

SURGICAL SCAR ENDOMETRIOSIS: A SERIES OF 14 PATIENTS AND BRIEF REVIEW OF LITERATURE

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Abstract

Background and aims. Endometriosis is a commonly encountered disorder in women of reproductive age, consisting of the presence of active ectopic endometrial tissue outside the endometrial cavity. Surgical scar endometriosis is a rare condition representing about 2% of all endometriosis cases. The purpose of this study was to assess the main characteristics, diagnostic tools and therapeutic options in abdominal wall endometriosis (AWE).

Methods. We have reviewed a series of fourteen cases with histopathological confirmation of AWE that were managed in our institution.

Results. The main characteristic of AWE were emphasized, showing that 78.57% of the patients had at least one previous caesarian section and that in only 57.14% of all cases an accurate diagnosis of AWE was established preoperatively.

Conclusion. A direct relationship between gynecological and obstetrical surgery and AWE is well established and as the caesarian section rates increase constantly, the awareness regarding AWE should also be increased.

Keywords: endometriosis, caesarian section, surgical scar, abdominal wall

Background and aims

Endometriosis is a benign disease in which functioning endometrial tissue is present outside the endometrium and the myometrium. It can affect up to 22 % of all females [1], though an estimated prevalence of 5–10% in the general female population is widely accepted [2]. Endometriosis is usually confined to the peritoneal and serosal surfaces of intra-abdominal organs (ovaries, fallopian tubes, peritoneum and recto-vaginal septum). Less common sites include the bowel, bladder, surgical scars, episiotomy, umbilicus, hernia sacs, lungs and pleura, kidneys and extremities [3].

Endometrial tissue has been identified in numerous procedure-related scars, as well as in the skin, subcutaneous tissues, and abdominal and pelvic wall musculature adjacent to the scars, a condition that is referred to as scar endometriosis [4]. The increasing rate of caesarean sections has raised concerns about both the short- and long-term maternal consequences of the procedure. The literature on

endometriosis as a sequel of caesarean section has focused on the risk of scar endometrioma, which has also been described at the episiotomy site in women who delivered vaginally [5].

Cesarean section scars are the most common site of anterior abdominal or pelvic wall endometriosis, with an estimated incidence of approximately 0.03%–0.4%. Some studies even report an incidence as high as 3.5% [2]. Performing a cesarean section before the onset of spontaneous labor may increase the risk for scar endometriosis, given that a higher incidence (1.1%) of scar endometriosis was reported after hysterotomy performed for midtrimester abortion [4,6].

Endometriosis is associated with substantial morbidity, being associated with a reduction in health-related quality of life, an increased risk for ovarian cancer, and possibly even malignant transformation within scar endometrioma. A previous caesarean section is known to be associated with an increased risk of chronic pelvic pain. Whether this condition is related to endometriosis is still unclear [5]. The main objective of our study was to assess the main characteristics, diagnostic tools and therapeutic options in abdominal wall endometriosis (AWE).

Manuscript received: 14.11.2016

Received in revised form: 13.12.2016

Accepted: 28.12.2016

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Material and methods

A retrospective observational cohort study was performed comprising all patients with histopathological confirmation of AWE after surgical excision performed at the 4th Surgical Clinic Cluj-Napoca, Romania and “Dominic Stanca” Clinic of Obstetrics and Gynecology, Cluj-Napoca, Romania. The patients’ medical records were identified from the comprehensive, noncomputerized surgical database of the two above mentioned hospitals from January 2011 to December 2015. Endometriosis was considered to be present when endometrial tissue was found in fibrous connective or skeletal muscle tissue in areas of focal hemorrhage or of active chronic endometriosis with fibrosis. The medical records of 14 patients diagnosed with AWE were reviewed following approval by the Local Ethics Committee of the “Iuliu Hatieganu” University of Medicine and Pharmacy, Cluj-Napoca, Romania, and clinical review board approval. We excluded patients in which endometriosis was limited to the peritoneal layer alone. The review data included age, parity, related past medical and surgical history, symptoms, asymptomatic time interval, location and tissue layer involvement, size, type of diagnosis, treatment, recurrence rate, specialty of primary surgeon, and incision type. The pathological diagnosis of endometriosis was confirmed in the presence of at least three of the following features: presence of endometrial stroma, endometrial-like glands and hemosiderin pigment [7].

