Third-time recurrence of CS scar endometriosis after one cesarean section: A case report and Review of literature

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Abstract

Background: Recurrent scar endometriosis was thought to be caused by implantation theory; this is a rare case to have recurrent CS scar endometriosis for the third time in different sites of scar of one CS, which could oppose this theory; we are presenting 34 years of patient complaint of recurrent painful swelling related to menses, previously excised twice and proved to be endometriosis by histopathology. Surgical excision of this swelling was done and proved to be endometrioma for the third time.

Conclusion: Implantation theory as a cause of scar endometriosis should be revised as it cannot support the recurrence of endometrioma for a third time.

Keywords: Caesarean section scar; Recurrent endometrioma

Scar endometriosis incidence has been reported to range from 0.03% to 1.7%. The most frequent symptom is cyclical or non-cyclical painful swelling presented in the abdomen at the site of previous obstetric and gynecological operations [1]. The occurrence of scar endometriosis is supported to be caused by the iatrogenic implantation theory. Secure-free margins must be obtained in surgical excision of scar endometriosis to prevent recurrence [2].

CASE PRESENTATION

A 34-year-old patient previously presented complaining of painful lower abdominal swelling at the left side of the CS scar with a typical presentation of endometriosis as it began before menses, increased in size and pain during menses, then disappeared after menses. The patient gave a history that she had one CS 6 years ago and a history of previous CS scar endometriomas twice, both excised and proved by histopathology to be endometriomas. On examination, left tender well-circumscribed swelling 5 x 5 cm in size at the site of scar of previous CS. The patient refused to have MRI as she had no medical insurance stating she had done it twice before, and it proved to be endometriosis. After routine preoperative preparation, we opened the scar and excision two swellings located subcutaneously not related to the rectus sheath (DD of desmoid tumor of rectus sheath); the first was 6 x 6 cm (Fig. 1.) and the second was 4 x 4 cm (Fig. 2.), and care was taken to remove any small swellings, ensuring hemostasis then closure of the wound.

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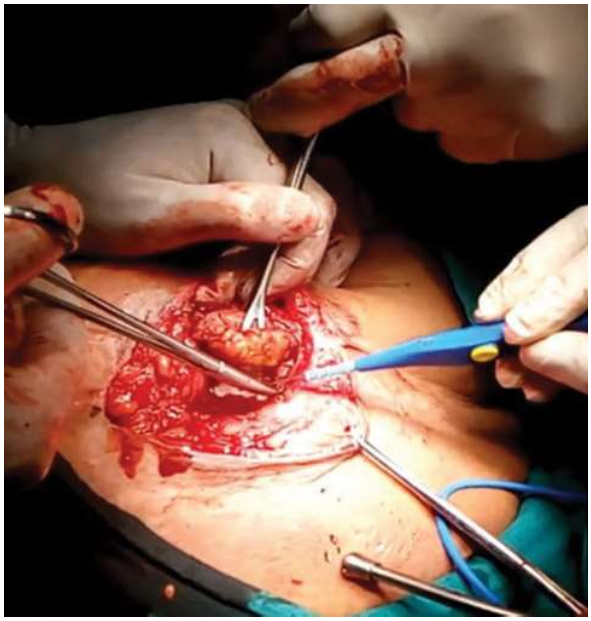

Fig. 1. Dissection of endometriotic mass with good surgical margin.

