

Endometriosis in a cesarean section scar: A series of 12 patients

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Abstract: Objective: Endometriosis is the presence of functioning endometrial tissue outside the uterine cavity. Both pelvic and extra pelvic endometriosis has been described. Extra pelvic endometriosis can occur in abdominal wall following obstetrical and gynecological surgery. Cesarean section scars are the most common sites of extra-pelvic endometriosis. It is mostly confused with other surgical conditions and should be considered in the differential diagnosis of lumps in the abdomen in females in reproductive age. The aim of this article is to increase the awareness regarding this rare condition and combine assessment using clinical, radiological and pathological findings to solve this diagnostic dilemma of abdominal wall endometriosis which is a frequently misdiagnosed condition. **Method:** We reviewed the case records of patients with the diagnosis of scar endometriosis seen in our hospitals from January 2010 to June 2015. We discuss the cases of the abdominal wall endometriosis following caesarian section. The incidence, pathophysiology, diagnosis, treatment and prevention of this condition are also reviewed. **Result:** Twelve patients of scar endometriosis were operated during the study period. The mean age of the patients was 31 years (range 22–46 years) and mean interval from previous surgery to onset of symptoms was 2.4 years (range 1/2 -5 years). Painful swelling at local site was the most common presenting symptoms. Ultrasound, CT scan and MRI examinations were important in the precise location of the disease. FNAC is a good tool in diagnosis of this condition. 10 patients underwent wide excision of the mass, while re-excision was needed in 2 patients. The patients progressed satisfactorily and histopathology confirmed the diagnosis of abdominal wall endometriosis in all cases. No recurrence was occurred at a follow up ranging from 6 months to 1 years. **Conclusion:** Abdominal scar endometriosis should be kept in mind while dealing with a case of the painful lump over the abdominal scar in any woman of childbearing age having a history of obstetric or gynecological procedure. Increasing awareness of this condition among doctors can help in early diagnosis and treatment with gratifying results.

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Keywords: endometriosis, abdominal wall endometriosis, scar endometriosis

1. Introduction:

Endometriosis is a rare condition and was first described by an Austrian pathologist, von Rokitansky in 1860[1]. It is defined as the presence of ectopic functional endometrial tissue outside the uterine cavity.

The most common locations occurs in the pelvic sites such as the ovaries, posterior cul-de-sac, uterine ligaments, pelvic peritoneum, bowel, and recto-vaginal septum. Extra pelvic endometriosis is a fairly uncommon disorder and can be found in unusual places like in the nervous system, thorax, urinary tract, gastrointestinal tract, and in cutaneous tissues unless its most frequent location is the abdominal wall [2].

Abdominal wall endometriomas often develop in previous surgical scars but there are case reports of

involvement of the rectus abdominis muscle in a virgin abdomen [3].

Many theories have been postulated regarding scar endometriosis, one of which is the direct implantation of the endometrial tissue over the wound edge during abdominal and pelvic surgery [4]. These cells may proliferate under cyclical hormonal stimulation in reproductive women and also cause metaplastic changes in the surrounding tissue and develop scar endometriosis (cellular transport and coelomic metaplasia theory). Another theory is the transfer of endometrial cell to surgical scar via lymphatic or vascular way and ultimately generates scar endometriosis [5].

Scar endometriosis patients are often referred to the general surgeons because the clinical presentation

suggests a surgical cause. Differential diagnosis includes abscess, lipoma, hematoma, sebaceous cyst, suture granuloma, inguinal hernia, incisional hernia, desmoid tumor, sarcoma, lymphoma and primary or metastatic cancer. The development of the endometriosis on a surgical scar may have a very late onset after the surgery, which often predisposes to incorrect diagnosis and inadequate surgery [6].

2. Methods:

The study was carried out at three private hospitals in Jeddah, KSA. In the period between January 2010 to June 2015, we retrospectively reviewed 12 consecutive cases who underwent surgery for pathologically confirmed endometriosis of the abdominal wall postoperatively.

