ECTOPIC CHORION-EPITHELIOMA

by

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Chorion-epithelioma is a rare and malignant neoplasm which arises from the epithelium of chorionic villi; hence it may arise at any site at which there are living chorionic cells. This growth may be associated with full-term pregnancy, abortion, extra-uterine pregnancy, hydatidiform mole, teratoma of the ovary and testicle, and it may occur at any site to which chorionic villi have metastases.

The rarity of the occurrence of chorio-carcinoma and the bizarre clinical symptoms with which this condition presents make it an interesting study. The incidence of chorion-epithelioma varies from 1:680 (Acosta-Sison) to 1:50,000. Ectopic chorio-carcinoma is even rarer. The following cases of ectopic chorio-carcinoma came under our observation in February 1963.

Case Report

C. P. D., a Hindu patient, 32 years of age was admitted on 12-2-1963 for:

1. Irregular menstrual cycle with inter-menstrual bleeding — 2 years.
2. Dull dragging pain in right iliac fossa —6 months.

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Patient was well 2 years ago. She had an abortion at 3 months 2 years ago. She started getting irregular menstrual cycles $\frac{(2 — 4 \text{ days})}{6 — 20}$ immediately after the abortion. She also complained of intermenstrual bleeding which lasted for a day or two. Her past menstrual cycle was regular, painless, moderate $\frac{(4 — 5)}{28 — 30}$. Her last menstrual period was 5 days back.

Obstetric History. 4 F.T.N.D. — L., 1 abortion at 3 months 2 years back.

Last delivery — 4 years back.

On general examination, she was thin, fairly nourished with slight anaemia. Blood pressure was 110/70 MM. Hg., no oedema of feet, no lymphadenopathy. Cardio-vascular and respiratory systems were normal.

Per-abdomen: A middle pelvic tumour was felt, size of 16 weeks' pregnancy; it was firm, nodular, irregular. Tenderness in right iliac fossa was present.

Per vaginam: cervix was downwards, backwards and irregular. Uterus: anteverted, anteflexed, 16 weeks' size, firm, nodular. Fornices: left clear, right side, irregular firm non-tender mass 1½" x 1" extending from the uterus and stopping short at the lateral pelvic wall.

Our clinical impression was that of multiple fibroids or endometriosis.

Pre-operative investigations:— HB: 60%.
RBC: 3.25 Mill/cmm.
Urine: nil abnormal.

An **exploratory laparotomy** was done on 13-2-1963 under general anaesthesia. Abdomen was opened by a midline sub-umbilical incision. There was blood in the peritoneal

cavity. An irregular firm haemorrhagic mass, extending from the uterus and stopping short of adnexa was seen on the right side. Right tube and ovary were infiltrated by the mass. Uterus was 12 weeks' size.

Total abdominal hysterectomy with right salpingo-oopherectomy done. Left tube and ovary were not removed as the exact nature of the tumour could not be determined macroscopically and the patient was young. While removing the tumour from its bed there was profuse bleeding. Patient lost 1500 cc of blood within ½ hour. She collapsed for 10 minutes during the operation. Venesection was done. Total 1800 cc of blood given during the operation along with Inj. Methedrine 0.5 mgm and 6 mgm. of nor-adrenaline in the drip. Abdomen was closed in layers.

Post-operative: Patient had a very stormy convalescence. Nor-adrenaline 4 amps in 5% glucose given continuously for 24 hours. It was gradually withdrawn. There was oliguria for some hours post-operatively. The temperature was (102°F) for 4-5 days.

After 24 hours, patient was breathless and had tachycardia with basal crepitations due to pulmonary oedema and acute left ventricular failure.

X-ray chest on 14-3-1963 revealed no pulmonary infarct. No enlargement of the heart. No secondary cannon ball nodules in the lungs were visualised.

On the third day digoxin 0.5 mgm. was given intra-venously.

