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#### CASE REPORT

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# Postpartum unscarred uterine rupture caused by placenta accreta: A case report and literature review

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#### **Key Clinical Message**

Our case and the literature review suggest that placenta accreta spectrum, with use of uterotonics and manual removal of placenta, could be risk factors for postpartum unscarred uterine rupture.

**KEYWORDS** 

placenta accreta, postpartum, retained placenta, unscarred uterine rupture, uterotonics

# 1 | INTRODUCTION

Postpartum unscarred uterine ruptures are exceedingly rare. A 29-year-old multipara with a retained placenta presented hypovolemic shock. An emergency laparotomy revealed that her uterus was ruptured where the placenta adhered to. Pathological examination confirmed the placental accreta that could be a risk factor for postpartum unscarred uterine rupture.

Uterine rupture is a rare obstetrical condition that can result in life-threatening situations for mothers and their babies. Although the main risk factors for uterine rupture are multiple cesarean sections and uterine trauma caused by myomectomy or adenomyomectomy, there have been several reports of unscarred uterine rupture.<sup>1,2</sup> The risk factors of unscarred uterine rupture are multiple gestations, congenital anomaly, uterotonics, and placenta accreta spectrum (accreta, increta, and percreta).<sup>2,3</sup>

Placenta accreta spectrum occurs when the placental villi directly attach to/invade the myometrium, a process known as abnormal placentation. Unscarred uterine rupture possibly occurs due to the weakness of uterine layers caused by placenta accreta spectrum in the third and even second trimester.<sup>4</sup> However, very few cases of postpartum unscarred uterine rupture possibly caused by placenta accreta spectrum have been reported. We here present a case of retained placenta due to placenta accreta followed by postpartum unscarred uterine rupture. We also review similar cases within the literature.

# 2 | CASE REPORT

The patient was a 29-year-old woman without any particular medical or surgical history. She had one vaginal delivery three years ago without complications at 40 weeks of gestation. She became pregnant by induction of ovulation using clomiphene citrate. Her pregnancy course was uneventful. She delivered a 2432-g baby (small-for-date infant) vaginally at 39 weeks and 5 days without the use of uterotonics or Kristeller maneuver. The delivery time until the end of the second stage was less than 4 hours. The umbilical cord was torn by umbilical cord traction and the placenta was retained in the uterus. Manual removal of the placenta was not successful because of shortness of the reach. The amount of bleeding during delivery was 380 mL and continuous bleeding was minimal.

The patient was transferred to our hospital the next day. Vaginal bleeding was minimal. On ultrasonography, the

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**FIGURE 1** Sagittal (left) and coronal (right) CT images with contrast enhancement revealed thinning of the muscular wall, suggesting placental adhesion (arrows)

border between the myometrium and placenta was unclear. Computed tomography (CT) revealed thinning of the muscular wall of the uterine fundus at the site of placental adhesion; however, leakage of the contrast medium from the uterus was not observed (Figure 1). Intravenous oxytocin (2 IU/h) and oral methylergometrine (0.75 mg/d) were administered for induction of placental delivery but failed.

On the second day after delivery, vaginal bleeding was minimal; however, the patient developed sudden abdominal pain with signs of hypovolemic shock. Rapid infusion and blood transfusion were performed. Intra-abdominal bleeding and uterine rupture were suspected on CT (Figure 2A), and an emergency laparotomy was subsequently performed. We observed massive intraperitoneal bleeding along with a 3-cm ruptured wound on the uterine fundus where the placenta adhered to (Figure 2B). The uterine muscular wall was thinned around the ruptured wound. Thus, we presumed that uterine preservation was impossible and performed a total hysterectomy. Blood loss during surgery was 3100 g; the total of blood transfusion was 14 IU TBC, 960 ml FFP, and 20 IU PC. The postoperative course was uneventful. She was discharged on the seventh postoperative day. Placental accrete was confirmed by pathological examination. Written informed consent was obtained from the patient.

# **3** | **DISCUSSION**

Unscarred uterine rupture is a rare condition. Gibbins et al<sup>5</sup> reported that the incidence of unscarred uterine rupture is 4.54 per 100 000 deliveries. There are various reports indicating that the risk factors for unscarred uterine rupture include high parity, uterotonics, uterine anomalies, advanced maternal age, dystocia, instrumental delivery, uterine fundal pressure, macrosomia, multiple gestation, placenta accreta spectrum (accrete, increta, and percreta), short interpregnancy interval, prior dilatation and curettage, manual removal of placenta, and congenital disorders, such as Ehlers-Danlos type IV.<sup>2,3</sup> Among them, placenta accreta, postpartum use of uterotonics, and manual removal of placenta apply to our case.

