AN INTERESTING CASE OF CHORIONEPITHELIOMA ASSOCIATED WITH NORMAL PREGNANCY

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Introduction

Chorionepithelioma is a very rare condition, even more uncommon than hydatidiform mole which, as Novak & Novak mention, occurs once in every 2500 pregnancies.

The invasive character of the trophoblast is indeed physiological though in normal pregnancy certain local and systemic defensive mechanisms keep its invasion into the uterine musculature and blood vessels within limits and do not permit the occasional emboli of the trophoblast from gaining a foothold anywhere else in the body. It is indeed surprising why such defence mechanisms break up so rarely that the condition of chorionepithelioma is so uncommon.

The distinguishing feature of chorionepithelioma is the invasion of the uterine musculature as also the blood channels by trophoblastic cells advancing in bulk, with destruction of the uterine tissues accompanied by coagulation necrosis and haemorrhage. Usually, little or no evidence of original villous pattern is detectable histologically though their presence does not exclude chorionepithelioma.

Metastases are common and are quite often responsible for drawing attention of the clinician to the existence of this condition. In the manner of spread to other organs this condition resembles sarcoma rather than carcinoma, in that it is chiefly by vascular invasion. Retrograde transport and occurrence of metastases in the vagina and even vulva are quite frequent. Novak estimates vaginal metastases occurring in about 50% of cases. Novak and Novak report that this condition is preceded by hydatidiform mole in 40% of cases, by abortion in another 40% and by normal pregnancy in 20% of all cases. Payne states that the interval between pregnancy and the disease varies from 6 days to $5\frac{1}{2}$ years and that in 91% of cases it is less than 12 months. Adair mentions that authentic cases have been reported upto 10 years after pregnancy. Hertig finds from study of 200 hydatidiform moles that 2.5% of these are followed by metastasizing chorionepithelioma. The tumour usually arises at the placental site but may also arise from the invasive areas in the uterus or other organs.

Normal pregnancy occurring concomitant with chorionepithelioma is a very rare and interesting condition. It is indeed remarkable that whereas

in the same subject remnants of old trophoblastic tissue turn malignant, yet the trophoblast of the later pregnancy remains within bounds.

Author came across one such interesting case of chorionepithelioma associated with normal pregnancy. It was therefore considered proper to report the case.

On looking up the records of the K. R. Hospital (where author is working) from April 1951 to April 1960, author found reports of 5 cases of diagnosed chorionepithalioma and another 5 cases in which, though the histological report of the curretting was very suggestive of this condition, no definite diagnosis could be done as the cases did not turn up for further investigations.

Out of the 5 cases, three followed hydatidiform mole; one normal pregnancy and one followed abortion. The case reported here had a hydatidiform mole before the epithelioma.

Case Report

The patient, a young Muslim lady, 20 years of age, was admitted to the maternity ward of the K. R. Hospital on 19-9-1958 with three months' amenorrhoea and bleeding per vaginam for 2 months.

Past History. On looking up her old record it was found that in May 1957 she had been admitted to this hospital with 11 weeks' amenorrhoea and bleeding per vaginam for 4 days and had turned out to be a case of hydatidiform mole. The mole was evacuated per vaginam and she was discharged. The patient, however, came back after a month with pain in abdomen and bleeding per vaginam. Currettage showed vesicular mole. She was discharged after currettage. The patient, however did not come for follow-up.

She was now (from 19-9-1958 onwards) treated on the lines of threatened abortion but inspite of all treatment slight bleeding continued. It was therefore considered proper to exclude hydatidiform mole in view of her past history. At 16th week skiagraphy of abdomen was done and it showed foetal shadow. The bleeding stopped and after observing for another 10 days she was discharged.

The patient came back to the hospital on 10th January 1959 with 32 weeks' pregnancy and slight bleeding per vaginam for about an hour. Obstetric examination showed uterus of the size of 32 weeks' pregnancy, foetus presenting by vertex in L.O.A. position and foetal heart sounds could be heard.

Speculum examination revealed no abnormality. Urine was normal.

