Scholars Journal of Medical Case Reports **ISSN 2347-6559 (Online)**
Sch J Med Case Rep 2017; 5(8):502-506 **ISSN 2347-9507 (Print)**

(An International Publisher for Academic and Scientific Resources) DOI: 10.36347/sjmcr.2017.v05i08.015

Fortuitous Discovery of A Primary Ovarian Choriocarcinoma During a Caesarean Section: About a Case

Tapa E[1], Rahali M[2], Lebbar K[3], Fichtali K[4], Benhessou M[5], Bouhya S[6]

[1-3]Resident doctor, Maternity ward of children's hospital Abderrahim Harouchi, Ibn Rochd University Hospital of Casablanca (Morocco)

[4,5]Associate Professor, Maternity ward of children's hospital Abderrahim Harouchi, Ibn Rochd University Hospital of Casablanca (Morocco)

[6]Head of service, Maternity ward of children's hospital Abderrahim Harouchi, Ibn Rochd University Hospital of Casablanca (Morocco)

***Corresponding author**
Dr TAPA Elisa
Email: elisatapa@yahoo.fr

Abstract: Pure ovarian choriocarcinomas are extremely rare and aggressive tumors that may be gestational or non-gestational in origin. Due the rarity of tumor, there is a ack of information on the clinicopathologic features, diagnosis, and treatment. We report a case of pure metastatic ovarian choriocarcinoma, to liver and lung, probably of gestational origin, which was accidentally discovered during a caesarean section, treated by surgery and whose chemotherapy could not be conducted on time.

Keywords: choriocarcinoma – ovary –caesaran section

INTRODUCTION

Ovarian choriocarcinoma is a rare and very aggressive tumor [1]. It is a germ cell tumor affecting especially young women and which is characterized by the presence of cytotrophoblastic and syncitirophoblastic malignant cells. It may develop as a metastatic gestational choriocarcinoma of the uterus or fallopian tubes (most common) or non-gestational (primary). The incidence of ovarian gestational choriocarcinoma is estimated to be 1 in 369 million pregnancies [2], while the non-gestational type accounts for less than 0.6% of all ovarian neoplasms [3]. Non-gestational choriocarcinoma is rare in its pure form, it is frequently associated with other tumors such as dysgerminoma, embryonic carcinoma and mature or immature teratoma, given their identical embryological origin. The gestational origin may be suspected in the presence of a gravid yellow body, however the final diagnosis will be based on genetic analysis [4]. Due the rarity of tumor, there is a ack of information on the clinicopathologic features, diagnosis, and treatment.

OBSERVATION

This was a 40-year-old patient, 2nd gesture and 2nd pare, with a 10-year-old male child born, followed by a 10-year unexplained secondary infertility. The patient was admitted to our facility for the management of a threat of premature delivery on a spontaneous, unplanned pregnancy, presumed at eight months. Clinical examination found normotensive patient, negative urine strips, positive uterine contractions with a vaginal touch a dilated collar dilated to 1cm.The obstetric ultrasound performed showed a progressive monofetal pregnancy in cephalic presentation with a fetal biometry corresponding to 33 weeks of amenorrhea. The amniotic fluid was sufficient for gestational age and the placenta was fundial. The procedure to be followed was hospitalization, a negative infectious test, a calcium channel blocker tocolysis and a betamethasone corticosteroid treatment. The evolution was marked by premature rupture of the membranes showing a meconium LA associated with a fetal bradycardia at 80 beats per minute, hence the indication of caesarean section for suspected fetal distress. This caesarean allowed the extraction of a male liveborn infant, Apgar to 8/10 in the first minute and 10/10 in the 5th minute, from birth weight to 2400g. In the presence of unexplained active bleeding, exploration of the pelvic cavity revealed a tumor at the site of the left ovary of about 10 cm, bleeding on contact, with the presence of a tumor graft at the colon sigmoid. A left adnexectomy was performed with good haemostasis. The postoperative follow-up was marked by the appearance at H36 of an acute pain in the right hypochondrium associated with a sensitivity of this zone, a distended abdomen with tympanic membrane, a transit not yet taken up and a drain to 150 cc of liquid sero-hematic. The abdomino-pelvic ultrasound performed revealed a liver of normal size, seated at the level of the right liver of an echogenic mass, heterogeneous seat of hypoechoic zones, vascularized with the color Doppler measuring 85/67 mm may be of secondary origin. Then, in the periphery, the presence of a heterogeneous hyperechogenous formation,

following the contours of the liver, deforming it in some places, measuring 3.1 mm in thickness, may be related to a subcapsular hematoma of the liver.An abdominal scan was indicated, which, in the state of shock presented by the patient, could not be achieved. Subsequent monitoring noted the presence of red blood drain at 900 cc of blood in 1 hour and 30 minutes. A surgical recovery for a subcapsular hematoma of the broken liver was indicated. This second exploration revealed a peritoneal effusion of great abundance, an enormous subcapsular hematoma taking the whole dome of the right liver with a suspicious lesion of 4 cm bleeding. A hepatic and epiploic biopsy was performed, then a three-field packing was performed. The patient stayed in obstetric resuscitation service where multiple bilateral lung lesions of secondary aspect were revealed.

