Rare Presentation of Chorioadenoma Destruens as Acute Haemoperitoneum Mimicking Ruptured Ectopic Pregnancy

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Citation

Sinha M, Kaur R, Gupta R, Rani R, Aggarwal A. Rare Presentation of Chorioadenoma Destruens as Acute Haemoperitoneum Mimicking Ruptured Ectopic Pregnancy. *Kathmandu Univ Med J* 2014;48(4):288-91.

ABSTRACT

Gestational trophoblastic neoplasms (GTN) are proliferative degenerative disorders of placental elements and include complete or partial mole (90%), invasivemole (5-8%), choriocarcinoma (1-2%) and placental site tumor (1-2%). Chorioadenoma destruens is a trophoblastic tumor, characterized by myometrial invasion through direct extension or via venous channels. We present a case of invasive mole eroding uterus and uterine vasculature, causing sudden rupture of uterus with massive haemoperitoneum mimicking ectopic pregnancy. A 20 year old G1P0 at 6 weeks gestation presented in Casualty of Kasturba Hospital complaining of severe acute onset lower abdominal pain for one hour. Clinical examination revealed shock. Sonography suggested ectopic pregnancy and immediate exploratory laparotomy was decided. On laparotomy, 2000cc of haemoperitoneum was noted. Grape like vesicles protruding through fundal perforation with profuse active bleeding was seen. Bleeding persisted despite evacuation. Step wise uterine devascularisation failed to achieve haemostasis. Total abdominal hysterectomy was performed as a life saving measure.

KEY WORDS

Ectopic pregnancy, haemoperitonem, invasive mole

INTRODUCTION

Invasive mole or chorioadenoma destruens comprises 15 % of all gestational trophoblastic neoplasia (GTN).[1] GTN is characterized by histological abnormalities of chorionic villi with oedema of villous stroma and varying degrees of trophoblastic proliferation. Absence or presence of fetal or embryonic elements classifies complete or partial mole. Molar pregnancies which fail to regress result in invasive mole, choriocarcinoma, persistent trophoblastic tumor or placental site trophoblastic tumor.[2]

Molar pregnancies become Invasive moles in 20% and are commoner in complete molar pregnancies.[3,4] Chorioadenoma destruens is a trophoblastic tumor, characterized by myometrial invasion through direct extension or via venous channels with persistence of edematous chorionic villi and trophoblastic proliferation invading myometrium.[5] The presence of villi in the trophoblastic tissue differentiates an invasive mole from choriocarcinoma Instead of vaginal bleeding (97%) chorioadenoma destruens may present rarely as ruptured ectopic pregnancy in shock, incomplete abortion, menorraghia or carcinoma endometrium.[6,7]

We present a case of invasive mole which gradually eroded in the uterus and the uterine vasculature, leading to sudden rupture of uterus, massive hemoperitoneum and shock, mimicking ectopic pregnancy.

CASE REPORT

A 20 year old G1P0 at 6 weeks gestational age presented to the emergency room of Kasturba Hospital complaining of severe lower abdominal pain, acute in onset and for the past one hour. Her pain was severe, colicky in nature and radiated to the back and thighs. She denied any nausea, vomiting or fever. Her past medical and surgical histories were unremarkable. She denied any known drug allergies and was not on any medications. She attained menarche at

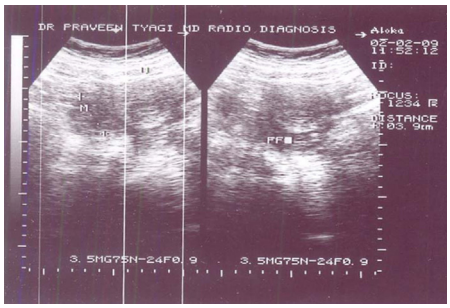

Figure 1. Ultra Sonography prior to surgery

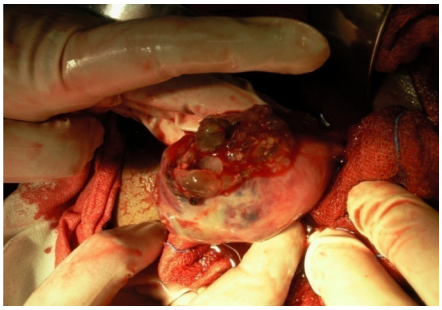

Figure 2. Uterine fundal perforation showing grape like vesicles protruding through the perforation

