



CASE REPORT

# Management of Isolated Umbilical Endometriosis in a Resource Limited Country: Two Cases and Review of the Literature

Dehi Boston Mian<sup>1,2,3,4</sup> · Védi Loue<sup>1</sup> · Alexis Yao<sup>2</sup> · N'guessan Koffi<sup>1</sup> · Boni Serge<sup>1,2</sup>

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## Introduction

Endometriosis (EM) is an enigmatic, relatively common benign gynecologic disease, affecting 6–10% women of reproductive age [1]. This frequency is underestimated in general population [1, 2]. The abnormal lesions are typically found in the pelvic cavity but can also occur in extrapelvic areas [1, 2]. Umbilical endometriosis (UEM) is the most common cutaneous form, likely secondarily to a surgical scar. This location of EM, also known as Villar's node, is a rare disease accounting for 0.5–1% of all extrapelvic forms [1–3]. The real pathogenic mechanism remains hypothetical and poorly elucidated; therefore, current medical therapeutic options are based more on symptomatology than etiology [1, 2]. The patient usually presents with cyclic pain and bleeding from an umbilical skin nodule. The objective of this retrospective case series evaluation is to report the presentation, pathogenic aspects, current diagnosis and management features in a low- and middle-income country (LMIC).

## Presentation of the Cases

### Case 1

A 32-year-old nulliparous woman consulted for a painful umbilical swelling of 3 years duration, with cyclic increase during menstruation. She did not have a history of a prior abdominal surgery or dysmenorrhea. Examination showed a black appearing umbilical nodule approximately 3 cm × 2 cm in size. This umbilical swelling was firm, mobile, painful and irreducible (Fig. 1a). This macroscopic appearance was highly suspicious for umbilical endometriosis. CA-125 was unremarkable (13.2 IU/ml). Surgery under general anesthesia enabled nodule removal and umbilical reconstruction. Histological examination of the specimen confirmed endometriosis (Fig. 1b). Post-op esthetic surgical result and follow-up were satisfactory. No recurrence was noticed 6 months after surgery, and since then she has not returned for consultation.

### Case 2

A 34-year-old woman came to our hospital for a painless bleeding nodule in umbilicus. She was desirous for further fertility cause her only living child was 3 years old. As stated by her, the lesion started growing 1 year ago with spontaneous cyclic frank bleeding during menstruation and appearance of secondary dysmenorrhea. She had a history of two surgical procedures: abdominal laparotomy myomectomy 4 years ago, and cesarean 2 years ago. Examination revealed a painless rigid node, with bleedings areas, approximately

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MIAN DEHI BOSTON is an Associate Professor of GO of CAMES and Specialist in GO ultrasound, medically assisted reproduction

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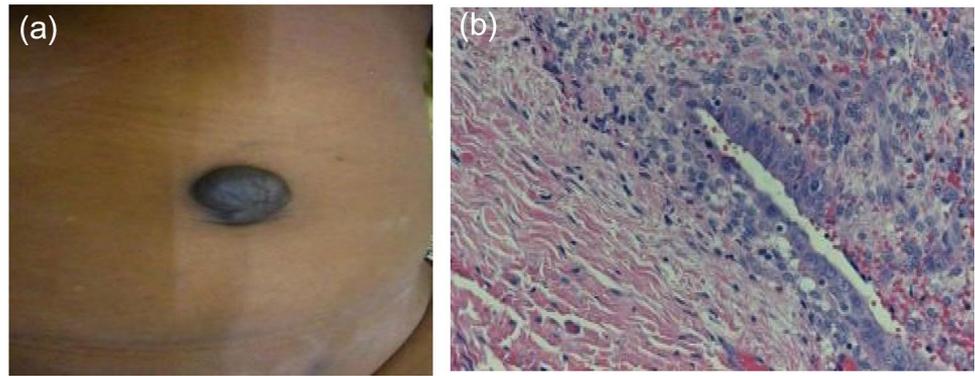
Dehi Boston Mian is a Professor; Védi Loue is a Doctor; Alexis Yao is a Doctor; N'guessan Koffi is a Professor, Boni Serge is a Professor.

