



Case series

The challenge in diagnosing abdominal wall endometriosis; a series of three cases

Mohammad S. El Muhtaseb^a, Hebah T. Daradkeh^a, Mohammed A. Sunoqrot^{a,*},
Naser Al-Husban^b

^a Department of General Surgery, School of Medicine, The University of Jordan, Amman, Jordan

^b Department of Obstetrics and Gynecology, School of Medicine, The University of Jordan, Amman, Jordan

ARTICLE INFO

Keywords:

Endometriosis
Endometrioma
Abdominal wall
Case report

ABSTRACT

Introduction: Abdominal wall endometriosis (AWE) is an understudied entity in which many women of child-bearing age who have undergone pelvic obstetric surgeries suffer. In this series, we will present three cases of AWE and discuss the diagnostic challenge in this uncommon disease.

Presentation of cases: These case series describe the different presentations of the entity and the various methods of diagnosing them. Many other reviews have discussed the possible preventative methods to decrease the chances of developing abdominal wall endometrioma.

Discussion: Women of childbearing age, with a previous C-section or hysterectomy, who present with chronic abdominal pain and bulging related to their wound should raise the suspicion and further go an abdomen pelvic CT scan at the time of menstruation. Diagnosis of abdominal wall endometriomas is usually delayed, as symptoms are non-specific, cyclical, and slowly progressive. After imaging, the patient must undergo surgical excisional biopsy to have a definitive diagnosis.

Conclusion: Abdominal wall endometriomas are only visualized on a CT scan during menstruation, increasing the challenge of diagnosing this entity significantly. To minimize the error in diagnosis, a raised suspicion of this pathology is the key to identifying this issue.

1. Introduction

Endometriosis is defined as the ectopic implantation of stromal and glandular uterine tissue outside the uterine cavity, which could be present adherent to any organ, such as ovaries, sigmoid colon, appendix, and other adjacent or remote organs [1,2].

Abdominal wall endometriosis (AWE) is a unique condition in the list of differentials of women with chronic pelvic pain, especially at a childbearing age, and with a history of gynecological surgeries done, mostly cesarean sections and hysterectomies. A previous diagnosis of endometriosis could aid in the diagnosis; however, cases are usually misdiagnosed with abdominal wall lipomas, sarcomas, and incisional hernias. A history of a mass that changes in size, shape, and pain with cycle should immediately raise the suspicion of this rare pathology.

This case series presents three AWE cases managed in a general surgery department. The cases showed a variety in presentations and were challenging to be diagnosed in a timely manner, which added

much labor and suffering upon the patients.

This Work has been done in accordance with the PROCESS Guidelines 2020 [3].

2. Presentation of cases

2.1. Case 1

A 31-year-old female, not known to have any medical conditions, with a history of an open appendectomy and two cesarean sections, presented to the clinic complaining of a right inguinal bulge that is persistent; increasing in size, pain and discomfort only with her menses, improves immediately with the resolution of the cycle. Her condition started four months after her last cesarean section and lasted two years before she presented to the general surgery clinic. The patient had regular cycles, and she denied the use of intrauterine birth control devices or any previous similar complaints.

* Corresponding author at: The University of Jordan, 11942 Amman, Jordan.

E-mail address: mohammad-si@hotmail.com (M.A. Sunoqrot).

<https://doi.org/10.1016/j.ijscr.2022.106787>

Received 10 December 2021; Received in revised form 15 January 2022; Accepted 20 January 2022

Available online 24 January 2022

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On abdominal physical examination, a well-formed mass was felt just superior to the right inguinal ligament measuring 3 * 3 cm; the mass was immobile when the abdominal muscles were put in action, suggesting infiltration of the underlying musculoaponeurotic structures. Tenderness was noted around the time of menstruation, but soon after, the mass became non-tender. The serological workup was unremarkable; a contrasted CT scan was done at the end of her menstrual cycle and showed a hyperdense lesion at the edge of the wound (as shown in the image), suggestive of a soft tissue lesion, which could be a sarcoma, lipoma or endometrioma (Fig. 1).

