CHOCOLATE CYST OF THE OVARY COMPLICATING TERM PREGNANCY

(Review of Literature and three Case Reports)

by

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Tumours complicating pregnancy are showing a higher incidence due to ever-expanding indications for caesarean section. McKerron (1903) estimated the frequency of ovarian neoplasms met with in pregnancy as 1 in 2,500 pregnancies. Grimes *et al* (1954) found ovarian enlargement of more than 6 c.m., once in every 274 pregnant women. Booth (1963) reviewing the records of patients seen at Queen Charlotte's Hospital over the period of 10 years (1950-59) found an incidence of 1 in 591 pregnancies. Greenhill (1965) states that ovarian neoplasms occupy the second place as a tumour complicating pregnancy, leiomyomas being the most frequent association.

Dermoid cysts are the commonest and about 50% of all types of ovarian neoplasms complicating pregnancy. (Carverly 1931, Booth 1963) Endometrial cyst of the ovary complicating pregnancy is a very rare association. Reported cases in the literature are very few and this is because endometriosis often produces sterility. But the situation has improved greatly with ever-increasing hopes for the women who were victims of endometriosis and is due to newer progestogens available for the treatment of such conditions. Up till now there are only seventeen reported cases of ovarian endometriosis associated with pregnancy that could be collected from the literature. In this paper three more cases of chocolate cyst complicating term pregnancy are reported. The cases reported here, occurred in this hospital and were diagnosed at caesarean section. All of them were asymptomatic throughout pregnancy or had mild symptoms that could be relieved by conservative treatment.

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Received for publication on 23-8-67.

Case 1

Mrs. S. B., para 0 + 0, married for 2 years. No contraception. Menarche 11 years, cycle 28 ± 2, duration 3/4 days, dysmenorrhoea severe type. Before marriage she had two attacks of sub-acute appendicitis which were relieved with conservative treatment.

She had antenatal care regularly under senior obstetricians and on 13th November 1965 she was admitted, being 15 days over-

due. In the antenatal period she had subacute pain in the right iliac region during the 2nd and 3rd trimesters which were treated and relieved by conservative measure. On the day of admission she was otherwise normal except that the presenting head of the baby was not engaged. Vaginal examination showed that cervix was ripe; stripping of the membrane was done. This was followed by castor oil and enema. Re-assessment after 12 hours revealed the os to be 1½ fingers, cervix partially taken up and bag of membranes flush with the head. Head was in L.O.P. position. Considering overdated pregnancy, high floating head in left posterior position and not much progress of labour after 12 hours of medical induction, caesarean section was decided on. A living male child, weighing 6 lbs. 10 ozs., was delivered by lower segment caesarean section. Before closing the abdomen uterus was eventrated to clean the pouch of Douglas when a sac burst containing tarry fluid; it was adherent to the right half of the posterior wall of the uterus and broad ligament. It appeared as if there was a big haematoma on the right infundibulopelvic ligament and the mass was connected to the ovarian ligament. It was diagnosed as right-sided ovarian cyst. The left ovary was then inspected and it also contained a chocolate cyst about the size of an orange. Partial resection of right ovary was done and then repaired with an omental graft for covering the raw areas. On the left side partial resection and repair of the ovary was done. There was doubt about the existence of ovarian tissue in the remaining portion.

Patient was given a blood transfusion of 600 c.c. as also three pints of 5% glucose. The puerperium was morbid in the first week. A stitch abscess was drained on the 6th day. Broad spectrum antibiotics were given but the temperature range rose up. A lower abdominal mass merging into the subinvoluted uterus was felt and confirmed by pelvic examination. Suspecting an infected haematometra cervical dilatation was done but no fluid could be drained. The postoperative period continued turbulent, abdominal distention increased, urinary output diminished and the patient expired three days later.

Case 2

Mrs. R. G., para 0 + 0, 22 years, married for 2 years, menarche at 13 years, no dysmenorrhoea. She did not use contraceptives. She was admitted on 7th December 1965 with dribbling of liquor at 39th week of gestation. Baby was presenting by vertex which was not engaged. Internal examination was not done and she was given antibiotics and sterile vulval pad. Next day she did not start labour pains and internal examination revealed that cervix was tubular and long. Os closed. Pelvis — subnormal with flattening of sacrum. Head was still floating. Considering subnormal pelvis with flat sacrum and premature rupture of membrane with floating head, caesarean section was decided on.

At laparotomy black tarry material was scattered on the anterior wall of the uterus. After completion of repair of the uterus, it was eventrated. Right ovary contained a chocolate cyst of about foetal head size, while the left ovary contained another chocolate cyst of duck's egg size. Right-sided ovariotomy and left-sided ovarian cystectomy were performed. Tarry thick material was adherent to the intestinal wall. On attempting to remove this the gut wall appeared to be damaged and so only superficial removal was done.

She was discharged on the 10th postoperative day. Follow-up after 8 months of operation — no complaints. She started menstruating in 5th month after childbirth. Internal examination showed uterus well involuted, and in midposition. Right fornix —clear, left fornix—a small mass 1½" X 1½", mobility slightly restricted.

Case 3

Mrs. P. S., aged 23 years, menarche at 12 years, dysmenorrhoea of severe type which started after 7 years of menarche. For two years she was having menorrhagia. She was married for 6 years. Prepregnancy — Anovlar-21 monthly for 6 months. She was having regular antenatal care and was admitted at 39th week with labour pains. Foetus was presenting by the vertex which was floating. Vaginal examination revealed, cervix partly taken up. Os closed. Pelvis adequate.

After 12 hours of labour, mother showed early signs of exhaustion and an internal examination was done—cervix completely effaced, os—2 fingers, head still floating and in posterior position. Caesarean section was decided on. On section tarry material was found on the uterine wall. When the uterus was eventrated after rapair, a chocolate cyst about the size of an orange was felt on the left side while on the right side a small cystic ovary was present. On the left side there were dense adhesions between the cyst, gut and omentum. Adhesions were separated and left-sided ovariotomy, with right-sided enucleation of the cyst was done.

She was discharged on 10th postoperative day. At home she had a stitch abscess.

Follow up: After 6 months of childbirth she had no complaints and she was still suckling her baby. Internal examination showed that the uterus was of normal size, in midposition and no mass felt in the fornices.

Histological examination of the tissue removed from the three cases showed the same picture. Lining of cubical epithelium in some places with large and deeply stained cells underneath. There were endothelial leukocytes heavily laden with blood pigments (pseudoxanthoma cells). This picture suggests decidual reaction in the wall of the endometrial cyst.

Accompanying figure shows the changes mentioned above. It is a section from the removed tissue of Case 1.

Discussion

Reviewing the literature of chocolate cyst with pregnancy, only seventeen case reports could be collected. These cases can be classified into three groups.

I. Cases of early uterine pregnancy, with chocolate cyst of the ovary/ovaries diagnosed at laparotomy.

Sampson (1922) first reported a case where laparotomy was done for suspected fibromyoma, but an exploration of abdomen revealed a chocolate cyst of the ovary in association with uterine pregnancy of about 14 weeks. Winestine (1924) did ovariotomy for left quadrant abdominal pain in the early months of pregnancy. Aschheim (1929) reported a case where there was right ovarian endometrial cyst in early pregnancy. Shanning (1930) reported an adherent endometrial cyst of one ovary removed from a patient who was carrying 16-18 weeks. Von Franque (1934) described fist size ovarian endometrial cyst removed from a patient who was gravid for 6 weeks, but the patient ultimately aborted. Scott (1944) reported two cases of cystic ovaries with uterine pregnancy. In one of his cases he did a laparotomy in the early months. This was a case of bilateral chocolate cysts with 12 weeks' gestation. He did right salpingo-oophorectomy, and resection of left ovary and appendicectomy. Patient delivered at 35 weeks. Barnes (1945) reported two cases where she did laparotomy in the early months; in one case she did enucleation of the cyst and in the other a salpingo-oophorectomy. In one of her cases (where enucleation was done) the affected ovary also contained corpus luteum which she could salvage during enucleation. Both the cases were of unilateral chocolate cysts in association with pregnancy. Both the cases carried to near term and caesarean section was done in one case; the other case was carrying 30 weeks when the cases were reported. Both the cases were sisters and another sister of theirs was also being investigated under Dr. Barnes for

secondary infertility. Pelvic findings in this case also revealed a round mobile swelling 3″ in diameter and seemed to be in the right ovary.

II. In this group of cases pregnancy was advanced and, due to associated complications in pregnancy, laparotomy was done and chocolate cyst was detected. This is the group of great clinical significance.

Scott (1944) reported a case who during routine antenatal care in the early months had uterine fibroid with left ovarian cyst in addition to pregnancy. Considering her long period of infertility and being asymptomatic she was being closely followed during the antenatal period. At 35th week she had a sudden severe, sharp and non-radiating pain in the lower abdomen in addition to constipation for 3 days. There was no muscle guarding or rigidity in abdomen, but there was tenderness in the right lower quadrant of the abdomen extending from umbilicus almost to Poupart's ligament. Tentative diagnoses were — (1) twisted ovarian cyst; (2) acute appendicitis; (3) degenerated fibroid. McBurney's incision was made for appendicectomy, which revealed brownish-black semisolid material. So the abdomen was opened again by a midline incision. Caesarean section was done and, on eventration of the uterus, bilateral chocolate cysts adherent to the uterus were found. Subtotal hysterectomy with bilateral salpingo-oophorectomy was done in this case.

