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Case Report

Bicornis unicollis uterus as a risk factor of preterm birth: A case of young woman with multiple premature births

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Abstract

Bicornis unicollis uterus is a rare congenital uterine abnormality that occurs due to the failure of Mullerian duct fusion early in the development of the female internal genitalia system. In this case report, we present a woman with bicornis unicollis uterus who had preterm birth. A 30-year-old female patient with two caesarean sections history with premature babies was presented to the hospital with a complaint of regular contractions for twelve hours in her third preterm pregnancy. The patient has no particular symptoms besides acute abdominal pain. The ultrasonography examination indicated a uterus didelphys with breech presentation fetus. Due to the patient's caesarean history and the fetal presentation, an emergency caesarean section was decided and performed. It was found that the gravid uterus was on the left and the baby was subsequently delivered with a complete placenta. Postoperative condition of the patient was shown to be stable while the baby underwent an intensive care at the neonatal intensive care unit. This case report highlights that early diagnosis in this rare case is critical since bicornis unicollis uterus are mostly asymptomatic. Caesarean section was chosen in the present case based on consideration of the fetal and maternal clinical conditions.

Keywords: Uterine abnormality, Mullerian ductus abnormality, uterus bicornis unicollis, preterm birth, premature

Introduction

Uterus bicornis unicollis is an extremely rare condition and the most common type of Mullerian ductus abnormality. It is a type of uterus didelphys disorder which is a congenital abnormality that occurs due to failure of the Mullerian duct fusion during the development of the female internal genitalia system [1,2]. The complete abnormality is characterized by two corpora uteri and two cervixes separated by a longitudinal vaginal septum, and many other variations of this uterine abnormality have been reported [3].

Uterine abnormalities including uterus bicornis unicollis affect 0.5% to 5.0% of the general population [2]. Uterine abnormalities are usually asymptomatic and the incidence is not widely known in general public [4-6]. Its diagnosis can be initiated by pelvic examination and supported by ultrasound, magnetic resonance imaging (MRI), sonohysterography or hysterosalpingography examinations [3,5,7-9]. Pregnancy with bicornis unicollis uterus is at high risk and causing various maternal and perinatal consequences [10,11]. These risks should be prevented to reduce maternal and infant mortality. The aim of this case report was to present a case of bicornis unicollis uterus in a 30-year-old female that had preterm birth.

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Case

A 30-year-old female presented to Dr. Zainoel Abidin Hospital, Banda Aceh, Indonesia with chief complaint of regular contractions for twelve hours before being admitted to the hospital. Blood from the birth canal was reported. Fetal movements were active, and the patient had a gestational age was 33 weeks six days. The patient had antenatal care (ANC) three times, with the result the last ultrasonography result before being presented to the hospital indicated the fetus was in good condition but had breech presentation. The bicornis unicollis uterus was found later during a subsequent ultrasonography examination.

The patient did not have a history of vaginal discharge. The present pregnancy was the third pregnancy, with the previous two had cesarean sections and resulting in premature births. Defecation and micturition were within normal limits. The patient had no history of allergies, and neither the patient nor the family had a history of hypertension, heart disease, diabetes mellitus, or bronchial asthma. The patient had not taken any medication except for vitamins during the pregnancy. The patient denied of smoking and drinking alcohol.

Vaginal examination found that cervix dilation of six cm, negative amniotic membrane, 75% effacement, breech presentation, and sacrum in Hodge II-III. Transabdominal ultrasonography results indicated singleton live breech presentation, fetal heart rate (FHR) was positive, biparietal diameter (BPD) 8.05 cm, head circumference (HC) 29.61 cm, abdominal circumference (AC) 29.38 cm, femur length (FL) 5.52 cm, estimated fetal weight (EFW) 1769 g, and amniotic fluid index (AFI) 3.96 cm. The placenta was located in the anterior corpus of the uterus. Laboratory results are presented in **Table 1**.

Table 1. Laboratory results of the patient during hospitalization

Parameter	Result
Hemoglobin	9.7 g/dL
Hematocrit	30 %
Red blood cells	4.4 million/µL
Platelets	292,000/µL
White blood cells	13,920/µL
Mean corpuscular volume (MCV)	69 fL
Mean corpuscular hemoglobin (MCH)	22 pg
Mean corpuscular hemoglobin concentration (MCHC)	32 g/dL
Differential blood count	
Basophils	0%
Eosinophils	0%
Band neutrophils	1%
Segmented neutrophils	82%
Lymphocytes	11%
Monocytes	6%
Kidney function test	
Urea	11 mg/dL
Creatinine	0.5 mg/dL
Blood sugar level	91 mg/dL
Hemostasis	
Prothrombin time (PT)	12.20 second
Activated partial thromboplastin time (APTT)	27.40 second
Hepatitis B surface antigen (HBsAg)	Non-reactive
Liver function test	
Serum Glutamic Oxaloacetic Transaminase (SGOT)	16 U/L
Serum Glutamic Pyruvic Transaminase (SGPT)	5 U/L
Electrolytes	
Sodium (Na)	142 mmol/L
Potassium (K)	3.7 mmol/L
Chloride (Cl)	113 mmol/L

