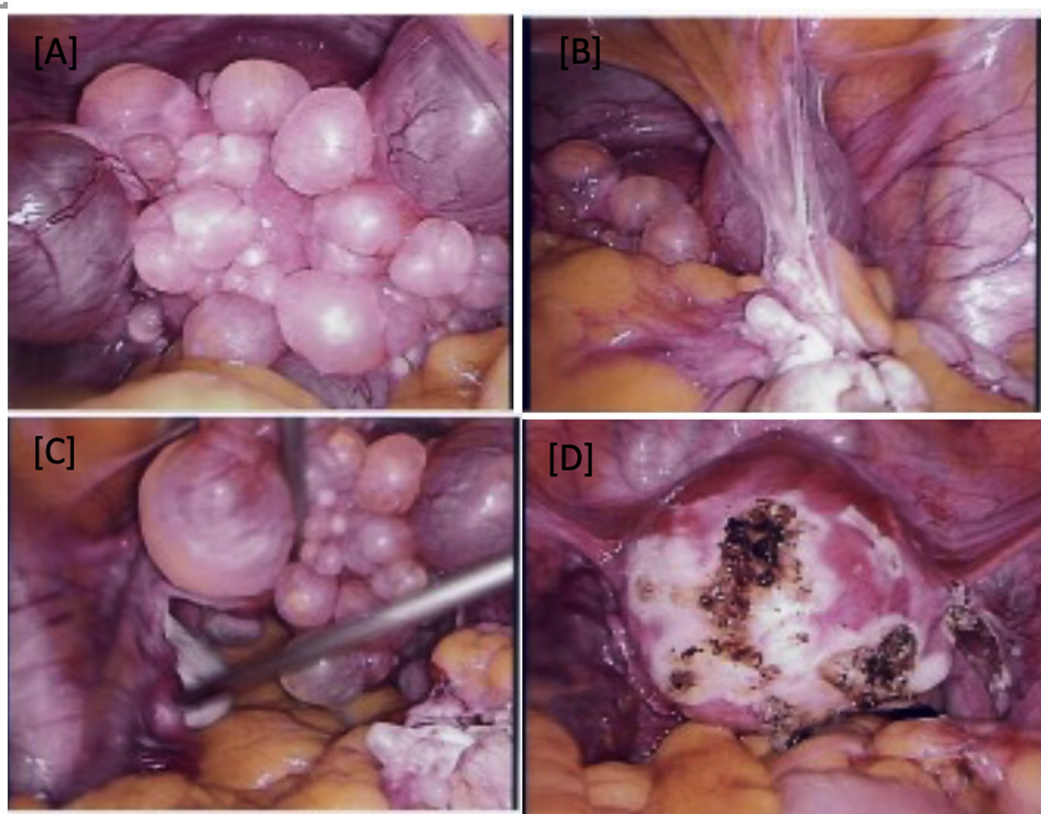


Fig. 1. Intraoperative images during laparoscopy showing [A] multiple subserosal cysts, [B] right tube and ovary buried in adhesion, [C] normal left tube and ovary, and [D] the uterus after excision of all the subserosal cysts.

A repeat ultrasound scan of the pelvis six months later showed a slight increase in the adnexal mass size, to 9.3 cm. The mass extended to the midline and there was vascularity seen at the periphery. Magnetic resonance imaging (MRI) of the pelvis showed a multiloculated cystic tubular structure draped over the right uterine wall. The right ovary appeared to be medial to the tubular cystic structure, with a radiological impression of a convoluted hydrosalpinx with underlying endometriosis. The CA 125 level was slightly elevated at 38.1kU/L. Carcinoembryonic antigen [CEA], alpha-fetoprotein [AFP] and beta-HCG [β hCG] were unremarkable.

She underwent a diagnostic laparoscopy. Intra-operatively, there were multiple subserosal cysts on the uterus (Fig. 1) and a 2 cm right

ovarian cyst that was adherent to the right tube and omentum. The left fallopian tube and ovary were grossly normal. Right salpingo-oophorectomy and biopsy of the subserosal cysts were performed. All the subserosal cysts were excised and cauterized intraoperatively.