Clinical diagnosis of minor degree of placenta praevia was made and she was put on conservative treatment. The bleeding per vaginam however continued.

On the 9th day after admission she suddenly started vomiting coffee ground material and developed incontinence of urine and faeces. Neurological examination by the physician showed an upper motor neurone type of hemiplegia (right side). The patient was semi-conscious. Blood pressure and pulse were normal.

Three days after developing hemiplegia she delivered a premature live baby weighing 3 lbs. The child, however, died later. The patient became comatose and the B.P. became 130/90 m.m. Hg. Lumber puncture was done. The C.S. fluid pressure was high but the fluid was otherwise normal. Lungs were found to be clinically normal.

On the 8th day after delivery the patient was transferred to the medical ward.

She was then found to have upper motor neurone type of quadruplegia. Her total W.B.C. count was 12,400 per c.mm.

After a couple of days (i.e. 10th day after delivery), she started getting excessive bleeding per vaginam and was referred to the gynaecologist. Internal examination revealed uterus of the size of 10 weeks' pregnancy. An irregular, purplish growth was detected in the vagina arising from the anterior wall. This growth bled easily on touch. Biopsy of the growth revealed that it was a metastatic growth from chorionepithelioma. Skiagraphy of the chest on 14-2-'59 showed non-homogeneous opacities all over both lungs. Right hilar shadow was enlarged. Trachea was

central. Skiagraphy of the skull showed no abnormality.

On 17th February 1959 she was transferred to gynaecology ward. Total hysterectomy was done under local anaesthesia. At the operation the uterus was found to be of the size of 10 weeks' pregnancy and irregularly enlarged at the fundus. On opening the uterus a purplish irregular growth, about 1″ x 2″, was seen on the left side of the fundus.

Right ovary was cystic and congested. Left ovary was also congested.

Vaginal growth receded after operation but the patient did not regain full conciousness. She ran temperature up to 102°F and gradually deteriorated and finally expired on 2nd May 1959.

Discussion

As already mentioned in the introductory remarks, chrionepithelioma is a rare condition and its association with a normal pregnancy is rarer still. Walthard (1907) reported such a case in which pregnancy was associated with chorionepitheliomatous vaginal nodules and although hysterectomy failed to reveal any chorionepithelioma of the uterus or placenta, the patient died 3 or 4 years later of chorionepitheliomatous metastases in lungs, liver and kidneys. Fickentscher (1941) reported a case of chrionepithelioma associated with pregnancy and cited a few others of a similar type. Cordua (1949) reported one case and so also Mathieu (1939), Dafoe (1939) described a case of chorionepithelioma occurring during an ectopic pregnancy. Mac Rae (1951) described one case of this disease associated with pregnancy. In this case the baby was alive and well a year later though the patient expired.

In Mac Rae's case there was no history of abortion, miscarriage or hydatidiform mole. The patient had had 3 normal pregnancies previously.

In Mac Rae's case the placenta of the child born alive was found to have a small yellowish excrescence on its surface which proved to be the primary chorionepitheliomatous growth. On the placental site in the uterus there was a raised area of about 3 cm. in diameter later shown to contain chorionepitheliomatous cells. In the case reported here, however, the placenta was normal in appearance. Its histopathological examination was, however, not carried out.

In the present case it appears that the chrionepithelioma followed the hydatidiform mole which occurred in 1957 and that the normal pregnancy occurred in the presence of this condition. It is, however, remarkable that the placenta of the foetus did not show any macroscopic evidence of chorinepitheliomatous involvement. Perhaps the normal trophoblast could not break up the local defensive maternal barrier which the hydropic cells of the old hydatidiform mole turned malignant could.

Summary

A case of chorionepithelioma of the uterus with metastatic secondaries in vagina, lungs and central nervous system accompanying a normal pregnancy, has been reported. The patient had had hydatidiform mole about 14 months previous to the pregnancy. The foetus was delivered alive though premature. The foetal placenta showed no macroscopic evidence of chorionepitheliomatous involvement.

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