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Incarceration of a gravid uterus with massive placental enlargement and fetal triploidy at 19 weeks of gestation – A case report on the simultaneous presence of two rare conditions

Judith Sarah Abel *, Bernd Morgenstern

University of Cologne, Faculty of Medicine and University Hospital Cologne, Department of Gynecology and Obstetrics, Germany

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ABSTRACT

This article reports a rare case of uterine incarceration in pregnancy concurrent with nonmolar fetal triploidy and massive placental enlargement in a 35-year-old primigravida. The patient presented with abdominal discomfort and peripheral edema at 19 weeks of gestation.

Diagnostic assessments revealed a retroflexed uterus with a massively enlarged placenta and a severely growth-restricted fetus. Uterine repositioning was successfully achieved after rectal filling. However, spontaneous fetal demise led to vaginal delivery. The fetal autopsy confirmed female triploidy. Histopathology of the placenta showed no features of a partial mole.

This case highlights the challenges in diagnosing uterine incarceration in pregnancy, the need for early intervention and the complexity of managing multiple concurrent pathologies.

1. Introduction

We report on the simultaneous presence of uterine incarceration and nonmolar fetal triploidy with massive placental enlargement in a 35-year-old primigravida. The patient presented at 19 weeks of gestation with back pain, abdominal pressure, urinary discomfort and edema. Clinical examination and further diagnostic assessment revealed an enlarged placenta with a distinct two-part structure and a fixed retroflexed uterus as well as a severely growth-restricted fetus. Uterine repositioning could be achieved after rectal filling. The case concluded with intrauterine fetal death and subsequent vaginal delivery. Fetal autopsy confirmed female triploidy (XXX). Histopathology showed no features of a partial mole.

Uterus incarceration is a rare event and early diagnosis is important to prevent serious complications.

2. Case Presentation

A 35-year-old primigravida presented at 19 weeks of gestation with intense back and flank pain. She also complained about a feeling of pressure in the lower abdomen and the urinary bladder as well as increasing peripheral edema. The pregnancy had been uneventful up to that point, and her medical history was unremarkable. In her medical files a normal early-screening ultrasound was documented. She had had neither first-trimester screening, nor invasive or non-invasive prenatal testing.

The cervix was palpable behind the symphysis and the Douglas cavity was had a bulging-elastic consistency. The cervical os was shifted far ventral-cranially and could not be visualized.

Ultrasound showed an intact intrauterine singleton gestation with severe fetal growth restriction: head circumference, abdominal circumference and femur length were far below the 3rd percentile, resulting in an estimated fetal weight of 128 g. Doppler ultrasound showed a reverse flow in the arteria umbilicalis and a pulsatile flow in the ductus venosus. As the peak systolic velocity in the middle cerebral artery was normal, no signs of fetal anemia could be found.

The placenta was massively enlarged and had a total size of 9 × 14 × 19 cm. Within the placenta, there was a two-part structure: a homogenous, hyperechoic larger portion and a smaller hypoechoic, inhomogeneous structure of about 9 × 5 × 9 cm in vicinity of the fetus (Fig. 1). There were no sonographic features of a molar placenta. Amniotic fluid was normal and there was no urinary congestion.

Taking all clinical and sonographic findings into consideration, the diagnosis of a fixed, retroflexed uterus was made. The uterine volume

* Corresponding author at: Department of Gynecology and Obstetrics, University Hospital of Cologne, University of Cologne, Kerpener Straße 34, 50931 Cologne, Germany.

E-mail address: judith.abel@uk-koeln.de (J.S. Abel).

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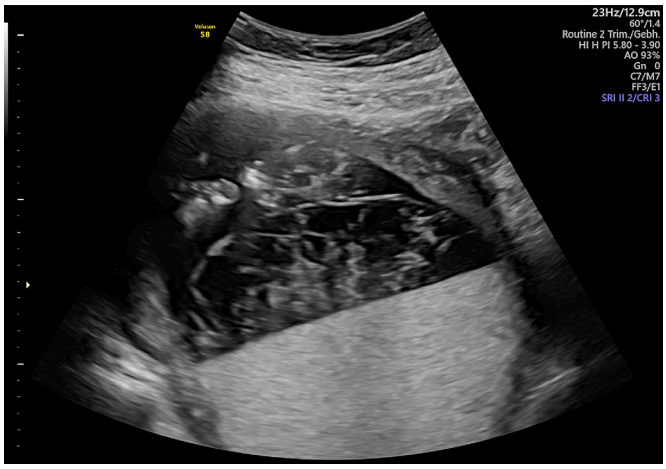

Fig. 1. Sonographic image of the two-part structure within the placenta: a homogenous, hyperechoic larger portion and a smaller hypoechoic, inhomogeneous structure of about 9 × 5 × 9 cm in vicinity of the fetus.

was significantly increased due to the abnormal enlarged placenta. The patient was hospitalized for initial pain treatment. Next day a sonographic follow-up was performed. It confirmed the finding of severe fetal growth restriction with pathologic fetal haemodynamics, specifically a high cardiac output failure without fetal anemia. The patient was counselled on invasive diagnosis to exclude aneuploidy. Because of the unusual placental structure, an acute placental abruption was suspected and chorionic villus sampling was postponed.

