

Cutaneous endometriosis

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ABSTRACT

Introduction: Cutaneous or subcutaneous endometriosis is a rare entity that is an often overlooked because of chronic abdominal pain.

Methods: We reviewed the ten cases of cutaneous endometriosis that presented to this hospital over a seven-year period.

Results: The mean age of patients at presentation was 36.3 years. All our patients presented with cyclical abdominal pain. There was a considerable delay in the diagnosis and offer of treatment, the mean length of time between onset of symptoms to surgery being 31.7 months (range 1–62 months). The onset was spontaneous in 40 percent of our patients and the rest had scar endometriosis. The patients with spontaneous onset of disease had a more severe pelvic disease compared to those with scar endometriosis. Complete surgical excision was curative but left a large defect requiring polypropylene mesh repair in two patients.

Conclusion: Cutaneous endometriosis should be suspected in any female presenting with cyclic or non-cyclic pain emanating from a mass in the vicinity of a previous surgical scar, the umbilicus or in the inguinal region. Surgical excision of the cutaneous endometriotic implants can be easily performed and is curative.

Keywords: cutaneous endometriosis, endometriosis, inguinal scar, scar endometriosis, umbilical scar

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INTRODUCTION

Endometriosis is defined as the presence of endometrial glands and stroma outside the endometrial cavity. It is a common gynaecological condition that affects up to 22% of all women,⁽¹⁾ about 20%–30% of patients presenting with subfertility,^(2,4) and up to 45% of women with pelvic pain.⁽³⁾ In spite of being a relatively common condition, endometriosis remains a diagnostic and therapeutic enigma even today, largely due to its variable presentations. The pelvis is the most common site of the disease, giving rise to the common presenting symptoms of pelvic pain,

dysmenorrhoea, dyspareunia, cyclical bowel or bladder symptoms, and infertility. Extragenital or extrapelvic endometriosis is less common but even more difficult to diagnose due to the extreme variability in presentation. Although the first case of cutaneous endometriosis was reported as early as 1885, not much has been published on this subject, and even today, ignorance on the part of doctors result in the diagnosis being often delayed or missed. Our case series of ten cases of cutaneous endometriosis that presented to the National University Hospital over a seven-year period (2000–2007) is examined together with a literature review on the subject.

METHODS

Cases of cutaneous endometriosis were selected from the computer records and clinical audits of the Department of Obstetrics & Gynaecology, National University Hospital, from January 2000 to July 2007. Only those cases with a confirmed histopathological diagnosis of endometriosis were included. During this period, there were 908 surgically-proven cases of endometriosis, giving an incidence of 1.1%. The medical records of these patients were reviewed and entered into a database and analysed using the Statistical Package for Social Sciences version 13.0 (SPSS Inc, Chicago, IL, USA).

RESULTS

The patient characteristics and symptomatology are summarised in Table I, while their clinical features are listed in Table II. The mean age of the patients at presentation was 36.3 years (range 27–45 years). All patients with scar endometriosis presented to the gynaecologist. Three of the four patients with spontaneous endometriosis initially presented to general surgeons. The mean duration of symptoms before presentation to a doctor was 23.5 months, and the mean length of time between onset of symptoms to surgery was 31.7 months (range 1–62 months). Cyclical pain during menstruation localised to a palpable mass in the abdominal wall was the main clinical feature in these patients. Two patients developed a palpable mass only during menstruation. Six of the ten patients had symptoms of pelvic pain or dysmenorrhoea besides the pain in the abdominal wall. Two patients had infertility and one complained of cyclical bleeding from the umbilicus.

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Table I. Patient characteristics and symptomatology.

