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CASE REPORT

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Sub-hepatic ectopic pregnancy in a fallopian tube of a non-communicating rudimentary uterine horn

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Abstract Ectopic pregnancy in a non-communicating rudimentary uterine horn is a rare occurrence. The authors describe a case of ectopic pregnancy in a fallopian tube of a non-communicating uterine horn. The unusually high position of the ectopic gestation into the sub-hepatic region affects the early clinical presentation. The patient had been treated successfully by laparoscopic salpingectomy.

Keywords Sub-hepatic · Non-communicating rudimentary horn · Ectopic pregnancy · Mullerian · Salpingectomy

Introduction

Ectopic pregnancy in a non-communicating rudimentary uterine horn is rare occurrence. The authors describe a case of ectopic pregnancy in a fallopian tube of a non-communicating uterine horn.

Case report

A 26-year-old nulliparous lady was referred by her GP because of vaginal bleeding for one day associated with right-sided abdominal pain. She had amenorrhoea for approximately 6 weeks and a positive urine pregnancy test. The pain consisted of cramps in the lower abdomen and in the right loin. She had been treated for urinary tract infection with cephalexin for 6 days before presentation. She also complained of nausea, tiredness and dizziness.

Past medical history was unremarkable apart from being under urological investigations for frequency of micturition and recurrent urinary tract infections (UTI). She had undergone urethral dilatation and was on oxybutynin to control the frequency of micturition. Intravenous urography was part of her investigation, which confirmed normal kidneys, ureters and bladder.

She was clinically stable at the time of admission. There was no tenderness in the lower abdomen and no palpable masses; the tenderness was mainly in her right flank. Speculum examination showed a bloody discharge. There was no adnexal tenderness or cervical excitation. Genital swabs were taken. The provisional diagnosis was threatened miscarriage and the patient was allowed home to return the next day for pelvic ultrasound.

Investigations included: urine dipstick, which revealed a trace of proteinuria and a positive pregnancy test, high vaginal swab (HVS) for culture and sensitivity (C&S) and endocervical swab for chlamydia, as well as midstream urine for C&S and full blood count, which were all within normal values (Hb 13.1 g/dl).

The next day, transabdominal ultrasound (USS) showed a normal uterus with a regular endometrium of 6 mm. There was no evidence of a gestational sac or retained products of conception. The right ovary appeared normal, the left was not clearly identified; however, no adnexal pathology was demonstrated. Serum beta hCG assay was added to the last evening blood sample. Generally, the patient was feeling well with no pelvic pain, but had persistent back ache. The pervaginal (PV) bleeding was very light. The possibilities of complete miscarriage, ectopic pregnancy or very early intrauterine pregnancy were discussed at that point with the patient. On the same afternoon, the pain worsened. She was haemodynamically stable, but still tender over the right flank; however, she now also had additional tenderness in right iliac fossa associated with voluntary guarding. There was positive cervical excitation with right adnexal tenderness and the hCG assay was 2,723 μ/l.

A provisional diagnosis of ectopic pregnancy was made and the patient prepared for diagnostic laparoscopy. Beforehand, the next day a transvaginal USS showed that the uterus appeared normal in size and texture, the endometrium measured 8 mm and there was no evidence of a gestational sac within it. The right ovary could not be

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identified this time, but the left ovary appeared normal. There was a moderate amount of free fluid to the right adnexa. Repeat serum beta hCG after 48 h was 3,098 μ/l (abnormal rise). Patient was prepared for an emergency diagnostic laparoscopy ± salpingectomy.

The laparoscopic findings included a unicornuate uterus with a normal left fallopian tube and ovary. There was approximately 400 ml of clotted blood in the pelvis. After suctioning, there was a right rudimentary uterine horn, which was displaced laterally and was adherent to the lateral abdominal wall. There was a thin elongated tubular band of tissue, connecting this horn to the uterus (Figs. 1, 2). Also, an elongated tube starting from this rudimentary horn, extending upwards, adherent to the abdominal wall, ending a few (3–4) cm below the lower edge of the right lobe of the liver. At the distal end of this distorted tube was an ectopic pregnancy, in close proximity with the lower edge of the right lobe of the liver. Normal-looking ovarian tissue was found close to this tube, fixed against the abdominal wall.

Laparoscopic right salpingectomy was performed with bipolar diathermy and scissors (Figs. 3, 4). Histology confirmed interspersed chorionic villi in the lumen of the fallopian tube. The appearances confirmed the presence of products of conception. No features of gestational trophoblastic disease were noted. The postoperative period was uneventful and the patient went home the next day after a thorough explanation.

Discussion

A unicornuate uterus with a rudimentary horn is a mullerian anomaly associated with endometriosis and pregnancy complications, including miscarriage, ectopic pregnancy, uterine rupture, pre-term labour and malpresentation [1].

