Delayed diagnosis is associated with changes in the clinical and ultrasound features of subcutaneous endometriosis near cesarean section scars

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KEYWORDS

Ultrasound;
Color Doppler;
Endometriosis;
Cesarean section.

Abstract *Introduction:* The aim of the present study was to highlight the sonographic and clinical characteristics of large (≥ 30 mm in diameter) scar endometriomas near Pfannenstiel incisions, assuming that large size is a marker of delayed diagnosis.

Methods: We compared clinical, ultrasound (US) and color Doppler findings in 13 patients (mean age 31.3 years) with 13 large scar endometriomas (L-SEs) (mean lesion diameter 41.8 mm; range 30–60 mm) and 17 women (mean age 30.7 years) with 19 small scar endometriomas (S-SEs) (mean lesion size 18.3 mm; range 7–26 mm).

Results: Compared with the S-SE group, the L-SE group had a significantly longer mean interval between the last cesarean section and hospital admission (5.5 vs. 3.3 years; $p < 0.01$) and longer mean duration of symptoms before admission (43 vs. 17.4 months; $p < 0.01$). The L-SE group also had a significantly higher percentage of patients who had undergone at least one inconclusive diagnostic examination (Computed Tomography, Magnetic Resonance Imaging, fine needle biopsy, or laparoscopy) (39% vs. 0%; $p < 0.05$). As for US findings, L-SEs more frequently displayed cystic regions and fistulous tracts ($p < 0.05$), loss of round/oval shape ($p < 0.05$), and increased vascularity ($p < 0.05$).

Conclusions: Delayed diagnosis of scar endometrioma reflected by a longer and more complex medical history results in larger than usual endometriomas with peculiar US findings, which are even more likely to be misinterpreted by physicians and radiologists.

Sommario *Introduzione:* Scopo dello studio è valutare le caratteristiche ecografiche e cliniche di endometriomi in prossimità di cicatrice di Pfannenstiel in rapporto a ritardo diagnostico segnalato da dimensioni ≥ 30 mm al momento della diagnosi istologica.

Metodi: Sono stati confrontati i dati clinico-anamnestici, ecografici e Color Doppler di 13 pazienti (età media 31.3 anni) con 13 endometriomi ≥ 30 mm ($\emptyset$ medio 41.8 mm; range 30–60 mm) e di 17 pazienti (età media 30.7 anni) con 19 endometriomi < 30 mm ($\emptyset$ medio 18.3 mm; range 7–26). In tutti i casi era disponibile il risultato istologico su campione operatorio.

Risultati: Nelle pazienti con endometrioma ≥ 30 mm risultavano più lunghi sia il tempo medio intercorso tra il ricovero e ultimo taglio cesareo (5.5 vs. 3.3 anni; $p < 0.01$) che la durata media

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dei sintomi prima del ricovero (43 vs. 17.4 mesi; $p < 0.01$). Inoltre, indagini diagnostiche diverse dall'ecografia (TC, RM, FNB, celioscopia) o interventi chirurgici incongrui si registravano nel 39% delle pazienti con endometriomi ≥ 30 mm, ma in nessuna di quelle con piccoli noduli ($p < 0.05$). Dal punto di vista ecografico gli endometriomi ≥ 30 mm mostravano più spesso ($p < 0.05$) perdita della morfologia ovalare, aree liquide interne, tramiti fistolosi e multipli poli vascolari con aumentata vascolarizzazione intralesionale.

Conclusioni: Il ritardo nella diagnosi dell'endometrioma su cicatrice di Pfannenstiel, segnalato da una storia clinica più lunga e complessa, modifica i caratteri ecostrutturali più tipici dell'endometrioma e può rendere ancora più difficile il riconoscimento di questa condizione patologica.

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Introduction

Endometriosis is defined as ectopic implantation of uterine mucosal tissue outside the uterine cavity (usually in the pelvis), causing dysmenorrhea, dyspareunia, menstrual irregularities, and infertility [1]. Only a small proportion of endometriosis patients have extrapelvic disease [2,3], and subcutaneous involvement is even rarer. Subcutaneous endometriosis of the abdominal wall generally develops at the site of previous abdominal surgery or pelvic procedures, and these lesions are referred to as "scar endometriomas" [4–6]. They develop near caesarean section scars in 0.03–1.5% of all women who have had caesarean deliveries [4–8]. The diagnosis of scar endometriosis is complicated by similarities between its clinical symptoms and those of other benign or malignant diseases of the abdominal wall, such as suture granulomas, incisional hernias, and primary or metastatic cancers [9,10]. Its comparative rarity and the uncertainties encountered in its diagnosis are the main reasons scar endometriomas are so frequently misdiagnosed by both clinicians [9,10] and radiologists [11,12].

