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Dr. Sandhyasri Panda
Department of Obstetrics and
Gynecology, Maharaja's
Institute of Medical Sciences,
Vizainagaram, India

**Dr. Apurva Ganeshprakash
Gupta**
Department of Obstetrics and
Gynecology, Maharaja's
Institute of Medical Sciences,
Vizainagaram, India

Corresponding Author:
Dr. Sandhyasri Panda
Department of Obstetrics and
Gynecology, Maharaja's
Institute of Medical Sciences,
Vizainagaram, India

A series of caesarean scar endometriosis with literature review

Dr. Sandhyasri Panda and Dr. Apurva Ganeshprakash Gupta

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Abstract

Introduction: Scar endometriosis is a rare form of endometriosis that usually develops in the scar after obstetric or gynaecological surgeries. With the increasing caesarean section rates, the frequency of caesarean scar endometriosis (CSE) is expected to rise. The symptoms and signs of scar endometriosis may be ambiguous and hence remains under diagnosed and under reported.

Aim and Objectives: To analyse the cases of CSE and to establish surgery as the mode of treatment.

Materials and Methods: This study involves review of records of nine cases with a clinical diagnosis of caesarean scar endometriosis. Seven of them received surgical management while two chose medical management. Diagnosis was made by clinico-imaging and cyto-histopathology as applicable.

Results: The mean age of patients is 30.5 years. All subjects had history of caesarean section; they presented with cyclical pain at the scar and dysmenorrhea (100%). Five (55.6%) subjects had a palpable lump. Seven/nine subjects received wide local excision whereas two opted medical therapy. Two subjects underwent hysterectomy, one in view of recurrence and the other a multiparous women with concurrent AUB. Three subjects were given postoperative medical management in view of margin positive histology.

Conclusion: The medical therapy was aimed the symptoms and to suppress the lesion; preferred in smaller endometriomas, desirous of future child bearing and in margin positive postoperative cases. However, surgical excision, which is both diagnostic and curative, remains the most effective treatment for scar endometriosis.

Keywords: Caesarean scar endometriosis, wide local excision, medical management

Introduction

Endometriosis is defined as the presence or growth of functional ectopic endometrial tissue [1]. Ectopic tissue responds to ovarian hormonal stimulation and tends to proliferate when stimulated by cyclic estrogens, thus, appearing "to menstruate" [2]. It affects 89 million women of reproductive age worldwide, approximately 5 to 10% of all women [3]. The ovary and pelvic peritoneum are the two most common pelvic locations for endometriotic tissue. The gastrointestinal tract, urinary tract, and respiratory system are extra-pelvic sites [4-6]. One of the rare type of extrapelvic endometriosis is abdominal wall endometriosis and its incidence varies from 0.03 to 3.5% [7]. AWE can be superficial (affecting only subcutaneous tissue above the fascia), intermediate (infiltrating the rectus sheath), or deep (affecting rectus muscles) depending on the layers involved [8].

Scar endometriosis refers to the development of abdominal wall endometriosis (AWE) at the surgical incision site following obstetric or gynecological surgeries [9]. The majority of scar endometriosis cases have been reported following procedures such as cesarean section, hysterotomy, hysterectomy, episiotomy, tubal ligations, and a few following myomectomy, appendectomy, in the laparoscopic trocar tract, and amniocentesis needle tract [10, 11]. Caesarean Scar Endometriosis (CSE) is the most commonly reported form of AWE [12].

The actual incidence is difficult to determine because it is under diagnosed and under reported. Meyer reported the first case in 1903(13). Scar endometriosis incidence is reported to be 0.03-0.4% after caesarean section [7], and 1.1% after mid-trimester abortion [10]. The prevalence of concomitant pelvic endometriosis and scar endometriosis has been estimated to be 14.3-26% [14].

Scar endometriosis is expected to become more common as Caesarean section rates increase [15].

