PRIMARY EXTRAUTERINE CHORIOCARCINOMA

(Two case Reports)

by

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Primary choriocarcinoma of ovary is a rare condition. In an extensive analysis by Hertig and Mansell (1952) the incidence of primary choriocarcinoma of uterus was estimated to be 1:40,000 pregnancies and the chance of choriocarcinoma arising primarily from an ovary was estimated as 1:329 million pregnancies.

On the other hand, choriocarcinoma of the tube is more common and its incidence perhaps would have been higher except that most tubal pregnancies degenerate, die or are surgically removed before the malignant changes of the chorion become manifest (Chatunzew, 1930).

Case report 1: Mrs. R. G., 30 years, was admitted with the complaints of having pain in lower abdomen and irregular, at times profuse, bleeding per vaginam since last three months. Cycles prior to three months were regular.

She had three term normal deliveries, all male children, living; last delivery was 4 years ago. Examination on admission revealed a satisfactory general condition. No lump was palpable per abdomen. On pelvic examination, cervix was hypertrophied and congested. There was slight bleeding through the external os. Uterus was anteverted and anteflexed. Close to the uterus on the right side, a mobile, well defined cystic mass was palpable about the size of a 10 weeks' gravid uterus. On the left side, vague induration of the adenexa was palpable. The patient was examined under anaesthesia and the clinical findings were confirmed. Uterocervical length was 9.5 cms. As the patient was bleeding profusely (like an abortion haemorrhage) a blunt curettage was done and an endometrial polyp removed. Endometrium was reported to be in proliferative phase. As the mass persisted, with the clinical diagnosis of an ovarian tumour, a laparotomy was performed under spinal anaesthesia. Uterus was about 6 weeks' size, soft and very vascular. Left adenexa were apparently normal. Right adenexa were very vascular and fixed. Gentle separation showed the right ovary to be enlarged, 7.5 cms. x 6.5 cms, vascular and adherent to the posterior surface of uterus. There was a dark-coloured cystic swelling which burst during separation. Panhysterectomy was performed thinking the mass to be a malignant ovarian tumour. Postoperatively the patient made an uneventful recovery. Gross features of specimen and the cut surface showed that the right ovary had been replaced by a poorly defined haemorrhagic mass, measuring 5 x 3 x 2 cms. (Fig. 1). Left ovary lodged a lutein cyst. There was no other gross pathology.

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Paper read at the 15th All-India Obstetric and Gynaecological Congress held at Margao-Goa in December 1969.

HISTOLOGY: Right ovary: H & E sections of numerous blocks from the haemorrhagic mass revealed large amount of haemorrhagic and proportionately small amount of tumour tissue. The tumour consisted of small and large sheets of neoplastic cytotrophoblast and syncytiotrophoblast (Fig. 2). In some areas the clumps consisted of anaplastic cytotrophoblastic cells. No chorionic villi were seen. The characteristic feature was presence of tumour emboli in blood vessels (Fig. 3). The poorly formed capsule consisted of ovarian stromal tissue. Left ovary showed a lutein cyst. The other organs had no significant pathology.

Case report 2: Mrs. Z. G., 34 years, was admitted on 18-3-68 as an emergency case with the complaints of pain in abdomen since 3 months, vomiting and a lump in the abdomen since 1 month. Her menstrual cycles were regular; last menstrual period was 9 days prior to admission.

She had 10 term normal deliveries and one abortion after 5 months' amenorrhoea. Last delivery was 3 years ago. She had six living children. Clinical examination revealed an irregular firm mass palpable in hypogastrium corresponding in size to 14-16 weeks' gravid uterus, mobile, extending more to the right side. Internal examination under anaesthesia showed the cervix having a papillary erosion on the posterior lip. A second degree prolapse was present. Uterus was retroverted and bulky. The abdominal mass moved when the cervix was pulled down. A groove was palpable between the mass and the uterus. Routine investigations were within normal limits. With a provisional diagnosis of tubo-ovarian mass, laparotomy was performed under spinal anaesthesia supplemented by general anaesthesia. On opening the abdominal cavity, a dark brown encapsulated mass 12.5 x 10 x 6.5 cms. was seen incorporating the site of the left tube. It was adherent to the omentum and sigmoid colon. A diagnosis of pelvic endometriosis was made. A total hysterectomy with left salpingo-oophorectomy and resection of 17.5 cms. of firmly adherent segment of sigmoid, with end to end anastomosis was performed. Patient was given 300 cc. blood and she made an uneventful recovery.

