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PLACENTAL SITE TROPHOBLASTIC TUMOUR: A CASE REPORT

Ruby Bhatia* — Professor and Head of Department of Obstetrics and Gynaecology, Maharishi Markandeshwar deemed to be university, Mullana, Ambala, Haryana, India. *Corresponding Author

Reena Bisht — Post Graduate Student, 3rd year, Department of Obstetrics and Gynaecology, Maharishi Markandeshwar deemed to be university, Mullana, Ambala, Haryana, India.

Sukhbir Pal Kaur — Associate Professor Department of Obstetrics and Gynaecology, Maharishi Markandeshwar deemed to be university, Mullana , Ambala, Haryana, India.

ABSTRACT Placental site trophoblastic tumour (PSTT) is a rarest type of gestational trophoblastic disease (GTD) with an incidence of 1/50,000–100,000 of all pregnancies. In contrary to other variants of GTD, it arises from the intermediate trophoblast at placental site, secretes minimal beta-hCG and very less sensitivity for chemotherapeutic agents. Patient presents with amenorrhoea and abnormal bleeding. Imaging modalities like ultrasonography and MRI helps in making the diagnosis but is confirmed after histological evaluation and immunohistochemistry (IHC) studies. Patients mostly present with tumour limited to the uterine body. The mainstay of treatment is total hysterectomy. The prognosis of patients with tumour limited to the uterus is good.

KEYWORDS : Gestational trophoblastic disease (GTD), Immunohistochemistry(IHC), Placental site trophoblastic tumour (PSTT), Serum beta - human chorionic gonadotrophin (HCG)

INTRODUCTION

Placental site trophoblastic tumor is an extremely rare type of gestational trophoblastic neoplasia, arising from intermediate trophoblastic cells without chorionic villi infilteration at the placental site. The incidence of PSTT is about 1/50,000–100,000 of all pregnancies and accounts for 0.23% to 3.00% of all Gestational trophoblastic neoplasia, while its mortality being 25% .[1,2] Studies show that only 300 cases have been reported in the literature.[3] It is characterised by modestly elevated beta HCG and slow growth.[4] Therefore it is usually resistant to chemotherapy. PSTT poses significant diagnostic dilemma to the clinician due to its uncharacteristic presentation, indeed only case of PSTT in 36years of clinical experience.

CASE REPORT

A 40-year-old Indian woman (Para 2, living 2, miscarriage 1) presented in the emergency department of a tertiary care hospital with chief complaints of irregular bleeding per vaginum since last six months. History dates back to one year (January 2019) when patient had full term vaginal delivery at a private hospital in Haryana of a healthy male baby with birth weight 2.8kgs. According to the records complete placenta with the membranes was removed. Postpartum period was uneventful. She had lactational amenorrhoea for six months followed by menstruation. Since then she had irregular, heavy bleeding per vaginum with passage of clots, persist for 2-3 days with no set pattern. It was not associated with dysmenorrhoea. There was no history of post coital bleeding. Patient kept taking over the counter medication for the same, but was not relieved till she reported in emergency department on 7th January 2020. Menstrual cycles prior to last childbirth were regular lasting 5-6 days every 28-30 days, using 2-3 pads per day, not associated with passage of clots and dysmenorrhoea.

Patient had two full term normal vaginal deliveries at home, her last being one year ago. She also reported undergoing Dilation and evacuation two and a half year back following a spontaneous abortion at the gestational age of one and a half months. There was no history suggestive of coagulation disorder or medical co-morbidities.On admission her was BP-110/80 mm Hg, PR-108/min, afebrile and maintaining a saturation of 98% on room air. Patient appear severely anaemic(pallor++). Her cardiovascular and respiratory system examination was normal. There was no organomegaly .On local examination external genitalia appeared normal. On per speculum examination vaginal mucosa was pale and cervix was healthy with minimal bleeding.On bimanual per-vaginum uterus was ten weeks size, soft, mobile, non tender and bilateral fornices were free. There was blood present on gloved finger.Laboratory examination: Urine pregnancy test was negative. Haemoglobin was 6.3gm/dl with peripheral blood film suggestive of microcytic hypochromic anaemia and serum beta-HCG level was 46.8 mIU/ml. Renal function test, liver function test and coagulation profile was normal. X -ray chest and ECG were normal. Colour Doppler ultrasound pelvis reported 9.5 x 5.4 cm uterus with multiple large cystic spaces in anterior myometrium showing abundant blood flow with high vascular score. MRI pelvis revealed a well-defined lesion in anterior myometrium measuring 64 mm x 45 mm x 54 mm, predominantly hyperintense in T2 with multiple serpiginous flow voids. Endometrium was pushed posteriorly. The flow voids were communicating with similar vessels in bilateral parametrium and hence to bilateral internal iliac vessels, with intense post contrast enhancement suggestive of Uterine AV malformation.

