

A Rare Adnexal Mass in a Teenager

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INTRODUCTION

Congenital anomalies of the Mullerian system are estimated to occur in 0.1–0.3% of the female population (1). Even more uncommon is the occurrence of a unicornuate uterus with a rudimentary uterine horn, caused by the failure of development of one Mullerian duct. The true incidence is hard to determine because patients may never develop gynaecological or obstetrical problems. We present the case of a fifteen-year-old *virgo intacta* who presented with right iliac fossa pain and was found to have this anomaly at laparotomy.

Keywords: Adnexal mass, haematosalpinx, Mullerian duct abnormality

CASE REPORT

The teenaged girl presented to the Accident and Emergency Department of the Sangre Grande Hospital with a one-day history of right lower quadrant pain, described as a constant but dull pain, associated with vomiting. Her last menstrual period was two weeks prior to presentation. She had, within recent months, begun experiencing progressive and worsening dysmenorrhoea as previously her menses were painless.

Physical examination revealed minimal tenderness in the lower abdomen with no guarding or rebound tenderness. An ultrasound scan reported a right tubular, loculated adnexal collection measuring 10 cm x 5.8 cm x 8.7 cm suggestive of a tubo-ovarian collection or an ovarian mass such as a cystadenoma. A normal-anteverted uterus (7.7 cm x 5.9 cm), normal endometrium and normal left adnexa were seen. There was no evidence of free fluid in the pouch of Douglas.

The patient's pain resolved over some days with analgesia and she was next seen in the outpatient clinic about one month later. She was mildly tender in the right lower quadrant and a repeat ultrasound reported a 4.6 cm x 7.5 cm septated mass in the right adnexa, most likely of ovarian origin. All blood investigations were normal including tumour markers (serum beta human chorionic gonadotropin (HCG), alpha-fetoprotein, cancer antigen (CA) 125 and carcinoembryonic antigen (CEA)). The patient was booked for a right ovarian cystectomy with the possibility of an oophorectomy, after counselling and informed consent was obtained from her mother.

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At laparotomy, a large right haematosalpinx, approximately 14 cm x 5 cm x 4 cm was discovered (Fig. 1). Con-



Fig. 1: Right haematosalpinx and uterine horn.

nected to this was an under-developed right uterine horn which was attached by non-communicating fibromuscular tissue to the fundus of a normal appearing left uterine horn (Fig. 2). The right horn had no connection with the cervix



Fig. 2: Right ovary, right uterine horn and fibromuscular band.

and therefore all menstrual blood flowed into the right tube which was blocked at the fimbrial end. The right ovary was grossly normal as was the left adnexa.

A right salpingectomy and hemihysterectomy were performed after identification of the uterine blood vessels

and ureter. She had no intra- or postoperative complications. Histological examination confirmed that the suspected rudimentary uterine mass contained uterine tissue with weakly proliferative endometrium.

The patient subsequently had a computed tomography scan of her abdomen and pelvis that showed no abnormalities of her urinary system.

DISCUSSION

The diagnosis of Mullerian duct anomalies can be very difficult and may be overlooked even at laparotomy (2). In this case, an ovarian cyst would have been the most likely diagnosis given the patient's clinical scenario. As two ultrasounds failed to detect the unicornuate uterus with the rudimentary uterine horn, it demonstrates the limitations of this diagnostic tool. Indeed, magnetic resonance imagery is significantly more accurate in detecting and classifying uterine anomalies although it too has its limitations (3).

The American Fertility Society (AFS) has classified the unicornuate uterus anomaly into four subgroups: rudimentary horn with cavity communicating with a unicornuate uterus (IIa), rudimentary horn with cavity non-communicating (IIb), with no cavity (IIc), and without horn [IIId] (4). Our patient had subtype IIb, the most common subtype and the most clinically significant. Her complaint of progressive severe dysmenorrhea is a common clinical presentation. Other clinical presentations of subtype IIb include haematometra, pyometra and endometriosis attributed to retrograde menstruation.

Delay in diagnosis of these cases can have serious implications. Tanaka *et al* reported a case of iatrogenic dysmenorrhoea in a non-communicating rudimentary horn, which was initially misdiagnosed at laparotomy as a bicornuate uterus (5). The removal of the rudimentary uterine horn is recommended once it is diagnosed and a laparoscopic hemihysterectomy can be performed (6). There is danger of a pregnancy in the rudimentary horn from transperitoneal migration of sperm or ovum from the opposite side. Rupture through the wall of the vascular rudimentary horn is associated with sudden and severe intraperitoneal haemorrhage and shock. Death can occur in a few minutes (7). Heinonen and Pystynen reported an ectopic pregnancy rate of 22% in a unicornuate uterus and rudimentary horn cases (8).

The index case had a right salpingectomy and excision of the rudimentary horn. She remains with two healthy ovaries and a unicornuate uterus. In terms of pregnancy prognosis, however, the unicornuate uterus carries the poorest fetal survival rate of 40% for all uterine anomalies (6). Akar

et al found an incidence of spontaneous miscarriage, preterm labour, intrauterine growth restriction and live birth of 29.2, 43, 10 and 29.2%, respectively in women with unicornuate uteri (9). The abnormal shape, the insufficient muscular mass of the uterus, the reduced uterine volume and the inability to expand may explain the poor obstetric outcome.

The embryological developments of the genital and urinary systems are closely intertwined and therefore abnormalities of both systems are often present. On the side opposite the unicornuate uterus, there may be a horseshoe or a pelvic kidney, or the kidney may be hypoplastic or absent (5). Therefore, when a uterine abnormality is detected, an evaluation of the renal system is indicated (10). Ideally, preoperative evaluation is an important prerequisite to help with the identification and dissection of the ureters before horn resection (11–13). This patient was fortunate as no abnormalities of her renal system were detected.

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