

# Tubocutaneous Fistula: A Rare Entity

Anjum Ara<sup>1</sup>, Kanika Kumari<sup>2</sup>, Sushma Rani<sup>3</sup>, Indu Chawla<sup>4</sup>, Vaddi S Babu<sup>5</sup>, Sunayana Misra<sup>6</sup>

## ABSTRACT

Fistula between fallopian tube and skin, i.e., tubocutaneous, is extremely a rare entity. Very few cases were reported in the literature. We report a case of a 38-year-old woman who presented with chronic discharging left inguinal wound secondary to genital tuberculosis (TB) for which she had taken 6 months antitubercular treatment (ATT). It was diagnosed as tubocutaneous fistula on computed tomography (CT) sinogram. Complete surgical resection in the form of salpingectomy or salpingo-oophorectomy with complete fistula excision and supplementing it with long-term ATT is the treatment of choice in such patients.

**Keywords:** Computed tomography sinogram, Genital tuberculosis, Tubocutaneous fistula.

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## INTRODUCTION

Fistulas routinely encountered by gynecologists are vesicovaginal, ureterovaginal, and rectovaginal fistulas; sometimes uterocutaneous fistulas are also reported but fistulas between fallopian tube and skin, i.e., tubocutaneous fistulas or salpingocutaneous fistulas, are relatively rare. They present as chronic discharging pelvic sinuses and causes can be surgical or gynecological.<sup>1</sup> There are very few case reports of tubocutaneous fistulas reported in the literature.

## CASE REPORT

A 38-year-old woman with a history of previous three lower segment cesarean section (LSCS) presented with chronic discharging wound in left inguinal region for 1.5 years. Discharge was serous in nature and used to be blood stained during menses associated with slight pain in the abdomen. She had a history of similar discharge from the right inguinal region also. The patient was evaluated for these bilateral sinuses in a periphery hospital where on CT scan bilateral tubo-ovarian (TO) masses were identified and antitubercular treatment (ATT) was started provisionally. After taking ATT for 6 months, her right inguinal sinus healed but active discharge continued from left inguinal sinus.

On gynecological evaluation, she had a history of previous three lower segment cesarean section last being done 7 years back. There was no personal or family history of tuberculosis (TB) or inflammatory bowel disease.

On examination, the patient had three infraumbilical vertical scars of previous LSCSs/rest abdomen was soft. A healed sinus scar was present in right inguinal region and an active discharging sinus was present in left inguinal region. On bimanual examination, bilateral cystic masses were present in both the adnexae. Contrast-enhanced computerized tomography (CECT) was suggestive of bilateral pyosalpinx, right side: 5.6 × 3.9 cm/left side: 5 × 4.8 cm. computed tomography (CT) sinogram showed an irregular linear tract of 9 cm from skin to the left adnexa (Fig. 1).

Specific investigations like culture, TB-PCR, CBNAAT of discharge, biopsy of sinus tract, and endometrium were negative for TB and malignancy. Smears for fungal staining were also negative. Other causes of chronic discharging sinuses were ruled out, and in joint consultation with surgeons, the patient was planned for laparotomy.

<sup>1</sup>Department of Obstetrics and Gynaecology, PGIMER and Dr RML Hospital, New Delhi, India

<sup>2</sup>ABVIMS and RML Hospital, New Delhi, India

<sup>3,4</sup>Department of Obstetrics and Gynaecology, ABVIMS and RML Hospital, New Delhi, India

<sup>5</sup>Department of Plastic Surgery, ABVIMS and RML Hospital, New Delhi, India

<sup>6</sup>Department of Pathology, ABVIMS and RML Hospital, New Delhi, India

**Corresponding Author:** Anjum Ara, Department of Obstetrics and Gynaecology, PGIMER and Dr RML Hospital, New Delhi, India, Phone: +91 1123741635, e-mail: life\_dranjum@yahoo.com

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On exploratory laparotomy, bilateral TO masses were present, and tubes and ovaries could not be delineated separately from the TO masses. Left side TO mass of 5 × 5 cm was adherent at the internal opening of deep inguinal ring and the fistulous tract traversed along the entire length of left inguinal canal. Right side TO mass was 3 × 4 cm in size. Total abdominal hysterectomy with the removal of bilateral TO masses with excision of fistulous tract was done (Fig. 2).

