

Umbilical and Juxtaumbilical Chronic Discharging Sinus as a Delayed Complication of Caesarean Section: A Case Series

SUKUMAR MITRA¹, SHYAMALI DUTTA², KRUTIKA SANDEEP SAWANT³, SOHINI SEN⁴, MD MOHSIN⁵

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ABSTRACT

Cutaneous or uterocutaneous sinus or fistula following caesarean section is a rare complication that can occur after the primary wound has healed. Only a few cases have been reported where a fistula or sinus tract forms after a caesarean section. These tracts can communicate externally through the scar line or the skin near the umbilicus (juxtaumbilical) and internally to the uterine scar or an unhealthy abnormal cavity. It is a delayed complication that presents with discharge from the skin opening and can be diagnosed using imaging techniques such as Ultrasonography (USG), Magnetic Resonance Imaging (MRI), Fistulogram, or Sinogram. These tracts often seek weak areas to establish external or internal connections with other organs. The management of these abnormal tracts can vary from conservative approaches to surgical excision. Herein, the authors presented a series of three cases of cutaneous and uterocutaneous fistula following caesarean section. In case 1, a 19-year-old female patient presented with a discharging sinus from the umbilicus, which was found to be a sinus tract communicating inside the abdominal wall to the external skin on imaging. The patient underwent surgery, which involved excision of the sinus tract and necrotic debridement. Two similar cases (cases 2 and 3) were infra and paraumbilical regions that presented with chronic discharges in the infra and para umbilical region following Lower Segment Caesarean Section (LSCS) delivery. Both cases were diagnosed with cutaneous sinus tracts in the juxtaumbilical region using USG and other imaging modalities. They were managed conservatively with appropriate antibiotics and achieved complete healing without any residual infection during follow-up.

Keywords: Abdominal wall, Cutaneous sinus, Paraumbilical region, Uterocutaneous fistula

INTRODUCTION

Cutaneous and uterocutaneous sinus or fistula are rare conditions noted after caesarean section or abdominopelvic surgeries. They communicate internally with the uterus and externally with the vagina or skin. Some may also communicate between the uterus and bowel or bladder [1]; others open externally through the skin without any definite internal communication, resulting in sinuses [2-4]. Most cases result from infection, inflammation, injury, incomplete closure of the incision line, and inappropriate wound healing in the perioperative and postoperative period [5].

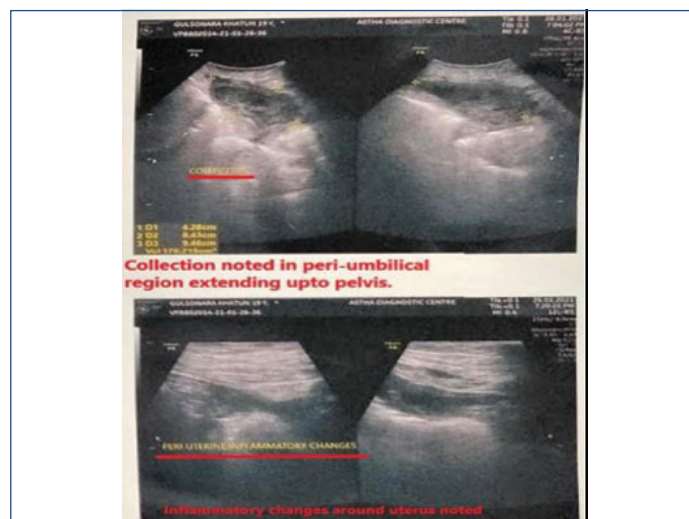
A thorough history, clinical examination, and detailed investigations including sonography, Computed Tomography (CT) scan, MRI, Fistulogram, or Sinogram are necessary to identify both complete and incomplete types (blind type/sinus) of abnormal tracts. Patients typically present with discharge through the external opening, with or without pain and swelling [2,5]. Because of uncommon presentation, the choice of treatment can be challenging. Suitable antibiotics and/or surgical intervention may be needed depending on the underlying causes, location, extent of the fistula tract, and signs and symptoms [2].

