

Mesothelial cyst of uterine corpus misdiagnosed as leiomyoma

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To the Editor: A 46-year old woman (gravida 3, para 1) presented at West China Second Hospital with complaints of a space-occupying lesion in the uterus that, 2 years before, had been diagnosed as a uterine cyst at West China Second Hospital. On that previous occasion, B-ultrasonography revealed a cystic mass with a compartmentalized structure (10.3 × 5.4 × 8.8 cm) on the left side of the pelvic cavity, and laparoscopic exploration found a cystic mass on the posterior uterine wall, which was diagnosed as “leiomyoma with cystic degeneration” based on pathology analysis of flash-frozen surgical tissues. Laparoscopic “myomectomy” was performed, and pathology analysis of paraffin-embedded tissue confirmed “leiomyoma with degeneration.” Immunohistochemistry showed the lesion to be positive for caldesmon but negative for CD10. Rate of Ki67 positivity was less than 1%, indicating a benign lesion. The woman recovered well after the operation. Follow-up ultrasonography at 3 months after surgery identified a uterine cystic mass of 3.0 × 2.5 × 1.7 cm.

At the present admission, 2 years after surgery, ultrasonography indicated that the mass measured 6.2 × 4.7 × 9.3 cm. The patient reported no abdominal pain or distention. Both menstrual cycle and blood volume were regular. Bi-manual examination revealed nothing remarkable except an enlarged palpable uterus, which was equivalent to what might be expected for a 4-month pregnancy. Laboratory tests showed mild anemia (hemoglobin, 91 g/L) and negative results for cancer antigens 125 and 19-9. Given this unusual situation, we consulted senior pathologists, who reviewed the paraffin-embedded sections from 2 years previously and concluded that they were “leiomyoma combined with mesothelial cyst,” not “leiomyoma with degeneration” as originally determined. A second operation involving hysterectomy was suggested, which the patient accepted. Laparoscopy under general anesthesia revealed an enlarged, irregularly shaped uterus. Total hysterectomy and bilateral salpingectomy were performed, and all specimens were checked carefully. The right myometrium of the uterus contained a thin-walled, 9-cm cyst with clear liquid inside [Figure 1A]. Analysis of paraffin-embedded sections from this repeat surgery showed

a single-layer cystic structure, lined with flat mesothelial cells, on the myometrial wall [Figure 1B and 1C]. The revised pathology classification of “mesothelial cyst of uterine corpus” was confirmed by positive immunostaining for the mesothelial markers calretinin [Figure 1D], mesothelin [Figure 1E], and cytokeratin 5/6 [Figure 1F]. The patient recovered well after the operation, and hemoglobin was normal at 1 month later. Follow-up at 3 months after surgery did not reveal any abnormal signs or complaints.

Mesothelial cysts appear as single or multiple, thin-walled inclusion cysts derived from benign mesothelioma.^[1] Such cysts can occur at any abdominal peritoneal surface, such as the round ligament, mesentery, and peritoneum.^[2] Uterine mesothelial cysts are rare. A search of PubMed entries since 1985 failed to identify a single case of mesothelial cyst in the uterine myometrium. Instead, we identified seven reports describing 19 patients with mesothelial cysts on the round ligament, 17 of whom were women of reproductive age.^[3-7]

What triggers growth of mesothelial cysts is unknown, although developmental disorder is generally suspected. Past abdominal surgery, pelvic inflammation, or endometriosis may be associated with mesothelial cysts of the round ligament.^[2] Some studies have explored a potential relationship between sex hormones and mesothelial cysts, but this remains controversial, especially given that mesothelial cysts on round ligaments do not immunostain for estrogen or progesterone.^[4]

Mesothelial cysts share similar appearance and histopathology characteristics with benign cystic mesothelioma.^[5] Both conditions can present as inclusion cysts in the pelvic cavity. Mesothelial cysts are usually solitary and benign, and they contain 1 to 3 cavities; the flat mesothelial tissue on the cyst wall is well-differentiated.^[1] Benign cystic mesothelioma, in contrast, often presents as multi-cavity cysts, which are considered to be a reactive tumor or a neoplastic tumor with negligible potential for recurrence and malignancy.^[1,2] If a patient is asymptomatic, serial ultrasonography can be used to assess cyst size and thereby monitor change in benign cystic mesothelioma. Surgical excision is the definitive treatment.^[2]

