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Case Report

Conservative management for adherent placenta after live birth in angular or interstitial pregnancies: A new entity “angular placenta attachment”

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ABSTRACT

Objective: Angular and interstitial pregnancies have been reported with live births and are often complicated by adherent placentas. Most cases had been treated with hysterectomy or cornual resection.**Case report:** We successfully treated four patients with conservative management (including one reported previously). Case 1 had a vaginal delivery, but the placenta remained attached. We maintained the patient under observation and delivered the placenta on postpartum day 9. Case 2 underwent a C-section. Uterine artery embolization controlled the hemorrhage without placenta removal. The placenta had disappeared by postpartum day 136. Case 3 underwent a C-section. The right uterine angle, where the placenta was attached, was bulging. We manually removed the placenta.**Conclusion:** We propose a new entity in angular or interstitial pregnancies called “angular placenta attachment” that could be diagnosed during C-sections or after vaginal delivery without placental separation. Expectant management may be considered for adherent placentas in these cases.© 2020 Taiwan Association of Obstetrics & Gynecology. Publishing services by Elsevier B.V. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

Introduction

Distinguishing angular (cornual) from interstitial pregnancies may be difficult, and these entities have been used interchangeably. Some of these cases have been reported with live births, and they are often complicated by adherent placentas. Only 11 cases of adherent placenta after live birth have been reported [1–11] (two of them by our group [1,10]). Most cases had been treated with hysterectomy or cornual resection. However, we reported a case of interstitial pregnancy with adherent placenta treated successfully with conservative management [1], and after that, we encountered three more cases treated likewise. We have two questions: Are all of these cases true angular or interstitial pregnancies? Can the cases with adherent placenta be treated conservatively? This is a first case series discussing conservative management for adherent placenta in cases that we have called angular or interstitial pregnancies. We present our cases and a literature review and propose a new entity called “angular placenta attachment.”

Case presentation

Case 1

A 30 year-old Japanese primigravida conceived spontaneously and started pregnancy checkups in China. She came to our hospital at 28 gestational weeks after moving to Japan. We observed no complications until she gave birth to a healthy 3212 g female infant by spontaneous vaginal delivery at 39 weeks. The patient received intravenous oxytocin after delivery, but the placenta remained attached even after a 30 min third stage. An ultrasound revealed the placenta located in the uterine fundus on the right side, with the myometrium in that region being extremely thin (Fig. 1a). Accordingly, we suspected retained placenta due to interstitial or angular pregnancy. Given the lack of severe hemorrhage and the patient's wish to retain her uterus, we decided to leave the placenta in situ and wait for its spontaneous separation. The diagnosis of interstitial or angular pregnancy was confirmed by both CT (Fig. 1b) and magnetic resonance imaging (MRI) (Fig. 1c), which both revealed an enlarged right uterine angle with a thin myometrium. On postpartum day (PPD) 6, the patient developed a persistent fever over 38.0 °C. We suspected intrauterine bacterial infection