Patient no.	Age (years)	Parity	Race	Site	Symptoms	Previous surgery	Duration before presentation (months)	Clinical finding	Imaging studies
1	43	2+0	Chinese	Umbilical	Painful umbilical nodule, cyclical bleeding	-	1	1 cm × 1 cm umbilical nodule	-
2	30	1+0	Chinese	Pfannenstiel	Painful nodule scar	LSCS at scar site	12	5 cm × 5 cm nodule in previous scar	-
3	36	0+1 Spontaneous complete miscarriage	Malay	Right inguinal	Cyclical right inguinal	Appendicectomy 15 years prior pain to presentation	36	1 cm × 1 cm lump in right inguinal region	CT scan
4	36	3+0	Thai	Midline infraumbilical	Dysmenorrhoea, cyclically painful nodule	LSCS	24	1.5 cm umbilical nodule	CT scan
5	28	1+0	Chinese	Pfannenstiel scar	Premenstrual scar pain	LSCS	12	2 cm × 2 cm nodule in previous scar	-
6	36	0+0	Chinese	Right inguinal	Subfertility & right inguinal pain	Laparoscopic excision of pelvic endometriosis in 2001	18	3 cm × 4 cm lump in right inguinal region	-
7	41	3+0	Chinese	Umbilical	Umbilical bleeding & tenesmus	-	24	1 cm × 1 cm umbilical nodule	-
8	27	2+0	Indian	Pfannenstiel scar	Scar pain during menses	LSCS	24	2 cm × 2 cm nodule in previous scar	-
9	45	2+0	Chinese	Pfannenstiel scar	Scar pain related to menses	Subtotal hysterectomy for fibroids	24	1 cm × 1 cm nodule in previous scar	-
10	41	2+0	Chinese	Pfannenstiel scar	Scar pain with menses	LSCS	60	6 cm × 5 cm nodule in previous scar	-

Six of the ten patients (60%) patients had a previous surgery, which could explain their scar endometriosis. For five patients, this was a caesarean section, while the remaining patient had a subtotal hysterectomy. The onset was spontaneous in the other four patients. The first patient was nulliparous and had spontaneous inguinal endometriosis, the second had had a previous first trimester miscarriage and presented with endometriosis on the pubic tubercle. The third patient presented with umbilical endometriosis following three previous vaginal deliveries. The fourth patient had a prior caesarean section done but had symptoms of umbilical endometriosis occurring ten years later; hence it was likely to be of spontaneous onset rather than due to implantation during the caesarean section. Three patients were offered medical treatment prior to surgery. Two had preoperative gonadotrophin-releasing hormone agonists (GnRH agonists) for three months to reduce the size of the endometriotic implant. Another patient was prescribed combined oral contraceptive pills, which failed to provide symptomatic relief.

Six of the ten patients (60%) presented with

endometriosis in proximity to the previous Pfannenstiel scar. All of them presented with a history of cyclical pain during the menstrual cycle and had a palpable mass on examination. Based on the classical symptom of pain and increase in the size of the lump during the menstrual cycle, all of them were clinically diagnosed as having cutaneous endometriosis. In five patients, this mass was in close vicinity to the previous scar; however in one case, the scar was found to be in the midline infraumbilical region, about 5 cm above the previous scar. Four of the six patients did not have any pelvic symptoms and did not undergo laparoscopy. The other two patients with dysmenorrhoea underwent concomitant laparoscopy, which revealed minimal to mild endometriosis. In all cases, excision was curative until the point of follow-up.

Two patients (20%) had endometriosis in the inguinal region. The first patient presented in 2001, with primary infertility, dysmenorrhoea and a lump in the right inguinal region which was painful during menstruation. Fine-needle aspiration cytology was done to aid diagnosis and it confirmed endometriosis. This patient underwent diagnostic laparoscopy twice with

Table II. Clinical features of the study patients.

Patient no.	Type of cutaneous endometriosis	Site & size	Depth of penetration	Repair of defect	Pelvic endometriosis	Histology of excised nodule	Follow-up	Recurrence
1	Spontaneous	Umbilical nodule 1.0 cm in diameter	Peritoneum	Primary	Asymptomatic hence not assessed	Endometriosis	3 years	Nil
2	Scar	5 cm × 5 cm nodule in previous scar	Rectus sheath	Primary	Asymptomatic hence not assessed	Endometriosis	2.3 years	Partial
3	Spontaneous	Right pubic tubercle lump 1 cm × 1 cm	Rectus sheath	Primary	Severe	Endometriosis	2 years	Partial
4	Scar	Midline infra-umbilical nodule 1.5 cm in diameter	Rectus sheath	Primary	Minimal–mild	Endometriosis	8 months	Nil
5	Scar	2 cm × 2 cm nodule in previous scar	Rectus sheath	Primary	Mild	Endometriosis	5 months	Nil
6	Spontaneous	Right inguinal lump 3 cm × 4 cm	Peritoneum	Mesh	Severe	Endometriosis	8 months	Nil, but painful nodule in the left inguinal region
7	Spontaneous	Umbilical nodule 1.0 cm in diameter	Rectus sheath	Primary	Moderate	Endometriosis	5 months	Nil
8	Scar	2 cm × 2 cm nodule in previous scar	Rectus muscle	Primary	Asymptomatic hence not assessed	Endometriosis	4 months	Nil
9	Scar	1 cm × 1 cm nodule in previous scar	Rectus muscle	Primary	Asymptomatic hence not assessed	Endometriosis	1 month	Nil
10	Scar	6 cm × 5 cm nodule in previous scar	Peritoneum	Mesh	Moderate	Endometriosis	1 month	Nil

