Open Access Library Journal
2022, Volume 9, e8566
ISSN Online: 2333-9721
ISSN Print: 2333-9705

Early Abortion of a Spontaneous Twin Pregnancy in a Unicervical Bicornuate Uterus: Accidental Discovery at the Principal Hospital in Dakar, Senegal

Yaye Fatou Oumar Gaye[1*], Mouhamadou Mansour Niang[2], Papa Malick Ngom[1], Mafing AyaSylla[1]

[1]Department of Gynaecology and Obstetrics, Hôpital Principal de Dakar, Dakar, Sénégal
[2]Department of Gynaecology and Obstetrics, Hôpital Institut d'Hygiène Sociale de Dakar, Dakar, Sénégal
Email: *yifigaye@gmail.com

How to cite this paper: Gaye, Y.F.O., Niang, M.M., Ngom, P.M. and AyaSylla, M. (2022) Early Abortion of a Spontaneous Twin Pregnancy in a Unicervical Bicornuate Uterus: Accidental Discovery at the Principal Hospital in Dakar, Senegal. *Open Access Library Journal*, **9**: e8566.
https://doi.org/10.4236/oalib.1108566

Received: March 4, 2022
Accepted: April 16, 2022
Published: April 19, 2022

Abstract

We report the case of a 27-year-old female, 3rd gesture, 2nd pare, with two live children born at term by vaginal delivery, referred for management of an unruptured left ectopic pregnancy. The emergency ultrasound showed two distinct uterine cavities containing a non-embryonic gestational sac of 7 SA on the left and a hyperechoic image on the right. Monitoring of plasma beta HCG kinetics showed no duplication. Medical treatment with misoprostol allowed evacuation of the uterine cavity. An MRI scan was performed and the diagnosis of a unicervical bicornuate uterus with fluid retention on the left was made, which was managed with a second course of misoprostol. A second misoprostol treatment allowed managing the uterus. The control ultrasound confirmed the vacuity of both uterine cavities. Through this case study, we highlight the diagnostic difficulties with the contribution of ultrasound and pelvic magnetic resonance imaging.

Subject Areas

Gynecology & Obstetrics

Keywords

Spontaneous Abortion, Bicornuate Uterus, Pelvic Ultrasound, MRI

1. Introduction

Congenital malformations of the female genital tract, particularly those involving the uterus, are relatively rare, with an estimated frequency of between 0.5

and 4% of women depending on the author [1]. This prevalence is significantly higher in the population of women with a history of recurrent miscarriage (15% to 25%) [1] [2] [3]. Their association with pregnancy would therefore be a source of gravidic complications which are mostly secondary to a decrease in the capacity of the uterine cavity and would be all the more frequent as the pregnancy is multiple. The occurrence of twin pregnancy in a woman with a uterine malformation would therefore be a high-risk obstetrical situation requiring special monitoring.

We report through this observation of a spontaneous twin abortion in a bicornuate uterus, the diagnostic difficulties and the prognostic and therapeutic aspects.

The aim of this publication is to show that a uterine malformation can go unnoticed even in a woman who has already given birth by vaginal delivery and that an abortion in a bicornuate uterus can simulate an ectopic pregnancy.

2. Observation

This is Mrs. O. S., 27 years old, 3rd gesture, 2nd pare with two live children born at term by vaginal delivery who was referred to us for the management of an unruptured left ectopic pregnancy. The onset of symptoms was reported to be one week old, marked by the onset of moderate abdominal pain without associated signs. She also had a secondary amenorrhea of one and a half months. Ultrasound examination at the referral facility showed a 7-week-old non-embryonic left ectopic sac in favour of an unruptured left EP.

On admission, vitals were normal with blood pressure at 10/7 cmHg and pulse at 95 beats per minute. The general condition was preserved with stained mucous membranes and conjunctiva. The abdomen was soft and there was no umbilical cry. On vaginal examination, the cervix was posterior, long and dehiscent, the vaginal cul-de-sacs were free and the fingernail came back clean.

Emergency endovaginal ultrasound revealed two distinct uterine cavities containing a non-embryonic gestational sac of 7SA on the left and a hyperechoic image on the right. The plasma beta HCG assay came back with a level of 74,402 IU/L. Inpatient expectant care was recommended with monitoring of plasma beta HCG kinetics which was 92,283 IU/L at H48 in favour of no duplication.

