

# Uterocutaneous Fistula- A Case Series

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## ABSTRACT

Uterocutaneous Fistula (UCF) is an uncommon condition, which usually occurs as a postoperative complication. Authors, hereby presented three cases of UCF. First case was of 40-year-old multigravida who presented with pus discharge from anterior abdominal wall for three months duration following an open uterine myomectomy. Her UCF was confirmed after methylene blue dye test and Magnetic Resonance Imaging (MRI). She had unsuccessful closure of UCF after three months of Gonadotropin-Releasing Hormone (GnRH) analogue therapy and complete excision of the fistulous tract was achieved later. She was asymptomatic after 24 months of follow-up. Second and third case was primigravida patients who developed UCF after caesarean section and were managed conservatively. Uterocutaneous fistula is a rare clinical entity, and its prevention is the best strategy with avoidance of postoperative infection and better surgical technique. Authors reviewed here the cases of UCF after abdominal myomectomy and caesarean section and its management.

**Keywords:** Caesarean, Diagnosis, GnRH analogues, Myomectomy, Obstetric fistula

## INTRODUCTION

Fistulous communication is an abnormal tract between two epithelial surfaces, whether it is between lumen to lumen or lumen to exterior. Obstetric fistula is a common occurrence after prolonged labour or postoperatively. Vesicovaginal, rectovaginal or combined fistula are common type [1]. Obstetric fistula usually lead to depression, sexual incompatibility, social stigma, divorce, ostracisation and marginalization of the patient in the society [2]. UCF is a rare condition, which usually occurs postpartum or postoperative complications after caesarean section [3]. Uterine fibroid is the most common benign tumour of the uterus and is usually treated with hysterectomy [4]. Abdominal myomectomy is a preferred choice for uterine fibroid in patients who want to preserve their fertility [4]. Authors report three cases of UCF after uterine myomectomy and caesarean section which is extremely rare.

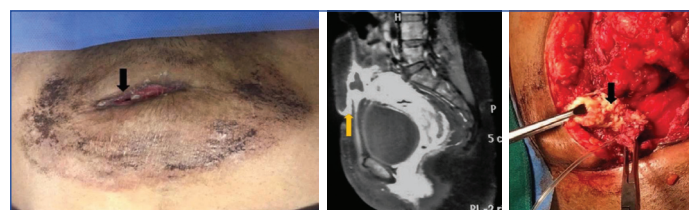
## CASE SERIES

### Case 1

A 40-year-old female, multigravida presented with secondary amenorrhoea for four months and pus discharge from anterior abdominal wall for three months duration. She had prior history of open uterine myomectomy five months back for multiple uterine fibroids. She also complained about mild suprapubic pain and intermittent mild fever for 20 days duration. Local examination revealed tender incision site and pus discharge from incision site on finger pressure [Table/Fig-1]. Pus culture was sterile and Ziehl-Neelsen stain for Acid Fast Bacilli (AFB) was non contributory. Initially, she was treated with broad-spectrum intravenous antibiotics for two weeks duration and as there was no improvement in her symptoms, and she was referred to our centre.

Her investigations revealed haemoglobin of 10.7 gm/dL, total leukocyte count was 5300/mm<sup>3</sup>; and liver and kidney function tests were within normal limits. Methylene blue dye test showed appearance of dye from pus discharge site from anterior abdominal wall, confirming uterocutaneous fistula. Magnetic resonance imaging showed a fistulous tract connecting between uterine cavity near fundus with the anterior abdominal wall and surrounding inflammation [Table/Fig-2].

After confirming the uterocutaneous fistula, she was started on injection leuprolide acetate 3.75 mg intramuscular in buttock region monthly for three months and broad spectrum antibiotics for 10 days. Her pus discharge decreased, however, not completely resolved. After three months, she underwent open laparotomy and excision of fistulous tract [Table/Fig-3]. The histopathological analysis showed fibrinous exudate with mixed inflammatory cells and Ziehl-Neelsen stain for AFB was negative. She is currently symptom free after two years postsurgery.



