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The One of its Kind- Non Healing Uterocutaneous Sinus- Managed Surgically and Medically

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Abstract: ***A uterocutaneous sinus is a very rare clinical entity. Here we report a case of uterocutaneous sinus tract following postpartum tubal ligation with recurrence after surgical management followed by spontaneous healing.***

Keywords: Uterocutaneous sinus, Tuberculosis, Tubectomy

1. Introduction

A fistula is an abnormal communication between two epithelial surfaces, they are usually lined by granulation tissue but chronic ones can get epithelialized [1].

A sinus tract is also a communication between two organs with one being the dead end; they can get infected and can become discharging. Uterocutaneous fistulas are rare and majority of them occur following uterine surgery; usually classical caseraen section. Literature is replete with articles on fistulas but no case of uterocutaneous sinus was found. We hereby report one such case which occurred following the minilap-sterilization and was managed surgically and medically.

2. Case Report

A 35 years old woman, P3L3 with previous three normal deliveries was referred with intermittent sero-purulent discharge through the minilaprotomy scar site following sterilization since four months. Patient had first noticed the discharge thirty-five days after tubectomy, which was done immediately in postpartum period. There was no history of puerperal sepsis or wound infection. There was no history of any mass or cyclical pain at the site of scar. Her menstrual history was normal. There was no history of tuberculosis in the past.

The general physical examination was normal. On abdominal examination infra-umbilical scar was present with purulent discharge from a cutaneous opening of 1.5 x 0.5cm in size. On vaginal examination, uterus was anteverted, normal size and mobile; with bilateral fornices free. On per speculum examination, cervix and vagina were healthy. Discharge fluid was sent for Gram staining, AFB staining and pyogenic culture which turned out to be negative. Due to scanty amount, it could not be sent for AFB culture.

The Ultrasound abdomen and pelvis was normal. On further evaluation with MRI revealed mild thickening of left rectus muscle with small subcutaneous fluid collection and fat stranding. Sinus tract was seen extending from collection till cutaneous surface (Fig 1). She underwent sinus-tract excision. Per-operatively sinus was seen extending from cutaneous surface to the anterior wall of rectus muscle, the tract was completely excised. The histopathology revealed nonspecific chronic inflammation.

On ninth postoperative day there was recurrence of discharge. The hysteroscopy was done to rule out uterocutaneous fistula, which was normal. On further evaluation with CT Sinogram, a sinus tract was seen extending from outer wall of uterine fundus till the abdominal skin (Fig. 2). She underwent redo surgery and complete excision of sinus tract was done, seven months after the first surgery. Biopsy of excised sinus tract again revealed non-specific inflammation. Culture reports were again negative for tubercular bacilli as well as for pyogenic and fungal elements.

On follow-up, patient again noticed sero-purulent discharge from the scar within a week of redo surgery. Considering performing a complete surgery and nonhealing nature of the sinus tract, she was empirically started on Anti-tubercular drug therapy. On anti-tuberculous drugs (cat I), discharge gradually started reducing over a period of ten days with complete cessation in 2 weeks. Patient has completed six month course of ATT with no recurrence of fistula.

3. Discussion

The reported cases of uterocutanous fistulas are associated with post-operative (casearen sections, uterine curettage, forceps delivery), infective (septic abortion) [2], actinomycosis, pelvic abscesses or rarely congenital malformations. The use of drains, incomplete closure of incisions and multiple abdominal surgeries are associated with post-operative uterocutaneous fistulas.

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In our patient there was history of mini-laprotomy for tubal ligation postnatally. As it was done on second day of delivery when the uterine size is 18 weeks, it is a possibility that while closing the rectus sheath a stitch accidently may have passed through the wall of uterus, which gradually was lined by granulation tissue and communication with skin was established. There is time delay between surgery and occurrence of uterine fistula, ranging between 2 months to 6 years. In our patient it was 35 days.

The diagnosis of uterine fistula can be made clinically if there is bloody discharge through the abdominal scar simultaneous with menstruation which is unfortunately not seen in our case. The investigations including MRI, CT scan, Hysteroscopy and Hysterosalpinography with Methlyene blue dye had been used in diagnosing the uterocutaneousfistula. In our case the ultrasound was normal. MRI also was not sensitive enough to detect whole length of sinus tract. Thus the CT sonogram/fistulogram is the modality of choice. On Hysteroscopy, in our case no opening was seen as it was a sinus tract.

