Case report

Term delivery of dicavity twins in a uterus didelphys by bilateral caesarian section

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Abstract

Twin pregnancy continuing up to term in a uterus didelphys is an extremely rare occurrence. The following is a case report of such an event at District Base Hospital, Dambulla, Sri Lanka.

Key words: uterus didelphys, twin pregnancy

Introduction

Uterus didelphys in the general population of women is very rare ranging from 1 in 1,000 to 1 in 30,000. Usually, due to the uterine anomalies, these women present with fertility problems and the overall reproductive performance of uterus didelphys is poor (Raga et al., 1997). However, there are no guidelines about follow up of pregnancy or selecting the mode of delivery as the incidence is very low (Arora et al., 2010).

Within this background, a mature woman with a uterus didelphys presenting with twins at term is an extremely rare occurrence with a chance of 1 in 5 million. Dicavity multi foetal (twin/triplet) gestations reported in literature has been numbering only 20 cases up to the dawn of the present millennium (Makoto et al., 2003), with only 1 case of triplets in a uterus didelphys recorded so far.

We present a similar a rare case of a woman with a uterus didelphys (bicornis unicollis) carrying dicavity twins up to 37 weeks of gestation giving birth to 2 healthy babies in our unit.

Case report

An 18 year old woman, married for 1 year, in her first pregnancy was admitted to the maternity ward of District Base Hospital, Dambulla for delivery. She had not used any contraceptives since marriage. At the time of admission her pregnancy was in to 37 weeks.

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She has had her booking visit at a different consultant led clinic at 6 weeks and thereafter had been managed at the local clinic till term. At the booking visit, she was diagnosed as carrying twins. No mention about the chorionicity or uterine anomalies noted in the records. All of her 3 trimesters have been uneventful and all investigations have been normal.

Upon her admission for delivery, she was well looking, not pale, with a pulse rate of 70/min and a blood pressure of 100/70 was noted. Her abdomen was distended asymmetrically roughly resembling a heart shape with fundus larger than dates at 42 cm. Twin fetuses were palpable and 1st of twin presenting in cephalic presentation and the 2nd of twin in breach presentation which were confirmed from ultra sound scan. On vaginal examination, a single cervix was visualized, partially effaced with the os closed. Therefore it was planned to deliver the babies by an elective caesarian section.

She underwent a caesarian-section the day after her admission. Upon safe entry in to peritoneal cavity, 2 separate uteri were noted each attached to its corresponding fallopian tube and ovary, in its lateral aspect. Therefore to deliver the babies, two separate lower segment incisions had to be made on both uteri and the babies were delivered separately. Twin 1 weighed 2180 g and Twin 2, 2480 g. Both were healthy and cried at birth and were handed over to the paediatric team. The separate uterine incisions were sutured with vicryl. Except for the two separate uteri, no gross anatomical anomalies were detected. Routine closure was done with 2 vicryl to rectus sheath and 2/0 vicryl to skin. Her post-op recovery was uneventful (Figure 1.)

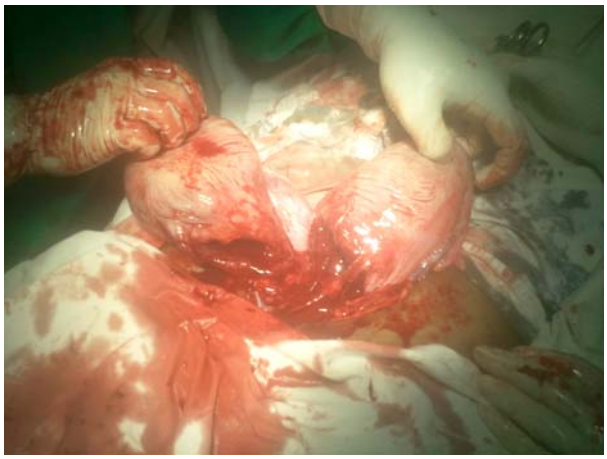

Figure 1. **Bilateral caesarian sections of the double uteri.**

As this was a case of undiagnosed uterus didelphys (bicollis unicollis) each carrying a viable foetus, further ultra sound imaging of her pelvis and abdomen was done at the radiology department 3 days after the caesarian delivery to detect any other anomalous pathology prior to discharge of patient. No such anomalies were noted and no evidence was found with regard to duplex renal systems (Figure 2).