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✉ Dehi Boston Mian  
bostondehimian@yahoo.fr

- <sup>1</sup> Department of Gynecology and Obstetrics, University Felix Houphouët Boigny of Cocody, Abidjan, Côte d'Ivoire
- <sup>2</sup> Department of Gynecology and Obstetrics, University Hospital of Cocody, Abidjan, Côte d'Ivoire
- <sup>3</sup> Research Unit of Medical Sciences, University Felix Houphouët Boigny de Cocody, BP Abidjan 03, Abidjan, Côte d'Ivoire
- <sup>4</sup> Unit of Gynecology and Obstetrics (GO) of University Hospital of Cocody, BP V 13, Abidjan, Côte d'Ivoire

**Fig. 1** Primary cutaneous umbilical endometriosis macroscopic visual aspects can be identified in image (a) and Histological characteristics in image (b)



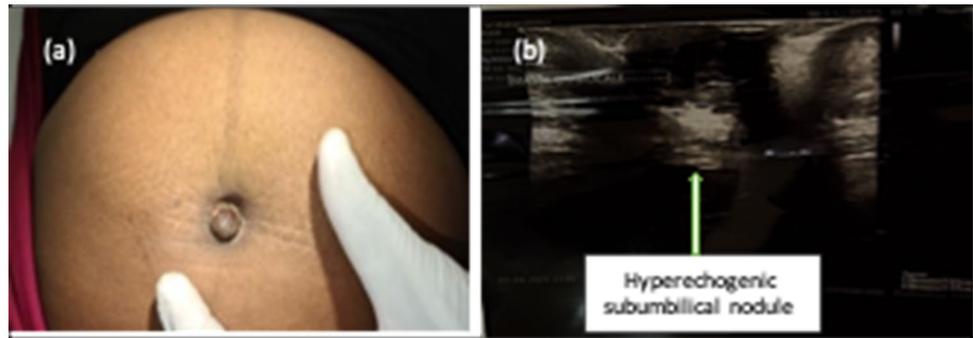
4 cm × 5 cm size. An abdominal ultrasound showed an umbilical, heterogenous hypoechogenic, ovoid mass (Fig. 2). We ordered a biopsy for histological analysis which was rejected by the patient due to psychological trauma related to previous surgeries. A continuous daily intake of norgestrel acetate (5 mg per day) for 2 months stopped the menses and umbilical bleeding. Symptoms reappeared 4 days after progestin was stopped, this led us to suspicion of diagnosis of endometriosis. The abdominal CT scan and MRI was not performed because it was too expensive and patient had low family income. The patient did not wish to receive GnRH analogues because she was desirous to have another baby. She has followed-up 3 months later with intermittent umbilical bleeding.

## Discussion

Endometriosis (EM) is characterized by the presence of endometrial tissue outside the uterus [1, 2]. It affects roughly 10–15% women or girls of reproductive age group globally and is responsible for pain and/or infertility [2–4]. This disorder causes a chronic inflammatory reaction and fibrosis that may result in the formation of abnormal scar tissue. It is commonly found within the pelvis but uncommon extrapelvic locations in the other parts of the body have been described [3, 4]. Cutaneous endometriosis (CEM) is one of the most infrequent sites, accounting for less than 5.5% [2, 5]. It has been frequently described in the umbilicus (Villar nodule), inguinal area and abdominal wall [1, 3, 5]. Cutaneous umbilical EM (CUEM) is a rare condition representing 0.5–4% of EM [3, 4]. In 4 years of practice, only two cases have been reported in our medical unit. CUEM appears as either primary (spontaneous) or secondary. The primary or spontaneous CUEM is less common (<30%) and observed in women with no past surgical history [1, 2, 5]. On the other hand, various risk factors pertaining to gynecological surgeries (laparoscopy or laparotomy, c-section, hysterectomy, myomectomy) are incriminated in onset of secondary CUEM [1–4]. Although few patients may not have clinical symptoms [1–5], the latter have been