Nelson *et al* (1950) and Steinberg *et al* (1962) reported two cases where rupture of endometrial cyst occurred during pregnancy and precipitated an acute abdomen.

Brill *et al* (1957) and Noel (1961) reported two cases of bilateral chocolate cysts in advanced pregnancy simulating concealed accidental haemorrhage. The reported case of Brill *et al* (1957), a multigravida, at term, was admitted with sudden excruciating pain. Uterus was hard and tender but there was no vaginal bleeding. Foetal heart sounds were 120 and mother's pulse was 72 per minute. The tentative diagnosis was abruptio placentae with concealed haemorrhage. After caesarean section, eventration of the uterus revealed bilateral chocolate cysts ruptured spontaneously. He did total hysterectomy with bilateral salpingo-oophorectomy.

Noel (1961) reported a case who had painless bleeding per vaginam at 24 weeks of pregnancy and it was diagnosed and treated as cervical polyp. At 34 weeks again she was admitted with intermittent pain in abdomen. Her blood pressure was 180/110 mm. of Hg. and the foetal heart sounds were 134 per minute regular with foetus presenting by vertex. Internal examination revealed cervix to be long and tubular and os closed; no vaginal bleeding. Probable diagnosis was mild concealed accidental haemorrhage. After section he found bilateral chocolate cysts ruptured. He failed to identify any ovarian tissue and he felt it unwise to attempt mobilization of adherent tissue. He just removed the chocolate coloured fluid and closed the abdomen. Patient made uneventful recovery.

III. In this group ovarian endometriosis was an association of ovarian pregnancy.

McKenzie (1943) first reported a case of ruptured ovarian pregnancy with endometriosis. The second case was reported by Durburg *et al* (1958). Their case was a primary ovarian pregnancy with bilateral endometrial cyst treated by subtotal hysterectomy and bilateral salpingo-oophorectomy. Modawi (1962) reported a case of primary twin ovarian pregnancy with ovarian endometriosis and this was diagnosed accidentally on histological examination. This is a very interesting and a rare case. Clinical significance of this group is not of much importance.

The co-existance of chocolate cyst and uterine pregnancy is very rare and seems paradoxical. The terms "Chocolate Cyst" and "Endometrial Cyst" of the ovary are now-a-days synonymously used, although the term 'endometrial cyst' is a better one as it reflects the actual pathology. Sampson (1922) called this type of ovarian cyst of endometrial origin as 'perforating chocolate cyst'. The cyclic haemorrhage and increased intracystic pressure tend to make these cysts more prone to leakage. During pregnancy, the cyclic haemorrhage being absent, and the cysts being thickwalled, the leakage of the distended cysts or sudden rupture seems to be unusual. But among the reported seventeen cases in the literature and the three cases reported here, only in four cases were symptoms present which at laparotomy proved to be either leakage or rupture. The usual process seems to be an occasional leak with rapid walling off by reacting inflamation and resultant extensive adhesions. The break-up of adhesions due to enlarging uterus seems to be the cause of leakage, in the absence of cyclic intracystic haemorrhage during pregnancy producing "sub-acute abdomen". This was the condition in most of the reported cases and in the first case reported in this paper.

The subacute abdomen in advanced stage of pregnancy also poses diagnostic problems. In the first case reported by Scott (1944) the possibilities were, twisted ovarian cyst, acute appendicitis, degenerated fibroid. This again proves the value of careful internal examination in the early months of pregnancy which is often avoided or done half-heartedly. In advanced stage of pregnancy it is very difficult to palpate an ovarian cyst which is placed posteriorly unless known in the early months. The test as described by Hingorani (1966) may be sometimes helpful in diagnosing difficult cases of pregnancy complicated by ovarian tumour.

The leakage of chocolate fluid into the peritoneal cavity which is highly irritant and semisolid in character can also create confusion with the diagnosis of concealed accidental haemorrhage. This occurred with Noel (1961) and Brill *et al* (1957). The presence of rebound tenderness as suggested by Scott (1944) might be helpful to exclude accidental haemorrhage in such a case favouring intestinal or intraperitoneal accident.

Chocolate cyst with pregnancy may often remain asymptomatic, being diagnosed accidentally during caesarean section. This was the condition with the second and third cases reported in this paper, suggesting the importance of examination of ovaries and tubes at caesarean section.

As the cysts, may remain asymptomatic during pregnancy and labour, they might produce acute/sub-acute abdomen in the immediate puerperium and in these cases prompt laparotomy instead of conservative attitude might bring better results.

The problem of surgical treatment, once diagnosed, will be solved by noting the extent of the lesion — unilateral or bilateral involvement, age and parity of the patient. Conservative treatment, by removing only the chocolate material and not tackling the cyst, does not seem to be a sound policy, as the chocolate material in the cyst wall is bound to produce irritative peritonitis and other complications. Conservative treatment was done in the first case reported in this paper and also in the case of Noel (1961). Result was satisfactory with Noel and also in the first case reported here. But the case ultimately had stormy postoperative period and succumbed to it following dilatation of cervix in order to drain a suspected infected haematometra which was thought to be the cause of her late rise of temperature not responding to antibiotic treatment. But the surgeon in this case did not feel happy with the conservative treatment.

As most of the patients are primiparas and in young child-bearing age, removal of uterus with both adnexae or of one side seems to be too radical. When bilateral—worst side should be sacrificed totally and on the other side enucleation and reconstruction of the ovary seems to be justified. This was the attitude during the operation on the second and third cases reported here, with uneventful recovery of the patients. The condition of the ovarian tissue is such that often it is very difficult to identify the anatomy unless one is aware of the possibility of chocolate cyst. This confronted the surgeon in the first case which was his first experience. In his subsequent two cases he was well experienced to tackle the condition scrupulously.

As the possibility of leaking chocolate cyst cannot be diagnosed, it is always better to open the abdomen by midline or paramedian incision instead of McBurney's incision while doing appendicectomy in pregnancy.

Conclusion

(1) Three cases of chocolate cysts with advanced pregnancy are reported in this paper. Review of literature revealed that only 17 cases are reported up till now, of which 3 were with ovarian pregnancy.

(2) Various clinical manifestations of leaking chocolate cysts in pregnancy are discussed.

(3) Surgical approach to such rare condition is also discussed.

(4) Importance of routine internal examination in the first trimester of pregnancy has been emphasised.

(5) During caesarean section eventration of the uterus and examination of the tubes and ovaries are also stressed to identify asymptomatic coexistence of chocolate cyst in pregnancy.

Acknowledgement

The authors are thankful to Prof. K. N. Mitra, Professor-Director, Department of Obstetrics and Gynaecology, Medical College, Calcutta, for his constant encouragement in pre-

Fig. 1(a)
Case 1. "Rama Lakshmi".

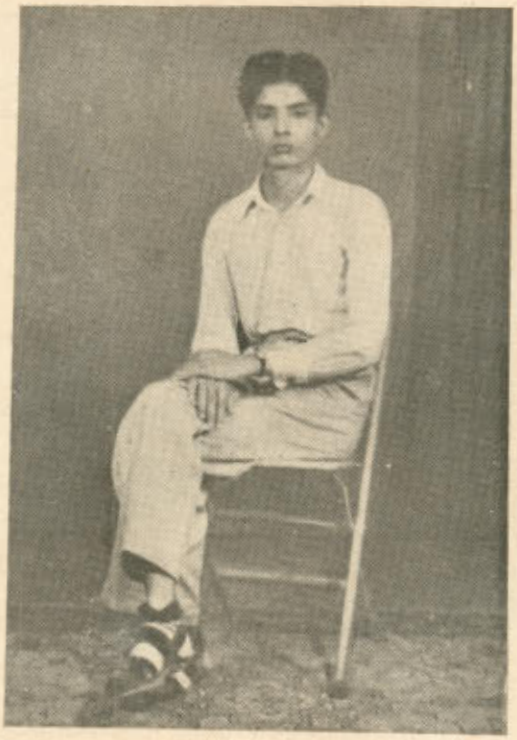


Fig. 1 (b)
Same case as in Fig. 1(a) "Ram Sharma"

Fig. 2 (a)
Ardanarishwar" (Meenakshi temple, Madurai)

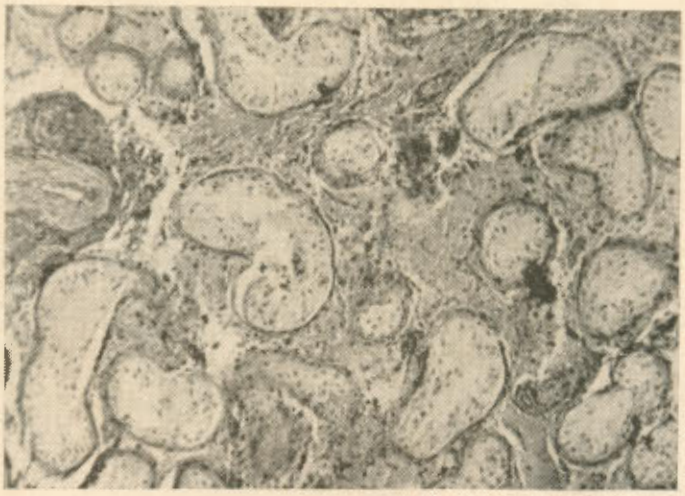


Fig. 2 (b)
Eris (Hermaphrodite) (Courtesy Scott & Jones 1958)

