

Cases of scar endometriosis after caesarean section

Namkha Dorji¹, Sangay Tshering², Sonam Choden³

^{1,2}Department of Obstetrics and Gynaecology, Jigme Dorji Wangchuck National Referral Hospital, Thimphu, Bhutan ³Department of Pathology and laboratory Medicine, Jigme Dorji Wangchuck National Referral Hospital, Thimphu, Bhutan

ABSTRACT

Introduction: The presence of functional endometrial tissue in the surgical scar, which commonly follows caesarean section is called scar endometriosis. It is a very rare condition which is commonly misdiagnosed. **Case report:** We report two cases of scar endometriosis which developed after caesarean section. One case was misdiagnosed as a keloid for a long duration (12 years) and other one was not diagnosed despite the characteristic presenting symptoms. Both the cases underwent surgical excision with free resection margins and histopathological confirmation of diagnosis. **Conclusions**: Clinicians must be aware of varying presentations of scar endometriosis to ensure timely diagnosis and treatment.

Keywords: Caesarean section; scar endometriosis; surgical excision; extra pelvic endometriosis.

INTRODUCTION

Endometriosis is the presence of functional endometrial tissues outside the uterus^{1,2}. It may be pelvic or extra pelvic; the common pelvic sites are ovaries, pouch of douglas, uterine ligaments, pelvic peritoneum and rectovaginal septum. The major extra pelvic sites include the lungs, pleura, kidneys, bladder, omentum, bowels, lymph nodes and abdominal wall³. Abdominal wall endometriosis is a common extra pelvic endometriosis occurring after caesarean section which is believed to be caused by direct implantation of endometrial tissues during caesarean section^{1,4,5}. The great variability in its symptoms and signs, and limited knowledge amongst clinicians often leads to unnecessary procedures, delayed diagnosis or misdiagnosis causing immense emotional and physical distress to the patients^{3,6}.

Here, we report two cases of scar endometriosis following caesarean section, its symptoms and signs, and preventive measures during caesarean section. This is the first such report from Jigme Dorji Wangchuck National Referral Hospital with the aim of increasing awareness amongst clinicians about this disease entity in order to take preventive measures and avoid diagnostic delays.

CASE PRESENTATIONS

Case 1:

A 34 year old woman with the history of a caesarean section in 2008 developed cyclical bleeding from the surgical site during menstrual bleeding three months after the caesarean section and lasted for two years. The bleeding site closed spontaneously

Corresponding author:

Namkha Dorji namji2002@gmail.com but she started feeling a painful lump when the severity of pain increased during menstruation. She didn't have dysmenorrhoea or dyspareunia. She was treated symptomatically for the last 12 years without a definite diagnosis. On consultation with a gynaecologist for secondary infertility, a tender hard lump of 6x5 cm was noted over the caesarean scar. With a clinical diagnosis of scar endometriosis, she underwent surgical excision under spinal anaesthesia.

Grossly, the cut section revealed a tan-white rubbery lesion (2.5x2.5x2 cm) in the subcutaneous tissue, located very close to overlying linear scar on the skin (Figure 1a). Microscopic examination confirmed the diagnosis of scar endometriosis, which revealed scattered foci of endometrial glands in proliferative phase, surrounded by endometrial type stroma with loose fibromyxoid change (Figures 1b-d). Some of the glands in dermis showed cystic dilatation. No malignant transformation was noted. The resection margin was free.

On follow up after six months, she continued to remain symptom free and there was no mass or tenderness on the scar.



Figure 1a. Tan-white rubbery lesion (black circle) in the subcutaneous tissue (yellow arrows), located very close to the overlying linear scar on skin (red arrows)



Figure 1b. Stroma poor endometriotic glands (yellow arrows) with cystic dilatation in the superficial dermis (H&E, 40x)



Figure 1c. Endometrotic glands (black circle) with fibromyxoid stroma (red arrows) in subcutaneous tissue (H&E, 40x).



Figure 1d. Endometrotic glands (black arrow) and stroma (red arrow) in proliferative phase (40x), mitotic figure (inset, 400x). ndometrotic glands (black circle) with fibromyxoid stroma (red arrows) in subcutaneous tissue (H&E, 40x)

Case 2:

A 37 year old, mother of three children, all of whom were delivered by caesarean sections in 2005, 2006 and 2016, developed a painful lump in the caesarean scar six months after her third caesarean section. The severity of pain increased during menstruation and the size of lump gradually increased. She didn't have dysmenorrhoea, dyspareunia or difficulty in conceiving. The lump was treated as keloid for the last one year with intralesional injection of Triamcinolone (40mg/ml) in the Department of Dermatology at Jigme Dorji Wangchuck National Referral Hospital.

