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Tubal Molar Pregnancy: A Case Report and Literature Review

F.Ouakka[1], A Elmoctar[1], M.K.Saoud[1], N.Mamouni[1], S.Errarhay[1], C.Bouchikhi[1] and A.Banani[1]

1 Deppartement of gynecology-obstetric 1, University hospital center of HASSAN II, FEZ, Morocco, faculty of medicine and pharmac

Abstract: *Gestational trophoblastic diseases (GTDs) constitue a spectrum of tumors characterizied by abnormal proliferation of pregnancy associated trophoblastic tissue with progressive malignant potential. GTD is classified into premalignant disease, termed complete and partial hydatiform mole (CM, PM), and malignant disorders (invasive mole, placental site trophoblastic tumor and choriocarcinoma)[1]. In most instances, moles develop within the uterine cavity, but may occur at any site. Ectopic molar pregnancy is a rare event, the first report on tubal mole was published in 1872 by Otto[2]. The incidence of ectopic hydatiform mole was found to be 1 per 1,000.000 pregnancies [3]. It's a rarity and less than 50 cases have been reported in the literature[4]. Patients with tubal molar pregnancy are very difficult to distinguish from patients with non-molar tubal pregnancy by means of presenting signs, symptoms or laboratory test[5]. Human chorionic (HCG) level is elevated in molar intrauterine pregnancies, but found to be in lower range in tubal molar pregnancy because implantation in the fallopian tube precludes adequate vascularization, and therefore may not be a good marker to diagnose this condition[6]. Accurate histopathology assessement of such cases remains the most reliable method of diagnosing these cases[7]. Management of ectopic molar gestation includes immediate removal of conceptus either via laparotomy or laparoscopically, followed by histological evaluation of the specimen and follow-up using serial hCG measurements similar to other trophoblastic tumors[2]. The outcome of the treatement of these patients is similar to those coming with non-molar ectopic gestation. We report a rare condition of ruptured tubal molar pregnancy.*

Keywords: Ectopic molar pregnancy, surgery, histology, HCG.

Résumé : *Les maladies trophoblastiques gestationnelles (TTG) constituent un spectre de tumeurs caractérisées par une prolifération anormale de tissus trophoblastiques associés à la grossesse, avec un potentiel malin progressif. Les TTG sont classées en maladies prémalignes, appelées taupes hydatiformes complètes et partielles (CM, PM), et en troubles malins (taupe invasive, tumeur trophoblastique du site placentaire et choriocarcinome) [1]. Dans la plupart des cas, les grains de beauté se développent dans la cavité utérine, mais peuvent se trouver à n'importe quel endroit. La grossesse ectopique des molaires est un événement rare, le premier rapport sur les taupes tubaires a été publié en 1872 par Otto [2]. L'incidence de la mole hydatiforme ectopique hydatiforme était de 1 pour 1.000.000 de grossesses [3]. C'est une rareté et moins de 50 cas ont été signalés dans la littérature [4]. Les patientes ayant une grossesse tubaire molaire sont très difficiles à distinguer des patientes ayant une grossesse tubaire non molaire par la présentation de signes, de symptômes ou par des analyses biologiques[5]. Le taux d'HCG est élevé dans les grossesses intra-utérines molaires, mais il reste inférieur à celui dans les grossesses tubaires molaires parce que l'implantation dans les trompes empêche une vascularisation adéquate, et peut donc ne pas être un bon marqueur pour diagnostiquer cette affection [6]. Une évaluation histopathologique précise de ces cas reste la méthode la plus fiable pour les diagnostiquer [7]. La prise en charge de la grossesse molaire ectopique comprend l'ablation immédiate du conceptus par laparotomie ou par laparoscopie, suivie d'une évaluation histologique de l'échantillon et d'un suivi à l'aide de mesures de la hCG sériques, comme pour les autres tumeurs trophoblastiques [2]. Le résultat du traitement de ces patientes est similaire à celui de la gestation ectopique non molaire. Nous rapportons un rare cas de de grossesse tubaire molaire rompue.*

Mots clès : mole hydatiforme ectopique, chirurgie, histologie, HCG.

