

Case Report

Scar Endometriosis with Rudimentary Horn: An Unusual and Elucidative Report of a Case Diagnosed on Histopathology and Immunohistochemistry

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ABSTRACT

Endometriosis is defined as the presence of functioning endometrial tissue outside the endometrial cavity. Scar endometriosis, also known as spontaneous abdominal wall endometriosis, is an unusual clinical presentation which often goes unnoticed. It usually develops after pelvic operations. The incidence has been estimated to be only 0.03%–0.15% of all cases of endometriosis. It can be either asymptomatic or present as abdominal wall pain at the site of surgical incision. It is most commonly diagnosed clinically or on ultrasonography. The treatment of choice predominantly remains surgical excision. We present a case of a 24-year-old female (known case of bicornuate uterus) who presented with chief complaints of abdominal pain for 1 month and 6 months after metroplasty. The patient was clinically diagnosed as a case of scar endometriosis with rudimentary horn and fistulous tract and taken up for surgery. Both the scar tissue and fistulous tract were removed and histopathology revealed only endometrial glands without stroma or hemosiderin-laden macrophages. Diagnosis of scar endometriosis was established on positive immunohistochemistry for estrogen and progesterone receptor in endometrial glands. Timely diagnosis and surgical excision of scar endometriosis along with close follow-up are necessary to prevent complications and recurrence.

KEYWORDS: *Bicornuate uterus, endometriosis, fistulous tract, rudimentary horn, scar endometriosis*

INTRODUCTION

Endometrial tissue growing outside the uterine cavity is known as endometriosis, which was first established by von Rokitansky.^[1] The most commonly involved site for endometriosis is pelvis besides which it is also found in bladder, kidney, bowel, omentum, lymph node, lungs, pleura, extremities, umbilicus, hernial sacs, and abdominal wall.^[2] The clinical presentation varies from asymptomatic to pain, dysmenorrhea, dyspareunia, infertility, fatigue, and painful micturition.^[3] It affects 10%–15% women of reproductive age group; scar or incisional endometriosis is known for its rarity as it affects <1% of these women. Diagnosing scar endometriosis is difficult leading to unnecessary procedures, delay, and misdiagnosis. The present case report is pertaining to a 24-year-old woman presenting to the gynecology outpatient department (OPD) with

chief complaints of abdominal pain after metroplasty procedure.

CASE REPORT

A 24-year-old woman (known case of bicornuate uterus) presented to the gynecology OPD with chief complaints of abdominal pain for 1 month. She was a known case of primary infertility with bicornuate uterus for which underwent metroplasty 6 months ago. Ultrasound findings of abdomen and pelvis suggested a diagnosis of bicornuate uterus with rudimentary horn. Magnetic resonance imaging (MRI) pelvis gave an impression of

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unicornuate uterus with noncommunicating rudimentary horn on the right side and ill-marginated T2-hypointense abdominal wall (rectus sheath) lesion at scar site more on the left side with further deep subserosal extension, likely scar endometriosis, mild left-sided hydrosalpinx with left ovarian stranding, likely inflammatory. Furthermore, contrast-enhanced computed tomography (intravenous contrast) confirmed the diagnosis as well. In addition, a hysterosalpingography was performed using 20 ml of urografin 76%, under fluoroscopic control which revealed only minimal spill in left fallopian tube.

On the present OPD visit, general examination was within normal limits. Per abdominal examination revealed a brownish mass measuring approximately 2 cm × 2 cm on the left side of metroplasty scar with slight tenderness, firm consistency, and restricted mobility. A provisional diagnosis of scar endometriosis with rudimentary horn was made and surgical excision was planned without any delay. Laparotomy was performed; intraoperative findings revealed dense adhesions between rectus sheath and peritoneal structures. Also seen was a fistulous tract with one end on left side of incision site and other end arising from metroplasty scar between two horns of uterus.

