

Uterocutaneous Fistula Following Conservative Management of Placenta Increta: A Rare Case Report and Review of the Literature

Donya Khosravi,¹ Maliheh Arab,^{2,3,*} Behnaz Ghavami,⁴ Maryam Shokrpour,⁵ Samaneh Sheibani,¹ and

Samaneh Saraeian¹

¹Assistant of Obstetrics and Gynecology, Imam Hossein Medical Center, Shahid Beheshti University of Medical Sciences, Tehran, IR Iran

²Professor of Gynecology-oncology, Imam Hossein Medical Center, Shahid Beheshti University of Medical Sciences, Tehran, IR Iran

³PhD of Medical Education, Shahid Beheshti University of Medical Sciences, Tehran, IR Iran

⁴Shariati Medical Center, Tehran University of Medical Sciences, Tehran, IR Iran

⁵Assistant Professor of Obstetrics and Gynecology, Arak University of Medical Sciences, Arak, IR Iran

*Corresponding author: Maliheh Arab, PhD, Professor of Gynecology-Oncology, Imam Hossein Medical Center, Shahid Beheshti University of Medical Sciences, Madani Street, Tehran, IR Iran. Fax: +98-21-77543634, E-mail: drmarab@yahoo.com

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Abstract

Introduction: This was a report of uterocutaneous fistula as a rare complication of expectant management of placenta increta, and a review of the literature.

Case Presentation: A 38-year-old pregnant woman in her third term of pregnancy was operated on in a secondary hospital in Arak (a city located in the center of Iran) in April 2015. As a result of placenta increta, the attached placenta was left in the uterus. On the 38th day, she was referred to our tertiary care hospital due to intermittent fever and bleeding. More examinations by MRI and hystrosalpingography revealed uterocutaneous fistula. She was operated on again, and surgical findings indicated coexistent uterine necrosis and uterocutaneous fistula.

Conclusions: This case was the first reported morbidity of fistula in the uterocutaneous pathway.

Keywords: Placenta Accreta, Fistula, Disease Management, Iran

1. Introduction

Placenta accreta is abnormally attached placental villi to the myometrium. The decidua basalis defect facilitates passage of the villi into the myometrium. This pathophysiology is a barrier to normal hemostasis in the postpartum period. Sometimes, bleeding is severe and hysterectomy is necessary to save life. If the placenta is detached forcefully, severe bleeding (more than 3 lit) may occur, as it has been reported in 40% of the placenta accreta cases (1). Severe bleeding after manual detachment of placenta is due to opening of spiral sinuses, which is regarded as an important morbidity (2). Transfusion is needed in about 90% of the patients, and more than 10 units of blood are transfused in 40% of the cases (3). The maternal mortality of placenta accreta in different studies is reported to be in the range of 0.3% to 7% (1, 3, 4).

There is a rise in the rate of placenta accreta parallel to the increase in the rate of cesarean delivery. There is a particular increased risk in placenta previa. Placenta accreta is reported in a range of about 1 out of 533 (5, 6). The 10-fold increase in placenta accreta risk during the past 50 years may be due to the increase in cesarean delivery and maternal age (6, 7). In addition to the strong risk fac-

tors such as previous cesarean scars and placenta previa, other known risk factors are multiparity, advanced maternal age, previous dilation and curettage, history of manual placenta removal, submucous myoma resulting in atrophy of endometrial mucosa, recurrent abortion, gestational product implanted in uterine diverticulum, and previous radium insertion (7-9). Placenta accreta-percreta can be diagnosed before delivery by sonography and MRI, especially in patients with risk factors such as previous cesarean as placenta previa. Some abnormal placentation cases are diagnosed just after delivery (10, 11). In a study, about 25% of the placenta accreta cases were not diagnosed antenatally. Another study indicated pelvic MRI harboring 19% false negative results (12). The American College of Obstetricians and Gynecologists confirmed hysterectomy as the optimal management of placenta accreta (13). Recent reports indicate similar morbidity of the conservative approach in selected cases with primary hysterectomy. Nowadays, conservative management is considered in selected cases. Fertility preservation is the main aim of conservative treatment (14, 15). Adjuvant treatment following conservative management including arterial ligation, uterine sutures, embolization of arteries, and methotrexate therapy are sometimes necessary (15, 16).

