

Inguinal Scar Endometriosis: A Rare Combination of Two Conditions

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ABSTRACT

Endometriosis has been observed throughout history, well before modern medicine developed taxonomy and treatment for this frequently seen benign condition. Endometriosis was discovered microscopically and described in the 19th century by Von Recklinghausen. These lesions commonly occur in females of childbearing age and can be found invading any organ in the human body. They present with cyclical pain that waxes and wanes with the menstrual cycle, dyspareunia, dysmenorrhea, and other symptoms that drastically lowers quality of life. Endometriosis may take many years to produce noticeable symptoms. A serious cause of concern is when these lesions remain unnoticed and undergo malignant transformation into clear cell carcinoma or endometrioid carcinoma. The prognosis for these malignancies is usually poor, which further stresses the importance of screening and early surgical intervention. We recommend that our patients be attentively heard and carefully examined during routine checkups, especially if they present with known or uncommon gynecological symptoms. There are many presentations of this disease with many theories to explain the pathophysiology, however, our case will focus on iatrogenic influence during abdominopelvic procedures. Our patient's case involves a rare case of "Inguinal-Scar Endometriosis" associated with progressively increasing burning pain along a surgical scar accompanied with a palpable subcutaneous mass.

Keywords: Endometriosis; Scar endometriosis; Inguinal endometriosis; Cyclical pain; Dysmenorrhea; Dyspareunia; Peritoneal seeding; Abdominal seeding

INTRODUCTION

Endometriosis is a benign condition that most commonly occurs in females of childbearing age that histologically presents as endometrial glands surrounded by endometrial type stroma in aberrant locations and is defined as a condition in which endometrium-like tissues are present in organs other than the uterus [1]. Endometriosis was first described by Von Recklinghausen in 1885 as functional endometrial tissue outside the uterine cavity. Endometriosis is a benign condition that can cause dysmenorrhea, dyspareunia, menstrual irregularity, and can lead to infertility. The incidence has been reported to be up to 8% in the general population but the frequency of endometriosis seen incidentally during any gynecological intervention in the reproductive years is as high as 20% [2]. Endometriosis most commonly presents with menstrual pain, sensitivity, swelling, and may even present as a mass [2].

The most common locations where endometriosis presents are the ovaries, sacro-uterine ligament, rectovaginal septum, and peritoneum. It may rarely be seen in the vulva, vagina, appendix, stomach, liver, thorax, bladder, umbilicus, and the inguinal canal. The etiology of endometriosis is unknown despite abundant literature and some predominant theories. The most widely accepted theory for the pathogenesis of endometriosis is retrograde menstruation through the fallopian tubes into the pelvic cavity. Another common theory of endometriosis is iatrogenic seeding during surgical procedures and has been reported to occur subsequent to caesarean section, hysterectomy, myomectomy, in the episiotomy line following birth, in the trochar location after laparoscopy, and following pelvic

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gynecological operations related to the uterus and endometrium. Once endometrial cells adhere to tissue surfaces, they can grow and invade under the influence of the hormonal environment, altered immunity, inflammatory responses, angiogenesis, and other factors [3]. Although pelvic endometriosis can be explained by this implantation theory, extra-pelvic endometriosis, involving the thoracic cavity and abdominal wall, is difficult to explain with this theory alone. Another hypothesis proposes that metaplasia of the celomic epithelium may contribute to the development of the disease [3].

The relationship between abdominal wall endometriosis and gynecological operations was first determined by Aimakhu et al. in 1975. The most frequent location is below the incision scar, in the umbilicus, within the rectus muscle, in the inguinal canal, and rarely in an inguinal hernia pouch. Right laterality is more common when endometriosis is within an inguinal hernia pouch because intraperitoneal circulation is clockwise [2]. The mechanism of inguinal endometriosis can be explained by the reflux theory of menstrual blood into the pelvis. The abdominal fluid containing the endometrium circulates clockwise in the abdominal cavity, and the sigmoid colon blocks the abdominal fluid from entering the left inguinal ring. As a result, the intraperitoneal fluid is more likely to enter the right inguinal ring than the left. It is also speculated that endometriosis propagates from the pelvis to the groin via the round ligament. Inguinal endometriosis occurs in several forms, including cystic lesions in the hernia sac and Nuck's canal, and solid mass lesions in the extra-pelvic round ligaments and subcutaneous tissue [3].

