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Case Report

Rudimentary horn fibroid with retrograde endometriosis in a patient with unicornuate uterus: A case report

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ABSTRACT:
We describe the case of a 37 year old female presented to ER complaining of pain in lower abdomen since 2 days associated with vomiting since one day. There was no history of constipation or loose stools or fever. She had irregular periods. Ultrasound demonstrated left sided retroflexed uterus with a right sided rudimentary horn showing fibroid within it. MRI findings depicted retroflexed hemiuterus deviated to the left with rudimentary right horn. An intramural fibroid with hemorrhagic / cystic degeneration seen within the rudimentary horn (U-4a C0V0 - ESHRE / ESGE classification).
Keywords Unicornuate uterus, Rudimentary uterine horn, Congenital mullerian anomaly.

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INTRODUCTION

The prevalence of congenital uterine malformations is about 0.5%. A malformation is a morphologic defect of a body region or organ resulting from an intrinsically abnormal developmental process. Reasons for uterine malformations include an incomplete midline unification of the para-mesonephric or Mullerian ducts. Depending on the incompleteness of fusion, there is a large variety of malformations. The American Fertility Society (AFS) suggest their specific classification of uterine malformations [1]. A unicornuate uterus is a rare uterine malformation with an incidence of 2.5–13% [2]. Incomplete fusion of the two Mullerian ducts may lead to a septate or bicornuate uterus; failure of Mullerian tube formation causes aplasia or atresia of one side, resulting in a unicornuate uterus. Incomplete atresia of a Mullerian duct leads to a rudimentary horn which is broadly connected or connected through streak tissue with the unicornuate uterus [3]. A unicornuate uterus may lead to various gynaecological or obstetric complications and diagnostics are often difficult and delayed to the fertile period or to pregnancy. Patients may present with painful menstruation, dyspareunia or malformations of the upper urinary tract, which are frequently inherited with a unicornuate uterus [4]. Rupture of a rudimentary horn is a life-threatening complication in pregnancy [5, 6]. We report a case of a MRI suggestive of Retroflexed hemiuterus deviated to the left with rudimentary right horn. An intramural fibroid with hemorrhagic / cystic degeneration seen within the rudimentary horn (U-4a C0V0 - ESHRE / ESGE classification). Note is made of adhered bilateral ovaries with endometriotic deposits in right ovary.

CASE REPORT

A 37 year old female presented to ER complaining of pain in lower abdomen since 2 days associated with vomiting since one day. There was no history of constipation or loose stools or fever. She had irregular periods. She was Conscious Oriented, with Pulse-80/min, BP-110/60 mmHg, sp02-98% on room air, Temp-99.7 F, Chest-B/L AE+, CVS- S1S2 +,P/A-Soft, Tenderness, Suprapubic region.

USG W/A WITH TVS.

Liver is mildly enlarged in size (16cm) and shows

grade-I/ll fatty changes. No focal intra-hepatic lesion is detected. Intra-hepaticbiliary radicals are not dilated. Gall bladder is partially distended and normal in calibre. Common bile duct is normal in calibre. In kidneys no focal lesion or calculus seen. Pelvicalyceal system is not dilated. Cortico-medullary differentiation is maintained.. Urinary bladder is partially Distended. Uterus is retroflexed. It measures 7.5 x 2.9cm. Myometrial echogenicity appears Uniform. Endometrial thickness is 2.47mm. A rudimentary horn is seen abutting the main uterine horn. This shows a hypoechoic lesion measuring approx. 3.5 x 2.7cm within it. Both ovaries are normal in size and echopattern. Right ovary measures 3.5 X 1.7cm. Echogenic area measuring 15mm seen in right ovary. left ovary measures 2.3 x 0.8cm, minimal free fluid is detected in pouch of Douglas.
IMPRESSION: Findings are likely suggestive of Left sided retroflexed uterus with a right sided rudimentary horn showing fibroid within it. **FIGURE 1,2 a,b**

