OVARIAN PREGNANCY

by

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Ovarian pregnancy is a rarity. The first case was reported in the 17th century. Later, in 1878, Spiegelberg, as quoted by Eastman laid down his classical criteria, for diagnosis of ovarian pregnancy, namely (1) that the tube on the affected side be intact, (2) that the foetal sac occupies the position of the ovary, (3) that it should be connected with the uterus by the ovarian ligament, (4) that definite ovarian tissue be found in its wall.

In 1902 Thomson recorded the first instance of ovarian pregnancy in the American literature. Since then many cases of ovarian pregnancy have been reported in the literature by various authors like Novak (1940), Hertig (1951), Baden & Heins (1952), Taber and Crossett (1952), King (1954), Upadhyay *et al* (1958), Subhadradevi (1960), Barberton (1963), Rakshit (1964), Rama Vaish (1965). At least 150 cases have been documented in the literature so far.

Incidence

Hertig (1951) reports an incidence of 1 in 25,000 to 40,000 pregnancies and 0.7 to 1.07% of all ectopic pregnancies. At Evanston Hospital, Chicago, over a period of 19 years ending January 1963, there were 4 ovarian pregnancies in 36,914 pregnancies, giving an incidence of 1 in 9,229 pregnancies and 4 cases in 146 ectopic pregnancies, giving an incidence of 2.74% as reported by Boronow *et al.* Dowling, Collier and Bretschneider (1960) reported one ovarian pregnancy in 59,740 pregnancies. During a survey of 10 years, from June 1956 to June 1966 at the Government General Hospital, Guntur, there were 4 cases of ovarian pregnancy in 31,512 pregnancies, giving an incidence of 1 in 7,878 pregnancies and 4 cases in 393 ectopic pregnancies, giving an incidence of 1.02% (Table I & II). The incidence given by various authors varies from 0.17% to 4.71% of all ectopic pregnancies. The wide variation in the incidence is probably due to the variation of the criteria laid by Spiegelberg and the adaptation of the same by various authors.

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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 and co-workers* ..	1955	36,978	1	1 : 36,978
Bobrow and Winkelstein* ..	1956	52,833	1	1 : 52,833
Dowling, Collier and Bretschneider	1960	59,740	1	1 : 59,740
Boronow, *et al*	1963	36,914	4	1 : 9,229
Govt. General Hospital series, Guntur	1966	31,512	4	1 : 7,878

* 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
Courtiss*	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 and Crossett*	1952	37	1	2.7
Hofman*	1952	65	1	1.54
Hayes*	1953	920	2	0.22
Bossert and associates*	1955	201	1	0.50
Bobrow and Winkelstein* ..	1956	587	1	0.17
Bercovici, Pfau and Liban* ..	1958	94	4	4.25
Ellis*	1959	85	4	4.71
Dowling, Collier and Bretschneider*	1960	186	1	0.21
Bacile and Nagler*	1961	316	1	0.32
Boronow, *et al*	1963	146	4	2.74
Govt. General Hospital series, Guntur	1966	393	4	1.02

* As cited by Boronow *et al.*

The original Spiegelberg criteria had no changes for 25 years, until in 1903, Williams in his early edition of his classic text book modified the 4th postulate, that ovarian tissue must be found in the wall of the sac in several places and at some distance from each other. The same was emphasized by Stander (1941). This was further modified by Baden and Heins (1942) that ovarian tissue should be present intervening between the foetal tissue and any adherant extraneous tissue, which will be useful in dealing with advanced pregnancy. In 1909, Norris modified the 1st postulate stating that the tube should be both macroscopically and microscopically normal and later Rubin emphasised removal of the tube in every case for study of the same.

The four cases which are reported below satisfied all the above postulates.

Case 1.

Mrs. K. K. aged 27 years, was admitted in the Maternity ward, Government General Hospital, Guntur, on 18.2.1959 at 1.25 P.M. with the history of 45 days' amenorrhoea and bleeding per vaginam since one week. Patient had a sudden attack of acute pain in the lower abdomen associated with vomiting and giddiness, 16 hours prior to admission. She had three such attacks before the pain became generalized.

Gynaecological history: Her periods were regular and painless. Since one year she was having painful periods occurring once in 40 to 45 days, flow lasting for 4 to 9 days, and last menstrual period was 45 days ago. Patient attained menarche at 12th year. Her married life was 10 years. Had one child, 9 years ago.