Introduction:

The hydatidiform mole is the most common gestational trophoblastic disease, its incidence is 0.1% among all pregnancies, it is due to a malformation of the placenta as a result of a genetic aberration of the city trophoblast. Its characteristic feature is the increased levels of human chorionic gonadotropin beta (β -HCG) which is useful for diagnosis as well as follow-up after treatment and for the prediction of recurrence. It develops in the uterus and its ectopic localization is very rare. We report a rare case of ectopic hydatidiform mole managed in our training.

Anatomopathology:

Hydatiform mole is a cystic involution of the placenta that invades the endometrium. The mole can be complete, in the absence of a fetus, or partial with the development of a fetus, normal or not. The anatomopathological examination of a complete (CM) finds very large

villi with a central edema. There is cystization and hyperplasia of the perivillosal trophoblast, composed of syncitiotrophoblasts of cytotrophoblasts and of intermediate cells with cytonuclear atypia. These villi are very rarely vascularized; some rare vessels not containing nucleated cells are sometimes seen in villous segments with little or no edema. CM results from the fecundation of a nucleated ovum by a haploid sperm that will duplicate or by two sperm [8, 9].

The anatomopathological examination of a partial Hydatiform mole (PM) finds a mixture of molar and nonmolar villi with sometimes a discreet hyperplasia of the perivillository trophoblast. The villi are vascularized and the vessels may contain nucleated red blood cells. An amniotic cavity associated with fetal debris is frequently observed. MHP results from the fecundation of a haploid ovum by two spermatozoa [10].

Case report:

The following case is about a 28-year-old with no pathological history, mother of three children delivered by the vaginal route, of low socio-economic status, presented for the management of acute right-sided pelvic pain associated with an 8 week amenorrhoea aggravating a vomiting episode.

General clinical examination found a conscious, hemodynamically unstable patient hypotensive at 08/06cmHg tachycardic at 120bpm, polypneic, with discolored conjunctiva, apyretic. Gynaecological examination shows a macroscopically normal cervix without metrorrhagia, slight right latero-uterine tenderness with a normal sized uterus.

After the patient's condition and stabilisation, a pelvic ultrasound scan was carried out, which revealed an empty uterus of normal size, the presence of a heterogeneous right latero-uterine echogenic image with a very abundant peritoneal effusion reaching the Morrison's space. A ruptured ectopic pregnancy was thus suspected.

Hence the decision to perform culdocentesis, which brought back incoagulable blood, and the patient was then sent to the operating room for laparotomy. Upon exploration, a large-abundance aspirated hemoperitoneum was discovered, estimated at 2 liters, the right tube ruptured with visualization of the product of conception containing vesicles, hence the decision to perform a right salpingectomy. The patient was transfused by 2CG with a hemoglobin at 8.6 g / dl, blood grouping A +. A BHCG dosage recovered a posteriori was elevated to 8940 IU/L.

The product of conception with the salpingectomy specimen was sent for an anatomopathological study returning in favour of a partial hydatiform mole, thus concluding in a very rare case of ruptured ectopic molar pregnancy.

The patient was placed on estrogen-progestogen contraception with clinical and biological monitoring of the quantitative level of BHCG in the same laboratory. The outcome was favorable with a gradual and significant decrease in the level of BHCG and negativation after two weeks. Monthly monitoring was subsequently maintained for 6 months as is the case for partial molar intrauterine pregnancy.