Inguinal endometriosis is rare, with an incidence of 0.3%-0.6% in all endometriosis cases. While pelvic endometriosis usually causes cyclical pain that is exacerbated during menstruation, inguinal endometriosis frequently presents as an inguinal mass with a constant pain, which is not associated with the menstrual cycle [1]. Therefore, inguinal endometriosis can mimic other diseases such as a hernia, lymphadenopathy, abscess, and cancer. Patients may have to visit multiple departments, including internal medicine, surgery, and gynecology, before receiving a diagnosis of inguinal endometriosis [1]. Scar endometriosis is often palpable as a subcutaneous mass around the surgical scar on physical examination. If the mass is palpable, the physical examination should focus on determining if the mass is attached to the anterior fascia. More detailed examination is required, especially if the patient does not complain of a palpable mass or pain. Ultrasonography, computed tomography (CT), and magnetic resonance imaging (MRI) are all useful for diagnostic imaging. Ultrasonography is a simple procedure, which often shows abdominal wall endometriosis (AWE) as a hypoechoic solid mass. MRI and CT are helpful for clarifying the extent of the lesion, fascia involvement, and the depth of lesion invasion. On MRI, the presence of blood components, such as hemosiderin, within the abdominal wall mass is suggestive of endometriosis. Fine-needle aspiration can confirm the preoperative diagnosis of AWE and can help excluded malignancy [3].

To prevent our patients the inconvenience of visiting multiple specialists and the emotional effects from enduring the pain for

CASE

Our patient is a 37-year-old female who presented to the emergency department (ED) for pain in her left groin as well as a "knot" along a surgical scar on her abdomen. She has a past surgical history of laparoscopic salpingostomy/salpingectomy for ectopic pregnancy, open myomectomy for uterine fibroids, and two C-sections that presented with left sided incisional pain. She noticed the knot along her C-section incision on the left side and was experiencing a burning pain at the site. Our patient has an intrauterine device in place but still has her menses irregularly that accompanies the pain. She also experiences a feeling of fullness when the pain occurs. She takes ibuprofen for the pain which usually mildly mitigates it. Our patient endorses that the pain is more noticeable and occurs more frequently as of recently. She denies history of abdominal hernias and endometriosis symptoms.

A transvaginal ultrasound (US) was performed during her ED visit which revealed a uterine fibroid that measures 0.7 cm. US also revealed a $1.4 \times 1.0 \times 0.8$ cm hypoechoic structure present in the same area which corresponds to the palpable "knot". The surgical team was consulted who decided to excise the mass under general anesthesia. The surgical note indicated that the mass was within abdominal wall and was 2 cm superior to the C-section incision on the left side. After discussing the risks, benefits, and alternatives of the procedure with the patient, she endorsed understanding and consented to the procedure.

The surgical team made a 3 cm incision on the left side of Csection incision and dissected superiorly in the subcutaneous spaces and was able to palpate a subfascial mass. They dissected around the mass to free it from the surrounding tissues down to the level of the abdominal wall muscles using a combination of blunt dissection and electrocautery. The mass involved the fascia of the abdominal wall and the rectus muscle. After removal of the mass from the surrounding structures, the specimen was sent to pathology for microscopic evaluation. The patient was smoothly awoken from anesthesia and tolerated the procedure well with no acute concerns.

The specimen was sent to pathology designated as a "left groin mass". Gross examination of the specimen in the pathology department showed 3 tan-pink, fibro-membranous fragments of soft tissue that were covered in adipose tissue. The fragments were serially sectioned which revealed variegated cut surfaces exhibiting layers of white fibrous tissue, yellow lobulations, and red congested vessels. Histologic examination of the specimen was performed after formalin fixation, paraffin embedding, and staining with hematoxylin and eosin. Microscopy revealed numerous endometrial type glands and stroma aberrantly located in a background of fibro-adipose tissue and blood vessels. The endometrial glands were infiltrating through and entire had replaced sections of fibrous tissue. Immunohistochemical stain for CD10 was utilized to highlight endometrial stromal cells that revealed moderately positive

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staining of cells that surrounded endometrial glands and lumina. The pathologist's final diagnosis report read "Endometriosis".



Figure 1: (A) Overview of the cut surface upon serial sectioning of the mass lesion revealing hemorrhagic cystic cavities surrounded by fibrous septae in a background of fibrous connective tissue and adipose tissue. (B) High power view of the central two tissue sections revealing dark red hemorrhagic cystic cavities surrounded by tan-white fibrous septae in a variegated tan-white to tan- brown background of fibrous connective tissue embedded within lobules of smooth and glistening goldenyellow adipose tissue. (C) High power view of the far right tissue section revealing dark red hemorrhagic cystic cavities of various diameters and depths surrounded by tan-white fibrous septae in a variegated tan-white to tan-brown background of fibrosis embedded in lobules of smooth and glistening golden-yellow adipose tissue.