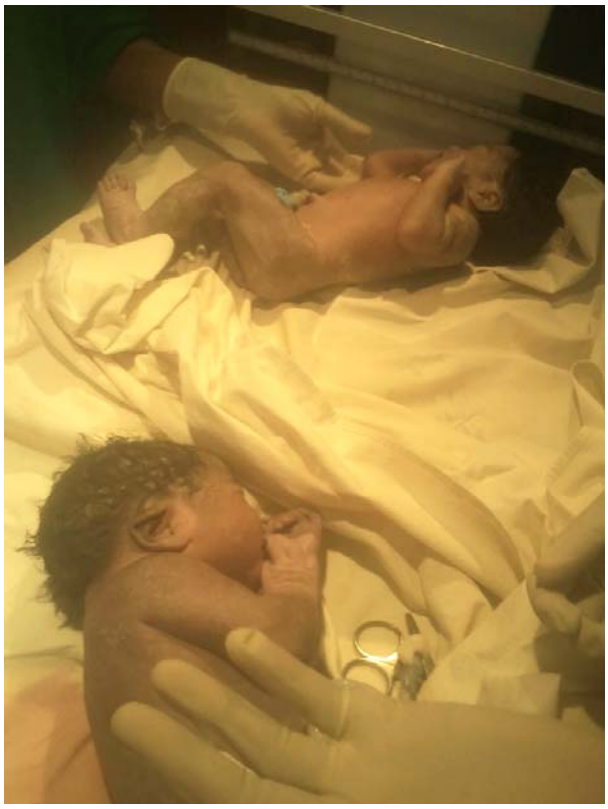

Figure 2. **The two healthy new born dicavity twins.**

Babies were examined by the paediatric team and were pronounced healthy. Patient was discharged on post-op day 3.

Field midwifery staff was notified to make further post natal visits to ensure the wellbeing of the mother and the babies.

Patient was reviewed 3 months post-delivery at the hospital clinic to arrange a contraceptive implant (Jadelle) and for further counseling with regard to any future pregnancies. Furthermore, a hystero-salphingogramme was arranged as follow up to the initial ultra sound imaging, which confirmed its reporting with regard to her cervical and uterine anatomy (Figure 3).

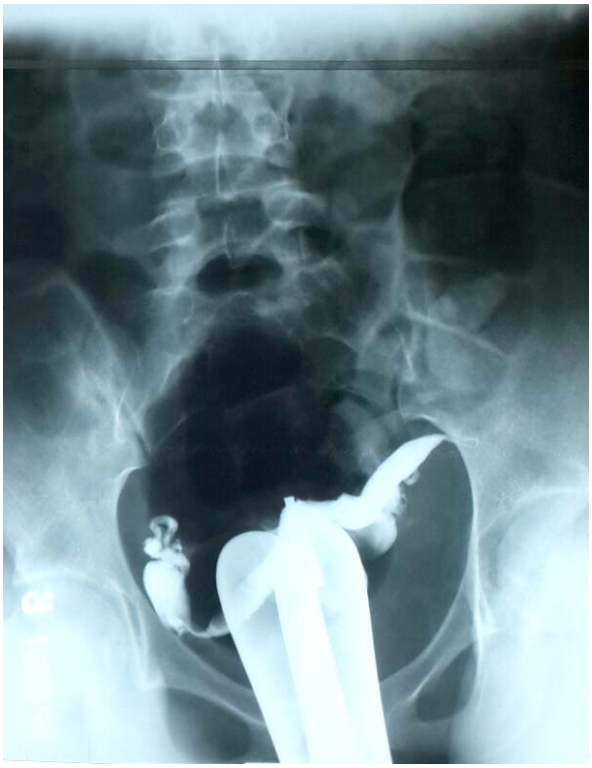

Figure 3. **Hysterosalphigogramme of the uterus didelphys at three months postpartum.**

Discussion

Uterine anomalies in a woman can vary from absence of uterus to fusion anomalies. The latter may present up to various degrees with or without clinical or pregnancy related outcomes depending on the severity of the anomaly. With the more common lesser degree fusion defects, the cornual parts of the uterus may remain separate giving rise to an arcuate shaped uterus. If the separation is of a minor degree it is unlikely to present with significant clinical implications. However, a presence of a complete rudimentary horn may give rise to a serious situation if a pregnancy is implanted within.