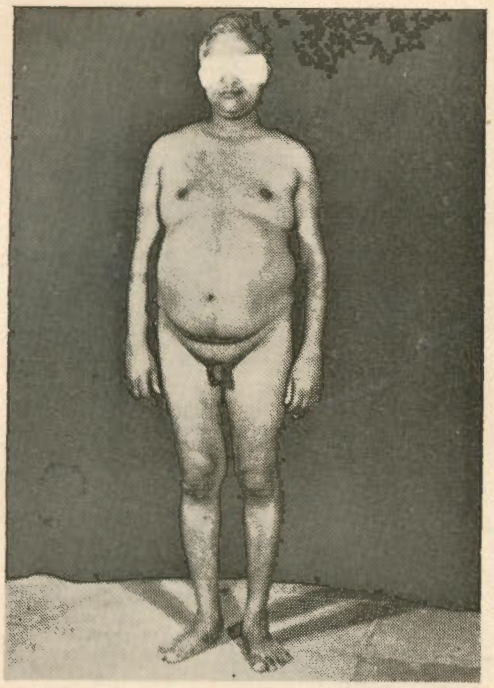


Fig. 2 (a)
Case 2. A case of Kline-felter's syndrome.

Fig. 2 (b)
Case 2. Testicular biopsy: Peritubular fibrosis.

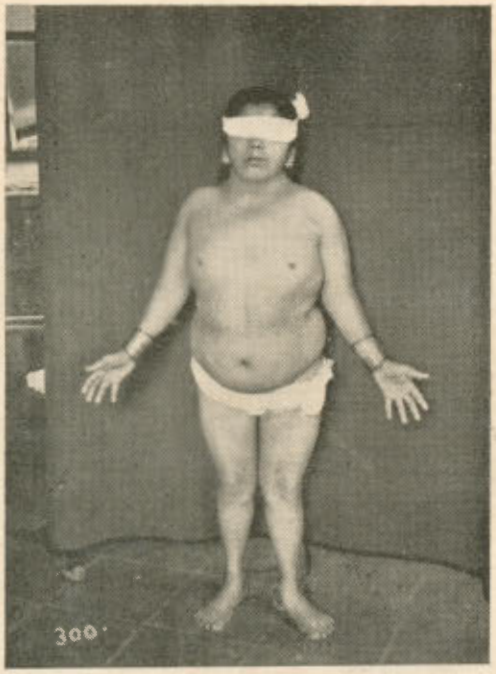


Fig. 3 (a)
Case 3. A case of Turner's Syndrome. Note marked carrying angle and webbing of neck (Courtesy J. Obst. & Gynec. Brit. Emp.).

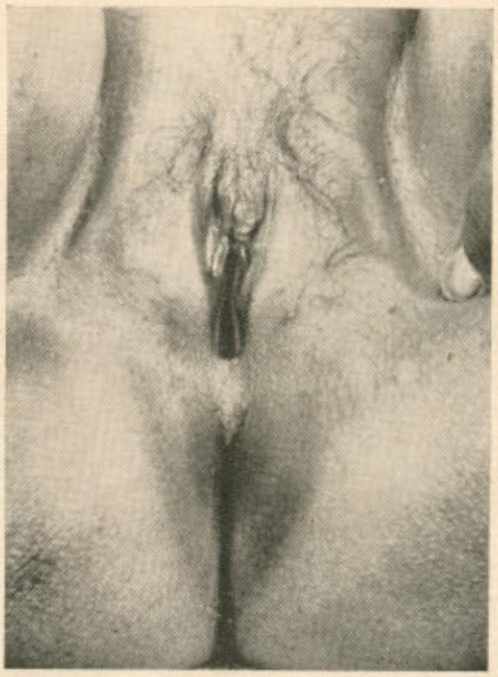


Fig. 3 (b)
Case 3. Note feminine type external genitalia with agenesis of gonads

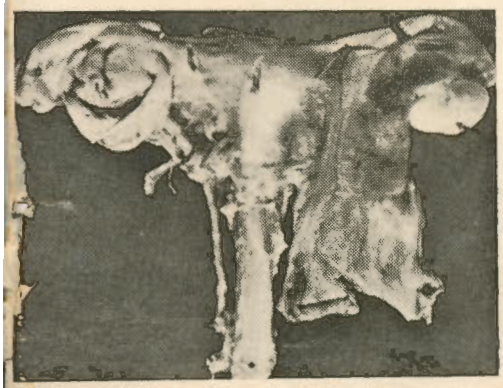


Fig. 4 (a)
e 4. Gross specimen of hernia showing rus, vagina with tubes. Left gonad looked e the testis with the tunica and the right ovary.

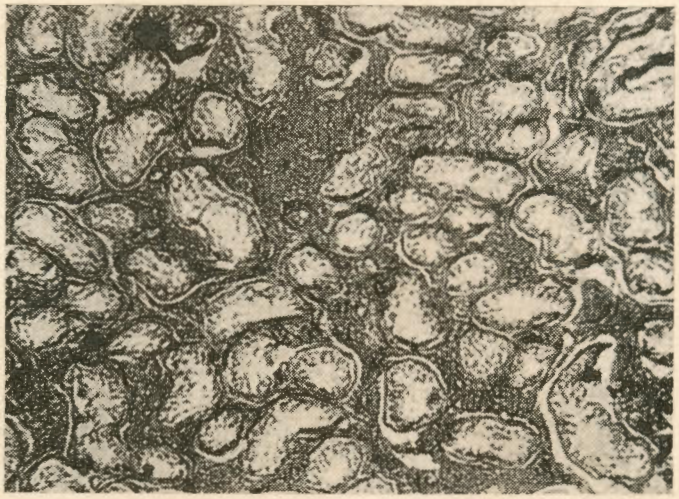


Fig. 4 (b)
Case 4. Photomicrograph of gonad showing testicular tissue.

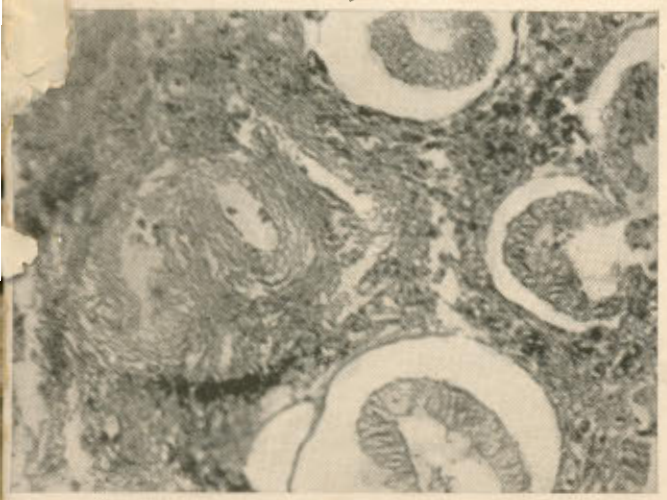


Fig. 4 (c)
Case 4. Endometrium from uterus showing simple tubular glands due to oestrogen effect.

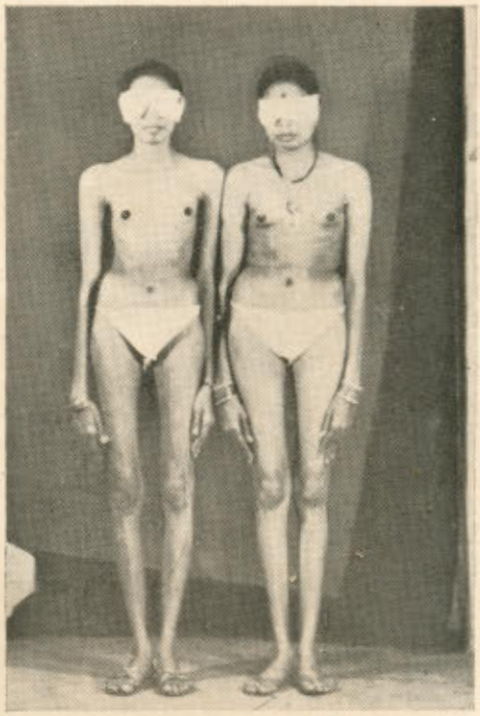


Fig. 5 (a)
Cases 5(a) and (b). Pure Gonadal dysgenesis —two sisters.