ablation of endometriosis, but was not offered excision of the cutaneous endometriosis until five years after the initial presentation. The third laparoscopy revealed endometriotic deposits along the right round ligament, contiguous with the right-sided inguinal lump. Ablation of the round ligament endometriosis was done together with complete excision of the inguinal lump, which left a defect in the fascia which was then repaired using a Prolene mesh. There was evidence of severe pelvic endometriosis, which was treated appropriately, as well as endometriotic deposits along the left round ligament, which were ablated. Ten months postoperatively, the patient complained of pain in the left inguinal region while remaining asymptomatic on the right.

The second patient had a history of an open appendectomy and a spontaneous first trimester miscarriage, and presented to the general surgeon with a three-year history of a palpable mass over the pubic tubercle, which was again characteristically painful during the menstruation. The presumptive diagnosis was adhesion colic. Diagnostic laparoscopy performed by the surgeon showed small intestinal adhesions at the site of the

appendectomy scar, but these were away from the site of symptoms and hence adhesiolysis was not performed. She was then referred to the gynaecologist, and based on the characteristic symptoms, was diagnosed as having cutaneous endometriosis. Laparoscopy revealed dense adhesions and deeply infiltrating endometriosis in the cul-de-sac, which was treated with ablation and excision. The nodule on the pubic tubercle was found to be overlying the rectus sheath and was excised. The patient was asymptomatic for two years after the surgery; however, mild inguinal pain at the site of the previous surgery returned subsequently indicating possible incomplete excision or recurrence.

Two patients (20%) presented with a history of cyclical bleeding from the umbilicus. The first, a 43-year-old woman (para two) with two previous caesarean sections presented with an umbilical lump that was noticed a month prior to presentation. There were no other symptoms suggestive of pelvic endometriosis, hence laparoscopy was not performed. During the excision, the nodule was found to be extending down to the peritoneum, and was excised completely with primary closure of the peritoneum and

rectus sheath. She continued to be asymptomatic six years following surgery. The other patient had three previous vaginal births and no previous surgery. She presented with a history of dysmenorrhoea and cyclical bleeding from her umbilicus. The 0.5 cm umbilical mass was excised completely two years following the first presentation. Concomitant laparoscopic evaluation revealed the presence of deeply infiltrating endometriosis in the pelvis, which was then treated with excision and ablation.

Interestingly, in our study, all patients with spontaneous endometriosis had concomitant symptoms of pelvic endometriosis, and laparoscopy revealed moderate to severe endometriosis in all of them. On the other hand, only two of those with scar endometriosis (33.3%) had symptoms of pelvic endometriosis and they had only minimal to mild pelvic disease. In three patients, the endometriotic implant extended down to the peritoneum. One of these patients had spontaneous umbilical endometriosis, and complete excision was carried out with primary closure of the resulting fascial defect. In one patient with spontaneous inguinal endometriosis, there was a 4-cm diameter defect in the rectus sheath following excision, and required placement of a Prolene mesh to prevent subsequent herniation. Another patient with scar endometriosis had a 6-cm defect following excision and similarly required an insertion of a polypropylene mesh for repair.

The histological appearance of all the specimens was consistent with endometriosis with both glandular and stromal elements. In all the patients, the endometrial glands and stroma were within in a background of fibroadipose tissue. However, in two patients with spontaneous cutaneous endometriosis (patient nos. three and six), skeletal muscle fibres were noted to be present.