At D4 of hospitalization, the patient presented metrorrhagia with red blood. The follow-up ultrasound revealed a unicervical bicornuate uterus with a clear egg in the left hemi uterus and a heterogeneous hyperechoic mass in the right cavity in favour of an incomplete abortion (**Figure 1**). Plasma beta HCG was slightly decreased to 83,380 IU/L.

Treatment with misoprostol at a dose of 600 micrograms orally was initiated and had resulted in expulsion of one of the products of conception. This was confirmed by pathological examination and ultrasound which showed a clear egg in the left hemi uterus and an empty right hemi uterus.

On the 7th day after medical treatment, the plasma beta HCG level was 49,004

IU/L.

A second course of 600 micrograms of misoprostol was instituted and resulted in expulsion of the second product of conception. Pelvic ultrasound confirmed the vacuity of both uterine cavities (Figure 2).

Following uterine evacuation, a pelvic MRI confirmed the diagnosis of a unicervical bicornuate uterus with no communication between the two cavities and showed fluid retention on the left (Figure 3).

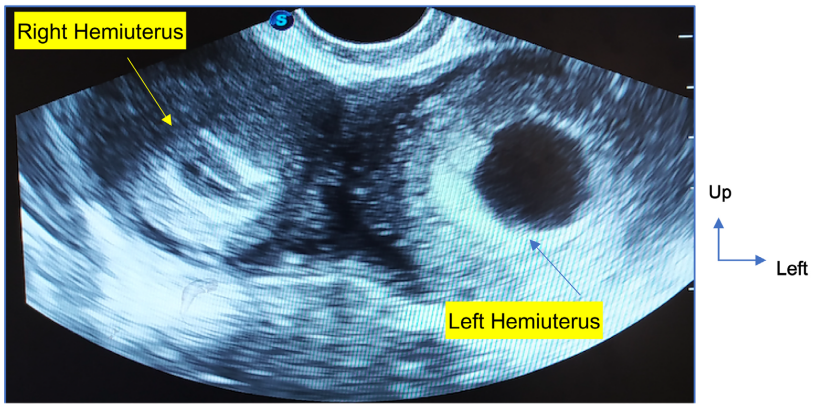

Figure 1. Pelvic ultrasound with two uterine cavities. On the left a clear egg of 7SA and on the right an incomplete abortion (Picture Hôpital Principal de Dakar).

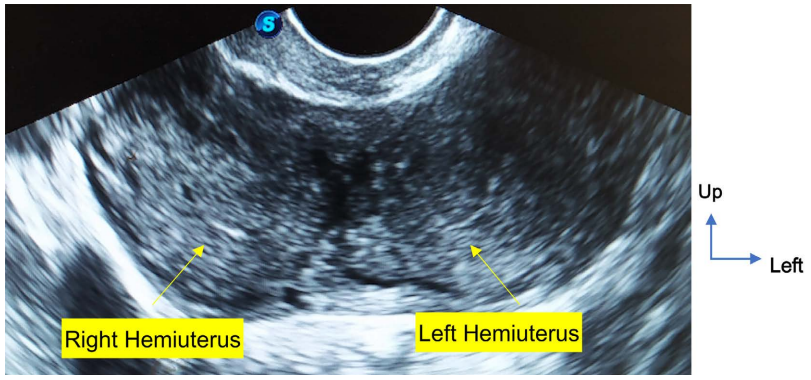

Figure 2. Vacuity of both hemi horns on ultrasound after medical treatment (Picture Hôpital Principal de Dakar).

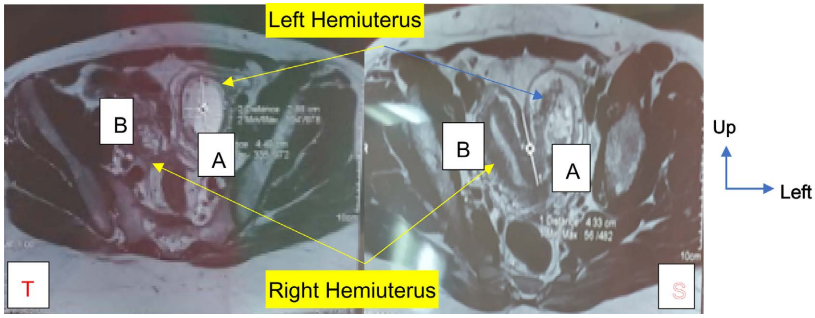

Figure 3. MRI image of bicornuate uterus with trophoblastic retention (A) in the left hemi horn and empty right hemi horn (B), cross section (T); sagittal section (S) (Pictures Hôpital Principal de Dakar).