Some refer to AWE as the "iatrogenic" subtype of endometriosis; however, the mechanism remains undetermined [16]. Various theories have been proposed regarding the development of scar endometriosis. The direct implantation theory states that during surgery, endometrial cells scattered around the abdominal wall proliferate or cause metaplasia of the surrounding fascia due to hormones. The second theory proposes that endometrial cells spread via lymphatic or hematogenous routes [10]. The third theory holds that pluripotent mesenchymal cells differentiate to form endometriosis [17]. Scar endometriosis may present with ambiguous symptoms and sign [9]. Patients typically report a triad of complaints including cyclic pain and swelling at the incision site with history of obstetric and gynaecological surgery [7]. A clinical examination may reveal a painful and palpable nodule with maximum tenderness over the scar during menstruation. Only half of them have classic symptoms and can be misdiagnosed as an incision hernia, abscess, hematoma, lipoma, carcinoma (primary), granuloma, lymphoma, neuroma, cyst, abscess, or soft tissue sarcoma [18]. This causes a significant delay in diagnosing and initiating treatment. It is hypothesized that obesity-related suboptimal closure of uterine incision or abdominal layers contributes to the development of AWE. Pelvic endometriosis increases the risk of developing AWE [15].

Ultrasound aids in the diagnosis of subcutaneous lesions by detecting the presence of a hypoechoic mass, which may occasionally reveal internal hyperechoic areas [19]. It also helps to rule out other possibilities, such as an abdominal wall hernia. Doppler may reveal increased vascularity. An MRI is required to determine the extent of the lesion beyond the subcutaneous plane and to identify any intra abdominal extension. FNAC may help distinguish between other diagnoses and metastatic disease, but it may be harmful in the case of abdominal wall hernia. FNAC helps to rule out malignant transformation of abdominal wall endometriosis [20]. Ectopic endometrial glands with cellular stroma, extravasated erythrocytes, and inflammatory infiltrate are common histological findings. However, fibrosis in the endometriotic tissue may lead to an inconclusive diagnosis [13].

Malignant change in abdominal wall endometriosis is extremely rare, with an estimated incidence of 0.3% to 1% [21]. Advanced age, postmenopausal status, and lesion diameter of more than 9 cm are all risk factors. Malignancy should be suspected in cases of multiple recurrences, failure to respond to treatment, and sudden rapid growth. Clear cell carcinoma is the most common histological subtype [15].

Medical treatment with combined oral contraceptives (COCs), progestins, androgens, or gonadotropin-releasing hormone (GnRH) analogues temporarily relieves symptoms, but they recur after the therapy is discontinued [18]. Surgery is frequently required for both treatment and definitive diagnosis, with a 95% success rate [15].

The purpose of this study is to examine the clinical aspects of caesarean scar endometriosis, to correlate clinico-imaging with histopathology findings and to establish wide local excision as the treatment of choice.

Materials and Methods

In this study, prospectively maintained case profiles of nine cases between 2015 and 2021 with a clinical diagnosis of caesarean site scar endometriosis were reviewed. Seven of

them received surgical management, which included a wide local excision, while two were given medical management. A detailed history about symptoms at presentation, indication and the stage of labor when caesarean delivery was conducted, examination findings, ultrasonography findings, and surgical notes describing the lesion were recorded. Any information about postoperative medical management was obtained. CSE is confirmed by the presence of endometrial tissue, focal hemorrhage, hemosiderin deposits and/or fibrosis on histopathology reports. Patients were followed up for two years.

