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PLACENTAL SITE TROPHOBLASTIC TUMOR COMPLICATING PREGNANCY- A CASE REPORT

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ABSTRACT
Placental site trophoblastic tumor (PSTT) is a very rare form of gestational trophoblastic diseases, which represent the neoplastic transformation of intermediate trophoblastic cells that normally play a critical role in implantation. They are commonly seen in reproductive age group and can occur after normal pregnancy, abortion or following gestational trophoblastic disease. We present this case of PSTT in an obstetric hysterectomy specimen done for severe postpartum haemorrhage with a fibroid complicating pregnancy.

Abbreviations: βHCG - beta Human Chorionic Gonadotrophin; FIGO - International Federation of Gynecology and Obstetrics; GTN - Gestational Trophoblastic Neoplasm; GTD-Gestational Trophoblastic Diseases; HPL - Human Placental Lactogen; pMBP - pregnancy- associated Major Basic Protein; PSTT - Placental Site Trophoblastic Tumor.

INTRODUCTION

Placental site trophoblastic tumour (PSTT) is a rare and unique form of gestational trophoblastic disease which constitutes 1 – 2 % of all gestational trophoblastic neoplasia (GTN) [1, 2] and accounts for 0.5% of GTD worldwide [3]. This entity was was first described in 1976 by Kurman et al as 'trophoblastic pseudotumor' [4,5,6] and was later adapted to PSTT in 1981, by the work of Scully and Young, who recognized its malignant potential [1,4,5,6]. It is important to make the distinction between PSTT and other GTD as it is less chemosensitive and adverse effects are more common [4].

CASE REPORT

A 36-year-old lady (G3P1L1A1), with history of previous caesarean section, came for safe confinement at 36 weeks of gestation. Her first child was delivered at full term by caesarean section 7 years ago. After 6 years she had an abortion at 3 months of amenorrhoea, following which she conceived immediately and was on regular follow up. The initial ultra-sonogram showed a fibroid in the anterior wall of uterus. Further scans done had no mention of the fibroid. She continued her pregnancy till term and baby was delivered by elective caesarean section following which she had uncontrollable postpartum hemorrhage for which emergency hysterectomy was done and the specimen was sent for histopathological examination.

Grossly the gravid uterus was hypertrophied. On sectioning, a poorly circumscribed grey white mass was seen in the myometrium, infiltrating up to the serosa. The endometrial cavity showed greyish black discoloration. [Figure 1].

Microscopically, the myometrium showed nodules of intermediate trophoblastic cells permeating

between the smooth muscle fibres and around blood vessels[Figure 2]. Invasion of the vessel wall with fibrinoid change and preservation of lumen were also seen. The tumour cells are medium to large sized and mononuclear, with mild nuclear atypia, prominent nucleoli, eosinophilic to clear cytoplasm, occasional mitoses [Figure 3]. Focal area of necrosis was also seen. A histopathological diagnosis of Placental Site Trophoblastic Tumor was given and the patient was referred to higher centre for immunohistochemistry. She came back after 3 months with immunohistochemistry report - tumor cells positive for HPL, negative for p63 staining and low proliferative index of 6% for Ki 67. Blood investigstions done at that time showed minimally elevated βHCG value of 50 mIU/ml (initial βHCG value done on day 10 postpartum was 33 mIU/ml) and USG did not show any peritoneal metastasis. Chest x-ray was normal.

Though the PSTT was confined to the uterus, without metastatic foci, but in view of minimally elevated βHCG levels, patient was given prophylactic methotrexate and advised regular follow up.

Figure 1. Gross specimen of gravid uterus with an ill-defined grey white mass in myometrium

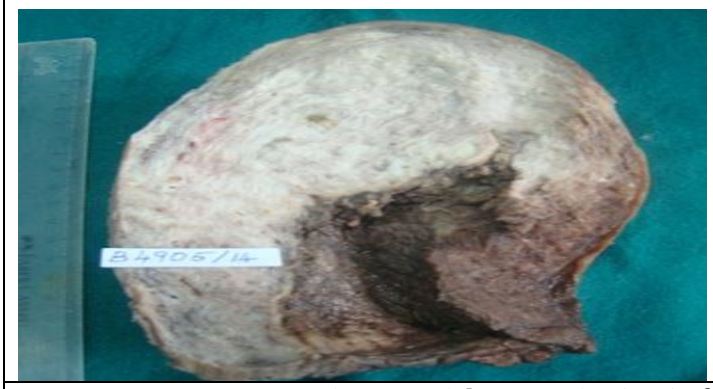


Figure 2. Microscopic examination (Low power, 10X) show nodules of intermediate trophoblastic cells infiltrating the myometrium

