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COMMUNICATION

A unique mullerian anomaly of bicollis uterus and vaginal septum with abnormal cervical smears—a case report of pregnancy in such an anomaly

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Introduction

Performing a cervical smear is technically difficult with certain mullerian anomalies. We encountered a case of double cervix and vagina who presented with difficulty in taking cervical smears and was reported to have an abnormal smear. This is the first reported case of pregnancy with this type of unique abnormality.

Case report

A 28-year-old nulliparous woman presented to her GP for a routine cervical smear. There was some difficulty in obtaining the smear. The smear revealed moderate dyskaryosis. She was referred to a hospital for further management.

Examination in the clinic revealed a double cervix with a vaginal septum. In view of the anomaly, colposcopy was performed under anaesthesia and she was confirmed to have a vaginal septum, two cervixes (Fig. 1) and one uterus. She underwent laparoscopy, which revealed one uterus and a normal adnexae. Hysteroscopy performed from both cervices revealed a common uterine cavity with a normal endometrium. Colposcopy showed findings suggestive of CIN 1–2 and loop-cone biopsies were taken from both cervices. The vaginal septum was excised with diathermy. The histology of the loop cones revealed inflammatory changes with no evidence of dysplasia. An intravenous urogram (IVU) was carried out, which did not show any abnormality.

She had a natural conception six months later. The pregnancy was uncomplicated. She went into spontaneous labour at 38 weeks gestation and delivered vaginally a 3-kg baby in good health. Her active labour lasted for 4 h and 18 min, and the third stage was prolonged at 1 h and 11 min, but the placenta was delivered spontaneously. No manipulation was done to the second cervix. She sustained a third-degree tear, which was repaired in theatre under spinal anaesthesia.

Discussion

This case represents a unique mullerian malformation in that it does not conform to our classical description of mullerian development [1]. We performed a Medline search and only four such cases have been reported in the literature, of which, one case had one of the cervices ending blindly and not communicating with the uterus [2–5]. This is the first reported case of pregnancy in an anomaly of this description.

The classical understanding of the mullerian development holds that mullerian ducts cross ventrally over the mesonephric ducts and their medial walls fuse in the midline at their caudalmost aspect, known as the mullerian tubercle. Fusion continues cranially and the fused medial walls are then resorbed, forming the uterine cavity. The mullerian tubercle induces proliferation of the adjoining urogenital sinus. This

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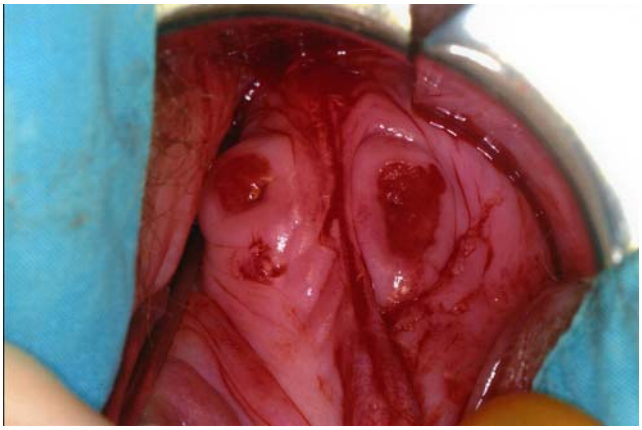

Fig. 1 Bicollis uterus with vaginal septum

results in the formation of the sinovaginal bulb, which, in turn, forms the solid vaginal plate. Canalisation of the vaginal plate forms a patent outflow tract. This classical description of mullerian development, which suggests a fusion in the cranial direction, does not explain the anomaly which we are reporting because it does not explain the two cervices and one uterus.

Musset et al. proposed an alternative mechanism which suggested that fusion first occurs at the level of the uterine isthmus and simultaneously proceeds in both directions [6]. Midline resorption also begins at the isthmus and is first directed caudally, unifying the cervix and vagina and, later, cephalically to eliminate the uterine septum. This alternative embryological mechanism explains both the standard classification of mullerian anomalies [6] and this unique anomaly as well. This isolated mullerian anomaly may be a consequence of a minor mesonephric defect [7].

As a part of the routine cervical screening, it becomes necessary to take smears from both cervices at screening, as oncogenic agents like human papillomavirus (HPV), which may be responsible for cellular changes, may affect both cervices. Her smear was reported as moderate dyskaryosis. Colposcopy and taking smears was technically difficult in this patient in view of the anomaly. Hence, she was examined under anaesthesia and a laparoscopy was performed to exclude uterine and other pelvic abnormalities. Loop-cone biopsies were performed for diagnostic and therapeutic purposes. Since the biopsy report did not suggest any CIN, further smears would be done at 3-yearly intervals in the future. In the event of technical difficulties in obtaining cervical smears, the same procedure may need to be carried out under anaesthesia.

Vaginal septae could become a source of vaginal dystocia or lacerations [8, 9]. Since it was excised, it prevented these potential obstetric complications. Excision of the septae may reduce the technical difficulty in taking cervical smears and general anaesthesia may not be required for cervical screening in the future.

Mullerian anomalies are known to present with cervical incompetence, preterm labour, malpresentations or cervical dystocia in labour. Due to the rarity of the case, we have not come across any other case of pregnancy reported in this anomaly. Considering the fact that she had an uneventful pregnancy, we can assume that women with this anomaly can be offered normal vaginal birth.

Summary

This unique mullerian anomaly which does not conform to our classical understanding of mullerian development may be due to minor mesonephric defects or to an alternative mechanism of mullerian development. Women with two cervices and a vaginal septum may pose difficulties during taking cervical smears and the process may be performed under anaesthesia. Since they are exposed to the same oncogenes, the risk of developing carcinoma exists in both cervices. This is the first reported case of pregnancy and, hence, we can assume that women with this anomaly can be offered normal vaginal delivery.

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