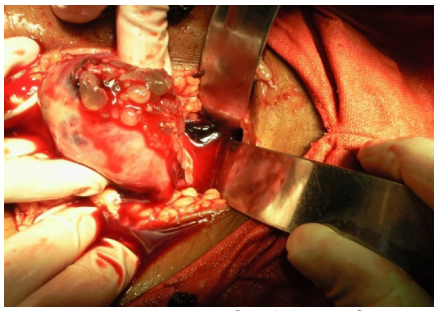

Figure 3. Uterine fundal perforation showing grape like vesicles protruding through the perforation& profuse bleeding

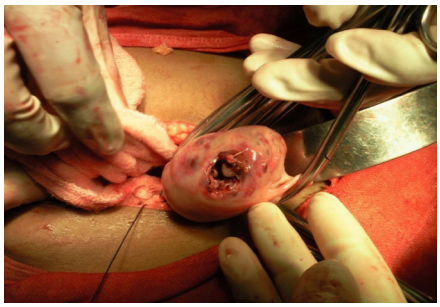

Figure 4. During hysterectomy- uterus showing large perforation

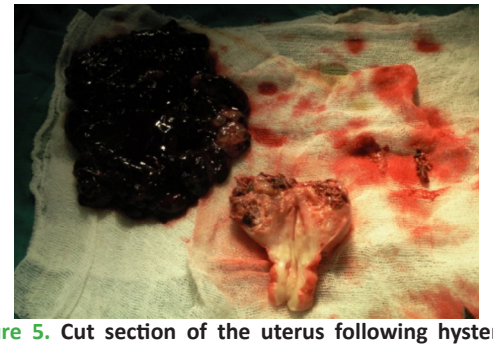

Figure 5. Cut section of the uterus following hysterectomy showing chorioadenoma destruens with tissue & Blood clot

the age of 12 years and her menstrual cycles were regular. She denied any pregnancies in the past and had a marriage history of one year.

On examination patient was awake, alert and oriented but was in apparent distress. She was hypotensive with BP of 90/60 mm of Hg, tachycardic with pulse of 112 b/min. She was pale clinically, afebrile and her respiratory rate was 16/min. Her cardiac, respiratory and neurological system evaluation was unremarkable

On abdominal examination, she was severely tender in bilateral lower quadrants, with presence of guarding, rigidity and rebound tenderness. On bimanual examination, cervical os was closed, the uterine size could not be assessed due to presence of guarding and tenderness. These findings were highly suggestive of ectopic pregnancy and emergency transvaginal ultrasound (TVS) was performed, which demonstrated anteverted uterus, measuring approximately – 46 x 60 x 76 mm with irregular echogenic products seen in uterine cavity. Pouch of Douglas showed moderate amount of free fluid. However, a definitive diagnosis of ectopic pregnancy could not be established.

During TVS, patient became drowsy and slipped into a semi comatose condition. Her vitals deteriorated with pulse of 140/min and systolic blood pressure of 60 mm of Hg. Patient was immediately rushed to operation theatre for Emergency laparotomy with a definitive suspicion of a ruptured ectopic pregnancy. On laboratory investigation, her haemoglobin and hematocrit was 6 gram and 20 mg percent, random blood sugar values of 130 mg/dl, Serum Bilirubin 1.2 mg/dl, and Serum creatinine 0.9 mg/dl. Her beta HCG could not be sent as emergency laboratory facilities of Kasturba Hospital does not include this investigation.

On, exploratory laparotomy, 2000cc of hemoperitoneum was noted, with peritoneal cavity full of blood and clots. Uterus was perforated at the fundus and profuse active bleeding was noted at the site of perforation. Grape like vesicles were protruding through the uterine perforation which measure approximately 3 cm x 3 cm (Fig 2, Fig 3). The uterus was eight weeks in size, soft and enlarged, bilateral adenxae were normal. On further abdominal exploration, no ectopic tissue or metastatic tissue was identified.