Sonographic examination was performed in 7 patients using L5-12 MHz linear-array transducers (iU22 MATRIX, Philips Ultrasound machine). Power Doppler sonography was used to assess the vascularity of the lesions. In addition to sonography, a CT examination that included IV contrast material was performed in 2 patients on an MDCT scanner (Philips Brilliance 6) axial cuts taken with 2 mm slice

thickness, 4.0×2.5 mm collimation at a table feed of 12 mm/rotation then followed by sagittal and coronal reconstruction. Two other patients also underwent MRI on a 0.3-Tesla scanner (Hitachiapertolucent); those examinations included spin-echo T1-, fast spin-echo fat-saturated T2, and fast spin-echo fat-saturated gadolinium-enhanced T1-weighted sequences and were performed with the patient in the supine position. Fine needle aspiration cytology (FNAC) was done in 2 patients.

Patient's demographic characteristics, clinical presentation, radiologic imaging findings, surgical treatment, and histopathology of surgical specimens were recorded and analyzed.

The ethical approval was obtained from our hospitals review board, and all patients were informed and agreed to participate in our research.

3. Results:

Over a period of 5 years, the data of 12 patients who underwent surgery for abdominal wall endometriosis were entered in this study. The demographic features of the patients are given in Table 1.

Table (1): Characteristics of the patients with abdominal wall endometriosis

	Age	Complaint	Period	Size	Investigation	Preoperative diagnosis	Treatment
1	33	Painful swelling	8 months	3 cm	-	Incisional hernia	Excision
2	30	Painful swelling	3 years	2 cm	U/S	Endometriosis	Excision
3	32	Painful swelling	1.5 years	2 cm	U/S	Lipoma	Excision
4	30	pain	6 months	1.8 cm	-	Suture granuloma	Incomplete Excision
5	34	Painful swelling	2 years	3 cm	U/S, CT	Endometriosis	Excision
6	28	Painful swelling	2 years	2.8 x 1.9 cm	U/S, MRI	Endometriosis	Excision
7	31	Swelling	3years	3 cm	-	lipoma	Incomplete Excision
8	24	Painful swelling	1 years	1.4 cm	U/S	Suture granuloma	Excision
9	46	Painful swelling	5 years	4 cm	CT, FNAC	Endometriosis	Excision
10	34	-	3 years	2 cm	-	-	Excision during CS
11	22	Painful swelling	4 years	2 cm	U/S	Endometriosis	Excision
12	29	Painful swelling, severe dysmenorrhea	3 years	6 x 4 cm	U/S, MRI, FNAC	Endometriosis	Excision

The mean age at presentation was 31 years (range 22 - 46 years), all patient had previous caesarean sections.

The main complaint in most cases (9 patients) was painful swelling in the abdominal wall. The pain was cyclic in 5 patients while it was non-cyclic in 4 patients. The complaint of the remaining three cases was as following: one patient had mild pain only at the lateral end of the caesarean section scar. The second one patient had a notable mass as a presenting symptom and the last patient did not have any

symptoms and abdominal wall endometriosis was discovered during the caesarean section.

The masses of the abdominal wall had mean diameter of 2.7 cm (range 1.4 - 6 cm), and localized around the abdominal scar in all cases.

The period between previous surgery and onset of clinical manifestations ranged from 6 months to 5 years with an average of 2.4 years.

The preoperative diagnosis was suspected in 6 cases. Based mainly on the presence of abdominal

mass located in a laparotomy scar worsening with menses and confirmed by investigation.

The other 6 patients were diagnosed with suture granuloma (2 patients), lipoma (2 patients), incisional hernia (1 patient) and incidentally discovered in one patient.

None of our patients had concomitant pelvic endometriosis whether diagnosed pre or post-operatively.

All patients with preoperative correct provisional diagnosis of endometriosis underwent surgical exploration with wide excision of endometriomas,

freeing limits of at least 1 cm. It was necessary to use a polypropylene mesh in the abdominal wall reconstruction in one case (case no 12). The final diagnosis was confirmed by histopathology of surgical specimens, where stroma and ectopic endometrial glands were found. Re-excision was required in 2 patients due to incomplete excision of the first operation.

The postoperative course of all patients was satisfactory, without any complications. All patients had regular follow-up ranging from 6 months to 1 year and there were no recurrences in any patient.