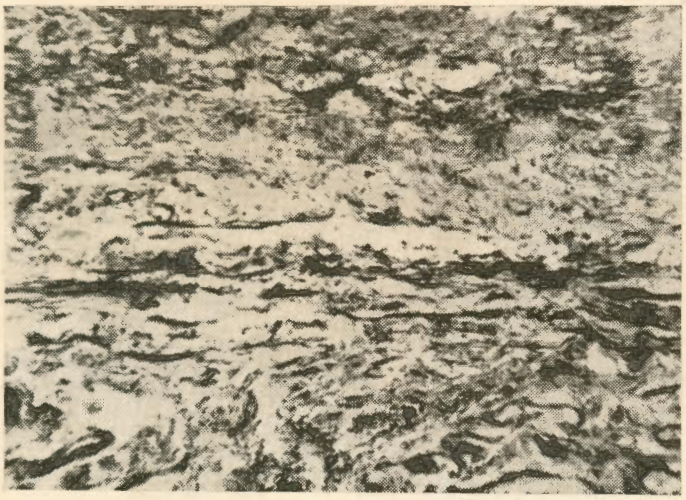


Fig. 5 (b)
Case 5. Photomicrograph of streak Gonad showing absence of follicles.

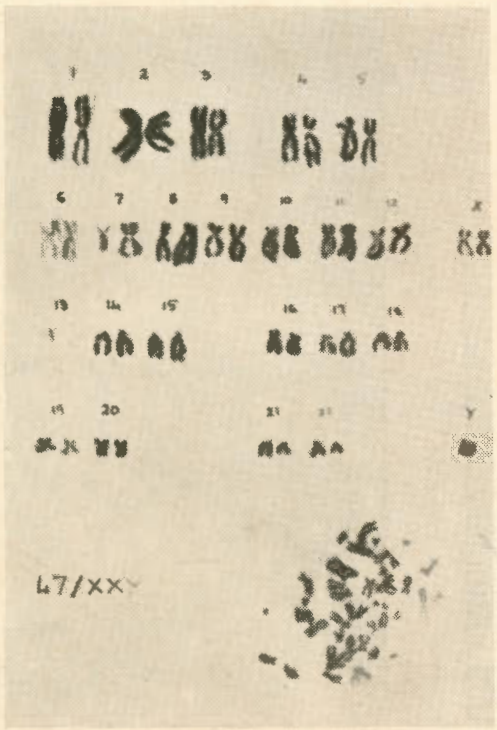


Fig. 5 (c)
Case 5. Karyotype showing 47/XXY.

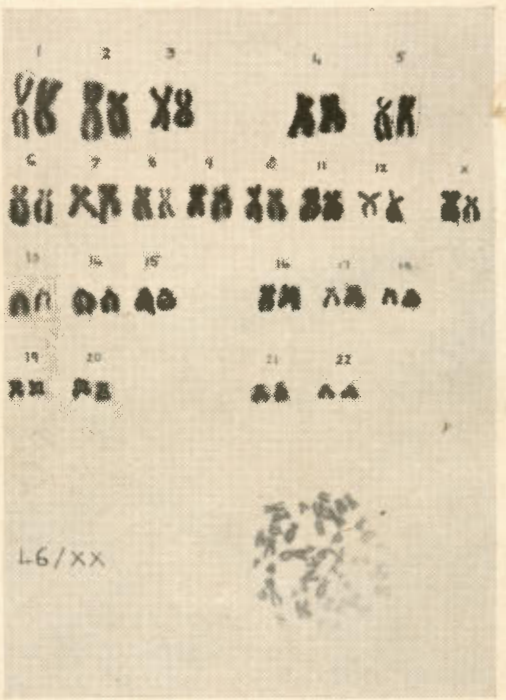


Fig. 5 (d)
Case 5. Karyotype showing 46/XX.

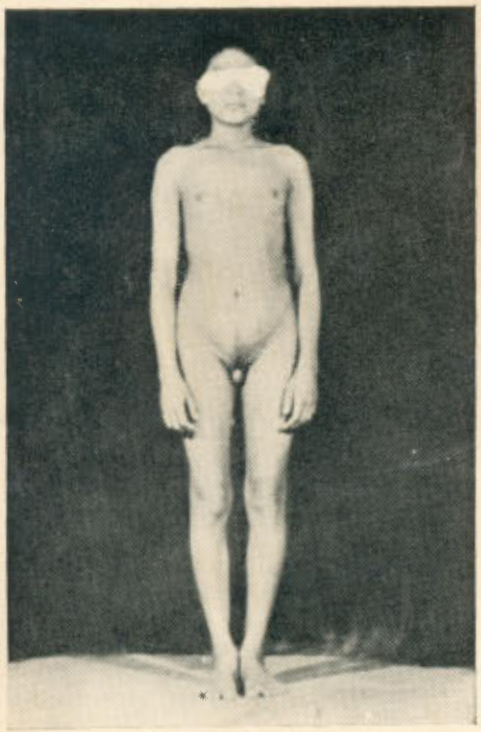


Fig. 6 (a)
Case 6. A case of testicular dysgenesis showing poor development of breasts.

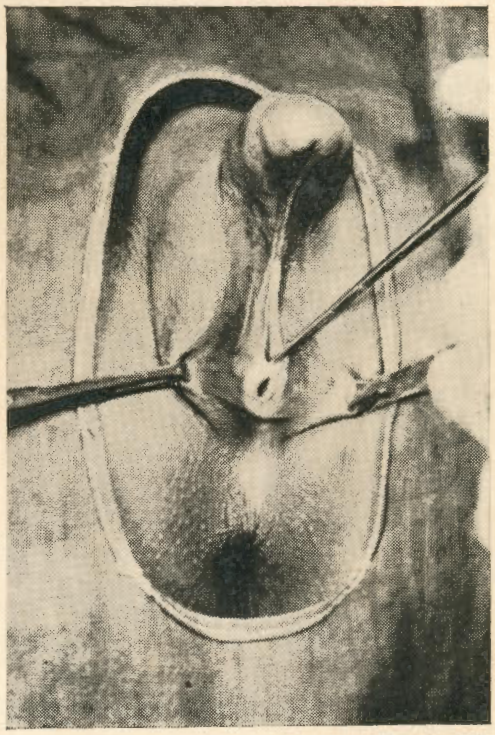


Fig. 6 (b)
Case 6. External genitalia showing enlarged phallus with a grooved urethra.

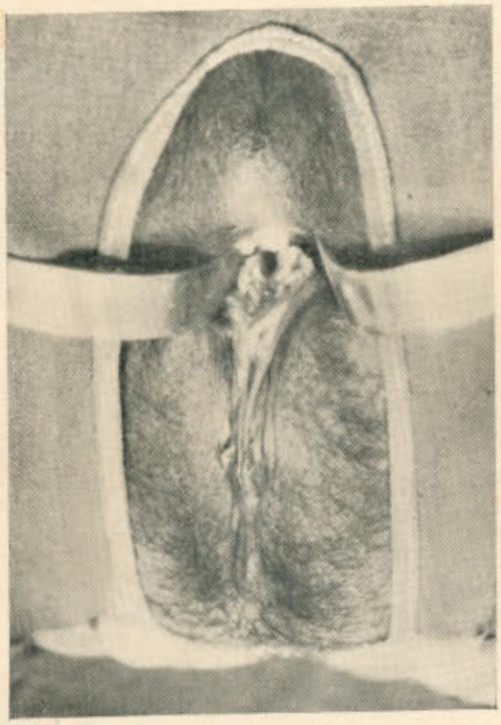


Fig. 6 (c)
Case 6. External genitalia after reconstructive surgery.

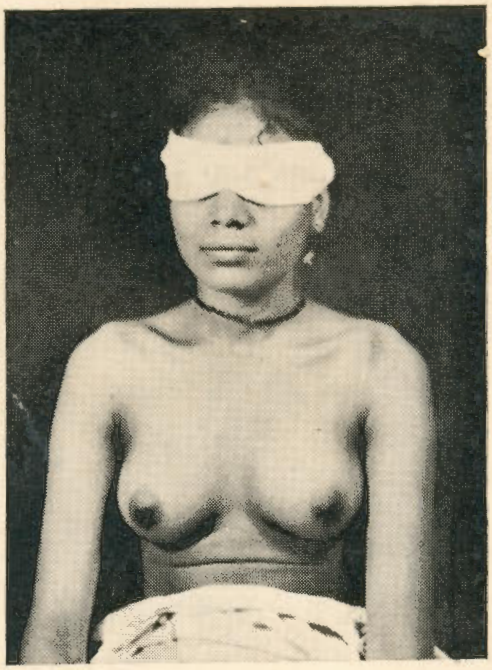


Fig. 6 (d)
Case 6. Breast development after hormone therapy.

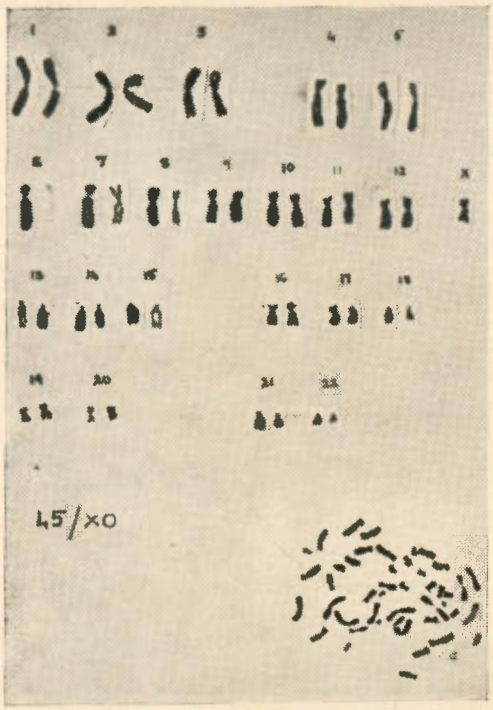


Fig. 6 (e)
Case 6. Karyotype showing 45/XO.

