Intramuscular Abdominal Wall Endometriosis Treated By Ultrasound-Guided Ethanol Injection

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Running title: Endometriosis treated by ethanol injection

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Received: June 17, 2013
Revised: September 18, 2013
Accepted: October 30, 2013
doi:10.3121/cmr.2013.1183
Abstract

Abdominal wall endometriosis (AWE) is a rare condition, which usually develops in a surgical scar resulting from a Caesarean section. While commonly seen in the cutaneous and subcutaneous fat tissue at the Caesarean scar level, its intramuscular localisation is quite rare. Its treatment options consist of the excision of the lesion and/or hormonal therapies, although wide surgical excision is the treatment of choice in the literature. Wide surgical excision may create a defect in the abdominal wall and the risk of hernia formation and mesh complications also increases. This case report describes the clinical and radiological findings and treatment modalities of endometriosis that has appeared in the rectus abdominis muscle of a 25-year old patient at the Caesarean scar level. Sclerotherapy may used for endometrioma. We present a new and alternative treatment method using ultrasound guided intralesional ethanol injection for AWE. Compared with the complications of surgical excision, the complications of sclerotherapy by ethanol are at a more acceptable level. So, sclerotherapy by ethanol injection may be an alternative treatment to surgery.

Keywords: Endometriosis, abdominal wall, sclerotherapies, intramuscular endometriosis, magnetic resonance imaging, fine needle aspiration cytology
Introduction

Endometriosis is described as the presence of endometrial tissue outside the uterine cavity. Endometriosis is most commonly located in the ovaries, bowel or the tissue lining in the pelvis. It may develop in virtually every organ in extrapelvic sites. The abdominal wall is an uncommon site of the extrapelvic location where it mostly occurs in an old surgical scar. AWE appears following the implantation of endometrial cells into soft tissues of the abdominal wall after open uterus surgeries such as Caesarean sections. The incidence rate is reported at 0.4-0.1% (1). The disease is characterised with the triad: mass in the abdominal wall, periodic pain associated with menses, and history of operation. Its treatment is recognised to be broad surgical excision. There are reports suggesting that successful results can be achieved in patients with pelvic endometriosis by injecting ultrasound-guided alcohol into the lesion and performing sclerotherapy as well as surgical treatment (2, 3). However, as far as we are concerned, the literature contains no cases of intramuscular AWE treated by injecting ultrasound-guided alcohol. This case report describes the clinical and radiological findings of a patient who had intramuscular AWE following Caesarean section and received sclerotherapy by injecting ultrasound-guided alcohol.

Case report

We report a 25- year-old G2 P2 A0 C0 female patient who underwent a Caesarean section 7 years ago for post term pregnancy and had not response to induction. The patient, who underwent a second Caesarean section 4 years ago, was admitted to another clinic 26 months after this operation with complaints of pain in the Caesarean scar and palpable mass
in the anterior wall of the abdomen. Hematoma in the rectus abdominus muscle was suspected in CT and US scans, and palliative treatments were performed at other hospitals. The patient’s complaints were not resolved with medical treatment, and the patient was admitted to the Clinic of Obstetrics and Gynaecology, Universal Malatya Hospital with complaints of increased pain and increase in mass size. The patient stated that the pain was cyclic, intensified particularly in the first 2 days of menstruation and significantly restricted her daily activities. The patient had no history of any chronic diseases, previous ovarian cyst surgeries, and endometriosis. The gynaecological examination was normal. Routine laboratory findings were in the normal range. Abdominal examination showed a hard and partially mobile, painful and palpable mass lesion, about 3x 1.5 cm, located in the right side of the Caesarean scar.

US was performed with LOGIQ 9 ultrasound system (GE Healthcare Technologies, Ultrasound, Milwaukee, USA) by using linear (12-14 MHz) and convex (2-5 Mhz) transducer. Superficial US scan showed a heterogeneous hypoechoic, irregularly marginated lesion, 30x14 mm, in the lower right quadrant in the rectus abdominis muscle (Image 1B). The patient’s contrast enhanced abdominal CT scan taken at another hospital showed an asymmetrical thickening at the inferior portion of the left rectus abdominus muscle and hyper- intense mass that would suggest hematoma (Image 1A).

