

PRIMARY OVARIAN PREGNANCY

(A Case Report)

by

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and

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The occurrence of primary ovarian pregnancy is a relatively uncommon phenomenon. The first authentic case of ovarian pregnancy was reported as early as 1614 by Mercurdus. Following this, in 1682, St. Maurice reported the post-mortem findings of a case of ovarian implantation. The entity came to be fully accepted in 1878 when Spiegelberg laid down definite criteria which are essential for the diagnosis of this condition. The criteria proposed by him were:

1. The tube including the fimbriated end must be intact and must be distinctly separate from the ovary.
2. The gestational sac must definitely occupy the position of the ovary.
3. The gestational sac must be connected to the uterus by the utero-ovarian ligament.
4. Unquestionable ovarian tissue

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must be demonstrated in the walls of the sac.

Morris (1909) insisted that the tube should not show any evidence of pregnancy. Stander *et al* (1941) modified the criteria by requiring ovarian tissue to be present at several places at some distance from each other in the wall of the sac. Baden *et al* (1952) consider that ovarian tissue be found interposed between extraneous adherent tissue and the foetal tissue without directly coming into contact with the foetal tissue.

Incidence

Wide variation has been observed in the incidence reported by various authors. Hertig (1951) calculated the incidence as 1 in 25,000 to 40,000 pregnancies, and 1 in 110 ectopic pregnancies. Bossert *et al.* (1951) reported one case among 36,978 pregnancies. Barrow and Winkelstein found one case among 52,833 pregnancies. Dowling *et al.* (1960) also give more or less the same figures. Boronow *et al.* (1955) reported four cases among 36,914 pregnancies, giving an incidence rate of 1 in 9,229 pregnancies. Sakuntala Devi *et al.* (1967) also reported four cases

among 31,512 pregnancies, an incidence of 1 in 7,878 pregnancies. In the present study only one case was observed during a period of five years from January 1963 to December 1967 among 27,385 pregnancies.

Among ectopic pregnancies ovarian pregnancy is the least common and different figures have been given by different authors for the frequency of this type of pregnancy among ectopic pregnancies. The figures vary from 0.22 to 5.12% of all ectopic pregnancies. This may probably be due to the variation in the criteria adopted by different authors for designating particular cases to one or the other group. Table I shows the figures given by the various authors.

Barrow and Winkelstein (1956) consider that ovarian implantation is not as rare as would be seen from the relatively small number of cases, as the majority of cases are either overlooked or unreported. Many of the cases which appear to be tubal pregnancies occurring in the close vicinity

of the ovary, on careful examination and histopathological studies may prove to be primary ovarian pregnancies.

Wittenberg and Ries (1948) have classified ovarian pregnancy as follows:—

1. Primary.
 - a. Deep
 - i. Intrafollicular
 - ii. Interstitial or juxta-follicular.
 - b. Superficial
 - i. True superficial.
 - ii. Supra-follicular.
2. Secondary
3. Combined.

In the intrafollicular type the ovum gets fertilised in the follicle itself, whereas in the extrafollicular (i.e. interstitial and superficial types of Wittenberg) the fertilised ovum implants itself in the superficial layer of the ovary or over its surface. It is very difficult to establish the type of pregnancy in most of the cases, especially where the pregnancy is an advanced one and the anatomical rela-

TABLE I

Authors	Year	No. of ectopic pregnancies	Ovarian pregnancies	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	1952	37	1	2.70
Hoffman*	1953	65	1	1.54
Henges*	1955	920	2	0.22
Bossert*	1956	201	1	0.50
Barrow	1959	587	1	0.22
Ellis*	1959	85	4	4.71
Dowling	1960	186	1	0.21
Bacile*	1961	316	1	0.32
Boronow	1965	146	4	2.71
Sakuntala Devi	1967	393	4	1.02
Present Study	1967	260	1	0.31

(*) As cited by Boronow (1965).

tions are distorted due to the pregnancy. Most of the ovarian pregnancies terminate in the first trimester. According to Baden and Heins 75% terminate in the first trimester, 12.5% in the second and 12.5% in the third trimester. According to Guxio *et al.* 81.3% of cases ended in the first trimester and 5% in the second trimester. Advanced primary ovarian pregnancy has been reported from India by Upadhyay (1955), Subhadra Devi (1960), Rakshit (1964) and Vaish (1965). Cases reported by King (1954), Upadhyay *et al.* (1955), Rakshit (1964) and Vaish (1965) had gone up to full term before termination. Modavi (1952) reported a case of twin ovarian pregnancy of approximately 12 weeks' duration.

