



Primary umbilical endometriosis presenting with umbilical bleeding: A case report

Ifeoma Ogamba^{a,*}, Samuel Napolitano^a, Linus Chuang^a, Deborah August^b, Kathleen LaVorgna^b

^a Danbury Hospital, 24 Hospital Ave, Danbury, CT 06810, United States of America

^b Norwalk Hospital, 34 Maple St, Norwalk, CT 06850, United States of America

ARTICLE INFO

Keywords:

Umbilical endometriosis
Umbilical bleeding
Cyclic umbilical swelling
Gynecology

ABSTRACT

Endometriosis is a condition in which endometrial tissue implants outside the uterine cavity, which can cause cyclic pain, dysmenorrhea, dyspareunia, and infertility. Endometriosis implants are typically seen on pelvic peritoneal surfaces and extra-pelvic disease is uncommon. We present an interesting case of primary umbilical endometriosis in a patient who presented with umbilical bleeding with a history of pelvic inflammatory disease complicated by bilateral tubo-ovarian abscesses. She was found to have an umbilical mass, which was resected, and the pathology was consistent with endometriosis. The objective of this case report is to detail the case and discuss diagnosis and management of umbilical endometriosis. It is important to recognize that umbilical endometriosis can occur in patients with no surgical history.

1. Introduction

Endometriosis is a benign gynecologic condition in which functional endometrial tissue appears outside of the endometrial cavity. The World Health Organization estimates that endometriosis affects 10% (190 million) of reproductive-aged women globally [1]. Presentation of endometriosis varies, but common symptoms in diagnosed cases include abdominopelvic pain, dysmenorrhea, and/or heavy menstrual bleeding [2]. Concordantly, endometriosis is prevalent in up to 50-70% of patients presenting with these symptoms [3,4].

Retrograde menstruation into the peritoneal cavity during menses is the most notable pathogenesis for the disease [5,6]. Peritoneal conditions must be such that fragmented endometrial cells evade immune clearance, attach to peritoneal epithelium, establish neuro-vascularity, and induce an inflammatory response for disease to exist [7-10]. These conditions may be a consequence of, but are not restricted to, genetic risk factors for endometriosis - as heritable genetic mutations can increase the survival and growth of refluxed cells [11-13]. Nonetheless, decreased progesterone exposure, as in nulliparity and prolonged endogenous estrogen exposure (i.e. early menarche, late menopause, short menstrual cycles), remains the most commonly observed risk factor in diagnosed cases [14-16].

Sites of refluxed cell implantation commonly occur in organs or structures within the pelvis, with the ovaries being the most common

[17]. Less common sites include the diaphragm, lungs, and anterior abdominal wall [18,19]. Extra-pelvic sites are rare, especially umbilical endometriosis, which has a prevalence of 0.5-1% [19]. The purpose of this report is to present a case of primary umbilical endometriosis and to discuss the management.

2. Case Presentation

The patient was a 37-year-old woman, gravida 1, para 1, with a past medical history of pelvic inflammatory disease complicated by bilateral tubo-ovarian abscesses who presented to the emergency room for the first time complaining of umbilical/periumbilical pain and umbilical bleeding. She described the pain as sharp and throbbing, radiating outward in all directions but worse at the umbilicus. The pain started 5 days prior to admission but the bleeding occurred on the day of admission, and had prompted her emergency department visit. Her last menstrual period started 4 days prior to admission and she reported that the bleeding ceased with cessation of her menses. She denied associated symptoms, including umbilical/abdominal trauma, fever, chills, vomiting, diarrhea, and constipation. Vital signs on admission were within normal limits. Labs included a hemoglobin of 11.7 g/dL, platelet count of $403 \times 10^9/L$, negative urine b-hCG, and an unremarkable urine analysis. CT of the abdomen/pelvis with contrast showed resolving bilateral tubo-ovarian abscesses with new accumulated fluid along

* Corresponding author.

E-mail address: Ogamba.maureen@gmail.com (I. Ogamba).

<https://doi.org/10.1016/j.crwh.2022.e00441>

Received 10 July 2022; Received in revised form 9 August 2022; Accepted 11 August 2022

Available online 14 August 2022

2214-9112/© 2022 Published by Elsevier B.V. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).



Fig. 1. Umbilical mass with swelling and discoloration due to endometriosis.

aspects of the right abscess, and potential colonic inflammation. The general surgery service was consulted and noted a mass beneath the umbilical skin on physical exam (Fig. 1). Umbilical endometriosis was suspected given that the umbilical pain and bleeding had a cyclic pattern and correlated with her menses. The gynecology service was consulted given concern for endometriosis.