Case 1

A 19-year-old female patient underwent Emergency Lower Segment Caesarean Section (Em-LSCS) due to foetal distress. On days 6-7 of the postoperative period, the patient developed a stitch line infection which was managed with parenteral antibiotics. The infection gradually resolved, and the wound healed. On day 14 postdelivery, she was discharged without any residual stitch line infection. Three weeks after the postnatal period, the patient visited the Outpatient Department (OPD) with a complaint of discharge from the umbilicus for five days. She did not report any cyclical bleeding or pain from or around the opening of the caesarean section scar. Additionally, she had no history suggestive of a drain,

mesh, or Intrauterine Device (IUD) placement for contraception during her surgery. On examination, a pus-like discharge was noted from the anterior abdominal wall at the level of the umbilicus. Palpation of the discharge opening revealed tenderness and induration. The uterus was substantially involuted, and both fornices were free with no palpable masses during pervaginal examination. Clinical examination did not reveal any collections. Perspeculum examination was normal.

Pelvic organ ultrasound showed a thick-walled multiseptated anechoic collection at the subumbilical region extending downwards up to the LSCS scar in the anterior abdominal wall. Additionally, a bulky puerperal uterus, distended bowel loops, and a mild collection in the Pouch Of Douglas (POD) were observed [Table/Fig-1].



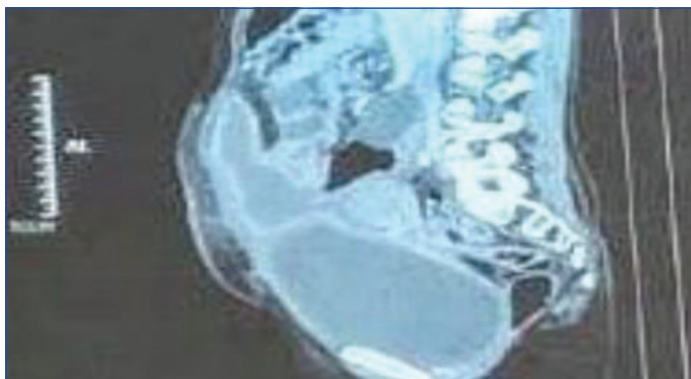
[Table/Fig-1]: Case1: Ultrasonography (USG) shows collections and inflammatory changes around the uterus (marked).

Contrast Enhanced Computed Tomography Scan (CECT) showed a large loculated thick-walled collection in the lower and mid abdominal wall, superior to the peritoneum, with an external communication to the umbilicus [Table/Fig-2].

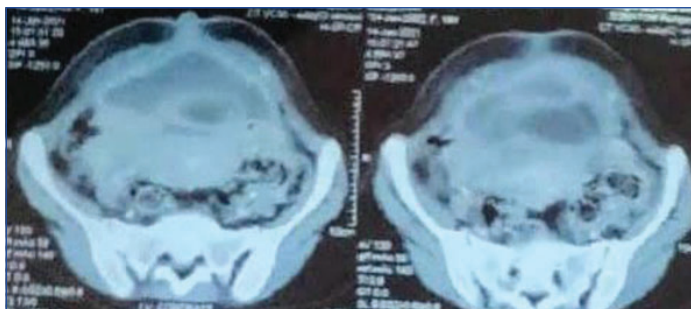


[Table/Fig-2]: Case 1: Contrast Enhanced Computed Tomography Scan (CECT) scan shows collections at lower and mid. abdominal wall.

The MRI of the pelvis showed a thin and irregular communication extending to the anterior abdominal wall, suggestive of a chronic sinus [Table/Fig-3,4]. The adjacent bowels appeared oedematous and adhered together with peritoneal fat. However, there was no communication between the uterus and the discharging tract.



[Table/Fig-3]: Case1: Magnetic Resonance Imaging (MRI) shows thin irregular communication with anterior abdominal wall.



[Table/Fig-4]: Case1: Magnetic Resonance Imaging (MRI) shows collections in the anterior part of uterus.

Sinogram showed a contrast-filled tract within the abdominal wall without any peritoneal or uterine connection [Table/Fig-5]. [Table/Fig-6] depicts the tract after 25 minutes of dye injection. Laparotomy was planned for the patient. During the procedure, a tract was identified superficial to the peritoneum below the



[Table/Fig-5]: Case1: Sinogram shows contrast filled tract.