Due to an increase in the tendency for conservative management and fertility preservation, in addition to the increased prevalence of placenta accreta, it seems reasonable to pay strong attention to complications and consequences of conservative management of placenta accreta. Previous studies found blood transfusion, prolonged hospital admission, infection, and secondary hysterectomy to be the main complications of conservative management. In the present study, a rare complication of conservative management of placenta accreta and the review of the literature were presented.

2. Case Presentation

A 38-year-old female, who was in her third term of pregnancy, was operated on in a secondary hospital due to total placenta previa and accreta in the background of her 2 previous caesarian sections in Arak (a city located in the center of Iran) in April 2015. She had been operated by Pfannenstiel incision in the abdominal wall and Kerr incision in the uterus.

Her surgical record indicated placental invasion into the lower segment of the myometrium and cervical region. About 8 cm³ of the placenta remained in the cavity. Ten units pack cell and 4 units fresh frozen plasma were infused and a drain was left for 48 hours. A lower uterine segment had been packed in the operation field with long gauze that was left for 24 hours and removed through the vagina. Because of continuous bleeding, intrauterine balloon packing was alternated to the previous packing for another 24 hours. Uterotonic drugs and multiple antibiotic therapies had been prescribed. Due to fever (38 - 39°C) in the postoperative period (days 1 - 3), triple antibiotherapy including Ampicillin, gentamycin, and clindamycin had been started. In the fourth day, due to persistent fever, a new regimen (including meropenem-vancomycin) was prescribed. On the fifth day, the results of the abdomino-pelvic sonography revealed a heterogeneous collection similar to the retained placenta about 6 cm in endometrium besides the fluid collection, and a few air bubble about 8.5cm³ in the subcutaneous region above the cesarean scar. On the seventh day postoperation, she was discharged without fever and in a good condition.

On the eighth day, the results of a postoperation abdomino-pelvic CT scan revealed a heterogeneous 10 × 6 cm mass in the lower segment of the abdominal cavity in favor of retained placenta. On the ninth day postoperation, 85 mg methotrexate was prescribed. On the 24th day (during the follow-up), a 5 mm incisional defect with bloody discharge resulted in antibiotherapy and transvaginal sonography. A mass of 8.5-cm, filling the

lower segment and cervical area, was reported in addition to a 4-cm fluid collection in the abdominal wall, with drainage pathway into abdominal wall. The result of the serum BHCG was negative. On the 25th postoperation, abdominal-pelvic MRI showed a heterogeneous mass of 6 to 7 cm in the left side of the uterus, which was extended to the near serosal surface. Extension outside the uterus was not possible. On the 38th day postoperation, due to the continuous abdominal incision drainage and her medical history, she was referred to our center, a tertiary hospital in Tehran, Iran. Our hospital is a 570-bed, government general hospital, which is a referral center and offers service to patients nationwide.

During admission, she was afebrile with normal vital signs. A 5 mm defect in abdominal incision drained a little bloody discharge. On the 41st day after operation, MRI was repeated in our center and revealed 2 heterogeneous masses of 2 and 6 cm³ in favor of myoma as a retained placenta. Another MRI finding was a fistula pathway extending from the uterus to the abdominal wall. Hystrosalpingography with dye injection confirmed fistula diagnosis due to the entrance of dye into the abdominal cavity extending to the abdominal wall (Figures 1 and 2). Uterocutaneous fistula as a rare complication was diagnosed using MRI and Hystrosalpingography. The patient was scheduled for laparotomy and hysterectomy because of the co-existent placental retention with fistula. Sound and metallic sound pathway of fistula from the skin was followed inside in 5 cm by a thin-rubbery material. Intraoperative findings confirmed the diagnosis of uterocutaneous fistula. A cherny abdominal cavity was exposed by Pfannenstiel incision. Severe adhesion of omentum, bowels, peritoneum, and uterus existed. Uterine incision was open (incisional necrosis) in about 4 cm length, retaining its configuration without rupture.