The following day the patient underwent a joint surgical procedure with general surgery and gynecology-oncology. The general surgery team excised the umbilical mass. Upon excision, dark red sanguinous fluid resembling old blood was noted, as was a 1-2 mm mass with extension into the peritoneal cavity. The gynecological team performed a diagnostic laparoscopy and found significant peritoneal/abdominal adhesions, a large adnexal cystic structure containing greenish discharge

upon rupture, which might have been a residual of the tubo-ovarian abscesses, two cystic uterine masses, and a normal liver edge and diaphragm. Lysis of adhesions and resection of cystic masses were performed. Then general surgery performed closure of the umbilical trocar site and umbilicoplasty.

The patient had an uncomplicated postoperative recovery and she was discharged on postoperative day 1. Upon discharge, she was started on combined oral contraceptives for endometriosis management. Pathology of the umbilical and pelvic masses revealed endometrial glands and stroma consistent with extra-pelvic endometriosis (Fig. 2).

3. Discussion

Extra-pelvic endometriosis sites are not common, especially the umbilicus. Cutaneous endometriosis can occur in scarred areas such as cesarean section scars and episiotomy [20]. Primary umbilical endometriosis occurs spontaneously and secondary umbilical endometriosis occurs after a surgical procedure, specifically after laparoscopy or open abdominal surgery. Due to the rarity, there is a paucity of data on umbilical endometriosis and most of the information is from case reports.

Diagnosis of umbilical endometriosis is initially clinical, but histopathology confirms the diagnosis. Clinical presentation includes cyclic pain, umbilical mass with discoloration and/or bleeding. A systematic review by Victory et al. found that pain was the most common symptom (77.93% of patients), along with bleeding and swelling, which is consistent with our patient presentation [21]. In the previously mentioned article, 73.1% of patients had no history of endometriosis. Interestingly, our patient had no prior history of endometriosis and denied history of dysmenorrhea, menorrhagia, cyclic pelvic pain or any other endometriosis symptoms. However, her pathology showed pelvic endometriosis.

One of the theoretical causes of spontaneous endometriosis is shedding of endometriotic cells which are transported through the lymphatic and vascular system to the umbilicus [22]. Interestingly, our patient had resolving bilateral tubo-ovarian abscesses which were drained via

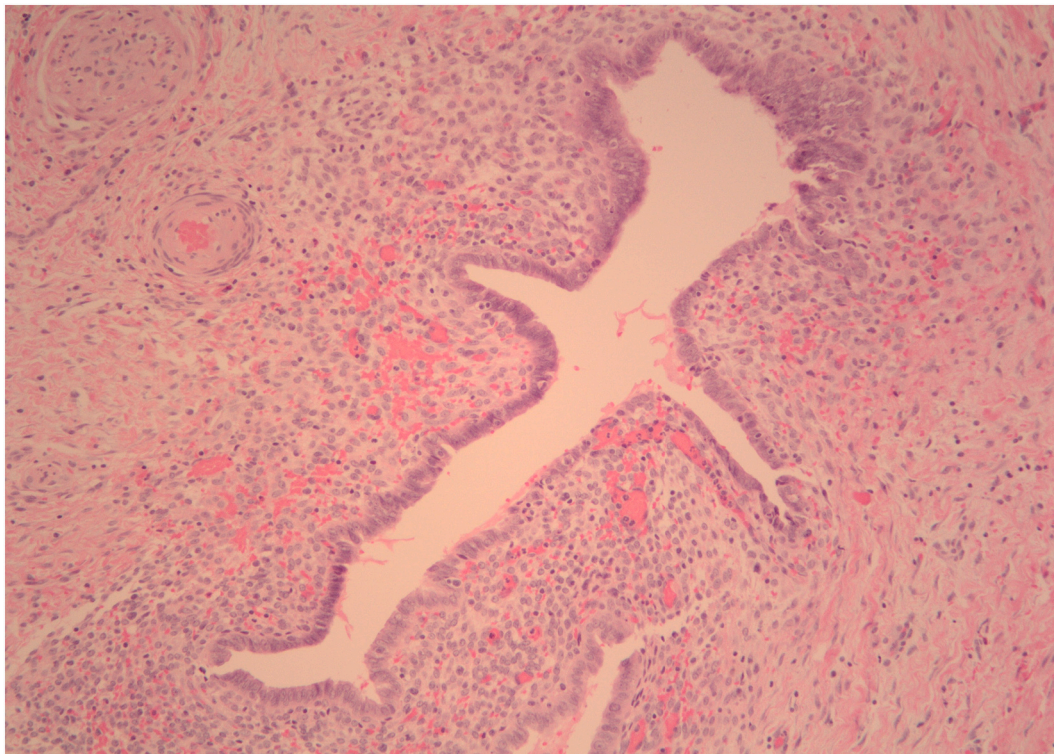


Fig. 2. Histology image of umbilical mass resection.