Results

The study group is comprised of 14 female patients who presented to our clinic mainly for a palpable mass at the level of a previous surgical scar, painful at palpation or associated with abdominal pain at the surgical scar level. Patient characteristics are presented in Table I, while the symptoms and lesion characteristics are presented in Table II. All the patients included in the study had at least two histopathological features of endometrioma, as described above.

The average age of the patients included in the study was 34 ± 6.50 years. Patient parities ranged from one to three. All patients included in the study had previous uterine surgery. Eleven patients (78.57%) had at least one previous caesarian section, and three patients (21.42%) had a previous myomectomy. The mean duration of symptoms was 10.28 ± 5.91 months, ranging from four to twenty-four months. The mean asymptomatic period between the previous surgery and diagnosis of AWE was 18.28 ± 12.91 months, ranging from two to forty-eight months. Symptoms at presentation included the presence of a palpable mass at the level of the scar (78.57%), non-cyclic and cyclic abdominal pain (50%, 42.85% respectively), bleeding form mass (7.14%) and swelling of the affected area (7.14%).

Preoperative examinations included ultrasonography in all patients; computed tomography (CT) scan was performed on one patient; magnetic resonance imaging

(MRI) scan was performed on another patient; one patient had a histopathological specimen from an incomplete resected abdominal mass. The accurate diagnosis of AWE was established preoperatively in only eight patients (57.14%), the other six patients being diagnosed with granuloma (21.42%), hernia (14.28%), and abdominal wall tumor (7.14%). In all fourteen patients, the histological examination of the surgical specimen showed irregular endometrial glands surrounded by stroma with hemosiderin-laden macrophages and chronic inflammation (Figure 1, 2) with disease free margins. The mean diameter of the masses was 22.46 ± 8.21 mm, ranging between 11 mm and 35 mm.

Table I. Patient characteristics.

Characteristics		No. of patients (%)
Age (years)		
Median	34 (± 6.50)	
Range	26-55	
Parity		
Mean	1.50	
Range	1-3	
Previous abdominal surgery		14 (100)
CS		11 (78.57)

CS-cesarean section

Table II. Lesion characteristics and symptoms.

Characteristic	No. of patients (%)	Range (Mean \pm SD)
Previous surgery	14 (100)	
CS	11 (78.57)	
Other	3 (21.42)	
Asymptomatic period (months)		2-48 (18.28 \pm 12.91)
Symptoms		
Mass	11 (78.57)	
Cyclic pain	6 (42.85)	
Noncyclic pain	7 (50)	
Bleeding from mass	1 (7.14)	
Swelling	1 (7.14)	
Duration of symptoms (months)		4-24 (10.28 \pm 5.91)
Mass		
Solitary	12 (85.71)	
Multiple	2 (14.28)	
Mass diameter (mm)		11-35 (22.46 \pm 8.21)
Diagnosis		
US	14 (100)	
CT	1 (7.14)	
MRI	1 (7.14)	
FNAC	0 (0)	
HP of incomplete resected mass	1 (7.14)	
Accurate initial diagnosis	8 (57.14)	
Wrong initial diagnosis	6 (42.85)	

CS-cesarean section; US-ultrasonography; CT-computed tomography; MRI-magnetic resonance imaging; FNAC-fine needle aspiration cytology; HP-histopathology; SD-standard deviation