Results

The mean age of patients is 30.5 years ranging between 26 to 36 years. Six subjects are para 2 with a history of two previous LSCS, while the remaining three are para1 with one previous LSCS. The mean duration of symptoms is 10 months, with a range of 6 months to 24 months. All nine subjects reported cyclical pain at the scar site (100%) along with dysmenorrhea (100%). Five subjects (55.6%) had palpable solitary lump at the scar site. Clinically inconspicuous abdominal wall endometriomas in four subjects were later supported by ultrasonography. Itching was an accompanying complaint in two of them (22.22%). The mean interval between caesarean section and diagnosis of abdominal wall endometriosis is 7.8 years, with a range of 3 to 14 years. Six subjects had a subumbilical midline scar (66.66%), while the other three subjects had pfannensteil scar (33.33%). The majority of the pathology occurred on the right side (88.88%) of the incision followed by left side (11.11%). All subjects underwent ultrasonography to aid in the diagnosis. It revealed the presence of a hypoechoic lesion with heterogenic spots or cystic lesions in the anterior abdominal wall. The mean lump size based on USG is 4.81 +/- 1.1 cm and intraoperatively is 4.43 +/- 1.1 cm, with fibrosis of adjacent tissue. Amongst the operated cases, there are four cases of superficial scar endometriosis (57.71%), which affects only the skin and subcutaneous layers, and three intermediate cases that involve the rectus sheath (42.85%) as well, while none had deep CSE i.e. none showed involvement of the rectus muscle or peritoneum (Table 1). Hence, it indicates the endometrioma of scar invades from superficial to deep layers. None of the subjects demonstrated signs of concurrent pelvic endometriosis. FNAC was performed in 3 out of 9 subjects, with one yielding inconclusive results and the other two revealing the presence of endometrial glands and stroma, thus confirming the diagnosis. As first-line management, seven of nine subjects underwent wide local excision with a 1cm free margin (Figure 1). In light of extensive intraoperative fibrosis and margin positive histology reports, three patients received postoperative DMPA (depot medroxyprogesterone acetate) 150mg single dose, followed by continuous dienogest 2mg OD orally or COC for three months in two of them respectively. These three did not show any relapse hence confirming cure. A 26year old, PIL1A1 conceived after 2 year of excision but had a spontaneous abortion at third month of gestation. One case underwent WLE along with TAH + BSO as first line management due to concurrent complaints of AUB and completion of childbearing (Figure 2). Only one subject (No. 6) experienced recurrence after 10 months of wide local excision. The subject also had an incisional hernia. As a result, a repeat excision was performed, along with a total

abdominal hysterectomy and incisional hernia repair with mesh. The most likely cause of recurrence in this subject could be due to incomplete excision of an extensive fibrosis, obesity with BMI (>35kg/m²); she also presented with one and half years of amenorrhea, and resumed menstruation immediately following the primary excision of the mass. Two subjects with relatively small endometriomas denied surgical treatment. One subject was treated with continuous COC for three cycles and the other was given only

symptomatic treatment with NSAIDS based on surgeon preference. Both reported symptom relief during two year follow-up period.

Histopathological diagnosis is confirmed by the presence of endometrial glands and stroma, focal hemorrhage, hemosiderin deposits and/or fibrosis as shown in figure 3. Hence, clinico-imaging diagnosis of CSE was proved to be 100% accurate by histopathology reporting in all of the surgically treated subjects.

Table 1: Caesarean scar endometriosis (CSE): Demographic details, symptoms and management

Subject no.	Age (in years)	Parity	No. of LSCS	LCB (in years)	C/F		Type of scar	Side	Size of mass (in cm) USG		FNAC	Location	Preoperative diagnosis	Management 1 st line Follow up	
					Cyclical pain	Lump			Intraop						
1	26	P1L1 A1	1	3	+	+	SUM	Rt	5X5	6x5	-	Subcutaneous	CSE	WLE	DMPA f/b dienogest for 3cycles
2	28	P1L1	1	8	+	-	SUM	Rt	2.6x2	3x4	-	Subcutaneous + RS	CSE	WLE	-
3	28	P2L2	2	8	+	-	SUM	Rt	3x3	3.5x2.5	Inconclusive	Subcutaneous + RS	CSE	WLE	DMPA f/b COC for 3cycles
4	35	P2L2	2	6	+	+	Pf	Rt	6.6x5	6x5	-	Subcutaneous + RS	CSE	WLE	DMPA
5	28	P1L1	1	3	+	+	SUM	Rt	4x5	5x5	Endometrial glands and stroma +	Subcutaneous	CSE	WLE	-
6	32	P2L3	2	13	+	+	SUM	Lt	5x5	6x6	Endometrial glands and stroma +	Subcutaneous	CSE	WLE	-
					+	+	-	Lt	3.5x3.2	4x3	-	Subcutaneous + RS	Recurr-ence after 10 months postop	DMPA 150mg x 2doses	WLE + TAH + Incisional hernia mesh repair
7	36	P2L2	2	14	+	-	SUM	Rt	4.4x5	5x5	-	Subcutaneous	CSE	WLE + TAH + BSO	-
8	30	P2L2	2	5	+	-	Pf	Rt	1.2x1.2	-	-	-	CSE	COC x 3cycles	Symptom relief +
9	32	P2L2	2	7	+	+	Pf	Rt	2.9x1.2	-	-	-	CSE	NSAIDS	Symptom relief +