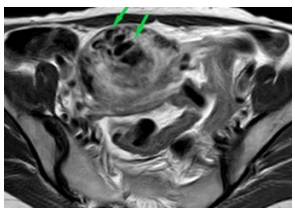

Figure 1:Axial T2 MRI image section showing multiple enlarged vascular flow in a serpiginous pattern (marked as arrow)

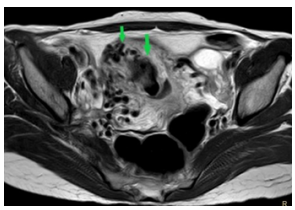

Figure 2: Axial T2 MRI image section showing multiple enlarged vascular flow (marked as arrow)

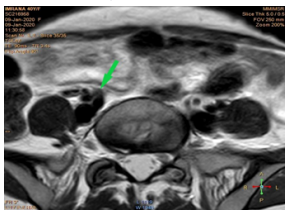

Figure 3: Connection between the enlarged serpiginous flow voids showing connection with the right iliac vessels (marked as arrow)

Patient was build up with four units of packed red blood cell transfusion and planned for hysteroscopic guided endometrial aspiration and biopsy under general anaesthesia after informed and

written consent. Patient started bleeding profusely during the procedure. Resuscitation with massive blood transfusion was initiated. Oxytocin infusion and balloon tamponade was tried to arrest the profuse bleeding per vaginum but to no success despite conservative management. She was taken up for emergency total abdominal hysterectomy after taking informed consent. On gross examination uterus was 10x5 cm, cut section uterine corpus showed grey-brown to grey-black nodular area in the endometrial cavity measuring 3x2x1.5 cm infiltrating less than one third of the myometrium. Histopathology examination was suggestive of Placental site trophoblastic tumour.

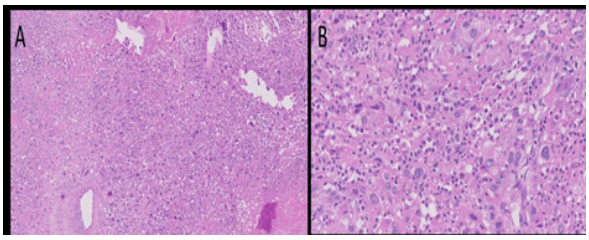

Figure 4-Microscopic findings-Section shows infiltrative tumor arranged in cords and sheets composed of round to polyhedral trophoblastic cells exhibiting marked nuclear atypia with large convoluted hyperchromatic nuclei and eosinophilic cytoplasm, infiltrating less than one third of myometrium. There was no significant increase in mitotic activity(3-4/10 HPF).

Figure 4 Histology of PSTT shows only intermediate to trophoblastic cells without chorionic villi infiltration at the placental site and no syncytiotrophoblast. On the contrary, histology of choriocarcinoma shows haemorrhagic mass consisting of both cytotrophoblast and syntitiotrophoblast along with intravascular growth. However our case started profusely bleeding at the time of hysteroscopy guided endometrial biopsy where in emergencytotal abdominal hysterectomy was performed. Thus confirming PSTT as retrospective diagnosis on hystectomy specimen.

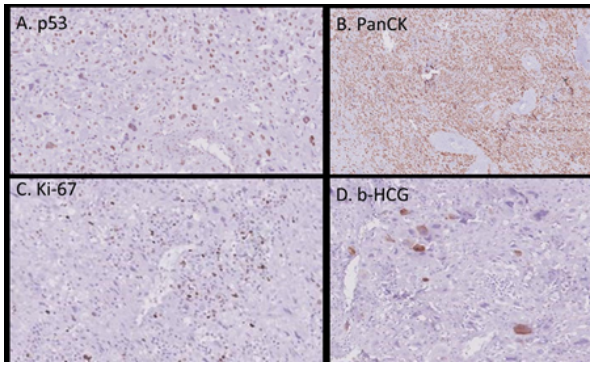

Figure 5-IHC Findings-A.-Overexpression of p53 seen; B. Tumor was strongly positive for PanCK; C. Low to intermediate grade index for Ki-67 was seen; D.Tumor was focally positive for b-HCG