The uterocutaneous fistula is a rare clinical entity and has variable etiology hence a standard treatment is not feasible in all cases. Previously it has been stated that all cases should be managed surgically including excision of the fistula tract or hysterectomy [3], preferring uterine preservation especially in young female. In recent reports, however, a combined surgical and medical treatment have been used and in yet another reported by Seyhan et al a patient was treated with gonadotropin-releasing hormone agonist (GnRH) alone [4].

Our is the rarest of rare case, as diagnosis of fistula may be easy, but this is the first case of sinus tract communicating between skin and myometrium at the level of fundus.

Tuberculosis is a crucial health problem in developing countries [5]. Although cutaneous mycobacterial infection is common, but surgical wound infection is rare [6, 7]. Secondary tuberculosis ; that arises in a previously sensitized host; may follow shortly, but presents more commonly decades after initial infection especially when host resistance is weakened [8] and, may result from exogenous re-infection, as occurs in geographical regions of high endemicity, or more commonly from reactivation of a latent primary focus with haematogenous spread to the site of the secondary infection or local reactivation at the secondary site [9]. Local reactivation may be precipitated by trauma or surgery (as seen in the above case), or any factor or insult that alters local tissue response; like injury, local vascular derangements, foreign body reactions and chronic inflammation [9]

The diagnostic dilemmas for tuberculosis have always existed, as in our case the gold standard test - AFB culture and biopsy of the sinus tract also turned out to be negative. The role of empirical ATT on the basis of clinical suspicion cannot be undermined in developing country like India where tuberculosis has been seen in varied forms involving all parts of the body. It is still considered an option in many cases of nonhealing wounds with good results, though the clinical experience with empirical ATT is passed on from the wisest to the youngest medical fraternity, the published data is difficult to find.

4. Conclusion

This case report will help the clinician in judging the best modality in delineating the sinus tract, the superiority of one over the other and will also alert the clinician about the elusive nature of tuberculosis, when the gold standard tests can also be inconclusive. Unfortunately, despite the efforts of Government of India in Revised National tuberculosis control program (RNTCP), we still lag behind in our diagnostic and therapeutic coverage with more than million cases being missed every year due to the silent nature of this disease, thus making place for empirical ATT.

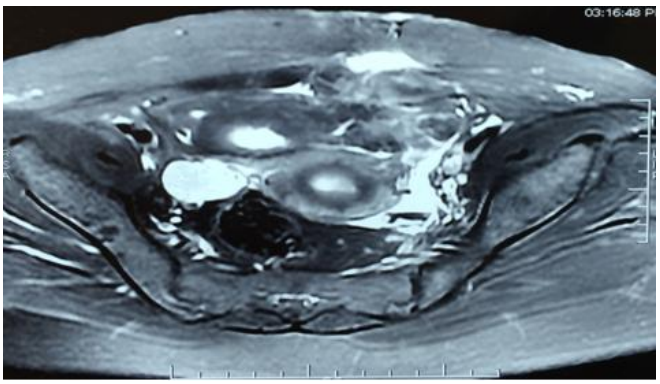


Figure 1: Axial view of MRI

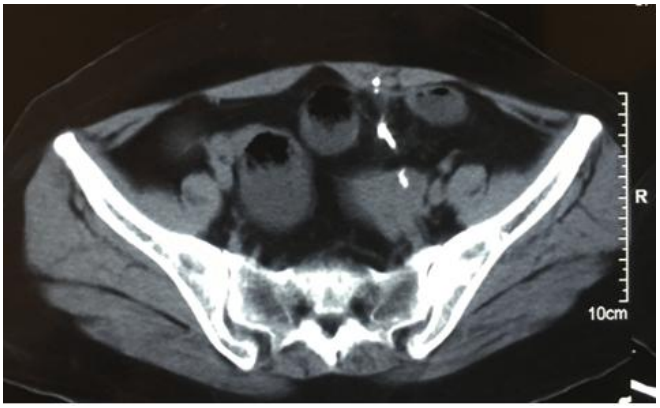


Figure 2: CT-fistulography

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