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Case Report

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EXAGGERATED PLACENTAL SITE REACTION: A RARE CAUSE OF MASSIVE POST PARTUM HAEMORRHAGE

Sidhu S.*, Ashima, Isaacs R. and Dhar T.[1]

Department of Pathology, Obstetrics and Gynecology, Christian Medical College and Hospital, Ludhiana.

***Corresponding Author: Dr. Sidhu S.**

Department of Pathology, Obstetrics and Gynecology, Christian Medical College and Hospital, Ludhiana.

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ABSTRACT

Exaggerated placental site reaction (EPSR) is a non-neoplastic condition of intermediate trophoblasts clinically presenting as post-gestational abnormal uterine bleeding (AUB), increase in beta- human chorionic gonadotropin (beta-hCG) or mass lesion in the uterus. In majority of cases a retrospective diagnosis is made after histopathological examination which is also essential to distinguish it from gestational trophoblastic diseases. Here we report a case of term pregnancy presenting with post partum hemorrhage (PPH) due to EPSR and a brief review of literature for this rare finding.

KEYWORDS: Exaggerated placental site reaction, intermediate trophoblast, post partum hemorrhage.

INTRODUCTION

Exaggerated placental site reaction is a non-neoplastic lesion characterized by extensive infiltration of extravillous implantation site intermediate trophoblasts (ISITs) into the endometrium, underlying myometrium and blood vessel walls of the normal placental implantation area.[1,2] Diagnosis of EPSR should be considered in women with AUB related to gestational event. Its incidence is 1.6% in spontaneous and elective first trimester abortions.[3] EPSR is also seen in molar pregnancy[4], cervical pregnancy[5] and intrauterine pregnancy.[1,6,7,8] Though presenting as heavy irregular uterine bleeding, it is an uncommon cause of massive postpartum hemorrhage associated with atonic uterus.[7] Here we present a case of EPSR as a cause of massive PPH diagnosed on histopathological examination.

CASE REPORT

A 30 years old $P_7G_5A_2$ lady was referred to our hospital in shock due to profuse per vaginal bleeding following normal vaginal delivery. Clinical examination revealed retained membranes along with atonic uterus. Membranes were removed but patient continued to bleed immensely in spite of adequate medical treatment. Hysterectomy was done as a life saving procedure to manage the post partum hemorrhage and specimen sent for histopathological examination. Gross examination of hysterectomy specimen sent showed an enlarged, gravid uterus measuring 15.5x10x7cm. There was no residual placenta or placental membranes on the placental site and no cervical tear or laceration were detected "Fig.1". The endometrial cavity was lined by adherent polypoidal hemorrhagic material. Microscopic examination showed infiltration of intermediate trophoblasts into the endometrium, superficial myometrium and vessel walls. The intermediate trophoblastic cells were mononucleated and multinucleated along with interspersed hyaline material. Mitosis was insignificant. Endometrial glands were surrounded by trophoblasts however their architecture was maintained. No villi were identified "Fig. 2". A diagnosis of EPSR uterus was made.

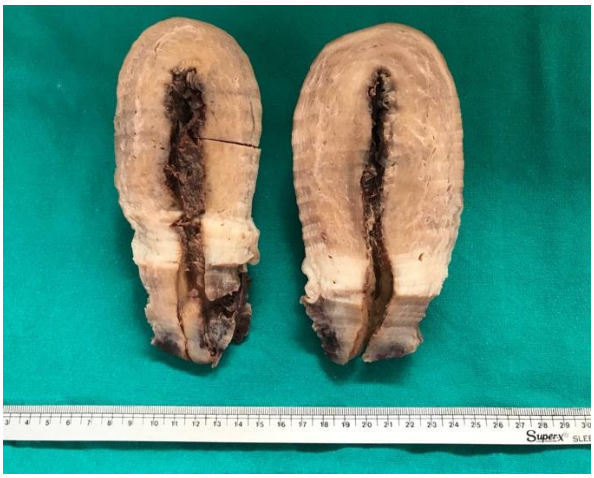

Fig. 1: Gross specimen of an enlarged, gravid uterus measuring 15.5x10x7cm. Dilated endometrial cavity filled with clotted blood. Thickened lining with adherent polypoidal hemorrhagic material.

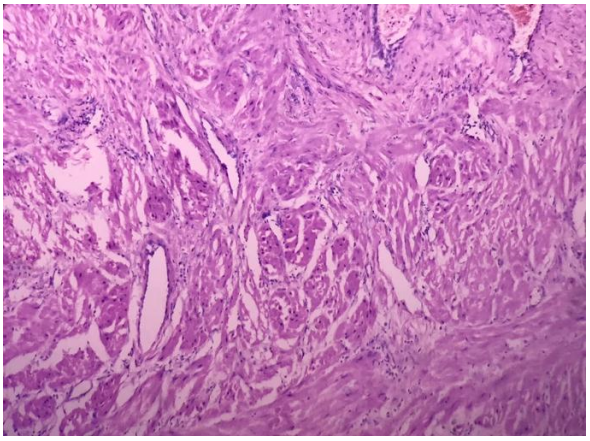

Fig. 2: Infiltration by mononucleated intermediate trophoblasts into the superficial myometrium with insignificant mitosis. Endometrial glands surrounded by trophoblasts but their architecture is maintained. (H&E 100X).

