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The clinical challenge of a uterine cotyledonoid dissecting leiomyoma with adenomyosis: A case report

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ABSTRACT

Cotyledonoid dissecting leiomyoma (CDL) is a rare uterine tumor with unique clinical and histological features. We present a case of a 46-year-old woman with a 3-month history of left-flank pain radiating to the back. The patient had a history of infertility and a previous miscarriage. Ultrasound revealed a solid tissue mass suggestive of a degenerated fibroid. Laparoscopy identified subserosal leiomyoma and leiomyoma in the broad ligament. Histologically, CDL is characterized by disorganized smooth muscle with hyaline degeneration and no evidence of malignancy. Clinically, CDL can present with a variety of symptoms, including heavy menstrual bleeding, pelvic pain, and infertility. The coexistence of CDL and adenomyosis is exceedingly rare. This case highlights the importance of considering CDL in the differential diagnosis of pelvic mass, malignant neoplasms, and infertility, even with atypical symptoms. It also emphasizes the value of cooperation between clinicians and pathologists for accurate diagnosis and management of CDL. Adenomyosis in this case further complicated the diagnosis and highlighted the need for an index of suspicion for this rare condition.

1. Introduction

Uterine leiomyomas or fibroids are common benign smooth muscle tumors [1]. They are diagnosed in approximately 70% of white women and 80% of black women [2]. These tumors may be asymptomatic or may cause a group of symptoms. The most typical symptom is heavy menstrual bleeding. Other symptoms include non-cyclical pain, abdominal distension, constipation, and urinary retention. Leiomyomas may also be associated with infertility [1]. Cotyledonoid dissecting leiomyomas (CDLs), also named Sternberg tumors, are a rare variant with unusual growth characteristics [3]. The term "cotyledon" is used to describe both mammalian placenta lobules and the leaves developed by embryos of seed plants. It comes from the Greek *kotulēdṓn* ("cup-shaped cavity") [3]. Macroscopically, CDLs resemble placental tissue [4–9]. Endometriosis and adenomyosis have been reported in women with leiomyomas. This report describes a woman with both CDL and adenomyosis. To our knowledge, this is the second case report of CDL coexisting with adenomyosis [6].

2. Case Presentation

A 46-year-old woman, gravida 0 para 0, presented with a 3-month history of left-flank pain radiating to the back. The pain had increased in the last month and did not resolve with analgesics. The patient reported regular menstrual cycles. She had a history of infertility and one previous in vitro fertilization (IVF) pregnancy that resulted in a miscarriage. She also had a history of ovarian cysts that had been treated with norethisterone without complete resolution.

The abdominal examination was normal, and no palpable masses were noted. The CA-125 level was 29.4 and her blood count was within normal limits. Ultrasound of the uterus (Fig. 1) showed a solid tissue mass measuring 5.4 cm × 5.2 cm seen at the posterior aspect of the uterus, indicative of a degenerated fibroid tumor. Subsequent diagnostic laparoscopy showed an irregular uterine wall with subserosal leiomyomas, severe distension of the left fallopian tube, and a leiomyoma in the broad ligament, prompting exploratory laparotomy.

Intraoperatively, a leiomyoma measuring 3 cm × 3 cm with cystic formation extended from the uterine wall to the broad ligament. It was

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excised, preserving the left adnexa, and the specimen was sent for pathological examination. The excision of the leiomyoma within the broad ligament likely alleviated the source of pain, leading to a positive outcome following the surgical intervention.

The macroscopic findings (Fig. 2) showed many pieces measuring 5. 9 cm × 5.6 cm × 2.6 cm, irregular in shape, firm in consistency, tan in color, and cut section revealing numerous hemorrhagic cysts. Histological sections (Fig. 3 a, b, c, d) showed multiple nodules consisting of disturbed swirls of benign smooth muscle cells with marked hyaline degeneration and hydropic changes, without atypia or atypical mitotic figures. Some nodules showed cystic structures lined by cuboidal to columnar epithelium with foci of siderophages and hemorrhage, surrounded by endometrial stroma.

At six-month follow-up, the patient mentioned a significant improvement in her symptoms. Her left-flank pain had resolved completely. She had not experienced any recurrence of pain postoperatively or other abdominal complaints. Her menstrual cycle remained regular. Since she had a history of infertility, it was important to continue monitoring and follow-up, to assess prognosis and risk of recurrence.