We recently described the clinical, sonographic, and color Doppler features of 12 cases of cesarean-scar endometriosis [13]. The most common sonographic presentation we observed was that of a subcutaneous nodule near a Pfannenstiel incision (average length 2 cm) with irregular borders, a heterogeneous texture characterized by scattered internal hyperechogenic foci, a peripheral hyperechoic ring of variable width, and scanty vascularity at color Doppler imaging. Clinical symptoms included cyclic pain and swelling of a usually palpable nodule. With increasing experience, however, we have noticed that large lesions (≥ 30 mm) are often associated with certain distinctive clinical and US characteristics, which are probably related to their delayed diagnosis. The aim of this study was to verify which sonographic and clinical characteristics of scar endometriomas are affected by delayed diagnosis, reflected by larger than usual nodules.

Methods

We enrolled 30 women (mean age 30.6 years; range 20–42) with 33 scar endometriomas, who were consecutively seen at our institution between June 1999 and April 2008. All of the endometriomas were treated with wide surgical excision, and pathology reports were available for all surgical specimens. The following information were collected from each participant:

- 1) Clinical data: number of caesarean sections; time elapsed since last caesarean section; time between symptom onset and hospital admission; known pelvic endometriosis; inconclusive diagnostic exams (other than sonography) previously performed at other institutions (Computed Tomography [CT], Magnetic Resonance Imaging [MRI]), fine needle biopsy [FNB], laparoscopy); previous pelvic/abdominal surgery related to painful symptoms; characteristics of pain (cyclic with menses vs. continuous); nodule palpability on admission.
- 2) Ultrasound and Color Doppler findings: morphology (round or oval vs. irregular shape); echotexture (hypoechoic with hyperechoic spots/strands; additional findings, e.g., cystic areas, fistulas); presence/completeness of peripheral hyperechoic ring; margins (spiculated/infiltrating vs. smooth/lobulated); presence of multiple vascular pedicles entering the mass from different points; presence of central vascularity; and the resistance index (RI) of arteries supplying the lesion.

All US examinations were carried out by a single operator (G.F.) using high-frequency transducers (7.5–14 MHz) with the US scanners available at our Institution during the study period (Logic 500, Logic 700 Expert Series, Logic 7, Logic 9, GE Healthcare, Milwaukee, WI).

Color Doppler parameters were set to detect low blood flow velocities (pulse repetition frequency 500–1000 Hz, wall filter 50 Hz, high filters for color-vs.-echo priority and color persistence). The RI of arterial flow (when detectable) was expressed as the mean of 2 measurements.

For comparison purposes, a large scar endometrioma (L-SE) was defined as a nodule whose maximum diameter was equal to or greater than 30 mm.

Fisher's exact test and the chi square test were used to assess categorical data. The unpaired *t*-test was used for continuous variables. *p* values < 0.05 were considered statistically significant.

Our institutional ethics committee approved the study protocol, and written consent to participate the study was obtained from all patients.

Results

The 33 scar endometriomas ranged in diameter from 7 to 60 mm (mean diameter 27.1 mm), and most (26/33, 78.7%) were located between subcutaneous fat and a muscle

sheath. The remaining endometriomas included 2 that were purely subcutaneous, 2 that were entrapped in the muscle layer abdominal wall, and 3 that had infiltrated both subcutaneous and muscular tissues.

Scar endometriomas did not show any preferential location with respect to the scar: 16 were located on the right, 14 on the left, and 3 on the midline.

Thirteen patients (mean age 31.3 years) had L-SEs. These 13 nodules ranged in diameter from 30 to 60 mm (mean 41.8 mm). One woman (whose 60-mm L-SE was the largest of this series) also had a 10-mm satellite nodule. Seventeen women (mean age 30.7 years) had small scar endometriomas (S-SEs) ($n = 19$) ranging in size from 7 to 26 mm (mean 18.3 mm).

Clinical findings

The mean number of caesarean sections per patient was 1.6 (range 1–3). The mean interval between the last caesarean section and hospital admission was 3.0 years (range 1–10 years), and the mean duration of symptoms before admission was 25.7 months (range 0.5–80 months). Only 2 women had histories of previous pelvic endometriosis. At enrolment, palpable abdominal wall lesions were found in all but 1 of the participants (an obese patient with a 7-mm nodule). Twenty-four (80%) women had cyclic pain associated with menses and a palpable mass. In the remaining 6 cases, pain was described as continuous.

