CASE REPORT

Uterocutaneous fistula following B-Lynch suture for primary postpartum haemorrhage

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SUMMARY

A 30-year-old woman, who had undergone emergency lower segment caesarean section (LSCS) for failed induction 2 months back, presented with a fistulous opening along with discharge from her previous incision scar. She had developed a massive primary postpartum haemorrhage at the time of LSCS 2 months back, which was managed with B-Lynch suture and vessel ligation. Fistulogram revealed a connection between the uterus and the skin. The diagnosis was confirmed by a contrast-enhanced CT scan. Patient was subjected to laparotomy. She was found to have an extensive necrosis of the anterior uterine wall. Total abdominal hysterectomy was done to avoid the risk of sepsis and haemorrhage. Postoperative period was uneventful. Histopathological examination confirmed the necrosis of the uterine wall. This case describes an extremely rare occurrence of uterocutaneous fistula as a result of uterine infarction following the application of B-Lynch suture for primary postpartum haemorrhage.

BACKGROUND

A fistula is an abnormal connection between two epithelialised surfaces. Most uterine fistulae occur between the uterus and either the bladder (uterovesical) or the colon (uterocolonic). They are usually caused by injury or surgery, but they can also result from infection or inflammation.¹ A uterocutaneous fistula, that is, communication between the uterus and the skin is an extremely uncommon condition, described only in a few case reports previously. Because of its rare presentation, the approach to management is not clearly defined.² B-Lynch suture is a form of compression suture

Figure 2 Ultrasound showing communication (black arrow) between uterus (horizontal white arrow) and subcutaneous planes (vertical white arrow).

used to mechanically compress an atonic uterus in the face of severe postpartum haemorrhage. It is generally considered a safe procedure with minimal complications.³ We report a rare case in which the application of B-Lynch suture for postpartum haemorrhage resulted in extensive uterine infarction and necrosis, ultimately leading to formation of a uterocutaneous fistula.

CASE PRESENTATION

A 30-year-old woman, para1 live1, presented with complaints of infected discharge through her previous Pfannenstiel incision. She had undergone a lower segment caesarean section (LSCS) 2 months ago for a failed induction at our institute. The caesarean was complicated by uterine relaxation resulting in persisting postpartum haemorrhage. The latter did not respond to systemic treatment with uterotonics; therefore, decision for surgical management was taken. We applied a pair of vertical brace sutures (B-Lynch) using vicryl no. 1, followed by bilateral uterine and ovarian artery ligation to control postpartum haemorrhage. Antibiotic cover was started immediately and patient received one unit of blood transfusion. The immediate postnatal recovery was apparently uncomplicated allowing discharge on the fifth postoperative day. One month later, she noticed a painful nodule at the suture line associated with pus-like discharge from it. She received medical treatment with antibiotics along with daily dressings at a nearby local hospital. Later on, she developed superficial 1 cm gap at the centre of stitch line for which resuturing was done. However, the discharge continued and she again presented to us. In addition, she had irregular bleeding per vaginum on and off for the past 1 month.

Figure 1 Picture showing fistulous opening in the skin (black arrow).

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Figure 3 Fistulogram showing fistulous tract (horizontal arrow) of dimensions 5.4 cm in length and 8 mm in thickness between skin and endometrial cavity (vertical arrow).

At presentation, her vitals were stable and general physical examination was unremarkable. On abdominal examination, a pinpoint opening was seen in the scar of LSCS, and granulation tissue was seen through that opening (figure 1). Per speculum examination was normal and per vaginal examination revealed a normal sized, anteverted uterus with restricted mobility.

INVESTIGATIONS

A transabdominal ultrasound was done, which showed an area of very unusual uterine echotexture with no visible endometrium. A suspicious discontinuity in the anterior myometrium in the fundic region was also noted which appeared to be communicating to the skin (figure 2). No drainable collection was noted in the pelvis. Bilateral ovaries were normal. Fistulogram revealed a 5.4 cm tract with thickness of 8 mm communicating with the endometrial cavity through the fundus of uterus (figure 3). Diagnosis of uterocutaneous fistula was confirmed by a contrast-enhanced CT (CECT) scan.

Figure 4 Intraoperative image showing communication of the fistulous tract with the endometrial cavity (black arrow).

Figure 5 Intraoperative image showing necrosis of the anterior uterine wall making endometrial cavity exposed (black arrow).

TREATMENT

Patient was subjected to laparotomy. An incision along the previous Pfannenstiel scar was given, and a probe was inserted into the fistulous tract to ensure tissue dissection around the probe. The tract was partially excised to enter the peritoneal cavity. There were dense adhesions between the anterior abdominal wall and the anterior wall of uterus, sigmoid colon and the omentum.