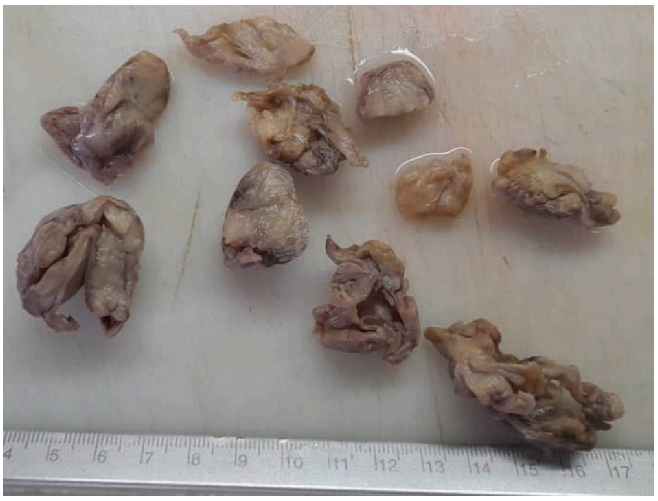

Fig. 2. Many pieces measuring around 5. 9 cm × 5.6 cm × 2.6 cm, irregular in shape, firm in consistency, tan in color, and cut section revealing numerous hemorrhagic cysts.

3. Discussion

CDLs commonly present as pelvic masses and with abnormal uterine bleeding. To our knowledge, this is the second reported case of CDL coexisting with adenomyosis in the English medical literature. The first case of CDL with adenomyosis was published in 2016. The 40-year-old parous woman presented with a history of heavy menstrual bleeding, anemia and an enlarging pelvic mass [6]. The pelvic mass had been diagnosed 6 years previously as a leiomyoma. The patient underwent hysterectomy. Our case report describes a 46-year-old nulliparous woman with a history of infertility. She therefore underwent fertility-conserving surgery. CDLs are considered to be benign and thus recognition of this unusual condition is required to avoid unnecessary intervention.

4. Conclusion

In summary, CDL of the uterus is a sporadic benign tumor. Recognizing this type of tumor is crucial for avoiding needless surgical interventions that could occur if it were mistakenly identified as a malignant neoplasm.

Contributors

Mohamad Moafak Hariri contributed to drafting the manuscript and revising the article critically for important intellectual content.

Mohamad Ali Farho contributed to drafting the manuscript and revising the article critically for important intellectual content.

Alaa Kourdy contributed to drafting the manuscript and revising the article critically for important intellectual content.

Hiba Allah AlHammoud contributed to patient care, conception of the case report, acquiring the data, and drafting the manuscript.

Kawthar Alawad contributed to patient care, acquiring and interpreting the data, and drafting the manuscript.

Lina ghabreau contributed to patient care, conception of the case report, acquiring and interpreting the data, drafting the manuscript, and

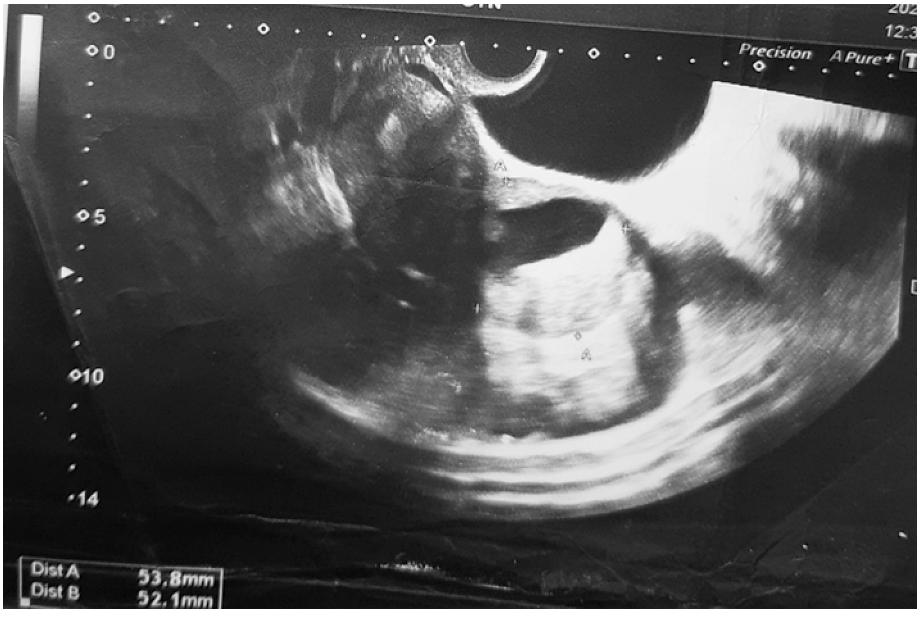

Fig. 1. The uterus is enlarged with a solid tissue mass measuring 5.4 cm × 5.2 cm seen at the posterior aspect of the uterus indicative of a degenerated fibroid tumor. The mass is not adherent to adjacent structures and does not infiltrate the cervix (compressing the cervical canal), and no free fluid is visualized behind the uterus.

