

DOI: <https://dx.doi.org/10.18203/2320-1770.ijrcog20240813>

Case Report

Operative hysteroscopy assisted evacuation of a non-viable pregnancy in the setting of a congenital uterine anomaly - bicornuate bicollis

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Received: 12 January 2024

Revised: 01 March 2024

Accepted: 02 March 2024

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ABSTRACT

The uterus is formed during embryogenesis by the fusion of the two paramesonephric ducts (mullerian ducts). The two mullerian ducts normally fuse to form the single uterine body. A didelphis uterus will have a double cervix and a double vagina. This case report describes the successful removal of retained products of conception following a spontaneous miscarriage in the setting of a bicornuate bicollis anomaly with a non-viable pregnancy using operative hysteroscopy. The successful removal of the retained products of conception is an essential step in the management of a spontaneous miscarriage. Gynaecologists should be aware of the potential complications associated with this condition and refer patients with a diagnosis of uterine didelphys to an expert in gynaecology for further evaluation and management.

Keywords: Uterine anomaly, Didelphys uterus, Uterus bicornisbicollis, Miscarriage, Hysteroscopy

INTRODUCTION

The development of the internal genital parts of the female reproductive system depends on the Müllerian, or paramesonephric, ducts. In females, the Müllerian ducts eventually give rise to the uterus, cervix, fallopian tubes, and the proximal portion of the vagina. The Müllerian ducts are initially seen in both sexes and are characterized by their appearance as paired channels that stretch inferiorly down the lateral face of the embryo to the sinus tubercle of the urogenital sinus. Under the influence of anti-Müllerian hormone (AMH), sometimes referred to as Müllerian inhibiting substance (MIS), Müllerian inhibiting hormone (MIH), or anti-Müllerian factor (AMF), these ducts usually recede in males. The Sertoli cells of the testis generates the anti-Müllerian hormone which inhibits the development of female genital tract. The Müllerian or paramesonephric ducts will grow into the uterus, uterine tubes, cervix, and superior 1/3 of the vagina in either sex if AMH is not present. Various developmental abnormalities of the female internal genitalia, such as

agenesis of the uterus or production of a unicornuate, bicornuate, didelphic, septate, or arcuate uterus, can be caused by disruption of these systems.

When the Müllerian ducts partially fuse at the fundus level, the result is a bicornuate uterus. It is identified by the existence of a cleft larger than 1 cm in the uterine fundus's exterior shape. Septum length determines whether a patient is placed in a whole or partial category. Bicornuate unicollis uterus and bicornuate bicollis uterus are terms used to describe complete uterine septa that extend to the internal or external cervical os, respectively. If the septum is limited to the fundal area, the uterus is regarded as somewhat bicornuate. This anomaly is often associated with infertility, miscarriage, and preterm delivery.

CASE REPORT

A 26-year-old primigravida married for 8 months presented with a history of amenorrhea for 3 months. She had done urine pregnancy test one month back at home

which was positive. She came to tertiary care hospital with chief complaints of bleeding per vagina for the past two days. She was corresponding to gestational age of 12 weeks according to her LMP. It was followed by the onset of foul-smelling, brownish-red vaginal bleeding with clots. The patient also reported abdominal pain. Her previous menstrual cycles were regular, 4-5 days duration, with moderate flow. History of typhoid in 2 months back, medically managed. She had conceived naturally. On examination her general condition was stable. She was afebrile and no history of fever. Her vitals were within normal limits with a pulse rate of 100/min and blood pressure of 110/74 mmHg. And other systemic examination was within normal limits. On pelvic examination two cervical openings were observed with minimal foul-smelling brownish discharge. Uterus was bulky corresponding to 10 weeks gestation size. An endovaginal ultrasound examination was done to assess the uterine anomaly and to confirm the presence of retained products of conception. Ultrasonography (USG) was suggestive of gravid bicornuate bicollis uterus with gestational sac in the right horn. Crown-rump length (CRL) corresponding to 9+1 weeks of gestation. Absent cardiac activity noted.

