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CESAREAN SCAR ENDOMETRIOSIS: PRESENTATION OF 10 CASES AND LITERATURE REVIEW

SEZARYEN SCAR ENDOMETRİOZİS: 10 OLGUNUN SUNUMU VE LİTERATÜR İNCELEMESİ

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ABSTRACT

Objective: Endometriosis, characterized by the presence of functioning endometrial tissue in non-uterine locations, is a relatively common disease in women of reproductive age (10-15%). Besides that, cesarean scar endometriosis (CSE) is rarely seen; the clinical presentation of the disease is mainly in the form of a painful mass at the cesarean incision site.

Method: Ten patients, whose diagnosis was confirmed histopathologically by surgery with CSE anterior chamber, were evaluated in terms of clinical and histopathological features.

Results: The mean age of patients was 35 (min-max:22-45). Six (75%) of patients with pain symptoms were cyclic, and 2 (25%) were noncyclic. The mean latency period (time from last cesarean section to the beginning of symptoms) was 44,6 (min-max:6-88) months, and the mean duration between symptoms and surgery was 28,9 (min-max:2-60) months. Six (60%) of endometriomas were located right corner of the incision, and 4 (40%) were at the left. All of the endometriomas were solitary. The mean hospitalization time was 2,5 (min-max:1-6) days.

Conclusion: Excision of endometriotic foci within 1 cm safety margins may be an appropriate curative treatment option.

Key Words: Cesarean Section, Endometriosis, Cicatrix, Pelvic Pain, Dysmenorrhea

ÖZ

Amaç: Endometriozis, endometrial dokunun uterus dışı yerleşimlerde varlığı ile karakterize olup, üreme çağındaki kadınlarda nispeten sık görülen bir hastalıktır (%10-15). Bunun yanında sezaryen skar endometriozisi (CSE) nadiren görülür; hastalığın klinik görünümü esas olarak sezaryen insizyon hattında ağrılı bir kitle şeklindedir.

Yöntem: CSE ön tanısı ile opere edilerek tanının histopatolojik olarak doğrulandığı 10 hasta klinik ve histopatolojik özellikleri açısından değerlendirildi.

Bulgular: Hastaların yaş ortalaması 35 (min-maks:22-45) idi. Ağrı semptomları hastaların altısında (%75) siklik, 2'sinde (%25) nonsiklik karakterdeydi. Ortalama hastalığın görülme süresi (son sezaryen ile semptomların başlangıcına kadar geçen süre) 44,6 (min-maks:6-88) ay ve semptomlar ile ameliyat arasındaki ortalama süre 28,9 (min-maks:2-60) ay idi. Altı hastada (%60) endometrioma insizyonun sağ köşesinde, 4 hastada ise (%40) soldaydı. Endometriomaların tamamı soliter yapıda izlendi. Ortalama hastanede kalış süresi 2,5 (min maks:1-6) gündü.

Sonuç: Endometriotik odakların, 1 cm güvenlik sınırı ile eksizyonu uygun bir küratif tedavi seçeneği olabilir.

Anahtar Kelimeler: Sezaryen, Endometriozis, Sikatris, Pelvik Ağrı, Dismenore

INTRODUCTION

Endometriosis is a common disease that affects 10–15% of all women of reproductive age [1] and is characterized by functioning endometrial tissue in non-uterine locations. Although ectopic endometrium tissue is usually located in the pelvis (ovaries, fallopian tubes, peritoneum, and recto-vaginal septum), it can be found at sites outside the pelvis such as the lung, brain, bowel, and abdominal wall [2–4].

While abdominal wall endometriosis (AWE) can be seen spontaneously, it usually develops secondary to a surgical operation such as a cesarean section (CS), hysterectomy, or laparoscopy [5–7]. Occasionally AWE cases have also been reported after amniocentesis [8]. The most common subtype of AWE is cesarean scar endometriosis (CSE), approximately 85% of all AWEs, and the reported incidence is 0.03-0.45% [9]. Although several mechanisms have been proposed to explain the development of CSE, metaplasia and cell migration in association with direct seeding are most accepted [10].