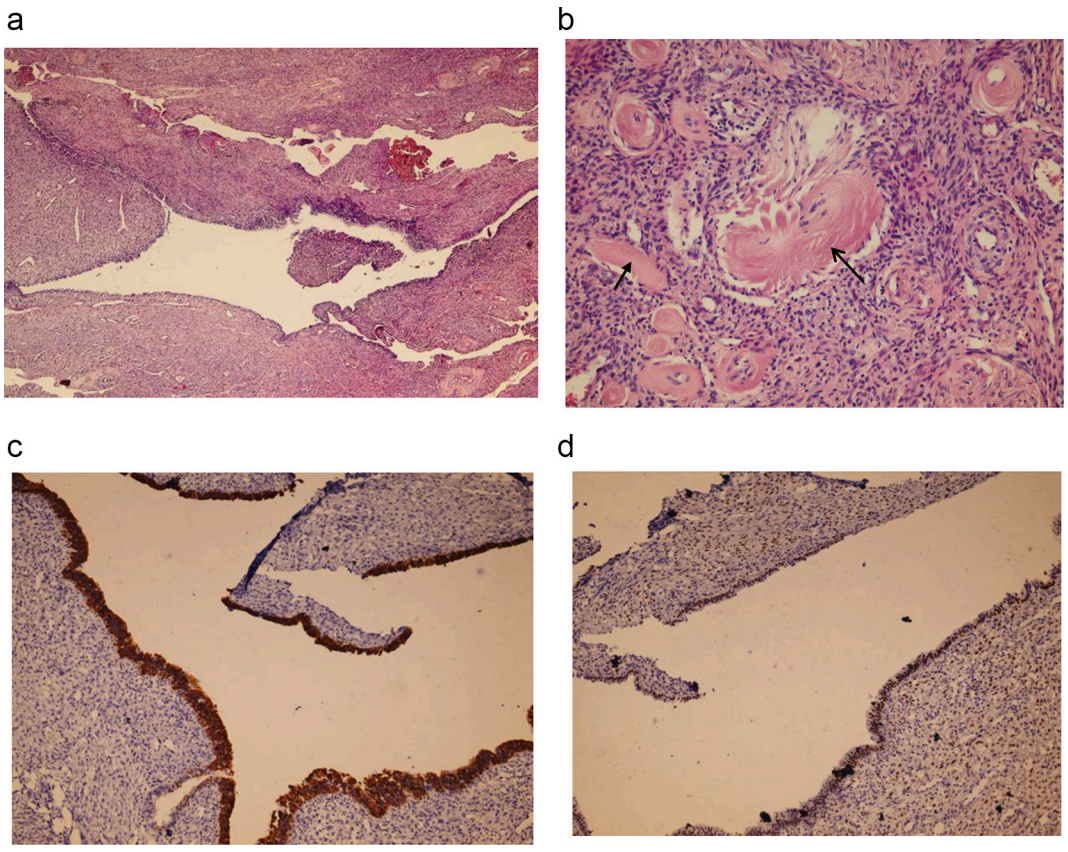

Fig. 3. a. H&E, magnification X200. The sections display multiple nodules consisting of disturbed swirls of benign smooth muscle cells with marked hyalinized degeneration and hydropic changes, without atypia or atypical mitotic figures. Presence of cystic structures lined by cubodial to columnar epithelium with foci of siderophages and hemorrhage, surrounded by endometrial stroma.
b. H&E, magnification X400. Hyalinized degeneration.
c. CK7 immunohistochemical stain magnification X400. Positive CK7 membranous immunostaining is observed in the endometrial epithelium of the cysts.
d. ER immunohistochemical stain magnification X400. Positive ER nuclear immunostaining is observed in the endometrial glands and stroma.

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Patient consent

Written informed consent was obtained from the patient for publication of the case report and accompanying images.

Provenance and peer review

This article was not commissioned and was peer reviewed.

Conflict of interest statement

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

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