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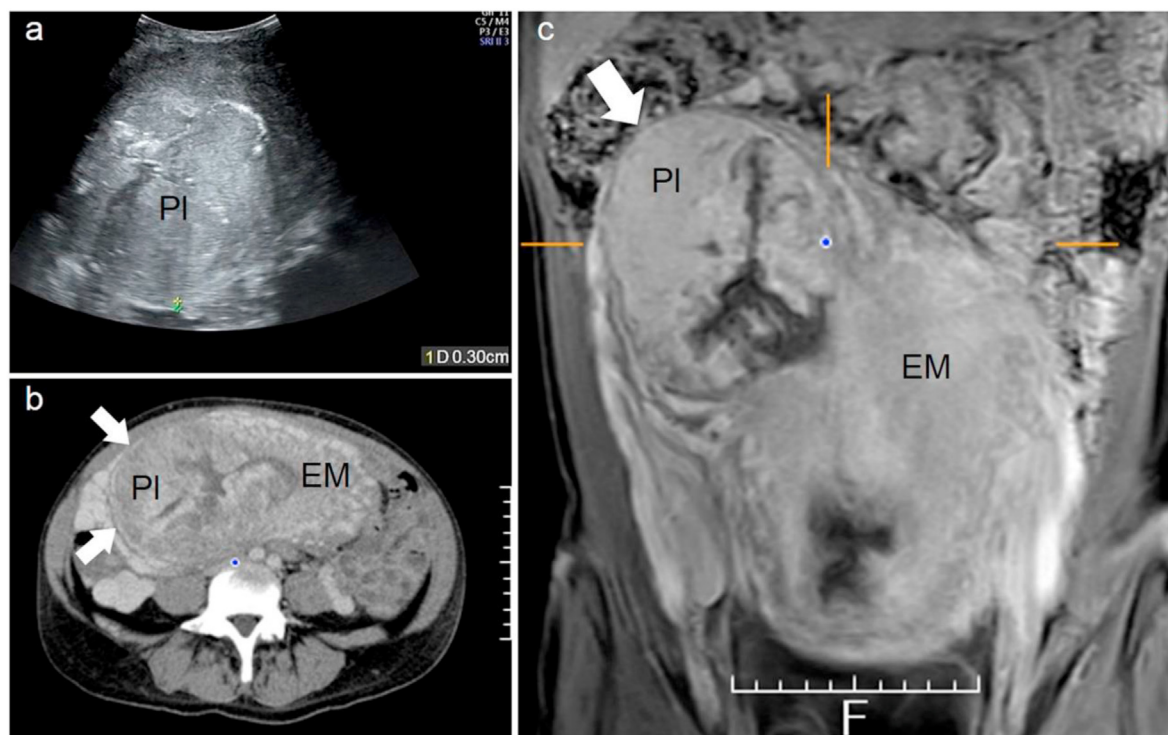


Fig. 1. Postpartum images of Case 1. a) Transabdominal ultrasound image of the uterine fundus after a vaginal delivery of the fetus. The myometrium is only 3 mm thick (arrow). b) Axial contrast-enhanced CT image on PPD 2. Note the strong placenta enhancement and the thickness of the lateral myometrium around the placenta (arrow). c) Coronal gadolinium-enhanced T1-weighted MRI image on PPD 9, showing the placenta at the right uterine angle. Arrow showing thin myometrium. PPD, postoperative day; PI, placenta; EM, endometrium.

and started intravenous antibiotic therapy. On PPD 9, the patient had a small amount of vaginal bleeding. We found the placenta had spontaneously moved toward the center of the uterus according to an ultrasound image. We infused oxytocin, and the placenta was delivered without significant bleeding. The patient's temperature normalized on PPD 11, and she was discharged on PPD 13.

Case 2

A 31 year-old primigravida was referred to our hospital at 27 gestational weeks because her fetus was suspected to have congenital anomalies. The ultrasound and the MRI revealed pulmonary agenesis of the fetal left lung and polyhydramnios. Although the placenta was on the left uterine angle, asymmetrical enlargement of the uterus was not apparent in the MRI (Fig. 2a). Induction of labor was attempted at 36 gestational weeks, but the patient underwent an emergency C-section due to induction failure, and an 1810 g female infant was delivered. During the operation, we observed an enlarged left angle of the uterus, medial to the round ligament (Fig. 2b). The myometrium on that region was so thin that we could see the placental tissue through it. Although the patient received intravenous oxytocin, the placenta failed to separate from the uterine wall easily. We suspected placenta accreta due to angular pregnancy. Since this patient also strongly hoped to preserve her uterus, we decided to leave the placenta inside the uterus and close the incision. At the end of the operation, the total blood loss reached 2000 g. Patient's hemodynamic state was normalized by blood transfusion, and uterine artery embolization after the surgery controlled the hemorrhage. MRI on POD 4 revealed a large residual placenta at the left uterine angle with diffuse gadolinium enhancement (Fig. 2c and d). The patient was discharged on POD 16. After that, she was checked once a week for

genital bleeding and fever, and she developed neither of them. MRI on POD 39 showed a reduction of placental size with only the periphery of placenta enhanced (Fig. 2e). On a POD 136 MRI, we found the placenta had almost disappeared (Fig. 2f).

Case 3

A 31 year-old woman with prior vaginal delivery was referred to our hospital at 20 gestational weeks because her fetus was suspected to have Tetralogy of Fallot. Her antenatal course was uneventful. We performed a C-section at gestational week 37 because of breech presentation. A 2550 g female infant was delivered but the placenta remained in the uterus. The right uterine angle region was bulging, medial to the round ligament (Fig. 3a and b), and the placenta was attached to that part. After the intravenous injection of oxytocin, the placenta failed to separate spontaneously. Therefore, we manually compressed the uterine myometrium and pulled the placenta, which separated gradually. The placenta had two lobes (Fig. 3c). The bleeding stopped, and we observed no complications and discharged the patient on POD 7.