3. Discussion

According to the European Society of Human Reproduction and Embryology (ESHRE) the unicervical bicornuate uterus corresponds to the partial persistence of the duality of Muller's ducts fused to the lower part of the uterus. There are four types of unicervical bicornuate uterus (the perfect or complete unicervical bicornuate uterus; the simple or partial unicervical bicornuate uterus; the unicervical bicornuate uterus with unequal horns and the uterus with indented fundus named corded uterus). Our patient presented the complete form (classified U3b by ESHRE 2013) with two distinct uterine bodies and isthmuses but which terminated in a single cervix [4].

The bicornuate uterus is the second most common uterine malformation. It accounts for 39% of uterine malformations [4]. Its worldwide frequency, estimated at about 0.4%, is largely underestimated due to asymptomatic forms that escape diagnosis [5] [6]. Although pregnancies with a malformed uterus are prone to obstetric complications, in some situations the pregnancy can be carried to term without complications [7]. Kachhawa *et al*. concluded that women with a bicornuate uterus can expect a favourable pregnancy outcome in 55% - 60% of cases, while 14% will have difficulties with their reproductive health [8].

A previous pregnancy without obstetric incident is often considered a good predictor of subsequent outcome, so that the diagnosis of a major uterine malformation in the next pregnancy is almost always an incidental finding. This was the case with our patient who had two full-term vaginal deliveries.

The case described by Henry Osazuwa *et al*. had a history of vaginal delivery at term [5]. The diagnosis of malformations is thus most often made during the etiological assessment of an obstetrical complication such as spontaneous miscarriage or premature delivery. In the observation of Alkhateeb *et al*., we noted the fortuitous discovery of a uterine malformation following a failed post-abortal treatment in a patient who had already given birth by vaginal delivery at term [2].

In our patient, the first diagnosis of an extra uterine pregnancy was wrongly made, which could have resulted in a laparotomy or laparoscopy. Indeed, one of the uterine horns was mistaken for an ectopic gestational sac. Medical imaging with endovaginal ultrasound and MRI had rectified the diagnosis and confirmed the uterine anomaly. According to the Collège National des Gynécologues et Obstétriciens Français (CNGOF), magnetic resonance imaging (MRI) is currently the gold standard for exploring utero-vaginal anomalies. It is superior to ultrasound, hysteroscopy and hysterosalpingography in the diagnosis of genital malformations [1] [6]. Ultrasound (endovaginal or even three-dimensional) remains a reliable and efficient means of detecting these anomalies during the first trimester of pregnancy for reasons of safety, availability and cost [5] [6] [9].

In the work of Doruk *et al*., the diagnosis of the malformation was made on first trimester ultrasound showing the importance of this examination in prenatal screening and follow-up [3]. Twinning facilitates the ultrasound diagnosis of

bicornuate uterus when each horn has a gestational sac as in Arzu Doruk *et al.* [3]. However, there is a risk of confusion with a heterotopic pregnancy or with an extra uterine pregnancy as in our patient's case.

Uterine defects expose women to a variety of gynaecological and obstetric complications. A systematic review has shown that defects of fusion, to which the bicornuate uterus belongs, do not appear to reduce fertility, but are associated with more frequent pregnancy complications. The bicornuate uterus is associated with a higher risk of early or late, recurrent abortion, preterm delivery and dystocic presentation [7] [10] [11]. The incidence of miscarriage and overall fetal loss in women with a bicornuate uterus has been estimated to be around 30% to 40% [1] [12]. The causes are mainly functional and histological. Indeed, adverse obstetric outcomes are due to inadequate musculature, insufficient uterine expansion capacity, cervical insufficiency, abnormal endometrial development and inadequate vascularisation [1] [3] [12] [13]. The obstetrical performance and gestational capacity of a bicornuate uterus are inversely proportional to the degree of muscle distortion. This result is in a high incidence of abortion and preterm labour [6] [9].

In twin pregnancies, these anatomical and functional defects can lead to even worse outcomes. Twin pregnancies significantly increase the risk of negative reproductive outcomes [11].