Due to the patient's condition, an emergency caesarean section was performed. Intraoperatively, after the peritoneum was opened, a double uterus (uterus didelphys) was observed, with the uterus on the right was asymmetrical in size, while the gravid uterus was on the left. The lower segment of the uterus was then incised semilunarly and bluntly extended laterally. The amniotic membranes were ruptured, and a small amount of clear amniotic fluid was obtained. The baby was delivered in breech presentation with a body weight of 1900 gram, body

length of 40 cm, and Apgar score of 6/8. The umbilical cord was gently retracted, and the placenta was delivered with complete cotyledons. The shape of the uterus was found to have two horns that merged at the bottom of the uterus confirming uterus bicornis unicollis. Both tubes and ovaries were within normal limits. The lower segment of the uterus was sutured in two layers and there was no active bleeding. Abdominal cavity was washed and abdomen was then closed layer by layer.

The patient condition during and after surgery was stable. Right after birth, the baby was treated in neonatal intensive care unit (NICU) with a diagnosis of respiratory distress syndrome, infection and low birth weight. Treatments were initiated and improved the condition of the baby. There was no sign of post-cesarean bleeding due to bad uterus contraction. Four days post-surgery the patient was discharged. A week after post-birth delivery, the patient was presented to the policlinic in a stable condition and the surgical sutures were removed.