In patients with L-SEs, the mean interval between the last caesarean section and hospital admission (5.5 vs. 3.3 years; $p < 0.01$) and the mean duration of symptoms before admission (43.0 vs. 17.4 months; $p < 0.01$) were both longer than those observed in patients with S-SEs.

In addition, a significantly higher percentage of patients with L-SE (39% vs. 0%; $p < 0.05$) had undergone at least one inconclusive diagnostic examination at another institution (CT in 1 case, MRI in 2, FNB in 1, and laparoscopy (twice) in 1 case).

Complaints of continuous rather than cyclic pain were also more common in the L-SE subgroup (30.7% vs. 11.7% in patients with S-SEs) although this difference was not statistically significant. In addition, 2 patients with L-SEs (15.3%) reported undergoing surgical interventions for “bowel adhesions,” which had not improved their symptoms. At the time of this report, recurrent endometriosis has not been observed in either group.

US and Color Doppler findings

On B-mode studies, most scar endometriomas (24/33, 73%) appeared as roundish, inhomogeneously hypoechoic nodules with hyperechoic spots or strands (indicative of fibrotic changes), a peripheral hyperechoic ring (complete or incomplete), spiculated margins, and a single vascular pedicle entering the mass at the periphery (Fig. 1). The mean RI of these 24 lesions was 0.78.

Fig. 1 A 28-year-old-woman with an 8-month history of cyclic pain that began two years after a cesarean delivery. Transverse sonographic scan reveals a small (2 cm) scar endometrioma with typical features: an ovoid nodule, hypoechoic with hyperechoic spots, spiculated infiltrating margins, and a hyperechoic peripheral ring (black arrows) (A). Color Doppler exam shows the typical single vascular peripheral pedicle without central vessels (B). The color Doppler examination reveals high-resistance arterial blood flow (C).

Table 1 Ultrasound and color Doppler findings in patients with large and small scar endometriomas.

	L-SE (13 pts with 13 nodules)	S-SE (17 pts with 19 nodules)	<i>p</i>
US findings			
Echotexture			
Hypoechoic with hyperechoic spots/strands	11	19	N.S.
Cystic portions and/or fistulous tract	7	1	<0.01
Peripheral hyperechoic ring (absent/present)	1/13	7/12	N.S.
Margins (spiculated/infiltrating)	9	14	N.S.
Loss of oval or round shape	4	0	<0.05
Color Doppler findings			
Resistance index (mean $\pm$ SD)	0.75 $\pm$ 0.08	0.66 $\pm$ 0.16	N.S.
Multiple vascular pedicles	8	2	<0.01
Central vascularization	8	2	<0.01

L-SE = Large Scar Endometrioma S-SE = Small Scar Endometrioma N.S. = Not Significant.

Compared with S-SE lesions, L-SE nodules more often presented the following features (Table 1): small internal cystic areas and fistulous tracts (53.8% vs. 5.2%; $p < 0.01$) (Figs. 2 and 3); elongated, irregular shapes as opposed to the usual oval or round shape (30.7% vs. 0%; $p < 0.05$) (Figs. 2 and 4); and multiple vascular pedicles associated with increased central vascularization (61.5% vs. 10.5%; $p < 0.01$) (Figs. 5 and 6). In 2 S-SEs (7 and 14 mm), no Doppler signals were observed.

During the study period, we also examined 7 other women with masses located near caesarean section scars. US excluded the diagnosis of endometriosis, suggesting different diagnoses (1 metastatic lesion, 2 incisional hernias, 1 abscess, 2 chronic inflammations, 1 desmoid tumor), which were subsequently confirmed by surgical pathology in all cases. This experience underlines the specificity of our diagnosis of endometrioma.

Discussion

Scar endometrioma is a rare form of extrapelvic endometriosis, and inadvertent transfer of endometrial cells during

caesarean section [1,14,15] is the most likely mechanism underlying its development. These lesions have also been reported with scars produced by other procedures, such as hysterectomy [15,16], laparoscopy (trocar tract scars) [17], needle tract amniocentesis (needle tract) [18], and perineal episiotomy [19].

