UTERUS PSEUDO-DIDELPHYS

(UTERUS BICORNIS BICOLLIS WITH SEPTATE VAGINA)

(Report of an interesting case)

by

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For the normal development of fallopian tube, uterus and vagina, the two müllerian (para-mesonephric) ducts of two sides of the developing embryo must come together and get fused completely except in their uppermost parts from which the tubes are developed. Absence or incomplete development of one or both müllerian ducts and their imperfect fusion or failure of fusion give rise to various malformations which are responsible for many important complications and interesting problems in obstetrics and gynaecology.

Why in some cases the two müllerian ducts fail to fuse normally and completely is not definitely known. But some defects in subperitoneal fibro-muscular tissues that normally bring them together, over-development of the round ligaments that tend to pull them apart and presence of a thick and tough recto-vesical fold or ligament in between the two ducts have been suggested as possible causes.

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Uterus didelphys or pseud-didelphys is duplication of uterus, including the cervix and vagina, and results from complete or almost complete failure of fusion of müllerian ducts. This type of malformation is very rare and unless the possibility is borne in mind, is liable to be missed during examination leading to interesting incidents. Chassar Moir (1956) mad mention of an instance of veheme dispute amongst a few specialists ov the degree of dilatation of cervix ir particular case, because, like bli men seeing the elephant, some them had felt only the one and s only the other of the two cervic the patient and none had palp both. Similarly, three experts failed to rupture artificially the me branes in a case of pre-eclamp toxaemia, because all of them ha tried to do so through the cervix of a non-pregnant second uterus. Jeffcoate (1962) cited instances where more than one caesarean section had been performed due to non-dilatation of the non-pregnant cervix. Treatment of spasmodic dysmenorrhoea, menorrhagia and vaginitis and also contraceptive measures have sometimes falied because the existence of a second uterus or a second

vagina was overlooked. An interesting case of uterus pseudo-didelphys, the first of its kind treated in the Gauhati Medical College Hospital, illustrating failure of contraception and continuation of menstruation during pregnancy is reported here.

Case Report

Mrs. S. P., aged 39 years, consulted the senior writer (R.K.D.) with the complaint of profuse and prolonged menstruation for two years. Excessive menstrual flow continued for 10 to 12 days in her 20-22 day cycles. Besides, something came out per vaginam after her first difficult labour 14 years ago and it has been dangling over her vulva ever since. She never had any urinary complaint, but suffered from dyspareunia for some time after marriage. The most interesting and unique part of her menstrual history was the fact that she continued to menstruate regularly all throughout her three pregnancies with practically no lactational amenorrhoea. Bleeding presumably occurred from the endometrium of the non-pregnant horn.

The patient had a difficult breech extraction and application of forceps to the aftercoming head during her first delivery at term and the asphyxiated baby died after two hours. The next pregnancy occurred while she was using contraceptives, the vaginal diaphragm and "Preceptin". The presentation was again breech but she had a spontaneous delivery of a healthy male baby at term. Her third pregnancy also ended in a normal labour. She gave no history of retained placenta or post-partum haemorrhage. There was nothing particular and relevent in her past medical history and family history. She had consulted several gynaecologists and had tried conservative treatment for her menorrhagia during the last two years, with no effects.

On examination, the patient was found to be in good health except for mild anaemia. She was of average stature and well nourished. A careful systemic examination could detect no abnormality in any of her systems.

On vaginal examination, a longitudinal fold of vaginal mucous membrane was seen hanging outside with its lowest margin about one and half inches below the introitus. A much smaller fold was also found attached to the mild-line of the lower part of posterior vaginal wall. There was no cystocele, rectocele or uterine prolapse. The bigger fold of vaginal mucosa was attached to the whole length of the anterior vaginal wall along the midline like the skin fold of the neck of an ox. Small tags of vaginal mucosa were also found on the posterior vaginal wall higher up. The cervix on palpation, at first, appeared to be one although it was unusually broad. But on inspection and on passing an uterine sound an external os for each and two cervical canals could be identified. Above the cervix, two almost equally developed uterine horns could be easily palpated and the cavity of each communicated with the cervical canal of the same side. Adnexa were not palpable. From the findings of the clinical examination and hysterosalpingography, a diagnosis of uterus pseudo-didelphys or uterus bicornis bicollis with septate vagina was made. In view of the severe and intractable menorrhagia telling upon her mind and body, failure of hormone therapy and other conservative measures, and as the patient and her husband had no desire for any further pregnancy, an abdominal hysterectomy and excision of the torn vaginal septum was advised.

On 27-3-68, the patient was admitted. Most of the routine laboratory investigations, including hysterosalpingography and pyelography, were already carried out; other investigations carried out at our hospital showed nothing abnormal.