[Table/Fig-6]: Case1: Sinogram after 25 minutes of dye injection, shows the sinus.

rectus sheath, extending from the umbilicus to the area of the caesarean scar. Local inflammatory reaction characterised by omental adhesion and induration was also observed. After adhesiolysis, the sinus tract was excised, and the abdomen was closed in layers. The postoperative period was managed with broad-spectrum antibiotics, and the wound healed without any complications. The histopathological report showed fibrocollagenous tissue and necrotic smooth muscle lined by epithelium with serosanguinous discharge, accompanied by acute inflammatory infiltrate with occasional groups of epithelioid-like cells. Occasional haemosiderin-like macrophages were also seen. No appreciable endometrial glands were observed. During the six months follow-up, the wound healed completely without any recurrence.

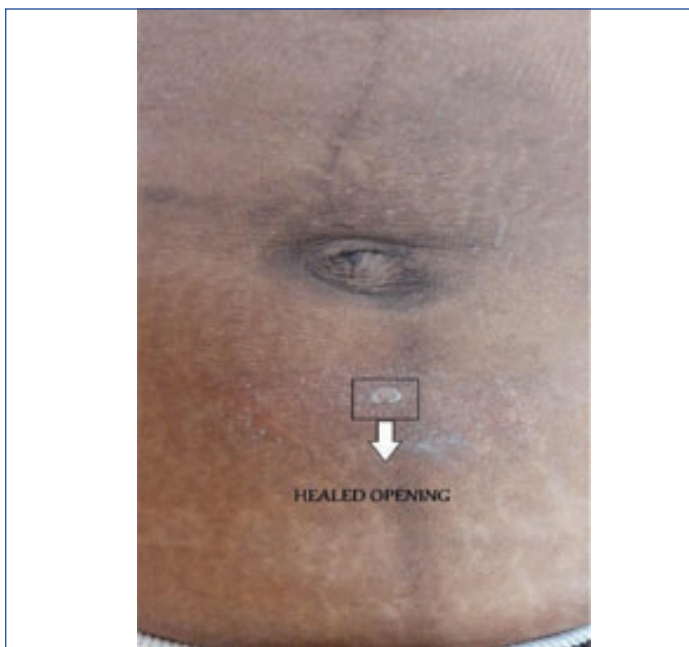
Case 2

A 26-year-old woman visited the OPD one and a half months after a caesarean section. The patient reported a one-week history of discharge through an opening located 2 cm below the umbilicus in the midline. Upon inquiry, she disclosed that she had a wound infection following the caesarean section, which was treated by a local doctor, resulting in complete healing of the incision. On examination, the infraumbilical cutaneous opening was found to be tender, erythematous, and oedematous, with purulent discharge. The patient had no fever, vaginal discharge, or vaginal bleeding.

Sinogram revealed an irregular sinus tract extending from the external wound in the infra and paraumbilical region, moving downwards [Table/Fig-7]. There was no communication with internal organs such as the gut, bladder, or uterus. The patient was planned for medical management. The pus was sent for culture sensitivity, and she was treated with appropriate antibiotics according to the culture sensitivity report. After two weeks, the sinus healed permanently [Table/Fig-8]. There was no recurrence after six months of follow-up.



[Table/Fig-7]: Case 2: Sinogram shows the sinus tract.

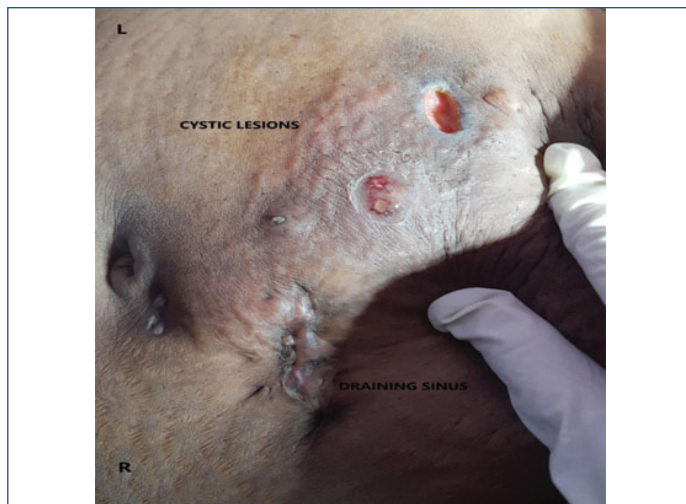


[Table/Fig-8]: Case 2: Healed opening of the discharging sinus after conservative treatment.

