

## SHORT REPORT



# Hydatidiform mole resulting from sexual violence

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## Abstract

**Background:** Hydatidiform mole (HM) is characterized by abnormal proliferation of human trophoblast with producers functioning tissues of human chorionic gonadotropin. It can evolve with ovarian cysts tecaluteínicos, hypertension of pregnancy or hyperthyroidism. The incidence of HM is variable and its etiology poorly known, associated with nutritional factors, environmental, age, parity, history of HM, oral contraceptives, smoking, consanguinity or defects in germ cells. There is no reference in literature on HM resulting from sexual violence, objective of this report.

**Method:** Description of two cases of HM among 1146 patients with pregnancy resulting from sexual violence treated at Hospital Pérola Byington, São Paulo, from July 1994 to August 2011.

**Results:** The cases affected young, white, unmarried, low educated and low parity women. Sexual violence was perpetrated by known offenders unrelated to the victims, under death threat. Ultrasound and CT of the pelvis showed bulky uterus compatible with HM without myometrial invasion. One case was associated with theca lutein cysts. The two cases were diagnosed in the second trimester of pregnancy and evolved with hyperthyroidism. There was no hypertension, disease recurrence, metastasis or sexually transmitted infection.

**Conclusion:** The incidence of HM was 1:573 pregnancies resulting from rape, within the range estimated for Latin American countries. Trophoblastic material can be preserved to identify the violence perpetrator, considering only the paternal HM chromosomes. History of sexual violence should be investigated in cases of HM in the first half of adolescence and women in a vulnerable condition.

## Background

Gestational impairments are important factors to be investigated [1,2], since it affects children development [3-5]. The hydatidiform mole (HM) is a variant of gestational trophoblastic disease, a generic term that brings together different conditions of proliferation of trophoblastic epithelium, with abnormal functioning tissue producing human chorionic gonadotropin (hCG) [6]. The gestational trophoblastic disease includes forms of HM, invasive mole, gestacional choriocarcinoma and placental site trophoblastic tumor, according to anatomoclinical criteria [7]. The choriocarcinoma is the most severe form of the disease with potential for invasion and metastasis, affecting every 20,000 pregnancies [8]. The etiology of HM is unknown. Studies associate it with nutritional deficiencies, environmental conditions, herbicides, parity, smoking, extremes of maternal age, intrauterine device, inbreeding, viral infections, oral hormonal contraception, history of HM or germ cell defects [9]. The incidence of HM varies. In Latin American countries may have frequency as low as 1:3.846 pregnancies, while in Asian countries its incidence reaches 1:99 [10,11]. This divergence probably results from a heterogeneous way of collecting data on population or hospital basis.

Sexual violence has a high prevalence and commits primarily young women of reproductive age [12-15]. Although the risk of pregnancy in these cases does not exceed 5% in each exposure, it is estimated that about



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30.000 forced pregnancies are carried out only in the U. S. every year [16]. In addition, even facing the large number of cases of pregnancy resulting from sexual violence, there is no reference in literature on its association with HM. A literature review indicating the occurrence of HM in age that features sex crime, according to the legislation of each country, would justify the communication to the competent authority and the investigation of sexual violence. Therefore, this study was undertaken to describe two cases of HM after rape.

## Method

Report of two cases of HM from 1146 women and adolescents with pregnancy resulting from sexual violence treated at Pérola Byington hospital, São Paulo, from July 1994 to August 2011, by consulting the medical records. All procedures were approved by the Ethical Committee in Research of our Institution (Protocol number: 003/ 08). The literature review to verify the association between sexual violence and HM consulted database Journal Citation Reports (JCR-ISI), Medical Literature Analysis and Retrieval System Online (Medline), Scientific Electronic Library Online (SciELO) and Latin American and Caribbean Literature in Health Sciences (LACLHS). The search strategy used DeCS/MeSH descriptors with syntax ("Hydatidiform Mole" [MeSH]) AND "Rape" [MeSH] and the syntax ("Hydatidiform Mole" [MeSH]) AND "Child Abuse, Sexual" [MeSH Terms] [17,18].

## **Case report**

No items were identified in databases relating HM and sexual violence. The incidence of HM was 1:573 pregnancies resulting from rape. The assaults were reported by two young, white and unmarried. Sexual violence was committed by a known abuser unrelated to the victim by threat of death. Ultrasound and non-contrastenhanced CT of the pelvis showed bulky uterus and compatible with HM content, with no signs of invasion of the myometrium or adjacent structures (Figure 1). In one case, the HM was associated with theca lutein cysts





with right ovary with 232 cc and left with 227 cc (Figure 2). The cysts showed complete spontaneous remission four months after uterine evacuation.