Adhesiolysis was done and the distal end of the fistulous tract was seen communicating with the uterine cavity through the fundus (figure 4). The anterior wall of uterus had been totally replaced by the granulation tissue (figure 5), so there was no scope for repair. Also the risk of sepsis was very high. Therefore, we decided to proceed with hysterectomy (figure 6), which was successfully performed using standard techniques.

OUTCOME AND FOLLOW-UP

The patient had a good postoperative recovery and was discharged on the seventh postoperative day. Histological examination confirmed the extensive necrosis of the uterus.

DISCUSSION

B-Lynch is recommended as a life-preserving and fertility-preserving technique for postpartum haemorrhage when medical and non-medical interventions fail. These compression sutures compress the uterine vascular sinuses mechanically without occluding either the uterine arteries or uterine cavity.³ Normally, there is significantly increased postnatal vascularity of the uterus with extensive anastomotic vessels within the pelvis, which make the uterus highly resistant to ischaemia and necrosis. Ideally, application of these compression sutures should not interfere with the uterine vascularity. However, a case of total uterine necrosis post B-Lynch suture has been reported in the past, which led to hysterectomy.⁴ Ochoa *et al* have also reported infectious complications like pyometra after application of B-Lynch suture.⁵ Erosion of the suture into the lower segment of the uterus in the postpartum period, resulting in anterior wall defects, has also been reported.⁶ The use of non-absorbable sutures increases the risk of potential complications. Monofilament suture with an absorption time of 90–120 days can decrease these complications.

Our patient suffered an extensive uterine infarction following the application of B-Lynch suture for primary postpartum haemorrhage. This resulted in the formation of a uterocutaneous fistula, which is a very rare condition. As per the existing

Figure 6 Specimen of uterus showing extensive necrosis of the anterior uterine wall up to the level of internal orifice (horizontal arrow). Note is made of fistulous tract opening in the fundus (vertical arrow).

Learning points

- ▶ Although, the B-Lynch suture is very successful in preventing the need for hysterectomy during intractable postpartum haemorrhage, the effects of B-Lynch suture on future fertility potential require further investigation.
- ▶ The risk of potential complications tends to be higher when non-absorbable sutures are used. Monofilament sutures with an absorption time of 90–120 days significantly decrease the complications related to B-Lynch suture.
- ▶ Incorrect application of B-Lynch suture can interrupt the uterine arterial blood flow to the myometrium and can rarely lead to uterine necrosis.
- ▶ This is the first case in literature, reporting the occurrence of uterocutaneous fistula secondary to uterine necrosis resulting from application of B-Lynch suture.
- ▶ In case of clinical suspicion of such a complication, hysteroscopic examination can be done to see the fistulous opening in the uterine cavity under direct vision.

literature, uterocutaneous fistulae have been reported after septic abortion,⁷ pelvic abscess,⁸ secondary abdominal pregnancy,⁹ uterovaginal malformation,¹⁰ migration of an intra-uterine device,¹¹ curettage, difficult vaginal delivery and the use of forceps.¹ However, none have reported formation of uterocutaneous fistula after ischaemic uterine necrosis as a result of B-Lynch suture application.

Fistulography is extremely useful for diagnosis of a uterocutaneous fistula as it establishes the connection between the skin and the uterus.^{2,7} Hysterosalpingography with methylene blue injection via the cervix can be used for diagnosis if the skin

opening is very small. Thubert *et al*² recommended the use of hysteroscopy for the detection of fistula opening in the uterine cavity under direct vision. CECT scan and MRI can also be used.^{1,2} We used both fistulography and CECT scan to establish the diagnosis in our case.

Because the complication is rare, there is no set protocol for optimal management. In most of the previously reported cases of uterocutaneous fistula, surgical management in the form of hysterectomy and excision of the fistulous tract was performed.² Recently, few authors have advocated the use of gonadotropin-releasing hormone (GnRH) agonist for the reduction in the risk of hysterectomy. GnRH agonist has been shown to be effective by causing fibrosis and closure of the fistulous tract through unclear mechanisms.² However, hysterectomy was mandatory in our case owing to the high risk of sepsis. This case thus highlights a rare complication of compression B-Lynch suture, leading to formation of uterocutaneous fistula and eventually resulting in hysterectomy in a young female.

Contributors MT, SSR and AJ were involved in patient management and made the diagnosis. MT wrote the manuscript. KM performed the literature search and corrected the manuscript. All authors read and approved the final version of the manuscript.

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