

Abdominal wall endometriosis in patients with a history of cesarian section



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OBJECTIVE: *The aim of this study is to review the characteristics, intraoperative and radiological findings of abdominal wall endometriosis (AWE).*

METHODS: *This retrospective observational cohort study was executed through analysis of the medical records of patients who underwent excision of AWE between January 2000 and June 2017. All the diagnoses were confirmed pathologically. Characteristics, intraoperative and radiological findings of patients with AWE were and analyzed.*

RESULTS: *Each of the 20 patients had a history of at least one prior cesarean section. The main presenting symptoms were pain (70%). Ultrasonography and/or magnetic resonance imaging was performed in 95% and 45 % of the patients, respectively. One patient (5%) was investigated by 18 Fluorodeoxyglucose positron emission tomography - computed tomography. The preoperative radiological diagnosis was correcting in 55 % of the cases. The mean diameter of the masses was 4.7 ± 1.53 cm. Recurrence was found only in one patient during 36-month follow-up.*

DISCUSSION: *Meticulous anamnesis, accurate clinical examination and proper imaging studies, are important guides for diagnosis.*

CONCLUSION: *AWE should be kept in mind when pain or mass is detected on the abdominal wall of women who have cesarean section history.*

KEY WORDS: Abdominal wall endometriosis, Cesarean section, Radiology, Scar endometriosis

Introduction

Endometriosis is defined as ectopic implantation of endometrial tissue outside the uterine cavity and is an enigmatic disease affecting 10-15% of women of reproductive age^{1,2}. Extrauterine endometrial lesions are com-

monly found in the genital organs and pelvic peritoneum, although they may also be seen in the gastrointestinal system, greater omentum, surgical scars, round ligament, mesentery, and occasionally in the kidney, lung, skin, umbilicus and rectus abdominis muscle³⁻⁶.

Abdominal wall endometriosis (AWE) is defined as endometrial tissue superficial to the peritoneum and is associated with previous surgical procedures^{7,8}. These lesions almost always lie in the territory of the previous surgical scars. Patients with AWE may initially apply to general physicians, surgeons or dermatologists instead of gynecologists because of atypical presentation patterns of the disease^{9,10} and this is an eligible reason to keep AWE on the agenda. The aim of the present study is to draw attention to scar endometriosis in the abdomi-

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nal wall. Moreover, it is anticipated that the detailed analysis of clinical features, intraoperative and radiological findings we put forth with this monocentric retrospective study, will contribute to the existing literature about this rare clinical entity.

Materials and Methods

This retrospective observational cohort study was performed through analysis of the medical records of patients who underwent excision of AWE between January 2000 and June 2017 in Goztepe Training and Research Hospital of Istanbul Medeniyet University. Approval from the local ethics committee was obtained before the execution of this study. Twenty patients with the diagnosis of AWE were included. All the diagnoses were confirmed histopathologically. Endometriosis in the cicatrix is being visible in routinely hematoxylin and eosin stained slides. It appears as a presence of endometrial stromal cell focusses usually with concomitant endometrial glands in deeper layers of the skin, subcutaneous tissue, sometimes also among skeletal muscle fibers. The endometriosis focus is usually embedded in fibrosing (reactive fibrosis) surroundings¹¹. The follow-

ing data were collected and analyzed: patient age, surgical antecedents, history of endometriosis, symptoms, duration of complaints, asymptomatic time interval, size, number and location of the masses, diagnostic imaging studies, initial diagnosis, recurrences, follow-up time and utilization of hormone therapy.

STATISTICAL ANALYSIS

Continuous variables are presented as the mean±SD (range) while non-continuous variables are presented as number (percentage). Statistical analyses were performed using R Statistical Software (www.r-project.org), a free software environment for statistical computing and graphics. $P < 0.05$ was considered statistically significant.

Results

Twenty patients with pathologically confirmed AWE were included in the study.

