

BMJ Case Reports

Successful pregnancy and delivery following surgical treatment of postmyomectomy uterocutaneous fistula

Journal:	<i>BMJ Case Reports</i>
Manuscript ID	bcr-2019-231594.R1
Manuscript Type:	Rare disease
Date Submitted by the Author:	n/a
Complete List of Authors:	Yesiladali, Mert; Yeditepe Universitesi Hastanesi, Saridogan, Erdinc; Zekai Tahir Burak Women's Health Training and Research Hospital, Department of Gynaecology Saridogan, Ertan; University College London, Institute for Women's Health
Keywords:	Obstetrics, gynaecology and fertility < Drugs and medicines, Reproductive medicine < Obstetrics and gynaecology

SCHOLARONE™
Manuscripts

BMJ Case Reports

Successful pregnancy and delivery following surgical treatment of postmyomectomy uterocutaneous fistula

SUMMARY

Uterocutaneous fistula is an extremely rare clinical condition which may be caused by postoperative or postpartum complications, such as infection or inflammation. Although fibroids and myomectomy are common clinical entities among reproductive age women, there are very few postmyomectomy uterocutaneous fistula cases in the literature. This article presents the first reported case of a successful pregnancy and live birth following treatment of a postmyomectomy uterocutaneous fistula. After laparoscopic adhesiolysis, a mini laparotomy was performed to excise the fistula tract completely from both the abdominal wall and uterus. The uterine wall defect was repaired in multiple layers. The patient had a good recovery after surgery and the uterocutaneous fistula resolved completely. Due to obliteration of both tubal ostia the patient was referred for IVF treatment. She conceived after the third frozen embryo transfer procedure and gave birth to a 4,4 kg baby at full term by caesarean section.

BACKGROUND

Uterocutaneous fistula is an extremely rare clinical condition which may be caused by postoperative or postpartum complications, such as infection or inflammation[1]. Although fibroids and myomectomy are common clinical entities among reproductive age women, there are very few postmyomectomy uterocutaneous fistula cases in the literature. This article presents the first reported case of a successful pregnancy and live birth following surgical treatment of a postmyomectomy uterocutaneous fistula.

CASE PRESENTATION

A 31 year-old nulliparous woman was referred to our clinic in 2016 with the diagnosis of uterocutaneous fistula. She was first diagnosed with a 4 cm submucosal uterine fibroid in 2010. The following year her fibroid was measured 7 cm and she subsequently underwent two hysteroscopic myomectomy procedures in which the fibroid was partly resected. In 2014, she was diagnosed with a 15 cm submucosal fibroid and underwent an open myomectomy operation. The procedure was complicated with post-operative infection and she was treated with intravenous antibiotics. Three weeks later she

started experiencing some discharge from her abdominal wound and this persisted over the following few months. On physical examination a 1 cm pocket in the middle of her low transverse abdominal incision was reported to be clearly visible. She was eventually diagnosed with uterocutaneous fistula three to four months after the operation.

INVESTIGATIONS

Magnetic resonance imaging (MRI) examination confirmed diagnosis of fistula between the uterine cavity and the abdominal wall (**Figure**). She also had a hysterosalpingography (HSG) which showed the contrast passage from the uterine cavity and the skin. There was no evidence of fill or spill of the fallopian tubes. Her last ultrasonography (USG) prior to surgery showed the fundal defect extending from the uterine cavity to the parietal peritoneum and a further communication with the anterior abdominal wall. She was given GnRH injections for few months to achieve amenorrhea with the hope that the fistula would seal off. Unfortunately, monthly blood stained drainage restarted when her periods returned after this treatment, thus a surgery was planned to repair the defect.

TREATMENT

Initial hysteroscopy showed uterine cavity was mostly regular with no obvious visible defect but tubal ostia were not visible. Laparoscopy showed omental adhesions to the anterior abdominal wall in lower abdomen and below these adhesions, uterus appeared densely adherent to the anterior abdominal wall. The uterus was separated from the anterior abdominal wall at laparoscopy and the suprapubic incision was extended to 4 cm using the existing low transverse incision scar. Abdominal wall part of the uterocutaneous fistula was excised. Uterus delivered through the incision and the fundal defect identified using methylene blue. Fundal fibrotic tissue around fistula was excised to the level of uterine cavity. Endometrium was repaired with No:2-0 polyglecaprone 25 and myometrium was repaired in 2 layers with no:1 polyglactin 910 sutures.