C/F: Clinical features; SUM: Subumbilical midline; Pf: Pfannenstiel; Rt: Right; Lt: Left; RS: Rectus sheath; CSE: Caesarean scar endometriosis; WLE: Wide local excision; DMPA: Depot Medroxyprogesterone acetate; COC: Combined oral contraceptive; TAH: Total abdominal hysterectomy; BSO: Bilateral salphigopherectomy



Fig 1: [a] Surgical exploration showing margins and extension of caesarean scar site endometrioma and [b] postoperative imaging of resected mass in respective subjects, with skin excision in case 6

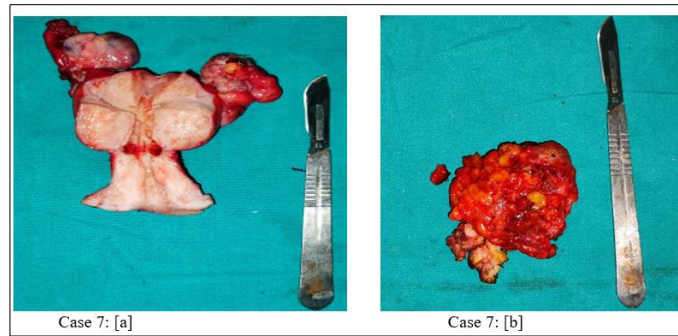


Fig 2: [a] TAH with BSO specimen [b] excised mass from abdominal wall layers

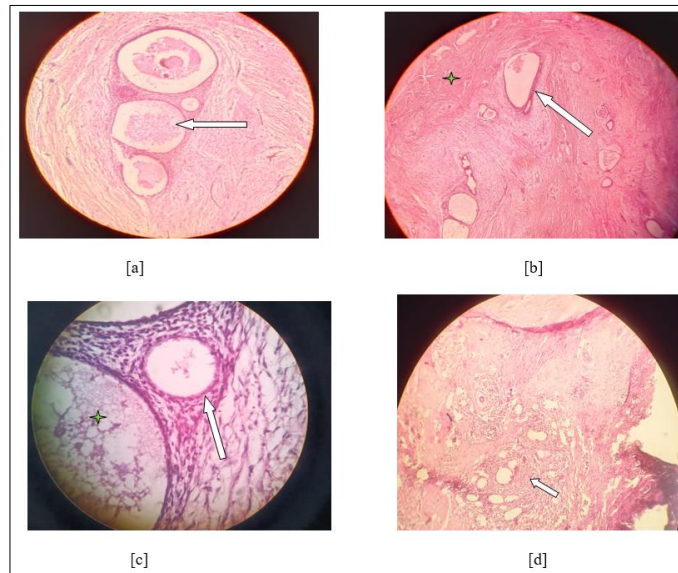


Fig 3: Hematoxylin and eosin (H&E) stained section of excised tissue displaying endometrial glands (arrow head) and stroma (arrow) in [a], [b], and [c], with fibrosis (star) in [b] and hemorrhage (star) in [c]. Figure [d] shows proliferating capillaries (arrow head) and hemosiderin-laden macrophages (arrow)

Discussion

CSE presents in reproductive age with mean age in our study being 30.5 years while that in the study by Faik Tatli *et al* was 32.71 +/- 8.61 years, Piriye *et al* as 36 +/- 5.4 years, Sumathy *et al* was 35.19 +/- 6.72 years [22-24].

It is important to note that a higher incidence is reported following early hysterotomy (end of second or beginning of third trimester), as early decidua appear to have more pluripotential capabilities, which can lead to increased cellular replication and endometriosis [25]. In theory, pregnancy, an altered immune response, and a caesarean section could increase the risk of developing endometriosis [26].