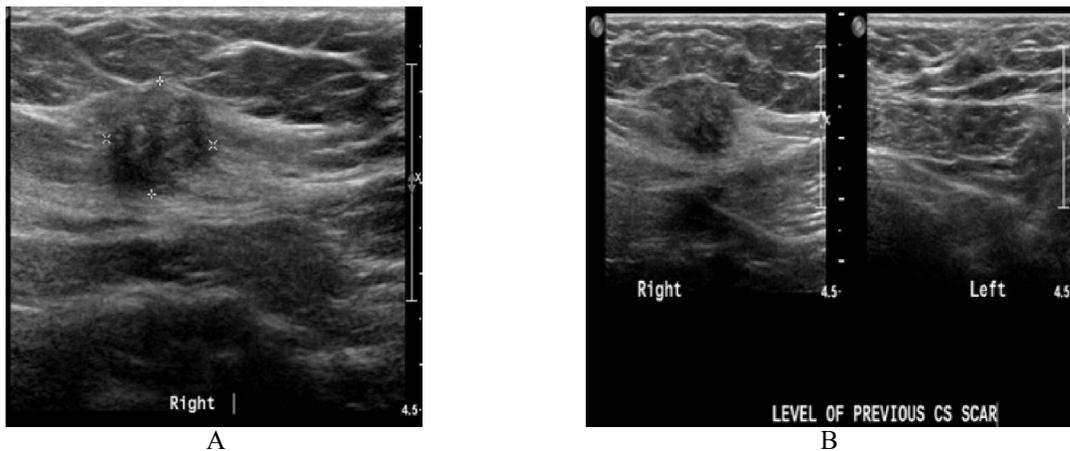


Figure (1): A) Transverse US scan of the right lower anterior abdominal wall in relation to the periphery of the CS scar revealed a fairly defined hypoechoic ovoid subcutaneous lesion. B) Transverse US scan comparison between the right and left lateral aspects of the CS scar.

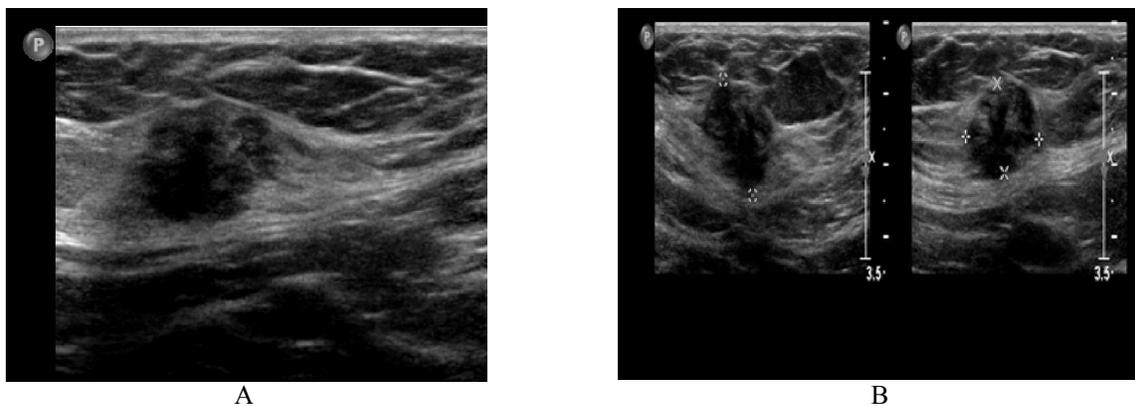


Figure (2): (A&B) follow up Transverse US scans of the lesion during menstruation revealed cyclic increase in the size of the previously seen subcutaneous lesion rising the probability of endometrioma.