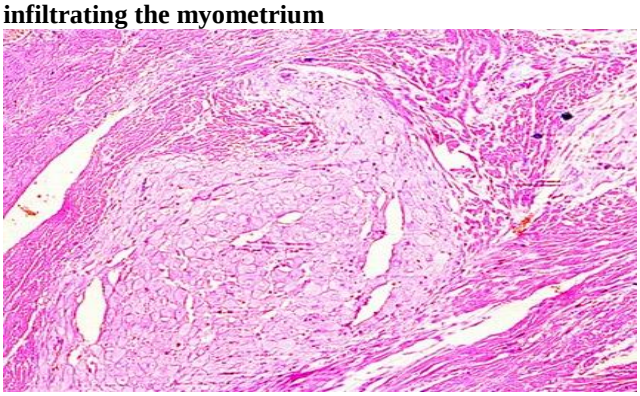
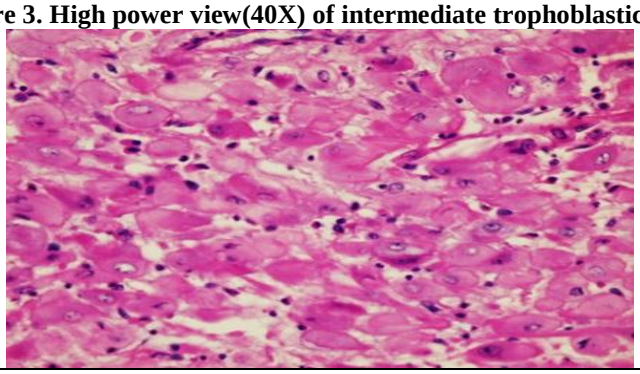


Figure 3. High power view(40X) of intermediate trophoblastic cells



DISCUSSION

Gestational Trophoblastic Diseases (GTD) are a spectrum of diseases that arises from placental trophoblast which consists of syncytiotrophoblast, cytotrophoblast and intermediate trophoblast. The syncytiotrophoblastic cells are polynuclear cells that form the external layer, mainly produce human chorionic gonadtropin (HCG) and invade the endometrial stroma. The cytotrophoblasts form the basal layer, whose cells can differentiate to syncytiotrophoblastic or intermediate trophoblastic cells. The intermediate trophoblastic cells leave the placenta to restructure the spiral arterioles in order to decrease the blood flow resistance toward the placenta [5]. PSTT arises from these intermediate trophoblastic cells at implantation site [7]. PSTT follows a normal pregnancy, a miscarriage, an abortion or a molar pregnancy [1,4],and during menopause [4] with varied interval between antecedent pregnancy and presentation [1,2,4,5]. The patient can present with irregular vaginal bleeding, amenorrhoea, nephrotic syndrome and rarely polycythemia or virilization [2] or symptoms due to metastasis [4,5,8].

In our case the patient presented 11 months after abortion, but as she conceived within 2 months, the mass which was noted on initial scan became inconspicuous with the growth of fetus and presented with severe postpartum hemorrhage after delivery. Grossly PSTT is seen as a poorly circumscribed mass diffusely infiltrating the myometrium [1,4] and histologically composed of intermediate trophoblastic cells without chorionic villi infiltrating muscle fibres and along vessel walls [1,4,5,9]. Mitosis is variable and is an important prognostic marker.

The differential diagnoses to be considered are exaggerated placental site reaction, epithelioid

trophoblastic tumor and choriocarcinoma [1, 5]. PSTT is associated with less vascular invasion, necrosis, and hemorrhage than choriocarcinoma, and it has a propensity for lymphatic metastasis [5,10].

The immunohistochemical analysis show strong positive staining for HPL; weak and focal positive staining for HCG [1,5,9]. The Ki 67 index, marker for mitotic activity is useful for differentiating PSTT(10-20%) from exaggerated placental site reaction(<1%) and choriocarcinoma(>50%) [1]. The pregnancy-associated major basic protein (pMBP), a marker of the intermediate trophoblast, is useful in differentiating PSTT from other forms of epithelioid trophoblastic tumors [5].

The diagnosis of PSTT is usually made after hysterectomy on histopathological findings [6]. Preoperative diagnosis may be aided by imaging and serum tumor markers [2]. Unlike choriocarcinoma, the level of serum βHCG in PSTT correlates neither with tumor burden, nor with the malignant behaviour. Thus, βHCG appears to have no predictive value and disease may still progress even if levels are not raised. The explanation of this moderated elevation of βHCG is that these cells produce primarily more HPL and less βHCG [3]. In our patient the diagnosis was made on hysterectomy specimen with characteristic histological features. The immunohistochemistry was positive for HPL with 6% Ki-67 index and negative for p63.

Metastasis may be seen in 10-15% of cases mainly to lung, lymphnode, brain, liver, kidney, vagina, stomach and spleen [6,9]. Adnexal metastases are rare (3%) [3].

PSTT is staged according to the FIGO staging of GTD, but the behavior of PSTT has not been well predicted by the FIGO 2000 scoring system and needs to be considered separately [3].

Poor prognostic factors are an interval of more than 2 years from known antecedent pregnancy, mitotic count >5/10 HPF, extensive necrosis and extension outside the uterus [1,5].

The preferred treatment for PSTT is essentially surgical and is based on hysterectomy with lymphatic sampling [5]. Chemotherapy used in the treatment of other GTT, is usually ineffective in PSTTs [3] as it is a slow growing tumor [5]. The role of adjuvant chemotherapy and the optimal treatment of metastatic disease remain uncertain [3]. In view of minimally elevated βHCG levels post hysterectomy, patient was given methotrexate and the patient is on regular follow up with βHCG levels.

GTN are now some of the most curable of all solid tumors, with cure rates -90% even in the presence of widespread metastatic disease [10].

CONCLUSION

PSTT is a rare and unique tumor, potentially curable with unpredictable behavior. Hence long term follow up is necessary.

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