Figure 5 showing Immunohistochemistry of the specimen. It was confirmed by IHC studies of tumor cells showing overexpression of p53, positive for PanCK, focally positive for beta-HCG, negative for inhibin, p63 and placental alkaline phosphatase (PLAP) and low to intermediate grade index for Ki-67. There were minimal areas of coagulative necrosis, no lymphovascular invasion and mitotic activity of 3-4/10HPF. Post operative period was uneventful. The serum beta-HCG levels were 38.1 mIU/ml and 0.07mIU/ml on postoperative day 2 and day 7 with discharge on post operative day 10. Patient is on three monthly follow up for one year. There is no complain of cough or bleeding per vaginum. General and local examination is normal. Serum beta HCG levels are negative, X-rays chest and ultrasounds pelvis and abdomen is within normal limits.

DISCUSSION

PSTT typically presents during reproductive age group after full-term delivery as in our case or rarely following therapeutic or spontaneous abortions, ectopic or a molar pregnancy.[5] Our patient presented 12 months after full term vaginal delivery. The most common presentation is amenorrhea, irregular bleeding per vaginum and enlarged uterus on local examination.

Serum beta-HCG is mildly elevated in PSTT. According to a study done at Charing Cross Hospital, the levels were below 1000mIU/mL in 79% and below 500mIU/mL in 58% of cases.[6] Our patient's beta-HCG was only mildly raised with value of 46.8 mIU/mL.

The diagnosis is confirmed by a histological examination along with immunohistochemistry studies of endometrial specimen.. Figure 1 shows only intermediate trophoblastic cells without chorionic villi infiltration at the placental site and no syncytiotrophoblast. On the contrary, histology of choriocarcinoma shows haemorrhagic mass consisting of both cytotrophoblast and syncytiotrophoblast along with intravascular growth. However our case started profusely bleeding at the time of hysteroscopy guided endometrial biopsy where in emergency total abdominal hysterectomy was performed. Thus confirming PSTT as retrospective diagnosis on hysterectomy specimen.

Figure 2 showing IHC staining generally shows weak and focal positive for beta HCG and diffusely positive for cytokeratin.[7] PLAP is secreted from syncytiotrophoblast cells and mostly absent in case of PSTT. Even in our case IHC studies revealed overexpression of p53, positive for PanCK, focally positive for beta-HCG, negative for inhibin, p63 and placental alkaline phosphatise (PLAP) and low to intermediate grade index for Ki-67. IHC confirmed the diagnosis of PSTT.

It is mostly benign and develops within the uterus. Metastasis is seen in around 30% of cases. Most common sites of metastasis are lungs, liver, vagina, gastrointestinal tract, brain and lymph nodes.[7]In our case there was no evidence of metastasis.

The most important predictor of prognosis is the extent of disease. Survival rate is more than 90% for nonmetastatic disease.[6] According to a study by Papadopoulos et al, showed that an interval of more than 2 years from antecedent pregnancy to the diagnosis of PSTT was associated with a mortality rate of 64%.[6] Thus prolonged interval from the antecedent pregnancy is associated with adverse outcome and poor survival. Pathological features associated with poor outcome are deep myometrial invasion (more than one third), extensive necrosis and high mitotic rate (>5/HPF).[7] In our case tumour occurred one year after term delivery with infiltrating less than one third of the myometrium and mitotic activity of only 3-4/10HPF. There was no evidence of metastasis or any high risk factors present. Hence probability of a good survival and low recurrence may be anticipated.

The main modality of treatment for patients with PSTT is surgery. Patients with disease localized to the uterine body should preferably undergo hysterectomy because PSTT is relatively resistant to chemotherapy.[8] In our case we performed emergency total abdominal hysterectomy as patient started bleeding profusely at the time of hysteroscopic guided endometrial biopsy. Metastasis to the ovaries is very unlikely. Hence oophorectomy does not improve the prognosis or prevent metastasis. Patients with metastatic disease to vagina, lungs and lymph nodes should undergo resection of the disease. Another option for patients not motivated for hysterectomy is hysteroscopic resection or trans-peritoneal local uterine excision.[8] All patients with metastatic PSTT and high risk factors on pathological examination should be considered for chemotherapy.[9]

Our patient did not require chemotherapy as the disease was limited to inner one third of the myometrium and no high risk factors present on pathological examination.

CONCLUSION

Differential diagnosis of GTD should always be kept in mind in all patients of reproductive age group presenting with abnormal uterine bleeding preceded by delivery. PSTT does not have any characteristic presentation. Hence, diagnosis can only be made after radiological and histological examination and confirmed by IHC studies.

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