After exclusion of an acute process, magnetic resonance imaging (MRI) was performed. Imaging confirmed the unclear changes in the placenta as well as the retroflexion of the entire uterus with incarceration of the caudal corpus caudal of the promontory (Fig. 2).

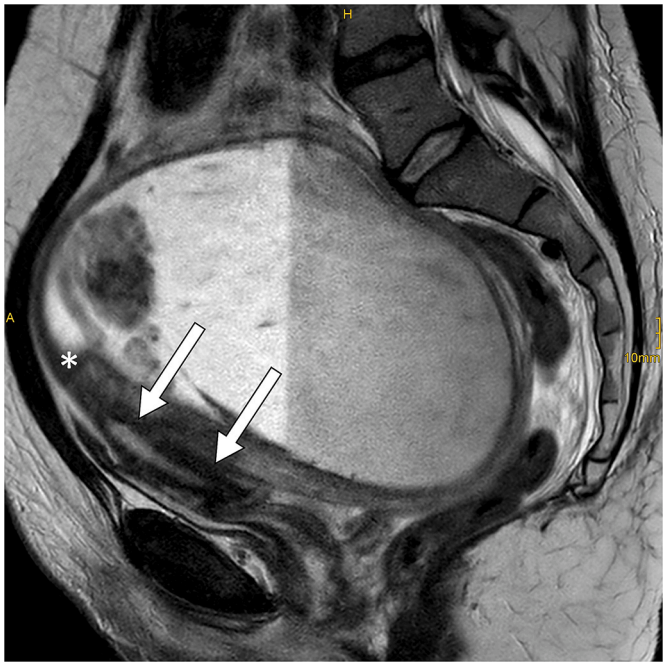

Fig. 2. MRI scan with sagittal section through the small pelvis with the patient in a supine position. The distinct two-layer placental tissue appears like a process of sedimentation. The darker part on the right corresponds to the homogenous, hyperechogenic structure seen on sonography (Fig. 1) and the left part corresponds to the smaller hypoechoic, inhomogeneous structure. The severely growth-restricted fetus can be identified on the left. The elongation of the cervix (arrows) starting from the internal os (asterisk) is visible behind the symphysis.

The patient and her partner were counselled in detail about the unclear placental pathology and the uncertain but most probably infaust fetal prognosis. An attempt at mechanical repositioning of the uterus was offered. The patient received intravenous analgesia before the procedure. The repositioning took place in an examination room without any special fetal or maternal monitoring. Digital-vaginal repositioning of the uterus was successfully performed after rectal filling with ultrasound gel in a head-deep-knee-elbow position (300 ml of ultrasound gel seemed sufficient). After the repositioning, the patient was free of complaints. Ultrasound showed no change in the fetal situation.

The following day an intrauterine fetal death was diagnosed and the patient developed endogenous labour. She had an uneventful vaginal birth. Due to placental retention, manual extraction was performed. According to the local clinic standard for cases before the 20th week of pregnancy, curettage with a blunt curette was subsequently performed.

Fetal autopsy showed a growth-restricted female fetus of 48 g with a crown-crump length of 12 cm. Fetal anatomy was normal.

In the histopathologic examination of the placenta, a broken basal plate with intraparenchymatous bleeding and several zones of infarction were found, as was a blood clot in the vicinity of the umbilical cord without direct association with the umbilical cord vessels. Moreover, no evidence of a chorangioma or other tumor could be found. In summary, the histopathologist suspected a chronic placental insufficiency and considered premature placental abruption as possible.

Molecular genetics of the placental tissue revealed female triploidy (XXX) (Fig. 3).

3. Discussion

Incarceration of the gravid uterus is rare, occurring in 1 out of 3000 pregnancies [1]. The main risk factor is uterine retroversion. Diagnosis is usually made in the first or second trimester, but there are also cases in which the incarceration was not detected before delivery [2–4].

Symptoms range from abdominal pain or back pain to disturbed urination or defaecation, urinary retention and urinary congestion to vaginal bleeding. Some authors report fetal growth restriction, which is attributed to the compression of the uterine arteries.