The anatomo-pathological study of the adnexectomy part showed a mass of 13/12/6 cm with a smooth surface, beige white, brownish, haemorrhagic and fleshy. On histological examination, it was an infiltrating tumor proliferation composed of trophoblastic cells associated with syncythiotrophobastic cells, without any other germline. No ovarian parenchyma or fallopian tube was found. The immunohistochemical study showed that the tumor cells expressed HCG and CK AE1 / AE3 and were negative for PLAP (Placental Alkaline Phosphatase), CD-30 and AFP (alpha foeto protein), concluding with ovarian choriocarcinoma pure. The different biopsies (epiploic and hepatic) were tumorous.

After a multidisciplinary staff meeting, it was decided to carry out chemotherapy first before the metastatic character of the choriocarcinoma, followed by a totalization surgery. Unfortunately the evolution was marked by the death of the patient.

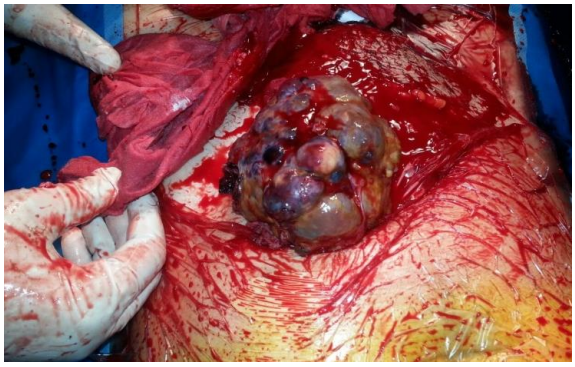

Fig-1: Ovarian choriocarcinoma of accidental discovery during a Caesarean section