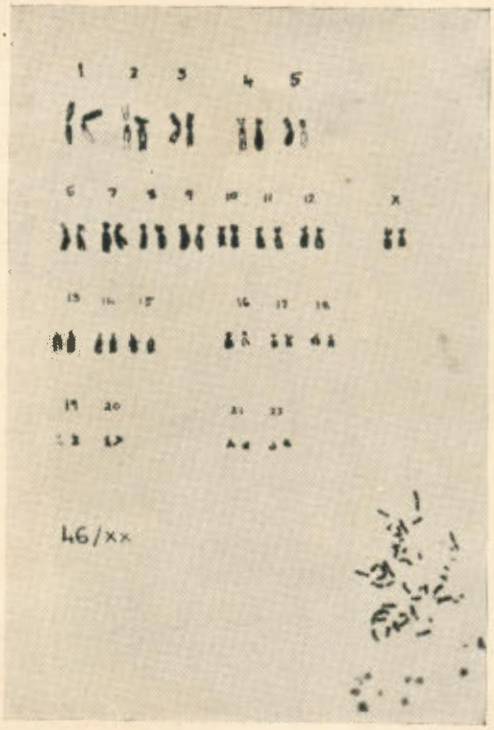


Fig. 6 (f)
Case 6. Karyotype showing 46/XX

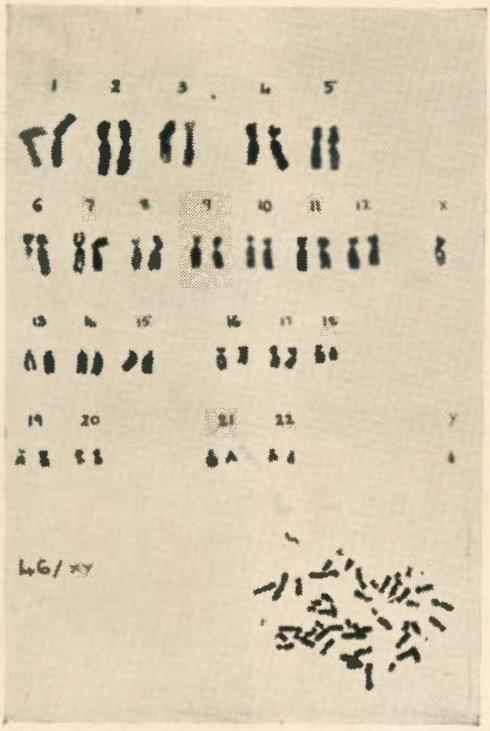


Fig. 6 (g)
Case 6. Karyotype showing 46/XY

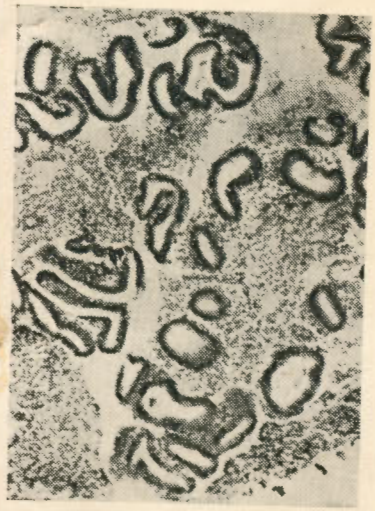


Fig. 1
Microphotograph shows adenomatous hyperplasia. There is active proliferation of endometrial glands with very little stromal space.

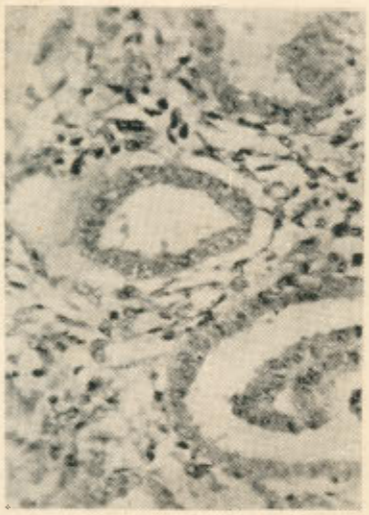


Fig. 2
Microphotograph shows cystic hyperplasia. The glands are irregularly cystic and lined by tall cells. There is sprinkling of inflammatory cells.

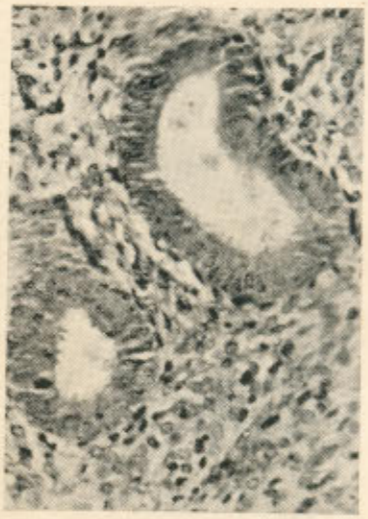


Fig. 3
Microphotograph shows proliferative type of endometrium.

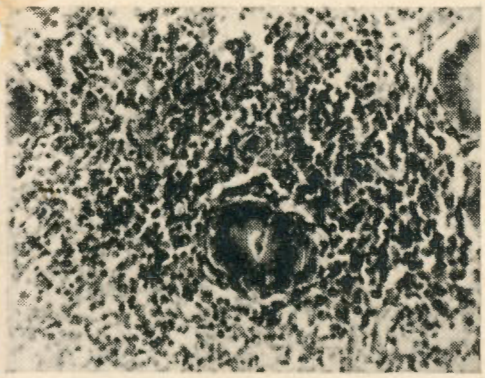


Fig. 4.
Microphotograph shows proliferative endometrium with very few glands which are non-secretory. There is considerable chronic inflammatory reaction in the stroma.

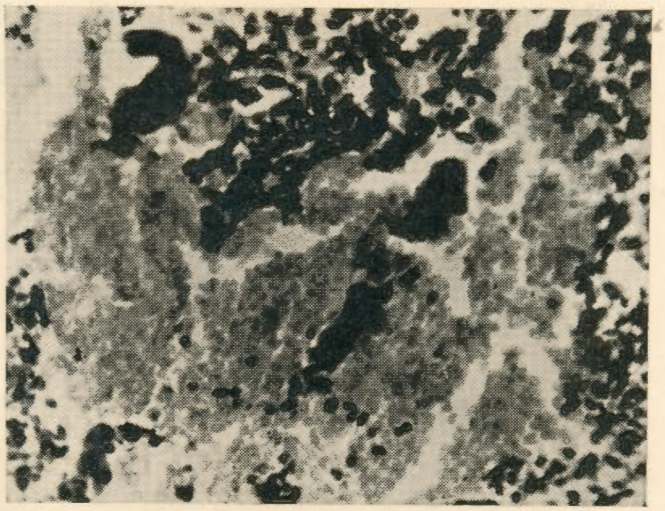


Fig. 5
Microphotograph reveals scanty hyperplastic endometrium. There is marked haemorrhage in the stroma.

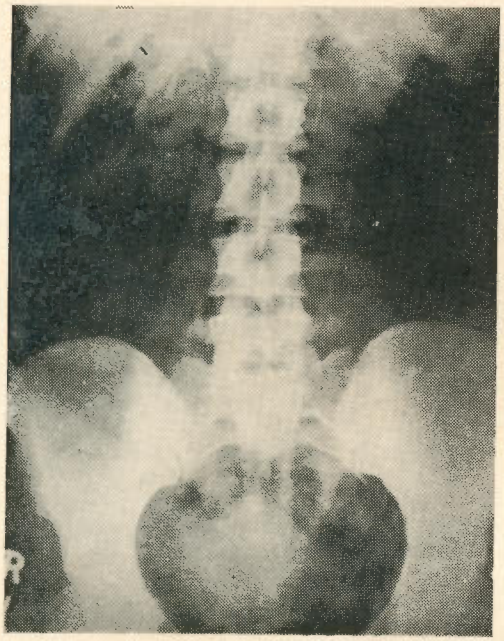


Fig. 1.
Photograph of non-pregnant woman. The calyces are cip shaped, the superior border of pelvis is concave. The diameter of right ureter is slightly greater than the left ureter.

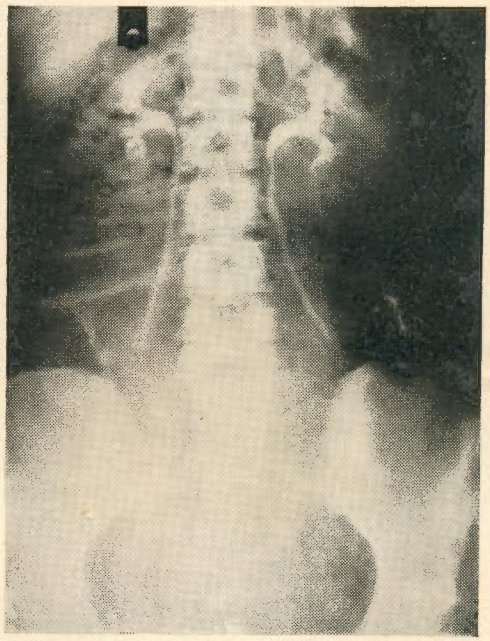


Fig. 2
Photograph of a pregnant woman without bacteriuria. Slight rounding of calyces and convexity of the upper border of renal pelvis is seen. The ureters are dialated.

