Spontaneous utero-rectal fistula formation following reconstructive genital tract surgery: An interesting case report

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ABSTRACT

A genital fistula is an abnormal communication between uterus with either urinary tract or gastrointestinal tract. Although genitourinary fistula is a common entity, rectovaginal or even uterorectal fistula is relatively uncommon. The etiology of fistula is usually due to an obstetric cause in a developing country, whereas, it is gynecological in the developing country. Whether etiology is obstetrical or gynecological, this develops following days of the procedure. Here, we present the case of a 17-year-old girl, who underwent vaginoplasty for cervical atresia and develop uterorectal fistula one and a half year following the first surgery. She was finally managed with hysterectomy.

Keywords: Cervical atresia, Reconstructive genital tract surgery, Utero-rectal fistula.

A genital fistula is an abnormal communication between the genital tract and either urinary tract or gastrointestinal tract, usually rectum. The prevalence of these genital fistulae varies widely world wise with regards to etiology. As per the World Health Organization estimation, worldwide 50,000 to 100,000 women develop obstetric fistula each year [1]. In developed countries, these fistulas are uncommon and are most often result of gynecologic surgery. Its incidence varies from 0.02% to 10% percent and depends upon the type of hysterectomy i.e. lowest in vaginal hysterectomy and highest in radical hysterectomy [2]. The common variety of fistulae are vesicovaginal fistula and rectovaginal fistula. So, we are reporting an uncommon variety of genital fistula, uterorectal, in an adolescent following vaginoplasty surgery.

CASE REPORT

A 17-year-old girl presented to gynecology OPD with complaints of severe lower abdominal pain of one-day duration and primary amenorrhea two years back. The pain was continuous, confined to the hypogastric region, without radiation and relieving factor.

On general examination, the patient was of thin built. Her vitals were stable and secondary sex characters, as per Tanner's staging, were appropriate for chronological age, suggesting normal endogenous sex hormones. Her bowel and bladder disturbance was normal. There was no palpable mass abdomen. Her external genital was normal without any bulging of the hymen. Per-rectal examination demonstrates well-defined mass felt above and anteriorly, suggestive of the uterus. Other systemic examinations were normal.

Transabdominal ultrasound (USG) examination revealed cervical atresia with hematometra which was confirmed with magnetic resonance imaging (MRI). Kidney, ureter, and bladder were normal. Her blood investigations were as follows: Blood grouping and Rh typing-A positive, Haemoglobin-11.3gm%, blood urea-32.4 mgm%, creatinine - 0.7 mgm% and random blood sugar-137 mgm%.

Our Plan was to attempt establishing uterovaginal communication through abdominal-vaginal approach after detailed discussion and counseling of the girl and her parents. They were informed regarding the procedure, chance of success, future requirement of hysterectomy if the communication gets fibrosis.

Intraoperative findings were as follows:-10weeks cystic uterus of normal shape with a thick fibrosed band in place of cervix between uterus and vagina, bilateral normal ovaries. Vagina measured 7.5cm using uterine sound. The lower part of the uterus and fibrous band excised after pushing the bladder downward. The hematometra drained through the lower uterine opening after excision. The upper part of the anterior and posterior vaginal wall stitched to the anterior and posterior aspect of the lower part of uterus respectively. A Foley's catheter was placed inside the uterine cavity and brought out through the vagina.

Postoperatively, the intrauterine catheter was kept for 6 weeks. Following surgery, she had a regular menstrual cycle without dysmenorrhoea for three months. Subsequently, the amount of flow started reducing with the appearance of lower abdominal pain.

