A non-classical presentation of scar endometriosis during pregnancy: Case report and review of literature

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ABSTRACT
Scar endometriosis is an uncommon condition in which endometrial tissue grows in a previous surgery wound site. The triad of scar endometriosis includes the history of caesarean section or any other gynecological surgery, cyclical waxing, and waning pain accompanied by the patient’s menstrual cycle and a tumor inside/near the scar site. Here, we present a case report in which the patient presented with endometriosis at the previous caesarean section scar site. Her body mass index (BMI) was 25.7kg/m2. An ultrasound scan revealed a well-defined hypoechoic and heterogeneous mass with internal echoes with smooth margins in the muscular plane of the suprapubic region measuring 6.23cm x 3.67cm x 2.28cm (Figure 1A). An endometrial cyst measuring 1.71cm x 1.66cm x 1.64cm and a clear cyst measuring 2.13cm x 1.77cm were found in her left ovary (Figure 1B). Her right ovary and uterus were normal. Ultrasound findings were suggestive of scar endometriosis. Magnetic resonance imaging (MRI) was performed to confirm the diagnosis. The scans revealed a relatively well-defined nodular lesion in the supra-pubic region within the bulky left rectus abdominus muscle, measuring 5.8cm x 3.3cm x 2.5cm. The lesion appeared heterogeneously hyperintense on T2WI (T2 weighted images) and STIR (short tau inversion recovery), showing a peripheral hypointense rim due to hemosiderin deposition (red arrow–Figure 2A, 2D) with post-contrast enhancement (Figure 2C).

Although the patient was offered surgery on account of the characteristic history and radiology findings, she preferred to undergo medical management. Hence, she was treated with dienogest tablets 2mg daily for 3 months, which improved her symptoms and reduced the size of the mass reduced (4.7cm x 3.5cm x 2.5cm). Since the patient was anxious to conceive, she was advised to discontinue treatment with dienogest and to try naturally for pregnancy. She conceived naturally after 4 months of stopping dienogest. She did not develop any symptoms during pregnancy, but could still feel the mass, as also seen on interval growth scans. Her pregnancy was uneventful and she underwent elective lower segment caesarean section with bilateral tubal ligation with wide excision of scar endometriotic tissue at 38±5weeks (Figure 3). Histopathology revealed fibro-collagenous tissue with striated muscle deeper down. There were vague fascicles of spindle cells with fusiform nuclei and scanty cytoplasm, in a densely collagenous stroma. Few extravasated red cells were seen in areas of hemorrhage. No endometrial glands were seen (Figure 4). The patient is in our follow-up program and has not reported recurrence of symptoms or mass.

DISCUSSION
Cesarean section appears to be an important risk factor of scar endometriosis, as it exposes a large number of endometriotic cells and these cells get entrapped into the wound (Wang et al., 2003). Moreover, amniotic fluid and excessive blood loss exacerbate the separation of these active cells and provide a rich nourishing environment encouraging the growth of endometrial tissue in the wound (Wang et al., 2003). Obesity is an additional risk factor as it provides a wide surface area for entrapment of endometrial tissue (Uçar et al., 2015).
Figure 1. 1A: Ultrasound scan illustrating a well-defined hypoechoic lesion with smooth margins in the muscular plane of the suprapubic region measuring 6.23cm x 3.67cm x 2.28cm. 1B: Ultrasound scan showing an endometrial cyst measuring 1.71cm x 1.66cm x 1.64cm and a clear cyst measuring 2.13cm x 1.77cm in the left ovary.

Figure 2. 2A (axial T2WI), 2B (coronal T2WI), 2C (T1WI-post contrast), 2D (STIR axial): the yellow arrows in these images reveal a relatively well defined nodular lesion in the supra-pubic region within the bulky left rectus abdominis muscle, measuring 5.8cm x 3.3cm x 2.5cm. The lesion appears heterogeneously hyperintense on T2WI and STIR, showing a peripheral hypointense rim due to hemosiderin deposition (red arrow- 2A, 2D) with post contrast enhancement (2C).