OUTCOME AND FOLLOW-UP

The patient had a good recovery after surgery and the uterocutaneous fistula resolved completely. Due to obliteration of both tubal ostia, she was referred for IVF treatment. A repeat hysteroscopy was performed prior to IVF and complete healing of the cavity without adhesion formation was confirmed. She conceived after the third frozen embryo transfer procedure and gave birth to a 4,4 kg baby at 38 weeks by caesarean section. The pregnancy was entirely problem free.

DISCUSSION

The most commonly seen uterine fistulas are uterovesical and uterocolonic[2]. Uterocutaneous fistulas are rarely seen and are defined as an abnormal connection between the uterine cavity and the skin. The typical sign of uterocutaneous fistulas is bloody discharge from skin occurring during menstruation[3]. For definitive diagnosis, the fistula can be shown by imaging methods which may be ultrasonography (USG), computerized tomography (CT), magnetic resonance imaging (MRI) and hysterosalpingography (HSG); or more simply by methylene blue test. In addition to these, a fistulography may also be performed; in which a contrast medium is injected into the opening, thus the fistula and uterine cavity may be demonstrated by direct radiography[4]. There is no gold standard test but HSG and MRI are both reliable tests to confirm diagnosis, in the presence of typical symptoms. In our case, the diagnosis was confirmed by MRI, USG and HSG, which all demonstrated the pathological passage between the uterine cavity and skin.

Possible etiologies of uterocutaneous fistula include multiple abdominal surgeries, incomplete closure of uterine wound during myomectomy or caesarean section, use of drains and postoperative complications such as infection and inflammation. Septic abortions, migration of intrauterine device, endometriosis and retained placenta after abdominal pregnancy have also been mentioned in the literature as more extreme etiologies[1,5,6]. The present case underwent two hysteroscopic myomectomy procedures and an open myomectomy surgery, which was complicated by postoperative infection. It is difficult to know if the hysteroscopic myomectomy procedures contributed to the fistula formation, but postoperative infection after open myomectomy is likely to be the main cause of fistula formation in this patient.

Since uterocutaneous fistula is an extremely rare condition, there is no standard approach for its management. Review of the literature revealed only 2 successful medical treatment cases[3,7]. Both studies used Gonadotropin releasing hormone (GnRH) analogues as medical treatment option and had successful results. As uterocutaneous fistula is thought to have a similar lining with endometrium, it is assumed that temporary menopausal state would cause atrophy of the fistula tissue and cessation of menstruation would contribute to the closure of the tract. However, medical treatment of uterocutaneous fistula may be insufficient in other cases, and has several limitations. First of all it is a long treatment process with an uncertain success rate. Secondly, the uterocutaneous fistulas, unlike utero-vesical fistula, have usually a larger size of tract which may be another obstacle for medical treatment.

Surgical management option appears to be the option of choice in the remaining cases reported in the literature, as a shorter treatment process with more accurate results. Additionally, if the patient wishes to become pregnant as in our case, surgical management should be considered to ensure integrity of the uterine wall. However, risks of surgery should be discussed with patient thoroughly and a decision between the medical and surgical treatment options should be made together with the patient.

As surgical management, mini laparotomy combined with laparoscopy, if available, appears to be the most rational option. It is reasonable to start with laparoscopy, since uterus and possibly other intraabdominal organs are expected to be densely adherent to anterior abdominal wall. In our case, after laparoscopic adhesiolysis, a mini laparotomy was performed to excise the fistula tract completely from both abdominal wall and uterus.

In conclusion, although a very rare entity, uterocutaneous fistula can be difficult to manage. Prevention could be possible with better uterine closure techniques, prevention of postoperative infections and avoiding multiple surgeries at the same site. When encountered, treatment modality should be discussed with patient considering fertility preservation desire, and successful pregnancy could be achieved with appropriate surgical treatment.

LEARNING POINTS/TAKE HOME MESSAGES

- Since there are no established guidelines for management of rare cases like uterocutaneous fistula, reporting and reading rare situations and their treatment outcomes may be crucial when these cases are encountered.
- When postsurgery discharge from skin incision is seen, uterocutaneous fistula should be considered among differential diagnoses.
- Multiple abdominal surgeries, incomplete closure of uterine wound during myomectomy or caesarean section, use of drains and postoperative complications such as infection and inflammation are some of the known risk factors for fistulas.
- Uterocutaneous fistulas can be treated by appropriate surgery.
- Successful pregnancies and live birth after complete recovery are possible.