Figure (3): Transverse US scan at the level of previous CS scar site shows a non-vascular hypoechoic solid focal lesion having a pine-like appearance seen subcutaneously and resting on the rectus muscle, with spike-like projections

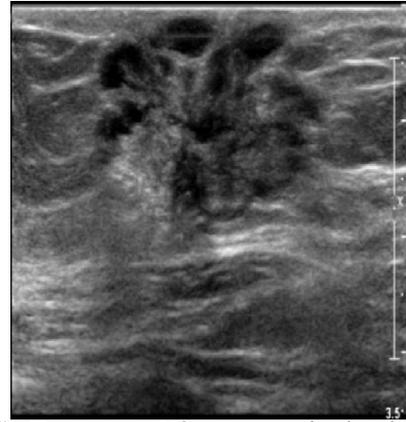
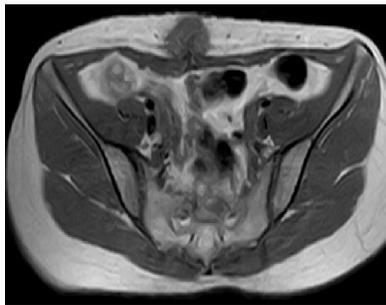


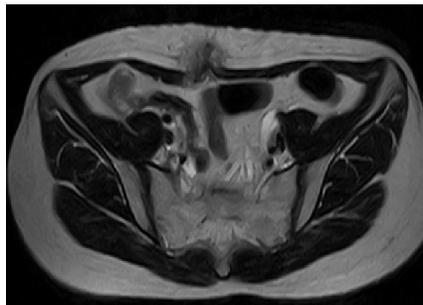
Figure (4): Transverse US scans at the level of the left lateral aspect of the S.C. scar showing a subcutaneous cauliflower echocomplex (with solid with cystic changes).



Figure (5): Trans-axial non contrast CT of the pelvis shows a rather well defined small ovoid relatively hyperdense focal lesion embedded in the deep subcutaneous tissue anterior to the right rectus abdominis muscle (57-58-HU in density).



A



B



C

Figure (6): Pelvic MRI sections at the level of the lesion: A (axial T1), B (axial T2), C (axial T1 post contrast) showing a well-defined irregular soft tissue lesion with spiculated margin seen in the anterior abdominal wall over longitudinal SC scar extending into the deep musculature of the underlying right lateral rectus abdominis muscle with intermediate signal intensity in both T1WI and T2WI and bright in T1W FATSAT & displaying enhancement after contrast. Findings suggestive of abdominal wall endometriosis versus desmoids tumor.

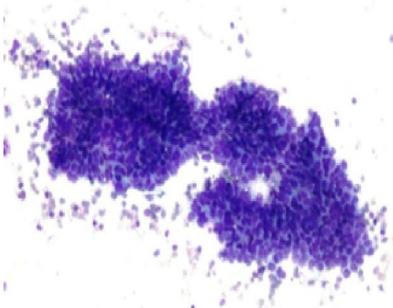


Figure (7): Fine needle aspiration cytology of CS scar nodule showing sheets of epithelial cells and many oval to spindle cells reminiscent of endometrial glandular and stromal cells (Geimsa, x200).

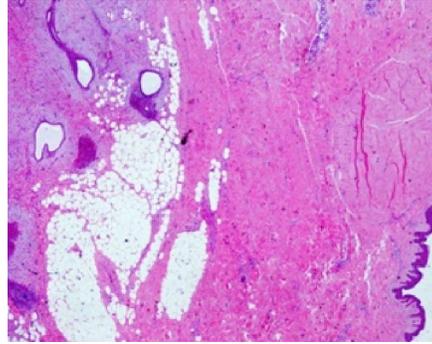


Figure (8) skin and subcutaneous tissue showing dermal cystically dilated endometrial glands and endometrial stroma (H&E, x40)

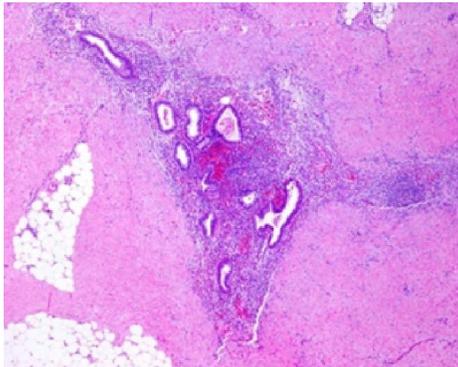


Figure (9) Endometrial glands and stroma with hemorrhage in subcutaneous fat and connective tissue (H&E, x100).