MRI PELVIS
MRI of whole abdomen was performed on 3-Tesla scanner using phased Array coil. High resolution on T1 and T2 sequences were obtained through the abdomen and pelvis in multiple planes. Post contrast scans were taker in T1 axial and coronal, T1 fat sat and thrive sequences.
Observations: Liver is enlarged in size (22cm). No focalparenchymal lesion is seen. No intra hepaticbiliary radicle dilatation seen. Portalvein is normal. Gall bladder is well distended with clear lumen. CBD is not dilated. Spleen is normal in size, shape and MR signals. Pancreas is normal in size, shape and MR signals. Both kidneys are normal in size and shape. No focal lesion is seen. The pelvicalyceal systems are not dilated. Uterus is retroflexed, deviated towards left. Endometrial thickness is 3.54mm. Another similar myometrial signal intensity area measuring 5 x 4.9cm is seen adjacent to the uterus, suggestive of rudimentary horn. A lesion measuring approx. 3.7 x 2.5cm with central cystic and haemorrhagic area within it is seen in the rudimentary horn. Bilateral ovaries appear to be adhered to the uterus. Few hyperintense signal intensity areas on T1W fat sat images suggestive of haemorrhagic foci are seen in right ovary. An endometriotic deposit measuring 1.5 x 1.3cm is also seen in right ovary. Minimal free fluid is seen in pelvis. The visualized bowel loops are normal. The rectum is normal. The perirectal fat planes are intact.
IMPRESSION: MRI findings showed Retroflexed hemiuterus deviated to the left with rudimentary right horn. An intramural fibroid with hemorrhagic / cystic degeneration seen within the rudimentary horn (U-4a C0V0 - ESHRE / ESGE classification). Note is made of adhered bilateral ovaries with endometriotic deposits in right ovary. **FIGURE 3-5**

FIG 1- TVS showing retroflexed uterus deviated towards left side. Myometrial echogenicity is uniform

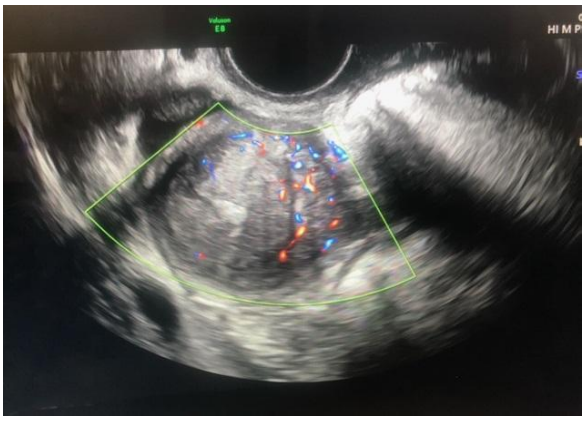
FIG 2A- The lesion in the rudimentary horn showing peripheral vascularity suggestive of a fibroid

FIG 2B-A rudimentary horn with a lesion is seen abutting the main uterine horn. The lesion shows whorled pattern suggestive of a fibroid.

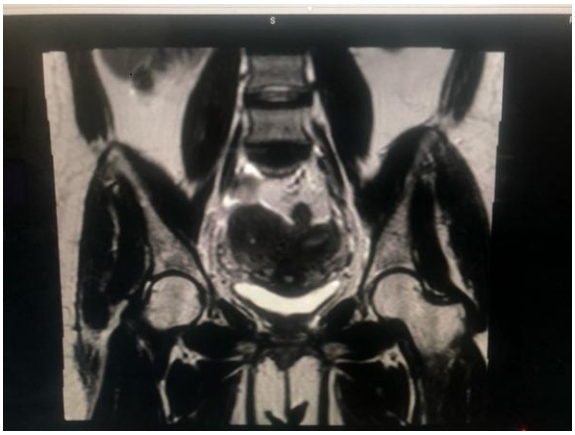

FIG 3- Coronal T2 W images show retroflexed uterus deviated towards left. Another similar myometrial signal intensity area seen adjacent to uterus denoting a rudimentary horn. A lesion with cystic and haemorrhagic areas is seen within the rudimentary horn

FIG 4- Axial T2 W images showing the rudimentary horn with a lesion showing cystic and haemorrhagic areas within it. Minimal free fluid is also seen in pelvis.