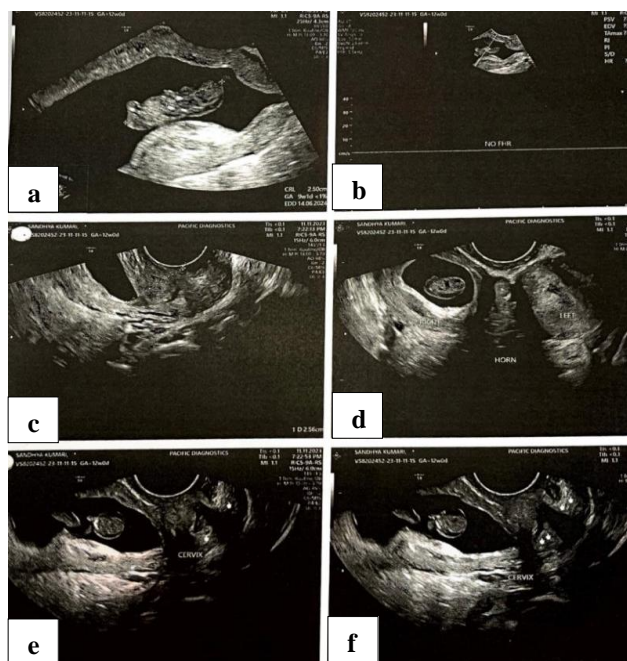


Figure 1: (a-f) Pelvic USG reveals a gravid bicornuate uterus with well-defined gestational sac in the right horn with no evidence of cardiac activity.

The patient was counselled regarding the risks and benefits of a diagnostic hysteroscopy and dilatation and evacuation (D&E) for removal of the retained products of conception. The procedure was performed under general anaesthesia. An attempt was made to evacuate the products of conception by doing manual dilation and evacuation. But it was unsuccessful as it was difficult to dilate the cervix of the gravid horn respectively. So, hysteroscopy with dilatation and evacuation of the non-viable pregnancy was

done. Upon visualization of the uterine cavity, it was noted to be divided into two separate horns, consistent with the diagnosis of bicornuate uterus with two cervixes. Intraoperative findings included right os: one ostium seen with retained products of conception (RPOC) at the fundus. Through left os: normal cavity with one ostium seen. The retained products of conception were successfully removed using a combination of hysteroscopy and manual evacuation techniques. The patient tolerated the procedure well and was discharged home the same day.

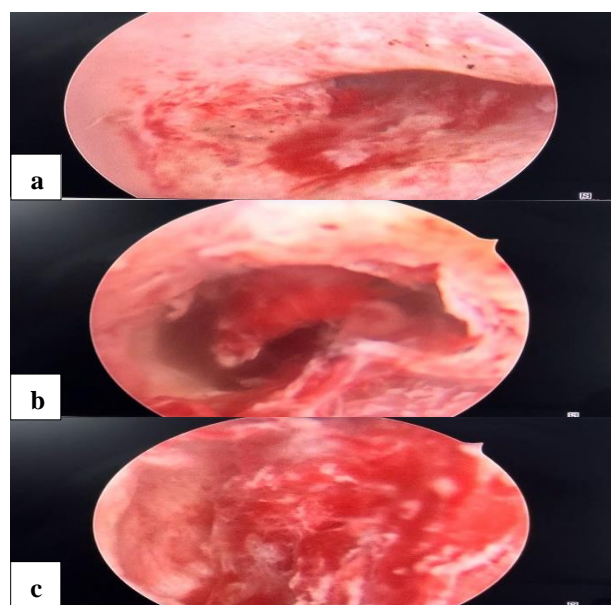


Figure 2: (a-c) Hysteroscopy view of right horn with products of conception.