Figure 2: (A) Low power (2x) photo of atrophic endometrial glands circumferentially surrounded by endometrial stromal cells that are infiltrating through and replacing a background on fibrous connective tissue, artifactual clefting, lobules of benign adipose tissue, and blood vessels. (B) Low power (4x) photo of atrophic dark blue endometrial glands with no appreciable columnar cells that are surrounded by light blue staining endometrial stromal cells, aggregates of glandless endometrial stromal cells, and blood vessels in a background of eosinophilic connective tissue. (C) Intermediate power (10x) photo of small atrophic endometrial glands and clusters of endometrial stroma infiltrating through a band of connective tissue surrounded by larger foci of endometriosis on either side. (D) High power (20x) photo of glandless aggregates of endometrial stromal cells infiltrating through bundles of collagenous connective tissue with extravasated red blood cells percolating throughout the field. (E) High power (40x) photo of an aggregate of plump blue spindle cell nuclei with small, central nucleoli, homogenous, benign appearing chromatin, and moderate to scant cytoplasm present arranged in whorls. (F) Low power (2x) photo of positive CD10 immunohistochemical staining of spindled endometrial stromal cells surrounding rims of atrophic endometrial glands.

(G) Intermediate power (10x) photo of positive CD10 immunohistochemical staining of spindled endometrial stromal cells surrounding an endometrial gland.

DISCUSSION

Our patient tolerated the procedure well and upon follow-up with the surgical team, it was found that she no longer experienced the burning pain and no palpable mass was felt in her left abdomen anymore. There has not been a recurrence of the disease and our patient's treatment was a success.

The case presented here is a case of scar inguinal endometriosis that is an exceptionally rare form of endometriosis that occurs in females of childbearing age. It presents as a constant burning pain that can intensify with menstruation. This can be very detrimental to a patient's quality of life and in turn, mental health. It is important to expeditiously diagnose and treat the condition and prevent any delays in the process because malignant transformation of scar endometriosis was reported to have a very poor prognosis. In a review of 48 cases, the mean age at diagnosis was 46 years, 87.5% were associated with cesarean section, and 12.5% were associated with open uterine surgery. The histological types of malignancy that endometriosis can transform into includes, clear cell carcinoma (66.7%), which is the most common, followed by endometrioid carcinoma (14.6%). The average time between initial surgery and diagnosis of endometriotic malignant transformation was 19.3 years, which suggested a slow evolution of the disease [3].

Malignant transformation involves numerous factors including, genetics, immunological effects, and environmental factors. It has been reported that some iatrogenic endometrioses have a cancer-driving mutation, which may be related to the mechanism of malignant transformation. Malignant transformation is associated with a poor prognosis, which further emphasizes the need to surgically remove the lesion [3]. If our patient had denied surgical treatment she would have been offered medical therapy with oral contraceptives (OC), progestin, or gonadotropin-releasing hormone (GnRH) agonists. These therapeutics may be effective in improving symptoms and can be an option for patients who do not want surgery. However, the symptoms are likely to recur after hormonal therapy is discontinued. Surgical treatment has a low postoperative recurrence rate and minimal invasiveness, and therefore may be recommended as the first choice of treatment [3].

Surgical resection is recommended for accurate diagnosis and appropriate treatment. Extensive resection is an effective method to avoid postoperative recurrence. Some authors have suggested that surgical excision with 1-cm margins on all sides of the lesion is optimal. Endometriotic lesions are reported to involve the adipose layer (91.4%-96.9%), fascia (65.7%-67.2%), and muscular layer (17.2%-20.7%). It is suggested that the mass be removed with a resection margin appropriate to the extent of the lesion. Imaging tests such as CT and MRI are useful to determine the extent of the lesion before surgery. The postoperative recurrence rate of AWE is 4.5%-11.2%, which is lower than that of ovarian endometriosis. To date, there has been no report demonstrating that hormonal therapy is effective for AWE [3].

Many cases of AWE have been reported to occur after surgical procedures that involve the gynecological pelvic organs in females. It has been reported that time elapsing from surgery to clinical presentation of endometriosis may range from 45 days to 20 years. To reduce the risk of endometriosis developing from mechanical transportation, it is recommended at the final stage of uterine surgery procedures, that the gloves, needles, suture materials and sponges are changed and removed from the operation area and contact with the wound area should be reduced to a minimum. Cleaning the surgical field with highflow saline solution before closing the incision line is thought to be of benefit in reducing the occurrence of post-operative endometriosis after C-sections [2]. Therefore, it is recommended that great care be taken regarding surgical technique and the surgical field should be thoroughly cleansed prior to closure to prevent the seeding of uterine contents in the pelvic cavity and surgical scar. We also recommend considering endometriosis in the differential diagnosis in a female of childbearing age that presents with recurrent or constant pain in the abdominal wall, regardless of the presence of a palpable mass [4-11].