REFERENCES

- 1) Dragoumis K, Mikos T, Zafrakas M, Assimakopoulos E, Stamatopoulos P, Bontis J. Endometriotic uterocutaneous fistula after cesarean section. *Gynecol Obstet Invest* 2004;57:90-2.
- 2) Eldem G, Turkbey B, Balas S, Akpinor E. MDCT diagnosis of uterocutaneous fistula. *Eur J Radiol Extra* 2008;67:e129.
- 3) Seyhan A, Ata B, Sidal B, Urman B. Medical treatment of uterocutaneous fistula with gonadotropin-releasing hormone agonist administration. *Obstet Gynecol.* 2008;111:526-8.
- 4) Gupta SK, Shukla VK, Varma DN, et al. Uterocutaneous fistula. *Postgrad Med J.* 1993;69:822–823.
- 5) Sonmezer M, Sahincioglu O, Cetinkaya E, Yazici F. Uterocutaneous fistula after surgical treatment

- of an incomplete abortion: Methylene blue test to verify the diagnosis. Arch Gynecol Obstet 2009;279:225.
- 6) Malhotra D, Vasishta K, Srinivasan R, Singh G. Tuberculous uteroenterocutaneous fistula: A rare post-cesarean complication. Aust NZ J Obstet Gynaecol 1995;35:342.
 - 7) Yadav P, Gupta S, Singh P, Tripathi S. Successful medical management of uterocutaneous fistula. Int J Gynaecol Obstet 2014;124:263-4

FIGURE/VIDEO CAPTIONS

Figure: T2 sagittal MRI image demonstrating the communication between the fundal myometrial defect and the skin incision

INTELLECTUAL PROPERTY RIGHTS ASSIGNMENT OR LICENCE STATEMENT

I, Ertan Saridogan, the Author has the right to grant and does grant on behalf of all authors, an exclusive licence and/or a non-exclusive licence for contributions from authors who are: i) UK Crown employees; ii) where BMJ has agreed a CC-BY licence shall apply, and/or iii) in accordance with the relevant stated licence terms for US Federal Government Employees acting in the course of the their employment, on a worldwide basis to the BMJ Publishing Group Ltd ("BMJ") and its licensees, to permit this Work (as defined in the below licence), if accepted, to be published in BMJ Case Reports and any other BMJ products and to exploit all rights, as set out in our licence [author licence](#).

Date: 23.10.2019

BMJ Case Reports

Successful pregnancy and delivery following surgical treatment of postmyomectomy uterocutaneous fistula

SUMMARY

Uterocutaneous fistula is an extremely rare clinical condition which may be caused by postoperative or postpartum complications, such as infection or inflammation. Although fibroids and myomectomy are common clinical entities among reproductive age women, there are very few postmyomectomy uterocutaneous fistula cases in the literature. This article presents the first reported case of a successful pregnancy and live birth following treatment of a postmyomectomy uterocutaneous fistula. After laparoscopic adhesiolysis, a mini laparotomy was performed to excise the fistula tract completely from both the abdominal wall and uterus. The uterine wall defect was repaired in multiple layers. The patient had a good recovery after surgery and the uterocutaneous fistula resolved completely. Due to obliteration of both tubal ostia the patient was referred for IVF treatment. She conceived after the third frozen embryo transfer procedure and gave birth to a 4,4 kg baby at full term by caesarean section.

BACKGROUND

Uterocutaneous fistula is an extremely rare clinical condition which may be caused by postoperative or postpartum complications, such as infection or inflammation[1]. Although fibroids and myomectomy are common clinical entities among reproductive age women, there are very few postmyomectomy uterocutaneous fistula cases in the literature. This article presents the first reported case of a successful pregnancy and live birth following surgical treatment of a postmyomectomy uterocutaneous fistula.

CASE PRESENTATION

A 31 year-old nulliparous woman was referred to our clinic in 2016 with the diagnosis of uterocutaneous fistula. She was first diagnosed with a 4 cm submucosal uterine fibroid in 2010. The following year her fibroid was measured 7 cm and she subsequently underwent two hysteroscopic myomectomy procedures in which the fibroid was partly resected. In 2014, she was diagnosed with a 15 cm submucosal fibroid and underwent an open myomectomy operation. The procedure was complicated with post-operative infection and she was treated with intravenous antibiotics. Three weeks later she

started experiencing some discharge from her abdominal wound and this persisted over the following few months. On physical examination a 1 cm pocket in the middle of her low transverse abdominal incision was reported to be clearly visible. She was eventually diagnosed with uterocutaneous fistula three to four months after the operation.