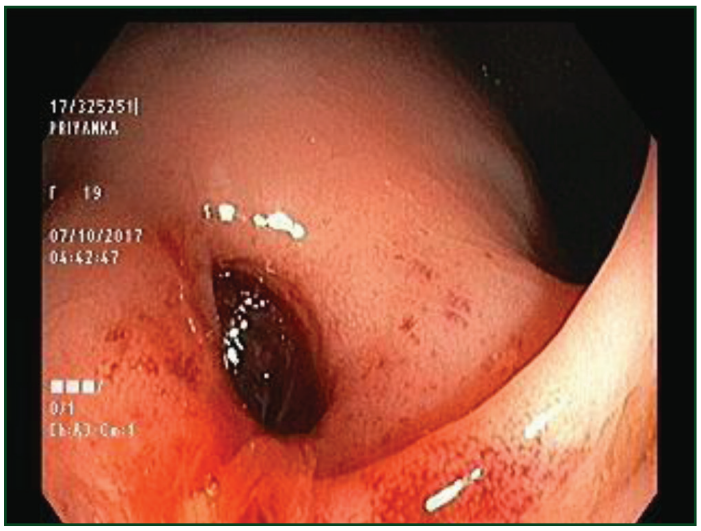

Figure 1: Sigmoidoscopy view of utero-rectal opening

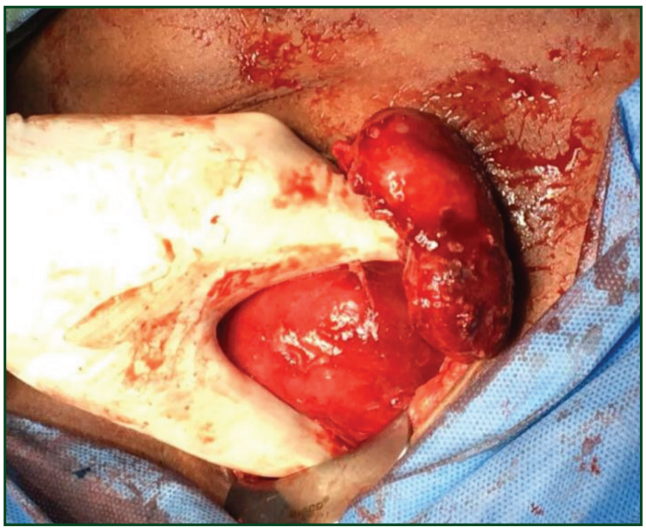

Figure 2: Distorted Left tube

Repeat ultrasound showed intrauterine collection. Examination under anesthesia done and hematometra drained vaginally with difficulty. The patient was put on Injection DMPA every two months. As she was symptom-free for a year, she stopped taking the injection of her own without any prior consultation.

After discontinuing the injection, she had amenorrhea for 6 months following which she had severe pain requiring admission for the second time. Her bowel and bladder habits were normal following the previous surgery prior to the second admission. On examination, her general condition was fair without any abdominal mass.USG revealed hematometra. So, occlusion of the previous uterovaginal surgical site was confirmed and MRI was planned to assess the length of that area to plan for further management. She was given injectable analgesic. MRI revealed hematometra. This time again it was planned to repeat the same procedure like that of first time mentioned and if failed, to do the same sitting hysterectomy. Accordingly, both the girl along with her parents counseled and informed consent obtained. Following 2 to 3 days of admission, she had one episode of bleeding per rectum and subsequent to it a fixed, tender mass of 10X14cm extending from left hypogastric region to the left iliac fossa appear, which was not there previously. We considered that mass as hematosalpinx. Sigmoidoscopy revealed an ulcer like area with minimal bleeding in the anterior rectal wall (Fig.1). Subsequently, bleeding per rectum stopped spontaneously and the patient was posted for the planned procedure mentioned before.

On laparotomy there was a tuboovarian abscess with omental adhesion of 10X14 cm on the left side, uterus was10 weeks, pouch of Douglas was obliterated with old clots, forming the pelvic hematocele with right side ovary buried in it. Left side tube was distorted (Fig. 2). The left side Tubo-ovarian abscess was removed (Fig. 3). The pus from this abscess sent for culture and sensitivity test. The uterine cavity opened through anterior midline incision to drain hematometra as described in the MRI. To our surprise, the uterine cavity was empty and the dilator passed to dilate the uterovaginal fibrosed part felt through the rectum. So, a diagnosis of uterorectal fistula was made and hysterectomy was performed along with repair of the rectal rent with diversion colostomy was performed with the help of the surgeon.

Following surgery, the patient was on piperacillin and tazobactam for five days. The pus was also sensitive to the same antibiotics. Postoperative period was uneventful. The suture was

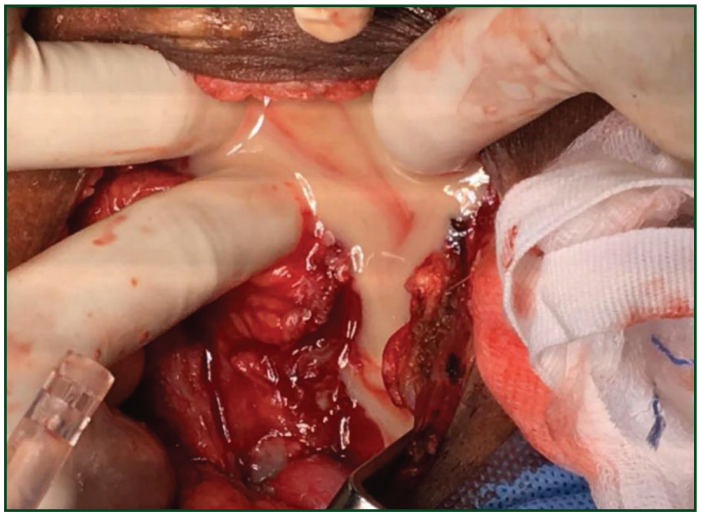

Figure 3: Left side tub-ovarian abscess (Ruptured)