The patient underwent surgical exploration to find a lesion attached to the Scarpa's fascia but not to the abdominal wall muscles, which was easily excised with a safety margin. Postoperatively, the patient had a smooth recovery without any surgical site complications, her previous symptoms were relieved, and she was able to resume her daily activities immediately after the recovery. She did not receive any hormonal or chemo-hormonal therapy after the procedure, and she was followed up every six months in the clinic. Histopathological exam revealed endometrial gland and stroma, suggestive of endometriosis with mucinous metaplasia (myxoid change), negative for malignancy.

2.2. Case 2

A 35-year-old female medically free, underwent two C-sections last one in 2012. Soon after her last surgery patient started complaining of surgical site pain and discomfort, but she was reassured several times that it was a normal postoperative recovery. However, the pain began to have a cyclical pattern with the menstrual cycle. A mass was felt at the

edge of the wound, which has increased in size during menses and decreased soon after. The patient denied discharge or ulcerations at the site of the mass. She had no alteration in her bowel habits, or bladder disturbance, but the patient was known to have some menstrual disturbances and irregularities; however, she was not diagnosed with endometriosis beforehand.

On her physical examination, a well-defined mass was felt at the edge of her wound, along with being mobile even with the contraction of the abdominal wall muscles suggesting no attachments to the underlying muscles.

Her serological workup was within normal range; no imaging was done in this case as the mass was superficial and had no obvious attachments. The differential diagnosis of the mass was most likely an abdominal wall lipoma.

The patient underwent complete surgical excision of the mass. It was a 5 * 5 cm mass with clear margins and well circumscribed. No mesh was inserted as there was no defect in the external oblique or recti aponeurosis. Postoperatively the patient was doing well with no postoperative complications of seroma or surgical site infection, and she was able to resume her daily activities shortly after her recovery.

After excision, the histopathology report showed a mass measuring 5 * 6 cm with multi hemorrhagic foci measuring 2 * 2 * 2.5 cm, containing endometrial glands and stroma confirming endometriosis negative for malignancy.

Her case was discussed in a multidisciplinary meeting with the gynecology department, and she was started on oral combined pills to regulate her menstrual cycle and control any upcoming similar implantations. The patient was followed up in our clinic and the