Esquivel-Estrada et al. (2004) described the triad seen in cases of scar endometriosis, which comprises history of caesarean section or any other gynecological surgery, cyclical waxing, and waning pain accompanied by the patient’s menstrual cycle, with a tumor inside/near the scar site as the clinical diagnostic sign for scar endometriosis. Our patient fulfilled all three criteria. Magnetic resonance imaging (MRI) and ultrasonography (USG) are the most commonly used non-invasive imaging modalities for endometriosis (Nisenblat et al., 2016).
The presence of two out of three features: 1) presence of endometrial glands, 2) endometrial stroma (often contains fine capillary network, long-standing cases may show fibrosis or decidual change, myxoid change), and 3) evidence of chronic hemorrhage (hemosiderin-laden/foamy macrophages) (Pathan et al., 2005). The histopathology findings were not classical in our case. However, evidence of fibrosis was noted, which is a characteristic feature of long-standing stromal endometriosis (Vigano et al., 2018). Vigano et al. (2018) have suggested the inclusion of fibrosis as an important part of the definition of endometriosis, not only for diagnostic but also for treatment purposes. In fact, 40% of the cases of ovarian endometriosis are devoid of endometrial epithelium, and in most of these cases, only the inner cyst wall demonstrates only fibrotic changes (Muzii et al., 2007). Moreover, pelvic adhesions classically seen in endometriosis are devoid of any endometrial components (Somigliana et al., 2012). In our case, the only finding evidenced in the excised tissue was fibrosis. Typical endometrial glands were not seen in our patient possibly because of the pregnancy-associated regression of endometriosis (Leeners et al., 2018). The presence of endometrial tissue in such patients can be confirmed with immunohistochemistry staining for ER (estrogen receptor) and PR (progesterone receptor).

The various indicators pointing towards the diagnosis of scar endometriosis in the case reported herein include history of cyclical pain increasing during menstruation and presence of a mass (confirmed by ultrasound and MRI scan) at the previous cesarean section scar site. The resolution of symptoms and the decrease seen in the size of the mass after the introduction of dienogest along with the absence of symptoms during pregnancy point towards the diagnosis of scar endometriosis.

The first-line management of scar endometriosis is wide surgical excision of the mass (Vagholkar & Vagholkar, 2019). Medical management can be considered for symptom management, which includes treatment with oral contraceptive pills, progestins, dienogest, or gonadotropin releasing hormone agonists (Dunselman et al., 2014). Since the symptoms were mild and the patient was planning for future pregnancy, she was treated medically with dienogest and surgical excision was deferred until the next caesarean section to prevent an additional surgery. Several preventive measures have been proposed in the literature to minimize the iatrogenic transplantation of endometriotic tissue, such as thoroughly cleaning and irrigating the abdominal wound before closure (Uçar et al., 2015). Also, it is a common practice amongst obstetricians to clean the endometrial cavity with a moist/dry sponge after placental removal and before uterine closure. If the same mop is used, it can cause the inoculation of endometrial tissue onto the abdominal wound site (Wolf & Singh, 1989). Moreover, the suture material used to close the abdominal wall should not be reused to close the abdominal wound (Uçar et al., 2015). The instruments and needles used in uterine wall closure should be replaced with new ones when the abdominal wall is sutured (Teng et al., 2008). Some studies have advocated that the closure of visceral and peritoneal layers might also help in the prevention of scar endometriosis (Chang et al., 2009).

CONCLUSION
Scar endometriosis is an uncommon disease caused by iatrogenic inoculation of endometrial tissue onto the abdominal scar site. Caesarean section is one of the common causes of scar endometriosis and adequate measures should be taken to prevent this iatrogenic complication. It is important to include fibrosis in the definition of endometriosis. Wide surgical excision is the first-line treatment option for scar endometriosis. If scar endometriosis develops after caesarean section and the patient is having mild symptoms and has plans to get pregnant in the future, excision may be deferred until the next caesarean section.
Support
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CONFLICT OF INTEREST
The authors have no conflict of interest to declare.

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