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Case Report

Ruptured arteriovenous malformation secondary to placental site trophoblastic tumor: A diagnostic dilemma and its successful management

Avantika Gupta[a,*], Usha Manaktala[a], Nita Khurana[b]

[a] Department of Obstetrics & Gynaecology, Maulana Azad Medical College, New Delhi, India
[b] Department of Pathology, Maulana Azad Medical College, New Delhi, India

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ABSTRACT

Placental site trophoblastic tumor is a very rare neoplasm and is a subtype of gestational trophoblastic neoplasm. Owing to its rarity and varied clinical manifestations, it can lead to a diagnostic dilemma. We report a very rare case of PSTT presenting as massive hemoperitoneum caused by rupture of secondary arteriovenous malformation. MR angiogram revealed the presence of arteriovenous malformation and the diagnosis of the neoplasm was confirmed on histopathology and immunohistochemistry. Hysterectomy was done and the patient was managed successfully.

1. Introduction

Placental site trophoblastic tumor (PSTT) is a rare neoplasm derived from intermediate trophoblast without a significant component of synctiotrophoblast. It accounts for 0.4–2% cases of gestational trophoblastic disease [1]. Owing to its rarity, the epidemiology, etiology and risk factors are still unclear. It usually occurs in women with childbearing age. Choriocarcinoma has been described as a cause of acquired uterine arteriovenous malformation (AVM) [2,3], but PSTT has never been described as a cause of secondary AVM in the literature. We are describing first case of ruptured AVM presenting as massive hemoperitoneum caused by PSTT.

2. Case report

A 28 year-old woman, para one presented to emergency room with chief complaints of acute onset abdominal pain and distension which developed & increased over 2 days. Her last child birth was two years ago and she had a preceding amenorrhoea for last one year. She also gave history of significant weight loss over 6 months, however there was no history of fever. On examination, she had kyphoscoliosis, was cachexic and had a mild pallor. She had a pulse rate of 100 per minute and a blood pressure of 110/68 mmHg. Her abdomen was distended, which revealed blood on paracentesis. A urine pregnancy test was positive. Her serum βhCG levels was 250 mIU/mL. An urgent ultrasound was done, which showed multiple degenerated fibroids with high vascularity along with one litre of hemoperitoneum, although there were no adnexal masses. In view of positive pregnancy test along with hemoperitoneum, she was taken up for exploratory laparotomy. Intraoperatively, she had a hemoperitoneum of around 2 L. Uterus was 14.0 × 8.0 × 5 cm in size. The surface was irregular containing multiple raised areas which appeared to be cystic. A 5 × 5 cm cavity was seen at the right cornual end containing necrotic material along with few bleeders at its margins (Fig. 1). The pelvic vasculature was dilated & tortuous. Margins of this cavity were trimmed and hemostatic sutures taken to control bleeding. The tissue was sent for histopathology and the necrotic material present inside was sent for bacterial as well as mycobacterial culture. An abdominal drain was inserted & the abdomen was closed. A gentle endometrial curettage was done & the tissue was also sent for histopathology. She received two units of blood & four units of fresh frozen plasma and was shifted to intensive care unit. Histopathology of the trimmed myometrial margin was mainly degenerated & showed few trophoblast cells in between myometrial bundles which stained for β hCG. Endometrial tissue obtained was inadequate for any opinion. Bacterial culture was sterile & there was no evidence of tuberculosis. A repeat β hCG level was 220 mIU/mL two weeks after surgery. A provisional diagnosis of placental trophoblastic tumor was made owing to a persistently low β hCG levels and trophoblast cells in the myometrium on histopathology. So, serum human placental lactogen levels was obtained which was raised to term pregnancy levels i.e., 9.6 ng/mL. This confirmed our diagnosis and a metastatic workup was also done including ultrasound pelvis along with Doppler study, MRI pelvis and abdomen along with MR angiography and CT chest and brain. There were no extrauterine metastatic deposits.

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* Corresponding author at: House No. 93/94, Pocket-2, Sector 22, Rohini, Delhi 110086, India.
E-mail address: dravantikagupta@gmail.com (A. Gupta).

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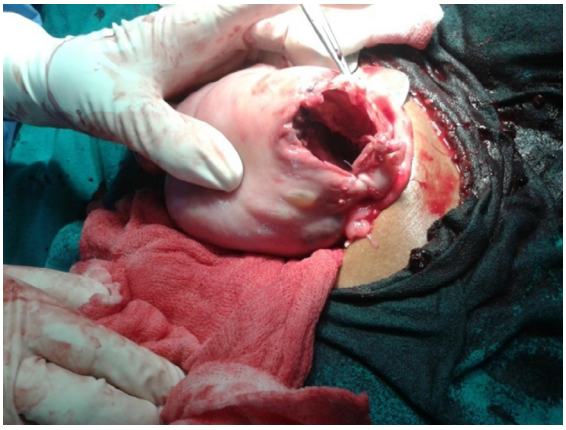

Fig. 1. An enlarged uterus showing multiple necrotic areas extending deep into the myometrium.

