

# Uterus-like mass in the right broad ligament

## A case report

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### Abstract

**Rationale:** Uterus-like masses (ULMs) are rare benign lesions that resemble the uterus.

**Patient concerns:** Here, we describe the case of a woman with a ULM in the right broad ligament. A 51-year-old woman with a 2-month history of irregular vaginal bleeding was found to have a mass in the right broad ligament. Imaging studies revealed a solid-cystic lesion, suggestive of an endometrial cyst with malignant transformation.

**Interventions:** She underwent prompt surgery for the removal of the mass. Intraoperatively, the uterus and ovaries appeared normal, and an 8-cm-long mass was observed in the right broad ligament without any connection to the uterus or ovaries. The mass was successfully excised.

**Diagnoses:** Postoperative histopathological examination showed that the cystic mass was filled with a blackish-brownish fluid and that it had thick walls resembling the uterine myometrium. The cyst center was lined by endometrial glands that were positive for cytokeratin as well as estrogen and progesterone receptors, and by stromal cells that were positive for CD10.

**Outcomes:** The patient recovered well and has had no further symptoms during 2 years of follow-up.

**Lessons:** We have reported a case of ULM in the right broad ligament in a Chinese woman. Although ULMs are rare, they should be considered in the differential diagnosis for pelvic masses.

**Abbreviations:** MRI = magnetic resonance imaging, ULMs = uterus-like masses.

**Keywords:** broad ligament, uterus, uterus-like mass

## 1. Introduction

Uterus-like masses (ULMs) are rare benign growths that morphologically resemble the uterus. ULMs are characterized by a central cavity lined with endometrial glands and stromal cells, and thick walls composed of smooth muscle cells. ULMs were first described by Cozzutto et al in 1981,<sup>[1]</sup> and to date, only about 30 ULMs have been described in the scientific literature. ULMs may originate from various organs and tissues, such as the ovaries,<sup>[2]</sup> uterine ligaments,<sup>[3]</sup> uterus,<sup>[4]</sup> pelvic wall,<sup>[5]</sup> obturator lymph nodes,<sup>[6]</sup> conus medullaris,<sup>[7]</sup> inguinal soft tissues,<sup>[8]</sup> vagina,<sup>[9]</sup> and colon.<sup>[10]</sup> Herein, we describe a ULM in a 51-year-old woman with irregular vaginal bleeding. We also review other cases of ULMs in the literature.

## 2. Case presentation

A 51-year-old postmenopausal Chinese woman (gravida 3, para 1, labor 1, abortion 1) with a 2-month history of irregular vaginal bleeding was found to have a pelvic mass. She was therefore referred to our hospital. A gynecological examination revealed normal vulvar development and an unobstructed vagina. The uterus was anteverted and of a normal size. A well-defined, movable, painless mass was palpated in the right uterine adnexa, posterior to the uterus. The mass measured 7 cm × 5 cm × 5 cm. There was no obvious abnormality in the left uterine appendages. Ultrasonography performed in our hospital revealed an 8-cm-long cystic mass on the right side of the pelvis. Magnetic resonance imaging (MRI) showed an irregular solid-cystic lesion measuring 74 mm × 44 mm × 38 mm on the right side of the uterus (Fig. 1). On both T1 and T2 images, we observed smooth thickening of the cyst walls, which appeared hypointense, similar to the uterine myometrium. In contrast, the cystic spaces appeared hyperintense on both T1 and T2 images. On the basis of these findings, we considered that the lesion was an endometrial cyst.

As we suspected that the lesion had undergone malignant transformation, we decided to promptly excise the lesion. The patient underwent laparoscopic surgery under general anesthesia. During the surgery, however, the uterus and ovaries were found to be normal, and an oval mass was observed in the right broad ligament. The mass was not connected to the uterus and was successfully removed. The patient was followed up for 2 years, and she is alive and well.

