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Uterocutaneous fistula communicating with dermoid cyst: A surgical bizarre

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Abstract
Uterocutaneous fistula is a rare entity. Surgeons are familiar with urinary and gastro-intestinal fistula. It occurs most often after pelvic surgeries. Cases have been reported following caeseraen section and infection is considered as the most common cause for formation of the fistulous tract. It can be managed medically and surgically. Only few cases have been reported till date. We are reporting one such case of utero-cutaneous fistula with dermoid cyst following caesarean section. Our aim is to discuss the diagnostic and treatment modalities of utero-cutaneous fistula. Although this is rare but will aware clinician about the rarest complication of caesarean section and emphasize on prevention of infection.

Keywords: uterocutaneous fistula, dermoid cyst, fistulectomy

Introduction
A fistula is an abnormal communication between two epithelial lined surfaces. This communication or tract may be lined by granulation tissue but may become epithelialised in chronic cases [1]. Fistulas may be congenital or acquired. Gynaecologists are usually familiar with vesico-vaginal fistula, uretero-vaginal fistula, vesico-uterine fistula. We are reporting a very rare case of utero-cutaneous fistula with dermoid cyst following caesarean section. Our aim is to discuss causes, diagnostic modalities, treatment options of utero-cutaneous fistula, to remind the clinicians about the rare complication of caesarean section and to share our experience with the present case.

Case Report
25 years Para 3 Live 2 Death 1 presented at 11 months postpartum period with chief complaints of pus discharge from midpoint of caesarean section wound since 8 months. She went under caesarean section 11 months back at district hospital for transverse lie. She discharged from hospital after stitch removal. After two months she developed serous discharge from wound along with pain over the stitch line. Dressing was done repeatedly and received course of antibiotic for a longer period but due to no improvement, resuturing was done at local hospital. Unfortunately serous discharge continuously pours from wound. Sonography was done which shows haematoma of size 13*12.5cm and she was referred to the Index Medical College, Indore, MP. Detailed history and examination was done. There was no history of blood transfusion, fever, foul smelling lochia and associated disorders. On inspection of abdomen transverse scar with a pin point hole in the centre of scar in lower abdomen with foul smelling pus discharge with suprapubic bulge. On palpation induration and redness was present around the stitch line, which was tender and cystic non tender mobile suprapubic swelling was palpable. On speculum examination cervix, vagina healthy. On bimanual examination cystic mass of approximately 10*7 cm which was mobile nontender, feel separately from the uterus suggestive of ovarian origin and uterus was anteverted, normal in size, non tender with restricted mobility. She was admitted and was further evaluated by blood investigations which were within normal range. Sonography showed 11.4*9.7 cm left ovarian dermoid cyst thick walled sinus tract in lower abdominal wall in the region of caesarean scar, right paramedian in location extending deep into peritoneum in between urinary bladder and dermoid cyst. uterus was normal in size. For further confirmation contrast CT scan was done which showed 9.6*7.4*8.9 cm left dermoid cyst communicated with anterior abdominal wall sinus tract evident by fluid-fluid level in the lesion. this anterior abdominal tract was also extending to the serosal surface of anterior wall of uterus. Mountex test was negative. To see for communication of tract with bladder, women was catheterized, catheter was clamped and looked for urine drainage through wound which was not there (excluding bladder involvement). Laparotomy was planned with prior consent of hysterectomy. Before giving abdominal incision dye (methylene blue) was injected to trace the sinus tract through hole in the scar and then via vertical incision abdomen was opened. Dense adhesion was present, dermoid cyst seen anteriorly to uterus communicating with the sinus tract. adhesion was released and cystectomy was done. Cord like tubular structure suggestive of sinus tract extending from anterior surface of utreus to the cyst and adhere to bladder base. Infant feeding tube was passed through caesarean section wound which comes out through serosal surface of uterus. Fistulectomy was done followed by hysterectomy to prevent the recurrence. post operative period was uneventful. Histopathology report showed hyperplastic stratified squamous epithilium with inflammatory infiltrate and foreign body giant cells in the sinus tract and over uterine surface, lining of cyst suggests communication of sinus tract with cyst and uterus. And ovarian cyst shows the feature of mature teratoma.