General condition on admission: Patient was grossly anaemic and restless. Tongue was moist and pale. Temp. 99.6°F., pulse 140/min. volume and tension fair. B.P. 120/80. Haemoglobin 30%. Urine: nil abnormal. Systemic examination showed no abnormality.

Per abdomen: Guarding of lower abdomen and shifting dullness was present. No masses were made out.

Pelvic examination: External genitalia were healthy. Cervix was pointing downwards and forwards, uterus was retroverted and retroflexed and of normal size. No masses were felt in the fornices. Slight fullness of right fornix was present. Speculum examination revealed a healthy cervix. Colpocentesis was done and old blood drawn. Pre-operative diagnosis of ruptured ectopic was made and laparotomy was done under general anaesthesia. On opening the peritoneal cavity free blood welled out and about 20 ozs. of blood clots were found in the pouch of Douglas. Uterus was found to be of normal size. Right adnexae and left tube were healthy. Left ovary was enlarged, congested and found to be the seat of ruptured gestation. A rent in the posterior aspect of the ovary was seen lined by amniotic membrane and bleeding was present at the hilar region. (Fig. 1). Left salpingo-oophorectomy was done. Corpus luteum was found in the left ovary. Postoperative period was uneventful. Patient was discharged well on 9th post-operative day. Biopsy report 589 to 592/59. Left tube: normal. Ovary: shows trophoblastic cell collections and syncytial masses amidst the ovarian stroma. They show degenerative changes and some are found in the vessels. Massive haemorrhage and necrosis present.

Case 2.

Mrs. V. S. aged 21 years, was admitted in the surgical wards of Government General Hospital, Guntur, on 21.10.59 with history of colicky attacks of pain in the right iliac fossa of 2 days duration with no history of amenorrhoea. Pain was not associated with vomiting or fever. Micturition and defalcation normal.

Gynaecological history: She attained menarche at 12th year. Menstrual periods were regular, painless, occurring once in 29 days and flow lasted for 5 days. Last menstrual period was five days ago. She had been married for 7 years. Had 2 children.

Last child was aged one year. No history of abortions.

General condition on admission: Moderately nourished individual, not anaemic. Pulse 116/min. Volume and tension good. Respiration 22/min. B.P. 110/70. W.B.C. 12,000/c.m.m. Urine: nil abnormal.

Per abdomen: There was fullness of lower abdomen and tenderness over Mc Burney's point with guarding in the right iliac fossa. No visible peristalsis was present.

Pelvic examination: External genitalia healthy. Cervix was pointing downwards and backwards. Uterus was of normal size, being anteverted. There was tenderness in the right fornix. No masses felt in the fornices. Movement of cervix was not painful.

In view of the above findings pre-operative diagnosis of acute appendicitis was made and laparotomy was done under spinal anaesthesia. On opening the peritoneal cavity there was haemoperitoneum with about 20 ozs. of frank fresh blood and clots. Exploration revealed normal viscera except for the right ovary which was congested and showed an area of rupture about the size of a pea. Blood was seen oozing from the area of rupture. Right tube was normal. Adnexae on the left side healthy. Uterus was anteverted and normal in size. A diagnosis of ruptured ovarian pregnancy was made and right salpingo-oophorectomy was done. Patient was discharged well on the 10th post-operative day.

Biopsy report: 3364 to 3366/59. Ovary shows corpus luteum of pregnancy with products of conception. (Fig. 2) Ovarian stroma oedematous. Right tube shows evidence of interstitial salpingitis.

Case 3.

Mrs. E. aged 30 years, was admitted in the maternity wards, Government General Hospital, Guntur, on 10.7.62 with history of 6 months' amenorrhoea and not feeling foetal movements since 10 days. She attended the antenatal outpatient clinic four days ago, with the same complaint. Examination then revealed an intra-abdominal well-circumscribed ovoid tumour in the right lumbar region measuring 6"/5" firm in consistency. No shifting dullness was elicited. Pelvic examination revealed a normal sized anteverted, mobile uterus with the tumour too high to be felt through the fornices. Speculum examination revealed a healthy cervix She was then referred to the surgical outpatient clinic with the provisional diagnosis of hydronephrosis. The surgeon, suspecting it to be a hydronephrosis, had an intra-venous pyelogram done, which revealed normal functioning kidneys with a foetal skeleton in the region of the tumour. So the case was referred back to antenatal clinic. On further probing into the history, the following salient features were elicited which were missed before, in the busy outpatient clinic. During the 3rd month of amenorrhoea, she had acute pain in the abdomen, associated with vomiting, slight vaginal bleeding for which she sought advice and was diagnosed as pregnancy by a local doctor. After 2 months she developed distension of abdomen and recurrence of vomiting and there was cessation of foetal movements, for which she was admitted in a mission hospital. She was told by the doctor that she was not pregnant and was discharged from the hospital, after which she sought admission here.