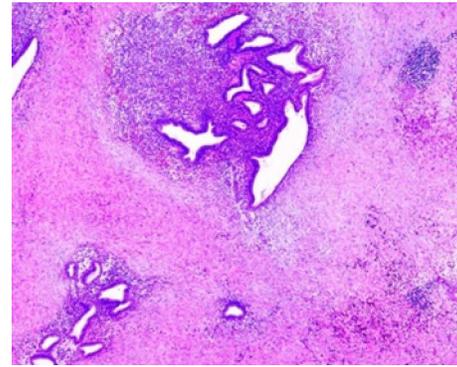


Figure (10) Multiple foci of endometrial glands, stroma and hemosiderin-laden macrophages within dermal connective tissue (H&E, x100).

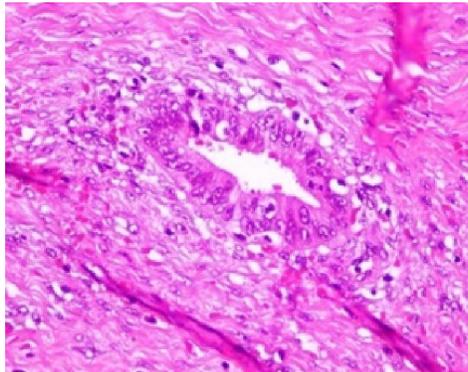


Figure (11) Atypical endometriosis showing atypical glandular component; cells exhibit enlarged vesicular nuclei and prominent nucleoli

4. Discussion:

Abdominal scar endometriosis is a gynecological pathology that often presents to the general surgeon rather than the gynecologist.

Endometriosis has been known for more than 150 years (Rokitansky, 1860) and it usually follows previous abdominal surgery, especially early hysterotomy and cesarean section

Its extra-pelvic location which involving the abdominal wall is much less common than the pelvic one, and should be considered in the differential diagnosis of abdominal wall masses in women of childbearing period [7]. No theory can fully explain the exact mechanism of endometriosis formation, it being likely to result from a combination of events.

Direct mechanical implantation seems to be the most plausible theory for explaining scar endometriosis. During caesarean section, endometrial tissue might be seeded into the wound, and under the same hormonal influences these cells proliferates [8].

Although spillage of endometrial cells into the surgical incision probably occurs quite frequently during gynecological or obstetric surgery, endometriosis occurs with much lower frequency. This implies that factors such as environmental, genetic, immune system, and hormonal abnormalities may confer susceptibility to the development of endometriosis in some people [9]. The fact that all the patients of our study had undergone a caesarean section, may be in favor of the transport theory.

The true incidence of scar endometriosis is difficult to determine. Frequency increases by induced number of caesarean section and laparoscopy performed in recent years [10]. Its incidence after caesarean section is estimated to be in range of 0.07%–0.47% [11].

Endometriosis involving the abdominal wall is an unusual phenomenon and difficult to diagnose. It is often misdiagnosed with other surgical conditions like hematoma, suture granuloma, fibrosis, lipoma, incisional hernia, desmoid tumor, or primary and metastatic malignancies lesions [12].

The usual clinical presentation is a painful swelling in a parous woman with a history of gynecological or obstetrical surgery. The Pain is classically described as cyclic pain but constant and non-cyclic pattern also have reported. Mass sensation, dysmenorrhea and bleeding are among the symptoms of endometriosis [13].

Most of the patients in our study (10 patients) had symptoms of abdominal mass sensation with or without pain (83.3%), Additionally, those 9 patients who had pain, 55% of them experienced cyclic pain (5 patients and one of them had severe dysmenorrhea as associated symptom), and 45% experienced noncyclic pain(4 patients). While one patient presented with pain only at the caesarean section scar, and the last one was discovered during the caesarean section.

The time elapsed between the previous operation and the onset of symptoms is very variable, ranging from 3 months to 10 years in different series [8,14,15]. In our study, the mean interval was 2.4 years.