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The primary symptom is a painful (commonly cyclic but also can be noncyclic) mass located in the cesarean scar area [11]. Due to the rarity and similarity of symptoms, CSE can easily be misdiagnosed. Hernia, lipoma, granuloma, sebaceous cyst, neuroma, hematoma, lymphadenopathy, abscess, dermoid tumors and sarcomas must also be considered at a differential diagnosis. Malignant transformation of CSE has also been sporadically reported [12-13]. In this study, we reported the clinical characteristics of our ten patients who underwent surgery due to scar endometriosis and aimed to discuss the surgical results in light of the literature.

METHOD

In this retrospective study, we analyzed patients' medical records with a histopathological diagnosis of surgical scar endometriosis in the department of Obstetrics and Gynecology of the Sakarya Research and Education Hospital between January 2018 and February 2021. Information about all patient's ages, parity, number and time of CS, symptoms, beginning time of symptoms, size, boundary and location of the lesion, diagnostic methods, surgical procedure, and hospitalization time were extracted (Table 1,2).

Statictical Analysis

Data concerning demographic and clinical characteristics were analyzed using descriptive methods (means, minimum-maximum). The statistical software used was SPSS for Windows, version 22.0 (SPSS Inc., Chicago, IL, USA).

Ethical Approval

The Non-Interventional Ethics Committee of Sakarya University Faculty of Medicine approved our study (Date: 30.06.2021, No: 39912).

RESULTS

The mean age of patients was 35 (min-max:22-45). The parity of patients ranged from 1 to 3. All of the patients had a history of CS with Pfannenstiel incision. Both numbers of patients were equal to 5 (50%) that has 1 CS and 2 CSs. The main symptom was a painful abdominal mass at the previous incision site. Six (75%) patients with pain symptoms were cyclic, and 2 (25%) were noncyclic. One patient had no symptom who was noticed incidentally while operating for myomectomy. The mean latency period (time from last CS to the beginning of symptoms) was 44,6 (min-max:6-88) months, and the mean duration between symptoms and surgery was 28,9 (min-max:2-60) months. Six (60%) of endometriomas were located right corner of the incision, and 4 (40%) were at the left. All of the endometriomas were solitary. The mean greatest diameter of masses was 30,2 (min-max:15-47). Ultrasonography (USG) was used for all nine patients to diagnose except one patient who noticed incidentally. Fine needle aspiration biopsy (FNAB) was used beside USG to support the diagnosis in one patient. We divided the abdominal wall into four layers to describe the boundaries of endometriomas: Adipose, fascial, muscular, and peritoneal. The Upper bound of all endometriomas was the adipose layer. The lower bound of 3 (30%) endometriomas were adipose layer while 2 (20%) was a fascial layer, 4 (40%) was a muscular layer, and 1 (10%) was peritoneal layer. All ten patients underwent total excision with a 1 cm safe margin, and one patient needed mesh repair for the significant fascial defect (Figure 1,2,3). The mean hospitalization time was 2,5 (min-max:1-6) days.

DISCUSSION

Even though endometriosis is a relatively common disease in women of reproductive age (10-15%) AWE is rarely seen. As most AWEs fallows a cesarean section, hysterectomy, tubal surgeries, appendectomy, and amniocentesis can lead to development. Even a case series about trocar site endometriosis has been reported [14]. Although abdominal endometriosis often occurs secondary to surgery near or within scar tissue, rare spontaneous cases without surgical incision have also been reported [15].