Adhesions were released and difficult supracervical hysterectomy was done. Severe bladder adhesion in the front and bowel adhesion in the posterior part could make the total hysterectomy traumatic. The retained placenta was completely resected from the lower segment and cervical region. In the post operation period, she was tachycardic and febrile (low grade) for 48 hours. She was discharged 6 days after operation. Moreover, she was found to be healthy, afebrile, and asymptomatic in the follow-up.

The current case report followed the principles of the declaration of Helsinki, and it was approved by the medical ethics review board of Imam Hossein hospital. The name and personal details of the patient have been kept private.

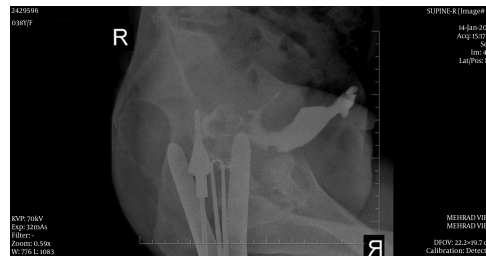


Figure 1. Hysterosalpingography Indicated Dye Continuing from the Anterior Part of the Uterus Into the Abdominal Wall.

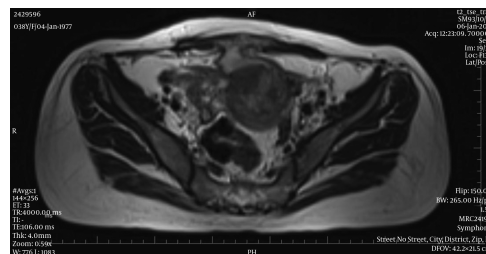


Figure 2. Magnetic Resonance Assessments (MRI) Indicated a Hetero- Signal Mass and Fistula in the Anterior Abdominal Wall Connecting to the Anterior Uterus (T2W- axilla view).

3. Discussion

Conservative management of abnormal placentation (placenta accreta-increta) is a known modality appropriate for specific cases. This method might preserve fertility, prevent severe bleeding, and other organs trauma (17).

Appropriate and conservative management of abnormal invasion of placenta can be done by removing spontaneously detached parts and leaving adherent parts in place. The less remaining placenta, indicate an increase in the success rate. Sometimes, complete resorption happens as spontaneous expulsion in the follow-up of the left placenta. In a review of 167 conservative management of placenta accreta, it was found that that the resorption of placenta took place in 13.5 weeks (range of 4 - 60 weeks). In a study, spontaneous resorption was mentioned in 87 out of 116 cases (75%). Another decision in leaving placenta was intervention by manual removal, hysteroscopic resection as curettage. In another study, intervention was suggested at 13 weeks (range of 4 - 60 weeks), and another opinion was at 2 months after delivery (17-21).

In the present case, about 8×3 -cm of the placenta was left in situ. One of the adjuvant treatments in conservation management of abnormal placentation is methotrexate (MTX), an immunosuppressive drug with cytotoxic activity. MTX reduces the placental vascularity and results in necrosis in placenta. There are different opinions about MTX efficacy. Some discuss against its use because of its op-

timal effect in rapidly dividing cells, which is not the case in term placenta. The study of Matsumura et al. confirmed the little effect of MTX in placental degeneration after term delivery. Another disadvantage of MTX therapy is drug-induced pancytopenia and nephrotoxicity. Although there is no documented support on MTX effectiveness, some authors believe it is a simple and perhaps effective treatment of left placenta, especially in minimal volumes of the remaining placenta. They discussed its effectiveness in reducing placental volume and infection rate. Discussion against its use is because of its immunosuppressive effect resulting in increased infection rate (17, 22-25).

In our case, single MTX therapy 50 mg/m^2 was prescribed in Day 9 after delivery.

Although the success rate of the conservative management of placenta accreta-increta is 78%, there are various reported side effects. The first group of side effects includes severe bleeding, transfusion, prolonged hospitalization, infection, and secondary hysterectomy (Table 1).

In the review of less common side effects, uterine necrosis, fistula, urinary system injury, and hematologic and thrombosis related disorders were observed (Table 2). According to our knowledge and the literature review, this case presentation of uterocutaneous fistula after the conservative management of placenta accreta was unique.