Based on our observation and literature review it can be hypothesized that decidua of early gestational age and at the same time elective caesarean or a caesarean section performed at early stage of labor has higher incidence of CSE; one of the contributing factor might be elective caesarean at earlier gestational age, as there is an increasing trend of caesarean section at maternal request. Some authors emphasized that having two caesarean sections did not increase the risk of being diagnosed with endometriosis when compared to having only one [26].

Vellido-Costelo *et al.* reported that there appear to be no link between pelvic and scar endometriosis development. In their study, 14% of patients had associated pelvic endometriosis, which is the incidence in the general population [27].

Studies by D.Poudel *et al* and Sumathy *et al* demonstrated presence of the endometrioma more to the left side on incisions unlike our study that showed more involvement on the right side [1, 24]. This right sided dominance was supported by Akbulut S *et al* [28].

In a series of 12 patients by Franciaca *et al.*, USG and color Doppler significantly contributed to the correct preoperative diagnosis, and the authors suggest that sonographic and color Doppler, when combined with clinical data, may significantly contribute to the preoperative diagnosis [29]. Older studies published a reporting in which FNAC was not diagnostic in any of the cases who underwent the procedure and use of this technique is debatable, as some authors have warned about the increased risk of producing new endometriotic implants at the puncture site, as well as visceral injury if the diagnosis is uncertain [25]. Whereas more recent studies by Sujaya Mazumder *et al.* and Katwal *et al* based on their experiences have argued that it is a quick, cost-effective, and accurate diagnostic tool to include in patients' management [13, 30] because of the practice of ultrasound guided FNAC. They supported the use of this technique to provide a tissue diagnosis prior to surgery and is also helpful in cases where the origin of the mass is uncertain. Vellido-Costelo *et al.* found that 52% of patients had a FNAC diagnosis prior to surgery, and one of them was diagnosed with cancer using this method, which led to a different therapeutic management [27].

Ucar *et al.* reported no recurrences over a follow-up period of 12-60 months [31]. Horton *et al.* in a series of 445 cases found a 4.3% recurrence rate [32], Zhang and Liu reported a 7.8% recurrence over an average of 20 (\pm 16) months in a series of 151 cases [33] while we had 14.28% (1/7 cases) recurrence rate over 24 months follow up period.

Medical treatment with progestogens, combined oral contraceptives, and danazol is ineffective in treatment of CSE and only provides partial relief from symptoms. Recently, there has been a report of the use of a gonadotrophin agonist, but only with immediate improvement in symptoms and no change in lesion size. These patients must be monitored because of the possibility of recurrence, which necessitates excision [3]. Surgical resection of the scar endometrioma remains the primary treatment option, even if the disease recurs. Because of the potential for recurrence (4.3% after surgery) [32] and malignant degeneration (0.3-1%) (21) of this condition, a local wide excision with at least a 1 cm resection margin is at present regarded as the best clinical practice. However, no studies have so far evaluated whether the surgical margin width affects the probability of recurrence rate.

To avoid endometrial inoculation after a caesarean section, it's important to clean the abdominal wound thoroughly, especially at the corners and on the surgeon's side which has been described in the older studies as well. It has also been suggested that at the end of surgery, particularly on the uterus and tubes, the abdominal wall wound be thoroughly cleaned by giving peritoneal wash or irrigated vigorously with a high jet solution before closure [1, 2, 34]. Other studies suggested that the absence of closure of the parietal and visceral peritoneum could significantly increase the risk of endometriosis in the skin incision scar [35]. Finally, instrument and needle replacement when suturing more superficial abdominal layers to avoid iatrogenic inoculation of endometrial cells is recommended [7, 34].

Conclusion

Despite the high probability of CSE due to rapidly increasing rate of abdominal birth and cut short technique of uterine and abdominal closure, still the diagnosis of scar endometriosis requires a high level of suspicion. It should be considered in all reproductive-age women with prior history of uterine surgery. They commonly present with cyclical pain and lump at the scar site. Clinico-imaging diagnosis was found highly specific as they correlated on histopathology. Medical therapy was aimed at both symptom relief and lesion suppression, with a preference in small endometriomas, in those who wish further child bearing and in margin positive postoperative cases. However, surgical excision, which is both diagnostic and curative, remains the most effective treatment for scar endometriosis.

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