OCCURRENCE OF A HYDATIDIFORM MOLE IN TWIN PREGNANCY AND LATER DEVELOPMENT OF CHORION-EPITHELIOMA

(Case Report)

by

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The coexistence of a well-formed foetus with placenta and a hydatidiform mole is an unusual obstetrical finding. On the basis of infrequent occurrence alone such a complication may present a problem in diagnosis and management. It is for this purpose such a case is reported in order to discuss its obstetrical implications. Hydatidiform mole associated with a well-formed foetus, though rare, has been reported by a number of authors like Acosta, Goddard, Herting and Herman, but the occurrence of hydatidiform mole in twin pregnancy followed by development of choriocarcinoma can be considered to be an even rarer condition. The present case, which emphasizes certain interesting features, is presented in order to augment the existing collection of cases of hydatidiform mole occurring in multiple pregnancy.

CASE HISTORY: Patients (S. K.) 28 gravida sixth, admitted on 4-6-1960 to the hospital with history of amenorrhoea 3 months and excessive vomiting for the last six days.

OBSTETRICAL HISTORY:

(1) 11 years-full-term normal delivery twins both female one alive.
(2) 6 months' abortion.
(3) 7 years-full-term normal delivery, died at 2 months.
(4) 5 years-full-term twins, both male, one alive.
(5) 2 years-full-term delivery, female alive.

History of vomiting in the early months was present in all previous pregnancies. On admission patient showed moderate evidence of dehydration. Blood pressure was recorded to be 120/80 and the pulse rate was 88 per minute. Cardiovascular, and respiratory systems showed no abnormality. The uterus was found to be enlarged to sixteen weeks' size. The patient was treated symptomatically and was discharged relieved after five days.

The patient was admitted on 11-6-1960, only two days after discharge from the hospital, again with the same complaints of vomiting. Blood pressure was 140/92. The uterus was enlarged to twenty weeks' size and foetal parts could be palpated. Patient was discharged on 16-6-1960 against medical advise with slight improvement.

The patient was re-admitted on 19-6-1963 for the third time with the same symptoms of vomiting. The blood pressure showed a slight rise of 140/100 but otherwise there were no relevant positive findings. A plain x-ray of the abdomen was taken on 24-6-1960 which revealed the presence of foetal parts. Patient was discharged after the vomiting was controlled on 5-6-1960.

She was again admitted on 29-7-1963 after twenty-four days with a history of amenorrhoea now four and half months and blood-stained discharge for the last

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two days and she gave the history that vomiting was continuing though less.

The uterus was enlarged to twenty-six weeks size and foetal parts were felt easily. Vaginal examination revealed that the cervix was closed and uterus was enlarged to twenty-six weeks size, slight bleeding was present. The patient was discharged against medical advise with slight bleeding still continuing.

The patient was admitted on 16-8-60 for the fifth time as an emergency with profuse bleeding per vaginam. The blood pressure was 160/100, pulse 110 per minute. It was noticed that the 'patient had slight edema on feet and for the first time albumin was detected in the urine. The uterus was thirty weeks in size and was felt to be hard in consistency. A provisional diagnosis of accidental haemorrhage was made. The patient expelled on the same day a live male child about sixteen weeks' gestation with placenta, which was followed by expulsion of a hydatidiform mole. As the mole was not completely expelled and the patient had moderate amount of vaginal bleeding evacuation was done under intravenous penthal anaesthesia and the rest of the mole removed. A transfusion of 400 c.c. of blood was given. Patient continued to have intermittent bouts of vaginal bleeding; a curettage was done on 24-8-1960.

Histopathology Report: Infected decidua present no evidence of hydatidiform mole or chorion-epithelioma detected.

Friedman's test done three weeks after the expulsion of the mole with undiluted urine, the test was positive. X-ray chest was done on 26-8-1963 showed no evidence of secondaries. As the vaginal haemorrhage continued off and on a second curettage was done on 24-10-1960, nearly two months after the expulsion of mole.

Histo-pathological Report: Scanty endometrium with glands in proliferative phase. No evidence of carcinomatous tissue. Vaginal examination under anaesthesia, uterus size about six to eight weeks. Right ovary about 2″ x 2″. No nodules felt or visible.

As the patient continued to bleed in spite of two curettages and in view of her parity it was decided to do a hysterectomy.

On 10-11-1960, nearly three months after the expulsion of the mole, a pan-hysterectomy was performed under spinal anaesthesia.