Mean patient age was 36.2 ± 6.7 years (min: 23, max: 52 years). Each case had at least one prior cesarean section with Pfannenstiel incision, three of the patients had

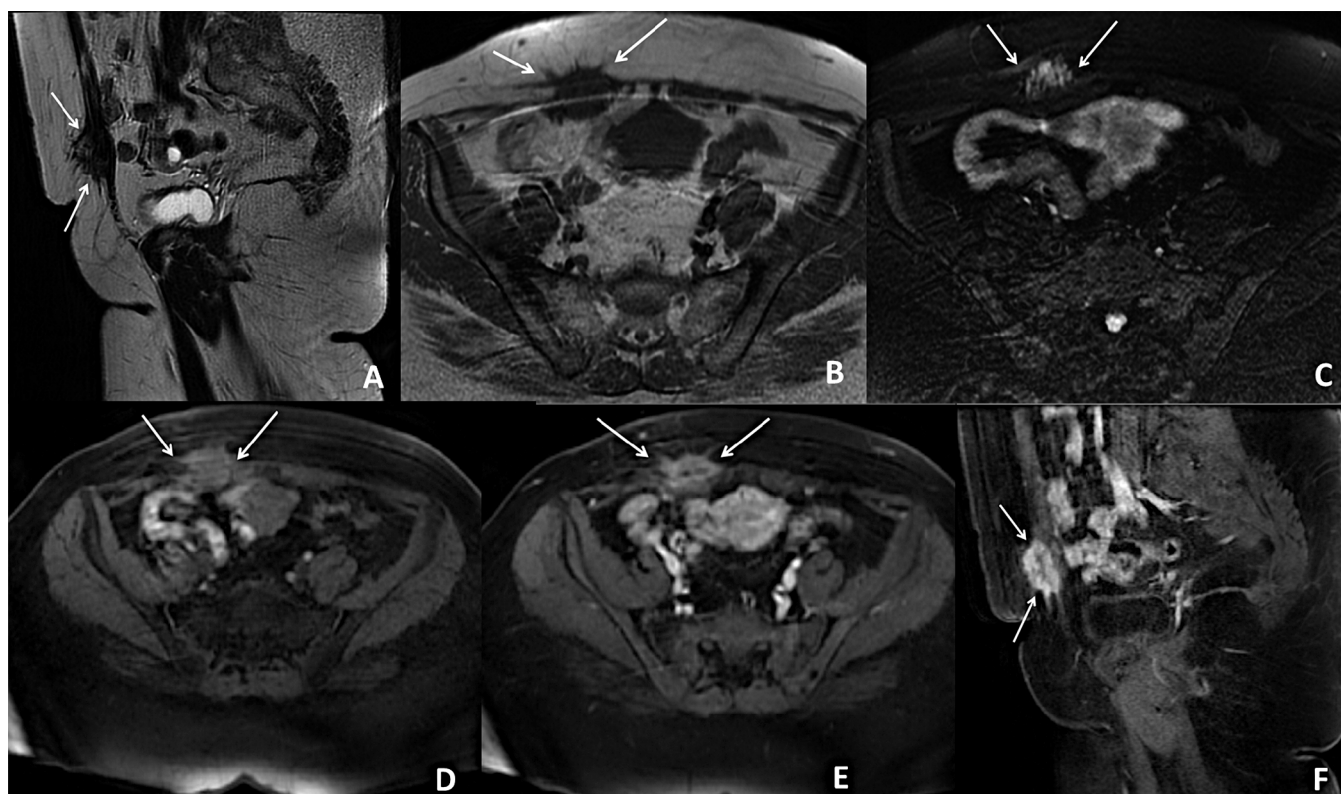


Fig. 1: 45-year-old-patient with cesarean section history 11 years ago. There is a 26x18x32 mm hypointense mass (arrows) above the right rectus abdominis muscle on the sagittal T2 weighted image (A). Mass has speculated margins and isointense on the axial T1 weighted image (arrows) (B). Mass is hyperintense on the axial fat saturated T2 weighted image, isointense on the axial non-enhanced fat saturated T1 weighted image, and markedly enhances after contrast admission on axial and sagittal fat saturated T1 images (arrows) (C, D, E, F).

