CASE REPORT

Case Report: Placental site trophoblastic tumor revealed by a clinical pelvic abscess [version 1; peer review: 1 approved with reservations, 1 not approved]

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Abstract

We report an uncommon clinical presentation of a placental site trophoblastic tumor. The patient presented initially with abdominal pain with, fever, bleeding and pelvic mass on ultrasonography leading to the wrong diagnosis of a pelvic abscess. Dilation and curettage were performed and pathological examination confirmed the diagnosis. of placental site trophoblastic tumor. Therefore, she underwent abdominal hysterectomy. Four years after surgery, the patient is still disease free. Gestational trophoblastic diseases should be considered in every patient presenting abnormal uterine bleeding after delivery or pregnancy loss despite the associated symptoms being very unusual.

Keywords

Gestational Trophoblastic Disease, Placenta Diseases, Trophoblastic Tumor, Placental Site, pathology.

This article is included in the **Oncology** gateway.

Open Peer Review

Approval Status ? X

	1	2
version 1		
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1. Antonio Braga , Maternity School of Rio de Janeiro Federal University, Fluminense Federal University, Rio de Janeiro, Brazil		
2. Maria-Gabriela Aniței , University of Medicine and Pharmacy, Iasi, Romania		
Any reports and responses or comments on the article can be found at the end of the article.		

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Author roles: **Nesrine S:** Conceptualization, Writing – Original Draft Preparation; **Bettaieb H:** Data Curation, Methodology, Resources; **Mbarki W:** Resources, Software; **Ehsen BB:** Investigation, Methodology; **Imen H:** Investigation, Methodology; **Mehdi BN:** Writing – Review & Editing; **Hedhili O:** Validation; **Najeh H:** Formal Analysis, Project Administration, Supervision; **Mbarki C:** Supervision, Validation

Competing interests: No competing interests were disclosed.

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Introduction

Placental site trophoblastic tumor (PSTT) is a rare tumor, representing 1% of gestational trophoblastic diseases.^{1,2} It mainly affects women of childbearing age after delivery or pregnancy loss.¹⁻³ In this paper, we report a case of placental site trophoblastic tumor diagnosed by an array of pelvic abscess associated with bleeding occurring two months after delivery. The evolution, after surgical treatment by hysterectomy, was favorable.

Case report

We report the case of an unemployed 32-year-old Arabic woman, gravida 1 and para 1, without medical history, gave birth naturally and without complications to a full-term healthy newborn. Two months after her delivery, she consulted our department with persisting metrorrhagia associated with fever of 39°C. The patient's examination revealed that the bleeding has been noted since the delivery day and it had increased two days earlier.

A physical examination revealed a fever of 39.5°C and abdominal tenderness. The blood pressure was 110/70 mm Hg and the heart rate was 82 beats/mn. Furthermore, there was no specific digestive or urinary symptom. Per speculum examination, the cervix was enlarged with purulent vaginal discharge. On digital pelvic examination, the uterus was increased in size accessed at 12 week gestation and its mobilization was painful. Blood tests showed an inflammatory syndrome with a C-reactive protein at 200 mg/l [≤ 6 mg/l] and white blood cells at 17000/mm³ [4000-10000/mm³]. The beta-human chorionic gonadotropin (β -HCG) rate was positive at 674 U/l [< 5 U/l]. The transvaginal sonography showed a large sized uterus containing an echogenic heterogeneous endometrial mass, vascularized on Doppler examination.

Figure 1. Sonographic appearance of the tumor of the placental site tumor. (A) Echogenic tumor occupying the uterine cavity (arrow) with infiltration of the myometrium; (B) appearance vascularized tumor at color Doppler.

This process seemed to invade the myometrium in part. In addition, ultrasound showed an average effusion in the Douglas' pouch (Figure 1) with a probable left adnexal abscess.

Laparoscopic adnexal drainage and endometrial curettage were performed. The infectious episode resolved promptly with antibiotics; gentamicin 160 mg per day for 5 days metronidazole 500 mg three times a day for 15 days and cefotaxim 1g three times a day for 15 days. Pathological examination of the endometrial curettage specimen reported a proliferation of large tumor cell with eosinophilic cytoplasm and nuclear atypia, infiltrating the myometrium. These cells were positive for pro inflammatory cytokine (IL6) in the immunohistochemistry studies and the final diagnosis was a placental site trophoblastic tumor.