Preoperative diagnosis of scar endometriosis is difficult to make and sometimes the diagnosis is made after excision only [16] and that what was happened in 6 patients in our study. Various non-invasive diagnostic imaging modalities like Ultrasound, CT and MRI, contribute fundamentally to precise location of the endometriotic lesion and differential diagnosis with other more common diseases, especially with

laparotomy hernias, but they not have a high sensitivity and specificity of certainty [17].

However, Francica et al; suggest that sonographic and color Doppler when combined with clinical data may substantially contribute to the preoperative diagnosis. They reported the diagnostic ultrasonography features of scar endometriosis as (i) a hypoechoic inhomogeneous echo texture with internal scattered hyperechoic echoes; (ii) regular margins, often spiculated, infiltrating the adjacent tissue and (iii) a hyperechoic ring of variable width and continuity. On colour Doppler examination, a single vascular pedicle entering the mass at the periphery is one of the diagnostic feature [18].

In our study we did ultrasonography for 7 patients, five of them (71,4 %) showed features suggestive of endometriosis (fig 1-4). However, 3 patients needed more investigations to confirm diagnosis.

CT scan usually shows a solid, well-circumscribed mass enhanced by contrast (fig 5) and may sometimes show hemorrhage. It also allows study of the whole pelvic cavity that may also be compromised by the disease. However, it may be difficult to distinguish scar endometriosis from bland scarring and other processes at unenhanced and contrast-enhanced CT, thus, correlation of clinical and imaging findings is important [19]. CT scan were done in 2 patients, one of them as further investigation after ultrasonography and the other as primary investigation and followed by FNAC.

MRI can be more helpful when the lesion is small because of its high spatial resolution. Furthermore, it performs better than CT scan in detecting the planes between muscles and abdominal subcutaneous tissue. It also may be used to accurately and safely plan surgical resection, particularly in patients who are symptomatic or have extensive lesions with deep infiltration[20]. In this series MRI were done in 2 patients as further investigation after ultrasonography (fig 6).

Lately many reports have documented the accuracy of FNAC from the mass as an important confirmatory investigation [21,22]. In our series, we could diagnose two patients accurately with this condition by FNAC (fig 7).

The treatment of choice of abdominal wall endometriosis is wide excision with at least 1 cm margin which is diagnostic and therapeutic at the same time [14,23]. We sent all excised specimens for histopathological examination which confirmed presence of endometriosis (fig 8-11).

Mesh repairs may be required as was done in one of our cases to fill large defects in the fascia, and these procedures require adequate counseling and precise surgical planning The hormonal treatment is a

controversial issue in treatment of scar endometriosis. Danazol and, more recently, GNRH analogues (Zoladex,) are used, but the results have not met the expectations, as it has significant side effect, gives only partial relief in symptoms and do not ablate the lesion [24,25]. Because of long treatment course, significant side effect and insignificant results no patient received medical treatment in this series.

Follow up of patients with scar endometriosis is important because of the chances of recurrence, which may require re-excision. Moreover, the lack of correct initial diagnosis, leads to recurrence due to inadequate excision. The renewal of the lesion, making it more extensive and destructive [26]. In spite of the malignant change of endometriosis in a cesarean scar is rare [11], but According to Han et al [27] it cannot be ruled out.

For prophylaxis of endometriosis after cesarean section Wasfie [28] recommends careful isolation of the wall incision and thorough lavage with saline before the closure of the wall. Thus, these measures are good surgical practice although their benefit has not been demonstrated [29].

Conclusion

In conclusion, scar endometriosis is a rare condition and can be confused with other surgical conditions. Correct diagnosis relies on careful examination, right questioning, and obviously taking endometriosis in consideration in all premenopausal women presenting with a painful swelling in the abdominal scar following a previous obstetric or gynecological procedures.

Efforts should be made to make a preoperative diagnosis with the help of imaging techniques and FNAC. The wide surgical excision remains the treatment of choice. Regular follow-up is necessary to detect recurrence.

The limitations of our study are its retrospective nature and the fact that no standardized protocol was predefined in order to scan the patients; this might have led to some lack of uniformity.

Ethical Approval

Ethical approval was provided through the Institutional Ethics Committee.

Conflict of Interests

The authors declare no conflict of interest

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