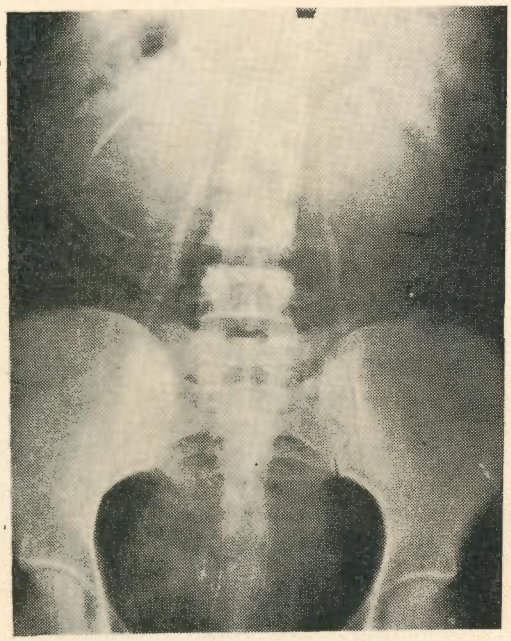


Fig. 3
Pregnancy with bacteriuria. The calyces are showing clubbing. The upper border of renal pelvises are convex and ureters are dilated.

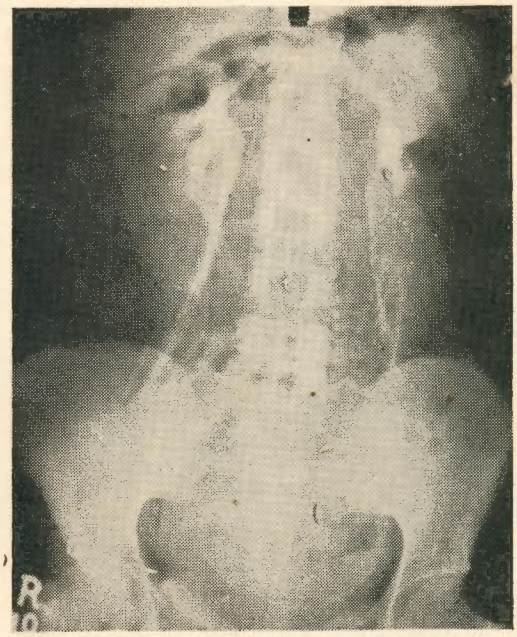


Fig. 4
Photograph of a pregnant woman with bacteriuria, urine showing above 100,000 colonies/cc. Intravenous pyelography shows loss of concavity of the calyces on the right side. The upper border of the right pelvis in convex. The right ureter is dilated; the left calyces, pelvis and ureter appear normal.

Scope and Limitation of Vacuum Extractor—Samadder pp. 654-662

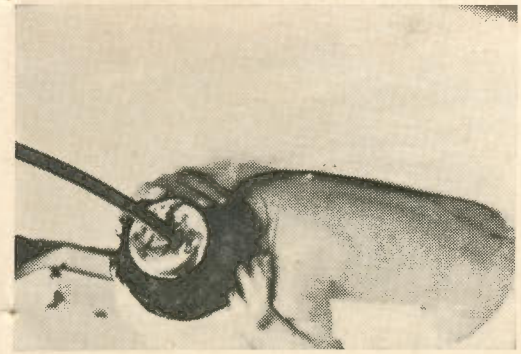


Fig. 1
Cup attached to the scalp. Delivery has just been completed by Ventouse.

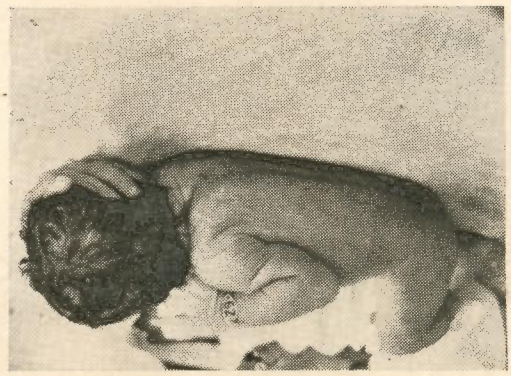


Fig. 2
The Chignon immediately after the removal of the cup.

Dystocia due to Foetal Abdominal Distention—Dhall and Indra Dhall pp. 709-712

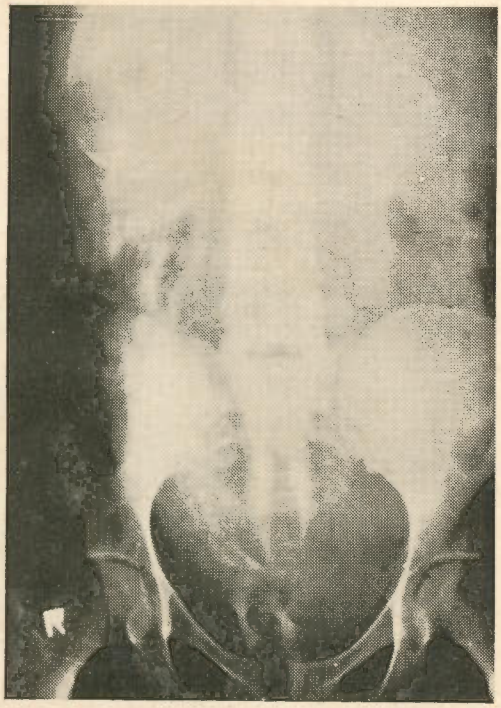


Fig. 1
Antero-posterior view showing displacement of foetus to the right side, straightened spine and unfolding of the foetal limbs due to a large soft tissue mass.

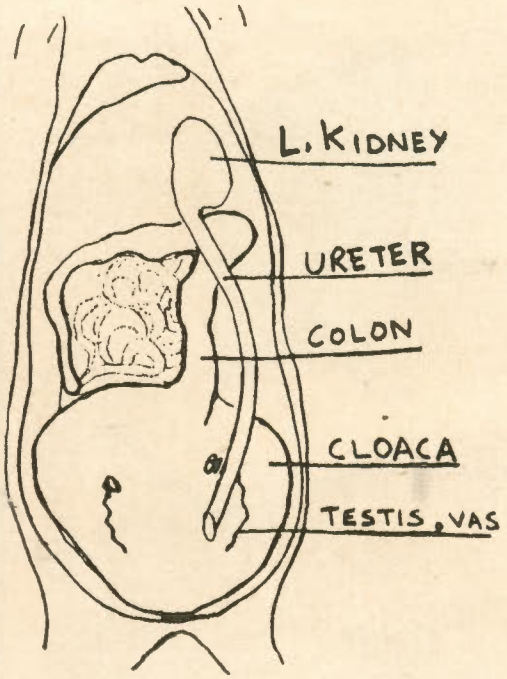


Fig. 2
The left ureter is seen opening into the grossly distended cloaca.

Dystocia due to Foetal Abdominal Distention—Dhall and Indra Dhall pp. 709-712

Fig. 3
Shows foetus after reinjecting 1600 cc of fluid into the abdomen.

Chocolate Cyst of Ovary and Term Pregnancy—Chakraborty and Banerjee pp. 673-679

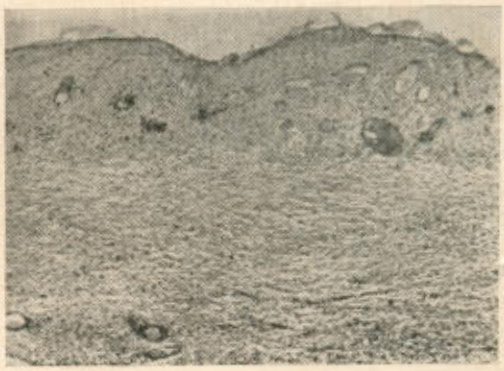


Fig. 1
Section from the chocolate cyst of right ovary of case No. 1

Velamentous Insertion and Rupture of Cord—Kavadia et al. pp. 715-717

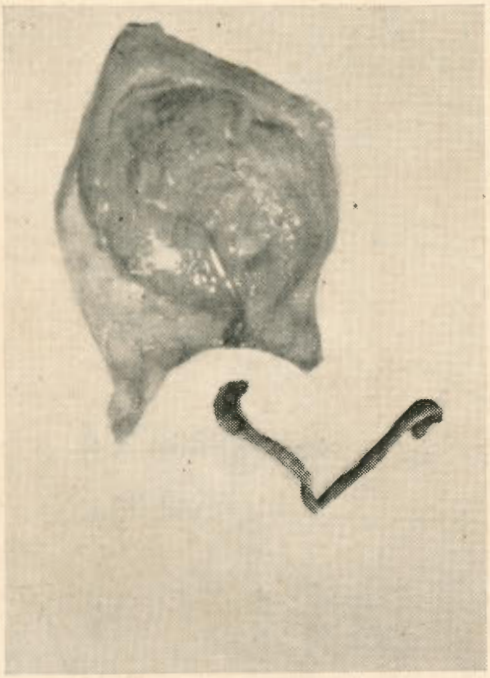


Fig. 1
Velamentous insertion of umbilical cord with short cord.

