Pregnancy luteoma presenting as ovarian torsion with rupture and intra-abdominal bleeding

Tan M L, Lam S L, Nadarajah S

ABSTRACT

We report a case of pregnancy luteoma, which had undergone torsion in a 33-year-old Indian woman, who presented with severe abdominal pain and decreasing haemoglobin levels at 33 weeks gestation. Ultrasonography showed a right adnexal mass, probably ovarian in origin, with suspicious intratumoral bleed. The pain was treated symptomatically, and the symptoms improved. A successful induction of labour was then performed at 36 weeks gestation. The pain recurred almost immediately after the delivery, and she experienced another intra-abdominal bleed. A diagnostic laparotomy and a right salpingo-oophorectomy were performed, and the diagnosis of luteoma was made based on histology. We discuss the clinical presentation of this unusual tumour, though often asymptomatic, can rarely present with severe abdominal pain from complications like torsion with rupture, leading to massive intra-abdominal bleeding. We also discuss the possible radiological investigations which can be done during pregnancy.

Keywords: intra-abdominal bleeding, ovarian torsion, pregnancy luteoma, pregnancy-associated tumours

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INTRODUCTION

Pregnancy-associated tumours are rare, for which benign conditions, like pregnancy luteomas, must always be considered. Though often symptomatic, they can be associated with acute abdomen secondary to torsion. We discuss the clinical presentation as well as the imaging findings that led to the diagnosis of this rare condition in our patient.

CASE REPORT

Our patient is a 33-year-old woman, with a known history of subfertility, and who underwent successful *in vitro* fertilisation. The pregnancy was uneventful until 33 weeks gestation, when she presented with abdominal pain. There was no associated bleeding per vagina. A full blood count was done, revealing a haemoglobin level of 10.2 g/dL, with a haemotocrit of 28.1%. The total white blood cell count was raised at 17.6×10^9/L, with a raised neutrophil count of 88%. Urinalysis showed a small amount of proteins, as well as red and white blood cells. Physical examination revealed the cervical os to be closed. She was admitted to our institution for observation and was treated symptomatically for possible gastritis or urinary tract infection, with subsequent pain improvement.

Ultrasonography (US) showed a single uterine pregnancy with foetal cardiac pulsations. There was also a large heterogeneous ovoid mass measuring 15.0 cm ×

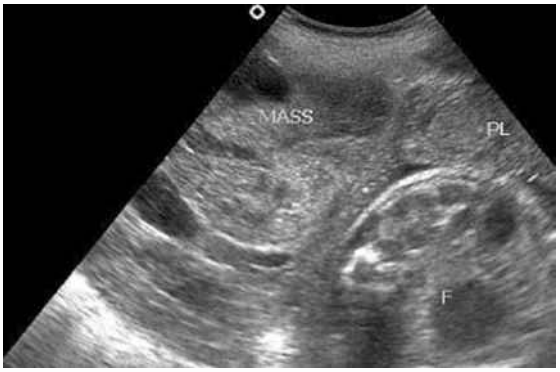

Fig. 1 Initial transabdominal US image obtained at 33 weeks gestation shows a large heterogenous abdominal mass which is separate from the uterus. The foetus (F) and the placenta (PL) are seen.

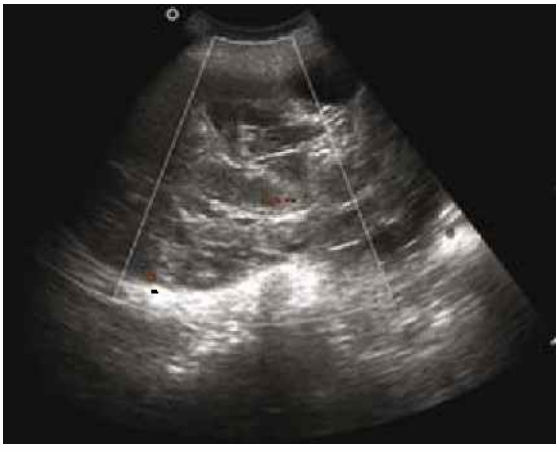

Fig. 2 Transabdominal US image obtained at 33 weeks gestation shows that the right hypochondrial lesion had a heterogeneous echotexture, with solid and cystic components. Echogenic foci were noted within the tumour, which was thought to represent intratumoral bleeding. Flow was demonstrated within the lesion.

