

## Primary Umbilical Endometriosis Masquerading as a Painful Umbilical Nodule: A Case Report

Ningi AB<sup>1</sup>, Jabbo MA<sup>2\*</sup>, Ibrahim JH<sup>1</sup> and Ibrahim HI<sup>1</sup>

<sup>1</sup>Department of Surgery, Abubakar Tafawa Balewa University Teaching Hospital Bauchi-Nigeria

<sup>2</sup>Department of Obstetrics and Gynaecology, 261 Nigerian Air Force Reference Hospital (NAFRH), Bauchi, Nigeria

### \*Corresponding author:

Mubarak Abdulkareem Jabbo,  
Department of Obstetrics and Gynaecology,  
261 NAFRH, Bauchi, Nigeria, Tel: +2348122292521;  
E-mail: mub2000us@yahoo.com

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Umbilical nodule; Cyclical pain; Extra pelvic endometriosis

## 1. Abstract

**1.1. Background:** Endometriosis is the presence of endometrial tissue outside the uterine cavity. Although the pelvis is the most common site of occurrence, extra pelvic areas like the umbilicus have previously been reported. Primary Umbilical Endometriosis is very rare; the reported incidence is 0.5-1% of all extra-pelvic diseases. It is often a second diagnosis, especially, in non-bleeding endometriosis. The cyclical monthly pain that coincides with the menstrual period often helps in establishing the clinical diagnosis.

**1.2. Case Report:** A 28-year-old married primipara who presented at the Surgical Out-patient Unit of 261 NAFRH Bauchi with 2-year history of recurrent umbilical pain and umbilical nodule noticed 3 months prior to presentation. The pain was dull in nature, aching, intermittent, worse during the menstrual period, and alleviated with nonsteroidal analgesia. The umbilical nodule was peanut-sized associated with cyclical pain that coincided with her menstrual flow. An abdominal Ultrasound Scan (USS) with a 5MHz probe indicated an umbilical soft tissue mass with benign features. An excisional biopsy was done and the histopathological analysis revealed an extra-uterine endometrial tissue with no malignant changes.

**1.3. Conclusion:** The clinical diagnosis of a cutaneous extra pelvic endometriosis is difficult, because, the potential differentials for such nodule are numerous. The presence of cyclical pain or bleeding concurrent with the menstrual flow should raise the index of suspicion. Histopathological diagnosis is often the final arbiter.

## 2. Introduction

A primary Cutaneous Endometriosis in the umbilicus was first re-

ported by Villar in 1886 [1]. Consequently, this condition is often referred to as the Villars's node [2]. Endometriosis is one of the biological quirks where an endometrial tissue is found growing outside the uterine cavity. It affects 10-15% of women, mainly, premenopausal [3]. It is mostly a benign lesion involving pelvic organs contiguous to the uterus. It is commonly found on the ovaries, ovarian fossa, utero-sacral ligaments, pouch of Douglas and the bladder [4]. Sometimes it is found as a cutaneous nodule in extra pelvic areas. The Villars's node is such an example. Umbilical Endometriosis is very rare, constituting about 0.5% to 1% of all diagnosed Endometriosis of extra genital sites [2]. Previous surgical scar is the most sited cause, although it may exist as primary umbilical endometriosis [5]. Various theories have been put forward to explain the aetiopathogenesis of Endometriosis. These include the possibility of a trans-coelomic migration of endometrial cells during menstruation, coelomic metaplasia or haematogenous and lymphatic metastatic spread of endometrial cells [6]. Differentiating umbilical nodules caused by Umbilical Endometriosis and other aetiologies is difficult clinically. However, it presents commonly as a reddish-brown umbilical nodule with cyclical dull aching pain or bleeding during the menstrual period [7]. Clinical examination often reveals a firm-hard, ovoid umbilical nodule with well-defined edges. Where there is endometriotic involvement of other sites such as the colon, rectum, or the small intestine, there may be associated colicky abdominal pain, abdominal distension, constipation, cyclical haematochezia or complete intestinal obstruction. Furthermore, if the endometriotic nodule is on the ureter the patient may present with cyclical, intermittent, colicky loin pain, haematuria or painful urination. Pulmonary endometriosis although