Lower abdominal magnetic resonance imaging (MRI) was performed on the patient with a 1.5-Tesla MR device (General Electric SignaExcita, GE Health care system Milwaukee, USA). The scan protocol included axial T1 fast spin echo, axial and sagittal T2-weighted
fast spin echo sequences, axial and coronal fat sat proton density sequences, diffusion weighted axial sequences and pre and post contrast axial and coronal T1-weighted fast spoiled gradient-echo fat-suppressed sequences. MRI imaging showed a weakly contrasting mass, 3x 1.5 cm, containing heterogeneous hypo- hyperintense areas in the suprapubic region on the right rectus abdominus muscle in the T1 and T2A sequences causing asymmetric thickening (Image 2 A, B, C, D). Diffusion-weighted images showed a slight signal increase secondary to restricted diffusion in the mass. ADC values were found to be $0.95 \times 10^{-3} \text{ mm}^2/\text{sn}$. With these findings, aspiration biopsy was performed on the patient with US considering AWE in the patient. The patient was diagnosed with intramuscular AWE after observing the endometrial glands, stroma and smooth muscle cells following post- biopsy microscopic analysis. Sclerotherapy was planned for the patient by injecting alcohol into the patient’s lesion. Approval was obtained from the patient and the hospital’s ethical committee. After local anaesthesia, 1 cc 95% ethanol was injected with the US device to various parts of the lesion using a 22 G needle (Image 3). After the operation, pelvic US and pelvic MRI were performed to check for any complications. Fine needle aspiration cytology confirmed the endometriosis. In the cytology the epithelial clusters showed orderly spaced nuclei arranged in a honeycomb pattern (Image 4). Oral contraceptives were started for 3 months to reduce the risk of recurrence. 3 months later, hormonal treatment was stopped, followed by a 3-month additional follow-up. The patient reported that her pain completely disappeared in the 9-month follow-up, and no recurrence was observed.
Discussion

The abdominal wall is an uncommon site of extrapelvic endometriosis, where it usually develops within the skin or subcutaneous tissues of the abdominal wall. Endometriosis involving the rectus abdominis muscle is rare. The differential diagnosis of the rectus abdominis muscle mass lesions includes hernias, lipomas, hematomas, abscesses, benign and as well as malignant tumors (4). Most of the abdominal wall endometriosis is located in the old surgical scars resulting from invasive abdominal-pelvic surgery. The aetiology of these foci of endometriosis is thought to be an iatrogenic transfer of endometrial cells into the surgical or procedural wound (5).

The literature mostly contains single case reports about abdominal wall endometriosis as well as case series. A study conducted on 445 patients has reported that the most important clinical finding in abdominal wall endometriosis was palpable mass, located particularly on the corners at the level of the Caesarean scar and that this finding was accompanied by a prevalence rate of 96%. In 86% of the patients, pain was described as the main symptom and with a cyclic characteristic in more than half of them (57%). The mean time between clinical presentation and surgery was found to be 3.6 years (4). A retrospective study by Leite GK et al. covering 33 patients reported that endometriosis incidence caused by Caesarean scar was 0.29%, and endometriosis incidence caused by episiotomy scar after vaginal delivery was 0.01%. It should be noted that pain cannot be cyclic at all times. It has even been reported that non- cyclic pain was seen more commonly in certain series. The main symptom has been reported to be cyclic at a rate of 66.7%. Caesarean section has been reported to be the major risk factor in abdominal wall endometriosis, and it has been observed that a previous Caesarean section increased the relative risk of AWE 27 times (6,
The palpated painful mass in our case was screened on the right side of the Caesarean scar. The pain was cyclic and increased during menstruation, particularly in the first 2 days, significantly reducing the patient’s daily activities. The time between previous cesarean section and clinical symptoms was 2.1 years.

Despite the use of US, CT and MRI for the diagnosis of the endometriosis, there were no pathognomonic imaging findings for endometriosis. Its appearance depends on the stage of the menstrual cycle, the proportion of stromal and glandular elements, the amount of bleeding and the degree of surrounding inflammatory and fibrotic response. Due to these non-specific findings, a wide spectrum of disorders such as hernias, lipomas, hematomas, abscesses, benign and as well as malignant tumors presenting as a mass lesion in the abdominal wall should be considered in the radiological differential diagnosis (8). In our case, US showed an irregularly marginated intramuscular lesion with a heterogeneous echogenicity. The lesion could not be clearly distinguished from the scar tissue, mass or chronic period hematoma. CT showed hypertensive areas suggesting haemorrhage in the lesion. Mass hematoma could not be clearly distinguished. The literature reports several kinds of signal patterns seen in endometrioma using MR imaging in AWE, due to the different stage of blood products found within these implants have been described. In these studies, endometriosis appeared homogeneously hypointense or isointense, or heterogeneous with focal areas of high and low signal intensity, suggesting old hemorrhage or fibrosis on T2-weighted and T1-weighted fat-suppressed imaging. Recent developments in MR imaging make it possible to obtain reliable diffusion-weighted images of the abdomen. Diffusion MRI is a magnetic resonance imaging (MRI) method which allows for
the mapping of the diffusion process of molecules, mainly water, in biological tissues. Water molecule diffusion patterns can therefore reveal microscopic details about tissue architecture, either normal or in a diseased state. Several studies showed that diffusion-weighted imaging (DWI) may be useful for differentiating tumors according to their different cellular construction (8, 9). Regarding endometriotic cysts (endometrioma), previous studies found a tendency towards lower apparent diffusion coefficient (ADC) values compared with other pelvic cysts, which might be more closely related to blood concentration (10).