Case 4

We have published the case of this woman with interstitial pregnancy and viable infant presenting placenta accreta [1]. We diagnosed interstitial pregnancy during a C-section, and the placenta did not separate until POD 9.

Discussion

Angular pregnancies are rare forms of ectopic pregnancy in which the embryo implants in the lateral angle of the uterine cavity.

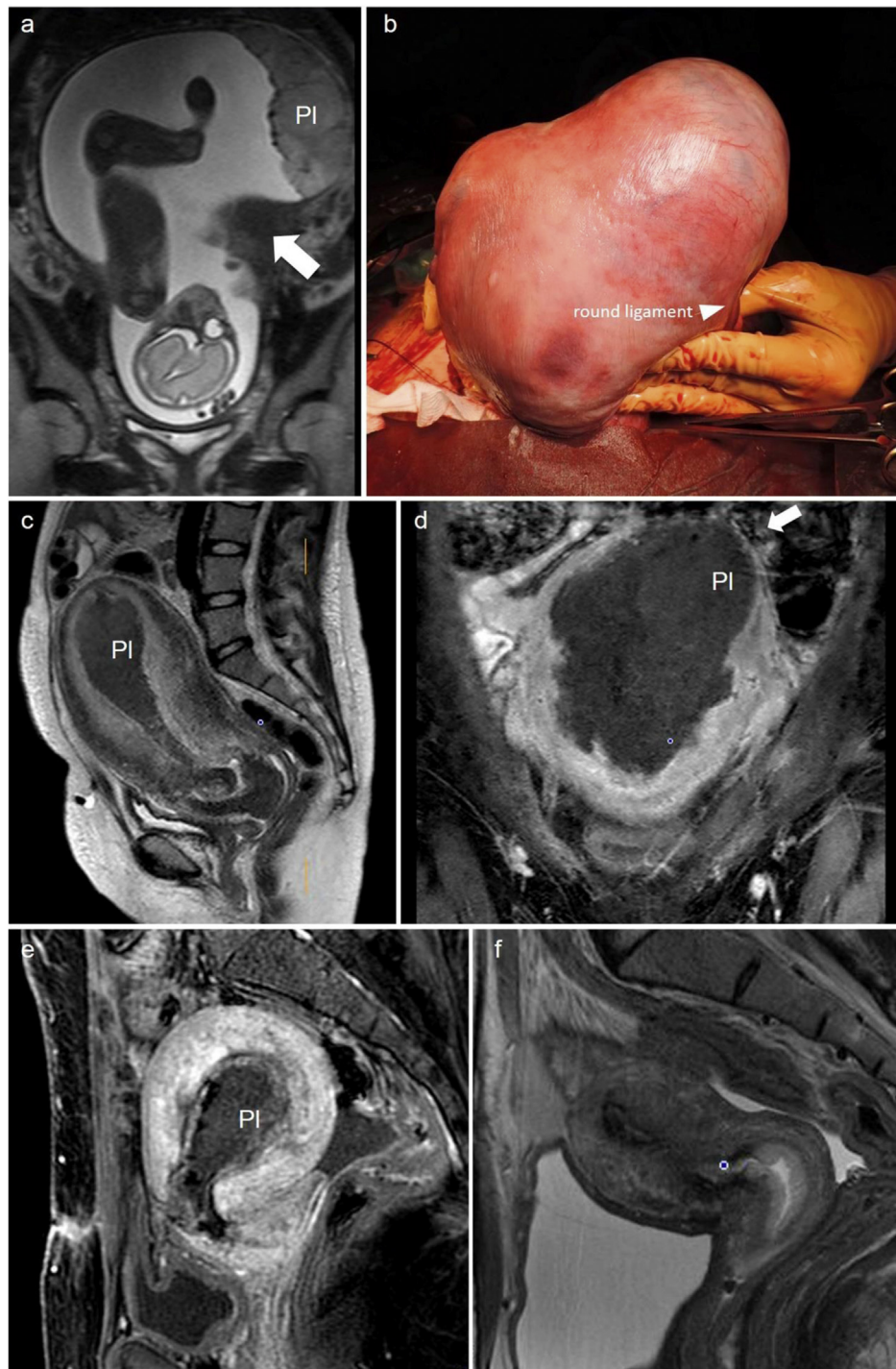


Fig. 2. MRI images and photograph at surgery of Case 2. a) Coronal T2-weighted image showing the placenta on the left angle of the uterus. Although asymmetrical enlargement of the uterus was not apparent, we observed an atypical retraction of the uterine myometrium (arrow). A fetus in the uterine corpus. b) Left anterior view of the uterus after fetal delivery showing a left angular bulge containing the adherent placenta. Sagittal T2-weighted image (c) and coronal gadolinium-enhanced T1-weighted image (d) showing a large residual placenta on the left uterine angle and the thin myometrium on that region (arrow). The placenta is weakly but diffusely enhanced. e) A sagittal gadolinium-enhanced T1-weighted image on POD 39. The placenta has been decreased in size and only its periphery is enhanced. f) A sagittal T2-weighted image on POD 136. The placenta has disappeared almost completely. PI; placenta.

The differential diagnosis of an angular pregnancy is an interstitial pregnancy, in which the embryo implants in the interstitial part of the fallopian tube. They both result in the lateral enlargement of the uterine angle, but the enlargement of an angular pregnancy displaces the round ligament reflection upward and outward, whereas in an interstitial pregnancy, the swelling locates lateral to the round