Profuse ongoing bleeding was seen from site of perforation following complete evacuation of the uterus and patient continued to remain in a state of shock despite adequate fluid replacement, 4 units of blood transfusion and 20 units of continuous oxytocin infusion. To achieve haemostasis, step wise systematic uterine devascularization was attempted but complete haemostasis could be not be achieved and patient continued to bleed profusely from the perforated site. Decision was made to proceed with total abdominal hysterectomy as a life saving measure. Post surgery patient became haemodynamically stable. Hysterectomized uterus was then sent for histopathological examination (Fig 4, Fig 5).

Her postoperative course was uncomplicated.

On postoperative day one, her vitals improved and her hemoglobin was 8 gm/dl. She received additional two units of blood transfusion and made an uneventful recovery.

The postoperative metastatic work-up included serum biochemistry, chest x-ray and upper abdominal Ultrasonography and no evidence of metastasis was detected. In view of low levels of serum ß-hCG, patient was kept under surveillance with serial quantitative ß-hCG estimation to consider chemotherapy if ß-hCG levels

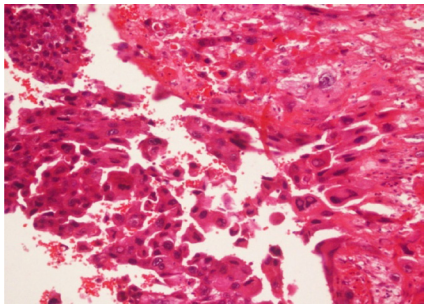

Figure 6. Section from uterus showing a tumour arising from endometrial surface and composed of intermediate trophoblasts and afew multinucleated syncytiotrophoblasts, arranged in sheets. (H&E, X40)

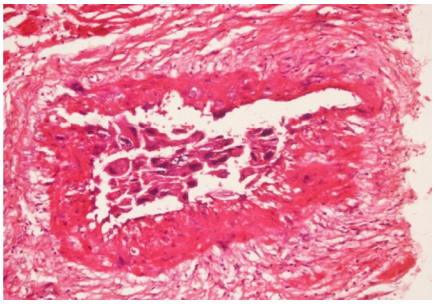

Figure 7. The tumour cells are showing vascular invasion. (H&E, X 20)

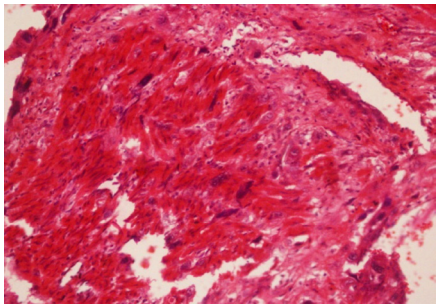

Figure 8. Syncytiotrophoblasts and intermediate trophoblasts are infiltrating the myometrium as single cell infiltrate. (H&E, X 20)

remains at a plateau or rises. Postoperatively ß-hCG level was 110 mIU/ml. Patient was discharged home in stable condition on postoperative day 3. Her ß-hCG level was <8 mIU/ml two weeks following surgery. The subsequent ß-hCG levels were negative on regular follow up visits.

Subsequently on detailed questioning patient revealed the history of evacuation for incomplete abortion about two months back, records of which were not available for review. It was debatable, if this presentation was a continuation of the previous conception or the patient conceived again.

Histopathological examination revealed Chorioadenoma Destruens with myometrial and blood vessel invasion. Section from endomyometrium showed trophoblastic cells with abundant eosinophilic cytoplasm and pleomorphic nuclei invading deep into myometrium and at places upto serosal layer. Few blood vessels are also seen invaded by these trophoblastic cells. This may explain the torrential bleeding in this patient.

DISCUSSION

Gestational trophoblastic neoplasms (GTN) are proliferative as well as degenerative disorders of placental elements and include complete or partial mole (90%), invasive mole (5-8%), choriocarcinoma (1-2%) and placental site tumor (1-2%) 15% of complete mole can develop into invasive mole. But only 2-4% of the partial mole transform into this variety of trophoblastic tumor.[8]

Complete hydatidiform moles are recognized to have a potential for developing uterine invasion or distant metastasis. Invasive mole may perforate through the myometrium resulting in uterine perforation and intraperitoneal bleeding.[9] Direct vascular invasion and metastasis rarely occurs in invasive moles, the most common site reported is the lung although an invasive mole is generally less malignant than choriocarcinoma,[10] it may be associated with fatal metastasis. Some degree of myometrial trophoblastic invasion is probably present in most moles; indeed, myometrial invasion is not exclusive to molar pregnancies. Hertig and Borrowing used the terminology used for myometrial placentation, classified invasive moles into accreta, increta and percreta.[11] Cases of gestational trophoblastic neoplasia with uterine rupture are often catastrophic owing to profuse bleeding, which could potentially be lethal.