In our case, MR imaging showed lesions, consistent with haemorrhage and fibrosis, containing heterogeneous hypo- hyperintense areas on the rectus abdominis muscle in the T1 and T2-weighted sequences. Weak contrast enhancement was observed in the lesion after contrast material injection. There was increased signal intensity due to restricted diffusion in DW sequences, with ADC values measured at $0.95 \times 10^{-3} \text{ mm}^2/\text{sn}$.

US-guided Fine needle aspiration (FNA) can be useful and less-invasive method to confirm abdominal wall endometriosis. The sample may show tubular structures which are indicative of endometrial tissue and stromal cells to confirm the diagnosis of the endometriosis (9). Despite extremely rare reports of malignant degeneration, ruling out malignancy and allowing for a quick diagnosis are the other important features of the FNA (11, 12).
Hormonal therapy and surgical excision are routinely used in the treatment of AWE. Hormonal treatment offering temporary alleviation of symptoms, but recurrence is common after cessation of treatment (13). The recurrence rate after surgery reported in the previous studies is 4.3% (4). To avoid recurrence, wide excision is recommended. Size of the lesion and extent of the mass, especially when it involves the rectus abdominis muscle or peritoneum, have shown to be risk factors for recurrence (14). Literature review revealed no studies about the incidence of recurrence in AWE patients treated with ethanol injection.

In wide surgical resections, complications including foreign substance reactions, mesh migration and eventual incidence of hernia may appear due to the propylene mesh used (14). In the literature, abdominoplasty by polypropylene mesh was recommended for abdominal wall reconstruction in large lesions in order to reduce hernia development (14).

A review of the literature revealed several studies that have reported positive findings after injecting 95% ethanol into the endometrioma in patients with pelvic endometriosis (15, 16). With this in mind, we planned sclerotherapy by ultrasound-guided ethanol to the patient with intramuscular AWE. The patient’s pain completely disappeared after the treatment and no recurrence occurred in the 9-month follow-up. We believe that the major factors in such a successful treatment were the absence of a large endometriosis focus (3x1.5 cm), its occurrence only in the muscles, and lack of an intraperitoneal extension. Intralesional alcohol injection may result in difficult-to-repair necrosis on the anterior muscles of the abdominal wall in large lesions. Also, in endometriosis foci extending into the intraperitoneal region, it may cause complications including chemical peritonitis and severe
pain as a result of alcohol penetration into the peritoneum. Therefore, injections may be given in several sessions instead of a single session, in such patients.

Compared with the complications of surgical excision, the complications of sclerotherapy by ethanol are at a more acceptable level. So, sclerotherapy by ethanol injection before surgical resection may be used as the first option in treatment.

AWE incidence also increases in association with increased numbers of Caesarean sections. Although minimising the contact of swabs used to clean the endometrial cavity within the scar site, quickly removing them from the operation area, avoiding the use of suture material that was used to close the uterus in order to suture the scar site, and thoroughly washing the scar site with saline before closing it are recommended in order to prevent the growth of endometriotic focus from the scar tissue. No prospective studies are available on this subject matter.

In conclusion, if a previous surgical history exists in cases with no primary pelvic endometriosis, endometriosis should be considered in the differential diagnosis of the palpable abdominal anterior wall masses at the scar site. Although excision is the conventional treatment in abdominal wall endometriosis, care should be taken about potential post-surgical complications. Sclerotherapy used for endometriotic cysts has been reserved for those patients who have high surgical risk, are pregnant or refuse surgical intervention. In the literature US-guided aspiration and sclerotherapy with 95% ethanol provides a valid alternative to surgery in treating endometrial cysts (17). Contrary to
conventional treatments, the patients’ complaints are eliminated by sclerotherapy by US-guided ethanol injection into the lesion, a minimally invasive method, and an accompanying short-term hormonal treatment, and no recurrence develops in the short term. 95% ethanol injection into the intraabdominal endometriosis may be an alternative method to surgery. To our knowledge, this is the first case in the literature to reveal the success of this condition. Further investigations of large series are needed to compare the surgical operation with ethanol injection treatment.
References


Image 1: A) An asymmetrical thickening compared with the left at the inferior of the rectus abdominus muscle as well as a hypertensive state that would suggest hemorrhage. B) A heterogeneous hypoechoic appearance in the rectus abdominus muscle in the transverse plane in the USG.
**Image 2:** A, B) Fast spin echo T1 and T2-weighted axial C) coronal fatsaturated proton density D) A mass lesion with asymmetric thickening in right rectus abdominus muscle and a heterogeneous intramuscular hypo-hyperintense signal pattern in fast spin echo T2 sagittal images (arrows).
Image 3: A) Needle echogenicity is noticeable in the endometriosis focus in the rectus muscle and at the center of the lesion (arrow) immediately before ethanol injection.
Image 4: FNAC findings of endometriosis of abdominal wall. The epithelial clusters showing orderly spaced nuclei arranged in honeycomb pattern (pap- stain x 20).