interventional radiology two months prior to her admission. The inflammation from the tubo-ovarian abscesses and subsequently the drainage could have led to seeding of pelvic endometriotic implants to the umbilicus. Providers should consider umbilical endometriosis in patients presenting with cyclic umbilical bleeding/pain and umbilical swelling; however, there are several other differential diagnoses. One should consider urachal malignancy, melanocytic nevus, umbilical polyp, and umbilical hernia.

Surgical management is the primary treatment of choice. Surgery may include resection of the umbilical mass and evaluating for pelvic endometriosis via laparoscopy if possible. Other treatment options include medical management using combined oral contraceptives (COCs) or progestins to reduce stimulation and inflammatory effects of the endometriotic implants. Our patient was started on COCs for long-term management of endometriosis. Gonadotropin-releasing hormone (GnRH) antagonists or agonists could also be considered for long-term management. However, our patient was not eligible for the medication due to costs.

4. Conclusion

Umbilical endometriosis is uncommon but should be considered in patients with umbilical swelling or bleeding and cyclic pain. It is important to recognize that it can occur spontaneously in patients with no surgical history as depicted in our patient. Hopefully, our case report will raise awareness of this rare condition and provide understanding of the clinical presentation and treatment.

Contributors

Ifeoma Ogamba was involved with patient care and writing of the case report.

Samuel Napolitano was involved with patient care and writing of the case report.

Linus Chuang was involved with patient care and editing of the case report.

Deborah August was involved with review of histopathology and obtaining images.

Kathleen LaVorgna was involved with patient care and review of the case report.

Funding

This work did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Patient consent

Obtained.

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 article.

