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Case Report

Uterus bicornis unicollis with multiple lieomyomas

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ABSTRACT

Uterine anomalies are congenital malformations caused by fusion or resorption defects during embryogenesis. Uterus bicornis unicollis (bicornuate uterus) is a rare condition which results from incomplete fusion of uterovaginal horns at the level of the fundus. Here we report a case of a 30 year old (P2L0) female, with uterus bicornis unicollis with multiple lieomyomas (fibroids). The patient underwent hysterectomy with adnexa removal. The specimen showed multiple lieomyomas with hyaline degeneration.

Keywords: Lieomyoma, Uterus bicornis unicollis

INTRODUCTION

Abnormal fusion of the Mullerian ducts during embryological life results in a variety of congenital uterine malformations; the etiology of such abnormalities remains unknown. The Mullerian duct anomalies are estimated to occur in 0.5 to 5% of women.[1] Uterus bicornis unicollis (bicornuate uterus) results from incomplete fusion of uterovaginal horns at the level of the fundus and are thought to represent 10 -35 % of Mullerian duct anomalies.[2] According to American fertility society, bicornuate uterus is class 4 Mullerian anomaly and may be associated with renal anomalies.

Cytogenetic abnormalities in the form of spontaneous chromosomal rearrangements are known to occur in uterine lieomyomas (fibroids). These chromosomal arrangements may be responsible for the initiation and progressive growth of the leiomyomas.[3]

Hysterosalpingography, hysteroscopy, laparoscopy, pelvic ultrasound using abdominal and transvaginal probes, CT scan, MRI of pelvic organs are increasingly being used as diagnostic modalities. 3-D ultrasound and MRI are standard methods of imaging uterine anomalies. 3-D ultrasound has the advantage of being non-invasive. In addition, it is mandatory to evaluate the urinary tract for concomitant anomalies and any altered anatomy due to mechanical effects of large leiomyomas.

CASE REPORT

A 30 year old P2L0 presented with menorrhagia since 4 months and distension of lower abdomen. She had a history of previous 2 cesarean sections done at 6 months and 8 months of gestation respectively. Intraoperative finding of uterus bicornis unicollis was noted. None of the babies survived despite neonatal intensive care. General and systemic examination showed no abnormality. On abdominal examination there was a firm non- tender, relatively fixed lump of approximately 28 weeks size and had limited mobility. On abdomino-pelvic examination above findings were confirmed and a single cervix felt. Hematological examination showed hemoglobin of 10gm/dl (hct 26%).

Ultrasound examination showed enlarged uterus with a distorted endometrial lining and multiple serous, interstitial and braod ligament fibroids arising from both horns of uterus. The right sided mass (cornuae and the fibroids) measured more than 14 x 9.4 cm, extending upto the right loin and left sided mass measured 10x 7.3

cm. bilaterally ovaries were identified and unremarkable. Both kidneys showed evidence of hydronephrosis due to pressure effect. CT scan confirmed the USG findings. There was no evidence of any enlargement of mesenteric or retroperitoneal lymph nodes or ascites.

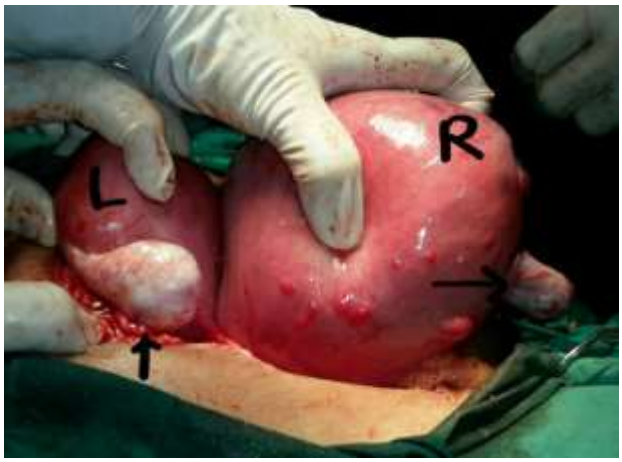

Figure 1: Uterus bicornis unicollis with lieomyomas (posterior view).

Cystoscopic bilateral ureteric catheterization was done prior to the exploratory laparotomy, so as to localize and safeguard the ureters during surgery. The surgery was done through a midline incision extending 4cm above umbilicus. Intraoperative, both cornuae were seen separate and having multiple fibroids. There was a broad ligament fibroid on left side (Figure 1).

The right mass was large extending high up to the under surface of liver. Both ovaries were unremarkable but adherent to the mass hence removed. The sigmoid colon was adherent at the cleft between the two cornuae (Figure 2).

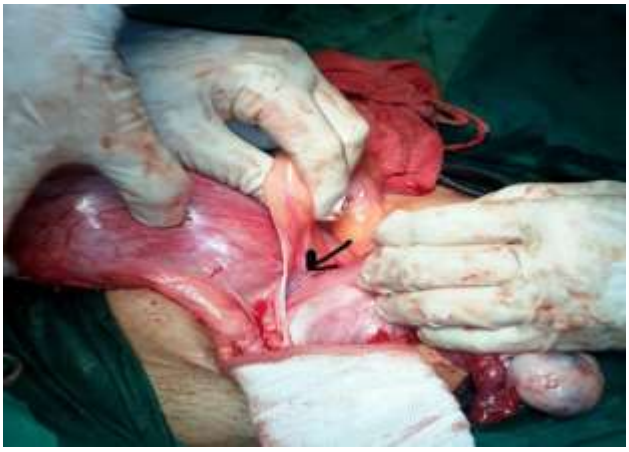

Figure 2: Sigmoid colon adherent in the cleft between the cornuae.

Adhesiolysis and bladder dissection was done. The lower part of cervix was difficult to approach hence a decision of sub-total hysterectomy was taken. The intra operative blood loss and the loss in the large specimen was substantial. However, based on pre-operative hemodynamic evaluation and post-operative clinical status it was recommended to give 4 units of blood. The ureteric catheters were later removed. The patient had an uneventful recovery. Histopathological examination of the specimen confirmed the presence of multiple lieomyomas with hyaline degeneration.

DISCUSSION

There are very few cases of lieomyomas in Mullerian anomalies reported in literature. Usually the diagnosis is not made clinically, because of its low incidence. Some cases of lieomyoma in bicornuate uterus has been reported previously.[4] The possible reason for its low occurrence could be a decreased concentration or sensitivity of the estrogen receptors or a lesser genetic predisposition for the clonal chromosomal abnormalities that are observed in women with normal uterus with lieomyomas.[3]

As the diagnosis of bicornuate uterus was already made in previous surgeries, in our case it was easy to delineate the location of various lieomyomas with respect to the uterine anatomy on USG. Both the ovaries were identified. Also, proximal hydroureter and hydronephrosis was noted due to the large lieomyomas. Histopathologically, the lieomyomas showed hyaline degeneration.

Patient consent was obtained and patient identity was not disclosed.

CONCLUSION

Uterus bicornis unicollis with lieomyomas is a rare but challenging situation. A proper pre-operative evaluation of the type of Mullerian anomaly, the number, location and size of lieomyomas along with mapping of the urinary tract is a must. A multi-disciplinary approach is essential to ensure safe recovery of the patient. Documentation of these rare cases can help us learn new concepts of management.

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