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Case Report

An ectopic pregnancy: a case report with undiagnosed uterus didelphys

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Abstract

Normally, pregnancy begins with a fertilized egg and it attaches to the lining of the uterus. An ectopic pregnancy occurs when a fertilized egg implants and grows outside the main cavity of the uterus. A growing ectopic pregnancy in any location can cause the tissue to become vascular, friable and eventually rupture resulting in internal bleeding. This situation can be life threatening and needs to be treated as medical emergency. The present case report is of a 22-year-old female who complained of multiple episodes of vomiting after eating food. She was admitted for one day as a case of unknown poisoning and was managed conservatively. She was referred to higher centre, where she was declared brought in dead. The interest in this case lies principally in the history of the illness, raising the suspicion of unknown poisoning and findings of ruptured ectopic pregnancy at post-mortem examination, which later on histopathological examination was confirmed as a case of uterus didelphys, a rare uterine malformation.

Keywords: Ectopic pregnancy, Poisoning, Haemorrhage, Uterus didelphys

Introduction

Ectopic pregnancy is the most life threatening emergency in pregnancy leading to maternal death.[1] Fallopian tube (97%) is the most common ectopic site of implantation, and the remaining 3 % in the cervix, ovary, peritoneal cavity, or uterine scars. In the fallopian tube, 80 % of ectopic pregnancies occur in the ampulla, 10 % in the isthmus region, 5 % in the infundibulum and about 3 % in the interstitial portion.[1] During the first trimester, ectopic pregnancy is the leading cause of maternal death in industrialized countries and, possibly the second common cause of death in developing countries. The extrauterine sites of pregnancy are most prone to rupture as these structures cannot expand enough to fit the growing embryo, leading to massive haemorrhage.

Over the past few decades, the incidence of ectopic pregnancies has been on the rise globally. In India, the incidence ranges from 1-2%.[2] There are some studies that show no mortality, while there are others that show fatality rate as high as 3.5%.[3] This disparity reflects variable quality of care and infrastructure available in different clinical settings across the country. There are a number of factors that increase the risk of ectopic pregnancy. Under normal circumstances, after fertilization in the fallopian tube, the egg travels down to the implantation site. Any mechanism that interferes with the normal functioning of the fallopian tube, i.e., anatomic or functional, increases the risk of ectopic pregnancy. The risk factors include: pelvic inflammatory disease, previous ectopic pregnancy, endometriosis, previous pelvic or tubal surgery, uterotubal anomalies, infertility and its treatments, history of in utero exposure to diethylstilbestrol and cigarette smoking.[4] Other factors that are considered to be of posing risk are multiple sexual partners, early age at first intercourse and vaginal douching, having an indirect mechanism of action.[5]

Suspicion of ectopic pregnancy is raised when any woman with child bearing age presents to the casualty with symptoms such as abdominal pain, amenorrhoea and vaginal bleeding.[6] Less

commonly, the ectopic pregnancy may present with pain in the shoulder, rectal pressure, urinary symptoms, and anaemia. The physical findings include a normal or slightly enlarged uterus, cervical motion tenderness and a palpable adnexal mass. Sometimes, the woman may present with symptoms such as tachycardia, hypotension, marked abdominal tenderness with guarding and rigidity suggestive of a leaking or ruptured ectopic pregnancy.