One more blood transfusion (300 cc.) was given. Patient improved considerably after digoxin. She was put on oral tablets of digoxin II 8 hourly. Injection of atropine 1/200 gr. given for lung congestion.

Patient improved considerably. She continued to get penicillin and streptomycin and later achromycin capsules 6 hourly for 5 days.

On the 7th day patient developed severe thrombophlebitis of the superficial saphenous vein. It was treated with Hirudoid ointment, crepe bandage, complete rest in bed.

Patient also had bilateral acute parotitis. This subsided gradually.

She was discharged on 19-3-63 i.e. 1¼ months later.

Follow-up. Patient developed haemoptysis within 1 week of discharge from hospital and temperature. She was given achromycin intravenously and developed a severe allergic reaction to this. She was hospitalised in skin department and put on cortisone. Gradually she developed cachexia and went downhill. She expired on 27-4-63 of paraplegia. Unfortunately a post-mortem examination could not be obtained.

Investigations

	H. B.	RBC.	Urine.
1. **Post-operative:**	42%	2.12 Mill.cmm.	5-6 pus cells.
2. **AZ test:** on 16-2-1963	— Qualitative:	+ Ve	
	Quantitative:	1 in 100 Weekly + Ve	
3. **Blood urea:**	14 mgm. %.		
NPN:	40 mgm. %.		
4. **X-ray chest:**	— NAD.		

Gross Pathology of Specimen

Hysterectomy with right-sided salpingo-oopherectomy. Uterus measures 9 cms. in length. A large brownish soft friable nectotic mass is seen in the broad ligament and infiltrating the right ovary. It measures 7 x 6 cms. It invades the myometrium from outside. The uterine cavity is smooth and entirely free from the tumour.

Posterior view of the uterus and the right tube with the mass seen (Fig. 1).

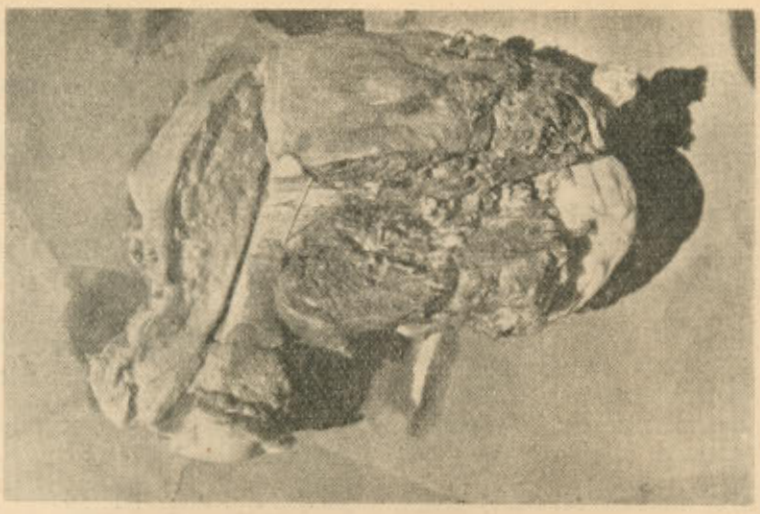

Fig. 1

Microscopic Examination. Specimen is composed of sheets of irregularly oval and polygonal Langhan's cells enmeshed in large areas of haemorrhage and necrosis. The cells show an ill-defined outline and large hyperchromatic pleomorphic nucleii, a few syncitial cells with multiple hyperchromatic nuclei are also seen. The tumour cells are infiltrating the outer walls of the ovary and uterus. The main cavity of the uterus and the musculature of the uterus are unaffected by the tumour as is seen microscopically by serial sections of the uterus (Fig. 2).