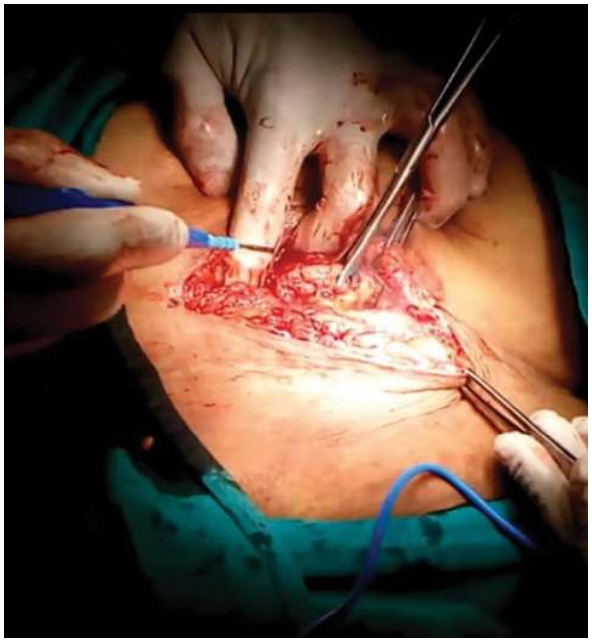

Fig. 2. Dissection of 2nd endometriotic mass with good surgical margin.

Both specimens were sent to histopathology and proved to be external endometriosis with free surgical margins (Fig. 3. is the pathology report to show free surgical margins). The patient follows up till the removal of stitches.

The pathology report revealed:

1. Fibrofatty tissue piece measured 7 x 6 x 5 cm, sectioning revealed circumscribed area, measuring 2.5 x 2.5 cm with a rubbery greyish-pink cut section.
2. Fibrofatty tissue piece measured 4 x 4 x 3 cm; sectioning revealed a circumscribed area, measured 2 x 2 cm with the same cut section as the previous specimen.

Microscopic: Sections examined from the BOTH specimens received revealed fibrofatty tissue showing endometrial glands with columnar non-secreting lining surrounded by stromal cells. There are foci showing excess hemosiderin. There is surrounding excess fatty tissue. Surgical margins are free. No evidence of malignancy in the sections examined (Fig. 4.).

PATHOLOGY REPORT

Gross:

Two undesignated containers were received:

1. Fibrofatty tissue piece measured 7x6x5 cm, sectioning revealed circumsribed area, measured 2.5x2.5 cm a with rubbery graish pink cut section, showing focal reddish areas and cystification.
2. Fibrofatty tissue piece measured 4x4x3 cm, sectioning revealed circumsribed area, measured 2x2 cm having the same cut section of the previous specimen.

Microscopic:

Sections examined from the BOTH specimens received revealed fibrofatty tissue showing
endometrial glands with columnar non-secreting lining surrounded by stromal cells. There are foci showing excess haemosidrin. There is surrounding excess fatty tissue.
Surgical margins are free.
No evidence of malignancy in sections examined.

Fig. 3. Pathology report stating free surgical margins.

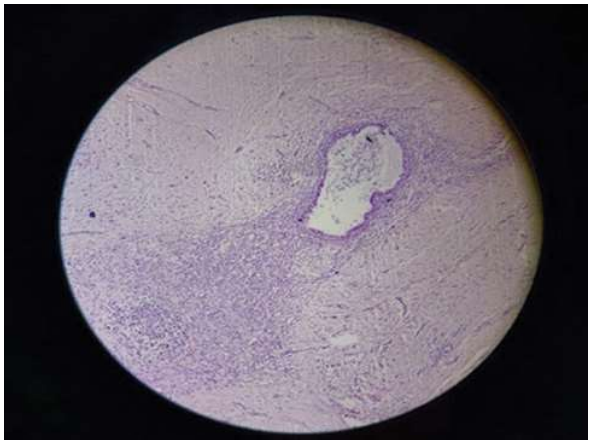

Fig. 4. Microscopic picture.

On contacting the patient to get her consent for publishing the case, she reported recurrence for the fourth time. No one can tell her the cause of recurrence; the cause of recurrence is unknown, especially since the pathology report proved free surgical margins.

Discussion

Cs scars endometriosis, although rare [1], is a distressing condition, especially if recurrent. In a retrospective study done by Yildirim et al. [3] on 29 patients in a period of 60 months with preoperative diagnoses of scar endometriosis, all were confirmed by histopathological diagnosis; they concluded that surgical excision of scar endometriosis is the gold standard treatment option but must be done with at least 1 cm surgical margin. In this case, although the patient had surgery three times for this condition, last time there was more than a 2 cm free surgical margin; when the patient was conducted to have consent for case publication, she said she had a recurrence for the fourth time again.