**[Table/Fig-1]:** At anterior abdominal wall, there is surgical scar, granulation tissue and fistulous opening with pus discharge is visible (black arrow).

**[Table/Fig-2]:** Magnetic resonance imaging showing fistulous tract (yellow arrow) in case 1. **[Table/Fig-3]:** Intraoperative photograph showing resected fistulous tract (black arrow) with infant feeding tube into the fistulous tract. (Images from left to right)

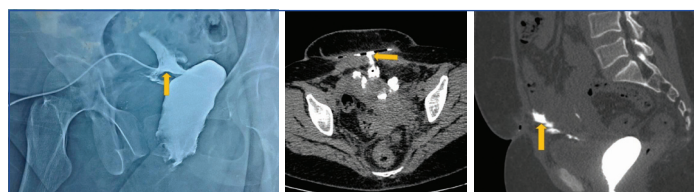
### Case 2

A 21-year-old female, primigravida at 37 weeks of gestation presented in gynaecology emergency as preeclampsia with foetal growth restriction with history of fever one week back. She underwent caesarean section in view of foetal distress. From postoperative day 3, patient started having fever with complain of wound discharge. On examination, pus discharge was seen, complete suture removal was done, and intravenous antibiotics (ceftriaxone and metronidazole) was given. Fever settled down from postoperative day 10 with persistent pus discharge from wound. Sinogram [Table/Fig-4] and Computed Tomography (CT) scan were done [Table/Fig-5], and patient was diagnosed with UCF. She was continued i.v. antibiotics, pus discharge slowly decreased. Patient was kept on close follow-up for 12 weeks and fistula resolved completely.

### Case 3

A 22-year-old female, primipara patient presented to emergency on postoperative day 18 of caesarean section with complains of discharge from wound and history of fever 10 days back. She underwent caesarean section at a private hospital in view

of meconium-stained liquor and patient had preeclampsia. On examination, superficial surgical site infection was present with minimal slough at the base. Pus was sent for culture sensitivity and i.v. antibiotics started. Computed tomography scan was done which revealed presence of UCF [Table/Fig-6]. Patient was discharged after fever subsided and was called for daily dressing. Wound healed gradually and fistula resolved. With conservative management, fistula resolved after eight weeks.



**[Table/Fig-4]:** Sinogram showing the fistula (yellow arrow) in case 2.

**[Table/Fig-5]:** CT scan showing Uterocutaneous Fistula (UCF) (yellow arrow) in case 2.

**[Table/Fig-6]:** CT scan showing Uterocutaneous Fistula (UCF) (yellow arrow) in case 3. (Images from left to right)

## DISCUSSION

Uterocutaneous fistula is a rare complication, usually occurs in postoperative period after caesarean section. Mostly, it was reported after classical caesarean section, however its incidence has decreased after improvement in technique of caesarean section [5,6]. There is no systemic study to determine the incidence and prevalence of this condition.

In the present case series, the first case, UCF occurred after abdominal myomectomy. Only four cases of UCF were reported after myomectomy [Table/Fig-7]. In one case, patient developed UCF after multiple abdominal myomectomies [5], in another case after removal of large sub-serosal fibroid [7] and after myomectomy [8,9]. Myomectomy can result in other type of fistula also like iliouterine, colouterine [10], uteroperitoneal [11], and uterine arteriovenous [12]. Uterocutaneous fistula formation has been reported after caesarean deliveries [3], endometriotic nodule from caesarean [13], infected placenta increta after caesarean [14], complicated appendicular abscess followed by caesarean section [15], gynaecological malignancy [16], septic abortion induced by introduction of Laminaria tent into the uterus, after surgical intervention for cryptomenorrhea [17], after an incomplete medical abortion or held placenta after caesarean [18], with uterine malformation [19]. UCF is reported in patients with term abdominal pregnancy [20] and after retained placenta.

In the present case series, the second and third case, UCF formation occurred after C-section. The duration of occurrence of UCF varies from weeks in postoperative period to 19 years after surgery in different case reports [21]. Tuberculous infection is particularly prevalent in Indian sub-continent and possibility of tuberculous UCF should be always kept as a possibility in these regions.