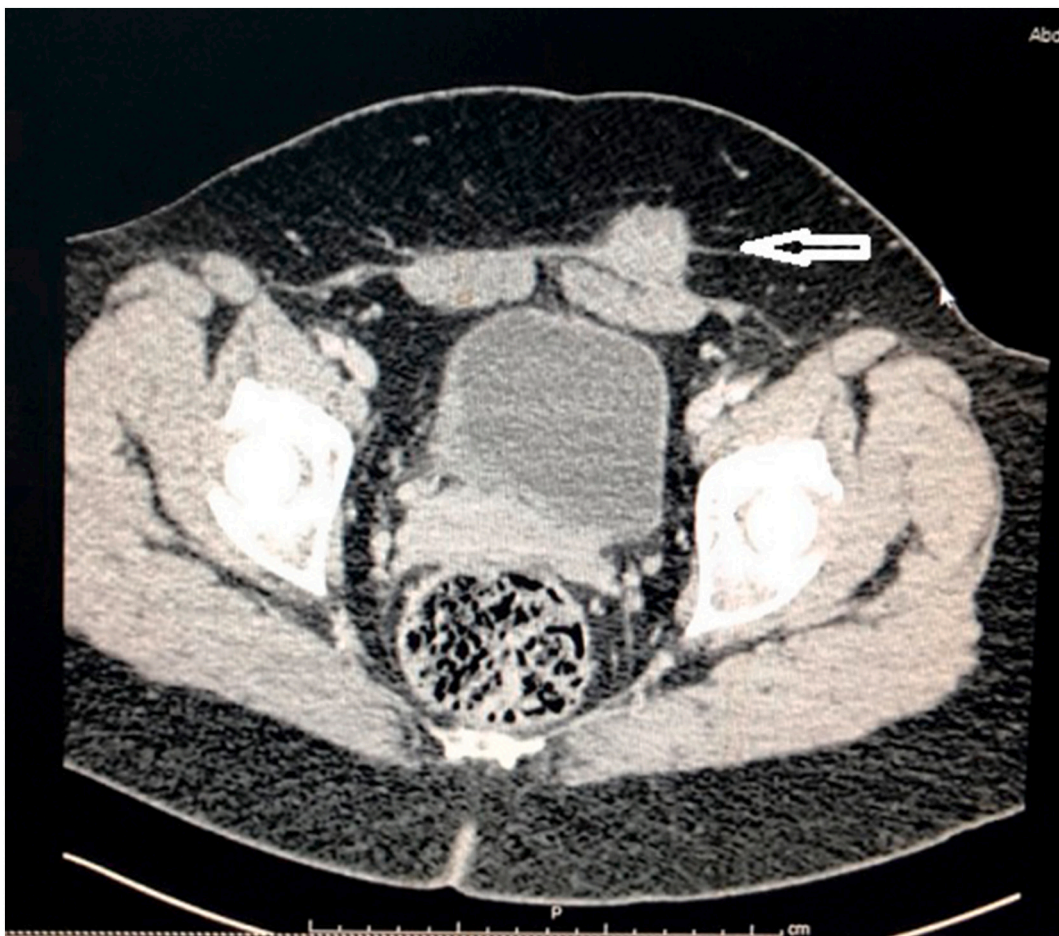


Fig. 1. The image shows an abdomen pelvis contrasted CT scan, white arrow points to the endometrial tissue implanted in the abdominal wall close to a previous C-section scar.

gynecology clinic every six months, where she did not show any recurrence of symptoms or complaints.

2.3. Case 3

A 28-year-old female presented to the general surgery clinic complaining of episodic abdominal pain and a non-healing ulcer close to the edge of a previous C-section scar done one year earlier. The ulcer was discharging clear fluid during the month, but at menses, the discharge changes into thick fluid with streaks of blood, resolving immediately soon after the menstruation period. The patient had no previous medical conditions or surgical history other than two previous C-sections and PCOS.

Upon examinations of the patient, a Pfannenstiel incision was noted at her lower abdomen, with a sinus inferior to it discharging clear fluid. There were no masses felt at the examination, and she had no abdominal distention or other GI or urinary disturbances. An abdominal CT scan was done mid-cycle, which did not show a mass; however, the tract and the sinus were closely related to the edge of the C-section scar, suggesting an involvement of the scar and raising the suspicion of abdominal wall endometrial tissue implantation.

We booked the patient for an elective excision of the sinus and tract at the beginning of her menses, looking for a mass or other changes that might aid in treating the condition. Intraoperatively, the sinus along its tract was excised, including some of the fibrous tissue at the edge of the scar giving a clear safety margin of 1 cm; primary closure of the wound was done, and the excised sample was sent to histopathology examination.

A week later patient presented to the clinic with no postoperative complications, good wound healing, and a complete resolution of her previous symptoms of discomfort. The patient was followed up in the gynecology clinic and was started on oral combined contraceptive pills for six months to treat her PCOS, and she was advised to reduce her weight by at least 10% in an attempt to regulate her cycles. Histopathology report showed endometrial tissue imbedded within the fibrous tissue sized 1 cm * 1.8 cm with clear margins, confirming the diagnosis of abdominal wall endometriosis.

3. Discussion

One of the most common complaints we came across in the general surgery department is the unspecific abdominal pain adding it to the complex anatomy of the female, sometimes leading to very challenging differentials and a handful of investigations to treat the patient. We came across a rare entity of cyclical abdominal pain throughout these years, with a mass or swelling that increases during the menstrual cycle and subsides soon after. Many patients were misdiagnosed and treated for other pathologies until the mass resection proved to be abdominal wall endometriosis; a very misleading condition in which the unfortunate female has to withstand agony for an extended period of time before she is correctly diagnosed, but until then, her countless visits to the ER, general practitioners, and gynecologists put her with a frustrating chronic pain.