Case Report

Mrs. S., a ninth gravida, aged 38 years, was admitted in the S.A.T. Hospital, Trivandrum, on 10-7-1967 with a history of amenorrhoea of 7 months' duration. Three months prior to admission, she had had an attack of acute abdominal pain accompanied by vomiting and slight vaginal bleeding lasting for five days which subsided with conservative management in a peripheral hospital. A week before admission she developed pain in the abdomen and a sero-sanguinous vaginal discharge.

She had a normal menstrual history. She was married at the age of 18 years and had eight full term normal deliveries and all the eight children are alive and healthy. Eight years ago she was operated for ectopic pregnancy on the right side in another hospital.

Examination on admission revealed an undernourished, moderately built, anaemic woman, with a blood pressure of 110/80 mm. of Hg. The temperature was normal. Abdominal palpation revealed a smooth, spherical mass of approximately the size

of a uterus of 28 weeks gestation which was mobile from side to side, but not in other directions. External ballotment was positive and foetal parts were made out. Foetal heart sounds were not audible. No free fluid was detected. Vaginal examination showed a normal vulva and vagina, the cervix was healthy, the uterus was anteverted and slightly bulky. The mass in the abdomen was apparently separate from the uterus and situated more anteriorly and to the right. Rectal examination revealed no additional information. A provisional diagnosis of secondary abdominal pregnancy was made.

Investigations

Her haemoglobin was 7.2 Gm%. RBC's-3 million cmm. Routine urine examination was negative for albumin and sugar and microscopy of the deposit revealed nothing abnormal. Her blood group was "O".

Straight films of the abdomen revealed a foetus situated in an abnormally high position, in an oblique lie, with the head towards the left side. Spalding's sign was positive and the ribs were crumpled together. Plain x-ray of the abdomen with a sound in utero revealed the same foetal shadow high up in the abdomen. The uterine cavity measured 4" in length. Lateral pictures revealed similar appearances.

A diagnosis of advanced extra-uterine pregnancy was made and a laparotomy was done on the 5th hospital day. The mass was found to occupy the right lower quadrant and was surprisingly mobile except for flimsy adhesions and measured 25 cms x 25 cms. It was attached to the lateral wall of the pelvis by the infundibulo-pelvic ligament and some hypertrophied ovarian blood vessels. On the medial side the mass was connected to the uterus by the ovarian ligament and some blood vessels. The fimbrial end and the ampullary portion of the right tube were absent, having been removed at the previous operation. The right ovary was about 3.5 cms long and was found closely applied as a thin mass over the sac on the upper and anterior surface. The left tube and the ovary were normal.

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stump of the right tube were removed en masse and the stumps were transfixed and ligated as usual. Sterilisation was done by a left salpingectomy and the abdomen closed in layers. The patient had an uneventful post-operative period and was discharged on the 10th post-operative day.

Specimen

On opening the sac a small quantity of inspissated liquor came out. The foetus was macerated and about 28 weeks size with no detectable deformity. The placenta was 16 cms. in diameter and attached to the inner wall of the sac anteriorly and showed infarction at two places.

Pathological examination

Sections from four different sites of the wall were taken. All of them revealed ovarian stroma compressed due to the pressure of the sac. Placental chorionic villi were demonstrated in two sections. One section revealed a small follicular cyst and a Graafian follicle. (See figures).