The postulated mechanism of development of UCF is either infectious process which disrupts the continuity of tissue barrier or dense adhesions between the uterus and the abdominal wall, which became thinner and less vascularised, results into the fistula formation. In most cases it is postoperative fistula formation, indicating a better surgical practices and postoperative care might reduce the chance of fistula formation. Another postulated mechanism is preoperative uterine artery embolisation before myomectomy lead to less blood supply and impair healing, resulting into the UCF. Genital flow obstruction in previously operated patient takes the path of least resistance and can develop UCF [17]. Red degeneration of fibroid complicated with infection can also lead to UCF due to decrease blood supply [22]. Endometriosis surgery might lead to implantation of endometriotic tissue in the scar, resulting in to the formation of UCF [3]. Other factors which might be implicated are non absorbable suture use which is nidus for infection, incomplete closure of caesarean section incision or non closure of visceral peritoneum [23]. Adhesion barrier is recognised to increase the risk of fistula and leak in abdominopelvic surgery [24], which probably hinders the natural process of wound healing. One should keep in mind that using adhesion barrier might increase the risk of fistula.

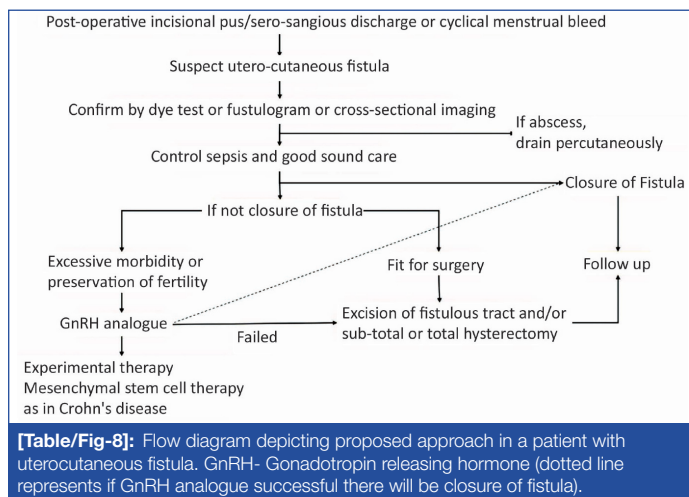
Diagnosis of UCF requires a high index of suspicion. It should be clinically suspected if cyclical menstrual discharge from the cutaneous site during menstruation or purulent, seropurulent or sero-sanguinous discharge from the previous scar site. Confirmation of the diagnosis can be easily done bedside by methylene blue dye test but can be fallacious in some cases due tight fibrotic stricture. Fistulogram/sinogram or hysterosalpingography is other radiographic techniques, which can confirm the diagnosis, but can be falsely negative also in some cases [22]. Cross-sectional imaging like Contrast Enhanced Computed Tomography (CECT) or Magnetic Resonance Imaging (MRI) has outmoded the conventional radiologic imaging and will give other information beyond the fistulous lumen, like anatomical variant, abscess, or evidence of lymphadenopathy in case of tuberculous aetiology. MRI has intrinsically superior soft tissue contrast and help in better delineating of the fistulous tract, which helps in better planning of the surgery preoperatively. Magnetic resonance imaging can also provide better information regarding activity of the disease and around the fistulous tract better than CT scan, as in perianal fistula or Crohn's disease [25]. On heavily T2-weighted MRI sequences, the UCF is seen as a fluid filled hyperintense tubular structure as in biliary tract imaging by Magnetic Resonance Cholangiopancreatography (MRCP) [25]. There is no consensus on imaging, because of rarity of the disease, however, in our opinion, MR imaging should be preferred due to non invasiveness, no radiation and better delineation of fistulous tract, but cost is prohibitive.