For histopathological examination, we received, a partially skin covered multiple gray-brown soft-tissue pieces together measuring 0.6 cm × 0.3 cm × 0.2 cm which was entirely processed. Also received another partly skin covered gray-white to gray-brown, irregular soft-tissue piece altogether measuring 6 cm × 4 cm × 1 cm which on cutting revealed a sinus tract measuring 2 cm in length. Microscopic sections from the scar tissue showed multiple tissue bits lined by epidermis. The underlying subepithelial tissues showed few glands lined by cuboidal to low columnar lining [Figure 1]. However, no

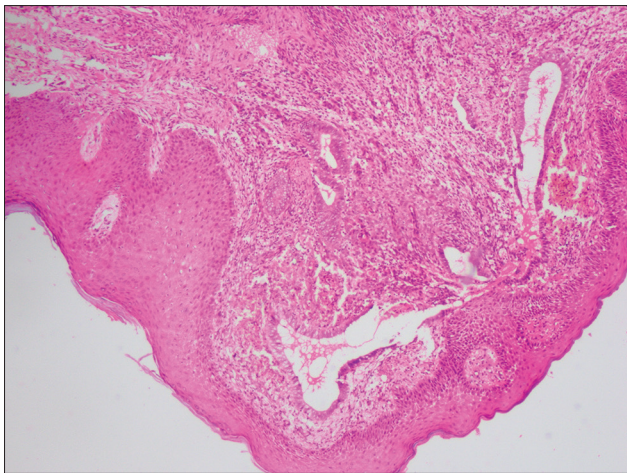


Figure 1: Low-power microscopic view of endometrial glands lined by cuboidal to low columnar lining

hemosiderin-laden macrophages or stroma were seen. This resulted in a pathological dilemma as in the absence of at least two of the three pathognomonic features (endometrial glands, endometrial stroma, and hemosiderin-laden macrophages), it was difficult to establish the diagnosis of endometriosis. Therefore to confirm the diagnosis, immunostaining for estrogen and progesterone receptors was done both of which came out to be positive in the glands [Figure 2]. This aided in arriving at a final diagnosis of scar endometriosis. Sections from the fistula revealed a tract lined by chronic nonspecific inflammatory granulation tissue with extensive foreign-body giant cell reaction, compatible with a fistulous tract.

DISCUSSION

Cases of scar endometriosis are primarily asymptomatic though, when presenting as a painful swelling, they resemble surgical lesions such as hernias, hematomas, granulomas, abscess, and tumors. Therefore, these patients generally first present to general surgeons rather than gynecologists. Scar endometriosis is a rare entity reported in the gynecological literature and presents in women who have undergone a previous abdominal or pelvic operation.^[4] The incidence has been estimated to be only 0.03%–0.15% of all cases of endometriosis.^[5] It most commonly occurs after an operation on the uterus and tubes. The most widely accepted theory in support of scar endometriosis is the iatrogenic transplantation of endometrial implants to the wound edge during an abdominal or pelvic surgery.^[5-7] Endometriosis is an important clinical entity seen in approximately 8%–15% of menstruating females, which often produces symptoms such as pelvic pain, dysmenorrhea, and also infertility in some cases.^[8]

In common scenarios, scar endometriosis is a consequence of previous abdominal or pelvic operations – cesarean

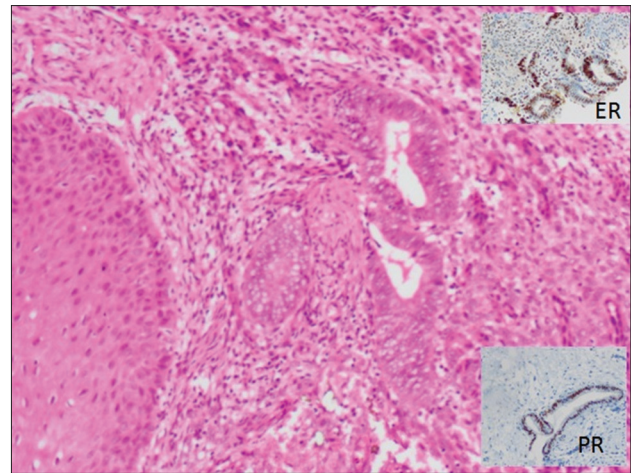


Figure 2: High-power microscopic view of endometrial glands, insets: Positive immunohistochemistry for estrogen and progesterone receptor