Case 3

A 33-year-old woman underwent a caesarean section for failed induction of labour at term. On the 6th day post-caesarean section, the stitches were removed, and the stitch line appeared healthy. The patient was discharged. Three months later, the patient presented with discharges from the paraumbilical region in the OPD. Prior to her visit, she had been prescribed local and oral antibiotics by her local physician, but her symptoms did not improve. Clinical examination revealed a swollen, indurated, firm to hard skin surface around the umbilicus. No masses or nodules were palpable. A few cystic lesions filled with pus were observed, some of which had ruptured. On the right side of the umbilicus, another indurated surface was noted, from which pus-like discharge came out upon pressure. The patient had no history of fever [Table/Fig-9]. Her other systemic examinations were normal. Ultrasound revealed a collection of fluid in the deep subcutaneous space in the paraumbilical region [Table/Fig-10]. A sinus tract in the anterior abdominal wall in the right paraumbilical region, filled with echogenic material suggestive of pus, was also observed.

The CT scan showed anterior wall cellulitis with a collection noted in the paraumbilical region. The patient was planned for medical management with sensitive antibiotics based on her



[Table/Fig-9]: Case 3: A clinical picture of the patient.



[Table/Fig-10]: Case 3: Ultrasonography (USG) shows sinus tract with echogenic material.

culture sensitivity report. The patient was found to be sensitive to trimethoprim/sulfamethoxazole (co-trimoxazole). She was prescribed this antibiotic at double the regular dose (160 mg/800 mg) orally twice daily for two weeks. After three weeks, the cystic skin lesions healed, and no discharge was noted through the sinus tract. The patient was followed-up for six months and there was no history or signs of recurrence.

DISCUSSION

Cutaneous fistulae and sinuses are rarely seen in clinical practice and usually occur congenitally or after caesarean section or other abdominopelvic surgeries [2]. Most uterine fistulae are vesicouterine or vesicovaginal fistulae, and umbilical fistulae can be of pilonidal, urachal, or enterocutaneous varieties [4]. Uterocutaneous fistula is defined as an abnormal communication between the uterus and the skin, resulting in umbilical or juxtaumbilical openings. These sinuses or fistulae occur due to chronic inflammation and the formation of an abnormal tract through which pus-like discharge drains out onto the skin [2,5]. The lower incidence of uterocutaneous fistulae is attributed to the type of operation and sutures used in modern obstetrics [3].

Most cutaneous or uterocutaneous fistulae or sinuses originate from postoperative infection of the uterine or abdominal wound or necrosis of the anterior abdominal wall due to persistent wound infection. Causes can include caesarean section, postseptic abortion, incomplete placental removal, pelvic abscess, placement of a drain, infected Intrauterine Contraceptive Device (IUD) or mesh, improper healing of the incision line due to the use of non absorbable sutures, adhesions resulting from repeated lower abdominal operations, multiple myomectomies, missed uterine perforation, scar dehiscence, prolonged bleeding leading to postpartum haemorrhage where a B-lynch suture has been placed, which may result in a uterine fistula later on [6,7]. After healing of the major wound has healed, any residual infective collection searches for an easy passage and ultimately opens externally, forming a fistula. Other causes may include endometriosis [8,9], as bleeding comes from the incision line, secondary changes due to red degeneration of fibroids, postsurgical interventions like cryptomenorrhoea, and malignancies like endometrioid adenocarcinoma, which can weaken

a caesarean scar and lead to fistula formation [10]. Risk factors for the development of chronic discharging tracts include persistent wound infections following abscess formation, repeated abdominal operations, improper closure of abdominal layers of the abdomen in abdominopelvic surgeries, and the use of non absorbable sutures. The skin surrounding the discharging sinus becomes swollen, tender, and indurated, with malodorous pus discharge coming out from the skin opening. USG, CT scan, MRI, and sinogram can help in the diagnosis of such abnormal tracts, and the course of the tract can also be visualised. Therefore, imaging modalities play a crucial role in the diagnosis of such sinuses or fistulae [11].