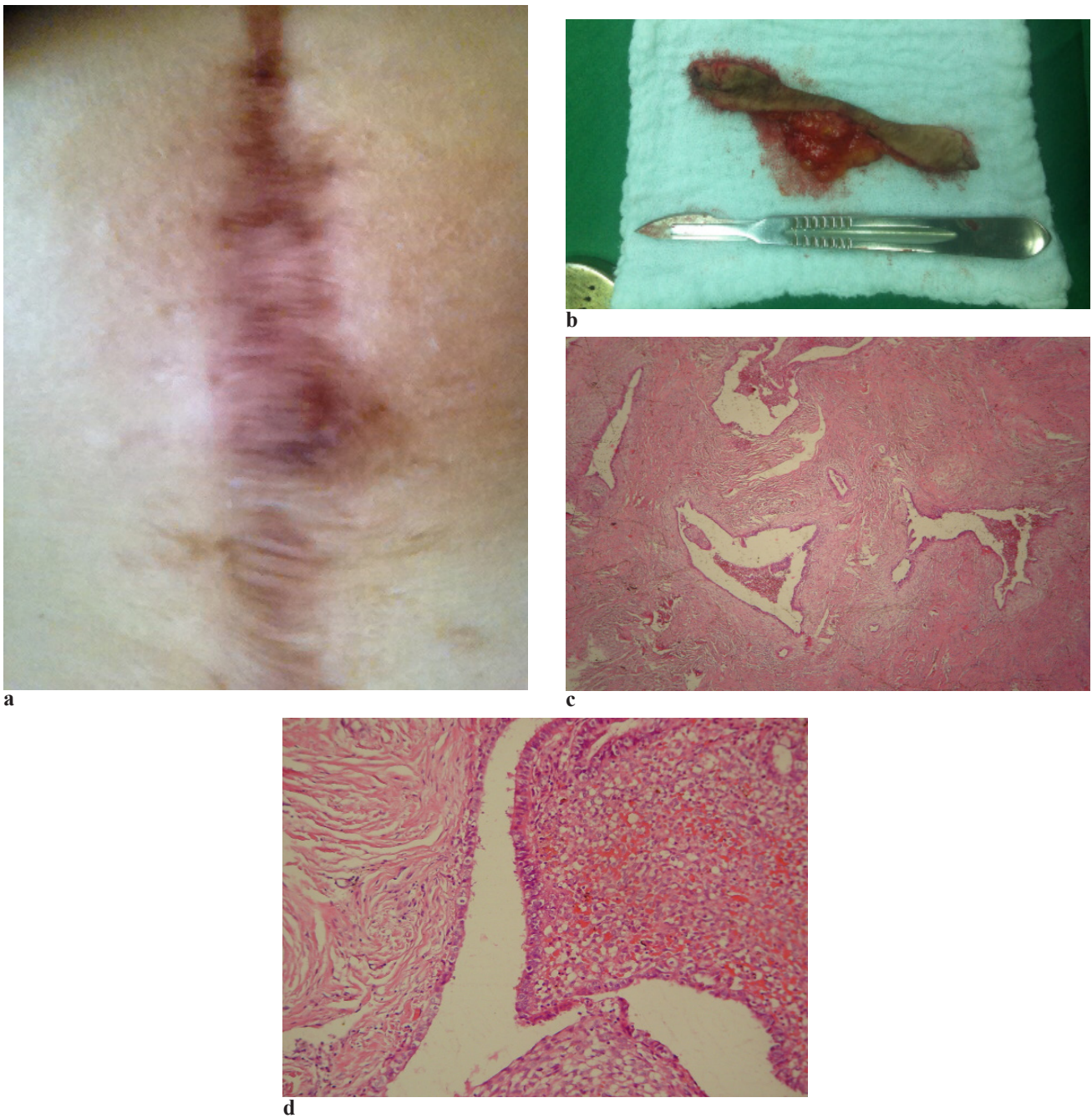


Figure 1. Abdominal wall endometriosis after caesarian section: **a)** Scar endometriosis in the region of caesarean section; **b)** Tissue removed after excision; **c, d)** Histological appearance of endometriosis.

