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Case Study

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Umbilical Endometriosis and the Unsuspecting Plastic Surgeon

Nura Y. Gouda[1] and Marshall G. Miles[2*]

Abstract

Our article describes the case of a 31 y.o. woman who presented with an unbiopsied, painful umbilical mass that had been enlarging over the previous two years. Surgical excision under local anesthesia was planned with subsequent pathology report revealing a diagnosis of umbilical endometriosis. Our paper discusses this specific case in addition to a brief literature review encompassing diagnosis, pathophysiology, medical and surgical management, and other common differential diagnoses.

Introduction

Endometriosis is a condition in which endometrial tissue and glands are located outside the uterine cavity, and currently effects about 5-15% of the female population [6]. It is the most common cause of secondary dysmenorrhea, which is defined as pain due to a simultaneous pathologic process. Typical anatomical sites include ovaries, fallopian tubes and uterine ligaments, but can include extra-pelvic sites as well. Extra-pelvic endometriosis is seen in 9-12% of women with the disease, with reported sites including the bladder, GI tract, skin, lungs, and brain [1, 9]. The most common reported extra pelvic location is the abdominal wall [4]. Umbilical endometriosis (UE), also known as Villar's nodule, was first described in 1886 by Villar and is seen in 30-40% of cutaneous cases making it the most reported cutaneous location of endometriosis [1]. Other reported cutaneous sites include the arm, groin, episiotomy wounds, appendectomy scars and prior C-section scars [1, 3]. With regard to UE, secondary is more common than primary. Secondary UE is defined by iatrogenic seeding of endometrial tissue occurring in an area of prior abdominal or pelvic surgery scar, most often from laparoscopic trocar insertion sites or C-section scars [1]. Conversely, primary UE happens spontaneously making it rarer. For Umbilical endometriosis, correct pre-operative diagnosis is surprisingly seen in only 25% of cases [6].

Case Report

A 31 y.o. female presented to the plastic surgery clinic with an unbiopsied enlarging umbilical mass. The patient reported that the mass was painful and had been enlarging for the past 2 years. She denied drainage or bleeding from the umbilicus and denied any prior history of skin malignancies. Review of systems was non concerning. Surgical history only included tonsillectomy and adenoidectomy; no prior abdominal surgeries were reported. Physical exam noted a large mass in the umbilical area taking up the entire umbilical stalk with associated pain. Keloid was suspected, as they often occur from even minor trivial trauma. The patient agreed to surgical excision with reconstruction with possible flap, total umbilectomy, and intralesional Kenalog injection. The procedure was done under local anesthesia with IV sedation. During the procedure, a 5x4cm mass was excised from the umbilicus

Affiliation:

[1]OMS-III, D.O. Candidate, Class of 2025, Philadelphia College of Osteopathic Medicine, Pennsylvania, USA

[2]Division of Plastic Surgery, Lehigh Valley Health Network Surgical Institute of Excellence, Allentown, Pennsylvania, USA

***Corresponding Author**

Marshall G. Miles D.O., FACS, Division of Plastic Surgery, Lehigh Valley Health Network Surgical Institute of Excellence, Allentown, Pennsylvania, USA

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and sent to pathology. The mass encompassed the entire umbilicus and stalk, and had to be excised down through the fatty tissue plane resulting in total umbilectomy. Dissection was completed using tenotomy scissors to avoid injuring neighboring structures. It was then determined that the defect was unable to be closed and therefore a local rotational flap was created in order to lessen risk of wound dehiscence. The flap was inset into the defect in a multi-layer approach, and care was taken to attempt to pucker down the deeper tissues in order to recreate a typical aesthetic "belly-button" appearance. Following that, Kenalog was then injected into the flap in case the pathology report confirmed a diagnosis of keloid since at the time there had not yet been a diagnosis, but keloid was highly suspected. Pathology resulted 4 days later and noted skin, dermis and subcutis with endometriosis and associated ulceration.