In Table 1 we observe the synthesis of two cases of hydatidiform mole resulting from rape.

The cases evolved with hyperthyroidism and the spontaneous normalization of thyroid-stimulating hormone (TSH) occurred in the first month after treatment. There was no hypertensive disorders of pregnancy (HDP). There were not identified significant changes in blood count, coagulogram, oxaloacetic transaminase (SGOT), pyruvic transaminase (SGPT), urea, creatinine, glucose, calcium, and thyroid ultrasound. Clinical and laboratory parameters, including CT control of the chest and pelvis, and the dose of beta hCG fraction, showed that cases were not associated with recurrent or metastatic disease. None of the cases underwent prophylactic chemotherapy.

Serology for hepatitis B and C, HIV, syphilis and HTLV I and II were negative when enrolled at the

service and for the six-month period for monitoring of sexual violence. There were no positive sexually transmitted infections in cervical scrapes and vaginal content. There were no complications that could compromise the reproductive future of the patients. The cases were formally reported to the police by the patients and the rape against the adolescent was reported by the hospital to the governmental agency for protection.

#### Discussion

In this study, we described the evolution of the two cases reported of HM after rape. The physical attacks were reported by two young girls. Also, sexual violence was committed by a known abuser.

Based on our data, the both cases presented hyperthyroidism and there was no HDP. Moreover, the spontaneous normalization of TSH was observed in the first month after treatment. We reported changes in blood components, such coagulogram, SGOT, SGPT,

	Case 1	Case 2
Age	22 years	13 years
Parity	Two	Zero
Gestational age	13 weeks	16 weeks
Uterus volume	740 сс	920 cc
Initial Beta hCG (mIU/ml)	320.080	960.320
Theca lutein cysts	No	Yes
Serum TSH levels	0,011 mcg/dl (0,4-5,5)	0,013 mcg/dl (0,4-5,5)
Evolution with HDP	No	No
Tomography of Thorax	Normal	Normal
Uterine evacuation	Manual vacuum aspiration	Manual vacuum aspiration
Anatomopathological	Complete hydatidiform mole	Partial hydatidiform mole

Table 1	1 Synthesis of two	cases of hy	ydatidiform n	mole resulting	from	rape
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cc (cubic centimeters) TSH (thyroid-stimulating hormone)

HDP (Specific Hypertension Disease in pregnancy)

urea, creatinine, glucose, calcium, and thyroid ultrasound. According to the literature, the HM can be complete when there is no formation of embryonic or fetal tissue, characterized by diffuse hyperplasia of the cytotrophoblast and syncytiotrophoblast. Partial or incomplete fetal material can be identified along tissue with stromal trophoblastic inclusions [19,20]. HM usually leads to vaginal bleeding in the first quarter and disproportionate uterine growth in relation to gestational age, accompanied by nausea and frequent vomiting, or hyperemesis gravidarum.

Based on clinical and laboratory parameters, including CT control of the chest and pelvis, and the dose of beta hCG fraction, the cases were not associated with recurrent or metastatic disease. None of the cases underwent prophylactic chemotherapy. In 15% to 25% of cases, the high concentration of hCG induces the formation of bulky ovarian theca lutein cysts, with more than 60 mm in diameter [21]. Early HDP is observed in 27% of complete HM and may end in eclampsia [22]. In addition, the HM can progress to full malignant forms of gestational trophoblastic disease in 20% of cases. Hyperthyroidism is found in less than 10% of the cases of HM, resulting from cross-stimulation of the TSH receptors by alpha chain of hCG [23].

Our findings are in agreement with the literature, since women at the extremes of reproductive age are more likely to develop HM. The relative risk is 7 to 10 times higher among those with more than 40 years [24]. However, the absolute number of HM cases after 40 years is small, due to decreased fertility and reduced frequency of pregnancy in this age group. The risk is 2 times higher in the first half of adolescence, although there are few records of HM under 15 [25].