Department of Diagnostic Imaging, KK Women's and Children's Hospital, 100 Bukit Timah Road, Singapore 229899

Tan ML, MBBS
Registrar

Lam SL, MBBS, FRCR, FAMS
Senior Consultant

Reproductive Medicine Unit

Nadarajah S, MBBS, MRCOG, FAMS
Senior Consultant

Correspondence to:
Dr Mark Tan
Tel: (65) 6394 2284
Fax: (65) 6315 6834
Email: minglong@cyberway.com.sg

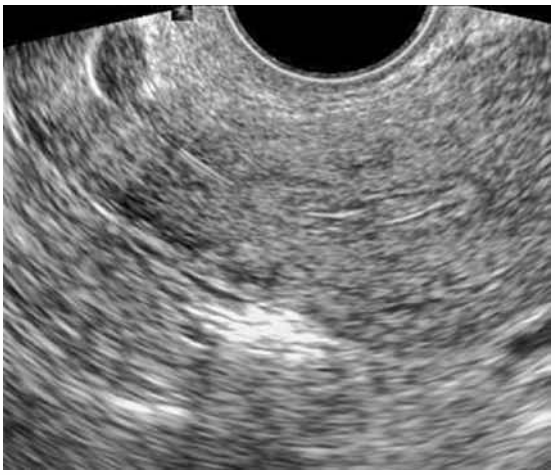

Fig. 3 Transvaginal US image obtained one year before the onset of symptoms shows a slightly bulky uterus with a heterogeneous echotexture, probably due to uterine fibroids.

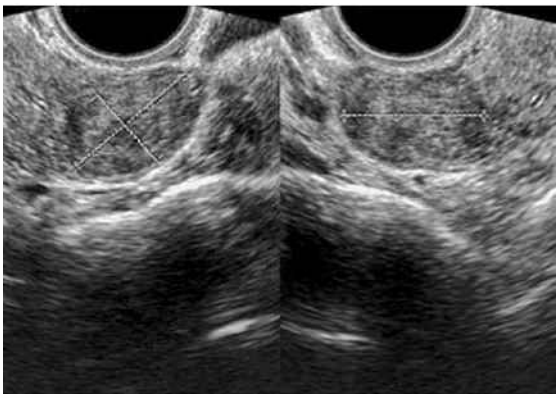

Fig. 4 Transvaginal US image obtained one year before the onset of the symptoms and pregnancy shows a 2.5 cm x 1.6 cm pedunculated right fibroid, with no other large adnexal mass.

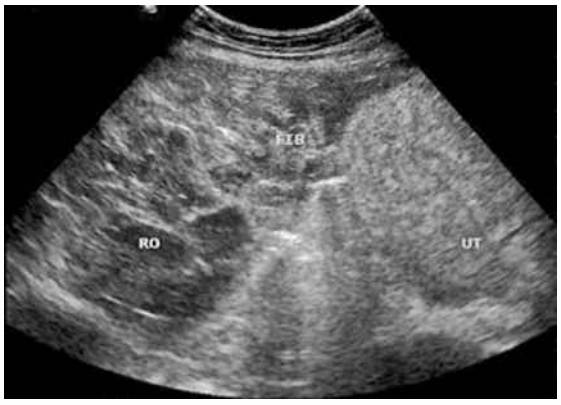

Fig. 5 Transabdominal US image obtained post-delivery because of persistent abdominal pain shows the previously-described lesion to be likely ovarian in nature, as the pedunculated fibroid could be identified separately from the lesion. Echogenic fluid around the tumour represesnts intraperitoneal bleeding.

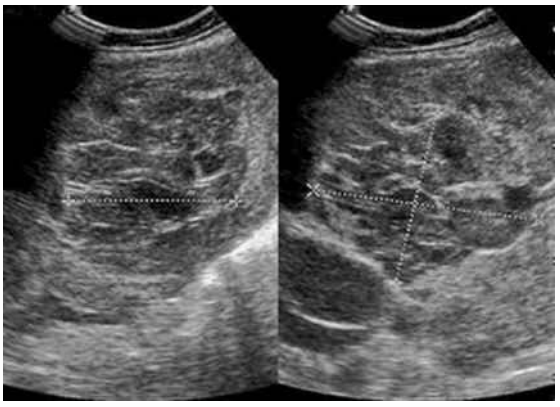

Fig. 6 Transabdominal US image shows the lesion to be arising from the right ovary and measures 7.5 cm × 9.3 cm. The heterogeneous nature of the tumour is clearly identified, with solid and cystic components. Adjacent echogenic foci were thought to be blood products.