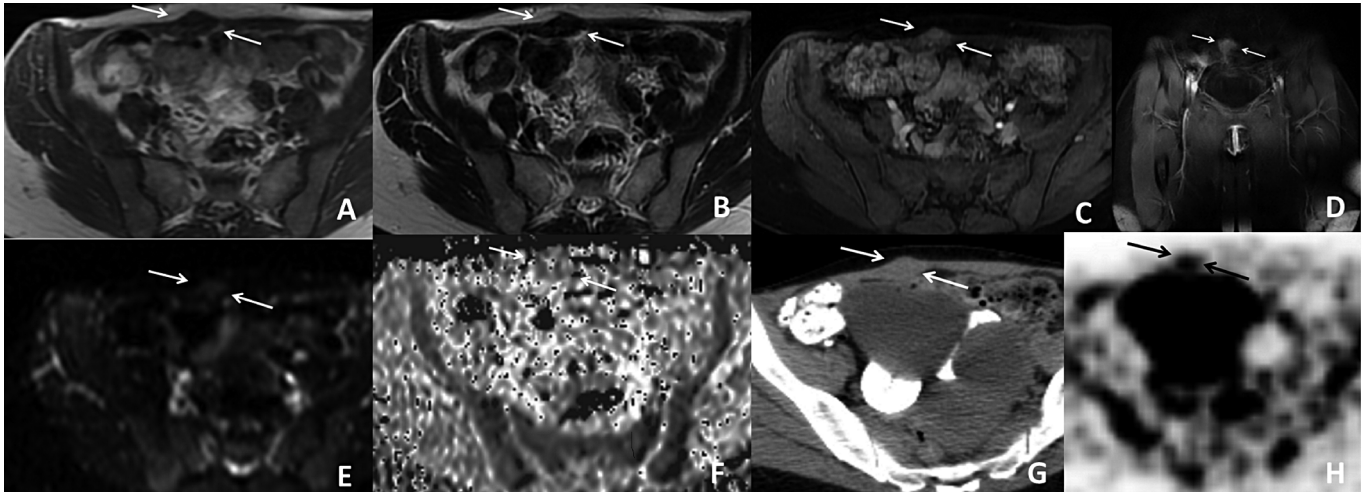


Fig. 2: 35-year-old patient presented with complaints of abdominal pain. On T1 weighted image a 21x11x17 mm isointense mass is located at right rectus abdominis muscle (arrows) (A). Mass is hypointense on the T2 weighted image and markedly enhances on contrast enhanced fat saturated axial and coronal T1 weighted image (arrows) (B, C, D). Mass is hyperintense on diffusion weighted images, and hypointense on ADC map compatible with restricted diffusion (arrows) (E, F). On 18- FDG-PET CT images, mass is isodense and shows mild FDG uptake (SUVmax=1.6) (arrows) (G, H).

TABLE I - Characteristics and symptoms of patients with abdominal wall endometriosis

	N	%	Range	Mean
Age (years)			23-52	36.2 ± 6.7
Previous operation and incision				
One C/S † with Pfannenstiel	16	80		
Two C/S † with Pfannenstiel	3	15		
Three C/S † With Pfannenstiel	1	5		
Asymptomatic time interval (years)			0.3-11	3.65 ± 3.05
Duration of symptoms (years)			0.08-11	2.65 ± 3.25
Endometriosis History Yes No	218	1090		
Symptoms Palpable mass Painful mass Cyclic pain Noncyclic pain	42104	20105020		
Diagnostic tests US ‡ MRI § PET/CT	1991	95455		
Radiological diagnosis Accurate Wrong	119	5545		