Uterine rupture caused by the use of uterotonics mainly occurs during pregnancy and delivery. When a fetus/fetuses are present in the uterus, administration of uterotonics could potentially induces excessive uterine contraction followed by collapse of the myometrium. However, in cases of retained placenta, intrauterine pressure does not increase since excessive pressure is released from the cervix. Therefore, it is unlikely that the case of unscarred uterine rupture described here was only caused by the administration of uterotonics. Uterine perforation or uterine rupture could be caused by manual removal of the placenta. However, in the current case, the attending physician refrained from manual removal of placenta as to not to risk uterine rupture and since there no signs that separation of the placenta would be easy. The uterine rupture occurred around the site of placental adhesion. In this case, it is unlikely that the myometrium was damaged by manual removal of the placenta. Therefore, placenta accreta is the main reason for unscarred uterine rupture but could also be affected by use of uterotonics and manual removal of the placenta.

To the best of our knowledge, only 3 cases of postpartum unscarred uterine rupture caused by placenta accreta spectrum have been reported (Table 1).<sup>6-8</sup> Uterine rupture occurred immediately after delivery (3 hours and 1 hour) in two of the three cases. These two cases underwent manual removal of the retained placenta. Placenta percreta was histologically confirmed afterward in both cases. In addition, the case described in Veenstra et al had a history of frequent dilatation and curettage and the case described in Morken et al have been administered with uterotonics for the induction of labor. In these two cases, uterine rupture occurred immediately after delivery. Thus, manual removal of the retained placenta percreta could be the direct cause of unscarred uterine rupture. **FIGURE 2** (A) Sagittal (left) and coronal (right) CT images with contrast enhancement revealed intraperitoneal bleeding and thus suspected uterine rupture (arrows). (B) Laparotomy findings. Ruptured wound on the uterine fundus and placenta thorough the wound (arrow). Uterine muscular wall was thinned around the ruptured wound



TABLE	1	Cases of postpartum	unscarred	uterine rupture
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Author Year	Age (y)	Gravida/ Para	Gestation (wk)	Time to uterine rupture after delivery	Histopathology	Treatment for retained placenta	Uterotonics
Veenstra (1995) <sup>6</sup>	38	G10/P0/ SA8/AA1	25	3 h	Percreta	MR (failed)	
Morken (2001) <sup>7</sup>	28	G2/P1	38	1 h	Percreta	MR	
Gherman (2004) <sup>8</sup>	34	G1/P0	32	13 d	Not assessed (clinical diagnosis is accreta)	Curettage	Intravenous oxytocin, intramuscular methylergonovine and prostaglandin F2, rectal misoprostol (after curettage)
Our case	29	G2/P1	39	2 d	Accreta	MR (failed)	intravenous oxytocin, oral methylergometrine (after delivery)

Abbreviations: AA, artificial abortion; MR, manual removal; SA, spontaneous abortion.

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On the other hand, in the case described in Gherman et al the patients suffered from uterine rupture 13 days after delivery. In that case, the placenta was removed by curettage, followed by the use of four types of uterotonics. In addition, the case was complicated by intrauterine infection, and infection and inflammation may weaken the uterine myometrium. No pathological examination of the placenta was carried out in that case, although the clinical diagnosis was placenta accreta.

Sturzenegger et al<sup>2</sup> reported that abnormal placenta is a risk factor for unscarred uterine rupture (adjusted odds ratio 20.82). The risk factors for abnormal placenta such as placenta previa with/without placenta accreta have been discussed in detail.<sup>9,10</sup> In our case, there were no risk factors of placenta accrete spectrum. However, the border between the myometrium and the placenta was unclear on ultrasonography. The CT revealed thinning of the uterine muscular wall, suggesting placental adhesion. These features are possibly implicated in the placenta accrete spectrum.<sup>11,12</sup> The risk factors for unscarred uterine rupture have been mainly discussed in the context of prepartum cases.<sup>3,13</sup> Considering our case and the literature review, placenta accrete spectrum may be a risk factor of postpartum unscarred uterine rupture. Therefore, when placenta accreta spectrum is suspected as the cause of retained placenta, a more careful management is required, and keeping postpartum unscarred uterine rupture in mind. This is of particular importance in cases of use of uterotonics and manual removal of the placenta.

In conclusion, we presented the case of postpartum unscarred uterine rupture with placenta accreta. The literature review suggests that placenta accreta spectrum, with use of uterotonics and manual removal of placenta, could be risk factors for postpartum unscarred uterine rupture.

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## **CONFLICT OF INTEREST**

None declared.

#### AUTHOR CONTRIBUTIONS

JO, DH and AI: wrote the manuscript. JO, DH, YU and MI: involved in the management of the patient. TK: supervised the whole management and preparation for the manuscript. All authors: discussed the results and contributed to the final manuscript.

### ETHICAL APPROVAL

Need for approval of the ethical committee was waived.

### DATA AVAILABILITY STATEMENT

The data of the study are available from the corresponding author upon reasonable request.

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