INVESTIGATIONS

Magnetic resonance imaging (MRI) examination confirmed diagnosis of fistula between the uterine cavity and the abdominal wall (**Figure**). She also had a hysterosalpingography (HSG) which showed the contrast passage from the uterine cavity and the skin. There was no evidence of fill or spill of the fallopian tubes. Her last ultrasonography (USG) prior to surgery showed the fundal defect extending from the uterine cavity to the parietal peritoneum and a further communication with the anterior abdominal wall. She was given GnRH injections for few months to achieve amenorrhea with the hope that the fistula would seal off. Unfortunately, monthly blood stained drainage restarted when her periods returned after this treatment, thus a surgery was planned to repair the defect.

TREATMENT

Initial hysteroscopy showed uterine cavity was mostly regular with no obvious visible defect but tubal ostia were not visible. Laparoscopy showed omental adhesions to the anterior abdominal wall in lower abdomen and below these adhesions, uterus appeared densely adherent to the anterior abdominal wall. The uterus was separated from the anterior abdominal wall at laparoscopy and the suprapubic incision was extended to 4 cm using the existing low transverse incision scar. Abdominal wall part of the uterocutaneous fistula was excised. Uterus delivered through the incision and the fundal defect identified using methylene blue. Fundal fibrotic tissue around fistula was excised to the level of uterine cavity. Endometrium was repaired with No:2-0 polyglecaprone 25 and myometrium was repaired in 2 layers with no:1 polyglactin 910 sutures.

OUTCOME AND FOLLOW-UP

The patient had a good recovery after surgery and the uterocutaneous fistula resolved completely. Due to obliteration of both tubal ostia, she was referred for IVF treatment. A repeat hysteroscopy was performed prior to IVF and complete healing of the cavity without adhesion formation was confirmed. She conceived after the third frozen embryo transfer procedure and gave birth to a 4,4 kg baby at **38 weeks** by caesarean section. **The pregnancy was entirely problem free.**

DISCUSSION

The most commonly seen uterine fistulas are uterovesical and uterocolonic[2]. Uterocutaneous fistulas are rarely seen and are defined as an abnormal connection between the uterine cavity and the skin. The typical sign of uterocutaneous fistulas is bloody discharge from skin occurring during menstruation[3]. For definitive diagnosis, the fistula can be shown by imaging methods which may be ultrasonography (USG), computerized tomography (CT), magnetic resonance imaging (MRI) and hysterosalpingography (HSG); or more simply by methylene blue test. In addition to these, a fistulography may also be performed; in which a contrast medium is injected into the opening, thus the fistula and uterine cavity may be demonstrated by direct radiography[4]. **There is no gold standard test but HSG and MRI are both reliable tests to confirm diagnosis, in the presence of typical symptoms.** In our case, the diagnosis was confirmed by MRI, USG and HSG, which all demonstrated the pathological passage between the uterine cavity and skin.

Possible etiologies of uterocutaneous fistula include multiple abdominal surgeries, incomplete closure of uterine wound during myomectomy or caesarean section, use of drains and postoperative complications such as infection and inflammation. Septic abortions, migration of intrauterine device, endometriosis and retained placenta after abdominal pregnancy have also been mentioned in the literature as more extreme etiologies[1,5,6]. The present case underwent two hysteroscopic myomectomy procedures and an open myomectomy surgery, which was complicated by postoperative infection. It is difficult to know if the hysteroscopic myomectomy procedures contributed to the fistula formation, but postoperative infection after open myomectomy is likely to be the main cause of fistula formation in this patient.

Since uterocutaneous fistula is an extremely rare condition, there is no standard approach for its management. Review of the literature revealed only 2 successful medical treatment cases[3,7]. Both studies used Gonadotropin releasing hormone (GnRH) analogues as medical treatment option and had successful results. As uterocutaneous fistula is thought to have a similar lining with endometrium, it is assumed that temporary menopausal state would cause atrophy of the fistula tissue and cessation of menstruation would contribute to the closure of the tract. However, medical treatment of uterocutaneous fistula may be insufficient in other cases, and has several limitations. First of all it is a long treatment process with an uncertain success rate. Secondly, the uterocutaneous fistulas, unlike utero-vesical fistula, have usually a larger size of tract which may be another obstacle for medical treatment.