We cannot find a cause of recurrence. This case report sends the message that incomplete excision is not the only cause of recurrence, and further research must be done to investigate other causes to prevent recurrence. And to investigate the efficacy of other treatment modalities than surgery, especially for recurrent cases.

Review of literature

1. Epidemiology of Endometriosis and Abdominal wall endometriosis

Endometriosis is characterized by endometrial epithelial and stromal cells in extra-uterine locations. Endometriosis is associated with chronic pelvic pain and infertility and affects 10% of women in their reproductive age, depending on the site of endometriosis. [4]. For instance, Pelvic endometriotic tissue's most common locations are the ovary and pelvic peritoneum. Sites of extra-pelvic localization include the gastrointestinal tract, the urinary tract, the respiratory system, and abdominal wall endometriosis (AWE), as those cesarean scar endometriosis (CSE). [4,5,6]

CSE has increased due to the increased rates of cesarean sections worldwide. This condition is only partially understood, and the diagnosis is often delayed or missed [7,8,9]. The effects of estrogen exposure after CS and endometrial seeding during the section are enhanced by altered immunity, chronic inflammation, and local growth factors [7,8,10]. This issue has many challenges since the preoperative diagnostic rate is low with no particular clinical risk factors, and the histological report remains the final diagnostic confirmation [9,10].

Abdominal Wall Endometriosis occurs after many obstetrical and gynecological surgeries as hysterectomy and laparoscopic surgeries, performed for non-surgical endometriosis as in the study of Akbarzadeh-Jahromi et al., where they reported trocar port site endometriosis in 18 patients [11,12]

Sumathy et al. reported CSE endometriosis in 18.9% in a case series of 16 women, while Tatli et al. reported no synchronous pelvic endometriotic lesions in 18 patients [13,14]. In 2 different case series studies, the mean age at diagnosis was 35 years, and the time from surgery to diagnosis of endometriosis varied from 3 months to 20 years [14,15]. The

reported incidence of CSE is 0.03-0.45%; however, this figure needs to be estimated due to the non-existence of consistent epidemiological data and the rarity of CSE [16,17]. Andolf et al. reported an incidence of 1.8% risk for developing endometriosis after CS. [18].

2. Pathogenesis and possible genetic role

Although many researchers describe AWE as a subtype of iatrogenic endometriosis, this did not explain why CSE happens. The pathogenesis is multifactorial, including immune, endocrine, and inflammatory pathways. Theories of cell migration in association with direct seeding or metaplasia have been proposed to explain this enigma [18]. Other theories of endometriosis as Sampson's theory (the retrograde menstruation hypothesis), cannot explain CSE but can explain pelvic endometriosis. Intra-operative implantation is certainly not relevant to non-surgical endometriosis (or "endogenous" endometriosis) [19-21]. Sumathy et al; Tatli et al. have identified pelvic endometriosis in cases of CSE [14,15]. In these endometriotic local implants [21]. Vascular Endothelial Growth factor abnormalities may be associated with this condition [22].

Genetic/epigenetic theory may explain the heterogeneity of endometriosis with a hereditary profile. Some Genome-wide studies have identified 12 nucleotide polymorphisms (single) at ten independent genetic foci associated with endometriosis. Two chromosomal areas with a significant linkage were observed on 7p13-15 and 10q26 (harboring genes such as INHBA, CYP2C19, HOXA10, and SFRP4). These changes include DNA methylation, de-methylation, and histone code modifications [23,24].

PPAR-γ is a nuclear receptor with neuroprotective and anti-inflammatory roles and is highly expressed in post-operative lesions [25]. Molecular biology studies of endometriosis have shown that Estrogen Receptors activation is a hallmark of the local changes occurring in CSE. Endometriotic lesions have estrogen and progesterone receptors. Hyperestrogenemia can occur secondary to defects of Methylation genes encoding transcription factors (steroidogenic factor-1, GATA6) and causing secondary inhibition of progesterone receptor. [26, 27].