Author/year	Place	Age (year)	Gravida	Procedure/aetiology	Diagnosis	Treatment	Follow-up
Loué V et al., 2013 [9]	Africa	30	Primigravida and Nulliparous	Multiple abdominal myomectomies with wound sepsis	Methylene blue dye test, Hysterosalpingography	Exploratory laparotomy with excision of fistulous tract; and high dose of norethisterone for three months.	Three months
Akkurt MO et al., 2015 [5]	Turkey	42	Nulliparous	Multiple abdominal myomectomies	Ultrasonography and Magnetic resonance imaging	Laparotomy and hysterectomy.	One month
Ilhan G et al., 2017 [7]	Istanbul	32	Nulliparous	Abdominal myomectomy	Contrast enhanced computed tomography	Exploratory laparotomy with excision of fistulous tract and primary repair of myometrium.	Three months
Min KJ et al., 2018 [8]	Korea	30	Nulliparous	Pelvicoscopic myomectomy	Clinical and hysterosalpingography	Explorative laparotomy repair of dehiscenced myometrium. Repeat total removal of fistulous tract and repair of dehiscenced myometrium and GnRH agonist for three months.	Three months
Present case series (2021)	India	40	Multigravida	Abdominal myomectomy	Methylene blue dye test, hysterosalpingography and Magnetic resonance imaging	GnRH agonist for three months and exploratory laparotomy with hysterectomy.	18 months

**[Table/Fig-7]:** Case reports of Uterocutaneous Fistula (UCF) after myomectomy [5,7-9].

GnRH: Gonadotropin releasing hormone

There is no consensus regarding treatment and mostly it was treated surgically [Table/Fig-8]. Spontaneous closure of fistula with conservative management is an exception [16,26]. In present case series, first case was treated surgically and second and third case was managed conservatively. Medical management can rarely lead to closure of the fistula. The most important is to contain sepsis by drainage of abscess, intravenous antibiotics, wound care and nutritional optimization for better wound healing. Two cases of UCF were successfully treated with Gonadotropin-Releasing Hormone (GnRH) analogues [27,28]. It require multiple doses at monthly interval and can be given upto six doses [29], however single dose of GnRH analogue can be sufficient to close the fistula [27].



The GnRH analogues cause cessation of menstruation, atrophy of uterine endometrium and decrease discharges leading to healing of the fistula [28]. Fistula healing can be permanent as reported in follow-up upto two years [28]. In patients, who want to preserve their fertility or those who are at high risk for further surgery, GnRH analogues can be tried. In our first case, also GnRH analogues was tried but due to persistent discharge after three months, it was stopped. Combined medical and surgical approach can be tried due to reducing the discharge by GnRH analogues and facilitating for further surgery [29]; and GnRH analogue or hormonal therapy can be used after surgery to prevent menstruation and augment better healing after surgery and prevent recurrence of fistula [8]. Endoscopic repair of UCF is reported to be successful, can be tried in suitable candidate [30].

If patients did not want to preserve the fertility or completed their family and in cases of failure, surgery is the best option. The surgical options include excision of the fistulous tract with or without hysterectomy. The surgical techniques include injection of methylene blue dye intraoperatively and identification of fistulous tract during surgery and it can also be identified by probe intraoperatively. Combined laparoscopy and laparotomy approach can also be attempted on individual basis [29]. In case of anatomical malformation one or two step surgery can be performed, first correcting the anatomical abnormality followed by correction of UCF [19].

In index case, the patient underwent total abdominal hysterectomy with fistulous tract excision, as she completed her family already. In case of UCF after abdominal myomectomy, total abdominal hysterectomy with fistulous tract excision or fistulous tract excision with closure of myomectomy site can be attempted to preserve the fertility. In future, medical therapy can be an option by further characterising the pathogenesis of the fistula if it is hormone dependent or Tumour Necrosis Factor (TNF)- $\alpha$  dependent as in inflammatory bowel disease [15]. Local injection of mesenchymal stem cell therapy can be exciting option as in perianal fistula of Crohn's disease. With rising rate of caesarean section, chances of

UCF will be higher. The best option would be the better surgical techniques and prevention of infection postoperatively.

## CONCLUSION(S)

Uterocutaneous fistula is a rare clinical entity and best would be its prevention to avoid postoperative infection and better surgical technique. However, in case of occurrence it can be treated medically or surgically depending on surgical risk and wish to preserve fertility.

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