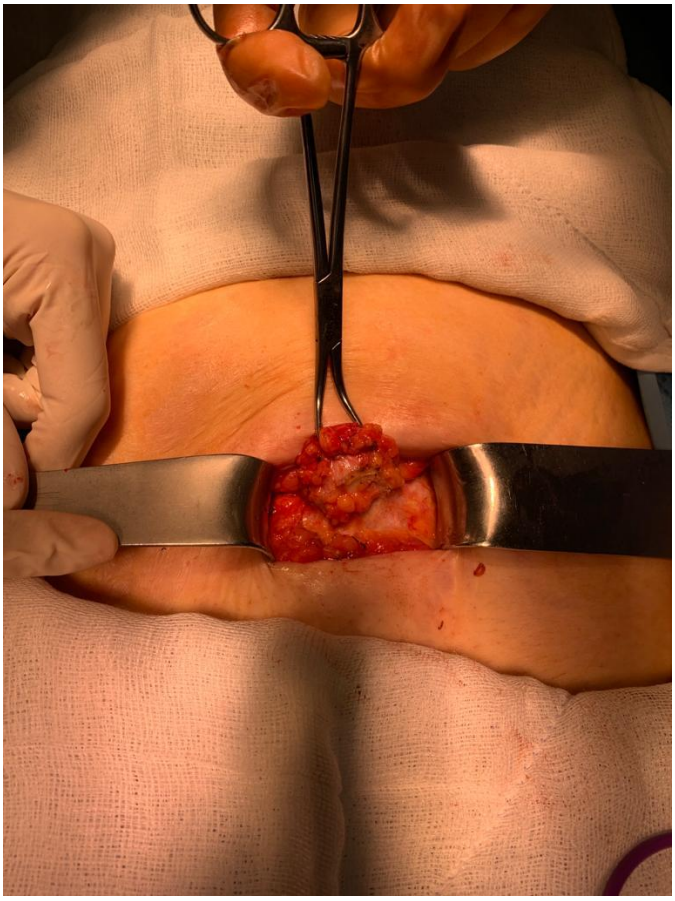

Figure 1. Appearance of the endometriotic focus after the incision

The reported incidence of CSE is 0.03-0.45%. The most frequently prominent theory in the pathogenesis of CSE is that implantation theory is caused by directly seeding of endometrial cells to the incision site [16]. Therefore, CSE is called by some 'iatrogenic' endometriosis. Low concurrent intrapelvic endometriosis rates support this theory [17]. No synchronous intrapelvic endometriosis existed in our study. However, most probably, pathogenesis is more complex than that. Endocrine, immune and inflammatory pathways must be considered. Under proper nutrient and hormone stimuli, endometrial cells survive and proliferate in the wound and eventually leads to CSE.

Pain and palpable mass at the cesarean scar site of the abdominal wall are the most characteristic symptoms of CSE. Patients with pain complaints mostly describe their pain as cyclic that increases during menstruation. Zhang et al. reported 86,9% cases to have cyclic type pain in their study, including 198 patients [11]. Noncyclic pain can complicate the diagnosis. Superficially located endometriomas may cause skin discoloration, discharge, or bleeding. It is pointed out that utero-cutaneous fistula formation is probable [18]. Also, some patients can be symptoms free like one of our patients who noticed during a myomectomy operation. Because of the slow-progressing nature of CSE, it is reported up to 17,7 years (mean 30 months) asymptomatic period (duration between last CS and onset of symptoms) in literature [19]. In our study mean asymptomatic period was 44,6 months (min-max:6-88). It can sometimes be difficult to diagnose correctly due to the nonspecific symptoms of CSE.

Many patients are followed up in general surgery clinics with pre-diagnoses such as hernia, lipoma, granuloma, sebaceous cyst, lymphoma, primary and metastatic tumors. In a study conducted by Yıldırım et al. with 24 patients, 54.2% were admitted to the general surgery clinic [20]. This can delay the appropriate treatment. Bektaş et al. reported the time between the onset of symptoms and surgery as 18.2 months (±23.4) in their study involving 40 patients [21]. This period means 28.9 (min-max:2-60) months in our study.

