

The path of least resistance: A case of cervical stenosis and uterocutaneous fistula☆



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ABSTRACT

Uterocutaneous fistula is exceedingly rare, and uniformly follows some type of operative procedure. In this case, a young woman underwent a cesarean delivery at an outlying clinic in rural Nigeria, following which she developed amenorrhea and cyclic pelvic pain. In attempts to resolve her condition, a second laparotomy was performed at the same medical center. She presented to us 2 weeks later, at which time an opening was present at the healing laparotomy scar, severe vaginal scarring and cervical stenosis were present, and marked hematometra was seen on ultrasound. Following a procedure to open her cervix, she began menstruating through a fistulous tract in her abdomen, which we subsequently excised and closed with no further problems for the patient. This case highlights the challenge in developing countries of surgical complications resulting from a lack of appropriately trained physicians in rural medical centers. We suggest that focus on excellent training of our young physicians and the creation of incentives to place and keep fully qualified physicians in such hospitals will improve this situation.

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1. Introduction

While vesicovaginal and rectovaginal fistulas remain common in developing countries, uterocutaneous fistula is very rare. The condition was first described in medical literature by Thomas Case in 1958 [1]. Since that time, only a small number of publications have addressed uterocutaneous fistula. Of these, all are single case reports in which the fistula followed an operative procedure. We present here an unusual case of uterocutaneous fistula.

2. Case

A 26 year old G1P1 presented complaining of amenorrhea following cesarean section 1.5 years ago performed at a private clinic near her village for suspected fetal distress. She was delivered of a severely asphyxiated infant who subsequently died. Amenorrhea followed delivery; however the patient experienced cyclic pelvic pain one week every month. Before coming to us she returned to the original clinic with these complaints and underwent laparotomy, though exact procedures performed were unknown to the patient and medical records are unavailable.

Two weeks postoperatively, she presented to our hospital where examination revealed a tender 16-week-sized uterus and a healing midline scar with a 2 cm opening which was slowly oozing blood near the superior edge of the incision. No guarding, rebound or rigidity was present. The cervix was palpable on vaginal examination, but extensive scarring in the proximal vagina obstructed visualization with a speculum. Ultrasound showed a bulky uterus with a large collection of blood within the endometrial cavity. The patient was scheduled for cervical dilatation under anesthesia.

Even with relaxation, visualization of the cervix was difficult due to the vaginal scarring, but with good retraction the external os was located and the cervix cannulated with Hegar dilators using ultrasound guidance to prevent misplacement or perforation. Upon reaching the endometrial cavity, copious brown blood flowed through the cervix, further confirming correct location. A 14-French Foley catheter was placed in the uterus for drainage and maintenance of cervical patency. The catheter was removed on postoperative day 12, by which time the patient was free from pain or bleeding.

Three weeks later she returned complaining of bleeding through her laparotomy wound. The previously noted bleeding at the healing abdominal wound had stopped following cervical dilatation until 3 days prior, when the patient began having pelvic cramping and bright red bleeding through a single point in the scar. A nine millimeter circular opening with granulated edges was noted in the laparotomy scar, though it was not presently discharging (Fig. 1). Ultrasound showed an empty uterus, adherent to the abdominal wall. Vaginal examination confirmed the cervix was still patent. A presumptive

☆ Informed consent: The patient whose case is described in this report has given written consent for the case to be used for publication, including the use of photographs used in the attached Figures. A copy of the written consent is available for review by the Editor upon request.

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Fig. 1. The granulated fistula evident in the now-healed laparotomy scar.

diagnosis of uterocutaneous fistula was made and the patient underwent exploratory laparotomy. A fistulous tract was identified prior to incision using a lacrimal probe, which was left in place to aid dissection (Fig. 2). The uterus was densely adherent to the anterior abdomen, requiring extensive adhesiolysis. The fistulous tract, inserting into the upper uterine segment, was excised from skin to endometrial cavity before repairing the uterine defect in layers. A portion of omentum was mobilized and tacked over the uterine repair as a barrier between the uterus and abdominal wall to reduce risk of recurrence.