This study was approved by the Ethics Committee of the Affiliated Hospital of Shandong First Medical University. All procedures performed in the study were in accordance with the ethical standards of the institutional research committee and with the

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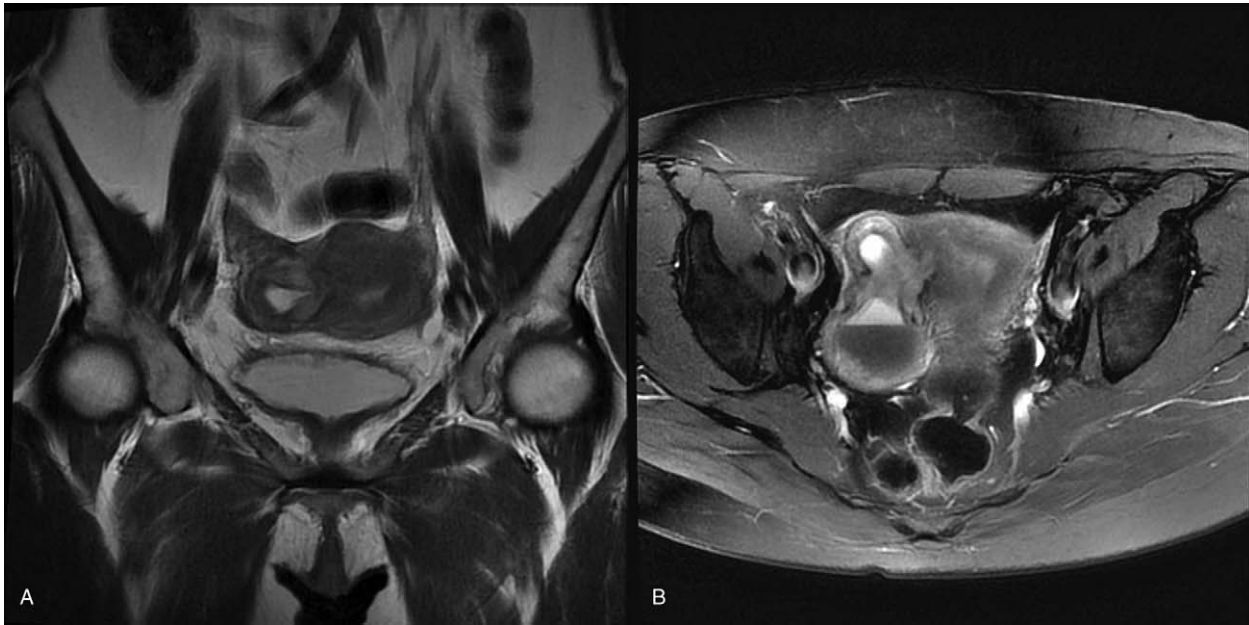
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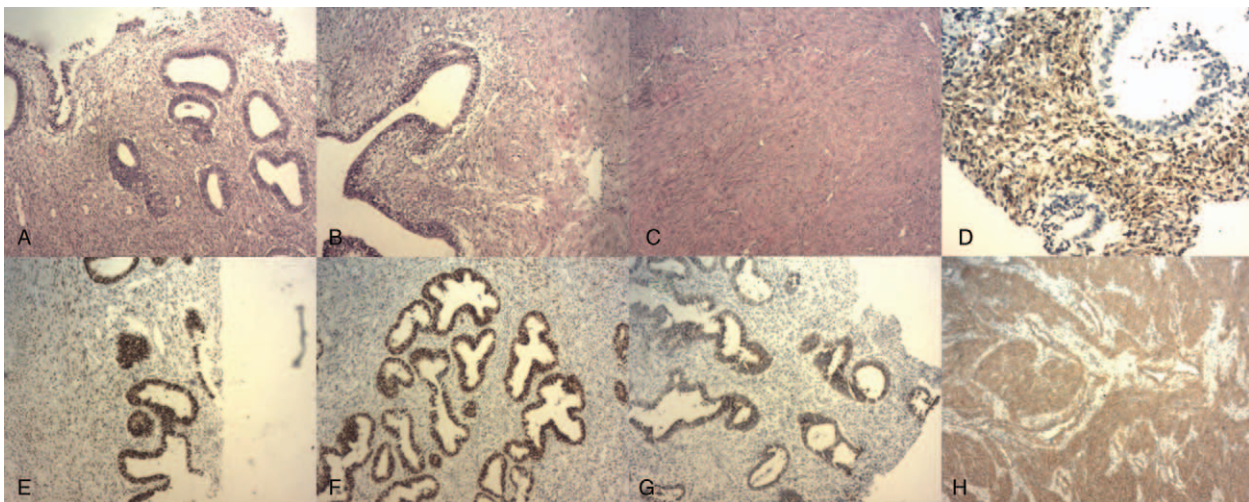
**Figure 1.** (A) A coronary T2-weighted image and (B) an axial T2-weighted image of the uterus of the patient in this case report. An abnormal lesion is seen to the right of the uterus. Its periphery appears solid and exhibits high signal intensity on both T1- and T2-weighted images. Low-intensity areas and fluid accumulation are present within the lesion. The lesion measures 7.4 cm × 4.4 cm × 3.8 cm. The boundary between the lesion and the right ovary is not well-defined.

