



## Case report

## Spontaneous twin pregnancy in uterus bicornis unicollis complicated with preeclampsia: A case report

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## ABSTRACT

**Introduction and importance:** Bicornuate uterus is a uterine malformation caused by abnormal Müllerian fusion, often leading to multiple obstetric complications. A twin pregnancy in this condition is extremely rare.

**Case presentation:** A 27-year-old patient (Gravida 3, Para 0) with a previous history of two spontaneous early miscarriages was diagnosed with uterus bicornis unicollis (unicervical bicornuate uterus). She presented with spontaneous twin pregnancy complicated with preeclampsia at 34 weeks + 6 days. Thus, she underwent cesarean section with two separate lower-segment incisions. Consequently, healthy twins were delivered successfully.

**Clinical discussion:** Bicornuate uterus is a rare congenital uterine malformation, and only 16 cases of twin pregnancy associated with uterus bicornis unicollis have been reported worldwide. This uterine malformation can be discovered spontaneously in imaging or during obstetric complications such as spontaneous abortions, premature labor, premature rupture of membranes, malpresentations, and intrauterine growth restriction.

In addition, the risk for preeclampsia in twin pregnancies is twice higher than singleton gestations.

**Conclusion:** Twin pregnancy in a bicornuate uterus is extremely rare; this phenomenon is yet to have guidelines for monitoring pregnancy and the mode of delivery.

## 1. Introduction

Congenital malformations of the female genital tract may involve the fallopian tubes, uterus, cervix, and vagina [1].

However, uterine malformations are the most common Müllerian abnormalities. They are associated with multiple obstetric complications, such as early and late abortions, preterm deliveries, malpresentations, pregnancy vascular pathologies, and intrauterine growth restriction.

Herein, we report an extremely rare case of a twin pregnancy in a unicervical bicornuate uterus (otherwise known as uterus bicornis unicollis) complicated with preeclampsia and a brief review of literature.

This work has been reported in line with the modified SCARE checklist, based on updated guidelines published in 2020 [2,4].

## 2. Case report

A 27-year-old patient (Gravida 3, Para 0) with no history of smoking or alcohol consumption, no chronic illnesses or family history of

congenital abnormality, and a previous history of two spontaneous early miscarriages, was diagnosed with uterus bicornis unicollis using pelvic magnetic resonance imaging (MRI) after the first miscarriage (Fig. 1). At 7 weeks of spontaneous twin pregnancy, she sought consultation for metrorrhagia with pelvic pain. Gynecological examination found a single cervix with minimal metrorrhagia.

First ultrasound examination showed a diamniotic dichorionic pregnancy; each gestational sac was located in a separate uterine cavity.

The parturient was prescribed 400 mg of progesterone per day with strict rest. At 12 weeks, she underwent cervical cerclage with non-absorbable sutures.

Fetal growth and cervical length were monitored and remained normal in regular ultrasound examinations.

At 34 weeks + 6 days, she manifested episodes of headache and was subsequently diagnosed with preeclampsia, thereby administered with methyl dopa, an antihypertensive drug.

Laboratory results were as follows: 24 h urine protein test, 600 mg; hemoglobin, 10 g/dl; platelet count, 250,000/mm<sup>3</sup>; aspartate amino transferase, 29 IU; alanine amino transferase, 24 IU; lactate

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dehydrogenase, 230 IU; total bilirubin, 10 mg/l; conjugated bilirubin, 1 mg/l; urea, 0.15 g/l; creatinine, 6 mg; total protein, 100%; and tricyclic antidepressant urine test, 30 s.

Moreover, the twin in the left horn showed intrauterine growth restriction via ultrasound, with normal fetal Doppler, amniotic fluid, and abnormal uterine Doppler with diastolic notches.

Unfortunately, headache and high blood pressure persisted despite receiving antihypertensive treatment. Hence, at 35 weeks, cesarean section was performed.

The intervention was performed by an experienced gynecological surgeon. The patient was administered spinal anesthesia, and two separate uterine horns were intraoperatively observed without any communication. By two separate lower segment Cesarean sections, the twins were successfully delivered (Figs. 2 and 3). Two placentas were observed, each in its respective horn. Birth weights were 2400 and 2100 g. The Apgar scores were both 10 and 10 at 1 and 5 min, respectively. These premature newborns were then admitted in the neonatal department, with good fetal evaluation. Maternal outcomes were also uneventful.

The sutures placed during transvaginal cervical cerclage were removed after the cesarean section.

The patient spent 3 days in the postnatal unit. The postoperative period was uneventful and the patient was discharged with satisfaction.

### 3. Discussion

Uterus bicornis unicollis is a congenital uterine malformation that usually results from an incomplete fusion of the Müllerian ducts to varying degrees of separation between the cavities [1].

Currently, the prevalence of bicornuate uterus is only 0.4% [3]. Twin pregnancies in a bicornuate uterus are extremely rare. Given its very low incidence, only few cases have been reported [4].

In a systematic review, only 16 cases of twin pregnancy associated with a bicornuate uterus were reported [5,6], and most of them were not spontaneous pregnancies.

Obstetric performance is related to bifurcation depth [7], influencing principally the maintenance of pregnancy without impacting conception [8]. This association is explained by several theories, such as decreased muscle mass, abnormal uterine blood flow, and cervical insufficiency [4].