ligament. Differentiating angular from interstitial or even normal pregnancies can be difficult. Some patients with angular or interstitial pregnancies may be diagnosed during early gestation during a transvaginal ultrasound examination, and 3-D ultrasound and magnetic resonance exams can facilitate these diagnoses [12]. The overall live birth rate of an angular pregnancy has been reported at

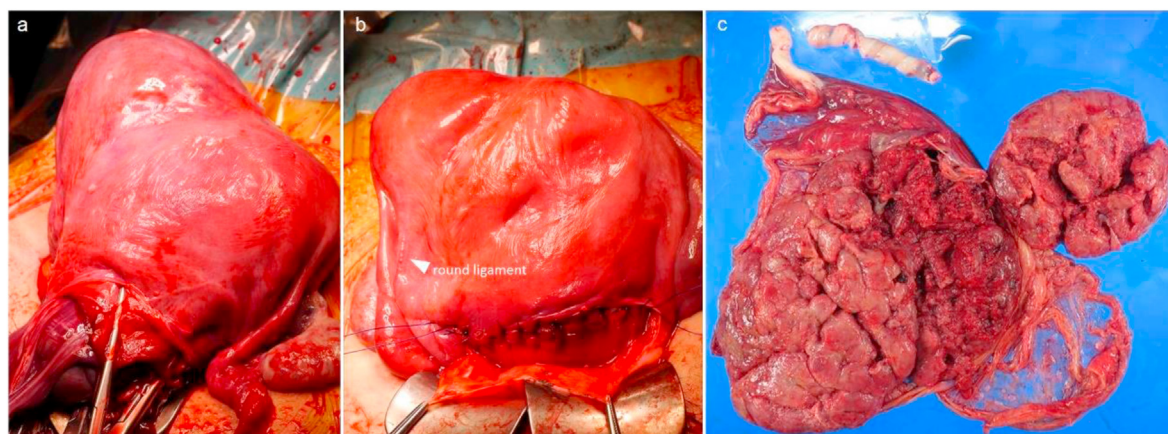


Fig. 3. Photographs at surgery and placenta of Case 3. a) Left anterior view of the uterus after fetus delivery showing an asymmetric angular bulge containing the placenta. b) A view of the uterus after the successful manual removal of the placenta. The enlargement at the right angular part has become smaller. c) Photograph of the placenta revealing two lobes.

25–80% [13,14]. More than half of the patients identified as angular pregnancy at first trimester ultrasound had normalized ultrasound findings at follow up about 2 weeks later [14], suggesting that the prognosis differs by the timing of diagnosis. Complications of angular pregnancy include uterine rupture (14%–29%), placenta accreta/increta/percreta (6%–10%), retained placenta or incomplete placenta removal (4%) [13], and increased risks of preterm delivery, placental abruption, and growth restriction ([15]). Interstitial pregnancies have worse prognoses than angular pregnancies, with much higher incidence of uterine rupture in the first trimester. However, the true incidence of each entity is unclear because the two have been used interchangeably.