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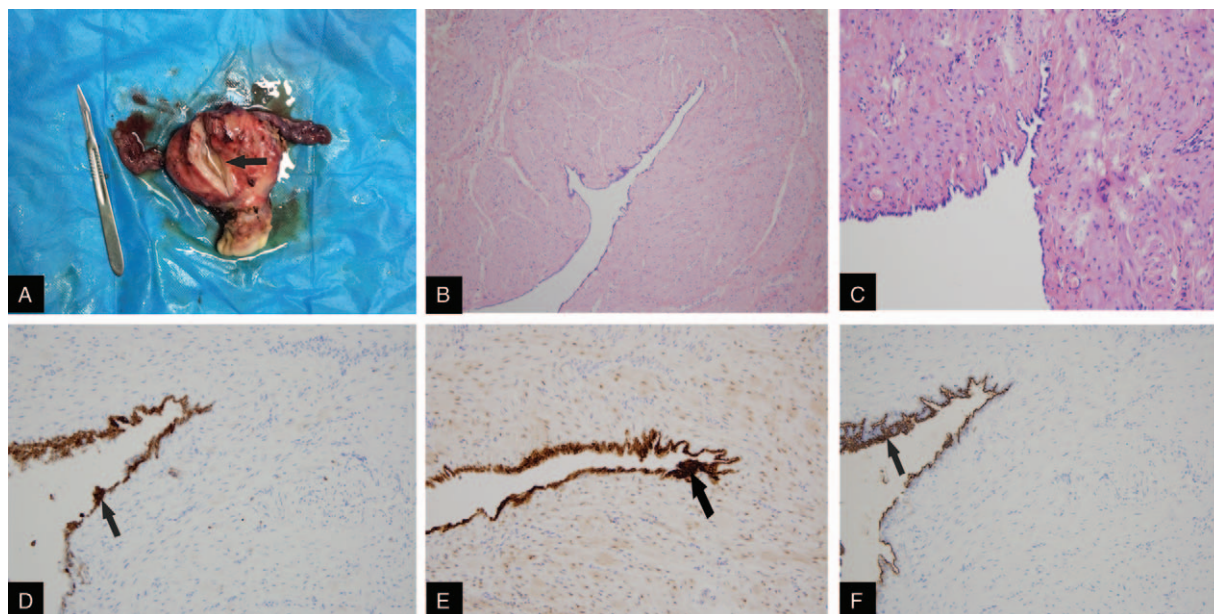


Figure 1: Mesothelial cyst in the uterine corpus. (A) Photograph of the mesothelial cyst (arrow) following hysterectomy. (B) Cyst with the myometrium resembles a fissure. Tissue was stained with hematoxylin-eosin (original magnification, $\times 100$). (C) Monolayer epithelial cells cover the cyst wall. Tissue was stained with hematoxylin-eosin (original magnification, $\times 200$). (D) Cyst-lining cells positive for calretinin immunohistochemical staining (arrow; original magnification, $\times 200$). (E) Cyst-lining cells positive for mesothelin immunohistochemical staining (arrow; original magnification, $\times 200$). (F) Cyst-lining cells positive for cytokeratin 5/6 immunohistochemical staining (arrow; original magnification, $\times 200$).

Uterine mesothelial cysts are not associated with specific clinical manifestations. Some patients may have a palpable abdominal mass or experience lower abdominal discomfort. Menorrhagia may also be detected in some women. Ultrasonography may reveal a cyst in the uterus. Diagnosis of uterine mesothelial cysts is particularly challenging because of the similarities between leiomyomas with cystic degeneration and congenital uterine cysts. Indeed, our patient was misdiagnosed as having leiomyoma with cystic degeneration after the first operation. Only after consultation with senior pathologists was the diagnosis revised to “leiomyoma combined with mesothelial cyst.” The typical microscopic appearance of a single layer of cuboidal cells lining the cyst wall and positive staining for specific markers such as calretinin may help diagnose mesothelial cysts in the pelvic cavity.^[2,5] Our case highlighted the need to be aware of the possibility of uterine mesothelial cysts in women of reproductive age.

No definitive treatment has been established for uterine mesothelial cysts, reflecting their rarity. Ultrasonography-guided aspiration of mesothelial cysts of round ligament can offer temporary relief from symptoms, but it leads quickly to fluid re-accumulation and symptom recurrence.^[5] Cyst removal may be an option, but the thinness and multi-cystic appearance of cyst walls make complete removal challenging, which leads to a high recurrence rate. The best way to eliminate lesion recurrence, at least in older women and women without childbearing plans, is total or partial hysterectomy.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient has given her

consent for her images and other clinical information to be reported in the journal. The patient understands that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflicts of interest

None.

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