1964 *Helsinki Declaration* and its later amendments. Written informed consent was obtained from the patient for the publication of this case report and the accompanying images.

### 3. Pathological findings

The resected mass was fragmented and measured 6 cm × 6 cm × 5 cm, in total. On gross examination, the cystic part of the mass was found to be filled with a blackish-brownish fluid and have thick walls resembling the uterine myometrium. No obvious sign

of malignancy was found on gross examination. Light microscopic examination of the tissue specimen after staining with hematoxylin and eosin revealed that the cyst center was lined by endometrial glands and stromal cells, and the cyst walls were composed of smooth muscle cells. On immunohistochemical examination, the glandular epithelium was positive for cytokeratin as well as estrogen and progesterone receptors, while the smooth muscle cells were positive for smooth muscle actin. The stromal cells were positive for CD10, which confirmed that they were endometrial stromal cells (Fig. 2).



**Figure 2.** Histopathological and immunohistochemical examination of the resected specimen. (A–C) On histological examination, the cystic mass was found to have a central cavity lined with endometrial glands and stromal cells as well as thick walls composed of smooth muscle cells (hematoxylin and eosin staining; magnification, ×100). (D) The stromal cells surrounding the endometrial glands as well as the endometrial glands were positive for CD10 (magnification, ×400). (E) The endometrial glands were positive for estrogen receptor (magnification, ×100) and (F) progesterone receptor (magnification, ×100). (G) The glandular epithelium was positive for cytokeratin (magnification, ×100). (H) The surrounding smooth muscle cells were positive for smooth muscle actin (magnification, ×400).

#### 4. Discussion and conclusions

We have described a case of ULM in a postmenopausal woman who was initially suspected to have an endometrial cyst. The lesion was successfully resected, and the diagnosis of ULM was confirmed on pathological examination. The patient recovered well after the surgery, and she has had no further symptoms during 2 years of follow-up. ULM is a rare benign lesion that is usually misdiagnosed as another tumorous or cystic lesion. ULMs have been reported in patients ranging in age from 11 to 59 years, though most patients are in their 40s or 50s.<sup>[11]</sup> ULMs can occur within the pelvic cavity as well as at extrapelvic sites. The most common site is the ovary, followed by the uterus, the uterine ligaments, and the pelvic wall.<sup>[11]</sup> The most common symptom at presentation is abdominal pain, which is often associated with menstrual symptoms; one-third of patients also have endometriosis or adenomyosis.<sup>[11]</sup>