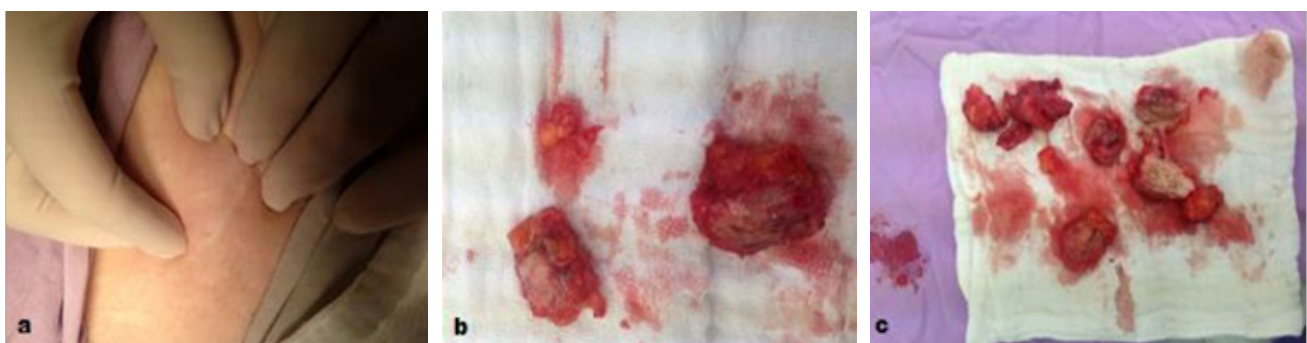


Figure 2. Abdominal wall endometriosis with a palpable mass: **a)** Caesarian section scar showing a palpable mass; **b, c)** Tissue removed after excision.

Discussion

Studies have shown a direct link between caesarean section and hysterectomy on the one hand and abdominal wall endometriosis on the other hand [8]. The surgical scar of the caesarean section is the most common site for extrapelvic endometriosis, known as caesarean scar endometriosis. This condition has an estimated incidence of 0.07%–0.47% [8]. In our study, all the patients had a previous obstetrical or gynecological surgical procedure involving the uterus, of which 78.57% caesarian section and 21.42% myomectomy. By comparison, one very recent study found surgical operations other than caesarian section in 26.7% of the studied patients. In contrast, all patients had at least one previous caesarian section in their medical history [8].

The pathogenesis of abdominal wall endometriosis is best explained by a combination of theories. One mechanism consists of the direct implantation of endometrial tissue during a surgical procedure on the endometrium. Another theory suggests the lymphatic and hematogenous routes as means of transportation of the endometrial cells to a cesarean section scar resulting in endometriotic foci. Finally, because the proliferative capacity of end-differentiated cells is limited, the most plausible cause of scar endometriosis is the transfer of endometrial stem cells into abdominal wall incisions at the time of uterine surgery, followed by proliferation at the new site [4,9].

Clinical symptoms of endometriosis are various, and some patients are asymptomatic. Patients with symptomatic scar endometriosis most often present a small tender abdominal or pelvic wall mass associated with a previous surgical incision. The surgical intervention is usually a cesarean section, hysterectomy, or another gynecological or obstetrical procedure. This association must raise the suspicion of scar endometriosis, especially if the pain is cyclical and corresponds to the patient's menstrual cycle [10]. Patients presenting with endometriosis often complain of dysmenorrhea, dyspareunia, irregular menstruation, chronic pelvic or back pain and infertility. Typically, clinical symptoms occur at the time of menstruation and include abdominal or pelvic wall pain and swelling; symptoms are less common between menstrual cycles. Rarely, cesarean scar endometriosis can take the form of an acute abdomen.

The clinical diagnosis is based on the patient's medical history and physical examination. A history of previous cesarean section, the presence of an expanding lump associated with the scar, symptoms of pain, bleeding and skin discoloration can be diagnostic clues for scar endometriosis. Cyclic symptoms such as bleeding or discharge from the scar during menstruation are not encountered in all cases, but, when present, they are pathognomonic for scar endometriosis [3]. All patients included in our study had at least one symptom associated with AWE. The most common situation was a palpable mass at the scar site (78.57%), followed by non-cyclic and

cyclic abdominal pain (50%, 42.85% respectively); more rarely bleeding from the mass (7.14%) and swelling of the affected area (7.14%) were noted. At the same time, a large recent study has found cyclic and non-cyclic pain as the main symptom of AWE in about 92.5% of the studied patients, abdominal mass in about 84.1% and dysmenorrhea in only about 4.4% of all patients [11].