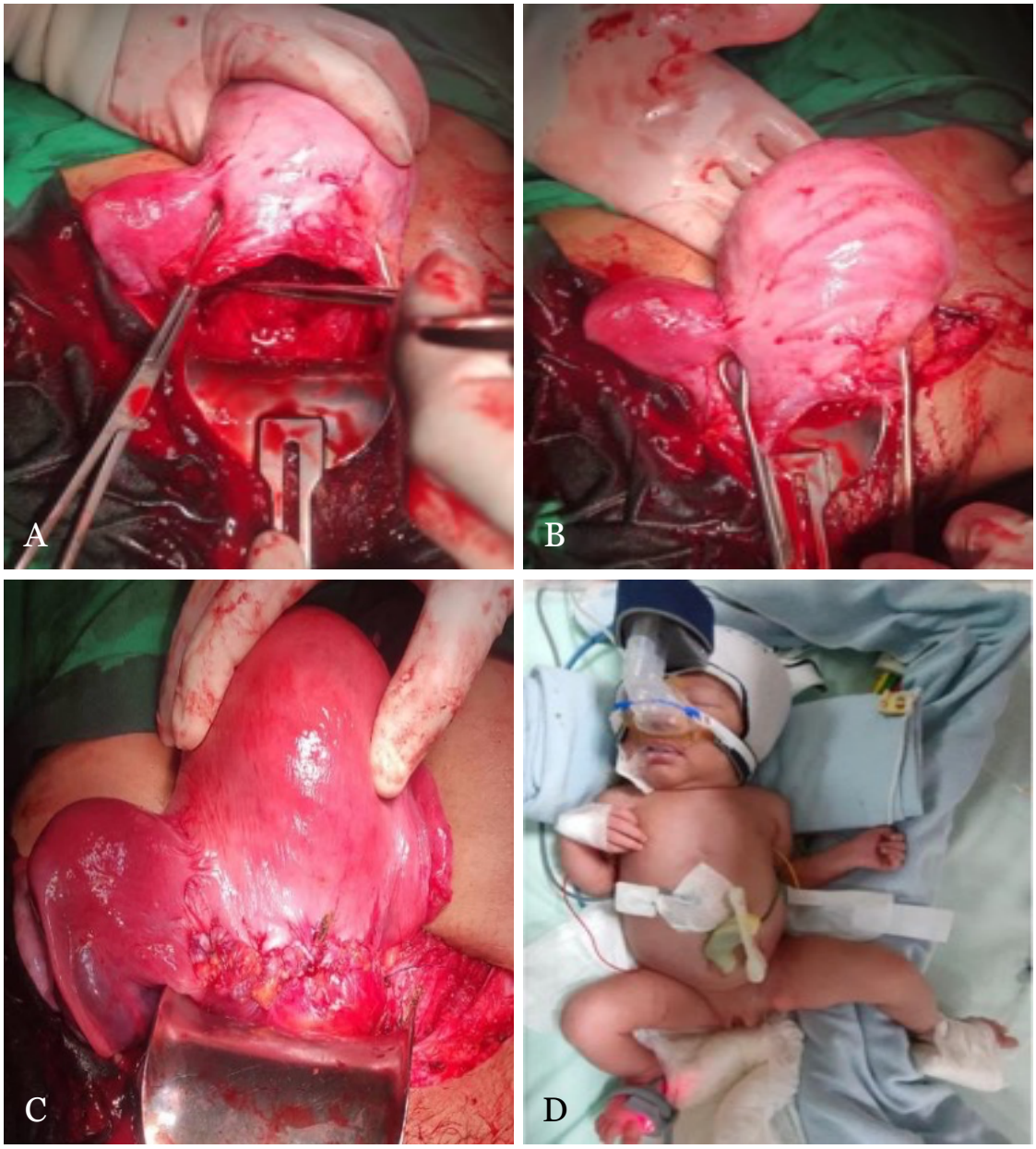

Figure 1. Presentation of bicornis unicollis uterus and the baby during post-surgery. Bicornuate uterus with one cervix (A); uterus didelphys on the right (B); gravid uterus on the left (C); premature baby installed with C-Peep in NICU (D).

Discussion

Uterus bicornis unicollis is a rare Mullerian ductus abnormality. It occurs when the midline Mullerian duct fusion is terminated, either completely or partially. Approximately 11% of uterine malformations are uterine didelphys, which includes uterus bicornis unicollis [12]. The complete form is characterized by two hemiuteri and two endocervical canals with cervix fused in the lower uterine segment. Each hemiuterus is associated with one fallopian tube [13,14]. Multiple malpositions of the ovaries and vagina can also occur [15]. Double vaginal manifestations include a longitudinal septum extending, completely or partially from cervix to introitus [16,17,18]. In 75% of these anomalies, complete longitudinal vaginal septum is present, although vaginal septa may also occur with other Mullerian duct anomalies [19,20].

In Indonesia, there is no available statistical data regarding pregnancies with bicornuate uterus specific suggesting its very rare incidence. According to a previous study, congenital uterine malformations may occur in up to 4.3% of women [21]. Patients with uterus bicornis unicollis who reach term have a probability of about 1 in 5 [2,21]. Pregnancy with uterus bicornis unicollis can cause various maternal and perinatal outcomes, including miscarriage (21%), prematurity (24%), ectopic pregnancy (2%), intrauterine growth restriction (11%), and an increased incidence of caesarean section (84%) [1]. The fertility of women with untreated uterus bicornis unicollis is better than women with other Müllerian duct disorders [6]. Risk factors for uterus bicornis unicollis include an increased risk of spontaneous abortion, fetal growth retardation, and prematurity [10,22]. The rate of preterm birth is higher compared to other abnormalities, such as septate and bicornuate uterus [14]. Some literature state that uterine didelphys increases the risk of preterm birth [21,23,24]. However, this is primarily because most patients with uterine didelphys are not detected in primigravida and multigravida patients.

Preventive measures and accurate diagnosis are crucial. Differential diagnosis is important for initial treatment. Uterus bicornis unicollis may mimic other congenital malformations during physical examination, particularly the septate or bicornuate uterus. Several key differences can help differentiate uterus bicornis unicollis from the latter two conditions. The septate uterus has normal fundal contours but contains a persistent longitudinal septum that may partially or completely divide the uterine cavity. On the other hand, the bicornuate uterus results from the failure of some ducts to unite, leading to the separation of one uterus into two horns. Similar to septate uterus, bicornuate uterus can be classified as complete or partial and can further be categorized as bicollis and unicollis [21]. In this patient's case, the shape of the uterus was found with two horns that merged at the bottom of the uterus, thus confirming that the patient possesses uterus bicornis unicollis.

Preventive measures for uterus bicornis unicollis cases cannot correct the congenital abnormality but can optimize health quality of life through education, physical examinations, routine ANC, and regular monitoring to prevent early and advanced complications in both mother and baby [1,3,4]. A study found that uterine didelphys does not necessitate a cesarean section, instead fetal presentation does [14]. In uterine didelphys, management of childbirth can be done either by vaginally or through cesarean section [6,25]. However, this condition is associated with fetal breech presentation and is the most common indication for cesarean delivery in women with uterine didelphys [1,4,13,21,26] Furthermore, if no coexistent symptom present, additional treatment to uterine disorder is deemed unnecessary [6]. In this present case, the patient showed no particular symptom prior and was decided to be given an emergency cesarean section based on her history with two previous cesarean deliveries and the fetus' breech presentation as seen in the ultrasonography examination imagery.

Conclusion

Uterus bicornis unicollis is a very rare case. The diagnosis can be made preoperatively using transvaginal ultrasonography as the main modality, but it is mostly diagnosed intraoperatively. Symptoms are not typical in many cases, and more than half of the cases are asymptomatic. Birth delivery can be performed either vaginally or through caesarean section. In present case, due to the fetus's breech presentation and the mother's history of two previous caesarean sections, caesarean section was chosen.

Ethics approval

The patient provided written informed consent to be published as a case report.

Competing interests

The authors declare that there is no conflict of interest.

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Underlying data

All data underlying the results is available as part of the article and no additional source data are required.

How to cite

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