sections, hysterectomies and appendectomies, to name a few, also seen as a sequelae to amniocentesis, episiotomy, and surgeries on fallopian tube.^[9] Isolated abdominal wall endometriosis has been reported as rarely as up to 4%; however, associated pelvic endometriosis was seen in 26% cases.^[3,10] No associated pelvic endometriosis was noted in our case.

Scar endometriosis needs to be assessed well, examined thoroughly, and treated promptly keeping in mind the consequences if left unattended. It has been more than often misdiagnosed as stitch granuloma, inguinal hernia, incisional hernia, Spigelian hernia, lipoma, organized abscess, desmoids tumor, neuroma, sarcoma, lymphoma, and occasionally as primary or metastatic cancer.^[11]

There is no set time interval between the surgery and onset of symptoms. In our case, duration between metroplasty and onset of pain and firmness in abdomen were 6 months.

In symptomatic cases, pain gets aggravated during menstruation with an increase in the size of lump/nodule. Cyclic nature of various symptoms during menstruation is pathognomonic of scar endometriosis though it is not seen characteristically in every case.^[12] Keeping in mind the frequency of misdiagnosis, severity of the condition, and complications, if left untreated a detailed clinical history of lump or nodule, its variation with cyclic menstruation and past surgical and gynecological history is a crucial tool in diagnosing scar endometriosis, to keep misdiagnosing episodes at bay. In our case, a fistulous tract was seen on one end, i.e., left side of incision site and another end, i.e., the metroplasty scar between two horns of the uterus. Pain was unrelated to menstruation.

Doppler sonography holds prime importance as a gold standard among various available radiological diagnostic modalities such as ultrasonography of abdomen and pelvis, computed tomography, and MRI. Due to the affordability and easy accessibility, ultrasonography remains one of the first investigations.

Histopathological examination of excised lump or nodule is the most reliable method to establish diagnosis. Grossly, endometriosis may present as small, dark red, black, or bluish cysts or nodules on the surface of peritoneal and pelvic organs. For histopathological confirmation of scar endometriosis, the presence of at least two of the following three microscopic findings are required – endometrial glands, stromal cells, and hemosiderin laden macrophages.^[13] Our case presented a diagnostic challenge as only endometrial glands were observed without any stroma or hemosiderin-laden macrophages; therefore, immunostaining for hormone receptors had to be done to confirm a diagnosis of scar

endometrium. In cases where sometimes hemorrhage, foamy cells, and hemosiderin-laden macrophages are seen obscuring the morphology of gland and stroma might be a tricky situation to diagnose. In such situations, a clinical correlation is of great importance.^[14]

Treatment of choice is wide excision of the lesion. Role of progestogens, oral contraceptive pills, and danazol is debatable and gives symptomatic relief. The benefits of gonadotropin have been found to be of limited relief in symptoms moreover it does not bring about much change in the size of lump/nodule.^[15] Owing to the recurrence, patients need to be kept under close follow-up. In cases of continual recurrence, the possibility of malignancy should be kept in mind. It has also been recommended that before closure, vigorous irrigation with high jet solution should be done on the abdominal wall wound.^[16]

CONCLUSION

A woman presenting to the OPD, essentially postgynecological/obstetrical surgery, with chief complaints of pain/swelling anywhere in the abdominal region should raise suspicion of scar endometriosis. All differential diagnosis should be ruled out effectively to prevent any chance of misdiagnosis. Through this case, we wish to highlight that immunohistochemistry supplements histopathology in diagnosis of endometriosis, especially in cases where only one of the three characteristic microscopic features is seen.

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Conflicts of interest

There are no conflicts of interest.

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