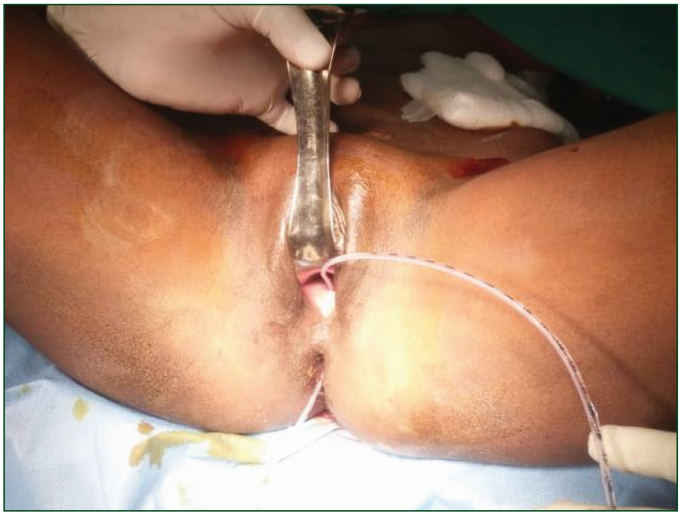

Figure 4: Rectovaginal Fistula

removed on the 8th postoperative day. The patient was trained to take care of the colostomy bag and discharged on the 10th postoperative day. On follow-up after three months, a small rectovaginal fistula was noticed (Fig. 4), which was closed in layers method followed by colostomy closure after three months of fistula repair ensuring intactness of the lower intestinal tract by barium enema. The patient is relieved of her cyclical pain abdomen, with normal passage of urine and stool till the date of reporting, under follow-up and is doing well.

DISCUSSION

Management of congenital cervical atresia is challenging. Treatment with hysterotomy and cervical canalization have been described as early as in 1900 [3]. However, hysterectomy was recommended in view of complications like re-stenosis of fibrous tissue, post-operative severe infection, or septicemia in occasional cases for this problem [4-6]. In recent years, to preserve the reproductive capability and relieve the menstrual symptoms conservative surgery is being attempted frequently in congenital cervical atresia. Canalization techniques are easier to perform, with a chance of re-stenosis and its complications in nearly 40–60%. Hence, the technique of uterovaginal anastomosis described by Deffarges *et al* [4]. Laparoscopically assisted uterovestibular anastomosis is also reported, but there is a paucity of data about post-procedure reproductive performance [7]. The success of uterovaginal anastomosis depends upon the size of the created ostium, the length of the new endocervical canal, the presence of vaginal mucosa adjacent to the end of the neo-ostium, and the duration of stenting [8].

The common complication following utero-cervical anastomosis in a case of cervical atresia with a functioning uterus is stenosis at the anastomotic site. Treatment for this is repeated canalization or if fails, hysterectomy to prevent retrograde menstruation resulting in pelvic endometriosis. Long term complication as uterorectal fistula is not reported in the literature. Usually, uterorectal fistulae are rare. Around 58 cases were reported over 200 years prior to 1955 [3]. Most common cause described in the literature occurred secondary to complications of diverticulitis in the elderly [9,10]. Other circumstances include sigmoid malignancy [11], radiotherapy and iatrogenic conditions such as insertion of intrauterine devices [12], endometrial curettage with uterine and bowel perforation [13], or obstetrical injury [14], following polymyomectomy [15]. Sometimes congenital fistula is possible with an anorectal anomaly, although the incidence is less than 1 % [16].

In the present case, the bleeding per rectum preoperatively was due to the uterorectal fistula. This occurred after one and a half year of vaginoplasty. As this complication is uncommon, rectal end opening of the fistulous tract was diagnosed as ulcer during sigmoidoscopy. It was inferred that haematometra described in the MRI opened into the rectum, as previous uterovaginal anastomosis was fibrosed completely, leading to the formation of a utero-rectal fistula. This fistula favored spontaneous drainage of hematometra, making the uterine cavity empty intraoperatively. However, ascending infection from rectum converted hematosalpinx into a tuboovarian abscess.

CONCLUSION

An enterouterine fistula is a rare entity and follows hysterectomy. It can occur following uterovaginal anastomosis in case of cervical atresia with a functioning uterus, especially, if the anastomosis gets fibrosed. This can occur even after years. So, it is essential to follow-up the patients, who undergo uterovaginal anastomosis, for outflow tract patency to avoid a complication like this.

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