

PRIMARY OVARIAN PREGNANCY

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This paper records an uncommon variety of ectopic pregnancy that almost invariably presents diagnostic uncertainty, and in the present case doubt existed even at operation.

CASE REPORT

The patient, Mrs. N.A., aged 42 years, was first seen 3½ weeks after onset of symptoms. She had experienced initially a sharp pain across the lower abdomen on 20 January 1966. This continued as a fairly severe pain for 2 days, then gradually eased off—she had relief from analgesics given by her own doctor, who had also administered antibiotics. Thereafter she had intermittent sharp pains with a dull ache in between.

Two days after pain had commenced she had vaginal bleeding, which then persisted as a dark slimy loss for nearly 3 weeks. The loss was not heavy, no clots were passed and it was followed by a brown discharge, with an occasional further slight bleeding.

She had not missed a period and her cycle was 2-3/26. Her last menstrual period was at the end of December 1965. The loss was normal and usually she had backache and numbness in her legs.

Dyspareunia had been present for a long time and she volunteered that decreased libido had been present for about 6 months.

She had 5 children, all full-term normal deliveries and the youngest was 15 years. She had 6 miscarriages at various intervals, and a D&C after her last miscarriage 5 years ago.

On Examination

Weight 110 lb. Hb. 12.5 G/100 ml. The abdomen was tender in the left iliac fossa low down, but no masses

were felt.

On pelvic examination the vulva and vagina were normal. There was a slight dark blood loss from the external os. Cervix multiparous, slightly bulky, with several follicles. A Papanicolaou smear was taken, which showed grade 1, normal cells. Uterus anteverted, bulky and mobile, but tender on movement. A tender thickening was present in the left appendage—this was thought to be tubo-ovarian and inflammatory in origin. Slight tenderness was present in the right appendage.

Diagnosis was thought to be salpingo-oophoritis causing prolonged menses. A broad-spectrum antibiotic was commenced. However, in view of her age and persistence of the bleeding it was decided to perform a curettage.

At curettage the uterus was anteverted, mobile, and 3¼ in. in depth. The right appendage was thickened and the left ovary palpable. The cervix was dilated and scanty endometrium curetted. Histology report: 'Sections of the curettings show non-secretory endometrium. There is no endometritis.'

Following curettage there was slight vaginal bleeding, and vague lower abdominal pains persisted. When she was re-examined a week later, moderate tenderness was still present in both adnexa—more so on the right side. Antibiotic therapy was continued, but as there was no improvement in the signs or symptoms she was advised to have a laparotomy with probable hysterectomy and bilateral salpingo-oophorectomy.

At operation, the right ovary was found to contain a haemorrhagic mass ± 1 in. in length and in width, and this was attached to the outer pole; the mass was continuous with the ovary and totally free of adhesions to the tube or any other tissue. There was no blood in the

peritoneal cavity. The right tube was normal and the left tube and ovary appeared normal. Nevertheless doubt existed about the benign nature of the firm haemorrhagic tumour on the right ovary, and it was decided to perform total hysterectomy and bilateral salpingo-oophorectomy. Apart from a mild urinary infection her postoperative course was uneventful.

Histology showed: 'The right ovary contains two large cysts, each measuring 1.5 cm. in diameter. One is filled with blood clot, the other contains clear fluid. Within the blood-filled cyst in the right ovary, placental villi are clearly recognizable, and in addition a cavity is present in the centre, with a thin collagenous wall lined by a single layer of flattened cells. This represents an ovarian ectopic pregnancy. The adjacent cyst is lined in part by luteinized cells (corpus luteum)' (Fig. 1).