As a relatively rare condition, abdominal wall endometriosis is often initially misdiagnosed. Most frequently AWE is mistaken for a suture granuloma, primary or metastatic tumor, hematoma, lipoma or hernia of the abdominal wall, thus resulting in unexpected findings at surgery or pathology [12,13]. One study has found a 63.3% rate of accurate initial diagnosis, abdominal wall tumor, incisional hernia and suture granuloma covering the other 36.7% [3]. In our series, we noted a very high rate of inaccurate clinical diagnosis (42.85%), patients being initially diagnosed with granuloma, hernia, or abdominal wall tumor.

Ultrasound (US) is usually the first imaging technique used to assess focal abdominal or pelvic wall thickening identified at clinical examination. Features of scar endometriomas at US vary and are nonspecific: lesions may be unilocular or multicystic, solid, or mixed cystic and solid. Ultrasound is useful for demonstrating the solid or cystic nature of the mass and its relationship with the fascia and skin [3,4,10]. CT and MRI are used in the assessment of a mass suspected to represent malignancy. In the study performed by Mistrangelo et al [3] ultrasonography was used to diagnose AWE in 93.3 of the studied patients, with CT scan and MRI being used 16.6% of all cases. In our study, US were performed in all patients, while CT and MRI were indicated in only one patient each.

Definitive diagnosis can be obtained by means of ultrasound guided needle aspiration cytology (FNAC) or excisional biopsy. US-guided fine-needle aspiration may help establish a preoperative diagnosis of scar endometriosis by excluding malignancy, thereby enabling pursuit of a definitive treatment [4,10,14]. In our series, the final diagnosis was obtained by excisional biopsy in all patients, except one case, in which the diagnosis was established by histopathological examination of an incompletely resected mass.

Management of AWE includes hormonal treatment and surgical resection. Surgery remains the mainstay of treatment, including disease recurrence. Due to the possible recurrence (4.3% after surgery) [15] and malignant degeneration (0.3–1%) of this condition, a local wide excision with at least a 1 cm resection margin is currently considered the best clinical practice [16], although no studies have so far evaluated whether the surgical margin width affects the recurrence rate [3]. Scar endometriosis incorporated into the muscular layer of the abdominal wall requires bulk resection of the underlying myofascial elements. As a result, surgeons should, therefore, be

prepared for the possibility of a coexisting hernia, and patients should be counseled that mesh repair may be necessary [3,17].

Other therapeutic options include pharmacologic therapy with hormonal suppression agents, such as progestogens or gonadotropin-releasing hormone (GnRH) analogs to down-regulate the hypothalamus-pituitary-ovarian pathway [10]. However, the success rate with medical therapy is low, offering only temporary relief of symptoms and is often followed by recurrence after the cessation of drug therapy [10]. Hormonal therapy with pituitary gonadotropin inhibitors is reserved for patients with concomitant pelvic endometriosis [4].

Preventive strategies for AWE are rarely mentioned in the literature. As direct inoculation during surgery is involved in the pathogenesis of AWE, it is recommended to follow good surgical technique during caesarean section and irrigation of the incision site before abdominal wall closure and to use separate sponges for cleaning the uterine cavity and skin wound [18].

The retrospective observational character represents a limitation of this study. The relatively small number of cases, as a limitation, is related to the rarity of this disease. In our series, none of the patients received pharmacological treatment, as surgery is considered to represent the best treatment.

Conclusion

In conclusion, as the direct relationship between gynecological and obstetrical surgery and AWE is well established and the caesarian section rates increase constantly, one can estimate that a greater number of patients with AWE will be diagnosed in the future. In case of preoperative suspicion of AWE, the treatment of choice should be local excision with clear margins, to prevent recurrence.