† C/S, cesarean section; ‡ US, ultrasonography; §MRI, magnetic resonance imaging; ||PET/CT, positron emission tomography-computed tomography

undergone cesarean section twice and one of them thrice. The mean time interval between the most recent surgery and the onset of symptoms was 3.65 ± 3.05 years (range, 4 months to 11 years). The mean duration of symptoms was 2.65 ± 3.25 years (range, 1 month to 11 years). Except for two patients who had endometriosis history, none of the patients received medical therapy until the time of excision of abdominal wall endometriomas. The main presenting symptoms were pain (70%, n=14/20), either cyclic (71%, n= 10/14) or noncyclic (29%, n= 4 /14); palpable abdominal mass (20%, n=4/20) and painful mass (10%, n=2/20). None of the patients had symptoms of pelvic endometriosis. Ultrasound was the only imaging study in 11 patients (55 %) and magnetic resonance imaging (MRI) was the only imaging study

in one patient (5 %). Seven patients (35 %) had both ultrasound and MRI (Fig. 1). One patient (5 %) was investigated by ultrasound, MRI and 18 Fluorodeoxyglucose positron emission tomography - computed tomography (18-FDG PET-CT) (Fig. 2). Correlation between preoperative radiological imaging and final pathological diagnosis revealed that the preoperative initial diagnosis was correcting in 55 % (n=11/20) of the cases. Remaining 9 patients had been initially diagnosed as desmoid tumor (35 %, n=7), suture granuloma (5 %, n=1) and fibroma (5 %, n=1). Characteristics and symptoms of patients are summarized in Table I. The mean diameter of the masses was 4.7 ± 1.53 cm (range, 3– 8.5 cm). According to the macroscopic observations during surgery, the exact locations of

TABLE II - Clinical data of patients and results of the study parameters

	N	%	Range	Mean
Mass diameter (cm)			3-8.5	4.7 ± 1.53
Depth of invasion Fat layer Fascia Muscle layer	848	402040		
Location of mass Right side Left side Middle	992	454510		
Repair of Defect Primary Prolene mesh	191	955		
Follow-up(months)			2-37	18 ± 15.38
Postoperative recurrence Yes No	119	595		

the masses were subcutaneous fat layer (40%, n = 8/20), the fascia (20%, n = 4/20), and the muscular layer (40%, n = 8/20). The mass was located on the left side of the wound in 9 (45%) patients; on the right side in 9 (45%) patients; and in the middle in 2 (10%) patients. Clinical data of patients and results of the study parameters were shown in Table II. All patients were treated surgically, with wide excision polypropylene (Prolene; Ethicon, Edinburgh, United Kingdom) mesh was used in one patient. After the first surgery for abdominal endometriosis, none of the patients were treated by medical therapy. Six patients were lost to follow-up. Regarding these 6 patients, the follow-up time after mass excision averaged 18 ± 15.38 months (range 2-37 months).

Recurrence was found only in one patient during 36-month follow-up who was treated by re- excision with safety margins.

Discussion

Abdominal wall endometriosis (AWE) is the functional endometrial gland transfer to the lower abdominal wall. Two main hypotheses are invoked to explain its cause. One suggests that multipotential mesenchymal cells undergo metaplasia under the proper circumstances, resulting in endometriosis; the other theory states that endometrial cells are transported to ectopic sites form an endometrioma^{3,12,13}. The most common site for extrapelvic endometriosis is the Pfannenstiel scar with an incidence of 0.07%-0.47% and is known as cesarean scar endometriosis^{7,14-16}. Our findings are compatible with the theory of iatrogenic cell transportation, since all the patients had a previous history of cesarean section procedure. It's a fact that some sporadic cases of scar endometriosis exists in women that did not previously have any type of surgery and it supports the coelomic metaplasia theory and suggests hematogenous spread and lymphatic dissemination as the possible explanations for its etiology^{8,17}. Scar endometriomas following cesarean sections occur more frequently than the ones in episiotomy wounds that follow vaginal labors; in both cases decidual cells keep in touch with the surgical incision¹¹. It might be explained by lower level of immune tolerance of the mother that had cesarean section compared to the ones that had vaginal delivery at term^{11,18-20}. Hui