13.7 cm × 10.5 cm in the right hypochondrium, separate from the uterus (Fig. 1). There were multiple irregular cysts seen within, and venous flow was detected within the mass (Fig. 2). Echogenic foci were also noted within the mass, which led to suspicions that it was intratumoral. The patient had US done a year ago which revealed a 2.5-cm pedunculated fibroid and a bulky uterus with a heterogeneous echotexture, probably due to small fibroids (Figs. 3 & 4). The ovaries were unremarkable and no large adnexal mass was seen. Given the rapid development of this large adnexal mass, the clinical impression was that of a possible enlargement of the previously-noted pedunculated fibroid, due to intratumoral bleeding.

With the working diagnosis of a bleeding pedunculated fibroid, the plan was for induction of labour at 36 weeks gestation. The patient's symptoms only improved marginally with the symptomatic treatment. A repeat blood count done two days later revealed that the haemoglobin had fallen to 7.2 g/dL, and the haemotocrit had also fallen to 23%. In view of the blood loss, the patient was given two pints of packed cell transfusion. The patient's haemoglobin improved to a value of 8.5 g/dL after the transfusion. The patient's haemoglobin levels were monitored with regular full blood counts. The haemoglobin levels then improved to about 13.2 g/dL. At 36 weeks gestation, the patient had a successful induction of labour, with a successful normal vaginal delivery of a baby boy.

Shortly after delivery, she again developed right hypochondrial abdominal pain. A full blood count was then repeated, and it revealed that the haemoglobin had fallen to about 9 g/dL. Repeat US showed a large postpartum uterus, with a large complex adnexal mass with cystic areas, as was previously detected (Figs. 5 & 6). Flow was detected in the tumour. On this occasion, the mass could be seen to be separate from the prenatally-detected large

pedundulated fibroid. It was concluded that the mass was probably more ovarian in origin than from the uterus (Fig. 5). The radiological impression was that of a possible hyper-stimulated ovary. There were echogenic foci noted around the mass, suggestive of intraperitoneal bleeding. The clinical impression was that a possible acute abdomen secondary to a ruptured ovarian cyst with intra-abdominal bleeding. Decision was made for an urgent laparotomy.

Intraoperatively, about 1 L haemoperitoneum was detected. The intraoperative haemoglobin fell to about 4.9 g/dL, and she was given about three pints of packed cell transfusion. A large 15-cm cystic ovarian tumour was also discovered, which appeared to have undergone torsion. It appeared solid with extensive areas of haemorrhage and oedema, probably accounting for the large amount of haemoperitoneum detected. No tumour deposits were noted in the subdiaphragmatic surface, omentum or peritoneum. The left ovary was unremarkable, and no large intra-abdominal lymph was detected. A right successful salpingo-oophorectomy was performed and haemostasis was secured.

Histological examination showed that the right ovary had extensive areas of haemorrhage with infarction and stromal oedema. There was a circumscribed tumour with a lobulated outline filled with multiple cells with features of luteinisation. Proliferation of spindle cells with large areas of oedema was also seen. The serosa tumour was intact. Despite the cellularity of the tumour, there were no histological features of cytologic atypia, tumour-type necrosis or increased atypical mitotic activity. The histological diagnosis was likely to be a lutenised form of the coma, which given the clinical history, was likely a pregnancy luteoma. The extensive haemorrhage, infarction and oedema accompanying the tumour were likely to be due to the effect of torsion. The chronic pain the patient experienced may have been due to irritation of the peritoneal lining by the blood. Postoperatively, in view of the profound intraoperative bleed, the patient was sent to intensive care unit for monitoring. She was found to be stable and was sent to the general ward shortly after and was discharged on the fifth postoperative day.