Case history

A 22-year old married woman complained of multiple episodes of vomiting and abdominal pain at around 6 P.M. on 23.07.2017 with alleged history of having eaten food from outside. She was taken to a private hospital at Kaithal, Haryana, where she was admitted as a case of unknown poisoning. She was being managed conservatively at the hospital. Gastric lavage was done and sample was taken owing to the history of the present illness. On 24.07.2017 at around 10 A.M., the patient was referred to a higher centre and reached GMCH 32, Chandigarh where she was declared brought in dead at 1.50 P.M. The body was shifted to mortuary and post-mortem examination was conducted on the next day. *Autopsy findings:* The body was that of a young well nourished woman. Rigor mortis was well marked and faint post mortem staining was present and fixed over the back. Generalized pallor was present over the body. No external injuries were present on the body. On opening the abdominal cavity, about 2.5 litres of clotted and unclotted blood was present. All the internal organs were pale. Pelvic peritoneum was apparently normal. The uterus measured 10.5 cm x 7 cm x 3 cm and the cavity was empty. Both the ovaries were normal. On careful examination, the dilated left fallopian tube measured 7 cm x 3 cm with a rent of size 8 cm which was present over the ampullary region. Intact amniotic sac with attached placenta was extruded out from the rent as shown in figure 1. On opening the sac, it contained a male foetus of length 16 cm. Pregnancy was around 14 to 16 weeks. Right fallopian tube was found to be patent. All the other internal organs were pale. The routine viscera were sent for chemical analysis. The uterus with bilateral ovaries and fallopian tubes was preserved for histopathological examination to confirm the tubal pregnancy.

The chemical analysis was negative for any poison. Histopathological examination confirmed ruptured pregnancy in one of the cornua in a didelphys uterus, a uterine malformation.

Discussion

Owing to the history of the presenting illness, though the present case was initially treated as a case of suspected poisoning, but during post-mortem examination death was due to haemorrhage and shock as a result of ruptured ectopic pregnancy. Ectopic pregnancy, being the most common cause of maternal death in the first trimester of pregnancy, usually presents as sudden natural death. The symptoms may mimic those of appendicitis, acute abdomen or poisoning. There are studies that reported ruptured ectopic pregnancy where the women were of 20-30 years of age. Sometimes, gastrointestinal symptoms like abdominal pain and nausea are common, which are treated with antacids, without any investigations.

This present case is of particular importance as the findings on post-mortem examination were consistent with ruptured ectopic pregnancy but histopathological examination confirmed morbid anatomical features that of uterine malformation (Didelphys uterus) with ruptured pregnancy in one of the cornua.

As compared to other anomalies, didelphys uterus is a very rare Mullerian duct anomaly. Mullerian duct anomalies are the congenital defects that arise from abnormal embryological development of the Mullerian duct. It normally occurs between 6 and 22 weeks in utero. They can result due to failure of development, fusion, canalization, or reabsorption. Studies shows that prevelance of such anomalies is 0.1 to 10% and account for 11% of uterine malformations.[7] They are associated with higher rate of infertility, spontaneous abortion, intrauterine growth retardation and post-partum bleeding.[7] Mullerian duct development begins at an approximately 6 weeks gestational age when the paired mullerian ducts invaginate and then grow caudally and cross over the Wolffian ducts to meet at the midline. If embryologic arrest occurs during the 8th week of gestation which ultimately affects the Mullerian and Metanephric ducts, it is suggestive of didelphys uterus. However, if one of the Wolffian ducts is absent, the kidney and ureter on the ipsilateral side will fail to fuse at midline. This process may occur completely or incompletely and if the fusion is

incomplete, then a uterine didelphys is formed. Renal agenesis with right sided prevalence is the most common renal tract anomaly associated with Mullerian duct anomalies.[7]

Mostly women with a didelphys uterus are asymptomatic, but dyspareunia or dysmenorrhea may be the presenting symptoms in the presence of a thick, sometimes obstructing, vaginal septum. This obstructing vaginal septum can lead to hematocolpos/hematometrocolpos and thus present as chronic abdominal pain as well. Rarely, genital neoplasms and endometriosis are reported in association with cases of didelphys uterus.[8] Most frequent abnormal presentation in uterine didelphys is frank breech. Management in such patients may need special attention during pregnancy, as premature birth and malpresentations are common. Cervical incompetence, PPH, uterine rupture are the other complications that can occur during pregnancy. Vaginal delivery should be considered first as a didelphys uterus is not an indication for cesarean delivery.