Acknowledgements

This paper was published under the project funded by "Iuliu Hatieganu" University of Medicine and Pharmacy, Cluj-Napoca, Romania, internal grant no. 4945/20/08.03.2016.

References

1. Moen MH, Muus KM. Endometriosis in pregnant and non-pregnant women at tubal sterilization. *Hum Reprod.* 1991;6:699-702.
2. Lu PY, Ory SJ. Endometriosis: current management. *Mayo Clin*

- Proc.* 1995;70:453-463.
3. Mistrangelo M, Gilbo N, Cassoni P, Micalef S, Faletti R, Miglietta C, et al. Surgical scar endometriosis. *Surg Today.* 2014;44:767-772.
4. Gidwaney R, Badler RL, Yam BL, Hines JJ, Alexeeva V, Donovan V, et al. Endometriosis of abdominal and pelvic wall scars: multimodality imaging findings, pathologic correlation, and radiologic mimics. *Radiographics.* 2012;32(7):2031-2043.
5. Andolf E, Thorsell M, Källén K. Caesarean section and risk for endometriosis: a prospective cohort study of Swedish registries. *BJOG.* 2013;120(9):1061-1065.
6. Francica G, Scarano F, Scotti L, Angelone G, Giardiello C. Endometriomas in the region of a scar from Cesarean section: sonographic appearance and clinical presentation vary with the size of the lesion. *J Clin Ultrasound.* 2009;37(4):215-220.
7. Dwivedi AJ, Agrawal SN, Silva YJ. Abdominal wall endometriomas. *Dig Dis Sci.* 2002;47:456-461.
8. Khamechian T, Alizargar J, Mazoochi T. 5-Year data analysis of patients following abdominal wall endometrioma surgery. *BMC Womens Health.* 2014 Dec 5;14:151. doi: 10.1186/s12905-014-0151-4.
9. Pados G, Tympanidis J, Zafrakas M, Athanatos D, Bontis JN. Ultrasound and MR-imaging in preoperative evaluation of two rare cases of scar endometriosis. *Cases J.* 2008 Aug 18;1(1):97.
10. Zhang J, Ding D, Liu X. Clinicopathological Features of Endometriosis in Abdominal Wall - Clinical Analysis of 151 Cases. *J Minim Invasive Gynecol.* 2015;22(6S):S174. doi: 10.1016/j.jmig.2015.08.644.
11. Ding Y, Zhu J. A retrospective review of abdominal wall endometriosis in Shanghai, China. *Int J Gynaecol Obstet.* 2013;121(1):41-44.
12. Savelli L, Manuzzi L, Di Donato N, Salfi N, Trivella G, Ceccaroni M, Seracchioli R. Endometriosis of the abdominal wall: ultrasonographic and Doppler characteristics. *Ultrasound Obstet Gynecol.* 2012;39(3):336-340.
13. Bazot M, Daraï E. Evaluation of pelvic endometriosis: the role of MRI. *J Radiol.* 2008;89(11 Pt 1):1695-1696.
14. Gupta RK. Fine-needle aspiration cytodiagnosis of endometriosis in cesarean section scar and rectus sheath mass lesions -- a study of seven cases. *Diagn Cytopathol.* 2008;36(4):224-226.
15. Horton JD, Dezee KJ, Ahnfeldt EP, Wagner M. Abdominal wall endometriosis: a surgeon's perspective and review of 445 cases. *Am J Surg.* 2008;196(2):207-212.
16. Honore GM. Extrapelvic endometriosis. *Clin Obstet Gynecol.* 1999;42:699-711.
17. Blanco RG, Parthivel VS, Shah AK, Gumbs MA, Schein M, Gerst PH. Abdominal wall endometriomas. *Am J Surg.* 2003;185(6):596-598.
18. Nigam A, Saxena P, Barla J, Pathak P. Scar endometriosis: the menace of surgery. *BMJ Case Rep.* 2014 Oct 15;2014. doi: 10.1136/bcr-2014-206693.