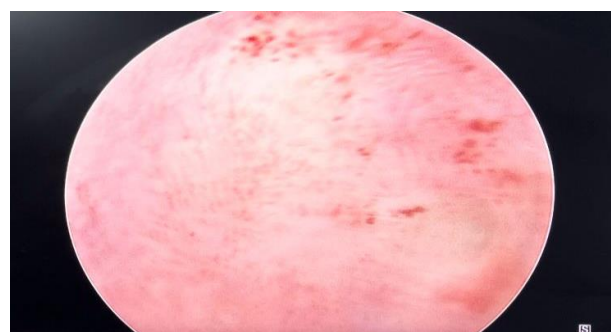


Figure 3: Hysteroscopy view of the left horn of uterus with normal uterine cavity.

DISCUSSION

Bicornuate uterus derives from deficient fusion of Müllerian ducts during fetal development; in this case, the two cavities are entirely or partially unified by caudal fusion.¹ Both endometrial cavities frequently open to a single vagina via a single uterine cervix (unicollis) or via separate uterine cervixes in rare cases (bicollis).² Bicornuate uterus competes with arcuate uterus for the place of third most frequent congenital Müllerian malformation in unselected populations (0.4%).³ It is often

asymptomatic and remains unidentified before puberty, showing a significant correlation with infertility and miscarriage.³

The rate of the premature rupture of membranes (PROM), preterm separation of the placenta, miscarriage, premature delivery, and intrauterine growth restriction (IUGR) is higher in cases of bicornuate uterus.⁴ Bicornuate uterus significantly contributes to uterine ruptures in first pregnancy patients and at any gestational age, as well as cervical incompetence, which can significantly increase the perception of birth risk.⁴ However, there are several reports of successful gestations involving bicornuate uterus patients. Prognosis remains debatable, since pregnancy may be compromised by cervical atresia, cervical mucus absence, upper congenital anomalies, and recurrence of the anomaly after cervical corrective surgery, and postoperative retrograde adhesions.⁵ Nevertheless, pregnancy may be possible after surgical corrections through natural or artificial insemination. In a rare case, a successful full-term twin pregnancy in each cavity of a bicornuate septate uterus of a patient with two cervixes and a longitudinal vaginal septum via natural insemination was published. Furthermore, a woman with bicornuate unicollis uterus with twins successfully delivered at 35 weeks of gestation through a bilateral caesarean section.¹ In addition, several cases of successful pregnancies of women with a bicornuate uterus have been reported without surgical correction of the anomaly. To our knowledge, surgical corrections or IVF procedures have not been reported in women with a bicornuate uterus.

A similar case was published by Ryu et al with ultrasonography suggestive of a missed abortion in a left-sided hemicavity of the septate uterus.⁶ Ultrasound-guided D&E was attempted but did not work as the gestational sac was blocking the curette's path into the uterus. Insufficient connectivity with the left-sided cavity right above the cervical internal os of the uterine septum was discovered during an operational hysteroscopy. Following increased communication, ultrasound-guided D&E was carried out effectively.⁶ In another case report published by Setchell et al, flexible hysteroscopy, in combination with trans-abdominal ultrasound, was used to facilitate the correct passage of the dilators during a successful dilatation and evacuation (D&E) followed by insertion of intra-uterine progestogen-only contraceptive system ('Mirena').⁷

CONCLUSION

One cannot deny that pregnancies of women with Müllerian anomalies associate with potential obstetric

complications. Hysteroscopy and manual evacuation combined can be used for evacuation. Considering the rare occurrence of such cases and the potential contributing risks, pregnancies of women with a bicornuate uterus, and twin pregnancies in particular, should be managed carefully, in a tailored fashion.

Funding: No funding sources

Conflict of interest: None declared

Ethical approval: Not required

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Cite this article as: Simpatwar S, Kumari B, Bhardwaj A, Afreen M. Operative hysteroscopy assisted evacuation of a non-viable pregnancy in the setting of a congenital uterine anomaly - bicornuate bicollis. Int J Reprod Contracept Obstet Gynecol 2024;13:1055-7.