

Scar endometriosis with uterocutaneous fistula- A rare entity - Case report with review of literature

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Abstract

Background: Uterine fistulae commonly occur between the uterus and bladder or bowel (utero-vesical or utero-colonic), as a sequelae of inflammatory / infective conditions, or following surgeries. Uterocutaneous fistula is an infrequently seen condition, which usually develops as a complication following caesarean section or pelvic surgeries. Rare causes of Uterocutaneous fistulae include endometriosis, infection and intra uterine contraceptive devices. **Case presentation:** We report here a case of uterocutaneous fistula with scar endometriosis, which was diagnosed preoperatively on MRI. A 34 year old Para 2, Living 2, previous 2 LSCS patient with no comorbidities, presented with complaints of pain and swelling at the LSCS scar site for the past two years, associated with serous and bloody discharge from the scar on and off for the past two months. On examination, her vitals were stable. Pfannenstiel incision was noted with a small swelling at the scar site. There was associated serous and bloody discharge from the swelling. Ultrasound evaluation revealed normal endometrial thickness with the endometrial cavity seen communicating with a complex hypoechoic collection anteriorly. Subsequent MRI not only diagnosed the presence of scar endometriosis, but also revealed the presence of a hitherto unsuspected uterocutaneous fistula extending through the endometriotic tissue. These findings were confirmed at surgery. The patient underwent hysterectomy, bilateral salpingectomy and excision of the uterocutaneous fistula with anterior abdominal wall mesh repair. Histopathological examination proved the diagnosis of endometriosis. Postoperatively, the patient was given GNRH analogues for 3 months. **Conclusion:** Accurate preoperative diagnosis of a complex entity like uterocutaneous fistula with scar endometriosis using a non invasive imaging modality [MRI] helps in optimal and appropriate patient management. Correct preoperative diagnosis in our patient facilitated good surgical outcome with an uneventful post operative period and resolution of symptoms on follow up.

Key Words: Uterocutaneous fistula, Scar endometriosis, Magnetic resonance imaging [MRI].

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a rare entity, usually occurs as a complication following pelvic surgeries or caesarean section or as a sequelae of infection / inflammation. Case reports have also been described following endometriosis¹ and laminaria tent placement². Only about 120 cases of uterocutaneous fistula have been reported in literature so far³. The peculiar presentation of uterocutaneous fistula makes the definitive treatment challenging². In this report, we seek to highlight the importance of MRI in successful preoperative diagnosis of scar endometriosis with UCF, so that appropriate management can be planned.

INTRODUCTION

Most uterine fistulae occur between the uterus and the bladder or bowel (utero-vesical or utero-colonic) due to postoperative injuries or following infective/inflammatory conditions. Uterocutaneous fistula, by itself

CASE PRESENTATION

A 34 year old Para 2 Living 2 previous 2 LSCS [lower segment caesarean section] presented with complaints of cyclical pain and swelling at LSCS scar site for the past two years, associated with intermittent bleeding and

serous discharge from the scar for the past two months. She has undergone the first LSCS 9 years back and second one 6 years previously. Her menstrual cycles were regular. She had no associated bowel or bladder complaints. On clinical examination, the patient was afebrile. Her vitals were stable. Her upper abdomen was soft with no distension /organomegaly. Suprapubic tenderness with guarding and rigidity was present. Pfannenstiell incision was noted with a swelling of approximate size 3 x 3 cm at the scar site. There was associated serous and bloody discharge from the swelling at the time of examination. Her routine laboratory investigations including CBC were within normal limits. The blood and urine cultures were negative. On per vaginal examination, the uterus showed restricted mobility with both fornices free and non-tender. Ultrasound pelvis revealed a normal sized uterus with endometrial thickness of 9 mm. The endometrial cavity was seen extending into a small hypoechoic collection/lesion anteriorly. Differential diagnosis of infection / inflammatory lesion like suture granuloma or endometriosis were offered. Subsequently, the patient was referred for MRI for better delineation and characterization of the lesion. MRI pelvis showed an acutely retroflexed and anteverted uterus adherent to the anterior abdominal wall. The adjacent anterior abdominal wall along with the previous LSCS scar including the recti muscles and subcutaneous plane showed heterogenous signal intensity lesion [Fig:1 B]with multiple hemorrhagic foci [Fig: 1D] within - features consistent with scar endometriosis [Fig 1B,C and D]. The