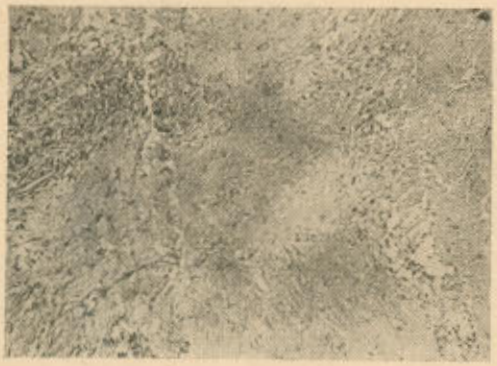

Fig. 2

Comments

There was in our case:

(1) Absence of primary tumour in the uterus. This chorio-epithelioma was seen entirely in the broad ligament and involved the fallopian tube extending medially into the uterus and laterally into the ovarian tissue.

Explanations for absence of the primary tumour are:—

i. Disappearance of primary tumour from the uterine cavity.
ii. Presence of teratomatous tissue in the broad ligament which has developed into a chorion-epithelioma.
iii. Development of chorion-epithelioma from an ectopic pregnancy in the fallopian tube or ovary.

(2) The second feature is that like most chorion-epitheliomas the diagnosis was in doubt till the abdomen was opened. This is often the case,

as chorion-epithelioma is so rare. Our clinical impression was that of multiple fibroids or endometriosis.

(3) The survival of this patient through the stormy post-operative period.

(4) Absence of clinically detectable secondaries.

According to Albert Mathieu ectopic chorio-carcinoma shows a tumour having no direct anatomical relation with the previous placental site. The uterus may be perfectly normal or may show such hyperplasia of the mucosa and musculature as usually accompanies tubal pregnancy. The site of chorion-epithelioma may be in the vagina, broad ligaments or intraperitoneally. The mass resembles a haematoma or collection of thrombi, tumour cells being usually found only in the periphery. The ectopic chorion-epithelioma probably arises from a primary uterine tumour that has regressed or has been expelled with a placenta or mole, or from the transplanted cells of a possibly normal intra-uterine placenta.

Chorion-epithelioma nearly always develops during the reproductive period. The tumour may arise from the ovary, testis or from mediastinum. Bell states that majority of cases develop in the uterus, though few tumours develop outside the uterus in various organs without any primary growth being found in the uterus. Probable types of tumours are:

(1) Teratomas.

(2) Tumours from chorionic ectoderm — in the latter case the tumour is of fetal rather than of maternal origin.

Chorion-epithelioma has been known to occur in the ovary. A few cases have been reported with the origin in the tubes. In rare cases the reproductive organs are free of the tumour the lesion being found in the viscera. This is referred to as an extra-genital chorio-carcinoma. Less than 30 such cases have been reported upto date.

Following are the possibilities of origin of the extra-genital tumours:—

1. Metastatic lesions — the primary having regressed or been expelled. This might be possible if the lesion in the placenta was completely cast off and no uterine growth remained. An unknown factor normally controlling chorionic growth might have caused regression.
2. Malignant transformation of trophoblastic emboli.
3. Teratomatous growth with the survival of only the chorio-epitheliomatous part.

Metastases have been seen in the lungs, vagina, liver, intestines, kidneys, brain and spleen.

Lawrence Berman (1940) has reported 17 cases of extra-genital chorion-epithelioma with multiple metastases. Novak and Koff, in 1930, have stated with regards to chorion-epitheliomas that errors in diagnosis are perhaps more common in this field than in any other in gynaecologic pathology. In 1902, Zagorjarski and Kissel collected 9 cases of chorion-epithelioma where no primary tumour in the uterus was found. In 8 of these, there was presumably a primary tumour in the vaginal walls and in one there were growths in the lungs and brain.

In cases with no definite history of pregnancy one might ask whether the uterus is actually the point of origin of the tumour and whether the distant growths are not of teratomatous nature.

Conclusion

A rare and interesting case of ectopic chorion-epithelioma in the broad ligament is presented where no tumour was found in the uterus. We removed the tumour along with the uterus and adnexa on the right side. Had this patient survived, she would have had Methotrexate therapy. As it was she went downhill and expired within 2 months of operation.

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