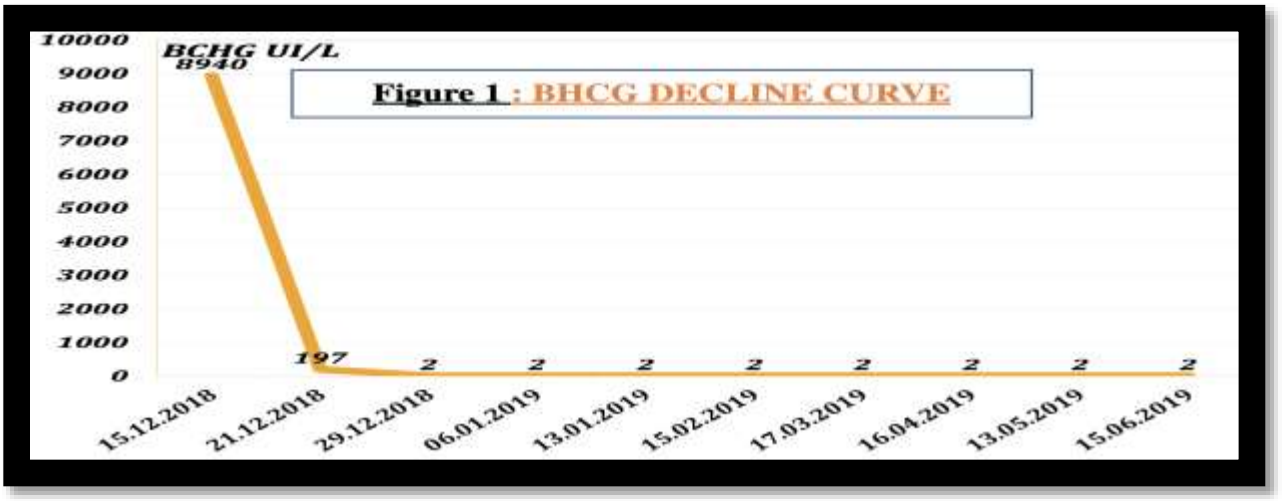

Figure 1 : BHCG decline curve :

Discussion:

Mole hydatidiform is a rare complication of pregnancy and is the most common gestational trophoblastic disease in women of childbearing age. Its incidence varies across the world, ranging from 1 per 1000 to 1200 pregnancies in the United States to 10 per 1000 in Indonesia[12].

It is a malformation of the placenta due to a genetic aberration of the villous trophoblast characterized by cystic swelling and varying degrees of trophoblastic proliferation[12].

Molar gestation commonly develops within the uterus but may also occur in sites of ectopic pregnancy, However, tubal ectopic hydatidiform moles are rare and less than 50 cases have been reported. The incidence of ectopic hydatiform mole was found to be 1 per 1,000.000 pregnancies [3]. Predisposing risk factors to development of tubal hydatidiform mole include, pelvic inflammatory disease, prior hydatidiform mole and advanced or young maternal age, nulliparity, oral contraceptive use and low socio-economic status[12]. Our patient was 28 years old with a low socio-economic status.

Clinically, tubal molar pregnancy mimics normal tubal ectopic pregnancy, thereby making diagnosis difficult. The diagnosis of a molar pregnancy is based on clinical symptoms, ultrasound, quantitative BHCG levels that mimic those of an ectopic pregnancy, in contrast to the high values found in an intrauterine molar pregnancy. MRI is a valuable tool for diagnosing ectopic hydatidiform moles due to the tiny cystic lesions resembling molar tissue, intratumoral hypervascularization, and increased density[14].

Tasha report in his study that tubal rupture occurred early in the molar group compared to the normal group. This may be due to the higher likelihood for invasion and penetration of trophoblastic tissue in GTD as compared to the trophoblast in a normal pregnancy[13].

The management of ectopic molar pregnancy consists of the surgical removal of the conceptus, preferably by laparoscopy. The trophoblast must be completely removed. The prognosis for ectopic molar pregnancy is the same as for other forms of gestational trophoblastic disease. Thus, molar pregnancy can potentially be complicated by persistent trophoblastic disease and malignant transformation as in intrauterine molar pregnancy[3].

Hisologically, enlargement of villi and marked hydrophobic changes, cistern formation and circumferential trophoblastic proliferation are the histological criteria for diagnosing a molar pregnancy in the literature, applicable to their more common uterine counterpart. Some studies have shown that there may be an over-diagnosis of ectopic molar pregnancy suggesting that the pathological criteria must be strict in the diagnosis of ectopic moles [15]. However, diagnosis can be challenging for the pathologist since the differences between molar pregnancies and hydropic abortions may be minimal, especially in early miscarriages, and the evaluation of the specimen depends on the experience of the examiner and can be highly subjective. On the other hand, immunohistochemical analysis using antibodies such as p57 can distinguish between the hydatiform mole with its different types and hydrophilic miscarriage. When available, cytometric DNA flow analysis to determine ploidy helps to differentiate the complete mole from the partial mole [15].

The risk of gestational trophoblastic disease (GTD) after salpingectomy in tubal moles is considered much lower compared to intrauterine counterparts because the lesion was completely removed. For intrauterine moles, approximately 20% of complete and 3.5% of partial moles progressed to GTD. There were only sporadic reports of tubal moles progressed to GTD after salpingectomy and all the reported cases achieved remission after methotrexate or second-line chemotherapy, yet the risk of GTD after salpingotomy in tubal moles is undefined [11].