The rate of twin pregnancies in women with uterine defects (2.7% - 3.1%) is close to that of the general population (2% - 3%) [14] [15] [16]. This phenomenon occurs following medically assisted reproduction or ovulation induction (38%), but also naturally, spontaneously, as in our patient [14] [17]. Twin pregnancies can develop in the same horn, as described by Adams *et al.* and Narlawar *et al.* [16] [18], or in two separate horns, as in our patient and Cruceyra *et al.* [19]. Twin pregnancy in both horns of a bicornuate uterus is a rare event. Its incidence is unknown, although several cases have been reported anecdotally. It is a combination of two rare events, uterine malformation (bicornuate uterus) and dizygotia, which occurs only exceptionally in spontaneous cycles with a frequency of between 1,300,000 and 2,000,000 pregnancies [17] [20].

The obstetrical outcome of this potentially risky pregnancy seems unfavourable. Twin pregnancies with uterine malformations are associated with a high rate of miscarriage [15]. Indeed, twin pregnancies individually increase the risk of complications; however, there is limited data to illustrate the effect when a twin pregnancy is coupled with a uterine malformation, particularly a bicornuate uterus. Studies in the literature have focused on the second and third trimesters [21]. Suh *et al.* report two cases of spontaneous miscarriage [12].

The management of this unusual pregnancy needs to be individualised due to the lack of consensus on the principles of management and the risk of potential complications in order to achieve better outcomes.

Elias and Amisi noted that there were no specific guidelines for management and therefore recommended the continued publication of case reports regarding

uterine malformations and twin pregnancy to provide more data for a global registry [7].

This could help to establish a protocol for the management of the combination of uterine malformation and pregnancy, particularly in twin pregnancies, in order to increase the fetal survival rate. Improved availability of diagnostic facilities would allow early detection and corrective surgery in the gynaecological period, or where appropriate early diagnosis of uterine malformation in the first trimester of pregnancy to improve prognosis.

4. Conclusion

The occurrence of spontaneous twin pregnancy with implantation in each horn of a bicornuate uterus is a very rare phenomenon with a guarded prognosis, hence the interest in systematic screening for uterine malformations in the preconception period. Endovaginal ultrasound and pelvic magnetic resonance imaging help to avoid diagnostic pitfalls thanks to optimal monitoring. The respect of the diagnostic tripod (clinical, biological and ultrasound) as well as the mastery of ultrasound anatomy, coupled with the dexterity and caution of the gynaecologist, allowed our patient to avoid white surgery in extremis.

Conflicts of Interest

The authors declare no conflicts of interest.

References

[1] de Peyruis, S.D.V. (2014) Malformation utérine et accouchement voie Basse: étude rétrospective de 139 grossesses. Université d'Angers, UFR des Sciences Médicales, Ecole de Sage-femme d'Angers, René Rouchy.

[2] Alkhateeb, H.M. and Yaseen, E.M. (2015) Twin Pregnancy in an Accessory Cavitated Non-Communicating Uterus. *International Journal of Surgery Case Reports*, **10**, 45-48. https://doi.org/10.1016/j.ijscr.2015.03.023

[3] Doruk, A., Gozukara, I., Burka, G., Bilik, E. and Dilek, T.U.K. (2013) Spontaneous Twin Pregnancy in Uterus Bicornis Unicollis. *Case Reports in Obstetrics and Gynecology*, **2013**, Article ID: 834952. https://doi.org/10.1155/2013/834952

[4] Grimbizis, G.F., Gordts, S., Sardo, A.D.S., *et al*. (2013) The ESHRE/ESGE Consensus on the Classification of Female Genital Tract Congenital Anomalies. *Human Reproduction*, **28**, 2032-2044. https://doi.org/10.1093/humrep/det098

[5] Osazuwa, H. and Ejenobo, D. (2012) Twin Gestation in a Septate Bicornuate Uterus. *Case Reports in Obstetrics and Gynecology*, **2012**, Article ID: 563085. https://doi.org/10.1155/2012/563085

[6] Arora, M., Gupta, N., Neelam and Jindal, S. (2007) Unique Case of Successful Twin Pregnancy after Spontaneous Conception in a Patient with Uterus Bicornis Unicollis. *Case Report. Archives of Gynecology and Obstetrics*, **276**, 193-195. https://doi.org/10.1007/s00404-007-0323-7

[7] Elias, H.E. and Amisi, J.A. (2020) Twin Pregnancy in a Bicornuate Uterus in Rural Kenya: A Case Report for Accidental Discovery and Successful Delivery. *African Journal of Primary Health Care & Family Medicine*, **12**, a2198.

