# CASE REPORT Open Access



# Coexistence of cutaneous endometriosis and ovarian endometrioma: a case report

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#### **Abstract**

**Background:** Umbilical endometriosis is a rare entity accounting for 0.5–4% of cases with endometriosis.

**Case presentation:** Here we report a rare case of umbilical endometriosis with concurrent ovarian endometriomas in a 37-year old primiparous Iranian woman.

**Conclusion:** This interesting coexistence reflects the importance of thorough gynecological assessment in patients with cutaneous endometriosis to enable appropriate management.

Keywords: Cutaneous endometriosis, Umbilical endometriosis, Endometrioma, Ovarian cyst

#### Introduction

Endometriosis is a common gynecologic condition characterized by the presence of endometrial tissue in anatomical sites other than the uterus and can lead to chronic pelvic pain or even infertility in women. The most common sites of involvement are ovaries followed by the Douglas pouch and pelvic ligaments, respectively [1]. It can be rarely found in other organs such as skin. Umbilical endometriosis is an unusual condition accounting for 0.5–1% of extra pelvic endometriosis [2]. Here we report a case of umbilical endometriosis with concurrent ovarian endometriomas.

### **Case presentation**

A 37-year old primiparous Iranian woman presented to our dermatology clinic with a complaint of an asymptomatic lesion in the umbilicus. The lesion appeared 1.5 years ago and had slightly increased in size. She denied previous piercing or trauma. However, she had had a laparoscopic cholecystectomy with concurrent ovarian

dermoid cystectomy about 10 years ago. She had regular menstrual cycles with normal flow.

Physical examination revealed a well-circumscribed.

Physical examination revealed a well-circumscribed, firm, bilobulated  $20 \times 22 \text{ mm}^2$ , nontender nodule on her umbilicus (Fig. 1a). An incisional biopsy was performed with differential diagnosis of adnexal tumor, umbilical granuloma, and metastasis, but surprisingly, histopathologic assessment showed the presence of a few dilated glands with stratified columnar epithelium in secretory phase (Fig. 2). The glands were surrounded by hypercellular stroma. Immunohistochemistry (IHC) staining showed positivity for estrogen receptor in the nuclei of epithelial cells lining glandular structure endometrial-like cells, CD10 diffuse and intense positivity in the stroma, and Ki67 positivity below 1% of epithelial cell nuclei (Fig. 3). These findings confirmed the diagnosis of cutaneous endometriosis. No signs of atypia or malignancy were observed. After consultation with gynecology service, abdominopelvic sonography was conducted, which showed an increased density at the umbilicus and also multiple ovarian cysts with appearance compatible with endometrioma (two  $15 \times 10 \text{ mm}^2$  cysts in right ovary and a  $35 \times 48 \text{ mm}^2 \text{ cyst in left one}$ ).

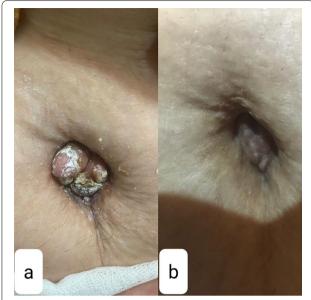
After establishing the diagnosis, total umbilectomy was suggested. However, due to the patient's phobia regarding that, just the nodule was excised

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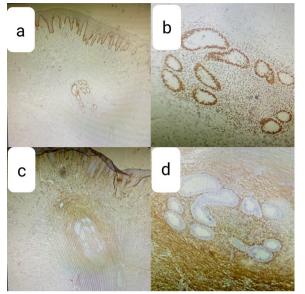


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**Fig. 1** A bilubolated umbilical mass, before (**a**) and after (**b**) treatment



**Fig. 3** Immunohistochemistry staining for estrogen receptor ( $\times$ 40,  $\times$ 100) (**a**, **b**) and CD10 ( $\times$ 40,  $\times$ 100) (**c**, **d**)

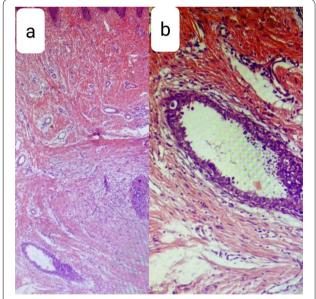


Fig. 2 Endometrial glandular structures surrounded by scant hypercellular stroma. **a** H&E  $\times$ 40, **b** H&E  $\times$ 100

as completely as possible by dermatologist, and oral progestin (Dienogest) was initiated. The remnants of the umbilical lesion had dramatically improved after 1 month (Fig. 1b). Follow-up is in progress.

#### Discussion

Endometriosis is a benign disease with incidence of 6–10% in women of childbearing age [1], usually being seen in the pelvic area. Cutaneous endometriosis can appear in less than 5% of cases [2], most of which have a history of former surgery [3]. Umbilical endometriosis appeared in 0.5–4% of affected cases, usually presenting with painful umbilical mass with periodic discharge or bleeding. An interesting feature of this case is that the lesion was totally asymptomatic. Another prominent feature is the presence of bilateral ovarian endometrioma, which reveals the importance of gynecological assessment in patients with cutaneous endometriosis to identify any pelvic involvement, which is reported to occur in 15% of these patients [4].