The histopathology of uterine cysts showed benign adenomyotic cysts involving the uterine wall. These cysts were lined by endometrial glandular epithelium and there was a small collection of endometrial stroma seen adjacent to the glandular epithelium (Fig. 2). Histology of the right fallopian tube and ovary showed a benign endometriotic cyst.

An ultrasound pelvis done 6 months after the surgery revealed no recurrence of the adenomyotic cysts. She remained asymptomatic with light menstrual flow while on the LNG-IUS.

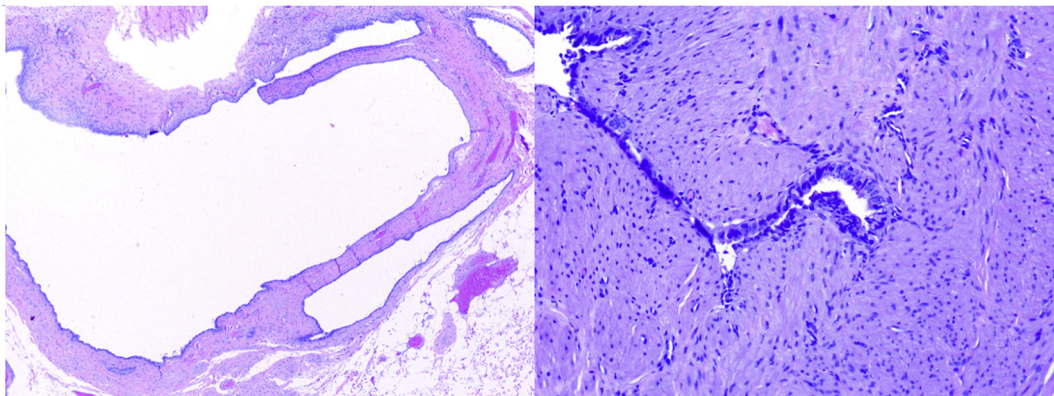


Fig. 2. Histology slides showing adenomyotic cyst wall lined by endometrial glandular epithelium with small adjacent collection of endometrial stromal tissues.

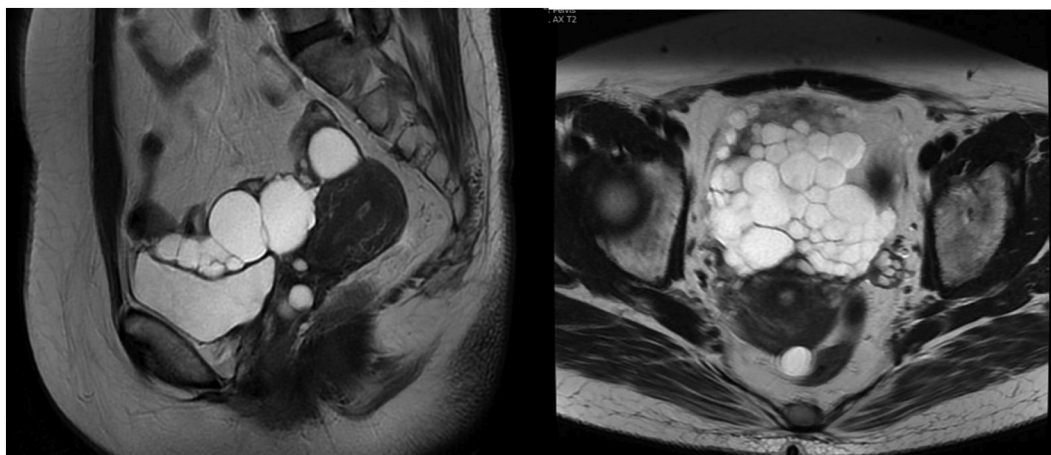


Fig. 3. Sagittal view (left) and axial view (right) on MRI scan showing multiloculated cystic lesions in the pelvis midline, anterior and superior to the uterus and bladder. There are intervening thin septations but no solid component noted.

3. Case 2

A 50-year-old nulliparous woman who had a history of subfertility and previous laparoscopic ovarian cystectomy for a benign cyst presented with oligomenorrhea. Ultrasound imaging of the pelvis showed a 8.7 cm by 5.3 cm multiloculated cystic mass anterior to the uterus and separate from both ovaries.