The presence of a septum extending over some length or all of the uterine cavity, which is called as a septate uterus, may be of normal external appearance or of bicornuate outline and is likely to present with clinical features such as recurrent spontaneous miscarriages or malpresentation. In more extreme forms of failure of fusion, could lead to two almost separate uterine cavities with a single cervix or to complete duplication of the uterus and cervix (uterus didelphys). However, uterus didelphys is much less commoner than other uterine malformations such as arcuate uterus, septate uterus or bicornuate uterus (Grimbizis et al., 2001). Usually, due to the uterine anomalies, these women present with fertility problems and the overall reproductive performance of uterus didelphys is poor (Raga et al., 1997). Even those who conceive could present with a high rate of complications such as spontaneous miscarriage (30%), breech presentation (43%), pre labour rupture of membranes (53%) and preterm labour (95%) (Heinonen, 1984) with an average live birth rate of 69% (Lin, 2004). In addition to above, in certain studies, uterus didelphys has shown to be associated with a higher rate of intra uterine growth retardation and postpartum haemorrhage (Pui, 2004).

However, interestingly, there is another school of thought suggesting that in more extreme forms of failure of fusion the clinical features may be less rather than more marked, and that out of congenital uterine anomalies, didelphys uterus offers the best chance for a successful pregnancy (57%) (Musich & Behrman, 1978), with a foetal survival rate as high as (64%) (Marshiach et al., 1981).

Anyhow, patients with double uteri may need special attention during pregnancy (Heinonen 1984; Reichmun & Laufer 2010). Mode of delivery can be abdominal or vaginal. However dystocia, malpresentation and possible risk of uterine rupture are major handicaps to avoid in vaginal delivery (Cruceyra et al., 2011). Caesarian section was preferred in 82% of patients reported by Heinonen (Heinonen, 1984). However, there are no guidelines about follow up of pregnancy or selecting the mode of delivery as the incidence is very low (Arora et al., 2010).

With the increasing trend of utilization of artificial reproductive techniques to tackle fertility problems, it would be a fair guess to predict an increase in the incidence of twin gestations in women with uterus didelphys in future. In this context it would be prudent to suggest that whenever a twin gestation is diagnosed especially in women with a past history of sub fertility, recurrent miscarriage or with persistent malpresentations, to pay special attention with imaging with regard to the diagnosis of a possible uterus didelphys or any other fusion anomalies of uterus in addition to the present practice of determining the chorionicity in multiple pregnancies. Moreover, as uterine malformations and genital tract anomalies could be closely associated with renal tract abnormalities, further imaging to confirm or rule out such co-existing anomalies, especially prior to planning of the delivery, need to be emphasized.

With regard to our case in discussion, as the initial booking visit was done at a different unit along with no reference to uterine anomalies in the notes and compounded by the fact that the patient had thereafter has been followed up exclusively at the local peripheral clinic (with no access to imaging) till delivery, the discovery of a fully grown term twin pregnancy in a uterus didelphys in the operating theatre presented as a complete surprise. Anyhow, as the babies and the mother made an uneventful recovery, the event was successfully concluded without any notable complications. However, to complete the picture, the patient was referred to the radiology department prior to discharge for imaging for possible associated hitherto undiagnosed genital or renal tract anomalies. The results came back as negative.

The patient was followed up after three months at the hospital clinic and underwent a hysteron-salphingogramme to confirm the postpartum ultra sound scan imaging results of her genital tract. Also, insertion of a Jadelle contraceptive implant was arranged.

Lastly it would be interesting to debate with regard to the place of post partum intra uterine contraceptive devices in such patients if requested by them, and the necessity and the success rate of having to use two devices on a single patient in such instances.

Author declarations

Declaration of interests: The authors report no conflicts of interests. The authors alone are responsible for the content and writing of the paper.

Author contributions: SW was the lead clinician who was in charge of managing the patient as well as the team leader of the surgical procedure. IS and DJ comprised the rest of the surgical team. IS, DJ and SS were involved with the pre-operative and post-operative patient care management up to the time of discharge from ward. SW developed the case presentation, editing and developing of the final draft.

Ethical considerations: Informed written consent has been obtained from the patient with regard to publication of the case report for academic and educational purposes. No patient identifiable details or images have been used in the case report and any information that could lead to the identity of the patient or the specific case scenario has been withheld in the publication.

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