Single Umbilical Artery—Rohatgi pp. 718-720

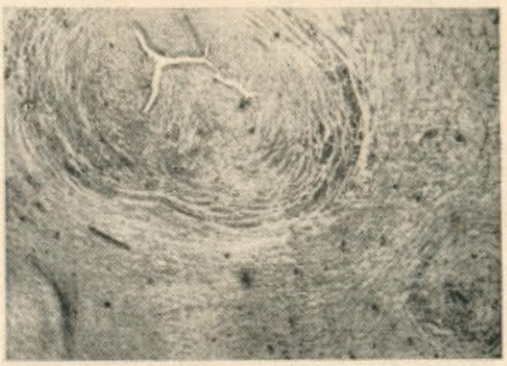


Fig. 1
Microphotograph showing a single umbilical artery.

Rhabdomyosarcoma of Ovary — Dubey and Agarwal pp. 724-725

Fig. 1
Bisected tumour mass showing uniform sarcomatous appearance with areas of haemorrhage.

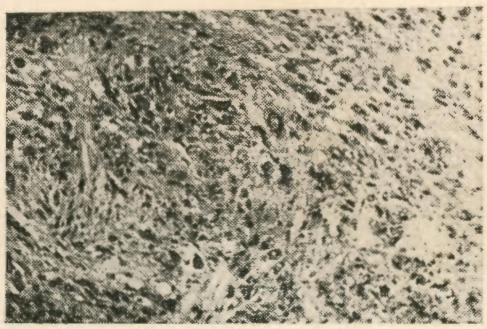


Fig. 2
Photomicrograph showing pleomorphic pattern (H. & E. x 100)

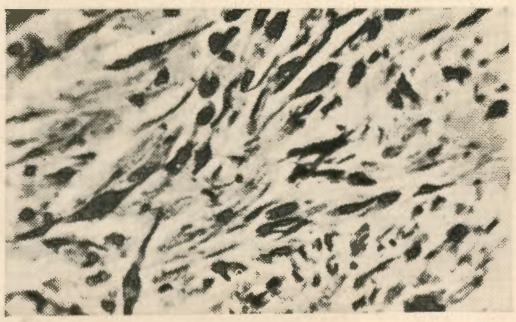


Fig. 3
Photomicrograph showing characteristic giant cells (H. & E. x 400).

pp. 726-729

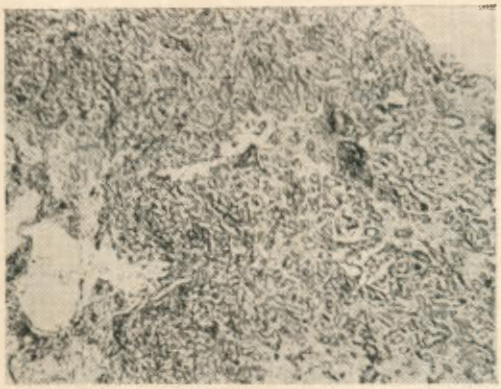


Fig. 1
Section of ovcary from a case of highly differentiated tubular variety of arrhenoblastoma; tubular pattern of the gland, is well seen (H. V. E. x 60).

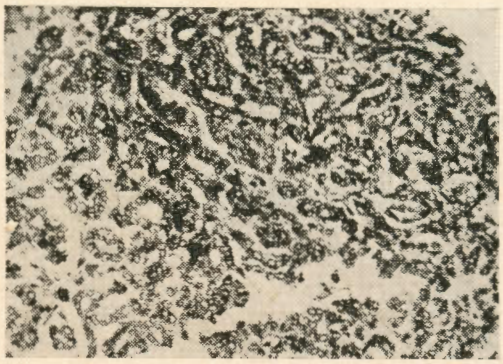


Fig. 2.
Same section under higher magnification showing glandular pattern mimicking seminiferous tubules. The interstitial tissue is scanty (H. V. F. x 240).

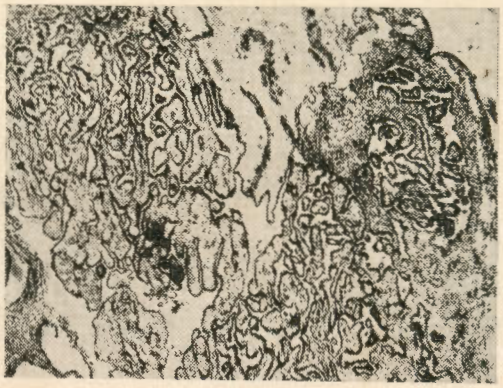


Fig. 3
Section of ovarian tumour from another area showing features mimicking rete testes (H. V. E. x 60)

Fig. 1

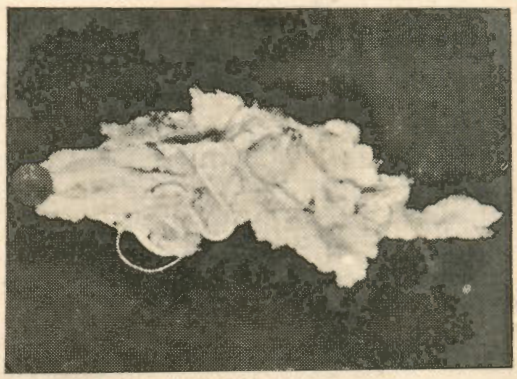


Fig. 3

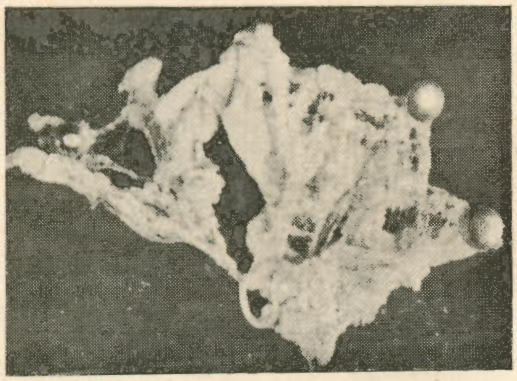


Fig. 2

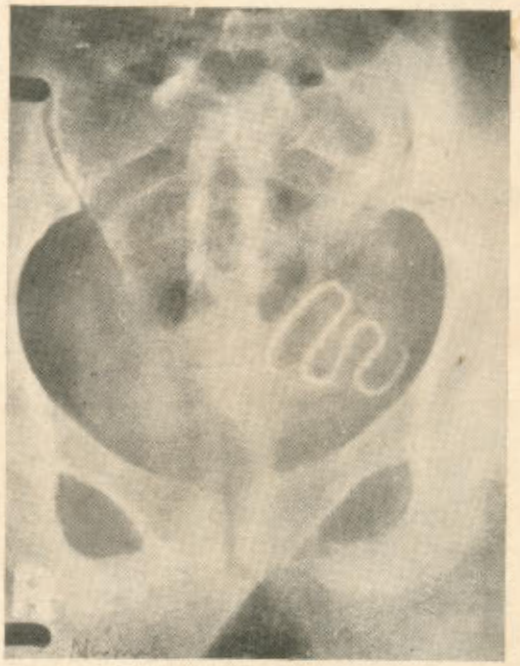
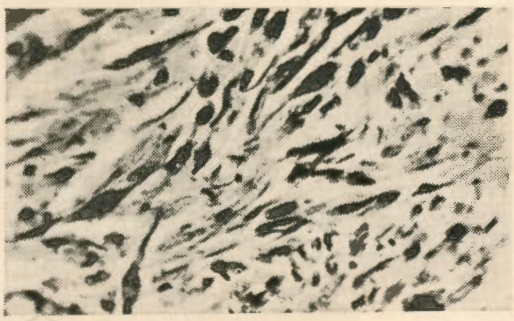


Fig. 4

Fig. 1
Bisected tumour mass showing uniform sarcomatous appearance with areas of haemorrhage.

Fig. 2
Photomicrograph showing pleomorphic pattern (H. & E. x 100)



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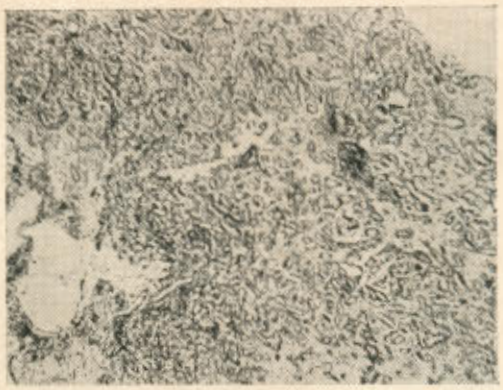
Photomicrograph showing characteristic giant cells (H. & E. x 400).



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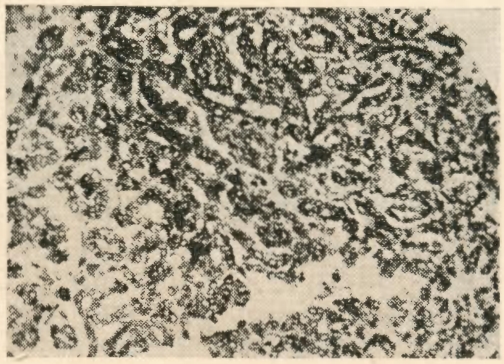

Fig. 2.
Same section under higher magnification showing glandular pattern mimicking seminiferous tubules. The interstitial tissue is scanty (H. V. F. x 240).