inferior 4 cm of the involved recti muscles with the endometriotic lesion were adherent across the midline, whereas superiorly rectal divarication was noted. Additionally, a fistulous tract was seen extending through the endometriotic tissue in the anterior abdominal wall from the endometrium up to the skin [Fig: 1A]. As the external fistulous opening was covered by the protruding soft tissue swelling, the presence of this uterocutaneous fistula had not been suspected prior to MRI. Intra-operatively, the fistulous tract was raised subcutaneously which led to an endometriotic mass of size 8 x 8 cm [Fig - 2A,2B]. As the endometriotic scar was found to be adherent to the anterior abdominal wall, the rectus sheath and muscle were cautiously opened about 5 cm above the mass. The upper quadrant of mass was slowly separated from the rectus sheath and the mass was found adherent to the lower border of the incision. Hence it was carefully separated away from the bladder upto the symphysis pubis. Subsequently the mass was seen adherent to the anterior wall of the uterus, and dissected. In view of extensive adhesions, hysterectomy with bilateral salpingectomy was performed in addition to fistulectomy and excision of the endometriotic tissue. Since the abdominal wall defect was large and difficult to approximate, prolene mesh was placed. Histopathological examination corroborated the diagnosis of endometriosis [Fig 3A, 3B and 3C]. Additionally the patient was given injection leuprolide [GNRH analogue], subcutaneously once monthly for 3 months. Postoperative period was uneventful and the patient did well on follow up after one year.

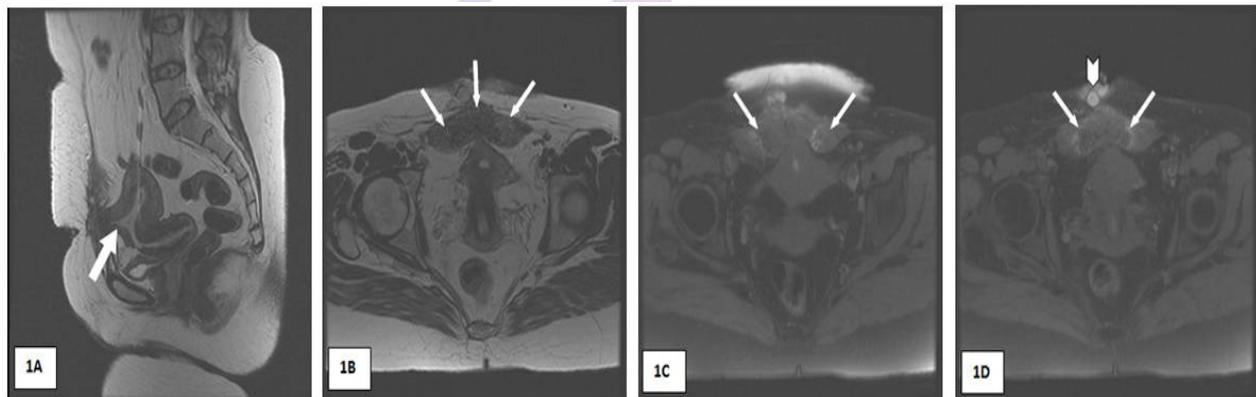


Figure 1: 1A, 1B: Sagittal and axial T2 weighted images reveal the presence of Uterocutaneous fistula [thick arrow] with scar endometriosis [thin arrows] 1C, 1D - Axial FST1 reveal the hemorrhagic foci[arrowhead] within the scar endometriosis.

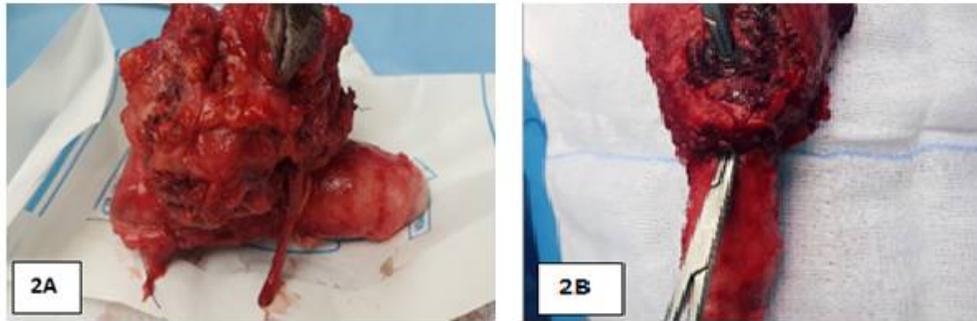


Figure 2A and 2B: Showing the excised endometrial lesion with uterocutaneous fistula, along with the uterus, cervix and bilateral fallopian tubes