Table 1. Review of the Side Effects (Morbidity) of the Conservative Management in Placenta Accreta-Increta

	Number	Bleeding Amount	Transfusion (Frequency-Units) ^a	Admission Duration	Infection ^a	Secondary Hyst. ^a
Our case	1	2.5 lit	10%	12	+	+
Morken and Kahn (24)	1	Unknown	Unknown	14	Unknown	Unknown
Warshak et al. (26)	99	2.570 ± 1.7 mL	Mens: 5.7 ± 2.1 (in 3/4 of cases)	6.6 ± 1.8	Unknown	Unknown
Timmermans et al. (17)	60	Unknown	Unknown	Unknown	11/60 (18)	11/60 (18)
Crespo et al. (22)	1	Unknown	No	9	No	No
Nijman et al. (9)	1	No	No	Unknown	No	No
Timmermans et al. (17)	31	Unknown	No	Unknown	10/31(32)	3/31 (10)
Eshkoli et al. (8)	139	Unknown (bleeding in 3.6%)	75/139 (54)	7.19 ± 0.4	14/139 (10)	2 (1.4)
Eller et al. (12)	76	Mean 2.52	62/76 (82)	6	24/76 (32)	6/76 (8)
Bretelle et al. (15)	50	Unknown	19/50 (38) mean: 4.9 unit	12 (± 9)	12/50 (24)	20/50 (40)
Eller et al. (27)	141	2.2 (Range = 0.1 - 23)	120/141 (85)	5 (9 - 54)	38/141 (27)	10/141 (11)
Resnik et al. (20)	434	230 (53)	Unknown	Unknown	26/434 (6)	83/434 (19)
Sentilhes et al. (21)	167	Unknown	70/167 (42) 15% more than 5 unit	Unknown	54/167 (32)	18/167 (11)
Panoskaltis et al. (28)	2	Unknown	4	Unknown	No	No
		No	No		No	No
Clement et al. (18)	2	Unknown	7	9	+	No
		Unknown	No	10	No	No

^a Values are expressed as No. (%).**Table 2.** Review of Less Common Reported Morbidity of Conservative Management in Placenta Accreta-Increta^a

	Fistula	Uterine Necrosis	Urinary System Injury	Hematologic-Thrombosis Related Disorders	Others
Our case	Utero-cutaneous	+	-	-	-
Warshak et al. (26)	-	-	Bladder injury 17/99 (17) ureteral injury 6/99 (6)	-	-
Timmermans et al. (17)	-	-	-	DIC 4/60 (7)	-
Eller et al. (12)	2/76 (3) vesicovaginal	-	Ureteral injury 5/76 (7)	Pelvic hematoma/pelvic thrombosis each 1/76 (1.3)	Pancreatitis/76 (1.3) incision hernia 1/76 (1.3) pelvic abscess 1/76 (1.3)
Bretella et al. (15)	-	1/50 (2)	Bladder necrosis 1/50 (2)	DIC 8/50 (16) leg ischemia and thrombosis 1/50 (2)	-
Eller et al. (27)	-	-	Ureteral injury 10/141 (7)	Coagulopathy 44/141 (31)	-
Timmermans et al. (17)	-	1/31 (3)	-	-	-
Sentilhes et al. (21)	1/167 (0.6) vesicouterin	2/167 (1.2)	Acute renal failure 1/167 (0.6)	Deep vein thrombophlebitis or pulmonary embolism 3/167 (1.8) Arterial venous malformation 1/167 (0.6)	Pulmonary edema 1/167 (0.6) adjacent organ injury 1/167 (0.6) septic shock 1/167 (0.6)
Provansal et al. (19)	-	3/46 (6.5)	-	-	1/46 (2) acute ischemia of lower limb

^a Values are expressed as No. (%).

3.1. Conclusions

The present case was a rare morbidity of uterine necrosis and uterocutaneous fistula. Uterine necrosis is reported in 2% to 3% of some series (Table 2). Uterine necrosis as a rare morbidity might be caused by many different factors including technical repair defects, hematoma, infection, diabetes, multiparity, old age, immunosuppression, and obesity (29).

In review of the case reports, it was found that the fistulas were vesicovaginal in 2 out of 76 cases (3%) in one study (12), and vesicouterine in 1 out of 167 cases (0.6%) in another study (21). This case was the first reported morbidity of fistula in the uterocutaneous pathway.

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Footnotes

Conflict of Interest: None.

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