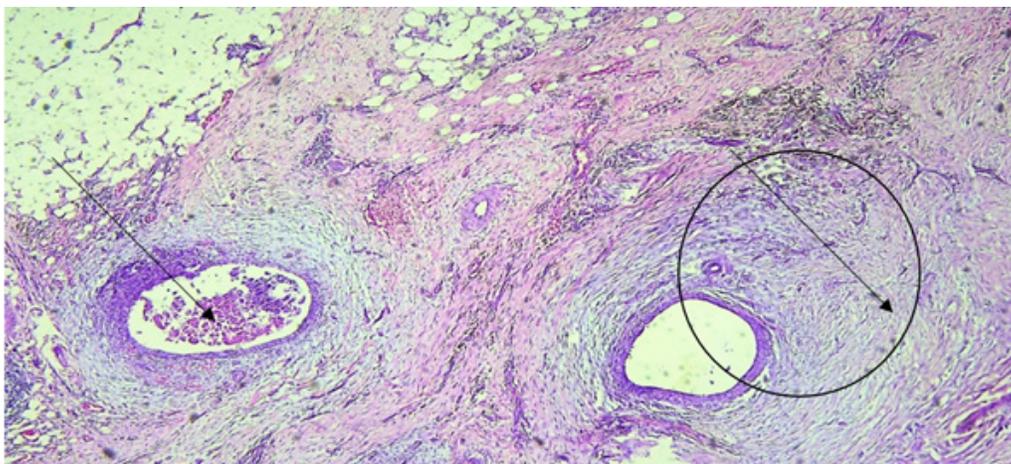
infrequent, may result in cough, haemoptysis, chest pain or dyspnoea during menstruation [8]. Histologic analysis is the definitive diagnosis of endometriosis. However, an abdominal USS or even MRI may be useful in the diagnosis of endometriosis. We present a case of a primary umbilical endometriosis in a 28-year-old lady who suffered a recurrent cyclical umbilical pain for 2 years and umbilical nodule noticed 3 months prior to presentation. She had spent a lot of money and resources for the evaluation and treatment of recurrent pelvic inflammatory disease, with no improvement of her symptoms.

### 3. Case Presentation

The patient is a 28-year-old married primipara whose last child birth was 3 years ago. Her last menstrual period was on 16th March, 2022. She was previously managed in the gynaecological clinic as a case of dysfunctional uterine bleeding characterised by both menorrhagia and dysmenorrhea of 2 months' duration. She was referred to the surgical out-patient department because 2-year history of recurrent umbilical pain and appearance of a painful umbilical nodule noticed 3 months prior to presentation. The umbilical swelling started as a peanut-sized swelling associated with cyclical dull aching pain that coincided with her menstrual flow. She also noticed a cyclical dark-bluish discoloration of the umbilical nodule during her men-

strual flow. There were no similar cutaneous nodules in other parts of the body. She had no history of bleeding from the umbilical nodule, cyclical haematuria, haematochezia, catamenial haemoptysis or seizure. There was no antecedent history of pelvic instrumentation or abdomino-pelvic surgery.

Physical examination revealed a dark-brown, ovoid shaped sub-umbilical mass, tender, smooth, firm to hard, measures 4x2 cm in dimension, with well-defined margins. It was entirely extra-peritoneal. Other parts of abdominal and pelvic examination were normal. An abdominal USS with a 5MHz probe indicated an umbilical soft tissue mass with benign features and no visceral or nodal enlargements. A pelvic scan showed a grossly normal endometrial plate, absence of leiomyoma and normal adnexae. An excisional biopsy was carried out and the histopathological analysis revealed an endometrial tissue with no malignant changes. There was presence of macrophages with hemosiderin pigments as shown in Figure 1. The patient was seen for follow up at the outpatient clinic at 2 weeks', 1 month and 3 months post-operative day. She reported complete recovery, the umbilical pain and nodule had disappeared. Furthermore, the surgical site had healed satisfactorily as shown in Figure 2.