Table 1. Patient characteristics, symptom, location and size of mass with hospitalization time

Cases	Age	Parity	No of CSs	Symptom	Lesion location	Size of mass(mm)	Size of mass (mm)	Diagnostic method	Hospitalization time (days)
1	29	1	1	Mass, pain	Right	21×15	30×20×20	USG	1
2	22	1	1	Mass	Right	27×25	35×35×30	USG	1
3	33	2	2	Mass, pain	Left	32×16	50×25×20	USG	3
4	36	2	2	Pain	Right	37×28	50×40×30	USG	2
5	43	3	1	Mass, pain	Left	29×20	40×35×30	USG	1
6	38	1	1	No	Left	-	30×20×20	Incidental	3
7	34	2	2	Mass, pain	Left	31×20	40v25×25	USG, FNAB	2
8	31	2	1	Mass, pain	Right	15×10	25×20×15	USG	1
9	39	2	2	Mass, pain	Right	33×25	90×50×40	USG	6
10	45	2	2	Mass, pain	Right	47×27	50×45×40	USG	5

USG: Ultrasonography, FNAB: Fine needle aspiration biopsy

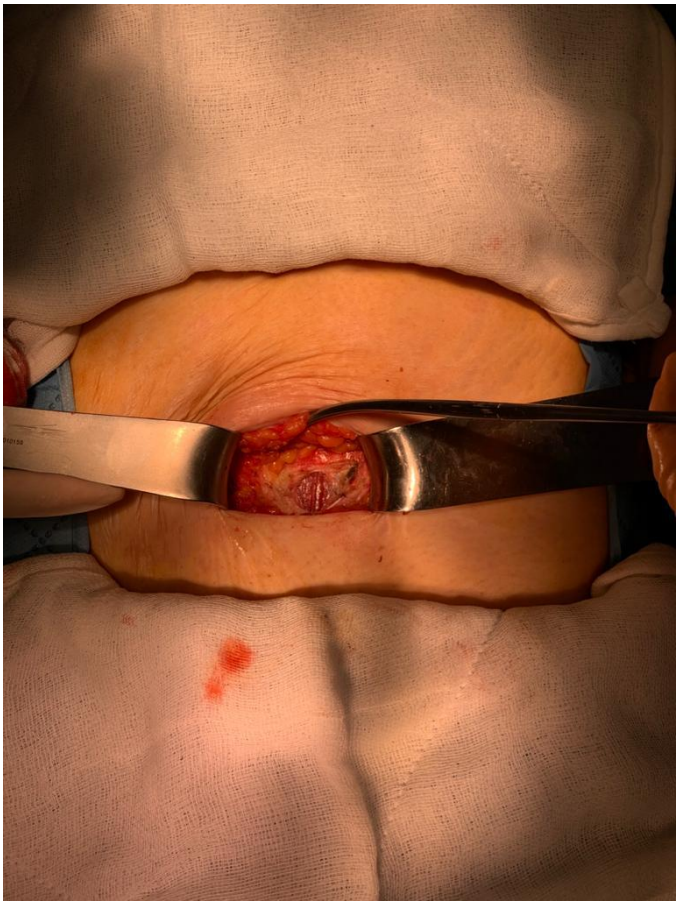

Figure 2. Fascial defect after removal of the endometriotic focus

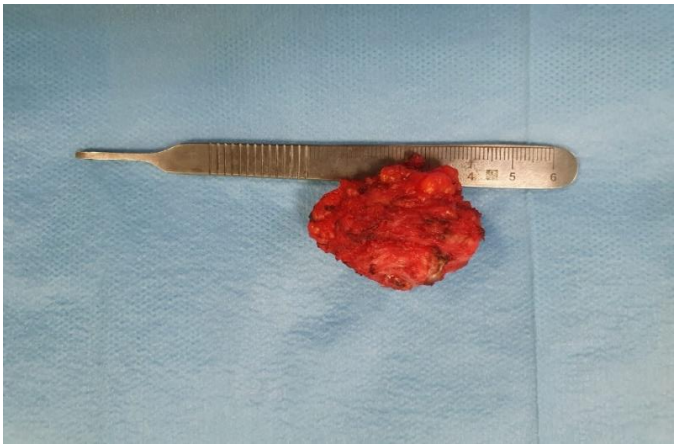

Figure 3. Macroscopic view of the removed endometriotic mass

Ultrasonography (USG) is the most commonly used and easiest to reach imaging method in cases suspected of CSE.

