PRIMARY OVARIAN PREGNANCY

(With a case report)

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Mercerdus (1614) first described the entity of ovarian pregnancy and probably the first record of an ovarian pregnancy is autopsy report of a case, conducted by Saint Maurice of Perigord, France in 1682. But the entity of this rare type of extrauterine pregnancy was accepted only after Spiegelberg in the year 1878, laid down the following classical criteria, namely:

1. The fallopian tube including the fimbriated end must be intact and must be distinctly separate from the ovary.
2. The gestation sac must occupy the position of the ovary.
3. The gestation sac must be connected to the uterus by the utero-ovarian ligament.
4. Unquestionable ovarian tissue must be demonstrated in the wall of the sac.
5. Well defined chorionic villi must be present in the substance of the ovary.

Norris (1909) insisted that the tubes must not show any evidence of pregnancy. Stander (1941). Baden *et al,* (1952) impressed on the presence of ovarian tissue in the sac wall and also its proper relation with the chorionic tissue.

In 1902 Thomson recorded the first case of ovarian pregnancy in the American literature. Later on cases of ovarian pregnancy have been reported by various authors like, Novak (1940), Hertig (1951), Baden and Heins (1952), Taber and Crosset (1952), King (1954), Upadhyay *et al,* (1955), Dalal (1964), Rakshit (1964), Rama Vaish (1965), Sakuntala Devi *et al* (1967), Rajaram (1967), Roychowdhury (1968), Kalyani Kutty (1969), Chaphekar (1970) and others. Yet the total number of cases reported do not probably exceed 200.

Case Report

Mrs. A.R., aged 25 years, mother of one child was admitted in Eden Hospital, Calcutta Medical College on 3-11-1955 with the complaint of acute lower abdominal pain with fainting attack and a history of nine weeks' amenorrhoea. There was no history of vaginal bleeding and the previous menstrual history was regular.

On examination there was moderate pallor, pulse rate was 120/minute. The temperature and blood pressure were normal. There was tenderness all over the lower abdomen.

Pelvic examination revealed extreme tenderness. The cervix was soft and the uterus slightly bulky and anteverted. There was a tender mass in the left fornix extending to the pouch of Douglas.

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Rectal examination confirmed the same findings.

Investigations

Haemoglobin was 7.2 gms%, WBC 9400/Cmm, polymorphs 76%, lympho 20%, eosino 4%, E.S.R. 20 mm/hr.

Blood group was group B; RhD-positive.

Urine examination showed no abnormality.

After the preliminaries the patient was prepared for operation with the provisional diagnosis of ectopic pregnancy.

First of all examination under anaesthesia was done which corroborated the previous findings.

Next diagnostic puncture of the pouch of Douglas was made which revealed internal haemorrhage.

On laparotomy the abdominal cavity was full of blood and blood clots. It was partly cleared off and the uterus was lifted up by placing the palm behind. The left ovary was found to be the site of sac of an ectopic gestation incorporated in it. The point of rupture could be seen from which blood was trickling. Clinically it appeared to be a case of primary ovarian pregnancy as:

(1) The tubes including the fimbrie were intact on both the sides and the tubes were clearly separated from the ovaries.

(2) The (?) gestation sac was occupying the normal position of the ovary.

(3) The (?) sac was connected with the uterus by the ligament of the ovary.

The tube on the left side was healthy and was distinctly separate from the ovary. Only left sided oophorectomy was performed. The right tube and the ovary were carefully inspected and were found to be healthy. The uterus was just bulky. After final inspection and toiletting the abdomen was closed in layers.

The patient received 1200 c.c. of blood transfusion. Antibiotics and other supportive measures were also instituted. The postoperative recovery was uneventful.

Pathological Examination of the Removed Specimen

Gross appearance: The ovary maintained its oval shape. There was dark brownish projection, like ruptured corpus luteum. The size measured about 1¾" × 1¼" × 1" (Fig. 1a).

Microscopical Examination: Section showed products of conception in ovarian tissue. There was a mass of chorionic villi and decidua like cells embedded in blood clot between the corpus luteum and surface of ovarian tissue (Fig. 2 and 3).

On the second postoperative day the patient passed a complete decidual cast which was also confirmed by histological examination (Fig. 1b and 4).

A biological test for pregnancy was done four days after the operation and it was found to be negative.

A hysterosalpingogram performed three months after the operation, showed patent condition of the tubes (Fig. 5).

The patient could be followed up satisfactorily. The menstrual cycles became normal from the second month postoperatively. The patient had an abortion in March 1957. Then the patient had a full term delivery in 1962. In 1966 she had dilatation and curettage for menorrhagia. Last time when she reported in February 1970, she was well.

Discussion

Primary ovarian pregnancy is not a common condition. Hertig (1951) reported an incidence of 1 in 25,000 to 40,000 pregnancies and 0.7 to 1.07% of all ectopic pregnancies. Boronow *et al*, (1963) in a period of over 19 years at Evanstone Hospital, Chicago found four ovarian pregnancies in 36,914 pregnancies. Dowling *et al*, (1960) reported one ovarian pregnancy in 59,740 pregnancies. In the Eden Hospital, Calcutta, there are more than 10,000 confinements per year. This case of primary ovarian pregnancy which is reported here with a long term follow up was encountered, in 1955. After that in 1963 another case of ovarian pregnancy was met with, as reported by Roychowdhury (1968). It is noticed that during eight years period from 1955 to 1962 only one case was observed when the total pregnancies were 94,782 and total cases

of ectopic pregnancies were 580. The variations of the incidence of this rare condition can be observed from Tables I and II where the reported incidence of ovarian pregnancies among total pregnancies and the reported incidence of

TABLE I

Reported Incidence of Ovarian Pregnancies Among Total Pregnancies

Authors	Year	No. of pregnancies (Intrauterine, ectopic abortion)	No. of ovarian pregnancies	Frequency
Hertig	1951			1:25,000 to 40,000
Bossert & Coworkers*	1955	36,978	1	1:36,978
Bobrow & Winkelstein*	1956	52,833	1	1:52,833
Dowling et al	1960	59,740	1	1:59,740
Boronow et al	1963	36,914	4	1: 9,229
Sakuntala Devi et al	1967	31,512	4	1: 7,878
Present study (Eden Hospital)	1955 to 1962	94,782	1	1:94,782

* As cited by Boronow et al.