Wound infections are common following caesarean section, and complications are often related to infections mostly near or at the incision line. Maddah G et al., reported a similar case of a fistula tract communicating with the uterus following caesarean section. The patient presented with an abdominal mass and a collection two months after surgery. The case was treated surgically by debriding the necrotic tissue and excising the fistula tract [2]. Jadib A et al., also reported a similar fistula, which developed seven months after a caesarean section. The patient presented with pain and blood discharge from the cutaneous scar for four months [7]. Dragoumis K discussed a case of uterocutaneous fistula that developed six years after a previous caesarean section. The cause was reported to be an endometriotic lesion resulting in inflammation and bleeding [8]. Lim PS et al., described a case of a uterocutaneous fistula where there was communication between the endometrial cavity and a skin lesion via a necrotic intramural fibroid following a caesarean section [12].

In case 1,2 and 3 of the present case series, the initial wound infection following caesarean section healed completely, but after a few months, chronic discharging sinuses developed at the umbilical or juxtaumbilical region. This may be due to the persistence of an indolent infective collection within the layers of the abdominal wall, which finds a weak tract and area to open. The authors proposed that the weak tract lies along the line of the median umbilical ligament, and the weak point for the tract to open is the umbilicus and juxtaumbilical region. In case 1, the sinus tract was excised through laparotomy, and intraoperative findings confirmed the location of the tract in the midline and extraperitoneally, supporting the proposal.

The management of cutaneous and uterocutaneous sinuses or fistulae depends on the type, symptoms, underlying cause, and extent of communication with other structures. After careful assessment, cases can be treated with medical interventions, surgical interventions, or a combination of both. Previous cases have been managed in various ways. Akkurt MO et al., showed the use of Gonadotropin Releasing Hormone (GnRH) analogues for spontaneous closure of the tract [11]. GnRH agonists induce atrophic changes in the epithelium and assist in the closure of the fistula. However, if the fistula opening is large, a surgical approach may be necessary. The exact treatment, as discussed by many authors in their cases, involved excision of the fistula tract with or without hysterectomy [12,13]. Thubert T et al., discussed

the combined use of conservative surgical and medical treatment for a uterocutaneous fistula. They administered leuprolide acetate to the patient for the first six months, followed by a laparotomy [14].

In the index cases, authors successfully managed one case (case 1, umbilical sinus) through a surgical approach. Excised the discharging tract, performed adhesiolysis, debrided necrotic, and administered antibiotics in the postoperative period. Other cases (cases 2 and 3 juxtaumbilical sinus), treated the patients with appropriate parenteral antibiotics.

CONCLUSION(S)

Cutaneous or uterocutaneous sinuses or fistulae are rare complications of caesarean section, with infection being the most common cause. They typically occur along a weak tract in a weak area. In these two cases, chronic discharging sinuses developed following caesarean section along the weak tract of the median umbilical ligament. One case opened at the umbilicus (umbilical sinus), while the other two opened slightly below the umbilicus (juxtaumbilical sinus). Depending on the clinical presentation and investigation findings, the sinus tract may be treated surgically by excising it or conservatively with appropriate antibiotics.

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PARTICULARS OF CONTRIBUTORS:

1. Associate Professor, Department of Obstetrics and Gynaecology, Medical College Kolkata, West Bengal, India.
2. Assistant Professor, Department of Obstetrics and Gynaecology, Medical College Kolkata, West Bengal, India.
3. Associate Professor, Department of Obstetrics and Gynaecology, Medical College Kolkata, West Bengal, India.
4. Resident, Department of Obstetrics and Gynaecology, Medical College Kolkata, West Bengal, India.
5. Resident, Department of Obstetrics and Gynaecology, Medical College Kolkata, West Bengal, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:

Dr. Sohini Sen,
Resident, Department of Obstetrics and Gynaecology, Golf Club Road Tollygunge,
Kolkata-700033, West Bengal, India.
E-mail: sohini.sen93@gmail.com

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