A laparotomic hysterectomy with preservation of ovaries was performed. On gross examination there was a mass of 6 cm localized to the myometrium of bluish appearance with areas of hemorrhage and necrosis. The tumors had ill-defined borders and invaded into the myometrium (Figure 2). Pathological examination was similar to that described in the curettage specimen, showing a proliferation of monomorphic intermediate trophoblasts (Figure 3A). The tumor cells demonstrated nuclear atypia, eosinophilic cytoplasm (Figure 3D) and myometrial invasion (Figure 3B,C). Extensive necrosis, hemorrhage and vascular invasion are observed (Figure 3E). The final pathology report confirmed the diagnosis of PSTT infiltrating the myometrium. The postoperative course was favorable. The patient had a regular follow-up every month for the first year combining a clinical examination and a serum level of β -HCG. She had a clinical check-up and pelvic ultrasound every year for two years, no sign of recurrence was noted. Four years after surgery, the patient is still disease free.

Discussion

In 1895, Marchand first described PSTT and named it atypical chorioepithelioma.⁴ In 1981, Scully and Young described the morphological details and recognized it as a neoplastic process, and named it PSTT.⁵ PSTTs are a rare subtype of gestational trophoblastic disease, accounting for less than 1% of all cases.^{1,2,6} It mainly affects women of childbearing age and it is seen in patients between 19-62 years, with an average age of 30 years.^{1-3,6,7} PSTT can occur after a normal pregnancy, abortion, term delivery, ectopic pregnancy or molar pregnancy.^{1-3,6-8} Here we reported a case of PSTT occurring after a normal pregnancy. This pathology may occur months to years after pregnancy with the average interval between the antecedent pregnancy and diagnosis being 16 to 18 months.^{1-3,6,9} Diagnosis of PSTT can be difficult due to the nonspecific clinical signs. Abnormal uterine bleeding is the most common presenting feature.^{1-3,6-9} Metrorrhagia can also be associated with amenorrhea.^{1,8} The peculiarity of our case lies in the fact that the tumor was manifested by an array of pelvic abscess associated with bleeding; such clinical presentation was rare in literature.⁸ On digital pelvic examination, the uterus can be enlarged or normal sized.^{2,8} A wide range of other rare symptoms have also been reported, including galactorrhea, virilization, nephrotic syndrome, and polycythemia.^{2,8} PSTT can also be revealed by metastasis.^{2,7} The serum levels of β -hCG are usually in the range of 1000-2000 mIU/mL.^{1-3,6,9} The origin of PSTT is the intermediate trophoblast cells which secrete little beta HCG and a lot of placental lactogenic hormone.^{6,9}

Figure 2. Macroscopic appearance of the placental site tumor. (A) Peri operative view showing increased uterine size, yellowish area seat (arrow); (B) appearance of the tumor to cut, yellowish tumor occupying the uterine cavity and infiltrating the myometrium.

Figure 3. Histological appearance of the placental site tumor. (A) Monomorphic cell proliferation of intermediate trophoblast; (B, C) tumor cells infiltrate the myometrium form of clusters, dissociating smooth muscle bundles (B: low magnification, C: magnification); (D) proliferation of large cells with eosinophilic cytoplasm and lobulated nucleus; (E) home of tumor cells with the presence of vascular emboli.

On gross examination, the tumors are located primarily in the endomyometrium, presenting as polypoid or nodular masses, with a variable diameter up to 10 cm.^{1,6} The sectioned surfaces of the tumors are solid, often fleshy, and usually yellow with necrosis, and hemorrhage.^{1,2,6} The tumor may extend into the cervix or infiltrate the serous, the adnexa or the round ligaments.² In our case, the tumor invaded the myometrium without reaching the serous. The diagnosis of PSTT is based on pathological examination.

On microscopic examination, we typically find a proliferation of intermediate trophoblastic cells without chorionic villi, which infiltrate muscle fibers.² Vascular invasion, necrosis and hemorrhage are also often observed.²

The tumor is composed of a relatively monotonous population of polygonal cells with nuclear atypia and moderately abundant cytoplasm that can be amphophilic, eosinophilic or clear.^{1,2} The distinctive pattern of vascular invasion and deposition of fibrinoid material are the key diagnostic features.¹⁻³ Usually, PSTTs are benign tumors. However, they have an unpredictable malignant potential and between 10 and 20% of patients have metastatic disease at the time of presentation.^{1,2,7,10} Metastasis site include peritoneum, vagina, lung, liver and brain.

Hysterectomy without bilateral oophorectomy is the gold standard in PSTT management.^{2,10} In fact, this entity is particularly less responding to chemotherapy than hydatiforme mole, invasive mole and choriocarcinoma.^{2,10} When there is extension beyond the uterus, surgical treatment should be associated to chemotherapy.^{2,10}

Actually, the FIGO anatomical staging system is the strongest predictor of patient outcome presenting with PSTT.^{2,7,10,11} After surgical treatment, patients should be followed up during years to optimize the chance of detecting a relapse or metastasis.^{7,10,11}

Strengths and limitations

This case report is interesting since it's the first reporting PSTT simulating a pelvic abscess. However, PTTT are rare which prevents researchers from realizing large scale studies in order to develop valid management protocols.