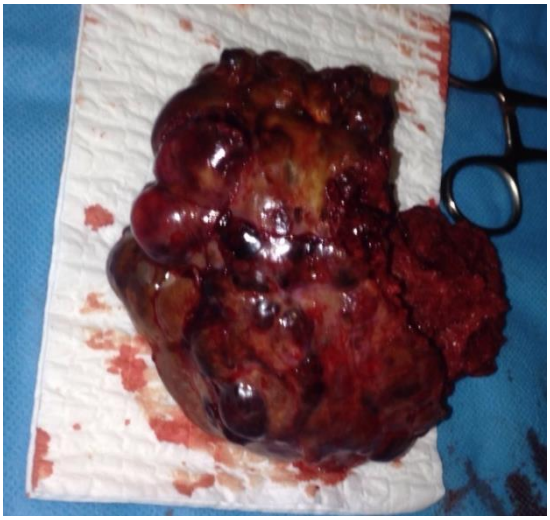

Fig-2: macroscopic appearance of choriocarcinoma

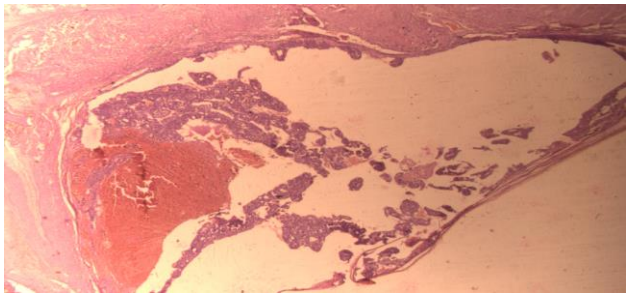

Fig-3: Tumor ovarian parenchyma

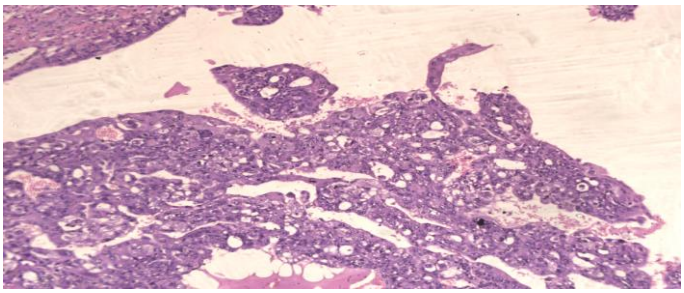

Fig-4: Tumor cytotoxic and syncitiotrophoblastic cells

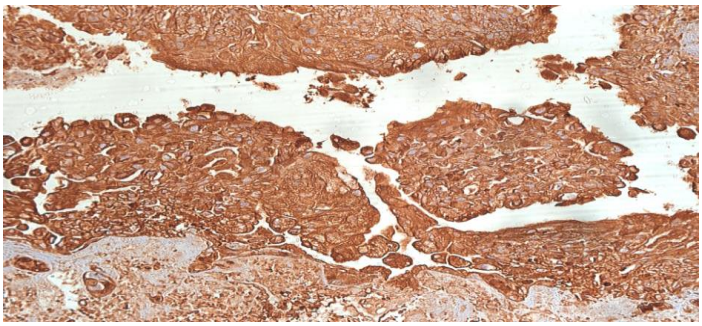

Fig-5: Positive AE1 / AE3 Cytokeratins

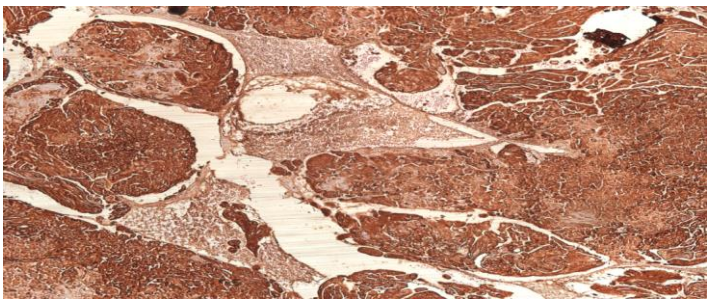

Fig-6: Positivity of beta hCG

DISCUSSION

Pure Ovarian choriocarcinomas are malignant germ cell tumors. They are defined by the presence of malignant cells, both cyto and syncitiotrophoblastic, closely entangled and without associated villus formation [3,5]. Their origin is either gestational or non-gestational. Gestational choriocarcinomas are the most frequent, whereas non-gestational choriocarcinomas are rare in their pure form [6]. They are often associated with other germinal tumors such as immature teratomas, dysgerminomas, embryonic carcinomas because their embryological origin is identical. Non-gestational choriocarcinomas may also be found outside the genital area (lung, abdomen) [7] However, diagnosis of primary Ovarian choriocarcinoma becomes extremely difficult during periods of genital activity and most of the clinical cases analyzed concern only very strong suspicions, because of the non-specific clinical symptomatology, which can mimic other more common pathologies. Thus, in the presence of an adnexal mass with increased plasma levels of βhCG and irregular vaginal bleeding, pure ovarian choriocarcinoma can easily be confused with an ectopic pregnancy [8, 9]. The distinction of this origin is important and often difficult because it is a question of different tumors, whose treatment and prognosis are different [10]. Thus, initially the diagnosis of non-gestational choriocarcinoma was retained, only if it

occurred in impuberated patients, incapable of conceiving or having no sexual relation [11]. The presence of a gravid yellow body adjacent to the tumor is suggestive of the gestational nature of the ovarian choriocarcinomas [12]. In our case, the fact of being associated with an evolutionary pregnancy supports us in this hypothesis. However, the search for paternal DNA in the tumor allows a clear distinction between gestational and non-gestational types. Indeed, tumors of gestational origin have a paternal genomic structure whereas non-gestational tumors have genomes of maternal origin without alleles of paternal origin [13]. In our case, the analysis of DNA polymorphism was not a routine technique in our laboratory, so we could not realize it. The treatment of germinal tumors of the ovary is medico-surgical and depends on the histological type. Proper surgical staging is essential during initial surgery. This should include peritoneal exploration with biopsies associated with omentectomy and pelvic and lumbo-aortic lymph node dissection to determine prognosis [14]. Some studies consider this staging a much more important determinant than gestational or non-gestational [15]. Because of its low incidence, Ovarian choriocarcinoma are treated with the same adjuvant chemotherapy regimens as those used for germinal tumors and gestational trophoblastic disease. Gestational forms should be differentiated from non-gestational forms in order to provide appropriate treatment. Indeed for the Gestational Choriocarcinoma, the chemotherapy is based on Methotrexate. On the other hand, for Non-gestational choriocarcinoma that respond less well to Methotrexate, a multidrug therapy according to the BEP protocol (bleomycin, etoposide, cisplatin) will be preferred because of the better tolerance and the effectiveness of etoposide. Chemotherapy should be undertaken as soon as possible (between 7-10 days) after diagnosis, as the growth and risk of tumor recurrence are very rapid [16]. Given the aggressive nature and the high risk of metastasis of pure Ovarian choriocarcinoma, close monitoring is essential by the regular dosage of hCG (human chorionic gonadotrophin) and its beta fraction, a highly sensitive specific marker [7], associated with Surveillance through both ultrasound and scanning imaging. The prognosis of ovarian choriocarcinoma is difficult to establish in the long term because of their rarity. Gestational Ovarian choriocarcinoma have a better prognosis than their non-gestational counterpart because of their sensitivity to Methotrexate [3,17].