described and include cyclic pain and/or bleeding, fatigue, depression or anxiety [1]. In cases of CUEM associated with pelvic EM, symptoms such as dysmenorrhea and dyspareunia are most common [1–3]. CUEM appears like a single multi-lobed swelling of either rubbery or firm consistency, ranging in size from a few millimeters to 6–9 cm (mean 2–2.5 cm) as described in both cases [1]. Lesion color varies from red or blue, to brown-black, depending on the level of bleeding, the depth of penetration of the ectopic endometrial tissue and patients' skin complexion [1, 5]. The main characteristics are tenderness, swelling, cyclic pain and/or bleeding [2, 5]. The diagnosis is confirmed by histopathological examination of surgical specimen [2, 4, 5]. In one of our cases histological analysis confirmed the presence of normal endometrial glands and stroma in the mid- or deep-dermis. Histological features may vary depending on the time of the menstrual cycle, as the presence of menses in the dermis leads to hemosiderin deposits, scarring and chronic inflammation [2, 4]. Diagnostic methods also include dermatoscopy, MRI, and ultrasonography. In both our cases, we used ultrasound scans due to their large availability and low cost. Ultrasonography and computed tomography tests are more readily available but less effective at diagnosing than MRI [2, 4]. Fine needle aspiration cytology can also be used in the event of inconclusive results [2, 5]. The differential diagnosis includes umbilical metastatic carcinoma, umbilical hernia, pyogenic or foreign body granuloma, melanoma, keloids developing on previous scars [1, 5]. Whereas the alleged pathogenetic mechanism of secondary or scar EM is iatrogenic implantation of endometrial tissue into the skin during a surgical procedure and that of spontaneous EM is still not well known [2, 5]. Several hypotheses have been proposed: the in situ transplantation theory, hematogenous/lymphogenous metastasis theory, induction theory, and a combination of the two former theories. Recently an immune theory involving the presence of adhesive endometrial cells (cadherins and integrins), impaired immune response in both T cell-mediated cytotoxicity and a decrease in natural killer (NK)-cell activity was highlighted [5]. Macrophages also seem to have key role by stimulating abnormal endometrial proliferation [5]. Hormonal and immune factors can create

**Fig. 2** Secondary cutaneous umbilical nodule, macroscopic visual aspects can be identified in image (a) and ultrasonographic features in image (b)



a pro-inflammatory microenvironment that facilitates persistence of these lesions [5]. Treatment options are medications and/or surgery depending on symptoms, lesions, desired outcome and patient choice [4, 5]. Surgical removal remains the main stay of treatment, and results in termination of menstrual flow at these sites [4, 5]. Surgical excision depends on tumor size and local extension. Two procedures can be used such as a simple excision of umbilical mass or laparoscopic large excision of the umbilicus [4, 5]. Contraceptive steroids, nonsteroidal anti-inflammatory medications, and analgesics are common therapies. Medical treatments for EM focuses on either lowering estrogen or increasing progesterone levels in order to alter hormonal environments that promote endometriosis. These medical therapies include the combined oral contraceptive pills, progestins, and GnRH analogues. However, none of these treatments eradicates the disease and endometriosis-related symptoms can sometimes reappear after therapy discontinuation. The choice of treatment depends on effectiveness in the individual, adverse side effects, long-term safety, cost, and availability. Although surgical management is the treatment of choice [1–5], it is not readily accepted by African women. Currently hormonal management is not suitable for persons suffering from EM who wish to get pregnant, since it affects ovulation. These treatments provide either an improvement in symptoms or a reduction in tumor size before excision [5]. A similar treatment was administered to both patients. Generally, the prognosis is good, and recurrences are uncommon if excision is performed with clean and wide margins [1, 4, 5]. Only one patient underwent surgical removal of the endometriotic nodule with a good short-term prognosis. Both patients have been lost to follow-up so long-term prognosis cannot be assessed.

## Conclusion

Cutaneous umbilical endometriosis (CUEM) is a rare entity which occurs primarily, or secondarily after surgery. The clinical presentation of the lesion is an umbilical swelling with cyclic pain and/or sometimes bleeding. Surgical

excision is the treatment of choice to confirm diagnosis by histological examination and prevent recurrence. It is difficult to do surgical management of endometriosis in LMICs because it is unavailable, expensive or declined. Likewise, medical treatment cost and consequences such as temporary infertility have also impeded this form of management because some women may be desirous of children.

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## Declarations

**Conflict of interest** The authors declare no conflict of interest with this publication.

**Ethics approval** was obtained from the teaching hospital of Abidjan University.

**Informed consent** Written and signed consent have been given by both patients for images used for scientific publication.

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