Gynaecological history: Patient attained menarche at the 14th year. Periods were regular, occurring once in 30 days' moderate, painless, flow lasting for 4 to 5 days. Her last menstrual period was six months ago. She had been married for 15 years. She had five children. All were full term normal deliveries and all alive. The age of the last child was three years.

General condition: Moderately nourished individual. Not anaemic. Temp. 98.4°F., pulse 80/min. B.P. 100/70 mm. Hg. Urine: nil abnormal. Systemic examination showed no abnormality.

Abdominal, bimanual and speculum examination findings were same as in the outpatient clinic.

Plain x-ray abdomen: Foetal skeleton about 6 months' size visualized on the right side.

I. V. pyelogram: Both kidneys functioning well. Right ureter dilated.

Hysterosalpingogram: Uterus and tubes

normal. Foetus was seen above the pelvis on the right side.

In view of an acute episode of pain and vomiting at the 3rd month, eccentric position of the foetus with a normal-sized uterus and absence of foetal movements, a diagnosis of secondary abdominal pregnancy was made and laparotomy was done on 16.7.1962 under general anaesthesia. On opening the peritoneal cavity and releasing few flimsy adhesions of omentum to the parieties, an oval mass about 6″/5″ well circumscribed came into view. Further exploration revealed it to be a gestation sac containing a 6 months' foetus. Uterus was found to be of normal size, both tubes healthy, long and tortuous. Left ovary normal. Right tube was adherant to gestation sac by flimsy adhesions which were released. The gestation sac was attached to the uterus by the ovarian ligament and occupied the site of the ovary. Flimsy adhesions of omentum were released (Fig. 3). On close inspection, ovarian tissue was found incorporated in the gestation sac. As the patient was not anxious to have more children bilateral salpingectomy and excision of the gestation sac was done. No evidence of corpus luteum was present on the left side. Post-operative period was uneventful. Patient was discharged well on the 13th post-operative day.

Biopsy report: 2356/62 organising products of conception and multiple foci of calcification seen in ovarian stroma, with haemorrhage and necrosis of vessels.

Case 4.

Mrs. A. aged 30 years, was admitted on 7.4.66 at 3 P.M. in the maternity wards of Government General Hospital, Guntur, with the complaint of 5 weeks' amenorrhoea, and acute colicky pain on the left side of lower abdomen from 19 hours prior to admission associated with fainting attacks and vomiting. Patient complained of difficulty in passing urine.

Gynaecological history: Attained menarche at 15th year. Periods were regular occurring once in 30 days' moderate, painless, flow lasting for 4 days. Last menstrual period was 37 days back. She had been married for 12 years. Had three children, all alive. The age of last child was five years.

General condition: Moderately nourished individual. Very anaemic Temp. 100.4°F., pulse 130/min. volume and tension fair. B.P. 100/70 H.B. 30%. Urine clear. Systemic examination showed no abnormality.

Per abdomen: Lower abdomen was rigid and tender to palpation and shifting dullness was present.

Pelvic examination: External genitalia were healthy, cervix was found to be pointing downwards and backwards. Uterus was found to be anteverted, slightly bulky. Extreme tenderness in both lateral fornices was present but more on the left side. Boggy mass was felt in the posterior fornix. Movements of cervix were painful. Colpocentesis revealed old fluid blood with small clots.

Pre-operative diagnosis of left tubal rupture was made and laparotomy was done under general anaesthesia. On opening the peritoneal cavity dark coloured fluid blood welled out. Plenty of clots removed from the pouch of Douglas. Uterus was found to be anteverted and bulky. Right adnexae healthy. On the left side, tube was found to be intact and healthy. Left ovary found to be enlarged to the size of 2″/1″, congested, with a ruptured sac lined by amniotic membrane on the inferior aspect. As the patient was not anxious to have any more children salpingo-oophorectomy on the left side and salpingectomy on the right side were done. A bunch of chorionic villi were recovered later from the blood clots removed from the pouch of Douglas. Post-operative period was uneventful. Patient was discharged well on 10th post-operative day.