suggested that because low levels of estrogen is maintained by lactation, breastfeeding can prevent the occurrence of postpartum abdominal wall endometrioma to a certain extent²¹. Zhang et al. found that postpartum breastfeeding, breastfeeding duration and menstrual recovery time were independent from the incubation period²¹. Accordingly, it has been suggested that local abnormal expression of estrogen and progesterone receptors in abdominal wall endometrioma leads to high estrogen producing levels in the ectopic endometrium²¹⁻²³. In our study, the mean age of patients was 36.2 ± 6.7, which is like other studies^{24, 25}. The younger mean age supports the observation that AWE affects young women of reproductive age²⁵. Pain caused by endometriosis is classically described as cyclic pain but constant and non-cyclic pattern also have reported (26). In the present study 80% of the patients had cyclic abdominal pain and painful abdominal mass, consistent with those reported in the literature^{4,8,21}. Also, in our study, the time from the most recent surgery to the onset of symptoms was 3.6 years which was like other series^{8,24,25,27}. Zhao et al. suggested that this latent period is positively related to patient's age at the onset of symptoms; as the more advanced the age of the patient, the longer the latency time²⁸. Time between the onset of symptoms and the definitive treatment with surgical excision was 2.65 ± 3.25 years (range, 1 month to 11 years) which parallels the findings in the literature^{10,25-27}. Previous studies have reported a delay in diagnosis ranging from 2.5 to 4.8 years²⁵. It might be explained that it is clinically often misdiagnosed. AWE is often confused with other pathologic conditions such as suture granuloma, abscess, inguinal or incisional hernia, soft-tissue sarcoma, desmoid tumor, lipoma, metastatic tumor and sebaceous cysts³. Especially, when the patient's symptoms are not cyclical and the history of endometriosis is unknown clinical diagnosis could be impaired. AWE is largely a clinical diagnosis²⁴. The diagnosis requires an accurate clinical examination combined with a detailed history. Palpation of the abdominal wall using superficial and deep palpation method is recommended. Supplementary diagnostic modalities may be necessary to confirm and clarify the diagnosis and to plan optimal surgical treatment options²⁹. The typical ultrasound finding is a hypoechoic nodule with speculated margins infiltrating the surrounding tissue³⁰. On color Doppler