Discussion

Endometriosis is a condition which affects 5-15% of the female population and is the most common cause of secondary dysmenorrhea and infertility. Umbilical Endometriosis (UE), aka Villar's nodule, is defined as a rare type of cutaneous endometriosis representing 0.5-1% of all ectopic sites of endometriosis. Patients with this pathology typically present to a plastic surgeon, general surgeon, dermatologist, or gynecologist often reporting a blue or violaceous nodule that may change in color to brown or skin colored [3]. The most common symptoms is pain, however swelling, bleeding, and discharge may or may not be present as well. Patients often do not become symptomatic until 1-2 years after, with mean a mean symptomatic duration of 17.8 months [2, 5, 6]. Interestingly, majority of patients with UE do not have concurrent pelvic endometriosis, with literature reporting only a third being simultaneously effected [1, 4, 5].

Diagnosis is typically based on objective findings and later confirmed by histopathology which is the gold standard and will reveal presence of endometrial glands and stroma with possible hemosiderin deposition in the mid to deep dermis [1, 2, and 6]. Pre-op imaging with doppler, CT, ultrasound, or MRI do not offer much clinical insight other than ruling out other more worrisome pathologies such as a large umbilical hernia or malignancy [6]. Since lesions typically infiltrated into the deep layers, they will not increase in size during Valsalva maneuver [9]. Other common differential diagnoses include keloid, umbilical hernia, Sister Mary Joseph Nodule malignancy, dermatofibroma, pyogenic granuloma, dermoid cyst, hemangioma, abscess, nodular melanoma, and embryogenic remnants. With such a long list of differentials with similar presentations, it makes sense why there is low accuracy in pre-op diagnosis. The size of the UE mass ranges from a few millimeters to 9 cm, with an average size of 2-2.5cm^2. UE can either be primary or secondary in etiology, but in general, published literature reveals that secondary is more common. In 2018, Lopez-Soto et al reported that only 109 cases of UE had been reported in the literature up to that point in time, only 30% of which were considered primary [3]. Secondary UE is typically caused by iatrogenic seeding of endometrial tissue during laparoscopic abdominal or pelvic surgeries [8, 11]. Conversely, primary UE is spontaneous. The most common theories on pathophysiology for UE include vascular or lymphatic migration, cellular metaplasia, or iatrogenic seeding. It is described by Victory et al, that microvascular endothelial injury may cause increased adhesion of tissue implants in the extragenital sites due to production of molecules such as e-cadherins and integrins. Afterwards, the tissue will then respond to hormonal and angiogenic factors such as VEGF, which provides further support for the association of UE with surgical scars. With regard to iatrogenic etiology, two studies interestingly suggested potential value of umbilical lavage after removal of umbilical trocars as a prophylactic measure to prevent UE as well [5, 8]. Seeding via hematogenous or lymphatic drainage is another plausible explanation as the umbilicus has a dedicated lymphatic supply with indirect pelvic drainage and can act as a physiological scar [5]. Additionally, implantation is more likely during menstruation, rather than during the proliferative or secretory phases [8].

Historically, management for umbilical endometriosis has included both medical and surgical treatment with surgical excision being the most definitive treatment. Of note, one case was reported by Heller et al which successfully used silver nitrate to treat UE but this has not been reproduced [8]. Medical management with oral contraceptive pills or gonadotropin releasing hormone analogs (danazol, leuprolide), are ineffective as patients must stay on them long term and this is not considered curative, as the lesion will grow back once medication is stopped [10]. However, they do provide benefit post excision to prevent recurrence and for women with UE who are near menopause [1]. GnRH analogues can also be used pre-operatively to help shrink the size of the lesion before removal by plastic surgeon. It is also recommended to perform the surgery at the end of the menstrual cycle [2, 6]. Surgical treatment with wide local excision is recommended with dissection of the umbilical peduncle down to the parietal peritoneum and subsequent umbilical reconstruction [4, 10]. Forgoing surgical treatment carries a 3% risk of malignant transformation, however occurrence is rare and there have not been many cases reported [4]. Lastly, in those that choose to undergo surgical excision, recurrence rate is reported to be around 4.7% [4]. Moving forward, it's important that we continue to consider umbilical endometriosis as a differential diagnosis in any female presenting with an umbilical mass. In addition, it may also be worthwhile to explore adjustments to technique during laparoscopic trocar removal in women

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undergoing abdominal and pelvic procedures during removal of uterine tissue. Although rare, UE causes significant discomfort and impacts the quality of life in those it affects and therefore attention should be given to decrease incidence, increase preventative measures, and increase awareness.

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