CONCLUSION

Pure Ovarian choriocarcinoma is a rare disease, of which the gestational or non-gestational origin must be known, in order to propose appropriate management. Its treatment combines surgery with chemotherapy. Because of its aggressiveness, its diagnosis must be rapid in order to start the chemotherapy as quickly as possible. Its monitoring consists of a regular dosage of the beta fraction of the hCG hormone. Its long-term prognosis remains to be assessed.

BIBLIOGRAPHIE

1. Lv L, Yang K, Wu H, Lou J, Peng Z. Pure choriocarcinoma of the ovary: a case report. Journal of gynecologic oncology. 2011 Jun 30;22(2):135-9.
2. Axe SR, Klein VR, Woodruff JD. Choriocarcinoma of the ovary. Obstet Gynecol 1985;66:111-4.
3. Vance RP, Geisinger KR. Pure nongestational choriocarcinoma of the ovary: report of a case. Cancer 1985;56:2321-5.
4. Mood NI, Samadi N, Rahimi-Moghaddam P, Sarmadi S, Eftekhar Z, Yarandi F. Pure ovarian choriocarcinoma: report of two cases. Journal of research in medical sciences: the official journal of Isfahan University of Medical Sciences. 2009 Sep;14(5):327.
5. Norris JH, O'Connor MD. "Pathology of malignant germ cell tumors of the ovary". second edition. In: Coppleson M, editor. "Gynaecologic oncology". Hong Kong: Longman Group (FE) Ltd; 1992.
6. Aucouturier JS, Bader G, El Fata G, Guyot B, Louboutin A, Camus E. Choriocarcinome ovarien: à propos d'un cas. Gynécologie obstétrique & fertilité. 2003 Jun 30;31(6):539-42.
7. Sugano I, Nagao K, Kondo Y, Hatakeyama Y, Numata T, Kohno H. Primary nongestational choriocarcinoma of submental region. Acta Pathol Jpn 1984;34(4):895.
8. Balat O, Kutlar I, Ozkur A, Bakir K, Aksoy F, Ugur MG. Primary pure ovarian choriocarcinoma mimicking ectopic pregnancy: a report of fulminant progression. Tumori 2004; 90:136-8.
9. Chen YX, Xu J, Lv WG, Xie X. Primary ovarian choriocarcinoma mimicking ectopic pregnancy managed with laparoscopy: case report. Eur J Gynaecol Oncol 2008; 29:174-6.
10. Simsek T, Trak B, Tunç M, Karaveli S, Uner M, Sönmez C. Primary pure choriocarcinoma of the ovary in reproductive ages: a case report. Eur J Gynaecol Oncol 1998; 19: 284-6.
11. Jacobs AJ, Newland JR, Green RK. Pure choriocarcinoma of the ovary. Obstet Gynecol Surv 1982; 37:603–9.
12. Russell P, Farnsworth A. 2nd sub ed. Philadelphia: W.B. Saunders Company; 1997. Surgical pathology of the ovaries; pp. 785–7.
13. Koo HL, Choi J, Kim KR, Kim JH. Pure non-gestational choriocarcinoma of the ovary diagnosed by DNA polymorphism analysis. Pathol Int. 2006;56(1):613–6.
14. Gueye A, Narducci F, Baranzelli MC, Collinet P, Farine O, Fournier C, Vinatier D, Leblanc E. Les tumeurs malignes de l'ovaire des cellules germinales : un essai de 36 cas. Gynecol Obstet Fertil 2007 ; 35 : 406-19.
15. Axe SR, Klein VR, Woodruff JD. Choriocarcinoma of the ovary. Obstet Gynecol. 1985; 66(1):111–4.

16. William SD, Birch R, Einhorn LH, Irwin I, Greco FA, Loerher PJ. Treatment of disseminated germ cell tumors: chemotherapy with cisplatin, bleomycin, and either vinblastine or etoposide. N Engl J Med 1987; 316: 1435-40.
17. Kaneta Y, Yoshiyama R, Inagaki N, Toyoshima K, Ito K, Nishino R, Kitai H, Kato H, Asanoma K, Wake N. Gestational choriocarcinoma whose responsible pregnancy was a complete hydatidiform mole identified by PCR analysis with new sequence tagged site primers. Japanese journal of clinical oncology. 1999 Oct 1;29(10):504-8.