Mullerian ducts fuse in the midline to form the uterus at about 10 weeks' gestation. Ectopic pregnancy in a rudimentary uterine horn is extremely uncommon. When one of the mullerian ducts fails to form, a single horn (banana-shaped) uterus develops from the healthy mullerian duct. This single horn uterus may stand alone. However, in 65% of women with a unicornuate uterus, the remaining mullerian duct may form an incomplete (rudimentary) horn. There may be no cavity in this rudimentary horn or it may have a small space within it, but no opening that communicates with the unicornuate uterus and vagina. In the latter case, a girl may have monthly pain during adolescence because there is no outlet for the menses from this rudimentary horn. This pain would lead to identification of this problem. In some cases, the rudimentary horn contains a cavity that is continuous with the healthy single horn uterus, but much smaller than the cavity within the healthy uterus. There is a risk that a pregnancy will implant in this rudimentary horn, but because of space limitations, 90% of such pregnancies rupture [2, 3].

There is a risk of pregnancy developing in the rudimentary horn from transperitoneal migration of the sperm or ovum from the opposite side. O'Leary and O'Leary found the corpus luteum to be on the side contralateral to the rudimentary horn containing a pregnancy in 8% of cases

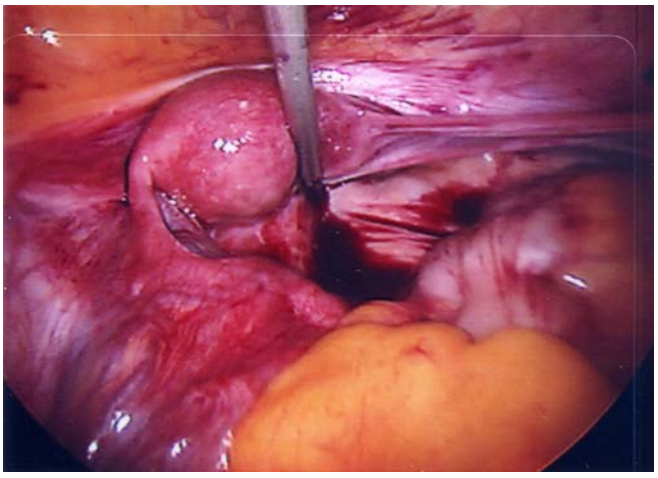

Fig. 2 Uterus with normal left adnexa and right elongated tubular band towards the rudimentary horn