Discussion
Utero-cutaneous fistula is rare entity with very few cases reported worldwide in literature till date. It results from post-partum and postoperative complications. Others causes are

history of multiple abdominal surgeries, use of drain, incomplete closure of uterine wound following caesarean section and myomectomy, intra-abdominal sepsis in previous scar, secondary abdominal pregnancy, migration of laminaria tent and intrauterine contraceptive devices, post-operative injuries and infections (tuberculosis) [2]. Most fistulae originate from trauma or some other type of inflammatory processes that disrupt the continuity of tissues involved [3]. In present case history of re-suturing was present which is suggestive of infective pathology. Non healing of superficial wound with persistent discharge from its deeper connection should raise a suspicion of development of fistula and presence of some local factor causing continuous infection. In present case dermoid cyst was also present which leads to adhesion and helps in the formation of fistula tract formation along with infection. Diagnosis of utero-cutaneous fistula is difficult to make clinically. Hence, demonstration of the fistulous tract is necessary for definitive diagnosis that can be done with a fistulogram, methylene blue injected trans-cervical, or use of contrast enhanced computed tomography (CT) scan as well as magnetic resonance imaging (MRI) or hysteron-salpingography (HSG) [4, 5]. Management of utero-cutaneous fistula is usually surgical, although spontaneous healing and medical management by gonadotropin releasing hormone agonist has been reported [6, 7]. Surgical management includes excision of fistula and hysterectomy. Decision of hysterectomy depends on the desire of future fertility. Though the women was young in present case but there was connection with the uterus and patient had completed her family so the decision of hysterectomy was taken along with dermoid cyst removal with the thought that if the source of fistula is present there will be high chance of recurrence, although cases have been reported with excision of fistula tract with the repair of uterine rent with postoperative period uneventful [8, 9].

Many cases of utero-cutaneous fistula have been reported till date but to our knowledge this was the first case of utero-cutaneous fistula communicating with dermoid cyst.

Fig 1: Transverse caesarean section scar with pin point hole in centre.

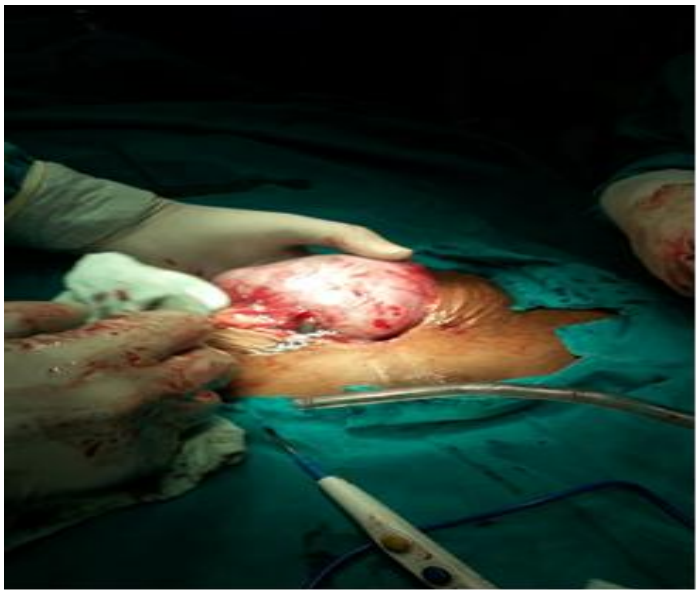

Fig 2: showing communication of fistula tract with dermoid cyst evident by methylene blue which was injected through skin.

Fig 3: After cystectomy fistula track visualised as a cord like structure with one end opening in uterus, adhere to posterior surface of uterus and open in skin.

Fig 4: showing fistula tract with infant feeding tube entering from skin to uterine end.

Fig 5: showing 1 fistulous tract, 2 uterus, 3 Dermoid cyst.

Conclusion

In recent years due to the rising caesarean section rates, it becomes important for the practitioners to be aware of such a rare complication and to give emphasis on prevention of infection.

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