Biopsy report: 1851/66. Tube: normal, Ovary: corpus luteum with haemorrhage and chorionic villi.

Discussion

The classification of ovarian pregnancy according to Baden and Heins is as follows:

I. Primary ovarian pregnancy:
   a. Intrafollicular.

b. Extrafollicular which includes juxta-follicular, interstitial, cortical and superficial ones.

II. Combined ovarian pregnancy, where ovary forms at least a portion of the tissue, lying adjacent to foetal tissues but not forming the entire wall.

In the intrafollicular type ovum gets fertilized in the follicle itself, whereas in the extra-follicular type the fertilized ovum gets implanted on to the surface of the ovary. A true intrafollicular type is difficult to prove and majority are extra-follicular in origin. Exact method of implantation was not established in our cases and this concurs with the findings of Berndt Johan and Vesanto (1966).

The average age of patients in this series was 27 years. Though it is stated that ovarian pregnancy is associated with primigravid state (27%), all the cases in this series occurred multiparous women. No history of ious operations nor evidence of metriosis could be elicited in any ese cases.

or ovarian implantation, obstruct- vulation has been proposed as one he causes. Pelvic inflammatory ase has been incriminated in 7% of these cases. In this series ere was only one case which showed evidence of chronic salpingitis.

The other causes are: tenacious granulosa cells and discus proligerous, low intrafollicular pressure, ineffective tubal function (ciliary and/or peristaltic), favourable surface phenomena, parthenogenesis and chance.

Ectopic endometrium is very rarely associated with ovarian pregnancy. Only 2 cases associated with ovarian endometriosis were encountered by Baden and Heins (1952) among 97 cases collected from literature. Animal work has shown that neither decidua nor endometrium are necessary for trophoblastic proliferation and implantation.

The signs and symptoms in our four cases were: 1. Variable period of amenorrhoea, except in one case. 2. Colicky pain in the abdomen associated with giddiness, guarding and rigidity of lower abdomen in three cases. 3. Palpable tumor mass in one case. 4. Vaginal bleeding and a positive colpocentesis in two cases which terminated in the early weeks of gestation. The pre-operative diagnosis of ovarian pregnancy was not made, as two were diagnosed as ruptured ectopic gestation, one as acute appendicitis and another as secondary abdominal pregnancy. This is in keeping with that found, in literature. Usually it is mistaken for a disturbed ectopic gestation if it occurs in the early months or a secondary abdominal pregnancy, when it occurs in the later months. Sometimes it may be even mistaken for multiple fibriods as in the case reported by Subhadra Devi (1960). As it presents diagnostic difficulties, it is hardly ever diagnosed pre-operatively and is always a laparotomy diagnosis.

According to Baden and Heins (1952) 75% terminate in the first trimestser, 12½% in the second trimester and 12½% in the third trimester. In the four cases reported above, the first and the fourth case terminated in the early weeks of gestation, one in the 5th week and the other in the 6th week while the third case reached 6 months' gestation. Cases have been

recorded in the literature where they go to term (Hubacker, 1963) or even after, which are usually operated on as secondary abdominal pregnancy, as in our third case. Most of the infants are still-born. Cases have also been recorded where there was simultaneous intra-uterine and ovarian pregnancy, twin ovarian pregnancy (Green and West, 1963) recurrent ovarian pregnancy and ovarian hydatidiform mole (Wittenberg and Ries (1948).

In all the four cases, tubes were healthy both macroscopically and microscopically except in one case, where there was evidence of chronic salpingitis with the ovary being the seat of gestation. Corpus luteum was found in all the four cases on the same side. Some of the authors believe that corpus luteum may be absent in the intra-follicular type.

The usual operation practised is removal of both tube and ovary on the affected side. When possible, conservatism is suggested, the tube on the affected side being preserved if grossly normal and wedge resection of the affected ovary done if possible.

All the four cases in this series were managed by laparotomy and salpingo-oophorectomy on the affected side. Two of the patients were sterilized by doing salpingectomy on the opposite side as the patients desired it.

Summary

1. Four case notes of proved ovarian pregnancy both macrocopically and microscopically have been recorded.

2. The incidence and diagnostic criteria have been discussed in detail.

3. The pre-operative diagnosis of all four cases was incorrect, thereby proving that ovarian pregnancy is a laparotomy diagnosis.

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Figs. on Art Paper III