Conclusion

Although PSTTs are extremely rare worldwide, the diagnosis should be considered during the post-partum and post-abortion periods if there is persistent metrorrhagia. After surgical treatment, the follow up is needed to be accurate, to help recognize a relapse or metastasis.

Data availability

All data underlying the results are available as part of the article and no additional source data are required.

Consent

Written informed consent for publication of clinical details and clinical images was obtained from the patient.

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Version 1

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Maria-Gabriela Aniței

University of Medicine and Pharmacy, Iasi, Romania

I was pleased to review the article **Case Report: Placental site trophoblastic tumor revealed by a clinical pelvic abscess**.

The methodology used by the authors is appropriate for the purpose of the study and conclusions are narrowly linked to available evidence. The title expresses clearly the content of the manuscript and highlights the importance of the study. As the authors note in **Strengths and limitations**, this case report is interesting since it is the first reporting of PSTT simulating a pelvic abscess. There was rapid evolution of tumor development, only 2 months after natural birth, apparently without a risk factor from her medical history. There are interesting images intraoperative and microscopy

The article respects the presentation algorithm that is required for a case report type article, but there is a number of medical details are missing, such as no detailed investigations (CT-scan, for example) to exclude metastases, both at the time of diagnosis and follow-up.

In the same direction, to improve the methodology of this study, the introduction section must be improved. It is too short, and I suggest that some information regarding diagnosis and prognosis of PSTT should be included. Likewise, Immunohistochemistry data and the FIGO score are missing from the anatomopathological presentation, this information would increase the value of the article.

Likewise, to be more convincing, the authors could image the patient at a distance through an CT-scan exam to confirm, through this high-performance investigation, the absence of local or distant disease. These tests would help to exclude metastases, both at the time of diagnosis and follow-up, especially as the anatomical-pathological examination shows a vascular invasion.

The conclusion of the article could be strengthened. **Although PSTTs are extremely rare worldwide, the diagnosis should be considered during the post-partum and postabortion periods if there is persistent metrorrhagia** - this means that, in front of a patient with metrorrhagia and febrile syndrome that appeared early after delivery, this differential diagnosis

(PSTT) must also be taken into account.

Other points:

1. It doesn't seem representative to me that **pathology** should be a Keywords
2. immunohistochemistry data and the FIGO score are missing from the anatomopathological presentation
3. Usually, 'PSTTs are benign tumors' – I am not agree with this affirmation
4. In figure 3- the microscopy details must be specified - is it an image of HE? , how many times is it amplified (which objective was used?) . As the area of interest is identified in image D, I recommend that the same thing be done for the other images (Figure 3- A, B, C, E)

Is the background of the case's history and progression described in sufficient detail?

Partly

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?

Partly

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?

Yes

Is the case presented with sufficient detail to be useful for other practitioners?

Partly

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: oncological gynecology

I confirm that I have read this submission and believe that I have an appropriate level of expertise to state that I do not consider it to be of an acceptable scientific standard, for reasons outlined above.

Reviewer Report 07 March 2022

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Antonio Braga

Rio de Janeiro Trophoblastic Disease Center, Antonio Pedro University Hospital, Maternity School of Rio de Janeiro Federal University, Fluminense Federal University, Rio de Janeiro, Brazil

Dear Authors and Editors,

I would like to thank you for the opportunity to review the paper entitled: "Case Report: Placental site trophoblastic tumor revealed by a clinical pelvic abscess", submitted to the F1000. Similarly, I would like to congratulate the authors for their interest in the topic.

In this case report, the authors presented a description of a PSTT presenting as abdominal abscess, reviewing the literature of this subject.

I would like to highlight some aspects that deserve attention.

1. The authors must describe in the case report the screening for metastatic disease, to justify the decision for TAH.
2. PSTT is always cancer, not a benign tumor as described.
3. Nowadays we have, besides FIGO anatomical staging system as prognostic factor for PSTT surviving, the interval of last pregnancy and the initiating of treatment of 48 months.¹
4. In the section 'Strengths and Limitations' there is a PTTT that should be corrected.

References

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Is the background of the case's history and progression described in sufficient detail?

Partly

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?

Partly

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?

Yes

Is the case presented with sufficient detail to be useful for other practitioners?

Yes

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Gestational trophoblastic disease

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

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