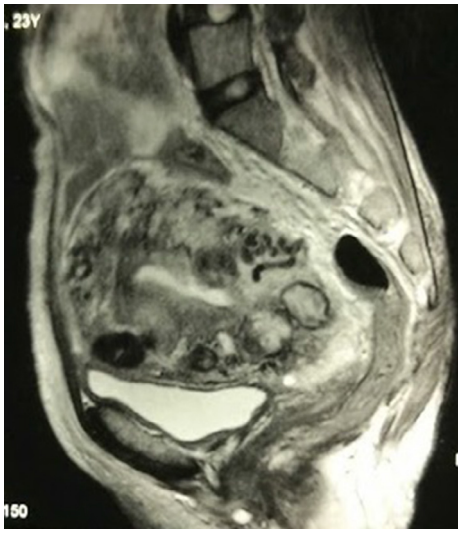

Fig. 2. MRI film showing enlarged uterus with multiple serpigineous flow voids and intramural dilated vessels involving whole myometrium.

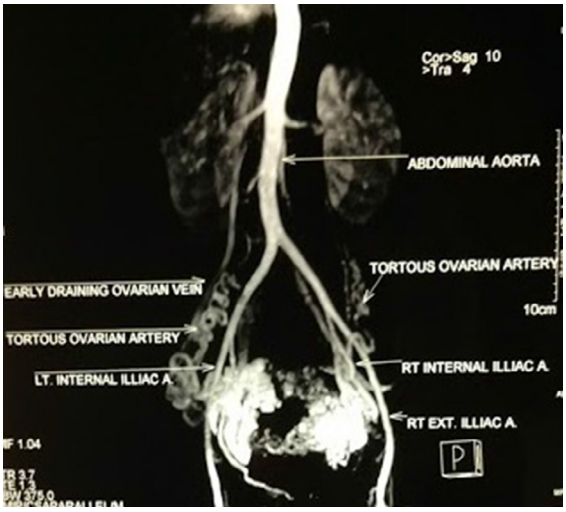

Fig. 3. MR angiogram showing bilateral tortuous ovarian, uterine and iliac arteries along with early draining veins suggestive of arteriovenous malformation.

Three weeks after surgery, a repeat ultrasound again showed highly vascular areas within myometrium on colour Doppler mainly arterial flow which had a high velocity and low resistance suggestive of arteriovascular malformation. MRI examination was performed in 3 Tesla scanner including T1 weighted, T2 weighted & fluid attenuated inversion recovery (FLAIR) sequences. It showed an enlarged & heterogenous uterus with evidence of multiple serpiginous flow voids involving whole myometrium extending into bilateral parametrium with indistinct endo-myometrial junction also suggestive of arteriovenous malformation (Fig. 2). On MR angiography, multiple tortuous vascular channels were seen in pelvis with feeders from enlarged tortuous bilateral ovarian & internal iliac arteries (Fig. 3). Early draining veins were also noted into bilateral ovarian veins & internal iliac veins (Fig. 3). Rapid and massive venous filling was noted through arteriovenous fistulae before contrast could reach the finer arterial branches.

A preoperative embolisation of the feeding vessels was done with polyvinyl alcohol particles two days prior to the surgery and a total abdominal hysterectomy was done thereafter. Consequently, the intraoperative blood loss was only 600 mL. The uterine specimen was sent for histopathology. On gross examination, multiple haemorrhagic ill-defined nodular areas were present in the myometrium & endometrium ranging from 1 to 3 cm. On histopathology, large areas of necrosis were seen in the myometrium, with intermediate trophoblast cells extending till serosa as well as endocervix. These intermediate trophoblasts seen as polyhedral cells showed nuclear atypia with high mitotic figures (Fig. 4). The tumor cells infiltrated between muscle fibers, and showed apparent tumor hemorrhage and necrosis (Fig. 4). Chorionic villi and cytotrophoblasts were not identified. Tumor cells expressed human placental lactogen (Fig. 5), epithelial membrane antigen, cytokeratin & focally expressed β hCG on immunohistochemistry. So, based on the above findings, a final diagnosis of placental site trophoblastic tumor was made. Patient did well in the postoperative period and was discharged on postoperative day 14 in good health. At three weeks, her human placental lactogen level was repeated which was 0.8 ng/mL, ie in the normal range. At six months follow up, patient is asymptomatic and is doing well.

3. Discussion

Placental site trophoblastic tumor is a rare variant of gestational trophoblastic disease. It can occur after a normal pregnancy, abortion, ectopic or molar pregnancy [4]. PSTT produces masses in which necrosis is marked. It differs from choriocarcinoma immunocytochemically because majority cells express hPL than hCG. The most common presenting features are vaginal bleeding followed by amenorrhoea. It is composed of a monomorphic population of intermediate trophoblasts of the placental bed which show invasion. The

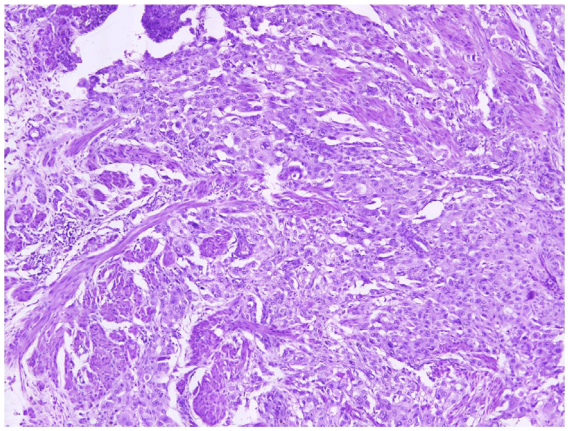

Fig. 4. Intermediate trophoblast cells seen in nests and large masses interdigitating between myometrial bundles.