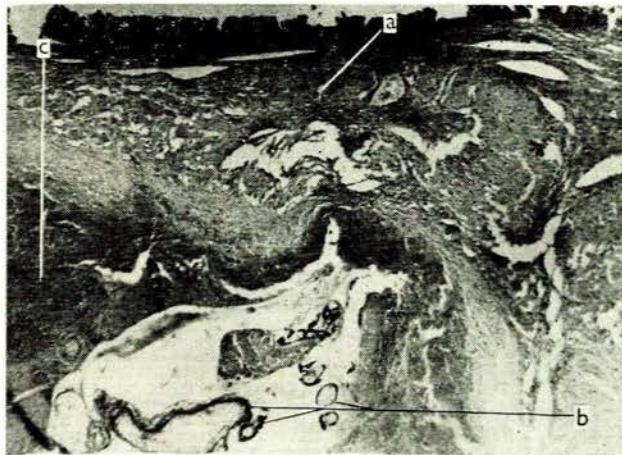


Fig. 1. Section of ovary showing: (a) corpus luteum, (b) chorionic villi and (c) blood clot.

DISCUSSION

The incidence of ovarian pregnancy was estimated by Hertig¹ to be 0.7-1% of all ectopic gestation, and on this basis he predicted an incidence of one ovarian pregnancy in 25,000-40,000 pregnancies. Boronow *et al.*² found one ovarian pregnancy in 9,229 pregnancies, and any apparent increase in incidence is really due to more careful section and study of haemorrhagic ovaries.

The diagnosis of ovarian pregnancy is based on the classical criteria put forward by Spiegelberg in 1878. These were:

- (a) that the tube on the affected side must be intact and separate from the ovary,
- (b) that the foetal sac must occupy the normal position of the ovary,
- (c) that the ovary and sac must be connected to the uterus by the ovarian ligament, and
- (d) that definite ovarian tissue must be present in the sac wall.

The last criterion was modified by Williams,³ who said that ovarian tissue must be found in several different areas in the whole of the sac at some distance from each other. A further modification,⁴ was the suggestion that ovarian tissue must be demonstrated between foetal structures and any tissue that was adherent. With advanced ovarian

pregnancies these modifications help determine the site of nidation.

A modification of the first criterion was made by Norris,⁵ who demanded not only a histologically normal tube (on the affected side), but that there must be no microscopic evidence of pregnancy; this would exclude a secondary ovarian pregnancy that had originated as a tubal pregnancy. Several authors⁶ have subsequently advised serial sectioning of the tubes. However, in young women it would seem unreasonable to remove a tube which looks and feels normal when operating for a haemorrhagic cyst on the ovary. Where there is doubt about the normality of the tube, it should be removed.

A perusal of the clinical data in various series^{2,4,7,21} indicate that the present case was atypical in respect of several factors, viz. age, parity, findings on examination and the absence of free blood in the peritoneal cavity. In Boronow's series² of 65 cases collected from the literature between 1950 and 1963, the average age was 28.7 years and the parity 1.3 (only 27% of cases were primigravidae). A pelvic mass was present in 90% of the cases, and a pre-operative diagnosis of ectopic pregnancy was made in 66% of cases, but none of these was labelled as ovarian pregnancy. The commonest incorrect pre-operative diagnosis was ovarian cyst. There was complete absence of intra-abdominal bleeding in only 3 of these cases; in 40%, the blood loss exceeded $\frac{3}{4}$ pint. The average period of gestation at termination of the pregnancy was $7\frac{1}{2}$ weeks, 91% of cases ending before 14 weeks. These findings are in substantial agreement with those of other series, though Baden and Heins⁴ found that 25% of their cases went on to the second trimester. About half of these would last to the third trimester or beyond. About 66% of the viable foetuses were stillborn and 18% of these were malformed. In all series, previous pelvic inflammatory disease, associated endometriosis and infertility were quite uncommon.

In general those cases reaching viability will usually have spurious labours and are then operated on as abdominal pregnancies. Most of the infants are stillborn, but a few have survived. Rarities recorded include a simultaneous intra-uterine and ovarian pregnancy, twin ovarian pregnancies,^{8,9} two consecutive ovarian pregnancies in two consecutive years¹⁰ and ovarian hydatidiform mole.¹¹

In 5 of the cases mentioned above,² colpo-puncture demonstrated intraperitoneal bleeding, and colpotomy in one case. In 4 cases, a biologic test for pregnancy was positive. Four cases were curetted; in only one was decidua found. In 115 cases of ectopic pregnancy Romney *et al.*¹² studied the curettings and only 19% showed decidua. In the remainder, decidua may have been shed or may not have developed at all, and they emphasized that failure to find decidua did not in any way rule out the possibility of ectopic pregnancy. Reid¹³ pointed out that both the biological pregnancy test and the presence of decidua depended on the trophoblast being viable and capable of secreting hormones.