This uterine malformation can be discovered spontaneously during radiological examinations (pelvic ultrasound, pelvic computed tomography scan, pelvic MRI, etc.) or during obstetric complications, including spontaneous abortions, premature labor, premature rupture of

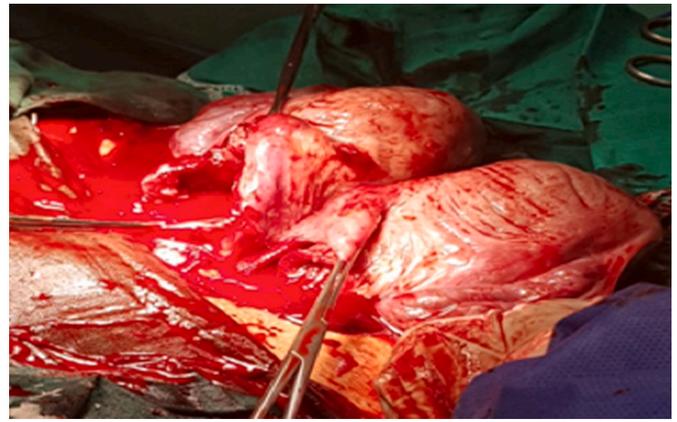


Fig. 2. Two incisions repaired separately during cesarean section.



Fig. 3. Final hemostatic sutures.

membranes, malpresentations, and intrauterine growth restriction [7].

Although preventive cervical cerclage has been associated with poor reproductive outcomes in uterine malformations and despite no apparent cervical anomalies, we still performed this procedure to our patient. Various researchers have postulated an abnormal ratio of muscle fibers to connective tissue in the cervix associated with Müllerian abnormalities [8].

This type of pregnancy still has no definite management, and vaginal

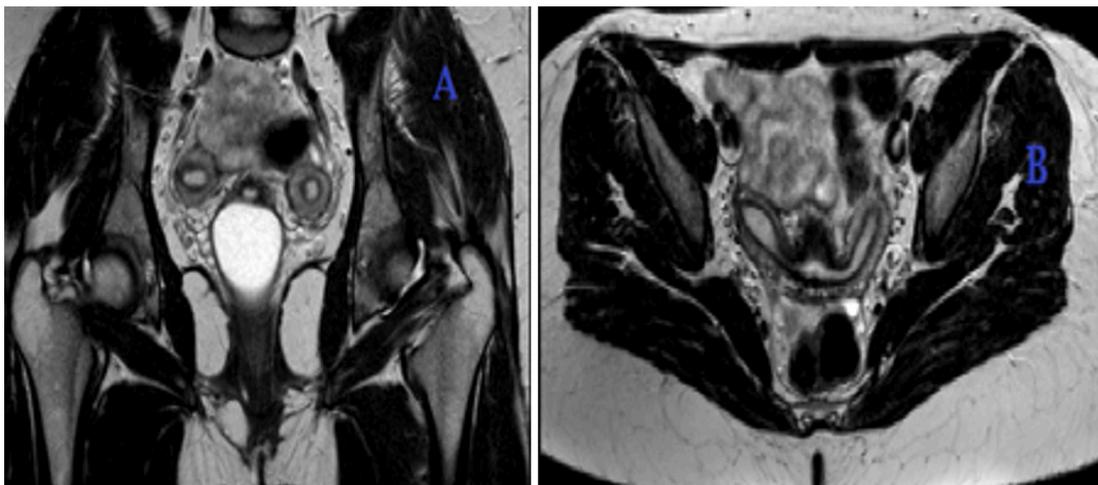


Fig. 1. View of the uterus in magnetic resonance imaging (MRI). A) Coronal T2-weighted MR image, B) axial T2-weighted MR image.

delivery is presently not contraindicated [7], although the 16 reported cases underwent cesarean section.

Preeclampsia is a hypertensive disease occurring during pregnancy, complicating 3%–5% of pregnancies worldwide. In fact, the risk of developing preeclampsia in twin pregnancies is twice higher than in singleton gestations [9]. In the present case, our patient underwent cesarean section because of unresolved severe preeclampsia.

Arora et al. reported a similar case of a twin pregnancy in a bicornuate uterus complicated with preeclampsia; likewise, the patient underwent cesarean section. However, they mentioned that vaginal delivery was also possible [4].

Furthermore, Doruk et al. [8] reported a case of a twin pregnancy in a bicornuate uterus delivered by cesarean section. They also concluded that cesarean section is preferable to avoid obstetric complications, including malpresentations and uterine rupture [8]. Other similar cases of twin pregnancy in a bicornuate uterus with cesarean section delivery have been reported [1,6,5]. In our case, twin pregnancy in a bicornuate uterus complicated with preeclampsia delivered successfully via cesarean section.

The Cesarean section was performed using the Pfannenstiel incision, the peculiarity in this case, i.e., the two separate lower segment Cesarean sections and the double hysterorrhaphies, will increase the duration of the operation, Doruk et al. [8] reported a similar case, intraoperatively. Each incision was repaired simultaneously by two different operators, and the aim was to prevent complications during delivery.

#### 4. Conclusion

Twin pregnancy in a bicornuate uterus is an extremely rare phenomenon. Ultrasound and MRI play an important role in the diagnosis. Serial ultrasound every two or three weeks is a feasible option to assess fetal growth and measure cervical length, for determining the risk of premature delivery. However, no guidelines for monitoring pregnancy and the mode of delivery are available.

#### Availability of data and material

The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

#### Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the consent form is available for review by the Editor-in-Chief of this journal on request.

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#### CRediT authorship contribution statement

Chaymae Chemlal, study concept and design, data collection, data analysis and interpretation, writing the paper.

Ahmed Mimouni; Hanane Saadi: data collection, supervision and final approval.

All authors read and approved the final manuscript.

#### Declaration of competing interest

The authors declare that they have no competing interests.

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