examination, a single avascular pedicle entering the mass at the periphery is one of the diagnostic features³¹. Endometriosis has no pathognomonic findings on computed tomography (CT), as appearances depend on the phase of the menstrual cycle, the proportions of stromal and glandular elements, the amount of bleeding, and the degree of surrounding inflammatory and fibrotic response. Owing to the relatively vascular nature of these lesions, enhancement often occurs on CT scans when intravenous contrast material is used^{7,32}. Preoperative MRI is valuable in defining the extent of disease, thus enhancing accurate and total excision³³. The hemorrhagic signal is characterized by the presence of hypointense lesions with hyperintense foci on T1- and T2- weighted sequences and T1-weighted sequences with fat suppression; these correspond to the areas of hemosiderin found in the endometriotic crypts^{1,32}. The study by Zawin et al. revealed an MRI sensitivity of 71% and specificity of 82% for abdominal wall endometriosis and the authors suggested that MRI seems to be the best method for use in preoperative diagnosis^{10,34}. In our study, abdominal wall endometrioma has no pathognomonic findings on 18 FDG-PET CT. The mass is isodense and shows mild FDG uptake (SUVmax=1.6). According to a recent study, PET/CT is a good diagnostic tool for malignant transformation of endometrioma (MTOE) in cases where differentiation of MTOE from endometrioma was difficult despite the use of transvaginal ultrasound and MRI. An SUVmax cut-off >4.0 can exclude endometrioma, with 75 % sensitivity and 100 % specificity³⁵. Fine needle aspiration is inconclusive in up to 75% of the cases with theoretical concerns about further tissue inoculation with needle passage³⁶. Malignant transformation of abdominal wall endometrioma is a rare complication (1%), but the existence of such cases should also be sought.^{7,37,38} Medical therapy with danazol and gonadotropin-releasing hormone (GnRH) agonists produces only partial relief of symptoms and usually recurrence occurs after cessation of the treatment³³. Surgical management offers the best chance for both definitive diagnosis and treatment. Resection should be at least 0.5 to two cm distant from the lesion²¹. As was required in one of our cases, fascial defect may need closure with synthetic mesh if the underlying sheath is found to be involved. Local recurrence is likely after an inadequate surgical excision and in our series recurrence was found only in one patient. It is still controversial whether to use postoperative medication to reduce the recurrence risk. The study by Zhang and Liu revealed that postoperative medical treatment could improve the prognosis, reduce the recurrence²¹. A combination of surgical re-excision and postoperative adjuvant medical therapy is recommended for patients with recurrent AWE, especially for those with a history consistent with pelvic endometriosis²⁹. In addition to the treatment strategies, there are prevention methods based on the implantation theory: Using a wound edge pro-

jector to separate the edges of the incision; careful flushing and irrigating before closure; suturing the uterine incision without endometrium; using separate needles for uterine and abdominal closure; not using a sponge to clean the endometrial cavity following complete delivery of the placenta; removing a functional corpus luteum simultaneously with a hysterectomy; and extending the breastfeeding period to delay menstruation²⁹. The current study has some limitations. Firstly, it is a retrospective analysis. Retrospective reviews are subject to information bias in the form of missing or illegible data and/or errors in data collection²⁴. Secondly, it involves a small number of patients which can be attributed to the rarity of this condition.

Conclusion

The present study draws attention to AWE to facilitate early diagnosis in cases of pain or mass detected on the abdominal wall of women that have cesarean section history. Besides being a rare entity, clinical importance of AWE is increasing in parallel with the increasing popularity of elective cesarean section procedures. In addition to clinical studies, AWE is open to further in-vivo and in-vitro investigations in order not only to enlighten its etiopathogenesis but also to find out alternative methods of treatment and prevention.

Riassunto

Si tratta di uno studio retrospettivo osservazionale finalizzato a riconsiderare le caratteristiche, gli aspetti radiologici ed intraoperatori della di endometriosi della parete addominale (AWE), sulla base delle cartelle cliniche di pazienti sottoposte ad exeresi chirurgica della lesione nel periodo compreso tra Gennaio 2000 e Giugno 2017. La diagnosi risulta confermata per tutte con l'anatomia patologica, e sono state analizzate i rilievi radiologici ed i reperti intraoperatori.

Ciascuna delle 20 pazienti della casistica presentava una storia di almeno un precedente taglio cesareo, ed il principale sintomo lamentato (70%) era il dolore. Lo studio diagnostico si è basato sugli ultrasuoni nel 95% dei casi e sulla RMN nel 45%. Una paziente (5%) è stata studiata con la PET mediante 18 Fluorodeoxyglucose.

La diagnosi radiologica preoperatoria è risultata corretta nel 55% dei casi. Il diametro medio della massa era di 4.7 ± 1.53 cm. Si è registrata una sola recidiva in una paziente lungo un periodo di controllo di 36 mesi.

Guida essenziale per la diagnosi solo l'anamnesi accurata, un attento esame clinico ed adeguate indagini strumentali per imaging.

L'endometriosi della parete addominale va sospettata in presenza di una massa nella parete addominale o dolore in donne già sottoposte a taglio cesareo.

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