Pathological examination: Gross feature of specimen showed that left tube was replaced by a tumour measuring 12 x 6 x 4 cms (Fig. 4). The outer surface of tumour was encapsulated, nodular, dark brown in colour. The cut surface revealed areas of haemorrhages and necrosis. There was no evidence of any foetal tissue or hydatidiform mole. The mass from broad ligament and the growth on the sigmoid also exhibited a similar picture. The mucosal surface of sigmoid was not affected.

HISTOLOGY: Tumour replacing left tube and present in the broad ligament consisted of sheet like masses of mononucleated tumour cells. The cytotrophoblast interspersed with many large multinucleated tumour giant cells, the syncytiotrophoblasts (Fig. 5 & Fig. 6). No chorionic villi or decidual reaction were seen. The tumour cells were infiltrating serosal and muscle coat of sigmoid colon. The left ovary lodged a corpus luteum. The remaining specimen did not exhibit any significant pathology.

On receiving this report, skiagram of chest was taken. It was negative for tumour metastases. Patient failed to report for the routine follow-up visit. She was readmitted on 7-12-1968 with acute abdomen and a lump the size of a 20 weeks' gravid uterus, arising from the pelvis. X-ray chest showed evidence of pneumonitis. Immunological test of pregnancy was negative. She received methotrexate, 15 mg. daily, for 9 days and 300 cc. of blood. The mass reduced in size to about 12 weeks' size of gravid uterus. She was readmitted on 30-1-69 for pain in the epigastrium and lump in hypogastrium. Liver was 4 fingers enlarged, firm, nodular and tender. Immunological test of pregnancy was negative, serum billirubin was 10.5 mg.%. Methotrexate was again started but the patient absconded in a moribund condition. No subsequent follow-up was available.

Discussion

Primary choriocarcinomas of the ovary are histogenetically classified into two types: (i) gestational and (ii) non-gestational (Novak, 1967).

According to Oliver and Horne (1948), the primary nature of choriocarcinoma of ovary could be assumed

with certainty in a prepubertal child, postmenopausal woman and in an undoubted virgin during the reproductive life. There is a general agreement that these are teratomatous choriocarcinoma.

Primary gestational choriocarcinoma of the ovary occurs during the reproductive period of a woman's life and its histogenesis is debatable. The condition is so rare that its diagnosis is usually not made till the histopathological report is received and hence chorionic gonadotropin studies have not been made in most of the reported cases.

In our case, curettage revealed the endometrium to be in an early regenerative phase. This would be expected as the patient had been bleeding per vaginam for some time.

The views regarding the origin of primary choriocarcinoma of the ovary are:

1. An ectopic ovarian pregnancy (Sunde, 1921).
2. Trophoblastic emboli which have become malignant (Willis, 1960).
3. Metastasis from a previous uterine or tubal choriocarcinoma in which the primary has been completely expelled, regressed or disappeared (Novak and Koff, 1930).
4. A one-sided development of an ovarian teratoma (Telium, 1946; Novak, 1967).
5. An ab initio malignant transformation of trophoblast without the formation of an embryo (Acosta Sison and Manila, 1955).

In the present case a thorough examination of uterus and fallopian tubes did not reveal any evidence of pregnancy or choriocarcinoma in the uterus. Yet the theoretical possibility of expulsion, regression or disappearance could not be ruled out. Thorough examination of the haemorrhagic mass did not reveal any teratomatous tissue, hence, preceding ectopic pregnancy or ab initio malignant transformation of trophoblast without formation of an embryo appear to be the likely histogenetic evolution. As this tumour occurred in the reproductive period, it would be reasonable to assume it to be a primary gestational choriocarcinoma of the ovary.

Histogenesis of choriocarcinoma of the tube remains unclear. Views put forth (Riggs *et al*, 1964) are:

1. Arises from ectopic pregnancy.
2. Intrauterine pregnancy that has spread to the tube via embolic transport of chorionic villi.
3. Arises from a teratomatous change within the tube.
4. Metastasis from uterine choriocarcinoma with disappearance of primary.

Fimarola (1947) studied 436 cases of ectopic gestation and found no choriocarcinoma. Heiss studied 540 tubal pregnancies and found one case of choriocarcinoma, i.e. 0.18%. In our study of 256 ectopic pregnancies over a period of 10 years, this is the first case, i.e. an incidence of 0.39%. Most patients present symptoms and findings of an ectopic gestation.

Our case is interesting in as much as the presenting clinical symptoms and sign were not suggestive of an ectopic pregnancy.

Summary

Two cases of primary extra uterine choriocarcinoma, one of ovary and one of tube are reported. The inci-

dence and probable histogenesis of these tumours are discussed.

Acknowledgments

We are thankful to the Dean, M.G.M. Medical College, Indore, and the Superintendent, M. Y. Hospital, for their kind permission to avail the library facilities and to use the hospital records for this report.

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See Figs. on Art Paper I