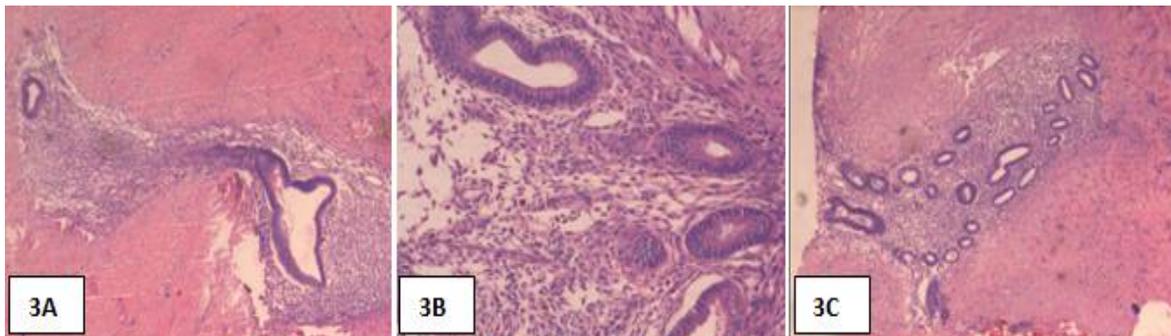


Figure [3A and C]: H and E 40x shows fibrocollagenous tissue infiltrated by benign endometrial glands and stroma; **[3B]:** H and E 100x shows endometrial glands, endometrial stroma and hemosiderin laden macrophages

DISCUSSION

In comparison to uterocutaneous and uterovesical fistulae, uterocutaneous fistulae are infrequently seen² Uterine fistulae including uterocutaneous fistulae usually develop as a complication following pelvic surgeries, infection and inflammatory conditions⁴ Majority of patients with uterocutaneous fistula have a history of caesarian section. In recent years, the frequency of UCFs secondary to caesarean section has declined as a result of the reduction in classical incisions for caesarian section⁵ Various isolated case reports have demonstrated uterocutaneous fistulae developing after septic abortion², pelvic abscesses⁶, true intra-abdominal pregnancy as a result of insufficient placenta removal⁷, Uterovaginal malformation³, infection with actinomyces due to intrauterine devices⁸, curettage, difficult vaginal delivery, or use of forceps⁴. Uterocutaneous fistula may be diagnosed preoperatively using methylene blue injection via the cervix at hysterosalpingogram, at hysteroscopy or on a CT study following instillation of contrast into a fistulous tract^{6,9}. However, all of these diagnostic methods are invasive in nature. The advantage of MRI in detecting uterocutaneous fistula lies in its non invasive nature, precise detection and localization of the fistulous tract and accurate delineation of surrounding soft tissues including possible infective and inflammatory changes. MRI is also useful in precise diagnosis of endometriosis

and excluding lesions like suture granuloma and abscess which can present as a complex hypoechoic / hypodense lesion on USG and CT respectively. Our patient had a prior history of two LSCS, last one 6 years previously. MRI study was performed following ultrasound which had revealed the endometrial cavity communicating with a small collection / lesion located anteriorly. The differential diagnosis included conditions like abscess, suture granuloma and endometriosis. MRI not only confirmed the diagnosis of scar endometriosis [Fig 1B,C and D] but also revealed the presence of a previously unsuspected uterocutaneous fistula [Fig 1A]. Diagnosis and treatment of uterocutaneous fistula may be challenging. Because of its rarity, there is no clear consensus as regards to whether medical treatment, surgical management or a combination of both is the definitive management⁹. Being a rare condition, definitive management of uterocutaneous fistula [less than 15 cases described in the last 20 years worldwide] is yet to be established.¹² Preoperative diagnosis of scar endometriosis with uterocutaneous fistula in our patient helped in planning appropriate treatment. In this instance, our patient [P2L2] had hysterectomy with bilateral salpingectomy, fistula excision [Fig 2A, 2B] and anterior abdominal wall mesh repair. Histopathological examination confirmed the diagnosis of endometriosis [Fig:3A,3B and 3C]. She also underwent

postoperative medical management with GNRH analogues [Injection leuprolide], once a month for three months. Thubert *et al*⁹ presented a case of UCF in a lady with previous h/o caesarian section and stage IV endometriosis with obliteration of the pouch of douglas. The fistulous tract was demonstrated on MRI and hysteroscopy, The patient was treated with combined approach comprising administration of leuprolide acetate [GNRH agonist] subcutaneously for 6 months along with surgery [laparoscopy / laprotomy]. Follow up at 6 months revealed a normal uterine cavity on hysteroscopy. Previously, surgeries ranging from excision of the fistula tract¹⁰ to hysterectomy^{2,6} was considered mandatory in the management of UCFs, however this could potentially be challenging in young patients especially when hysterectomy is being considered. Seyhan *et al* reported a patient treated with gonadotropin-releasing hormone agonist (GnRH) alone, which led to atrophic changes in the epithelium and aided the healing of the fistula. Nonetheless, a larger size of fistula opening in patients prompt surgeons to prefer the surgical approach¹¹ However recent articles have described a combined management plan comprising medical and surgical options which would reduce the chances of hysterectomy⁹. Two case studies have reported successful outcome in patients with uterocutaneous fistula and endometriosis, treated with GNRH analogues alone^{11,13}.

CONCLUSION

Uterocutaneous fistula is an extremely rare clinical entity. Accurate preoperative diagnosis of uterocutaneous fistula with its probable cause [in this instance, scar endometriosis] may be possible using a non invasive imaging modality like MRI. This would help in optimal patient management with a good outcome.

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