**Figure 1:** Histologic sections of umbilical nodule showing endometrial glands (left arrow) within a fibrotic stroma (right arrow). Areas of hemosiderin pigment (circled area) are also noted. (H&E x100)



**Figure 2:** Image of the surgical site (arrow) taken at the outpatient clinic 2 weeks' post-operative day.

#### 4. Discussion

Any organ or tissue may be involved in extra-pelvic endometriosis. Endometriotic tissue has been found in the Colon and Rectum causing cyclical per rectal bleed, in the bladder, the Lungs and the Brain, resulting in catamenial haematuria, haemoptysis and seizure [6]. Primary cutaneous/spontaneous endometriosis develops *de novo*, in contrast to secondary cutaneous endometriosis which appears following iatrogenic or traumatic scar [5,9].

There is no consensus on its pathogenesis, as various theories have been put forward to explain this bizarre phenomenon. The theories put forward suggested the migration of endometrial cells through the abdominal cavity, the lymphatic system, or embryonic remnants in the umbilical fold (the urachus and umbilical vessels); genetic predisposition; and immunologic defects [5,6]. Furthermore, the Umbilicus is considered to be a physiologic scar, therefore, has tropism for endometriosis like other scar tissues. This may be the case in our patient, because, she had no preceding history of pelvic nor abdominal surgery.

The age of our patient at presentation was 28 years which is similar to cases reported elsewhere [10,11]. This indicates that endometriosis is an estrogen-dependent condition affecting women of reproductive age. The differentials for an umbilical nodule are numerous. The cyclical pain, variation in colour or size that coincides with menstruation, is often the only hint for a clinical diagnosis [12]. The index patient also presented with the monthly cyclical pain associated with changes in colour and size. A constant pain in an Umbilical Endometriosis has also been reported [13]. In the absence of pain, during pregnancy and menopause, establishing a clinical diagnosis may be difficult. It is therefore pertinent to keep in view other differentials such as: umbilical polyp, pyogenic granuloma, melanocytic nevus and epithelial inclusion cyst, desmoid tumour, haemangioma, granular cell tumour and seborrheic keratosis [14]. An incarcerated umbilical hernia, keloid, malignant lesions of the umbilical skin such as melanoma, squamous and basal cell carcinoma, and metastatic Sister Mary Joseph's nodules from the gastrointestinal tract, may also present as umbilical nodule [14]. Although majority are diagnosed based on clinical suspicion, radiological assessment could improve the chance of making the right diagnosis. An ultrasound evidence of an umbilical hypoechoic mass with foci of hyperechoic regions and evidence of increased vascular flow on Doppler interrogation is very helpful [15]. An ultrasound scan can also help the surgeon to estimate the size of the nodule, involvement of the peritoneum, pelvic organs and other viscera organs like the colon [15]. In difficult cases, an MRI may be done, especially if a pelvic endometriosis is suspected and to rule out transcoelomic spread of metastatic nodules from the gastrointestinal tract. The MRI features of an umbilical endometriosis reported is the presence of a homogeneous hypointense lesion on T1-weighted sequence with low signals on T2 weighting [16, 17].

The rarity of umbilical endometriosis has made its treatment to be

mainly dependent on the experience of the surgeon. Medical treatment has been tried in some patients, but no randomised controlled trial has been done to assess its efficacy [12,18]. The drugs commonly used include progesterone analogues, norethisterone, androgens like Danazol and Gonadotrophin Releasing Hormone (GnRH) analogue, such as Goserelin and Boserelin. The most consistent effect reported is the symptomatic relief of pain and reduction in the size of the nodule after medical treatment [14]. Our patient had surgical excision of the umbilical nodule. Surgery has so far remained the most promising treatment for Umbilical Endometriosis [18]. The goal should be a complete resection with no recurrence, but this is an uphill task in a disease with tropism for scar tissue like endometriosis. Surgical umbilical resection with or without repair of underlying fascia and peritoneum, or enucleation of the endometrial nodule with preservation of the umbilicus are mostly done. Total resection of umbilicus is however, preferred, as it carries lower risk of recurrence [14].

#### 5. Conclusion

A Primary Umbilical Endometriosis is rare and requires histopathological analysis for a definitive diagnosis. However, high index of suspicion is required regarding any umbilical nodule with cyclical symptoms. Due to the potential surgical differentials, a general surgeon plays a crucial role in the diagnosis of suspected Primary Umbilical Endometriosis.

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