TABLE II

Reported Incidence of Ovarian Pregnancies among Ectopic Pregnancies

Authors	Year	Ectopic pregnancies (No.)	Ovarian pregnancies (No.)	Incidence (%)
Eckerson*	1941	339	1	0.30
Courtise*	1942	106	1	0.97
Kuzma*	1944	206	3	1.45
Nucci*	1946	150	1	0.67
Isbell*	1947	110	1	0.91
Manton*	1950	78	4	5.12
Hertig	1951	110	1	0.91
Taber et al*	1952	37	1	2.7
Hoffman*	1952	65	1	1.54
Hayes*	1953	920	2	0.22
Bossert et al*	1955	201	1	0.50
Bobrow et al*	1956	587	1	0.17
Ellis*	1959	85	4	4.71
Dowling et al*	1960	186	1	0.21
Bacile et al*	1961	316	1	0.32
Boronow et al	1963	146	1	0.21
Sakuntala Devi et al	1967	393	4	1.02
Kalyanikutty et al	1969	260	1	0.31
Present study (Eden Hospital)	1955 to 1962	580	1	0.18

* As cited by Boronow et al.

ovarian pregnancies among ectopic pregnancies are shown. Wide variations in the incidence is probably due to the variation of the adaptation of the criteria by various authors also.

The pathogenesis of ovarian pregnancy is poorly understood. The ovum may be fertilized before extrusion from the graffian follicle or fertilization of the ovum outside the ovary with its subsequent nidation on the ovarian substance may take place. Endometriosis and presence of embryonic Mullerian tissue in the ovary has been quoted as fertile soil for reimplantation. Kheng Khoo Tan *et al*, (1968) opined that oophoritis with or without thickened tunica albugenia is a factor in retaining the fertilized ovum in the ovary or corpus luteum. Ovarian pregnancy is classified as:

(i) Primary (true)—where ovarian tissue forms complete layer around the foetal tissue.

(ii) Secondary.

(iii) Combined—where ovary shares in the formation of at least a portion of the tissue lying adjacent to the foetus, tubo-ovarian pregnancy.

Primary ovarian pregnancy may be (A) Intrafollicular—the fertilized ovum develops in the graffian follicle. (B) Extra follicular—fertilized ovum implants and develops in the ovarian tissue other than the graffian follicle.

According to the various types of implantation it may be, (a) juxta-follicular, (b) interstitial, (c) cortical, (d) superficial. Some also call the former varieties as deep.

As stated by Hohne (1923), presence or absence of corpus luteum is determined by the position in which the ovum becomes embedded. In intrafollicular pregnancy the corpus luteum is pressed upon and may completely disappear. Similarly, there may be complete absence of decidual reaction of the stromal tissue of the ovary.

A true intrafollicular type is difficult to prove and the majority are extra follicular in origin. In this case as already described in the histological report there was a mass of chorionic villi and decidua like cells embedded in blood clot between the corpus luteum and the surface of the ovarian substance. So it can be described as primary (true) ovarian pregnancy of the extrafollicular variety.

The signs and symptoms of ovarian pregnancy are similar to ectopic pregnancy. The appearance of the haemorrhagic ovary seen at laparotomy may simulate a picture of a ruptured corpus luteum haematoma but the associated history of amenorrhoea should arouse the suspicion of this rare variety of ectopic pregnancy, to be proved only by subsequent histopathological study. At operation healthy condition of both the tubes must be carefully noted. In this case only oophorectomy was undertaken and subsequently the patent condition of both the tubes could be demonstrated by hysterosalpingogram. The passage of a complete decidual cast on the second postoperative day also indirectly proved that it was a case of ovarian pregnancy before the histopathological report of the sections from the ovary could be received. The nature of the cast passed was also confirmed by histological examination. The fertility in cases of patients after an ovarian (ectopic) pregnancy is deemed to be subnormal. This patient had one term normal delivery three and half years before this ovarian pregnancy. The patient could be followed up thoroughly. Subsequently she had one abortion within 1½ years after the operation and another term normal delivery 5 years after the

abortion. This patient had no evidence of pelvic inflammation or endometriosis. She had some functional menorrhagia diagnosed and probably relieved by curettage. She is reported to be doing fine with two healthy babies even after a long follow-up of over fifteen years.

Summary

1. A case of primary ovarian pregnancy is presented.

2. The case was primarily diagnosed as a case of disturbed ectopic (tubal) pregnancy.

3. The diagnosis was apparent only after laparotomy, which was confirmed by subsequent histopathological examination.

4. Passage of a decidual cast in the early postoperative period was also an important evidence pointing towards the diagnosis of ectopic ovarian pregnancy.

5. The healthy condition of the tubes pointed to the primary nature of the lesion. Oophorectomy alone wth conservation of both the tubes was performed. Subsequent hysterosalpingogram confirmed patent condition of both the tubes.

6. A follow up of more than 15 years helped us to study the subsequent fertility and other features.

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See Figs. on Art Paper IV-V