DISCUSSION

This study reviewed the ten patients diagnosed with cutaneous endometriosis at National University Hospital during a seven-year period (2000–2007). During this period, there were 908 surgically-proven cases of endometriosis, giving an incidence of 1.1%. This is the first case series reported from the region, with no comparative figures from the region to date. However, this incidence seems to be much lower than other figures quoted from Glasgow,⁽⁴⁾ where 5.2% of endometriosis has been reported as being cutaneous. It is possible that a comparatively low incidence of this condition in the region is due to a lower rate of recognition. The mean age for diagnosis in our study was 36.3 years, which is slightly older than that reported by Singh et al (34 years) and by Douglas and Rotimi in the Glasgow study (33.7 years).^(4,6) The average duration between onset of symptoms and

presentation was quite long (23.5 months), and this together with the delay in recognition could account for this difference.

Among all the cutaneous sites of endometriosis, scar endometriosis has been shown to be the commonest.^(5,6) Our findings were similar, with 60% of all cutaneous endometriosis being in an abdominal scar; and in keeping with other reports, the most common antecedent surgery was caesarean section. Cyclical pain with a palpable mass is the most commonly presenting symptom of this condition. In our study, all patients had these classical cyclical symptoms. In spite of this, the diagnosis was delayed by an average of 11 months (range 1–72 months), with significantly shorter delays as our experience grew. It is essential to point out that cyclicity is not always demonstrable and is not essential for diagnosis. Other authors have described non-cyclical pain as being more common,^(5,7) and hence the diagnosis of endometriosis must not be disregarded if the pain is not cyclical.

In spite of the classical presentation, misdiagnosis is not uncommon. The most common differential diagnoses include stitch granuloma, hernia and cellulitis. Umbilical endometriosis in particular can pose a diagnostic dilemma as it can simulate a malignant melanoma or the “sister Mary Joseph nodule”—a manifestation of intra-abdominal malignancy. While there are no pathognomonic radiological findings, owing to a change in appearance according to the phase of the menstrual cycle, and the degree of surrounding inflammatory and fibrotic response, some recent reports have shown that magnetic resonance imaging⁽⁸⁾ or epiluminescence microscopy⁽⁹⁾ may be useful in differentiating between umbilical endometriosis and other pigmented skin lesions. Although rarely required, fine-needle aspiration cytology⁽¹⁰⁾ may help resolve the diagnostic dilemma.

Most of our patients reported having visited several doctors, both general practitioners as well as specialists, before a diagnosis of cutaneous endometriosis was made. Even in a tertiary hospital setting like ours, there was a delay in the diagnosis and management of the first few cases owing to clinical inexperience. In the first few cases, imaging modalities, such as computed tomography, were used to aid diagnosis. However, with increasing clinical experience, it was evident that imaging was unnecessary in most cases, and the more recent cases were diagnosed based on clinical features and were offered prompt surgical treatment. In all our patients, histology of the excised tissue confirmed the diagnosis, and based on these findings, we do not deem any imaging studies to be required once there is a clinically palpable nodule, unless there are symptoms suggestive of a malignant transformation.

Excision is the mainstay of treatment of this

condition, and local wide excision to ensure complete removal of the disease is curative. Preoperative treatment with GnRH agonists has been advocated,⁽¹¹⁾ and was used in two (patient nos. two and eight) of our ten patients. Although it did provide relief from symptoms, it led to incomplete excision and partial recurrence of symptoms in one of the patients, and hence, we do not recommend its routine use. A second patient that was seen early in our series also had recurrence of symptoms 1.5 years after surgery, probably due to incomplete excision at the time of surgery. Two patients, one with scar endometriosis and the other with spontaneous inguinal endometriosis, had a large defect in the rectus sheath requiring a Prolene mesh insertion to prevent future herniation. As the exact depth of the cutaneous endometriosis can only be determined at the time of surgery, and complete excision is the only way to ensure a cure, it is essential to counsel patients preoperatively regarding the possible placement of a mesh to repair the defect in the rectus sheath. So far, there have been very few reports of the use of mesh following excision of cutaneous endometriosis⁽¹²⁾ and no reports of recurrence of endometriosis in these patients. Longer follow-up on these patients is required to demonstrate this. While spontaneous endometriosis is not preventable, scar endometriosis is likely to be preventable. Hence, routine irrigation of the abdominal wall wound before wound closure following any uterine surgery is recommended, to prevent implantation of endometriotic cells.