Ovarian tumour markers (CA 125, CA 19-9, CEA, AFP) were unremarkable. An MRI scan of the pelvis showed a multiloculated cystic lesion in the midline of the pelvis, anterior and superior to the uterus and bladder (Fig. 3). There were also cystic exophytic lesions seen on the posterior aspect of the uterus. The radiological impression was possible peritoneal spread of the cystic mass.

She underwent a laparotomy total hysterectomy, bilateral salpingo-oophorectomy and bladder peritonectomy. Intra-operatively, there were multiple cystic lesions seen over the anterior and fundus of the uterus, bladder peritoneum, and pelvic side walls (Fig. 4). The uterus, fallopian tubes, and ovaries were normal. Frozen section of the peritoneal cystic lesions showed cystic structures lined by bland columnar epithelium without overt malignancy seen.

An inadvertent cystotomy occurred during surgery. The urinary bladder was repaired, and she was discharged with an indwelling urinary catheter (IDC). A cystogram was performed two weeks later, which showed a normal urinary bladder contour without extravasation.

Final histology showed cyst wall lined by attenuated to low columnar-type epithelium, with underlying fibromuscular stroma (Fig. 5). No surface epithelial proliferation, cytological atypia or malignancy was seen. There were also no features of endometriosis or thyroid-type follicles to suggest stroma ovarii. The lining cells stained positive for calretinin on immunohistochemistry. These findings favoured a diagnosis of multi-locular benign peritoneal inclusion cyst.

An ultrasound pelvis done 1 year after the surgery revealed no recurrence of the cystic lesions.

4. Discussion

The patient in case 1 had a rare presentation of adenomyotic cyst, as most reported cases present as a singular myometrial cyst [1–3,5,6]. There was only one case reported in the literature on laparoscopic excision for a patient with symptomatic multicystic uterus [4]. Histopathologic analysis of the uterine cysts showed endometrioid cystadenomas without atypia. The findings are similar to the patient in case 1.

Uterine adenomyotic cysts are lined with endometrial epithelium and stroma [2]. They can be found within the myometrium, in the

submucosal or subserosal layer of the uterus. The main clinical feature of cystic adenomyosis is dysmenorrhea, but patients can also present with AUB, similar to our patient discussed in case 1. However, her AUB is more likely attributed to the underlying endometrial hyperplasia, which was successfully treated with LNG-IUS. Treatment approaches proposed include hormonal methods similar to the treatment of adenomyosis, or surgical excision of these cysts, which can relieve symptoms and exclude malignant transformation of adenomyosis. Even though uncommon, malignancy has been identified in a few case reports [13,14].

As the disease is more common among younger, pre-menopausal women, it is important to take into consideration the fertility wishes of these patients. Laparoscopic excision of the lesion can be done for intramural or subserosal lesions [15], while hysteroscopic resection can be

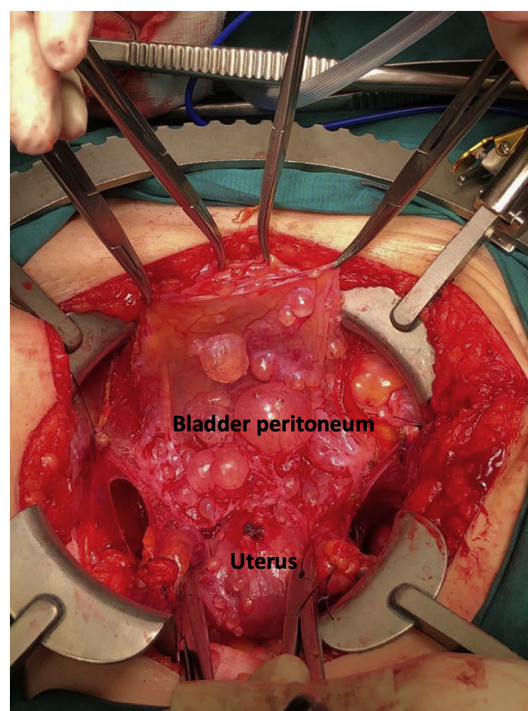


Fig. 4. multiple cystic lesions noted over the anterior surface and fundus of the uterus, as well as on the bladder peritoneum.