Many case report didelphys uterus as a part of a syndrome, more specifically called, Herlyn Werner-Wunderlich (HWW) syndrome, also known as obstructed hemivagina and ipsilateral renal anomaly (OHVIRA). It is a very rare congenital anomaly of the urogenital tract, and is characterized by the triad of didelphys uterus, obstructed hemivagina, and ipsilateral renal agenesis.[9] This condition can cause hematometrocolpos or hematocolpos on the side of the obstructed hemivagina which produces a mass effect with subsequent lower abdominal pain.[10,11] Most cases present after menarche as intense lower abdominal pain and/or a protruding mass over the vaginal introitus.[10,11]

Invasive methods such as hysteroscopy, hysterosalpingography, and laparoscopy/laparotomy are frequently used for diagnosis. However, they are used based on the clinician's subjective interpretation rather than strict diagnostic criteria.[12] The first type of imaging that is done is a 2D ultrasound; however it cannot reliably differentiate between subtypes of MDAs. Nowadays, 3D ultrasound is being more commonly used for diagnosis as it is not only noninvasive, but it also provides a coronal view that enables examination of both the endometrial cavity and uterine fundus, thus giving all the information needed for morphological classification.[13] Pelvic examination will typically reveal a double vagina and a double cervix. Transvaginal ultrasonography and sonohystero grapghy, hysterosalpingograpghy, MRI and hysteroscopy, laparoscopy/laparotomy are other useful techniques to investigate the uterine structure. More recently 3-D ultrasonography is used as a method to evaluate uterine malformations. It accurately analyses uterine structure, contour of fundus, muscular thickness and septum length and is best performed during secretory phase of menstrual cycle. A large retrospective longitudinal study of 3181 patients by Raga et al. demonstrated poor reproductive performance in women with didelphys uteri with a higher rate of preterm delivery, spontaneous abortion, and the lowest chance of having a term delivery than the other MDAs.[14] Another long term retrospective follow-up of 49 women with didelphys uterus found no impairment with fertility and decreased rate of spontaneous abortion; however the rate of prematurity was increased in comparison to other known studies on septate and bicornuate uteri.[15]

The present case is important as it presented as a case of suspected poisoning base on the history and presenting symptoms, causing the death of young married woman and the role of histopathological examination in ascertaining the rare uterine anomaly. Clinical examination and certain investigations can help in making early and correct diagnosis in such rare Mullerian duct anomalies.

Conclusion

Mullerian duct anomalies are rare and usually associated with various clinical manifestations which range from dysmenorrhoea at puberty to preterm delivery and increased rate of caesarean section during reproductive age group. Very sparse literature is available on the didelphys uterus at the present time. It is important for clinicians to rule out such anomalies in a woman of reproductive age group presenting with complaints such as abdominal pain and vomiting. Clinicians should have high index of suspicion of uterine anomaly to make early diagnosis of uterus Didelphys. Pregnancy in a uterus Didelphys deserves early diagnosis and meticulous care in pregnancy and delivery to avert the associated adverse outcomes.

Conflicts of interest/Competing interests: None

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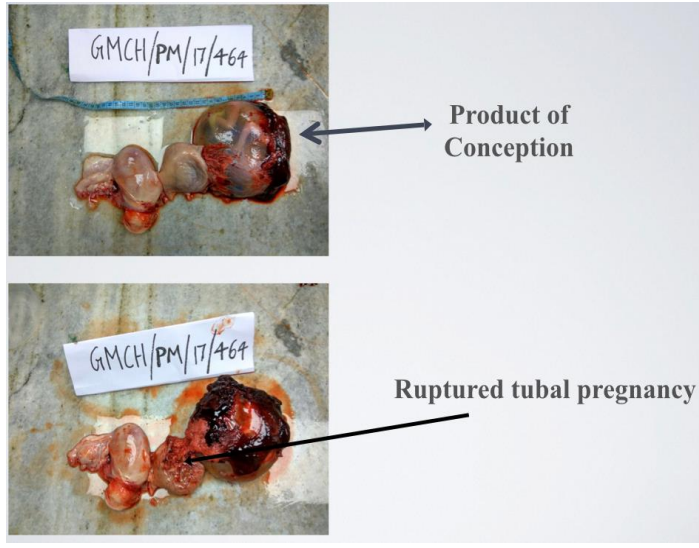

Figure 1: Intact amniotic sac with attached placenta extruded out from the rent