On opening the abdomen under spinal anaesthesia on 30-3-68, a beautiful specimen of a bicornuate uterus could be seen. Both the horns were well developed with a thick recto-vesical fold of peritoneum in between the two (Fig. I). Each horn had a normal tube and a normal ovary on its lateral side. A total hysterectomy was performed and the vaginal septum was excised at the same time. The patient had an uneventful recovery and was discharged from the hospital on the 12th day of the operation.

It is a rare and typical specimen of uterus bicornis bicollis with septate vagina

and has been preserved in the departmental museum (Fig. 2). From the degree of development and parous appearance of external os of both the cervices, it seems that both the horns had been pregnant with full-term babies.

Discussion

According to Chassar Moir and others, the term uterus didelphys should be reserved only for those cases of extreme degree of duplication where two complete sets of genital organs e.g. tube, uterine body, cervix, vagina and even vulva are developed separately on two sides. As the organs of the uro-genital system develop together from the mesonephric and para-mesonephric ducts in close relation, developmental anomalies of one system are frequently associated with those of the other. A true uterus didelphys is combined also with duplication of the bladder and urethra. This type of malformation is, of course, extremely rare and not more than 20 cases have so far been reported.

In uterus pseudo-didelphys or uterus bicornis bicollis with septate vagina, of which the present case is an example, the duplication of the organs is a bit less complete. Two cervices are usually joined medially, the vagina may be double or septate but the vulva is single. Other varieties of malformations due to imperfect fusion of müllerian ducts, namely planiform or anvil uterus, cordiform uterus, septate or sub-septate uterus, uterus bicornis unicollis, etc. are not so rare. One or other type of these developmental anomalies has been reported to be present in 1.1 to 3.5 per cent of all women (Strassmann, 1966) and in 0.3 per cent of all deliveries. (Baker, *et al* 1953). Many women are unaware of their existence as they often give rise to no symptoms. However, menorrhagia and spasmodic dysmenorrhoea are more common. Fertility is usually not affected. As a matter of fact, most of the cases are first detected during the investigations for complications of pregnancy and labour. Repeated abortions, pregnancy in rudimentary horn, especially when it has no communication with the main cavity or cervix, malpresentation, premature labour, abnormal uterine action, obstructed labour, rupture of uterus, post-partum haemorrhage, and retained placenta are some of the common and important obstetric complications requiring careful management. Cornual pregnancy or rupture of a rudimentary pregnant horn closely resembles tubal pregnancy or tubal rupture. One of the two horns is often mistaken for a fibromyoma of the uterus. Angular pregnancy and pregnancy in uterine diverticulum also come in for differential diagnosis.

The diagnosis of this type of malformation due to defective fusion of müllerian ducts can sometimes be extremely elusive. While the presence of a vaginal septum or two cervices can be seen and felt, two uterine horns or a dimple on the fundus may be palpated and an unusually broad uterus may arouse suspicion, diagnosis can be confirmed only by hysterosalpingography especially in cases like septate or sub-septate uterus. In many cases it is an accidental and unexpected finding at laparotomy.

A number of cases of twin pregnancy in bicornuate uterus have been

recorded in the literature and one of us (R.K.D.) has the personal experience of a young girl who had one foetus in each horn of her bicornuate unicollis uterus. One horn contains usually one foetus, but occasionally both. One of the most interesting aspects of twin pregnancy in a bicornuate uterus is that the deliveries of two babies may take place at different times at quite a long interval, varying from two weeks to fourteen weeks. A number of such cases also have been reported (Bainbridge, 1924; Colaco, 1949; Bruce and Cummings, 1953). One of Bainbridge's cases gave birth to two full-term babies — one white and one black — at an interval of two months. These cases bear contradictory evidence against some of the generally recognized conceptions or ideas about ovulation, menstruation, pregnancy and onset of labour.

For treatment every case should be considered on its own merit. Uterine malformations giving rise to no symptoms and no complications during pregnancy or labour should better be left alone. Vaginal septa and rudimentary horns may require excision. Resection of septa in cases of septate or sub-septate uterus gives satisfactory results (Way, 1945). A bicornuate uterus responsible for repeated abortions, menorrhagia or other symptoms can be successfully treated by an operation of unification of the two horns (metroplasty or utericuloplasty) after the technique of Strassmann. In his latest series of 263 operations, Strassmann (1966) claimed success in 197 (74.9%). During subsequent pregnancy and labour after the operation, the patient must be kept under careful observation. Although spontaneous vaginal delivery does occur in some cases, according to Jeffcoate (1962), it will be safer and wiser to perform an elective caesarean section because of the risk of rupture of the scar.

Summary

A case of uterus pseudo-didelphys (uterus bicornis bicollis with septate vagina) recently treated in the Medical College, Gauhati, has been reported with comments.

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