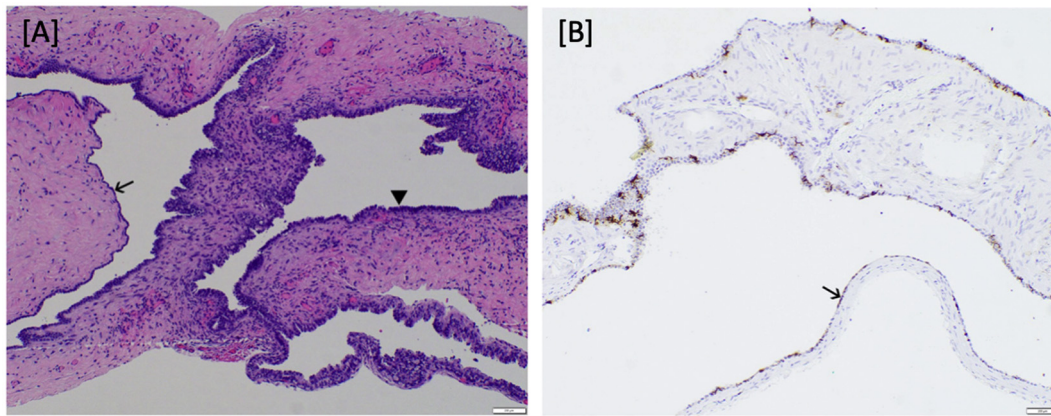


Fig. 5. Histology slides in case 2 showing [A] low-power view of paraffin sections of cystic structures (arrow head points to columnar epithelium and arrow points to flattened epithelium) & [B] immunohistochemical staining with calretinin (arrow points to an area of positive staining).

considered for submucosal lesions [7]. For patients who do not desire future fertility, a total hysterectomy can be considered. The patient in case 1 was pre-menopausal and nulliparous, and was not ready to lose her fertility. Consequently, a laparoscopic resection of adenomyotic cysts was done. The patient did not have a stable partner or active intention for pregnancy post-operatively. Therefore, the LNG-IUS was kept in situ for prevention of endometrial hyperplasia and treatment of AUB.

In view of the rarity of the adenomyotic cysts, there is currently inadequate data on the prognosis, influence on fertility, and the use of postoperative medication to prevent recurrence of the disease [4].

Peritoneal inclusion cysts, on the other hand, are benign aggregate masses of variable-sized, fluid-filled, mesothelial-lined cysts of the pelvis and abdomen. A history of insult to the peritoneum was found in 70.6% of cases [16]. As such, many authors believe the pathophysiology to be reactive in origin.

Peritoneal inclusion cysts are often a diagnostic challenge because the clinical findings and presentations can be varied. On radiological imaging, they may be hard to distinguish from an ovarian tumour. Therefore, surgical excision is the most commonly reported management option, often for histological diagnosis, as was with our patient in case 2.

Even with surgery, the overall recurrence rate for all surgical procedures is 28%, with unilateral oophorectomy with or without hysterectomy having the highest recurrence rate, of 50% [12]. Medical therapy may be useful in the treatment of peritoneal inclusion cysts. A case report in Japan describes successful treatment with combined hormonal pills for recurrent peritoneal cysts in two patients [11]. Considering the high morbidity of surgery and risk of recurrence, the goal for such a chronic disease should be symptomatic relief through individualization of treatment [12].

As both conditions are associated with recurrence after surgery, these two patients were scheduled for follow-up with interval pelvic ultrasound scans, which did not demonstrate any recurrence.

5. Conclusion

Adenomyotic cysts and peritoneal inclusion cysts can present as a suspicious multicystic pelvic mass that may mimic malignancy. Both conditions are unusual differential diagnoses of multicystic peritoneal masses. Detailed history taking and clinical correlation are important. MRI can be used to improve characterization. Increased awareness of these conditions among clinicians, radiologists and pathologists can allow for better counseling and management of these patients. In view of the high recurrence rate, long-term follow-up with imaging should be considered.

Contributors

All authors were involved in patient care and contributed to the preparation of this case report.

Declaration of Competing Interests

The authors declare that they have no conflict of interest regarding the publication of this case report.

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Patient Consent

Obtained.

Provenance and Peer Review

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