In this series, we found that spontaneous cutaneous endometriosis was associated with more severe pelvic disease than scar endometriosis. Possibly, patients with severe disease have several manifestations of the disease and cutaneous endometriosis could be one of the many extragenital manifestations. Thus far, this association has not been reported by any other authors. There is little doubt now that scar endometriosis results from iatrogenic implantation of endometrium (decidua). Scar endometriosis has been reported in abdominal scars following uterine operations, like caesarean section, myomectomy, hysterotomy and metroplasty, at the trocar site following laparoscopic treatment of endometriosis,⁽¹³⁾ in perineal scars of episiotomies,⁽¹⁴⁾ colporrhaphies, and Bartholin's gland excision,⁽¹⁵⁾ as well as along the needle tracks of amniocentesis or intrauterine injections for abortions. In contrast to scar endometriosis, the pathogenesis of spontaneous cutaneous endometriosis is yet unknown. Several aetiological theories have been proposed. These include coelomic metaplasia, congenital presence of developmentally-displaced endometrial tissue, direct extension through the round ligament or the patent omphalomesenteric duct, or mechanical seeding of

endometrial tissues via the lymphatic or venous system transfer via lymphatics or blood vessels.

Interestingly, endometriosis of the right inguinal region is more commonly reported than the left,⁽¹⁶⁾ and this was true for both our patients as well. Various theories have been proposed to explain this phenomenon. One of these proposes the presence of a clockwise⁽¹⁷⁾ intraperitoneal fluid circulation secondary to intestinal peristalsis and hydrostatic pressure changes from diaphragmatic movement, as was first described by Foster et al.⁽¹⁸⁾ As the endometrial cells remain in the right iliac fossa for a longer duration due to gravity, there is a greater chance of these cells being transported along the right round ligament through the inguinal canal to the inguinal region. Because of the transfer of endometrial cells along the round ligament, it has been suggested that the complete excision of inguinal endometriosis should also include the extraperitoneal portion of the round ligament to prevent recurrences.⁽¹⁹⁾

Histology is the mainstay of diagnosis of cutaneous endometriosis. Usually, a standard haematoxylin and eosin stain is sufficient for diagnosis, however, occasionally extragenital endometriosis may be atypical resulting in diagnostic difficulties.⁽²⁰⁾ Also, there may be surrounding, often pronounced, fibrosis, confusing the diagnosis. In case of a diagnostic dilemma, immunohistochemical analysis to detect oestrogen and progesterone receptors may be necessary to confirm the diagnosis.⁽²¹⁾ In all the patients in this series, the endometrial glands and stroma were within in a background of fibroadipose tissue. However the presence of skeletal muscle fibres in two patients with spontaneous cutaneous endometriosis, could indicate a deeper depth of the endometriotic deposits in these cases. There have been a few case reports of malignant transformation in cutaneous endometriosis especially in patients with the long-standing, recurrent endometriosis. Clear-cell carcinoma is the most common histological subtype, followed by endometrioid carcinoma.⁽²²⁾ Other subtypes, such as serous adenocarcinoma and adenosarcoma, have also been reported in both young and older postmenopausal women, especially following unopposed oestrogen therapy. Hence, the possibility of malignant transformation should be considered in rapidly-growing or recurrent cutaneous abdominal masses.⁽²³⁻²⁵⁾

In conclusion, cutaneous endometriosis is an increasingly diagnosed and reported condition with various sites of presentation, the most common being the abdominal wall. Due to a variable presentation, a high index of suspicion is essential and the condition should be included in the differential diagnosis of any patient who presents with pain and/or a palpable mass in the

abdominoperineal region, especially in those who have had previous gynaecological surgery. While symptoms are classically cyclical, the diagnosis of cutaneous endometriosis must not be disregarded if cyclicity is not demonstrable. Education of all doctors, including the primary care physicians, is important to help early diagnosis and treatment of this agonising condition. Surgery remains the mainstay of treatment. Complete excision prevents recurrence and should be the goal of treatment, even if it results in large fascial defects requiring primary closure using a mesh.

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