Patients typically present with a small (≤ 2 cm), palpable mass near a caesarean section scar that becomes painful with menses [6,11,13]. The US and Doppler characteristics of scar endometrioma described in the literature [13,20] are quite different from those observed in most cases of pelvic endometriosis. Classic adnexal endometriomas are round cyst-like masses with regular margins, thick walls, and homogeneous low-level internal echoes [11,21–23]. In contrast, scar endometriomas present as solid, inhomogeneously hypoechoic masses with fibrotic changes reflected by tiny hyperechoic spots or thick hyperechoic strands, and their margins are irregular, often frankly spiculated. Color Doppler reveals vascularization in most of these masses whereas cystic changes have rarely been reported [13,20,24]. In most cases, there is also a perilesional

Fig. 2 A large (60 mm) endometrioma in a 27-year-old woman. The lesion is irregularly shaped (black arrows), and a small cystic area (white arrowhead) is seen in the deepest plane.

Fig. 3 A 30-year-old woman with an 84-month history of continuous pain in the lower abdomen. This transverse scan reveals a linear fistulous tract emerging from the posterior aspect of the endometrioma running through the muscle layer (between black arrows).

Fig. 4 A 38-mm scar endometrioma involving subcutaneous and muscular tissues in a 25-year-old woman. The nodule was associated with cyclic pain, and on US it displayed an irregular shape, spiculated margins, and hyperechoic strands of fibrotic tissue (A). Macroscopic findings of the surgical specimen (B).

hyperechoic ring (complete or incomplete) [13], which is thought to reflect inflammatory changes triggered by monthly hemorrhage into adjacent tissues [24].

The diagnosis of scar endometriomas can be challenging. These lesions need to be distinguished from a number of other lesions (including suture granulomas, incisional hernias, benign tumors such as desmoids, primary or metastatic cancers), and their features on diagnostic imaging studies (including US, CT and MRI) are usually nonspecific [20,22,25,26]. Some authors have proposed FNB for diagnosis of scar endometriomas [3,20,27] but even this invasive method can produce inconclusive results [28], as it did in one of our patients.

These uncertainties and the consequent delay in diagnosis explain why large endometriomas accounted for 43% of our series. The risk of misdiagnosis is also reflected in the fact that women with L-SEs had longer medical histories that often included costly, inconclusive diagnostic procedures (CT, MRI, FNB, laparoscopy), some of which were invasive.

Delayed diagnosis also seems to modify both the pain characteristics and US features of scar endometriomas. Compared with women who had small nodules, those with L-SEs more often complained of continuous (rather than cyclic) pain, and their tumors more frequently presented

abnormal shapes, small cystic areas, fistulous digitations, and increased vascularization. These sonographic features are most likely related to long-standing cyclic hemorrhage that produces inflammatory and fibrotic changes in surrounding tissues. The increased blood-vessel density is another obvious result of increasing nodule size over time. The atypical presentation of these older lesions can add to the confusion, further delaying the diagnosis. To break this vicious cycle, a detailed clinical history and accurate physical and US examination are, in our opinion, the most appropriate and effective tools. In our institution, the work-up of women presenting with a palpable nodule near Pfannenstiel incisions (with or without pain) is based solely on clinical evaluation and US. Over the last 9 years, this approach has proved to be very cost-effective: after the first 3 misdiagnosed cases (suture granulomas and sarcoma), endometriomas were correctly diagnosed in the remaining 27 patients, and 7 other lesions near cesarean section scars were correctly identified as non-endometrioma. CT, MRI, and FNB were not used in the diagnosis of any of these lesions, and surgery, the only effective treatment for abdominal wall endometriosis [15,16], was never delayed.

Fig. 5 A 32-year-old woman with L-SE. In this transverse scan, color Doppler reveals multiple vascular poles entering the mass from different points.

Fig. 6 A 26-year-old woman developed continuous pain six months after a caesarean delivery 3 years before. In this 48-mm scar endometrioma, the color Doppler examination demonstrates increased internal vascularization along with multiple vascular pedicles.

In conclusion, delayed diagnosis of scar endometriomas, reflected by a longer and more complex medical history, results in larger tumors with peculiar US findings, which are even more likely to be misinterpreted by physicians and radiologists. In most cases, confident preoperative diagnosis of scar endometrioma can be made on the basis of the clinical presentation (palpable mass near cesarean section scar and pain) along with a careful sonographic examination of the abdominal wall, but it is important to remember that the US features of these lesions may vary according to their size.

Conflict of interest statement

The authors have no conflict of interest.

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