CONCLUSION

We recommend that clinicians have endometriosis on their differential when their female patient of childbearing age presents with constant or cyclical pain and irregular menstruation. This will prevent our patients from the inconvenience of visiting various specialties before receiving a correct diagnosis and treatment. It is suggested that clinicians consider their patient's past surgical history if it includes cesarean sections, myomectomies, or other pelvic gynecological procedures. Furthermore, if identified sooner, the early symptoms of inguinal endometriosis will allow for improved understanding of the disease. Early detection is important because of the risk of progression to malignancy. This form of malignancy is readily preventable. The risk for endometrial cell seeding is high because this cell type can express adhesive proteins, influence its hormonal environment, and invade surrounding tissues. Subcutaneous masses may form, as in our patient's case. It was fortunate that their history and presentation with "Inguinal-Scar Endometriosis" was easily distinguishable. Scar endometriosis may present with unambiguous clinical signs upon physical examination which facilitates an accurate work-up and diagnosis. Inguinal endometriosis may present in various manners and when not in the subcutaneous form, this clinically ambiguous lesion may be more difficult to correctly diagnose. Moreover, since inguinal endometriosis presents with similar pain patterns seen in other diseases, the condition becomes increasingly difficult to narrow on a differential diagnosis. Our patient did not present with the typical right laterality and if not for their past surgical history, palpable mass, and left groin pain along their surgical scar, our arrival at their diagnosis may have been delayed.

Scar and inguinal endometriosis demonstrate that surgical procedures may inadvertently cause seeding of endometrial cells. These situations are complex, especially because of the uniquely adherent and proliferative nature of endometrial cells. Our patient's mass was adherent to abdominal wall fascia and rectus abdominis muscle, and histologically presented as endometrial glands and stroma in a background of connective and adipose tissue, and blood vessels. The topographical nature of these masses demonstrates need for improved surgical techniques and the need for new surgical field cleansing procedures in gynecological operations. Appropriate strategies are recommended for development and utilization for in and around surgical fields during gynecologic abdominopelvic operations to prevent seeding. A separate, sterile set of tools are suggested to be utilized during the closing process, as well as, thorough irrigation and suction of the surgical field prior to approximation and closure. Our goal to improve our patient's quality of life and liberate them from their suffering is onerous if we do not significantly minimize the possibility for recurrence. Furthermore, we recommend that further research be performed pertaining to iatrogenic causes of scar and inguinal endometriosis.

REFERENCES

- 1. Fujikawa H, Y Uehara. Inguinal endometriosis: An unusual cause of groin pain. Balkan Med J.2020;37:291-292.
- Zihni I, Karaköse O, Özçelik KC, Pülat H, Eroğlu H, Bozkurt KK. Endometriosis within the inguinal hernia sac. Turk J Surg. 2020;36:113-116.

- Hirata T, Koga K, Osuga Y. Extra-pelvic endometriosis: A review. Reprod Med Biol.2020;19:323-333.
- Mu B, Zhang ZQ, Liu C, Zhang K, Li SH, Leng JH, et al. Long term follow-up of inguinal endometriosis. BMC Womens Health. 2021;21:90.
- Azhar E, Mohammadi SM, Ahmed FM, Waheed A. Extrapelvic endometrioma presenting as acute incarcerated right inguinal hernia in a postpartum patient. BMJ Case Rep.2019;12.
- 6. Basnayake O, Jayarajah U, Seneviratne SA. Endometriosis of the inguinal canal mimicking a hydrocele of the canal of nuck. Case Rep Surg.2020:8849317.
- Longras C, Braga SF, Carrilho C, Mesquita A. Endometriosis unusual aetiology of inguinal swelling. Rev Port Cir Cardiotorac Vasc.2020;27:237.
- 8. Nagama T, Kakudo N, Fukui M, Yamauchi T, Mitsui T, Kusumoto K. Heterotopic endometriosis in the inguinal region: A case report and literature review. Eplasty 19.2019:ic19.
- Nagase S, Ogura K, Ashizawa K, Sakaguchi A, Wada R, Matsumoto T. Hydrocele of the canal of nuck with endometriosis: Right- side dominance confirmed by literature review and statistical analysis. Case Rep Pathol.2020:2567267.
- Prodromidou A, Pandraklakis A, Rodolakis A, Thomakos N. Endometriosis of the canal of nuck: A Systematic Review of the Literature. Diagnostics.2021;11:p3.
- Swatesutipun V, Srikuea K, Wakhanrittee J, and Thamwongskul C. Endometriosis in the canal of nuck presenting with suprapubic pain: A case report and literature review. Urol Case Rep.2021;34:101497.