Regarding treatment of cutaneous endometriosis, complete excision is considered as the treatment of choice with or without hormonal therapy for ameliorating the symptoms [5–7].

Ovaries are the most common sites of endometriosis [8], and ovarian endometrioma accounts for 35% of all benign ovarian cysts [9]. Interestingly, ovarian endometriomas are more frequent in the left versus right ovary, possibly due to anatomical asymmetry and compression leading to venous congestion and hypoxia in the left side, affecting release of cytokines and sex hormones [10, 11]. In our case, endometriomal cyst in left ovary was considerably larger than on the other side, which might be in favor of the above-mentioned theory.

In addition to pain, discomfort, and fertility issues, an increased risk of infection, rupture, or transformation into ovarian cancer [12] obligates surgical intervention in larger ovarian endometriomas [9]. Nonsteroidal anti-inflammatory drugs, gonadotropin-releasing hormone (GnRH) agonists, and progestins are also considered as mainstream therapeutic options [9].

#### Conclusion

Umbilical endometriosis is a rare entity that might occur with concurrent pelvic endometriosis. Hence, thorough gynecological assessment is necessary in such patients to enable proper management.

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#### **Author contributions**

FM: visiting the patient. PH: writing drafts. PR: histopathological evaluation. ZA: supervision. All listed authors participated equally in preparing the manuscript. All authors read and approved the final manuscript.

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#### Availability of data and materials

The data that support the findings of this study are available from the corresponding author upon reasonable request.

#### Declarations

## Ethics approval and consent to participate

Ethical approval from the Medical Ethics Committee of Isfahan University of Medical Sciences was provided.

#### Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

#### **Competing interests**

The authors declare that they have no competing interests.

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#### References

- 1. Giudice LC, Kao LC. Endometriosis. Lancet. 2004;364:1789–99.
- Kyamidis K, Lora V, Kanitakis J. Spontaneous cutaneous umbilical endometriosis: report of a new case with immunohistochemical study and literature review. Dermatol Online J. 2011;17:5.

- Choi JK, Bae HA, Sang JH, Chung SH. Postmenopausal spontaneous umbilical endometriosis: a case report. J Menopausal Med. 2020;26(1):44– 6. https://doi.org/10.6118/jmm.19016.
- 4. Gin TJ, Gin AD, Gin D, Pham A, Cahill J. Spontaneous cutaneous endometriosis of the umbilicus. Case Rep Dermatol. 2013;5:368–72.
- Cucinella G, Granese R, Calagna G, Candiani M, Perino A. Laparoscopic treatment of diaphragmatic endometriosis causing chronic shoulder and arm pain. Acta Obstet Gynecol Scand. 2009;88(12):1418–9. https://doi. org/10.3109/00016340903314088.
- Obata K, Ikoma N, Oomura G, Inoue Y. Clear cell adenocarcinoma arising from umbilical endometriosis. J Obstet Gynaecol Res. 2013;39(1):455–61. https://doi.org/10.1111/j.1447-0756.2012.01964.x.
- Gopalan A, Sharp DS, Fine SW, et al. Urachal carcinoma: a clinicopathologic analysis of 24 cases with outcome correlation. Am J Surg Pathol. 2009;33(5):659–68. https://doi.org/10.1097/pas.0b013e31819aa4ae.
- Gordts S, Koninckx P, Brosens I. Pathogenesis of deep endometriosis. Fertil Steril. 2017;108:872–86. https://doi.org/10.1016/j.fertnstert.2017.08.036.
- Gałczyński K, Jóźwik M, Lewkowicz D, Semczuk-Sikora A, Semczuk A. Ovarian endometrioma—a possible finding in adolescent girls and young women: a mini-review. J Ovarian Res. 2019;12(1):104. https://doi. org/10.1186/s13048-019-0582-5.
- Sznurkowski JJ, Emerich J. Endometriomas are more frequent on the left side. Acta Obstet Gynecol Scand. 2008;87:104–6. https://doi.org/10.1080/ 00016340701671929.
- Matalliotakis IM, Cakmak H, Koumantakis EE, Margariti A, Neonaki M, Goumenou A. Arguments for a left lateral predisposition of endometrioma. Fertil Steril. 2009;91:975–8. https://doi.org/10.1016/j.fertnstert.2008.01. 059
- 12. Suryawanshi S, Huang X, Elishaev E, Budiu RA, Hang L, Kim S, *et al.* Complement pathway is frequently altered in endometriosis and endometriosis-associated ovarian cancer. Clin Cancer Res. 2014;20:6163–74. https://doi.org/10.1158/1078-0432.CCR-14-1338.

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