The aging of the eggs may explain in part the increased susceptibility to abnormal gametogenesis and fertilization in women over 40 years, but does not justify

the HM in younger women. Most studies suggest that the higher maternal age increased the incidence of choriocarcinoma [26]. In addition to maternal age as a recognized risk factor, personal history of HM increases up to 40 times the risk of the disease in following pregnancies [24]. Parity does not seem to have a decisive influence on the incidence, with 12% to 78% of cases of HM between nulliparous. Few studies suggest that the incidence grows with parity, or that relates to increased risk of choriocarcinoma [26].

Information on other risk factors are limited and conflicting, and may vary when the HM is complete or partial. However, in Asian countries there is sharp decrease in the incidence of HM in recent decades related to the improvement of socioeconomic conditions of the population, suggesting the importance of environmental or nutritional factors in its etiology [27].

In our study, the both cases were formally noticed to the police by the patients and the rape against the adolescent was reported by the hospital to the governmental agency for protection. Sexual violence is not associated directly or indirectly, with most known risk factors for HM. There is exception for age and parity, since the majority of rape victims are young women or adolescents, often nulliparous [12]. There is also no evidence that male factors have influence on the etiology of HM except inbreeding. Therefore, it cannot be attributed to the sexual violence's author who is unrelated to the victim a risk factor for HM, which can be corroborated by the incidence of HM 1:573 pregnancies resulting from sexual abuse observed in this report, within the limits of variation in Latin American countries [24,26].

However, the relationship between the perpetrator and HM may have important legal implications. In 90% of complete HM the karyotype is 46XX, and 10%, 46XY. In partial HM triploid karyotype is shown in 90% of cases, 69XXX or 69XXY with tetraploidy in 10%. All of

these are exclusively paternal chromosomes [19,20,26]. The proper preservation of part of the molar tissue allows to keep for a long period, genetic material that can be used for DNA analysis and determination of paternity. This issue is relevant to the identification and accountability of perpetrators of sexual violence, often not guilty by the courts for lack of evidence [12].

The absence of reports in the literature linking HM to sexual violence must be interpreted with reservation. HM cases reported in the first half of adolescence could be due to sexual violence, to the extent that the laws of many countries typifies sexual activity as a crime under certain age limits [25]. In Brazil, for example, it is considered a crime to have sex with children under 14 years. The underreporting of sexual violence, estimated at almost 90% of cases, can also interfere with the lack of records. The embarrassment and the threat of death can lead many women to choosing not to disclose to health professionals that the HM arises from sexual violence [12].

### Conclusion

In summary, the occurrence of HM in age that features sex crime, according to the legislation of each country, justifies the communication to the competent authority and the investigation of sexual violence. Our findings indicate that reported cases of sexual violence in HM require additional health professionals care with an investigation of sexually transmitted diseases, psychological treatment and support for legal action seeking and protection.

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#### Authors' contributions

All authors participated in the acquisition of data and revision of the manuscript. All authors determined the design, interpreted the data and drafted the manuscript. All authors read and gave final approval for the version submitted for publication.

#### **Competing interests**

The authors declare that they have no competing interests.

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#### References

 Moutasim KA, Shirlaw PJ, Escudier MP, Poate TW: Congenital heart block associated with Sjögren syndrome: case report. Int Arch Med 2009, 2:21.