References

- [1] Organization, W. H, Endometriosis, Retrieved from, <https://www.who.int/>, 2021, March 31, <https://www.who.int/news-room/fact-sheets/detail/endometriosis>.
- [2] K.D. Ballard, H.E. Seaman, C.S. de Vries, J.T. Wright, Can symptomatology help in the diagnosis of endometriosis? Findings from a national case-control study—Part 1, *BJOG Int. J. Obstet. Gynaecol.* 115 (11) (2008) 1382–1391, <https://doi.org/10.1111/j.1471-0528.2008.01878.x>.
- [3] N. Sinaii, K. Plumb, L. Cotton, A. Lambert, S. Kennedy, K. Zondervan, P. Stratton, Differences in characteristics among 1,000 women with endometriosis based on extent of disease, *Fertil. Steril.* 89 (3) (2008) 538–545, <https://doi.org/10.1016/j.fertnstert.2007.03.069>.
- [4] B. Eskenazi, M.L. Warner, Epidemiology of endometriosis, *Obstet. Gynecol. Clin. N. Am.* 24 (2) (1997) 235–258, [https://doi.org/10.1016/s0889-8545\(05\)70302-8](https://doi.org/10.1016/s0889-8545(05)70302-8).
- [5] R. Burney, L. Giudice, Pathogenesis and pathophysiology of endometriosis, *Fertil. Steril.* 98 (3) (2012) 511–519, <https://doi.org/10.1016/j.fertnstert.2012.06.029>. Retrieved from, <https://escholarship.org/uc/item/8nz1b9dw>.
- [6] A.E. Dastur, P.D. Tank, John A Sampson and the origins of endometriosis, *J. Obstet. Gynaecol. India* 60 (4) (2010) 299–300, <https://doi.org/10.1007/s13224-010-0046-8>.
- [7] D.J. Oosterlynck, F.J. Cornillie, M. Waer, M. Vandeputte, P.R. Koninckx, Women with endometriosis show a defect in natural killer activity resulting in a decreased cytotoxicity to autologous endometrium, *Fertil. Steril.* 56 (1) (1991) 45–51, [https://doi.org/10.1016/s0015-0282\(16\)54414-8](https://doi.org/10.1016/s0015-0282(16)54414-8).
- [8] A.W. Nap, P.G. Groothuis, A.Y. Demir, J.W. Maas, G.A. Dunselman, A.F. de Goeij, J.L. Evers, Tissue integrity is essential for ectopic implantation of human endometrium in the chicken chorioallantoic membrane, *Human Reprod.* (Oxford, England) 18 (1) (2003) 30–34, <https://doi.org/10.1093/humrep/deg033>.
- [9] A. Asante, R.N. Taylor, Endometriosis: the role of neuroangiogenesis, *Annu. Rev. Physiol.* 73 (2011) 163–182, <https://doi.org/10.1146/annurev-physiol-012110-142158>.
- [10] J.L. Shifren, J.F. Tseng, C.J. Zaloudek, I.P. Ryan, Y.G. Meng, N. Ferrara, R.B. Jaffe, R.N. Taylor, Ovarian steroid regulation of vascular endothelial growth factor in the human endometrium: implications for angiogenesis during the menstrual cycle and in the pathogenesis of endometriosis, *J. Clin. Endocrinol. Metab.* 81 (8) (1996) 3112–3118, <https://doi.org/10.1210/jcem.81.8.8768883>.
- [11] J.L. Simpson, S. Elias, L.R. Malinak, V.C. Buttram Jr., Heritable aspects of endometriosis. I. Genetic studies, *Am. J. Obstet. Gynecol.* 137 (3) (1980) 327–331, [https://doi.org/10.1016/0002-9378\(80\)90917-5](https://doi.org/10.1016/0002-9378(80)90917-5).
- [12] R.K. Jones, R.F. Searle, J.N. Bulmer, Apoptosis and bcl-2 expression in normal human endometrium, endometriosis and adenomyosis, *Human Reprod.* (Oxford, England) 13 (12) (1998) 3496–3502, <https://doi.org/10.1093/humrep/13.12.3496>.
- [13] M. Wingfield, A. Macpherson, D.L. Healy, P.A. Rogers, Cell proliferation is increased in the endometrium of women with endometriosis, *Fertil. Steril.* 64 (2) (1995) 340–346, [https://doi.org/10.1016/s0015-0282\(16\)57733-4](https://doi.org/10.1016/s0015-0282(16)57733-4).
- [14] E. Jenabi, B. Fereidooni, S. Khazaei, The association between parity and the risk of endometriosis: a meta-analysis, *Erciyes Med. J.* 43 (3) (2021) 228–232.
- [15] L.B. Signorello, B.L. Harlow, D.W. Cramer, D. Spiegelman, J.A. Hill, Epidemiologic determinants of endometriosis: a hospital-based case-control study, *Ann. Epidemiol.* 7 (4) (1997) 267–271, [https://doi.org/10.1016/s1047-2797\(97\)00017-3](https://doi.org/10.1016/s1047-2797(97)00017-3).
- [16] S.A. Treloar, T.A. Bell, C.M. Nagle, D.M. Purdie, A.C. Green, Early menstrual characteristics associated with subsequent diagnosis of endometriosis, *Am. J. Obstet. Gynecol.* 202 (6) (2010) 534.e1–534.e5346, <https://doi.org/10.1016/j.ajog.2009.10.857>.
- [17] A. Audebert, S. Petousis, C. Margioulas-Siarkou, K. Ravanos, N. Prapas, Y. Prapas, Anatomic distribution of endometriosis: a reappraisal based on series of 1101 patients, *Eur. J. Obstet. Gynecol. Reprod. Biol.* 230 (2018) 36–40, <https://doi.org/10.1016/j.ejogrb.2018.09.001>.
- [18] D.B. Redwine, Diaphragmatic endometriosis: diagnosis, surgical management, and long-term results of treatment, *Fertil. Steril.* 77 (2) (2002) 288–296, [https://doi.org/10.1016/s0015-0282\(01\)02998-3](https://doi.org/10.1016/s0015-0282(01)02998-3).
- [19] A.J. Dwivedi, S.N. Agrawal, Y.J. Silva, Abdominal wall endometriomas, *Dig. Dis. Sci.* 47 (2) (2002) 456–461, <https://doi.org/10.1023/a:1013711314870>.
- [20] L.E. Albrecht, V. Tron, J.K. Rivers, Cutaneous endometriosis, *Int. J. Dermatol.* 34 (1995) 261–262, 1:STN:280:ByqA3M%2FgvVI%3D 7790141.
- [21] R. Victory, M.P. Diamond, D.A. Johns, Villar's nodule: a case report and systematic literature review of endometriosis externa of the umbilicus, *J. Minim. Invasive Gynecol.* 14 (1) (2007) 23–32.
- [22] K. Obata, N. Ikoma, G. Oomura, et al., Clear cell adenocarcinoma arising from umbilical endometriosis, *J. Obstet. Gynaecol. Res.* 39 (2013) 455–461.