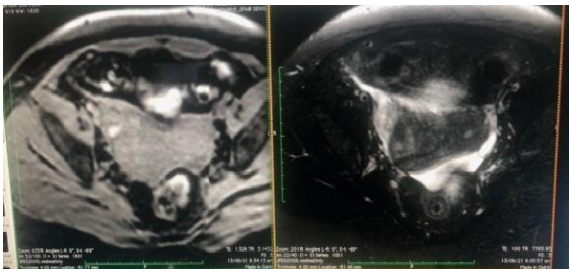

FIG 5- Axial T2 W and contrast images showing right ovary adhered to the uterus with an endometriotic deposit also seen n right ovary

DISCUSSION

A unicornuate uterus with a rudimentary horn is a rare uterine malformation, which explains why most general gynaecologists have little experience with this disease. Because of the lack of experience and incorrect interpretation of clinical and ultrasound findings, diagnosis is difficult and often found by chance only when the patient presents with sterility or complications [6]. As the literature demonstrates, early diagnosis is of great importance in order to avoid consecutive damage of the reproductive system and further painful complications [4, 7, 8]. To get to the correct diagnosis, the patient's history is crucial: increasingly painful periods, dyspareunia and sterility are common symptoms [2, 8, 9]. Our patient complained of increasing post-menstrual pain, which, in retrospective, we consider to be due to the increasing size of the rudimentary horn. To obtain the proper diagnosis, endovaginal grey-scale sonography is mandatory. As an additional method to investigate congenital abnormalities of the uterus, Salle et al. recommended hydrosonography [10]. Three-dimensional ultrasound and magnetic resonance imaging (MRI) may also give additional information, as well as hysteroscopy [2]. Often, laparoscopy is able to lead to the exact diagnosis of a uterine malformation [11] The classification of uterine malformations is described by the ASF [1]. A unicornuate uterus with a rudimentary horn is rather rare; case-control studies have demonstrated that rudimentary horns are mainly right-sided, as in our patient, which is the case in about 60% of patients [5, 12]. There is no evident explanation for this finding. Usually, the ipsilateral ovary is of normal function, as it is not of Mullerian duct origin but may be abnormally located. Extra pelvic localisations have been described [5]. If undetected, a unicorn uterus with a non-communicating cavitated horn is associated with a high incidence of complications. The fact that haematometra, haematosalpinx or even endometriosis are consequences of retrograde menstruation has already been described. The reason our patient developed endometriosis is due to retrograde menstruation. The diagnosis and appropriate treatment of a rudimentary horn should be carried out prior to pregnancy. Indeed, several complications have been described in the literature. Tubal pregnancy located in the tube near the rudimentary horn might be due to intra-peritoneal sperm or oocyte migration [2, 13]. Even pregnancies in the rudimentary horn have been described and show a higher incidence for abortion and rupture of the horn, particularly in the second or third trimester. The reason for uterine rupture is either due to a thinned muscular wall or an unphysiological implantation. This is why the rudimentary horn and the connecting tube should be removed in the course of a therapeutic laparoscopy [5]. A sub-mucosal fibroid was suspected, although the patient did not complain of any bleeding disorders. The differential diagnosis was adenomyosis. The endovaginal ultrasound demonstrated suggestive of Left sided retroflexed uterus with a right sided rudimentary horn showing fibroid within it. These malformations are, in 30–40% of cases, associated with further urogenital malformations. Therefore, we performed an MRI,

which did not show any pathological findings of the kidneys. MRI findings showed Retroflexed hemiuterus deviated to the left with rudimentary right horn. An intramural fibroid with hemorrhagic / cystic degeneration seen within the rudimentary horn (U-4a C0V0 - ESHRE / ESGE classification). Without the thorough knowledge of the pre-operative findings, we could have missed the uterine malformation, which has been described in the literature before, despite the experienced surgeons [7]. A detailed pre-operative diagnosis is crucial for the surgeon, as the removal of the rudimentary horn might be technically difficult: the layer between the rudimentary horn and the uterus is often barely visible [3, 8]. We used indigo carmine to dye the cavum of the rudimentary horn, which then was removed with a triangular excision without damaging the uterus. Case reports have described the use of hysteroscopic luminoscopy or intra-operative ultrasound with the use of specific transducers [14]. To avoid excessive bleeding during removal of the rudimentary horn, the knowledge of arterial blood supply is essential. Not only the ipsilateral uterine artery but also contralateral arcuate arteries inside the myometrium supply the rudimentary horn, which require coagulation [5, 6]. Finally, after the removal of the rudimentary horn, the muscular defect has to be closed, for which, the laparoscopic technique needs to be elaborated [15].

CONCLUSION

Particularly in pre-menopausal patients who present with abdominal pain, adnexal masses of unknown origin and severely painful periods with or without any signs of endometriosis, we must consider an anomaly of the Mullerian duct. Early diagnosis of a uterine malformation is essential to prevent complications. Pre-operative diagnosis is of great importance to enable adequate treatment to be undertaken. Such operations should only be performed by experienced surgeons with a high level of technical skill

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