Caesarean scar endometriosis: Is it time for us to pay more attention to this complication?

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ABSTRACT

Caesarean scar endometriosis is the result of inoculation of endometrial tissue in the scar during surgery. Due to the increase in the numbers of caesarean sections being performed world-wide, what was once a rare complication of endometriosis is now fast becoming more prevalent. Physicians' knowledge of its existence and preventive methods are, however, lagging behind: much focus has been placed on the diagnosis and treatment of endometriosis per se, but not of its complications. We present the case of a patient who faced a delay in diagnosis and management but was eventually treated with surgical excision of her caesarean scar endometriosis, with effective relief of her symptoms. Additional imaging and a multi-disciplinary input were beneficial in contributing to her management. This case highlights the urgency of increasing awareness and emphasizing the need of taking additional steps during surgery to prevent this complication.

KEYWORDS

Caesarean scar endometriosis, abdominal wall endometriosis, endometriosis, endometriotic nodule.

Introduction

Endometriosis is a pathological gynecological condition first described in 1860 by Karl Von Rokitansky. However, its official name was coined only 65 years later in 1925 by Sampson. Endometriosis is a chronic gynecological disorder in which the endometrial glands and stroma are present outside the uterine cavity [1]. Although the prevalence of endometriosis has been quoted to affect about 5-15% of women in their reproductive age [2], and up to 50% of sub-fertile women [3], the real incidence of this disease remains uncertain as menstruation disorders are often under-reported and definitive diagnosis requires surgery and histology. However, in recent years, increasing attention has been paid to endometriosis, generating more public interest and awareness. Despite this, much focus has been placed on the diagnosis and management of endometriosis, and less on the complications caused by this condition. Additionally, most physicians have not been aware of these complications and hence may not be cognizant of them when patients present in their clinics.

Abdominal wall endometriosis is a complication mostly occurring in old surgical gynecological or obstetrical operative scars [4]. Literature from the past has quoted the incidence of caesarean scar endometriosis (CSE) as ranging from 0.03-1.73% [5]. Globally, caesarean section rate is currently estimated to be around 21.1% of live births with numbers going up to 42.8% in Latin America and the Caribbean region [6]. This is a marked increase from the estimated global rate of 7% observed in 1990. With the number of caesarean sections increasing steadily over the years, one would likewise expect the number of CSE to increase accordingly and that the incidence is currently much more than previously quoted. This higher incidence justifies the urgent need for more attention to be paid to the swift management and prevention of this complication. We report a case of a woman with CSE who presented to our center after multiple visits to various other specialists and being inadequately treated.

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Case presentation

A gravida 1 parity 1 woman in her 20s comes to our gynecological clinic after having seen multiple specialists for chronic abdominal pain. She had an oesophago-gastro-duodenoscopy and colonoscopy performed that yielded no abnormal findings. A recent pelvic ultrasound also reported a normal uterus and ovaries. Upon anamnesis it was found that she had previously undergone 6 years back an emergency preterm caesarean section at 34 weeks due to antepartum hemorrhage. Subsequently, she reports noticing a discrete lump on the right side of her caesarean section scar; occurring at about 3 years after the surgery. This became more prominent over the last 3 years. In the preceding years she had been presenting cyclical abdominal pain which was more pronounced during her menstruation. However, in recent years, this pain became constant. She declared having regular menstruations and otherwise no dyspareunia, dysuria or dyschezia. A gynecologist, whom she was previously consulting before the current visit, had prescribed her a year of progestogen treatment for endometriosis, but this did not ease her pain and progressively stronger analgesics had also

proven ineffective, culminating to a point in which the pain was negatively impacting her quality of life.

Investigations

Clinical examination revealed a tender, 3 cm wide subcutaneous mass at the right corner of her Pfannenstiel scar. The mass was tender on palpation and immobile with no overlying skin changes. An ultrasound of the abdominal wall reported a heterogenous hypoechoic mass of about 3.8 x 3.1 x 2.2 cm with irregular margins and internal vascularity in the subcutaneous tissue, indenting the rectus abdominis muscle (Figure 1). A magnetic resonance imaging (MRI) of the pelvis was subsequently performed to evaluate the depth of the invasion for better surgical planning. This revealed a 3.3 cm enhancing soft tissue mass along the right caesarean scar, which likely represents subcutaneous endometriosis with a differential diagnosis being a desmoid tumor. The mass was shown to be predominantly immerse in the deep subcutaneous layer with broad contact inseparable from the adjacent rectus, indented up to 5 mm in depth (Figure 2).

Treatment and follow up

As she was symptomatic, a tissue biopsy for histology was omitted and surgery was planned. An incision of about 5 cm was made directly on the caesarean section scar above the mass. The mass appeared to be surrounded by fibrotic tissue with an extension to the underlying rectus sheath. The mass was enucleated and *en-bloc* excision was performed, ensuring clear margins with non-fibrotic tissue remaining. This left an approximately 3 cm defect in the rectus sheath. As the defect was small, it was deemed appropriate for primary closure without the use of a mesh. The wound was given copious lavage with warm normal saline and hemostasis achieved before the release and careful closure of the rectus sheath with continuous sutures. The fascia was once again washed with normal saline before a second clean needle was used to close the fascia.

After excision, the mass was cut into half and endometriotic contents was seen to extrude from the middle (Figure 3). Histology confirmed the presence of endometriosis involving soft tissue, associated with chronic active inflammation yet no malignancy detected. Post-operative recovery was uneventful and a six-month clinic review revealed a satisfied patient with no recurrence over 18 months of follow-up.