Management often entails removal of the uterus, but among patients in the reproductive age group, uterine resection of localized disease can be performed as reported by Mittal et al.[7] The diagnosis of invasive mole rests on the demonstration of complete hydatidiform mole invading the myometrium or the presence of villi in the metastatic lesion. Myometrial invasion is difficult to document on pelvic ultrasound and also in uterine curettings unless there is a sufficient myometrium to demonstrate the invasion Transvaginal sonography combined with HCG titers is non invasive and hence a useful diagnostic tool.[8] Color flow Doppler can define lesions of increased vascularity as seen in invasive disease. Myometrial invasion is difficult to diagnose on pelvic ultrasound unless there is sufficient myometrium to demonstrate the invasion. In a study conducted by Branka et al,[12] sonography failed to show any specific changes in the uterus even one month after the development of GTN, and he concluded that USG is of limited value in detection of partial moles and malignant GTNs. However, massive tissue destruction, hot spots (hypervascularization), and low resistance index are characteristic USG findings of malignant GTNs. The role of magnetic resonance imaging studies or positron emission tomography in the evolution of women with GTN is not yet defined.[7]

Our experience showed that a high index of suspicion should be kept in women with history of abortion and irregular bleeding. Intra-operative management options are limited due to the acute presentation. Uterine evacuation is the treatment of choice in many circumstances but use of oxytocin to attain haemostasis is controversial. Mitani et al recommended partial resection for young women if invasive moles are complicated by internal haemorrhage.[13] They have reported five women treated this way, four of which subsequently delivered healthy babies by caesarean section. Goldstein et al used local uterine resection together with bilateral internal iliac artery ligation in an attempt to achieve haemostasis and preserve fertility.[14]

Chemoprophylaxis may be particularly useful in patients with a high-risk complete mole when hormonal follow-up

is either unavailable or not affordable and chances of losing the woman to follow-up are high. The recommendations for post-molar follow-up include serum βhCG levels every 1–2 weeks after evacuation until normal, hCG levels 2–4 weeks after the first normal level, and hCG surveillance every 1–2 months for 6 months after the first normal hCG level. Serial hCG monitoring of women with disease in prolonged remission (greater than 1 year) often provides early evidence of recurrence, though GTT may rarely be associated with undetectable levels of hCG.[8]

In our patient, we were unable to achieve adequate haemostasis by step wise uterine devasculaization attempted with uterine and ovarian vessel ligation. Unfortunately the surgical emergency team was not well versed with the technique of Internal Iliac Ligation, hence it was not attempted and total abdominal hysterectomy was then performed. Although the development of effective chemotherapy has resulted in improved survival of patients with gestational trophoblastic tumor, hysterectomy remains an important adjuncts in the treatment of a selected subset of patients.

Use of chemotherapy in the management of invasive mole is still debatable, with the evidence of spontaneous regression of metastatic mole in the literature. Chemoprophylaxis may be particularly useful in patients with a high-risk complete mole when hormonal follow-up is either unavailable or not affordable and chances of losing the woman to follow-up are high. We did not consider chemotherapy in our case in view of no evidence of metastasis and low ß-HCG levels on subsequent follow up. The potential risk of patient being lost to follow up was very much there but did not happen in our case.

CONCLUSION

Chorioadenoma destruen has a potential for myometrial and vascular invasion, leading to uterine perforation and massive hemorrhage. Therefore, to avoid adverse consequences it is necessary to identify such cases by early first trimester ultrasound. Continued reporting of these cases are important so that the obstetricians are aware about the possibility of ruptured invasive mole and it should be kept as a differential diagnosis in all the young pregnant women presents with acute onset lower abdominal pain.

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