3. Discussion

Among published cases of uterocutaneous fistula, all instances follow an operative procedure. Etiologies include abortion [2,3], post-cesarean infection [4], myomectomy [1], dilation and curettage [5], and vaginal agenesis [6]. To our knowledge, no case has been reported of uterocutaneous fistula following laparotomy for hematometra following cesarean section.

This case highlights two important points. First, successful management of uterocutaneous fistula requires first ensuring a patent outflow tract. It has been documented that attempts to rectify hematometra without also creating or confirming a route for egress of menses is liable to fail [3,6]. It was fortuitous for us that our first encounter demonstrated hematometra and outlet obstruction without obvious fistula. This shifted our focus toward opening and maintaining the cervical canal, so when the patient returned with a more obvious uterocutaneous fistula, repair could be pursued knowing that subsequent menstrual bleeding could exit through the patent cervix. Menses followed the fistulous tract in this case despite an open cervix simply due to the lesser resistance of an open fistula versus the muscular sphincteric-type path of the cervical canal.

Secondly, we suspect that this case also highlights a growing problem being faced in medical care in resource-poor areas of the developing world — complications related to suboptimally trained individuals. In the past years it was correctly identified that many people faced disease and death due primarily to inadequate access to medical care. This was particularly true in rural areas. Therefore efforts were undertaken, many with great success, to establish medical centers in underserved areas. However staffing of these centers with adequately trained providers has proven frequently difficult or impossible. As such, some are being run by undertrained physicians while others are staffed solely by non-

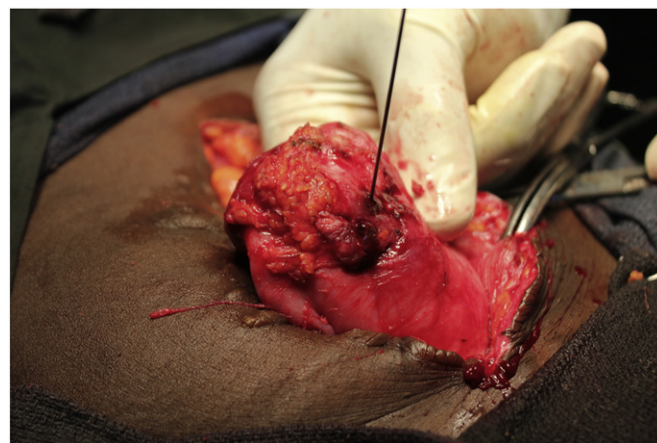


Fig. 2. A lacrimal probe placed through the fistula at the beginning of the procedure to aid in dissection is seen here entering the upper segment of the uterus. (All pictures are the primary author's personal photographs).

physician personnel. Risks increase for developing surgically-induced complications such as post-cesarean fistulas when the providers delivering care are undertrained, a problem that has also been identified by Onsrud in the Democratic Republic of Congo [7].

The locality from which this patient presented has previously referred patients to our hospital with complications indicative of limited surgical knowledge and technique. In this case history, examination, and surgical findings led us to suspect that during our patient's cesarean she likely had an extension of the hysterotomy into the cervix, and through efforts to stop the bleeding and repair it, the cervix and proximal vagina were actually sewn closed. In the subsequent surgery, laparotomy may have been performed with intentional entry into the uterus to relieve the hematometra. In the face of cervical obstruction, this likely led to the uterocutaneous fistula formation.

It is our belief that focusing on educating our young physicians and creating incentives to place fully trained physicians in underserved areas will improve outcomes and may prevent cases such as this.

In follow up, this patient healed without complication or recurrence. She has resumed normal monthly menstruation per vagina.

Conflict of Interest Statement

The authors declare that they have no conflict of interest.

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