Three cases were referred to the clinic with a long history of abdominal swelling and pain at the site of previous cesarean section, both of which emphasize with menstruation and improve soon after. Symptoms have lasted months to years, affecting the patients' lifestyle and daily activities. After multiple tests and visits to the ER, gynecologists, and general practitioners, two of the cases were mistakenly diagnosed as incisional hernia and were referred to follow-up. Their main symptoms were swelling and pain that increased at the time of the menstrual period and improved soon after; even one of the patients developed cyclical bleeding ulceration at the site of the swelling. Patients were operated on, and an excisional biopsy was done in all cases. The lesions' size ranged from 3 to 6 cm and were all found close to the cesarean section scar, which has raised the suspicion of abdominal wall

endometriosis even further. The wounds were closed primarily without complication, and the excised samples were sent to histopathology for evaluation and confirmation. Soon after, a histopathological examination of the excised swelling reported endometrial glands and stroma with foci of hemorrhage.

Endometriosis is defined as the ectopic implantation of stromal and glandular uterine tissue outside the uterine cavity, which could be present adherent to any organ, such as ovaries, sigmoid colon, appendix, and pleural cavity [1,2]. Endometriomas tend to occur near the site of a previous surgery violating the uterine cavity, such as a C-section or a hysterectomy, increasing the likelihood of implantation [4]. Regarding abdominal wall endometriosis, most patients present with abdominal pain, bulging, and skin irritation that is usually cyclical along their menstrual cycle. The pathophysiology of this entity was determined to be under investigation. However, multiple theories have been suggested; the retrograde reflux of the endometrial tissue into the peritoneal cavity through the fallopian tubes has been the most prominent one [5].

The incidence rate of implantation of endometrial tissue into a surgical scar is 0.03–1.7% and is increasing since the rate of C-sections is increasing exponentially [6], leading to the investigation behind the risk factors and the possible prevention methods that could be implemented intraoperatively. Many methods have been discussed to decrease the risk, such as wound protectors, rigorous irrigation of the wound, and careful removal of the instruments such as sponges used in the uterine cavity [7].

Various imaging techniques such as ultrasound, contrasted CT scan, and MRI could be sensitive, and they have been used to diagnose most of the cases, but the excisional biopsy seems to be the most specific one [8]. A CT scan was requested for each patient to be done only at the time of menses, which was able to confirm the presence of a mass within the wall of the abdomen without the presence of an incisional hernia, raising the suspicion of abdominal wall implantation of endometrial tissue. Diagnosis requires a high index of suspicion as patients often are juggled between gynecologists and surgeons before this pathology is even thought off and spotted. Their symptoms are cyclical and progress slowly, in addition to the fact that the mass shows well only during menstruation and hence visualized on a CT scan, increasing the challenge of diagnosing this entity significantly.

Patients may be offered the option of conservative management with a trial of progesterone, oral contraceptive pills, and danazol, however soon after stopping the medicine risk of recurrence increase (recurrence rate 1.5–9.1%), so the benefit will solely be with symptomatic relief. Hence, patients who choose to be on hormonal therapy must be closely monitored for recurrence or incases of incessant cutaneous manifestations, malignancy must be ruled out [9,10].

4. Conclusion

This entity needs to be considered a cause of any cases with a painful swelling after cesarean section before a diagnosis of incisional hernia is made. With a raised suspicion, a CT scan is better performed at the time of menses to help with the diagnosis to minimize the error in diagnosis, and save the patients the suffering. More future studies and investigations must focus on emphasizing the causes and modifying the risk factors that may have potentiated the patients to develop this underacknowledged entity.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Ethical approval

The study is exempt from ethical approval in our institution.

Consent

Written informed consent was obtained from the patients for publication of this case series and accompanying image.

CRediT authorship contribution statement

Mohammad S. Elmuhtaseb: Conceptualization, methodology and validation. Hebah T. Daradkeh: Investigation and Writing - Original Draft. Mohammed A. Sunoqrot: Writing - Review & Editing. Naser Al-Husban: Supervision.

Registration of research studies

N/a.

Guarantor

M.S Elmuhtaseb.

Declaration of competing interest

None.

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