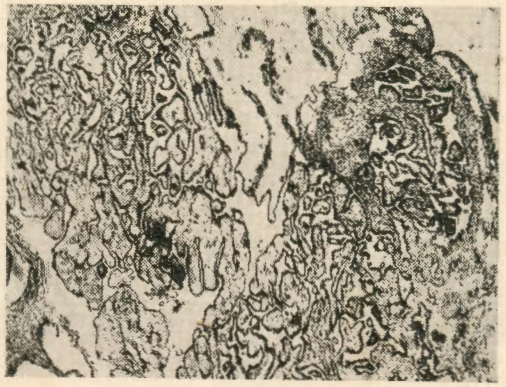

Fig. 3
Section of ovarian tumour from another area showing features mimicking rete testes (H. V. E. x 60)

Fig. 1

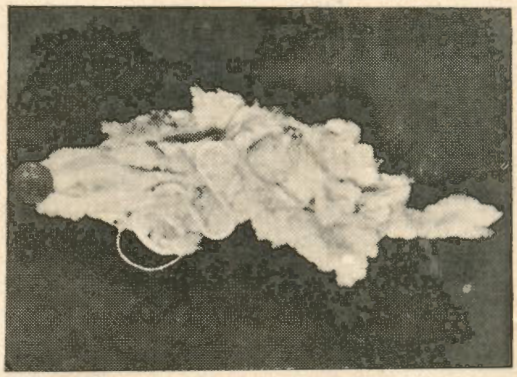

Fig. 3

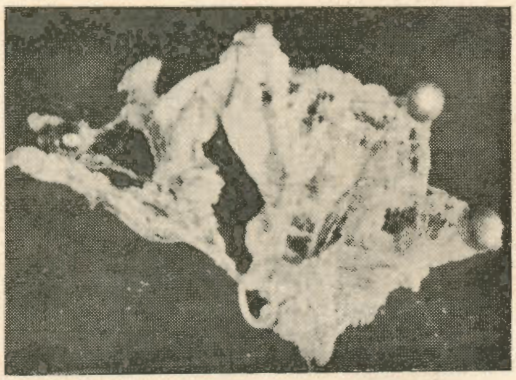

Fig. 2

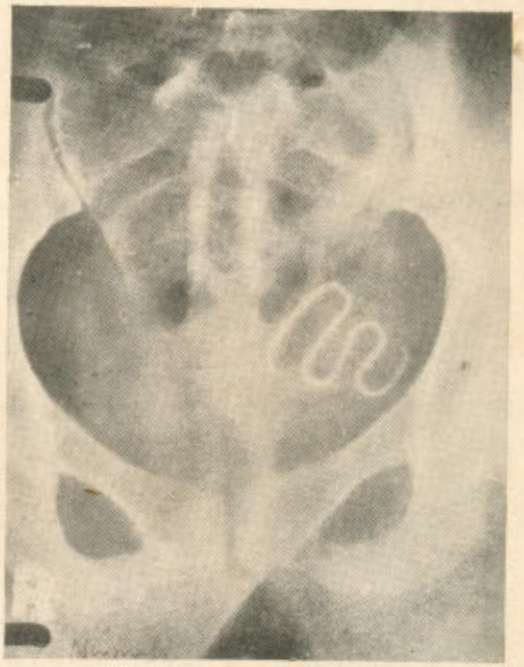

Fig. 4

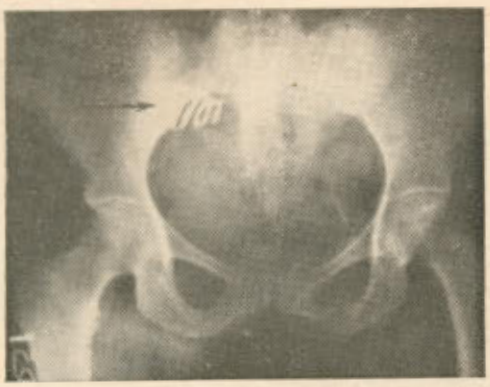

Fig. 1
Plain skiagram of abdomen. A. P. view of case No. 1 showing extrauterine displacement of loop into the peritoneal cavety (Rt. side)

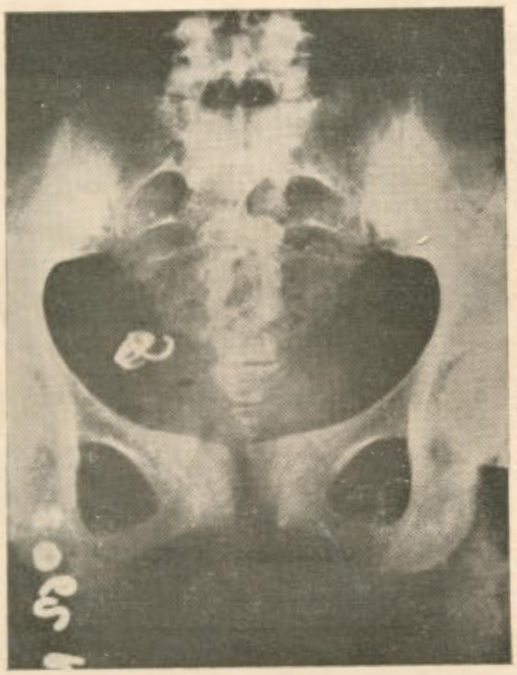

Fig. 2.
Plain skiagram of abdomen. A. P. view showing extrauterine displacement of loop into right broad ligament

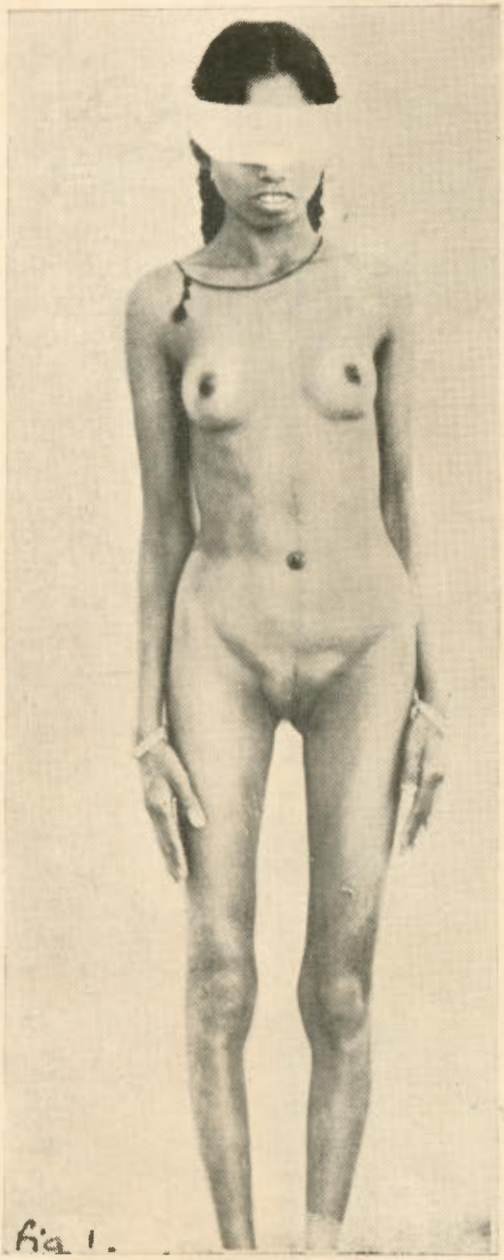

Fig. 1
Photograph of the patient in standing position.

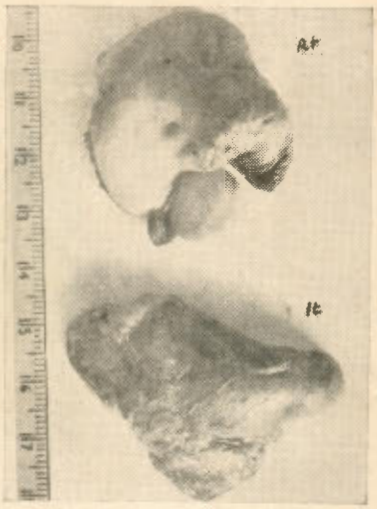

Fig. 2
Gonads removed at operation.

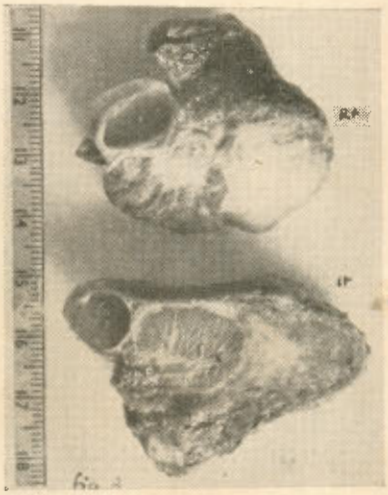

Fig. 3
Gonads on cut section.

Fig. 4
Microphotograph of the gonad.

paring this paper. The authors are grateful to Dr. K. C. Sarbadhikari, Principal-Superintendent, Medical College & Hospitals, Calcutta, for allowing them to present this material.

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Fig. on Art Paper X