A further aid in pre-operative diagnosis is the Arias-Stella phenomenon or reaction. Described in 1954 by Arias-Stella,¹⁴ the reaction consists of typical histological changes in endometrial glands and epithelium under the hormonal influence of chorionic tissue. These changes may be present with or without decidual transformation, are

usually focal, and of varying intensity. The epithelial cells in the endometrial gland are enlarged and their nuclei are hypertrophic, hyperchromatic, lobular, elongated or bizarre in shape. A characteristic feature is absence of polarity. The nuclei often occupy the luminal part of the cells. The cytoplasm may be vacuolated and look foamy. Mitoses are occasionally present (Fig. 2).

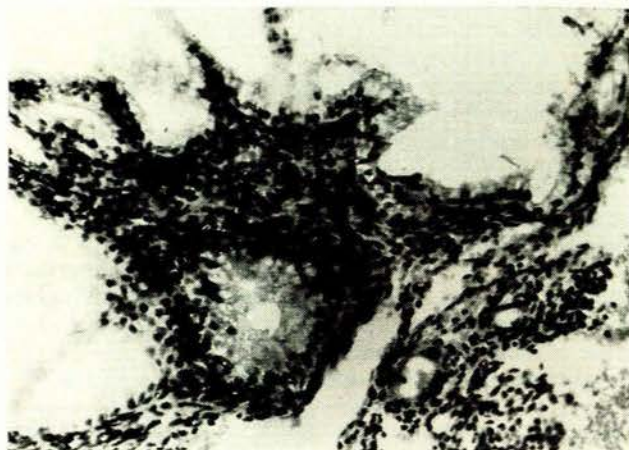


Fig. 2. Endometrium showing hyperchromatic nuclei and cytoplasmic swelling and vacuolation.

This phenomenon has been found in extra-uterine pregnancy, abortion, hydatidiform mole, chorionepithelioma and endometritis. In ectopic pregnancy a positive Arias-Stella reaction occurs in from 26 to 81% of cases, and most authors are agreed that these atypical cells are more frequently found than decidua.^{15,16} When both these features coexist, the diagnosis is virtually certain. The practical value of the Arias-Stella reaction is that, in the absence of decidua, it will suggest the presence of an undiagnosed or atypical extra-uterine pregnancy, provided chorionic villi are absent and the clinical findings have excluded a previous abortion. In the present case these atypical cells were found on re-examination of the curettings after histological examination of the operative specimen.

Only a positive atypical cellular reaction is of diagnostic value, since both atypical cells and decidua may be absent in 8 - 25% of cases.

Physiological Mechanisms of Fertilization

As the follicular ovum is not mature enough for fertilization, most authorities have felt that fertilization occurred elsewhere. Rock and Hertig¹⁷ showed that the follicular ovum may occasionally undergo meiosis and thus be ready for ovarian fertilization. Again Rock¹⁸ and Huffman¹⁹ have postulated that fertilization occurs in the fimbriae, and then secondarily implants on the ovary when the ciliary current and tubal peristalsis are collectively insufficient to convey the zygote. It has also been pointed out²⁰ that the fimbrial end of the tube may roam over the ovarian surface and fertilization could occur at such a time both within the fimbria and on the surface of the ovary.

In the aetiology, inflammation is a factor in only a small percentage. Ineffective tubal function may play some part. However, all the mechanisms of fertilization would seem to be explained by chance alone.

SUMMARY

A case fulfilling the diagnostic criteria of primary ovarian pregnancy is presented, indicating some of the difficulties in diagnosis. Emphasis is laid on histological aids to diagnosis in cases of unsuspected extra-uterine pregnancy.

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