The role of the ultrasound examination in atypical placental site trophoblastic tumour differential diagnosis and management. A case report

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Abstract
Placental site trophoblastic tumour (PSTT) is a very rare and unique form of gestational trophoblastic tumour, representing about 1-2\% of all gestational trophoblastic tumours. Usually, the pattern is a slow growing nodule implicating the endometrium and myometrium, accompanied by abnormal uterine bleeding. Three ultrasound types of PSTT are described, but there is no specific characteristic for diagnosis. We present the case of a patient with an atypical placental site trophoblastic tumour diagnosed two months after a caesarean scar pregnancy. In the presented case there are several particularities, such as the rapid growth and progression of the tumour, the limitation to the myometrium and the difficulty of the differential diagnosis and approach.

Keywords: trophoblastic tumour; ultrasound; differential diagnosis

Introduction
The placental site trophoblastic tumour (PSTT) is a very rare and unique form of gestational trophoblastic disease, representing about 1-2\% of all gestational trophoblastic tumours [1]. Several characteristics of this type of tumour are its unpredictability, the absence of the specific aspect of the syncytiotrophoblast and cytotrophoblast and also its malignant potential, implicitly its resistance to chemotherapy and subsequently the poor outcome [1]. An important prognostic factor is the depth of myometrium invasion and the lesion volume greater than 1 cm\textsuperscript{3} [2]. Usually, the pattern is a slow growing nodule limited to the endometrium and myometrium, accompanied by abnormal uterine bleeding, in a woman of reproductive age with history of vaginal or caesarean delivery, induced or spontaneous abortion, hydatidiform mole or another form of gestational trophoblastic disease (GTD) occurring several months or years after any of these events [3]. Also, typically, the serum beta- human gonadotropin (hCG) level is very low, a false negative pregnancy test is possible due to the secretion of human placental lactogen (hPL) and the diagnosis can be confirmed only by histopathological exam [4].

Three types of ultrasound (US) appearance of PTTS have been described [5]: Type I, characterized by the presence of an intrauterine heterogeneous solid mass with moderate vascularization; Type II, the intra-myometrial heterogeneous mass presents minimal to high vascularization; and Type III in which the lesion appears intra-myometrial, cystic, of lacunar-type, with high degree vascularization. The differential diagnosis with other types of GTD is difficult, considering that the hemodynamic and volume parameters do not differ significantly,
so, computer tomography (CT) and magnetic resonance imaging (MRI) are widely used especially for determining the extent of the lesion [6].

We present the case of a patient with an atypical placental site trophoblastic tumour diagnosed only two months after a caesarean scar pregnancy.

Case report

A 32-year-old woman, with caesarean delivery 10 years before presentation, referred for a second opinion in the context of a uterine nodular lesion, diagnosed two months after the surgical management of a caesarean scar pregnancy and metrorrhagia. No histopathological exam of the extracted material after curettage was performed.

Complete blood analysis showed mild anaemia (haemoglobin 9.0 g/dl) and negative serum beta-hCG. The US examination revealed a well delimited heterogenic anterior isthmic mass, at the level of the caesarean scar, of approximatively 64/50 mm, hypervascular on colour Doppler examination (fig 1a), limited to the myometrium and atypical placental-like and lacunar aspect (fig 1b). The extent to the endometrium was excluded trough a US guided curettage (fig 1c) as the histopathological examination of the extracted tissue did not detect any abnormal cell.

Pelvic MRI examination described in the anterior isthmic region, at the level of the caesarean scar, a heterogeneous, well individualized, with a shady outline mass (62/65/43 mm) with low T1 signal and T2 non-homogeneous and turgescent, peripheral vascularization. The mass deformed significantly the uterine cavity and the endocervix, with no signs of obstruction and no signs of bladder or parametrial invasion (fig 1d). The described aspect could not exclude the diagnosis of choriocarcinoma.

Macroscopically, the anterior uterine lesion appeared of increased consistency, abnormally adherent to the bladder serosa (fig 2) and with a brownish, friable, placental-like content. The subsequent histopathological examination described the tumour as necrotic placental villi, hyalinized at the level of the caesarean scar, with the suggestive aspect of intermediate trophoblastic cells. The diagnosis of PSTT was clarified.

Discussions

There are no specific risk factors or known aetiology for PSTT. The differential diagnosis includes choriocarcinoma, invasive mole, organized retained placental fragments, leiomyosarcoma or placental site nodule [7].

Although a clear diagnosis of PSTT cannot be clarified through US, this investigation is an essential tool. PSTT cases are particularly difficult to diagnose, considering the absence of specific biologic markers, as the neoplastic intermediate trophoblast cells produce a very low quantity of β-hCG. The US aspect varies consider-ably, but, as a particularity, high vascularization is always present. Specialized Doppler examination can predict the malignant nature, the stage and guide the further manage-
ment [8]. Most reports on PSTT describe the presence of cystic spaces along with prominent vascularization of the poorly defined lesion. There are only a few cases of PSTT [8,9] described in literature as well as individualized lesions, confined completely to the myometrium with atypical vascularization, as was in this case.

The rapid growth, in two months, and the changed aspect of the tumour after the caesarean scar pregnancy was a particularity of this case and made the diagnosis more challenging. In literature, a median interval from antecedent pregnancy of 12.5 months for PSTT cases was reported [8]. Next, the limitation to the myometrium, classified as type II, with very well-defined margins on transvaginal US is an atypical and rare presentation. Also, only in 3% of cases, PSTT occurs after ectopic pregnancy. Most commonly, in over 60%, this pathology follows a normal term pregnancy [9]. The surgical treatment is defining for the prognosis of PSTT cases, as the response to chemotherapy is relative and variable [10]. In the presented case, the complete myometrium invasion was regarded as a negative prognostic factor, but abdominal and pelvic lymphadenectomy was not performed due to the fact that there have been no studies that showed a positive influence on the survival rate related to this procedure [11].

**Conclusion**

There are no specific characteristics for PSTT diagnosis, but transvaginal US in combination with history and clinical exam, clearly contributes to the decision making regarding the appropriate approach. This case is presented in order to highlight the importance of a correct evaluation with a detailed US examination, management and follow-up of each clinical case as PSTT cases do not follow a time related, clinical or imagistic pattern.

**References**