As there was no improvement, she was referred to the Obstetrics and Gynaecology outpatient department at Jigme Dorji Wangchuck National Referral Hospital for further assessment. On examination, a tender hard lump (7x5 cm) was noted over the scar site. Based on typical clinical history and examination finding, a provisional diagnosis of scar endometriosis was made. She underwent surgical excision with 2 mm free margin under spinal anesthesia. The endometriosis had extended up to rectus sheath (Figure 2a). Microscopic evaluation confirmed the diagnosis of scar endometriosis. The endometriotic glands were in proliferative phase (Figures 2b-c). Evidence of recent and old hemorrhage was evident within the lesion.

There was no recurrence six months after the excision.



Figure 2a. Grey-white fibrotic lesion (blue arrow)



Figure 2b. Endometrial glands (black arrow) and stroma (red arrow) (H&E, 40x)



Figure 2c. Cystic dilatation of glands (black arrows) filled with red blood cells and amorphous debris (yellow arrow) (H&E, 40x) DISCUSSION

Scar endometriosis is a very rare condition with an incidence of 0.03-0.4%⁶. Although there are several postulated theories on the etiology of scar endometriosis, direct iatrogenic implantation of endometrial tissue to the wound during pelvic or abdominal surgery is known to be the most widely accepted theory⁵.

A case series involving 12 women with cesarean scar endometriosis in Turkey reported that these patients were women aged 24-44 years with a median age of 34.6+/-5.43 years following caesarean section. The presenting symptoms included a palpable subcutaneous mass under the incision scar, cyclical pain, non-cyclic pain, enlargement of nodule during menstrual period, menstrual-related dark brown leakage, cyclic hypogastric pain and right iliac fossa pain without radiation⁸. The mean latency period for scar endometriosis after caesarean section has been reported to be 31.6+/-23.9 months and duration between initial symptoms and surgery was 28.3+/-25.0 months⁴.

In our report, one case presented with cyclical menstrualrelated dark brown leakage from previous caesarean scar for 12 years and the second case presented with a cyclical painful mass at the caesarean scar more than 3 years. The diagnosis should have been made on time with the given awareness of the typical cyclical symptoms. Clinicians should be aware of this rare entity as the definite treatment depends on the correct diagnosis.

Imaging studies like ultrasonography, MRI and CT scan can be useful to study the depth and extent of lesions to plan surgery^{7,9}. Ultrasound is usually the first imaging technique used to study the identified mass on the abdominal scar. However, ultrasound imaging shows varying nonspecific features like unilocular or multi cystic lesions, solid, or mixed solid and cystic lesions. Ultrasound maybe useful in demonstrating solid versus cystic nature of lesions and its relationship with skin and fascia¹⁰. In the two cases that we have reported, imaging studies were not performed as the symptoms and signs were indicative of diagnosis. Needle aspiration cytology can also be performed preoperatively if the lesion is suspicious of malignancy⁹. FNAC was not done in our cases as the clinicians were not aware of its role during the clinical situation then.

Most literature reports the treatment of scar endometriosis with surgical excision and final confirmation of diagnosis by histopathology examination^{6,8,11}. Medical management with oral contraceptives, progestational and androgenic drugs have also been tried and reported. However, hormonal suppression is believed to be effective only partially and surgical excision of the scar is considered as the definitive treatment¹². In the 2 cases that we've reported, surgical excision with free margin was performed. Both cases continued to remain recurrence free on follow up.

Since scar endometriosis is known to develop following iatrogenic implantation of endometrial tissue during caesarean section, we believe that standardization of the techniques of caesarean section would reduce its incidence. Some of the standard good surgical techniques to be employed includes suturing of uterine incision without involving endometrium, using different sponges for cleaning the endometrium and the abdominal wound during caesarean section, using separate needles to close the uterine and abdominal wound, and thorough irrigation and cleaning of abdominal wound prior to closure^{7,9,13}.

CONCLUSIONS

Clinicians must be aware about the varying clinical presentations of scar endometriosis and appropriate referrals to gynaecologists must be done for definitive treatment. Caesarean section should be performed using standard surgical techniques to reduce occurrence of scar endometriosis. Radiological imaging modalities are useful to study the size and extension of lesions. Preoperative FNAC should be performed in cases of diagnostic uncertainty or when a suspicion of malignancy exists.

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