Surgical management option appears to be the option of choice in the remaining cases reported in the literature, as a shorter treatment process with more accurate results. Additionally, if the patient wishes to become pregnant as in our case, surgical management should be considered to ensure integrity of the uterine wall. However, risks of surgery should be discussed with patient thoroughly and a decision between the medical and surgical treatment options should be made together with the patient.

As surgical management, mini laparotomy combined with laparoscopy, if available, appears to be the most rational option. It is reasonable to start with laparoscopy, since uterus and possibly other intraabdominal organs are expected to be densely adherent to anterior abdominal wall. In our case, after laparoscopic adhesiolysis, a mini laparotomy was performed to excise the fistula tract completely from both abdominal wall and uterus.

In conclusion, although a very rare entity, uterocutaneous fistula can be difficult to manage. Prevention could be possible with better uterine closure techniques, prevention of postoperative infections and avoiding multiple surgeries at the same site. When encountered, treatment modality should be discussed with patient considering fertility preservation desire, and successful pregnancy could be achieved with appropriate surgical treatment.

LEARNING POINTS/TAKE HOME MESSAGES

- Since there are no established guidelines for management of rare cases like uterocutaneous fistula, reporting and reading rare situations and their treatment outcomes may be crucial when these cases are encountered.
- When postsurgery discharge from skin incision is seen, uterocutaneous fistula should be considered among differential diagnoses.
- Multiple abdominal surgeries, incomplete closure of uterine wound during myomectomy or caesarean section, use of drains and postoperative complications such as infection and inflammation are some of the known risk factors for fistulas.
- Uterocutaneous fistulas can be treated by appropriate surgery.
- Successful pregnancies and live birth after complete recovery are possible.

REFERENCES

- 1) Dragoumis K, Mikos T, Zafrakas M, Assimakopoulos E, Stamatopoulos P, Bontis J. Endometriotic uterocutaneous fistula after cesarean section. *Gynecol Obstet Invest* 2004;57:90-2.
- 2) Eldem G, Turkbey B, Balas S, Akpinor E. MDCT diagnosis of uterocutaneous fistula. *Eur J Radiol Extra* 2008;67:e129.
- 3) Seyhan A, Ata B, Sidal B, Urman B. Medical treatment of uterocutaneous fistula with gonadotropin-releasing hormone agonist administration. *Obstet Gynecol*. 2008;111:526-8.
- 4) Gupta SK, Shukla VK, Varma DN, et al. Uterocutaneous fistula. *Postgrad Med J*. 1993;69:822–823.
- 5) Sonmezer M, Sahincioglu O, Cetinkaya E, Yazici F. Uterocutaneous fistula after surgical treatment

- of an incomplete abortion: Methylene blue test to verify the diagnosis. Arch Gynecol Obstet 2009;279:225.
- 6) Malhotra D, Vasishta K, Srinivasan R, Singh G. Tuberculous uteroenterocutaneous fistula: A rare post-cesarean complication. Aust NZ J Obstet Gynaecol 1995;35:342.
 - 7) Yadav P, Gupta S, Singh P, Tripathi S. Successful medical management of uterocutaneous fistula. Int J Gynaecol Obstet 2014;124:263-4

FIGURE/VIDEO CAPTIONS

Figure: T2 sagittal MRI image demonstrating the communication between the fundal myometrial defect and the skin incision

INTELLECTUAL PROPERTY RIGHTS ASSIGNMENT OR LICENCE STATEMENT

I, Ertan Saridogan, the Author has the right to grant and does grant on behalf of all authors, an exclusive licence and/or a non-exclusive licence for contributions from authors who are: i) UK Crown employees; ii) where BMJ has agreed a CC-BY licence shall apply, and/or iii) in accordance with the relevant stated licence terms for US Federal Government Employees acting in the course of the their employment, on a worldwide basis to the BMJ Publishing Group Ltd (“BMJ”) and its licensees, to permit this Work (as defined in the below licence), if accepted, to be published in BMJ Case Reports and any other BMJ products and to exploit all rights, as set out in our licence [author licence](#).

Date: 30.05.2019