Taking angular pregnancies and interstitial pregnancies together, only 11 cases of adherent placenta after live birth were reported in the past 50 years (Table 1). Four cases were reported as angular pregnancies [2–5], and seven cases as interstitial pregnancies [1,6–11]. In some cases, however, the authors failed to mention the association between the enlarged part and the round ligament, making it hard to ascertain whether the diagnoses were accurate. Three cases out of four angular pregnancies were diagnosed after an uncomplicated vaginal delivery when the placenta failed to separate [2–5], and one case during a C-section [5]. The placenta was attached to the uterine angle in all cases. Manual

removal of the placenta and/or sharp curettage were attempted in all cases with vaginal delivery, but they all failed. As a result, hysterectomy was performed in one case and cornual resection in two cases. As for the case with C-section, manual removal of the placenta followed by three square compression sutures was performed successfully. On the other hand, C-sections were performed for obstetrical indications in all seven cases of interstitial pregnancy (including our previous case). Five were treated with hysterectomy [6–10], and two were successfully managed by uterine conservation (one with an intramuscular methotrexate injection [11], and the other (our case) was managed expectantly [1]). Other than expectant management, cornual resection (hysterotomy) can be a substitute option for those hoping to preserve future fertility. However, since the incidence of uterine rupture and dehiscence in subsequent pregnancy after corneal resection is high [16], the choice requires careful consideration.

Although only one case was suspected as being an interstitial pregnancy after a 26-gestational-week MRI, the other 10 cases were diagnosed as angular or interstitial pregnancies after vaginal delivery of the fetus or during C-sections. This shows the difficulty in detecting these cases before labor. As for our cases, all four had started the routine pregnancy checkup during the early gestation, and an antenatal MRI was further performed in Case 2, and yet we

Table 1

Previous publications of angular pregnancies and interstitial pregnancies with adherent placenta after live birth.

Reference No.	types of reported pregnancy	Reported year	Maternal age (y)	Previous surgery	Gestational age (wks)	Uterine rupture	Mode of delivery	Birth weight (g)	Type of treatment
[2]	angular	2000	33	Cesarean delivery	38	No	VD	2685 g	Cornual resection
[3]	angular	2004	27	none	41	No	VD	3150 g	Subtotal hysterectomy
[4]	angular	2010	26	laparoscopic left adnexectomy, right cystectomy	30	No	VD	*	Cornual resection
[5]	angular	2016	34	Cesarean delivery	32	No	CS	1650 g	Manual removal, 3 square compression sutures
[6]	interstitial	1988	38	Myomectomy	38	Yes	CS	3010 g	Subtotal hysterectomy, Salpingo-oophorectomy
[7]	interstitial	1997	*	Myomectomy	33	Yes	CS	2100 g	Subtotal hysterectomy
[8]	interstitial	1998	31	Cesarean delivery	37	No	CS	2786 g	Supracervical hysterectomy
[9]	interstitial	2012	30	none	28	No	CS	1000 g	Hysterectomy
[10]	interstitial	2018	33	Cesarean delivery	38	No	CS	3148 g	Hysterectomy
[11]	interstitial	2007	25	none	38	No	CS	*	Methotrexate
Our case 1	angular/interstitial		30	none	39	No	VD	3212 g	Expectant management
Our case 2	angular		31	none	36	No	CS	1810g	UAE, Expectant management
Our case 3	angular		31	none	37	No	CS	2550 g	Manual removal
Our case 4 [1]	interstitial	2014	35	none	32	No	CS	1038 g	Expectant management

* Not available.

did not diagnose them as angular/interstitial pregnancies. The asymmetrical enlargement of the uterus becomes apparent after the delivery of the fetus when the uterus begins to contract. However, whether these cases were true angular or interstitial pregnancies cannot be confirmed because all cases were missed after first trimester ultrasound examinations.

In conclusion, we propose a new entity of angular or interstitial pregnancies called “angular placenta attachment,” in which only the placenta is located in the uterine angle or in the interstitium while the fetus grows into the intrauterine cavity. This entity could be diagnosed during C-sections or should be considered as a differential diagnosis when the placenta does not come off after vaginal delivery. Expectant management could be considered for adherent placenta in these cases.

Statement of ethics

The institutional review board of Osaka University approved this study on Jun 13, 2019 (approval number 15240). We also obtained written informed consents from the patients.

Declaration of Competing Interest

The authors indicated no potential conflicts of interest.

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