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Spontaneous endometriosis associated with an umbilical hernia: A case report and review of the literature

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ABSTRACT

INTRODUCTION: Umbilical endometriosis occurring in the presence of an underlying hernia is extremely rare and presents a diagnostic challenge for the general surgeon. We present an interesting case and perform a comprehensive review of the literature.
METHODS: Medline and PubMed were queried for all cases of spontaneous umbilical endometriosis associated with an umbilical hernia. Data was analyzed and is presented along with an interesting case.
RESULTS: Only 7 cases have been reported in the literature. Median age was 38 years. Time to presentation was long (up to 5 years) and the majority had cyclical symptoms related to menstruation. All patients, including our case, were treated surgically.
DISCUSSION: Spontaneous umbilical endometriosis with an underlying hernia is often missed preoperatively. Preoperative suspicion warrants axial imaging for better operative planning and patient counseling. Surgery consists of enbloc excision of the umbilicus, implant and the hernia sac to avoid residual disease and reduce recurrence. The hernia defect can be repaired primarily or using mesh and the umbilicus reconstructed using skin flaps if necessary.
CONCLUSIONS: Surgery is the mainstay of therapy for umbilical endometriosis associated with an underlying hernia. Clinical suspicion warrants preoperative imaging, and follow-up with a gynecologist is essential to address any pelvic disease.

1. Introduction

Endometriosis, defined as the presence of functional endometrial glands and stroma outside the uterine cavity, is a common gynecologic condition affecting up to 22% of women [1,2]. It usually affects pelvic organs causing dysmenorrhea, dyspareunia, pain and infertility [3]. Extra-pelvic endometriosis is less common, but has been described in almost every area of the female body including the bowel, lungs brain, umbilicus and surgical scars [3,4].

Umbilical endometriosis is a rare condition accounting for 0.5–1% of endometrial ectopia. It usually develops in previous surgical scars but very rarely presents as primary or spontaneous umbilical endometriosis [5]. The first description of an umbilical endometrioma is credited to Villar in 1886, hence the term "Villar's nodule" [6].

The pathogenesis of endometriosis is widely debated but the most accepted theory is the "hypothesis of migration". This explains that the dispersion of endometrial tissue occurs by direct extension, vascular and lymphatic channels, and surgical manipulation [3,7]. While surgery can result in the direct inoculation and implantation of endometrial tissue in surgical incisions, the pathogenesis of primary cutaneous endometriosis is less clear. Lymphatics connecting the peritoneal cavity and the umbilicus course along the obliterated umbilical vessels. It has been postulated that the umbilicus serves as a physiologic scar with a predilection to implantation as endometrial cells course these lymphatic channels [8].

Umbilical hernias account for 3–8.5% of abdominal wall hernias [9]. The occurrence of a primary umbilical endometrioma in the presence of an underlying hernia is extremely rare and can present a diagnostic challenge to the general surgeon. We present a case of spontaneous endometriosis associated with an umbilical hernia along with a comprehensive literature review.

2. Case report

The patient is a 35 year old morbidly obese female who presented to clinic complaining of a 7 months history of cyclical umbilical bleeding. The bleeding would start 2 days before and last throughout her menses. She denied umbilical pain, dysmenorrhea, dyspareunia, infertility or a history of endometriosis. Her past surgical history included 2 caesarean sections through a lower abdominal (Pfannenstiel) incision. Her blood work was unremarkable except for iron-deficiency anemia (Hemoglobin of 11.4 g/dl). She had central obesity (Body Mass Index of 45.5 kg/m^2) making

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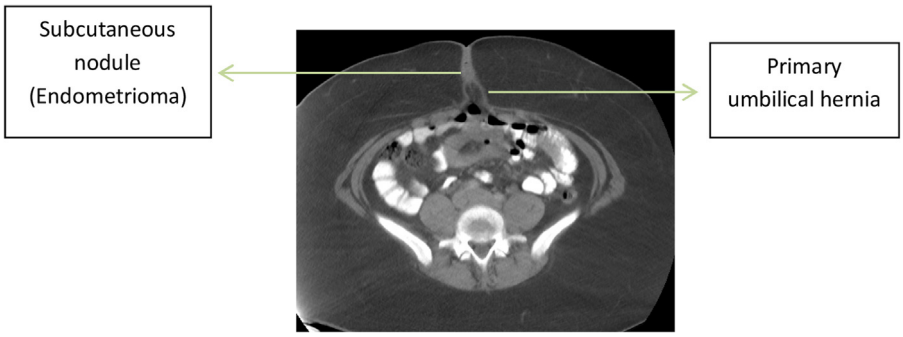

Fig. 1. CT scan demonstrating the subcutaneous nodule with an underlying umbilical hernia.