[8] Kachhawa, G. and Kriplani, A. (2017) Management of Reproductive Tract Anoma-

lies. *The Journal of Obstetrics and Gynecology of India*, **67**, 162-167. https://doi.org/10.1007/s13224-017-1001-8

[9] Enebe, J.T., Omeke, C.A. and Chukwubuike, E.K. (2020) Retained Second Twin Secondary to an Undiagnosed Bicornuate Uterus in a Poorly Supervised Labour: A Case Report. *Proceedings of Singapore Healthcare*, **30**, 162-165.

[10] Dasari, M. (2012) Bicornuate Uterus: A Case Report. *International Journal of Biomedical Research*, **3**, 395-396. https://doi.org/10.7439/ijbr.v3i9.416

[11] Shim, S., Hur, Y.M., Kim, D.H., Seong, S.J., Kim, M.L. and Shin, J.S. (2016) Evidence for No Significant Impact of Müllerian Anomalies on Reproductive Outcomes of Twin Pregnancy in Korean Women. *Twin Research and Human Genetics*, **19**, 146-153. https://doi.org/10.1017/thg.2016.4

[12] Suh, C.S., Jee, B.C., Ku, S.Y., Kim, S.H., Choi, Y.M., Kim, J.G. and Moon, S.Y. (2005) Twin Gestation Induced by Clomiphene Citrate and Bromocriptine in Both Horns of a Bicornuate Uterus. *Fertility and Sterility*, **84**, 756. https://doi.org/10.1016/j.fertnstert.2005.02.048

[13] Kong, W.Y., Zhao, S.R., Deng, K., Zhang, Q., Liu, W. and Yan, L. (2021) Effects of Bicornuate Uterus on Pregnancy and Obstetric Outcomes of *in Vitro* Fertilization/Intracytoplasmic Sperm Injection. *European Journal of Obstetrics & Gynecology and Reproductive Biology*, **258**, 132-138. https://doi.org/10.1016/j.ejogrb.2020.12.046

[14] Heinonen, P.K. (2016) Twin Pregnancy in the Congenital Malformed Uterus. *Journal of Obstetrics and Gynaecology*, **36**, 571-573.

[15] Ilyas, M., Dar, M.S., Rafiq, S. and Khan, I. (2018) Twin Pregnancy in Bicornuate Uterus: One Fetus in Each Horn. *Journal of Fetal Medicine*, **5**, 59-61. https://doi.org/10.1007/s40556-018-0156-4

[16] Adams, K. and Minuzzo, L. (2019) Successful Delivery of Spontaneously Conceived Twins in a Single Horn of a Bicornuate Uterus: A Case Report. *Case Reports in Women's Health*, **21**, e00103. https://doi.org/10.1016/j.crwh.2019.e00103

[17] Guennoun, A., Mamouni, N., Errarhay, S., Bouchikhi, C. and Banani, A. (2017) La grossesse hétérotopique spontanée: À propos de deux cas. *Pan African Medical Journal*, **28**, Article No. 306.

[18] Narlawar, R.S., Chavhan, G.B., Bhatgadde, V.L. and Shah, J.R. (2003) Twin Gestation in One Horn of a Bicornuate Uterus. *Journal of Clinical Ultrasound*, **31**, 167-169. https://doi.org/10.1002/jcu.10143

[19] Cruceyra, M., Iglesias, C., De La Calle, M., Sancha, M., Magallón, S.L. and Gonzàlez, A. (2011) Successful Delivery of a Twin Pregnancy in a Bicornuate Uterus (Uterus Bicornis Unicollis) by Bilateral Caesarean Section. *Journal of Obstetrics and Gynaecology Canada*, **33**, 142-144. https://doi.org/10.1016/S1701-2163(16)34800-9

[20] Rajaonarison, J.J.C., Rakotomahenina, H., Tohaina, D.V., Andrianandraina, M.C.G., Rabesandratana, N. and Randaoharison, P.G. (2015) Un nouveau cas de grossesse gémellaire sur utérus bicorne à Madagascar. *Journal Malgache de Gynécologie-Obstétrique*, **1**, 16-18.

[21] Fox, N., Roman, A., Saltzman, S., Klauser, C. and Rebarber, A. (2013) Twin Pregnancy in Patients with a Uterine Malformation. *American Journal of Obstetrics & Gynecology*, **208**, S222. https://doi.org/10.1016/j.ajog.2012.10.683