The histogenesis of ULMs is unclear, but 3 hypotheses have been suggested: the congenital anomaly theory, the heterotopia theory, and the metaplasia theory. The congenital anomaly theory was suggested by Rosai, who postulated that ULMs result from the malformation of the Müllerian duct system. This theory is supported by several case reports showing the coexistence of developmental abnormalities in the gastrointestinal tract and the urinary tract in ULM patients.<sup>[12]</sup> The heterotopia theory, proposed by Peterson et al, states that ULM results from heterotopic Müllerian duct tissue; the authors based this theory on their case report of an ileal ULM patient with multiple anomalies.<sup>[13]</sup> Cozzutto et al put forward the metaplasia theory, which states that ULMs originate due to the metaplasia of ovarian stromal cells into smooth muscle cells.<sup>[1]</sup> Subsequent reports of ULMs originating in the female genital tract support the metaplastic transformation theory.<sup>[6]</sup> In our patient, no other coexisting malformations or rudimentary Müllerian structures were found. Furthermore, our patient had no history of pelvic masses. These findings do not support the theory of malformation or heterotopias. Most ULMs have a good prognosis and can be cured by surgical excision.<sup>[10]</sup> To date, only 2 cases of a malignant tumor originating from a ULM have been reported: a case of endometrioid carcinoma<sup>[14]</sup> and a case of clear cell carcinoma.<sup>[15]</sup> Thus, ULMs, like endometrial cysts, may undergo malignant transformation. Serial ultrasonography and MRI may enable the early detection of malignant transformation in ULMs. Surgery is indicated in symptomatic patients, those suspected to have malignant transformation, and those with large masses.

In summary, we have reported a case of ULM in the right broad ligament in a Chinese woman. Although ULMs are rare, they should be considered in the differential diagnosis for pelvic masses. As the ULM in our patient was not associated with other identifiable malformations, this case provides new evidence that supports the metaplasia theory of ULM pathogenesis.

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#### References

- [1] Cozzutto C. Uterus-like mass replacing ovary: report of a new entity. *Arch Pathol Lab Med* 1981;105:508–11.
- [2] Pai SA, Desai SB, Borges AM. Uteruslike masses of the ovary associated with breast cancer and raised serum CA 125. *Am J Surg Pathol* 1998; 22:333–7.
- [3] Matsuzaki S, Murakami T, Sato S, et al. Endomyometriosis arising in the uterosacral ligament: a case report including a literature review and immunohistochemical analysis. *Pathol Int* 2000;50:493–6.
- [4] Tijani el H, Meryem T, Lamy GA, et al. Giant uterus-like mass of the uterus. *Indian J Pathol Microbiol* 2010;53:793–5.
- [5] Carinelli S, Motta F, Frontino G, et al. Multiple extrauterine adenomyomas and uterus-like masses: case reports and review of the literature. *Fertil Steril* 2009;91:1956e1959-1911.
- [6] Rohlfing MB, Kao KJ, Woodard BH. Endomyometriosis: possible association with leiomyomatosis disseminata and endometriosis. *Arch Pathol Lab Med* 1981;105:556–7.
- [7] Sharma MC, Sarkar C, Jain D, et al. Uterus-like mass of Müllerian origin in the lumbosacral region causing cord tethering. Report of two cases. *J Neurosurg Spine* 2007;6:73–6.
- [8] Seki A, Maeshima A, Nakagawa H, et al. A subserosal uterus-like mass presenting after a sliding hernia of the ovary and endometriosis: a rare entity with a discussion of the histogenesis. *Fertil Steril* 2011;95: 1788e1715-1789.
- [9] Redman R, Wilkinson EJ, Massoll NA. Uterine-like mass with features of an extrauterine adenomyoma presenting 22 years after total abdominal hysterectomy-bilateral salpingo-oophorectomy: a case report and review of the literature. *Arch Pathol Lab Med* 2005;129:1041–3.
- [10] Na KY, Kim GY, Won KY, et al. Extrapelvic uterus-like masses presenting as colonic submucosal tumor: a case study and review of literature. *Korean J Pathol* 2013;47:177–81.
- [11] He J, Xu J, Zhou HY. Uterus-like mass: a very rare and elusive entity a case report. *Medicine (Baltimore)* 2016;95:e4961.
- [12] Rosai J. Uteruslike mass replacing ovary. *Arch Pathol Lab Med* 1982;106:364–5.
- [13] Peterson CJ, Strickler JG, Gonzalez R, et al. Uterus-like mass of the small intestine. Heterotopia or monodermal teratoma? *Am J Surg Pathol* 1990;14:390–4.
- [14] Rahilly MA, al-Nafussi A. Uterus-like mass of the ovary associated with endometrioid carcinoma. *Histopathology* 1991;18:549–51.
- [15] Nakakita B, Abiko K, Mikami Y, et al. Clear cell carcinoma arising from a uterus-like mass. *Pathol Int* 2014;64:576–80.