Postoperative follow-up is mandatory. It requires full monitoring of βhCG and contraceptive use. Biological monitoring is essential based on patients'serum HCG levels until they become undetectable according to the recommended protocol: once a week for 3 consecutive weeks, then once a month for 6 consecutive months, in order to prevent recurrence and to facilitate early diagnosis of persistent trophoblastic disease or its malignant choriocarcinoma. However, a single undetectable level of HCG after evacuation is sufficient to monitor partial môle[12].

The outcome of the treatement of these patients is similar to those coming with non-molar ectopic gestation.

Conclusion :

Ectopic molar pregnancy is a rare event. Only a pathological examination of the resected tubal specimens can confirm the diagnosis and clarify the form of the molar pregnancy and thus initiate appropriate follow-up.

Références :

1. Mickael JS, Edward SN. Gestational trophoblastic tumours. In: Shaw RW, editor. Gynaecology. 3rd ed. London (UK): Elsevier Science Limited 2003. P. 653-64.
2. Rees HC, Paradinas FJ. The diagnosis of hydatidiform mole in early tubal ectopic pregnancy. Histopathology 2001;39:320-1.
3. Gillespie AM, Lidbury EA, Tidy JA, Honcock BW. The clinical presentation, treatment and outcome of patient diagnosed with possible ectopi molar gestation. Int J Gynaecol Cancer 2004;14:366-9.
4. Petersen R.W, Ung K, Holland C, Quinlivan J.A. The impact of mo- lar pregnancy on psychological, symptomatology, sexual functions and qualit of life. Gynecologic Oncology 2005; 97: 535-42.
5. Shamshiri-MilaniH Molar Pregnancy and husband's occupation:Do soil and dust have any role?East Mediterr HealthJ 2008;14:183-8.

6 Christopher PC. The female genital tract. In: Kumar V, Abdul K, Nelson F editors. Pathologic bases of disease. 7th ed. Philadelphia (US): Elsevie Inc; 2005. p.1059-119 Eqwuatu VE, Ozumba BC. Observation on molar pregnancy in Enugu, Nigeria. Int J Gynaecol Obstet1989;29:219-25.
7. Eqwuatu VE, Ozumba BC. Observation on molar pregnancy in Enugu, Nigeria. Int J Gynaecol Obstet1989;29:219-25.
8 Lawler SD, Fisher RA, Dent J. A prospective genetic study of complete and partial hydatidiform moles. Am J Obstet Gynecol 1991;164:1270
9. Lage JM, Sheikh SS. Genetic aspects of gestational trophoblastic diseases: a general overview with emphasis on new approaches in determinin genetic composition. Gen Diagn Pathol 1997;143:109–15.
10 Altieri A, Franceschi S, Ferlay J, et al. Epidemiology and aetiology of gestational trophoblastic diseases. Lancet Oncol 2003;4:670–8.
11. Zhao T, Hou X, Su C, Wu Q. Tubal hydatidiform mole treated with salpingotomy: A case report. Clin Case Rep. 2019;7:653–655.
12 Ruptured tubal hydatidiform mole, Modupeola Omotara Samaila[1], Adebiyi Gbadebo Adesiyun[2], Calvin Bifam, J Turkish-German Gynecol Assoc 2009; 10: 172-4
13 Tasha I, Kroi E, Karameta A, Shahinaj R, Manoku N. Preva- lence of gestational trophoblastic disease in ectopic pregnancy. J Prenat Med 2010; 4: 26–29.
14 Yasushi Yamada, Satoshi Ohira, Teruyuki Yamazaki and Tanri Shiozawa (2016) Ectopic Molar Pregnancy: Diagnostic Efficacy of Magnetic Resonance Imaging and Review of the Literature. Case Reports in Obstetrics and Gynecology 2016: 7618631.
15 Nora Mohamed, Laila Moharram, Hedaya Atalla M Alblewi. Ectopic Hydatidiform Mole a Case Report. Nov Res Sci. 4(3). NRS.000586. 2020. DOI: 10.31031/NRS.2020.4.000586.