Discussion

Abdominal wall endometriosis is predominantly seen in patients with previous surgeries. They may develop after both obstetrical (caesarean sections, hysterotomy) or gynecological (hysterectomy, cystectomies) surgeries and in some rare cases, in episiotomy wound sites after a normal vaginal delivery [7]. Due to the complex nature of endometriosis itself, the pathogenesis of CSE is ambiguous and surmised to be based on the implantation theory [8], in which the endometrial tissue is inoculated iatrogenically in the caesarean section incision itself during the surgery. With adequate blood supply providing a source of nutrients and hormones, the implanted endometrial tissue, when stimulated by estrogen, will become activated and proliferate, resulting in CSE [9]. Although rare, Leng *et al*. [10]

Figure 1 Ultrasound image showing heterogenous hypoechoic mass at the right of caesarean section scar.

Figure 2 MRI image of transverse view of the right soft tissue mass (yellow arrow) along the caesarean section scar.

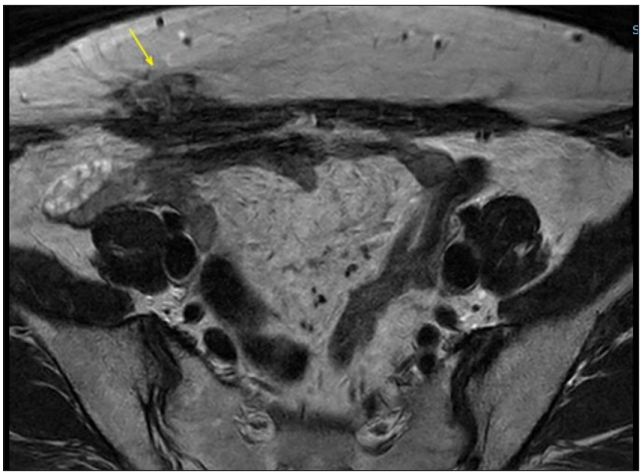
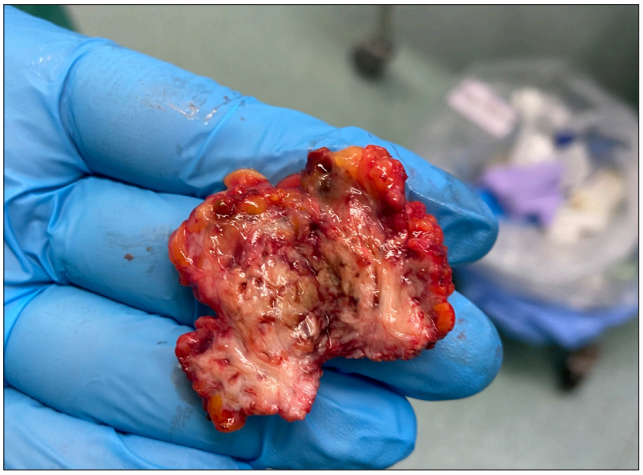


Figure 3 Excised mass cut into half with endometriotic contents.



and Alberto *et al*. [11] have also reported case studies of malignant transformation of CSE into carcinosarcoma and clear cell carcinoma respectively, proving that histological diagnosis of CSE is crucial for those with a rapidly enlarging mass but who refuse surgical removal. With the presence of CSE, patients may experience symptoms of cyclical pain and swelling. In other cases, leakage of the endometriotic content from the subcutaneous mass has been reported during menstruation. Despite this, a majority of women present with a clinically palpable mass upon examination [12].

Ultrasound remains the most cost-effective and efficient way to diagnose CSE. Imaging, together with a detailed clinical history and examination, will allow us to rule out other differential diagnoses such as a hernia, lipoma or cyst. However, in some cases with an inconclusive ultrasound, or for those with suspected deep infiltrating lesions, further evaluation with MRI may be helpful for both, diagnosis and pre-surgical planning.

Although hormonal medication may only offer a temporary cessation of symptoms with no or slight reduction of lesion size [13], the side effects from these medications can reduce patients' compliance. The definitive treatment for CSE would be *en-bloc* surgical excision of the lesion. Some authors [14] have advised a margin of at least 1 cm around the lesion for adequate clearance and to prevent recurrence. Depending on the size, location and number of CSE lesions, the inclusion of a multi-disciplinary team would be prudent and required. This comprises involving a general surgeon in the event of large fascia involvement which would require mesh placement.

Based on the implantation theory, some additional steps during the caesarean section can be proposed to reduce the risk of subsequent CSE. For instance, while cleaning the uterine cavity with gauze after delivery of the placenta, it would be judicious not to reuse the gauze during the closure of the uterine cavity and the abdominal wall. New suture needles should be used for each layer to prevent further inoculation of endometrial cells. Suture placement should also be precise and not involve unnecessary deeper tissues.

Mingaglia *et al*. [15] have also hypothesized that not closing the parietal and visceral peritoneum can significantly increase the risk of CSE. This should however be balanced with the increased operative time required to close the additional layers. Copious lavage of the pelvic cavity before closure of the abdominal wall has also been suggested [16]. In addition, it is important to ensure that suction of lavage is adequate to minimize the risk of leaving endometrial cells behind. Other authors [17,18] have recommended washing of the subcutaneous layer with normal saline prior to closure of the abdominal wall. Zhang *et al*. [12], in one study reported that 83.3% of CSE lesions were mainly found in the corner sites of the wound. With this knowledge in mind, particular attention should be paid at washing the corners of the wound and ensuring that the subcutaneous layer is dry prior to closure.

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