DISCUSSION

EPSR is characterized by infiltration of intermediate trophoblasts into the superficial myometrium. It represents one extreme of a normal physiological process rather than a true lesion.[2] EPSR was earlier termed as syncytial endometritis or benign chorionic invasion– both being misnomers as its not an inflammatory condition nor is it limited to the endometrium. In addition, there is no dominant syncytial arrangement of cells.[2]

Implantation site intermediate trophoblasts are important for the maintenance of pregnancy. They infiltrate the decidua, myometrium and spiral arteries during early gestation. This normal physiological process helps in establishing the materno-fetal circulation.[9] During the first trimester, infiltration into the upper half of myometrium takes place. As the pregnancy progresses infiltration by the intermediate trophoblasts regresses. The postpartum role of ISITs is not known.[7,10] In a term pregnancy they are confined to the superficial layers only. When exaggerated infiltration occurs or it fails to involute, the condition is known as EPSR.[2,10] The parameters for quantification of trophoblastic infiltration at different times of normal gestation are not well defined, hence the cut off to call it as EPSR is arbitrary.[11]

Four distinct trophoblastic lesions are known to arise from the intermediate trophoblasts. They include: Exaggerated placental site reaction(EPSR), Placental site nodule (PSN), Placental site trophoblastic tumor (PSTT) and Epithelioid trophoblastic tumor (ETT). EPSR and PSN are non neoplastic proliferations. PSTT and ETT on the other end have metastatic potential and can cause local invasion. PSN and ETT are lesions of intermediate trophoblasts of chorionic laeve while EPSR and PSTT arise from intermediate trophoblasts at the implantation site.[2] Correct diagnosis is a necessity as each of these lesions have different treatment approach.

The most important differential diagnosis of an exaggerated implantation site is placental site trophoblastic tumor.[2,11] PSTT is a neoplastic transformation of intermediate trophoblastic cells.[2,11] It presents with amenorrhea or gestation related abnormal bleeding with enlarged uterus. Microscopy shows proliferation of intermediate trophoblasts which are arranged in nests and interdigitating pattern invading deep into the myometrium. Invasion of blood vessels is seen along with deposition of fibrinoid material. Mitosis is present. No cytotrophoblast or villi seen.[2] EPSR is to be considered when the lesion has no increase in mitosis and interspersed with the trophoblasts is hyaline material. Villi can also present in EPSR.[2,11] The trophoblast of EPSR are cytologically similar to the intermediate trophoblasts of the normal implantation site. They have hyperchromatic nuclei with irregular nuclear margins and abundant amount of eosinophilic cytoplasm. Multinucleated forms are also seen.[2] Our case also showed intermediate trophoblasts invading the myometrium with interspersed hyaline material and absent mitosis. Villi were not seen.

EPSR can present as spontaneous or induced first trimester abortions[3], in association with molar pregnancy[4] and cervical pregnancy.[5] It is occasionally seen on HPE of curettage specimens of induced abortion.[3] Clinical presentation includes excessive uterine bleeding, elevated beta-hCG or uterine mass. Kase et al. highlighted EPSR in cervical pregnancy presenting with uterine bleed and intrauterine mass.[5] In 1999 Menczer et al. while reporting a case of EPSR in association with molar pregnancy found the patient to have increased beta-hCG levels post curettage along with intrauterine nodule.[4] Kadian et al. concluded that follow up of beta-hCG should be done in all women diagnosed with EPSR as PSTT is its most important differential.[8]

Liu et al. 2013 reported a case of EPSR in term pregnancy presenting as postpartum hemorrhage at the time of caesarian.[12] Similar to our present case, Takebayashi et al. 2014 reported a case of severe postpartum hemorrhage associated with atonic uterus following post vaginal delivery. Hysterectomy was carried out and a retrospective diagnosis of EPSR was made after histopathological examination of the hysterectomy specimen.[7] Both these cases share the same clinical presentation as our case, namely, EPSR manifesting as atonic post partum hemorrhage with no mass lesion in the uterus. The patient being reported was grand multipara and hence, atonic PPH was clinically considered to be the cause of PPH.

EPSR can also present as delayed irregular uterine bleeding as long as 7 months[11] to 15 years post normal delivery.[6]

Takebayashi et al. 2014 postulated that uterine atonia following delivery resulted due to vasodilation in the decidua and myometrium by excessive proliferation of

ISITs or due to unknown substances secreted by them preventing myometrial cell contraction.[7] EPSR should be considered in cases of postpartum hemorrhage caused by uterine inertia not responding to medical management. This should be confirmed by histopathological examination and followed up with serum beta-hCG levels.

CONCLUSION

The relation of EPSR with atonic uterus and its exact pathogenesis needs to be investigated. EPSR though one of the rare causes of PPH should be considered and confirmed by careful histopathological examination of the hysterectomy specimen.

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