Specimen: Uterus uniformly enlarged to six weeks' size. A blue nodule about half an inch in diameter present on the anterior surface of uterus near the cornua. Both tubes normal. Both ovaries cystic, size 2″ x 2″. Cut section showed two bluish areas in the myometrium suggestive of chorion-epithelioma.

Histo-pathology Report: Massive invation of myometrium by a mass of Langhans cells with large areas of haemorrhagic necrosis. Nuclei show hyperchromatism. The cells appear to be anaplastic. No villi visible.

DIAGNOSIS: Chorion-epithelioma

X-ray chest, carried out on two separate occasions before and after the operation, revealed no secondary lesions.

Post-operative deep x-ray therapy was given to the pelvis. The patient has been followed up for the last three years, every month, for the first three months, and every three months onward up till now. Friedman test was negative one month after the operation. The patient is well and healthy and there are no evidence of secondaries any where uptil now.

Comment

The etiology of hydatidiform mole is not known, but the primary defect is thought to be in the ovum. In twin pregnancy one embryo may become a hydatidiform mole and the other develop normally. Hydropic swelling of the placental villi may be noted in early stage of pregnancy though most commonly seen in early abortion specimen.

These cases of so-called "partial hydatidiform mole" are reported from time to time but their precise etiologic background is a matter of doubt. Blend has expressed the opinion that the presence of well-formed foetus and well-formed mole in uterus at the same time would seem physically

impossible except in plural pregnancy. Others regard them as a result of single ovum pregnancies in which the hydatidiform process involves only part of the placenta leaving a sufficient number of functioning villi to support foetal life. Still others consider them as aberration in simultaneous fertilization of two ova. Less than a dozen cases have been reported in the entire literature of such type. Smalbrak, in his valuable comprehensive volume on "Trophoblastic Growth", has thrown interesting side-light on this question by reporting two cases of partial hydatidiform mole with foetus. Both foetuses had identical malformation, such as spina bifida, harelip, cleft palate and club foot. Smalbrak, commenting on these malformations, suggests a common damaging agent, affecting the chorionic villi and the growing embryo and considers it to be a viral etiologic factor. This etiology of hydatidiform mole has been suggested by a few students who are considering the problem of greater frequency of hydatidiform mole in Asian population. Except for Smalbrak's two cases, the infants born in association with hydatidiform mole are reported to be normal and occasionally viable, though rarely.

The present case deals with many interesting problems in diagnosis. Hyperemesis graviderum, persistent inspite of active treatment, and pre-eclampsia before 24 weeks of pregnancy, uterine enlargement out of the proportion to the period of gestation, with signs of threatened abortion often associated with hydatidiform mole. Hypertension in these patients appears to be related to an elevated chorionic gonadotrophin levels. Pre-eclampsia has been reported in approximately 35 per cent of the patients with hydatidiform mole often occurring quite early in the course of disease. The patient reported, displayed all the above features. But the presence of foetus demonstrated by x-ray mis-led our original diagnosis of hydatidiform mole. The unduly large uterus was attributed to hydramnios, an error which has been made by others.

A clinical finding frequently associated with hydatidiform mole is a significant elevation or rise in the level of chorionic gonadotrophins. Due to our confusion at finding a foetus within the uterus, a quantitative assay on chorionic gonadotrophins was not performed.

In addition to the clinical and pathological similarities which may exist between a patient such as we have presented and the one with hydatidiform mole, the principles of management are same, including general supportive measures, replacement of excessive blood loss and evacuation and curettage of uterus to ensure complete removal of the diseased tissue and obtain suitable material for pathologic examination. Patient developed chorion-epithelioma, and was treated by surgery, and deep x-ray therapy. A well-supervised follow-up extended over a period of three years and the patient is free from secondaries. Another interesting feature of the case was the repeated history of plural pregnancy.

A search made in literature revealed two cases similar to ours. one reported by Logan (1957) where the patient was followed for five years

without any evidance of malignant transformation. Sitaratna and Sarma (1960) reported twin pregnancy with a well-formed hydatidiform mole associated with viable child of four pounds and a normal placenta. None of the cases reported by these authors showed development of chorion-epithelioma or the history of previous twin pregnancy.

Conclusion

(1) A case is presented as a rare occurrence of a twin pregnancy in which well-formed foetus with placenta occurred in conjunction with a hydatidiform mole, having all the clinical similarities of a hydatidiform mole, and later developing chorion-epithelioma. Patient has had two twin pregnencies prior to present gestation, which occurrence is not reported in the literature.

(2) Stress is laid on diagnosis as the case was misdiagnosed as a case of accidential haemorrhage of toxaemic variety, by the presence of foetus.

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