The sonographic appearance of CSE is nonspecific, and it changes due to the phase of the menstrual cycle, chronicity of lesion, amount of inflammation, and proportion of glandular and stromal cells. It can be cystic, solid, or heterogeneous. Intramuscular endometriomas appear as isoechoic with muscle or mild hypoechoic masses. Hyperechogenic halo may be seen in surrounding tissues due to edema and inflammation. Peripheral or internal vascularization can be observed in doppler at large lesions. The findings of CSE are also nonspecific on computed tomography (CT) and magnetic resonance imaging (MRI), but they can give valuable information about the exact size and extension of masses. The most prominent finding on CT examination is a soft tissue mass with intense contrast enhancement.

Table 2. Distribution of the patients according to the pain type and the localization layer of the mass, latency period and duration between symptoms and surgery

Features		n(%)	Mean(min-max)
Type of pain	Cyclic	6(75)	
	Noncyclic	2(25)	
Latency period(months)			44.6 (6-88)
Duration between symptoms and surgery(months)			28.9 (2-60)
Lower bound of endometriomas	Adipose layer	3(30)	
	Fascia layer	2(20)	
	Muscular layer	4(40)	
	Peritoneal layer	1(10)	

MRI is superior to CT in distinguishing masses from muscle and subcutaneous tissue and is more effective in detecting small lesions [22]. Also, MRI can give information about if there is concurrent intrapelvic endometriosis. In addition to imaging methods, FNAB may be helpful to confirm the pre-treatment diagnosis and exclude malignancy risk histologically. However, it should be borne in mind that there is a risk of creating a new focus in the entry line [23]. Surgery is the primary treatment for CSE, including recurrences. Wide resection with a 1 cm margin is the accepted method. Postoperative hernia risk is increased in patients with significant myofascial defects after resection. Therefore, it would be an appropriate approach to use synthetic proline mesh in such cases. We performed mesh repair for this purpose in one of our patients. The probability of recurrence after surgery is 4.3% [24]. Recurrences usually occur in patients whose lesion margins cannot be precisely determined, so the mass could not be removed with a safe margin and multiple lesions. Medical therapy has low efficacy in CSE, unlike intrapelvic endometriosis, and provides only temporary relief. It has been reported that nonsteroidal anti-inflammatory drugs, combined oral contraceptives, gonadotropin-

releasing hormone analogs progestins can be used to relieve pain to gain time and reduce the size of the mass before surgery [24,25].

Also, in the literature, some authors suggest newer implementation, such as high-intensity focused ultrasound ablation. Moreover, results have been reported to be satisfactory compared with surgery [26-28]. Since the primary pathophysiology of CSE is the direct seeding of endometrial cells on the abdominal wall during surgery, it has been recommended as preventive measures to repair the uterus outside the abdominal cavity, not to use compresses to clean the endometrial cavity, not to use sutures and instruments used to close the uterus on the abdominal wall, and wash the subcutaneous layers with saline [24].

CONCLUSION

Although its etiopathogenesis has not been fully elucidated, iatrogenic transplantation of endometrial tissue to the wound site after obstetric and gynecological interventions is the most accepted theory in the development of AWE. Therefore, clinicians have a higher chance of encountering CSE due to the increasing number of cesarean deliveries. History plays an essential role in diagnosis. It should be kept in mind in the differential diagnosis of women with complaints of cyclic pain and a mass at the scar line after a previous cesarean section. Surgical removal of the endometriotic focus with a safe margin appears to be the only effective treatment. The relatively rare nature of CSE limits what we know about it, and therefore studies involving more patients are needed.

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