Discussion

The case reported above satisfies all the criteria laid down by Spiegelberg (1878), The right tube was absent, having been removed at a previous operation for ectopic pregnancy on that side. The ovary had undergone pressure atrophy, and unquestionable ovarian tissue and placental chorionic villi were demonstrated at different places in the walls of the sac.

The mechanism of ovarian pregnancy is not clearly understood. According to Leopold, primary ovarian pregnancy results from fertilisation of the ovum before it escapes from the follicle. The rarity of such a phenomenon is explained by the fact that the ovum requires a degree of maturation before which it cannot be fertilised, by which time it automatically finds itself in the tube. Rock and Hertig (1944), however, state that intra-

follicular eggs may occasionally undergo meiosis and become mature enough for fertilisation. Anything which interferes with the expulsion of the ovum from the ruptured follicle may also permit a sufficient passage of time to allow the ovum to mature in the ovary itself and becoming capable of being fertilised.

In the present case the removal of the tube and the closure of the exits may probably explain the occurrence of primary ovarian pregnancy. No thickening of the tunica or evidence of inflammation of the ovary was found on pathological examination.

Summary

A case of advanced primary ovarian pregnancy has been reported. The patient had undergone salpingectomy for a previous ectopic pregnancy which may probably explain the intraovarian implantation in the present pregnancy. The relevant literature is reviewed.

References

1. Baden, W. F. & Heins, O. H.: *Am. J. Obst. & Gynec.* 64: 353, 1952.
2. Bhaggamma, Purshotham: *J. Obst. & Gynec. India.* 14: 4, 1944.
3. Borrow, C. R., Mcelin, W. T., West, H. R. and Bukingham, C. J.: *Am. J. Obst. & Gynec.* 91: 1095, 1965.
4. Bossert, L. J., Hadon, G. B., Coitti, R. and Tiosco, E. L.: *Am. J. Obst. & Gynec.*, 62: 920, 1951.
5. Courtise, M.: *Am. J. Obst. & Gynec.* 44: 128, 1942.
6. Dowling, E. A., Collier, F. C. and Bretschneider, A.: *Obst. and Gynec. Abst.* 15: 58, 1960.
7. Eckerson, E. B.: *Am. J. Surg.* 54: 487, 1941.

8. Hertig, A. T.: Am. J. Obst. and Gynec. 62: 920, 1951.
9. King, G.: Am. J. Obst. & Gynec. 67: 712, 1954.
10. Kuzma, J. F. and Lillie, O. R.: Am. J. Obst. & Gynec. 48: 268, 1944.
11. Leopold: Arch. Gynaec. 58: 525, 1899.
12. Modavi, O.: J. Obst. and Gynec. Brit. Comm. 70: 743, 1963.
13. Morris, C. C.: Surg. Gynec. and Obst., 9: 123, 1909.
14. Peel, J., Quoted by Bourne A. (ed) British Obst. and Gynec. Practice, London, Hennmann, p. 433.
15. Rajaram, P.: J. Obst. and Gynec. India, 17: 5, 1967.
16. Rakshit, B.: J. Obst. and Gynec. India 12: 851, 1964.
17. Rock and Hertig: Am. J. Obst. and Gynec. 23: 262, 1932.
18. Sakuntala Devi: J. Obst. and Gynec. India. 17: 3, 1967.
19. Spiegelberg: Ark. F. Gynec. 13: 73, 1878.
20. Stander, H. T.: William's Obstetrics ed. 8, 1941, N. V. Appleton Century Co. Inc.
21. Taber, R. E. L. and Crosset E. S., Am. J. Surg. 83: 41, 1952.
22. Upadhyay, S. N., Bhattacharya G. R. and Prakash, B. J.: J. Obst. and Gynec. India, 6: 76, 1955.
23. Vaish, Rama: J. Obst. and Gynec. India. 15: 417, 1965.
24. Wittenberg, S. S. and Ries, R. G.: Am. J. Surg. 75: 618, 1848.
25. Wollner, A.: Am. J. Obst. and Gynec. 23: 262, 1932.

Figs. on Art Paper V