Overall, CSE is developed only in some females. The postulated mechanisms involve the local environment at the implant site, including metalloproteinase activation due to local growth factors and inflammation, increased estrogen production through stimulation of estrogen receptors, and potential epigenetic changes. [28]

3. Diagnosis

The most common complaint is pain at the site of the CS scar during menstruation. Chronic pain may be experienced unrelated to the menstrual cycle, including the pelvic, lumbar, and abdominal regions [29,30]. Rarely, the patient may present with skin changes, ecchymosis, or hyperpigmentation of the scar during menstruation [13]. A palpable lump may be felt at the abdominal wall during menses [30]. The clinical triad includes cyclical pain, a mass at or near the level of the CS scar, and a history of CS. [3,11]. In the study of Zhang et al., 98.5% of the patients presented with abdominal swelling, followed by cyclic pain in 86.9%of patients. Khan et al. performed a case-control study at Mayo Clinic, in which 2539 women had endometriosis-surgery were enrolled, showing that 1.34% of the patients had Abdominal wall endometriosis; CSE was recorded in 59% of cases with AWE. [31,32].

4. Pre-operative investigations

If EEE is suspected, we can use ultrasound, magnetic resonance imaging (MRI) of the abdomen, and computed tomography (CT) to examine the abdominal wall. (28)

MRI is better for discovering small lesions,

while CT is better in cases with subcutaneous layer and muscle involvement. Ultrasound is still the best screening method. By ultrasound, the lesions of CSE have a hyperechoic or isoechoic pattern (46.7%), with peripheral vascularization (61.5%), and are hyper-vascular or homogenous on CT scan [33]. MRI is the most common method for preoperative endometriosis staging [34].

Wozniak et al. have concluded that sonoelastography significantly improved ultrasound accuracy in evaluating the depth of infiltration of CSE, even in women with high. [35]. Fawzy and Amer evaluated transabdominal sonoelastography in 34 patients with CSE. They found that it is particularly useful in endometriomas [36]. Fine-needle aspiration (FNA) has been used for ultrasound-guided aspiration of superficial lesions [37,38]. FNA is a non-invasive and simple procedure. Lopez-Soto et al., in their theses of 33 patients, used FNA in 72% of patients [32]. Fine-needle aspiration is useful in the diagnosis and for differential diagnosis. The differential diagnosis of CSE includes hernia (incisional or inguinal), hematomas, lipomas, granulomas, and desmoid tumors [28].

5. Pathological report

The definitive diagnosis is the final histological report, where grossly, the mass is well-defined and manifests as endometriomas. The endometrial cells are implanted in the dermis and rectus abdominis muscle. [28]

6. Therapy

CSE may need a multidisciplinary approach. Usually, endometriosis is treated by hormonal drugs in addition to painkillers and, finally, surgery, depending on the pain management and/or desire for fertility. surgery is the only curative therapy in cases of CSE to resolve chronic pain. During surgery of endometriotic nodules, A wide incision is recommended to decrease the risk of recurrence, which is described in 5-9% of cases [28]. Sclerotherapy with ultra-sound guided ethanol injection into the lesion of scar endometriosis has been reported to be effective in isolated cases to prevent abdominal wall defects after wide excision [35]. High-intensity focused ultrasound ablation (HIFA) has been used as an alternative to surgery, with a recurrence rate of 3.9%. The study of Lee JS et al. showed that HIFA had fewer side effects, such as blood loss and parietal defects after surgery [38]. Combined oral contraceptives, progestins, and gonadotrophin-releasing hormone (GnRH) analogs are used in the postoperative period to delay new growth and reduce the risk of recurrence. [28]

Conclusion

CSE represents a dynamic multidisciplinary topic with an increasing incidence due to the increasing CS. The clinical manifestations range from a mass to local pain at the cesarean scar. Ultrasound and MRI may help diagnose, but the definitive diagnosis remains the histological report. The best management is the surgical removal of the implant.

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