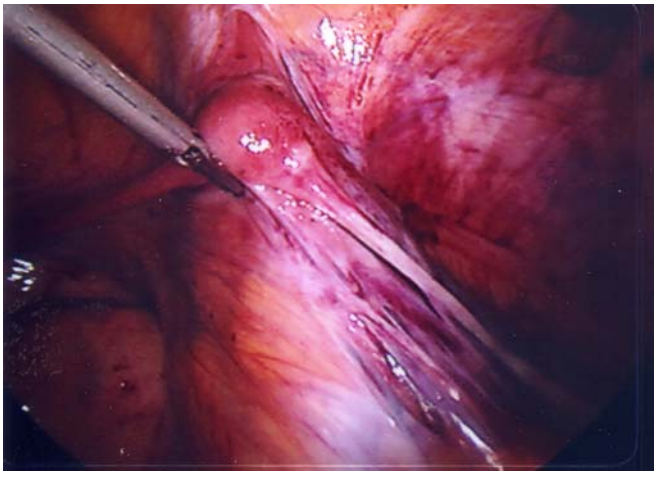

Fig. 1 Rudimentary horn in right pelvic side wall

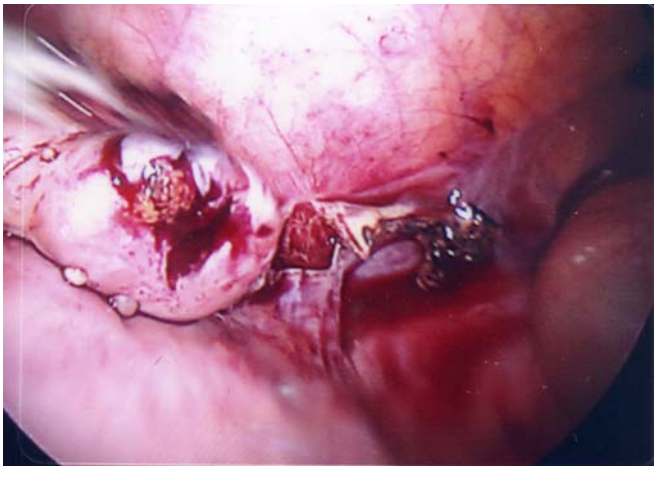

Fig. 3 Tubal pregnancy location just below the right lobe of liver

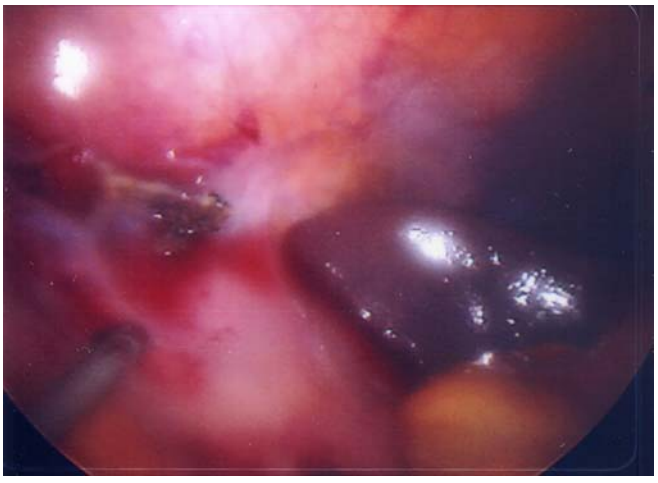

Fig. 4 Subhepatic location of removed ectopic pregnancy

[4]. Signs and symptoms of an ectopic pregnancy will develop with eventual rupture of the horn if the pregnancy is not detected early. Rupture through the wall of the vascular rudimentary horn is associated with severe intraperitoneal haemorrhage and shock. Therefore, removal of the rudimentary horn is required as soon as a pregnancy is confirmed.

The authors described a rare case of a tubal pregnancy in a non-communicating rudimentary uterine horn. We are aware that hundreds of cases of rudimentary horn pregnancy have been published, but this case is unique. Firstly, the pregnancy was in the tube connected to the rudimentary horn; secondly, the rudimentary horn with its tube was in an abnormally high position; and thirdly, the long convoluted fallopian tube was in a sub-hepatic position.

Cases have been reported of tubal pregnancy in a unicornuate uterus with rudimentary horn on the side of the rudiment [5]. Image diagnosis and pathological examination of the rudimentary horn revealed that this uterine malformation was a unicornuate uterus with a non-communicating, non-cavitary rudimentary horn, corresponding to class IIc of the American Fertility Society classification of mullerian anomalies.

The incidence of tubal pregnancy of a rudimentary horn is unclear. Our review of the literature showed that a significant amount of data on pregnancy in a rudimentary horn, however, consisted of very scattered case reports on tubal pregnancy of a rudimentary horn. Therefore, the incidence of rudimentary horn pregnancy is 1 in 100,000–150,000 [6], but the incidence of tubal pregnancy in a rudimentary horn is unclear. The same is applicable to what are recommended as the best diagnostic and therapeutic modalities.

It was postulated that transperitoneal sperm migration constitutes the mechanism by which ectopic pregnancy occurred in non-communicating uterine horn or occluded tube. In women with non-communicating rudimentary uterine horn pregnancies, the ratio of total uterine horn pregnancies to prior contralateral hemi-uterine pregnancies was calculated to infer the overall transperitoneal sperm transmigration rate [7]. The authors concluded that intraperitoneal sperm transmigration occurs approximately half the time in effecting spontaneous human pregnancies. To minimise the risk of ectopic tubal pregnancy in women with unilaterally damaged fallopian tubes, salpingectomy should be the preferred surgical treatment, rather than attempting tubal salvage and repair [7].

Since rudimentary horn pregnancy is a very rare condition, it is not easy to gain experience in diagnosing this entity with ultrasound investigation. Although ultrasound is reported to be a useful tool in diagnosing rudimentary horn pregnancy, this may not be the case in inexperienced hands.

Most pregnancies in a rudimentary horn rupture in the first or second trimester. However, in tubal pregnancy, presentation will be earlier. In our case the localisation of pain was unusual as it was mainly in the right upper abdomen even before rupture and the cause was obvious retrospectively due to the relatively sub-hepatic position of the ectopic pregnancy.

There is a well-known association between mullerian and renal anomalies. Unilateral renal agenesis was found in 13 (38%) of cases [9]. In our case this was ruled out by intravenous urography (IVU).

Diagnosis is usually based on clinical suspicion and aided by transvaginal ultrasound and b-hCG values. In our case, however, the abnormally high position of the tubal ectopic pregnancy meant that it was outside the transvaginal ultrasound field, but there was evidence of pelvic collection. For obvious reasons the time of presentation of our case was similar to that of any other tubal ectopic pregnancy and not delayed as in rudimentary horn pregnancy.

Ectopic pregnancy in a rudimentary horn with a unicornuate uterus has been estimated to be 22%. This indicates removal of rudimentary horn and its tube when diagnosed [8]. In our case, which was non-communicating, we think salpingectomy with diathermy of the distal tubal stump may be appropriate to circumvent future ectopic pregnancies.

Laparotomy has been the treatment of choice when resection of a rudimentary horn is indicated. However, laparoscopic surgery of a rudimentary horn pregnancy is a reasonable alternative in selected cases. Laparoscopy, in these exceptional cases, is the most accurate diagnostic tool that carries significant advantages with effective surgical management, thereby avoiding laparotomy [9].

The prognosis of intrauterine pregnancy is not impaired in the unicornuate uterus, although prematurity may threaten.

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