Abdominal ultrasound seems to be superior to transvaginal sonography because of the better presentation of the cervical course and the localization of the os internum. If the placenta is located on the posterior uterine wall it can be mistaken for placenta praevia totalis [5]. For more precise diagnosis, ultrasound can be supplemented by MRI.

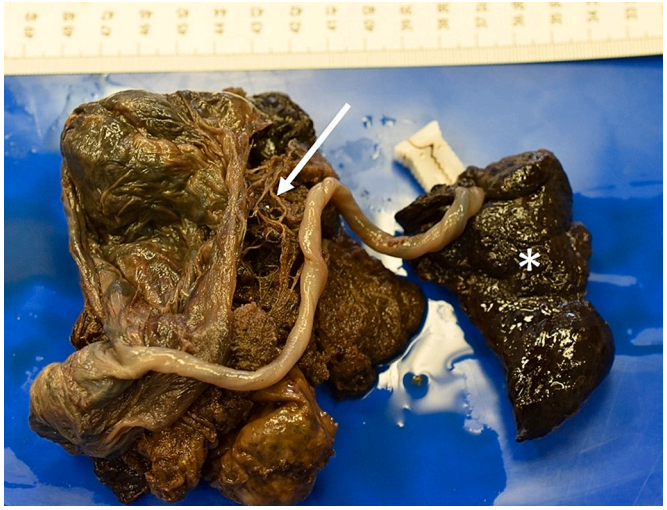

Fig. 3. Formalin-fixed placenta with a postnatal separated coagula (asterisk), which caused a partial detachment of the chorionic plate, leaving the small fetal placental vessels uncovered (arrow).

Many techniques for repositioning are described in literature. A precondition for all of them is initial emptying of the urinary bladder. In cases of intolerable pain, epidural anesthesia may be considered before repositioning is attempted [6]. In addition to the digital-rectal repositioning in a deep-knee-elbow position, successfully performed in the present case, some authors have described, for example, repositioning after vaginal manipulation with pressure on the dorsal fornix or after amnioreduction to reduce the uterus volume after repositioning [7]. Surgical repositioning after laparoscopy or laparotomy can be considered in the last resort if other manoeuvres are unsuccessful [8].

Vaginal delivery in patients with uterus incarceration is mechanically impossible. If the diagnosis is made just before delivery, caesarean section should be performed. If the specific anatomy is unknown, opening the supposedly lower uterine segment accidently opens the ventral and dorsal vaginal wall. The urinary bladder might also be injured.

Probably, there is a certain risk of recurrence in the same patient [9].

In the present case, fetal triploidy occurred as a concurrent pathology. Its incidence is estimated to be 2% of all conceptions [10]. However, between 16 and 20 weeks of gestation, the incidence of triploidy decreases to only 0,002%, as most pregnancies end in early miscarriage [11]. In paternal triploidy the placental structure is typically abnormal (hydropic, vacuolated and with a cloudy echogenic pattern). Usually, the fetuses have numerous anatomic abnormalities but can also develop normally [11]. When the additional chromosomal set comes from the mother, there is typically fetal growth restriction and the placenta is small. In 69XXX karyotype, as in the present case, the additional chromosomal information can either be maternal or paternal – unfortunately further molecular genetic testing for definitive diagnosis was not performed. Based on the sonographic findings, it is not possible to make a clear determination as there are signs for both diandric (abnormal placenta) and digynic triploidy (severe growth restriction). However, the placenta in the present case did not show typical molar or hydropic features; instead, the placenta was enlarged and showed an unusual two-part structure. In the histopathologic examination there were also no indications for a partial hydatiform mole. Therefore, we considered the risk for gestational trophoblast neoplasia in this case to be extremely low.

4. Conclusion

In the present case the simultaneous presence of two different pathologies made diagnosis difficult. In addition to the pathologically enlarged placenta with severe fetal growth restriction, the patient suffered from uterine incarceration.

It is questionable whether there was a causal relationship between the two pathologies. A rapid increase in the volume of the uterus, e.g. through intraplacental haemorrhage, may explain the failure of the uterus to straighten up. The changes in the seemingly two-layered placenta could be explained after birth as an intraplacental bleeding within the chorionic plate.

Contributors

Judith Sarah Abel contributed to patient care, drafting the manuscript and revising the article critically for important intellectual content.

Bernd Morgenstern contributed to patient care, drafting the manuscript and revising the article critically for important intellectual content.

Both authors approved the final submitted manuscript.

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Patient consent

Obtained.

Provenance and peer review

This article was not commissioned and was peer reviewed.

Conflict of interest statement

The authors declare that they have no conflict of interest regarding the publication of this case report.

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