- Metaferia AM, Muula AS: Stillbirths and hospital early neonatal deaths at Queen Elizabeth Central Hospital Blantyre-Malawi. Int Arch Med 2009, 2:25.
- Shoeps DO, de Abreu LC, Valenti VE, Nascimento VG, de Oliveira AG, Gallo PR, Wajnsztejn R, Leone C: Nutritional status of pre-school children from low income families. *Nutr J* 2011, 10:43.
- Nascimento VG, Machado TC, Bertoli CJ, de Abreu LC, Valenti VE, Leone C: Evaluation of mid-upper arm circumference in pre-school children: comparison between NCHS/CDC-2000 and WHO-2006 references. J Trop Pediatr 2011, 57:208-212.
- Torigoshi MF, Valenti VE, Siqueira AAF, Reis AOA, Leone C, Abreu LC: Outcomes of newborns admitted in the intensive care unit at a public hospital. *Health MED Journal* 2011, 5:295-300.
- Bagshawe KD: Introduction. In *Gestacional trophoblastic disease*. 2 edition. Edited by: Hancock BW, Newlands ES, Berkow RS, Cole LA. Chapman 2004:1.
- Genest DR, Berkowitz RS, Fisher RA: Gestacional trophoblastic disease. In World Health Organization classification of tumors. Pathology and genetics: tumors of the breast and female genital organs. Edited by: Tavassoli FA, Deville P. Lyon: IARC Press; 2003:250-256.
- Feltmate CM, Genest DR, Wise L, Bernstein MR, Goldstein DP, Berkowitz RS: Placental site trophoblastic tumor: a 17-year experience at the New England Trophoblastic Disease Center. *Gynecol Oncol* 2001, 82:415-419.
- Bracken MB: Incidence and etiology of hydatidiform mole: an epidemiological review. Br J Obstet Gynaecol 1987, 94:1123-1135.
- Rolón PA, de Lopez BH: Epidemiological aspects of Hydatidiform mole in the Republic of Paraguay (South America). Br J Obstet Gynaecol 1977, 84:862-864.
- Poen HT, Djojopranoto M: The possible etiologic factors of hydatidiform mole and choriocarcinoma: preliminary report. Am J Obstet Gynecol 1965, 92:510-513.
- Drezett J, Junqueira L, Tardelli R, Antonio IP, Macedo H Jr, Vertamatti MAF, Pimentel RM, Abreu LC: Influence of forensic examination on the accountability of sexual violence authors in teenagers. *Rev Bras Cresc e Desenv Hum* 2011, 21:189-197.
- Hayati EN, Hogberg U, Hakimi M, Ellsberg MC, Emmelin M: Behind the silence of harmony: risk factors for physical and sexual violence among women in Rural Indonesia. *BMC Womens Health* 2011, 11:52.
- Osinde MO, Kaye DK, Kakaire O: Intimate partner violence among women with HIV infection in rural Uganda: critical implications for policy and practice. *BMC Womens Health* 2011, 11:50.
- Lamichhane P, Puri M, Tamang J, Dulal B: Women's status and violence against young married women in rural Nepal. BMC Womens Health 2011, 11:19.
- Holmes MM, Resnick HS, Kilpatrick DG, Best CL: Rape-related pregnancy: estimates and descriptive characteristics from a national sample of women. *Am J Obstet Gynecol* 1996, 175:320-324.
- Ferreira Filho C, Abreu LC, Valenti VE, Ferreira M, Meneghini A, Silveira JA, Riera AR, Colombari E, Murad N, Santos-Silva PR, Silva LJ, Vanderlei LC, Carvalho TD, Ferreira C: Anti-hypertensive drugs have different effects on ventricular hypertrophy regression. *Clinics* 2010, 65:723-728.
- Ferreira M, Santos-Silva PR, de Abreu LC, Valenti VE, Crispim V, Imaizumi C, Filho CF, Murad N, Meneghini A, Riera AR, de Carvalho TD, Vanderlei LC, Valenti EE, Cisternas JR, Moura Filho OF, Ferreira C: Sudden cardiac death athletes: a systematic review. Sports Med Arthrosc Rehabil Ther Technol 2010, 2:19.
- 19. Bentley RC: Pathology of gestacional trophoblastic disease. *Clin Obstet Gynecol* 2003, **46**:513-522.
- Lawler SD, Povey S, Fisher RA, Pickthall VJ: Genetic studies on hydatidiform moles. II. The origin of complete moles. Ann Hum Genet 1982, 46:209-222.
- Montz FJ, Schlaerth JB, Morrow CP: The natural history of theca lutein cysts. Obstet Gynecol 1988, 72:247-251.
- Belfort P, Braga A: The changing clinical presentation of molar pregnancy. *Rev Bras Ginecol Obstet* 2004, 26:483-488.
- 23. Amir SM, Osathanondh R, Berkowitz RS, Goldstein DP: Human chorionic gonadotropin and thyroid function in patients with hydatidiform mole. *Am J Obstet Gynecol* 1984, **150**:723-728.
- 24. Smith HO: Gestacional trophoblastic disease epidemiology and trends. *Clin Obstet Gynecol* 2003, **46**:541-556.

- 25. Chapman GW Jr: Characteristics of molar pregnancy in a 12 year old. J Natl Med Assoc 1996, 88:806-807.
- Belfort P: Doença trofoblástica gestacional. In Obstetrícia. 12a ed. Guanabara Koogan, Rio de Janeiro Edited by: Rezende JM, Montenegro CAB 2011, 380-400.
- Kim SJ, Bae SN, Kim JH, Kim CJ, Han KT, Chung JK: Epidemiology and time trends of gestational trophoblastic disease in Korea. Int J Gynaecol Obstet 1998, 60:S33-S38.

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