DISCUSSION

Pregnancy luteoma is a rare non-neoplastic lesion characterised by ovarian enlargement during pregnancy.[1] The lesion occurs usually in the third and fourth decades of life, and about 80% are associated with multiparous women. The tumour is characterised by well-circumscribed, soft, fleshy tumours with frequent foci of haemorrhage. It ranges from a microscopic size to over 20 cm in diameter. One-third of the tumours are bilateral, and half are multiple. The aetiology is postulated to be due to replacement of the normal ovarian parenchyma by solid proliferation of luteinised stromal cells, under the influence of human chorionic gonadotropin. These lesions are not associated with excessively high levels of human chorionic levels, and are only seen in the pregnancy. This is in contrast to gestational trophoblastic disease, which is associated with very high levels of human chorionic gonadotrophin.

It is the stromal cells which are stimulated, and hence these lesions may be associated with high levels of androgen.[1-2] Virilisation of mother and foetus can occur in about 25%–30% of cases, of which 25% occur in the latter half of pregnancy. The testosterone levels are often elevated 70 times above normal, even in non-virilised patients. Of note, only two-thirds of the female babies of virilised patients are virilised, as the placenta aromatises androgens to progesterone. Maternal serum testosterone often returns to normal two months after delivery.[3]

Other pregnancy-associated tumours include, solitary lutenised follicular cysts, hyperreactio luteinalis, corpus luteum of pregnancy, granulosa tumours and steroid cell tumours.[4] Hyperreactio luteinalis is characterised by bilateral ovarian enlargement in the presence of numerous follicular cysts, but there is no association with virilisation of the newborn. The tumour is classically more cystic than when compared to pregnancy luteomas. All the other benign conditions are also not associated with virilisation, and all resolve after pregnancy. Malignant virilising tumours do not regress after pregnancy.

Pregnancy luteomas are often asymptomatic, and are often discovered incidentally at the time of the caesarian section. They are all self-limiting, and commonly resolve after three months of pregnancy. Given the size of some of these tumours, they may be associated with mass effect on adjacent organs like the ureter, presenting with symptoms of obstructive uropathy,[1] or are complicated by infarction, which can lead to ovarian scarring. They can rarely be associated with torsion, as in our case, and present with severe abdominal pain. The tumours can rupture, leading to massive blood loss, hence necessitating surgical management, as seen in our case. There have also been rare reports of pregnancy luteomas presenting like malignant tumours with massive ascites and raised CA-125.[5]

Diagnosis of pregnancy luteomas should be made with minimally-invasive modalities. It is important to provide appropriate medical/surgical intervention without disturbing the pregnancy iatrogenically or causing unnecessary maternal morbidity.[6] Modalities of investigation which can be used in this condition include magnetic resonance (MR) imaging and US.

For MR imaging, there have been several case reports on its usefulness.[2] It can be used as an adjunct to determine if the tumour is virilising or not virilising, as

virilising tumours often show relative high signal intensity on T1-weighted images and low signal intensity on T2-weighted images (due to the colloid material in the cells), while other neoplastic solid tumours show characteristic low signal intensity on the T2-weighted images. Cystic non-hormonally active tumours show high signal intensity on T2-weighted images with thick septa. Though some virilising pregnancy luteomas show characteristic features of MR imaging, it must be pointed out that in most cases, the MR imaging findings were similar to those of other cystic tumours or tumour-like lesions of the ovary.[6] Despite its limitations, with good clinical assessment, combined with MR imaging findings, pregnancy luteoma can be diagnosed, and obviates the need for unnecessary operations or interruption of pregnancies.

Similarly, for US, with careful consideration of the clinical history, accurate diagnosis of pregnancy luteomas can also be made. The classical appearance of pregnancy luteomas is solid, and can be identified as a solid tumour on US.[7] However, these tumours can also have a marked cystic appearance due to extensive necrosis, and mimic the appearance of other tumours,[4] reinforcing the need for clinical correlation. Pregnancy-associated tumours are rare, for which benign conditions, like pregnancy luteomas, must always be considered, especially when associated with virilisation. Though often symptomatic, they can be associated with acute abdomen secondary to torsion and rupture with a large associated intra-abdominal bleed, as was seen in our case. Investigations must be minimally invasive with no significant detrimental effects to the developing foetus, and imaging modalities like MR imaging and US, with sound clinical judgment, have shown promise in being able to help diagnose the tumour, and prevent unnecessary interruption of pregnancies.

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