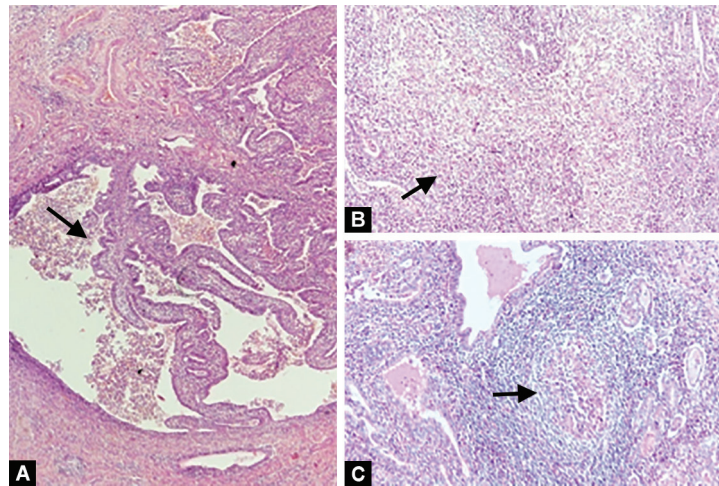
Histopathology was suggestive of granulomatous salpingitis. ATT was started in postoperative phase. Left-sided sinus healed completely in her subsequent follow-up visits.

## DISCUSSION

Fistula is an abnormal communication, connecting two hollow organs or connecting a hollow organ with skin or mucous membrane. Gynecologists are familiar with vesicovaginal, ureterovaginal, and rectovaginal fistula. Fistula between fallopian tube and skin is extremely rare.<sup>1</sup> They can present as chronic discharging wounds. Causes of persistent pus discharging sinuses can be dermatological like cryptococcosis, actinomycetes, surgical, or gynecological. Tubocutaneous fistulas are mainly seen after the complication of



**Fig. 1:** CT sinogram sagittal oblique view—nonionic iodinated contrast agent measuring approximately 10 mL through an opening in left iliac fossa showing a linear irregular tract of approximately 9-cm-long tract extending from skin to left adnexa



**Figs 2A to C:** Dense lymphoplasmacytic to histiocytic aggregates with well-defined granulomas s/o granulomatous salpingitis. Connective tissues showed lymphocytic aggregates

underlying pathologies like pelvic inflammatory disease, genital TB, endometriosis, inflammatory bowel disease, postradiotherapy, and pelvic surgeries.

Female genital TB is a chronic disease with low-grade symptoms affecting fallopian tubes in almost all (90–100%) of the cases and involvement is usually bilateral. The age of presentation in 80% of women is 20–40 years age group, especially in the developing countries. Its prevalence is underreported due to asymptomatic cases, paucibacillary nature, and lack of reliable confirmatory investigation. The diagnosis is made by the detection of acid-fast bacilli (AFB) on microscopy or culture on endometrial biopsy or on histopathological detection of an epithelioid granuloma on endometrial biopsy. Because of its paucibacillary nature, it sometimes becomes difficult to demonstrate AFB.<sup>2</sup> Ultrasound, CECT, and magnetic resonance imaging (MRI) are helpful for diagnosing hydrosalpinx and pyosalpinx and TO masses. CT sinogram is the preferred investigation in chronic sinus cases.

Treatment depends upon age and fertility options of the women. Medical management with ATT is the mainstay of treatment in female genital TB. Sometimes the response could be slow or paradoxical with the development of abscess or sinuses and

sometimes there is partial response. In such situations, surgical treatment can be considered. In the present case, the patient had already taken ATT and biopsy with other investigations which were negative for TB. Bilateral TO masses were removed surgically along with the resection of fistulous tract and postsurgery after histopathology (HPE) patient was started on ATT.

There are no standard guidelines or protocols described for the management of tubocutaneous fistula; very few case reports are available in the literature.

Tubocutaneous fistula was first reported by Wittch et al. in 1982 which was secondary to tuberculous TO abscess.<sup>1</sup> In 2008, Choe et al. reported a similar case secondary to Crohn's disease.

Sheikh et al. in 2014 reported tuboenterocutaneous fistula secondary to surgical site wound infection of cesarean section; there was no evidence of TB or endometriosis. They did salpingectomy, resection, and excision of fistulous tract.<sup>3</sup> Nayani et al. in 2015 reported a case of tubocutaneous fistula secondary to pelvic inflammatory disease for which they did salpingectomy and excision of fistulous tract.<sup>4</sup> Krychowska-Cwikla et al. in 2015 reported a similar case secondary to peripartum hysterectomy and postoperative sepsis.<sup>5</sup> Lopes et al. in 2017 reported a tubocutaneous

fistula secondary to endometriosis that developed after cesarean section. They did resection and excision of entire fistulous tract with salpingectomy and hormone supplementation with desogestrol.<sup>6</sup>

Rajkumar et al. in 2018 reported laparoscopic excision of tubocutaneous fistula secondary to post-cesarean complication.<sup>7</sup>

All of these cases advocate excision of fistulous tract along with salpingectomy or salpingo-oophorectomy as definitive treatment with the treatment of the underline cause.

## CONCLUSION

Tubocutaneous fistulas can present as chronic discharging pelvic sinuses. Strong clinical suspicion after ruling out other causes can confirm the diagnosis. CT sonogram is the investigation of choice. Complete surgical resection and treating the underlying pathology can prevent long-term complications in these patients.

## ORCID

Anjum Ara  <https://orcid.org/0000-0003-1582-1576>

Sunayana Misra  <https://orcid.org/0000-0001-5091-4992>

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