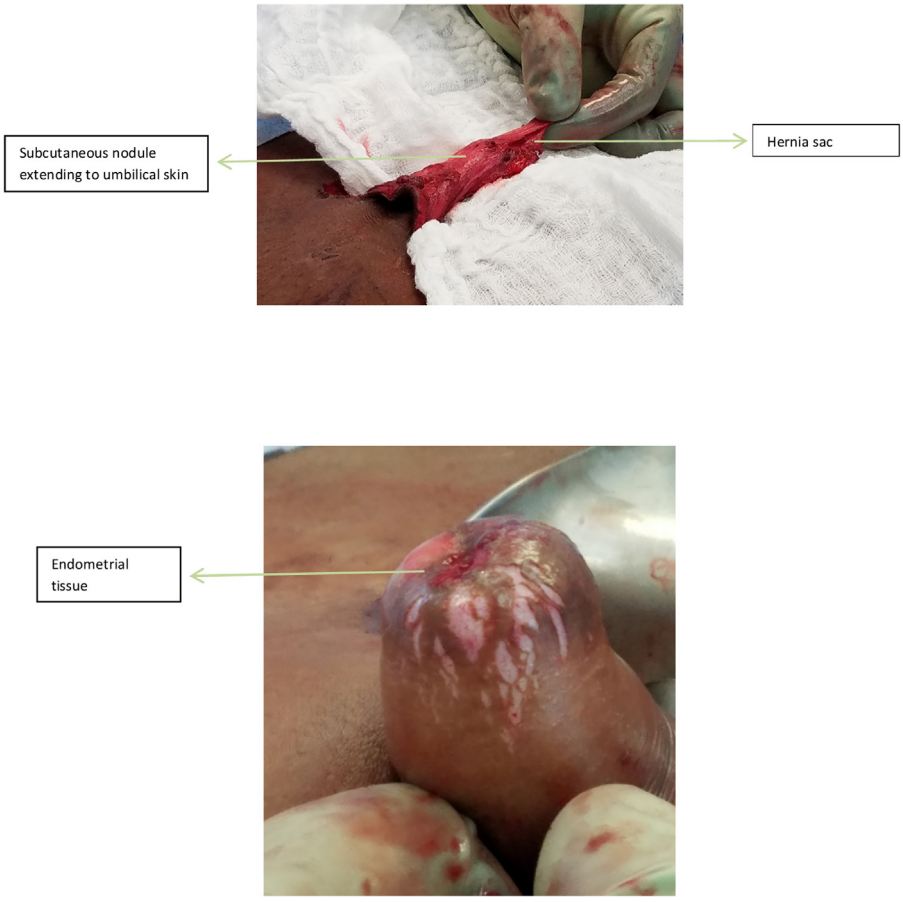

Fig. 2. Intraoperative images of endometrial tissue, subcutaneous mass and hernia sac.

the assessment of an umbilical nodule, mass or hernia difficult by physical exam. A CT scan was ordered to help with the differential diagnosis and demonstrated a subcutaneous nodule with an underlying umbilical hernia (Fig. 1). The nodule measured 1.9 × 1.67 mm. The patient was taken to the operating room and underwent a primary hernia repair with excision of the subcutaneous mass and umbilical reconstruction.

A vertical incision was made around the umbilicus and the wound was deepened using electro-cautery down to the abdominal wall fascia. The hernia sac was dissected and divided at the level of the fascia leaving it attached to the overlying subcutaneous nodule. The umbilicus was inverted and red-purple endometrial tissue was seen implanted at its base (Fig. 2). An Incision was made to include the involved skin and the specimen (skin, endometrioma and hernia sac) was pulled through and sent to pathology (Fig. 3).

The hernia was fixed using interrupted # 1 Polydiaxanone sutures. The umbilical skin was reconstructed using interrupted subcuticular 4-0 Monocryl sutures and then tacked down to the fascia using 3-0 Vicryl. The patient tolerated the procedure well and was discharged home from the recovery unit. The pathology demonstrated endometriosis (Fig. 4). She was referred for a gynecologic evaluation which was unremarkable. There was no disease or hernia recurrence at her 6 month visit.

3. Discussion

Primary cutaneous endometriosis is rare and poses a diagnostic challenge when associated with an underlying hernia. A comprehensive review of the English literature using PubMed and Medline was performed and only 7 of these cases have been described