KRUKENBERG TUMOUR IN PREGNANCY WITH VIRILIZATION

(A Case Report)

by

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Krukenberg tumour is a rare malignant ovarian tumour. Its exact incidence in pregnancy is not known. Incidence of malignant ovarian tumours in pregnancy is 1:18,000, and about one seventh of these tumours are Krukenberg tumours (Hertig and Gore, 1971). One out of the 11 cases of Krukenberg tumour reported in the recent Indian literature had a pregnancy of 4 months' duration at the time of operation (Konar, 1967; Tyagi *et al*, 1967; Talib *et al*, 1974; Rao *et al*, 1975; Phillips and Kaur, 1976 and Mathur *et al* 1976). Krukenberg tumour with evidence of endocrinal activity especially showing evidences of virilization during pregnancy is very rare; only a few such cases are reported in the world literature. Spadoni *et al* (1965) collected 9 cases of Krukenberg tumours associated with signs of virilization from the world literature and 1 of them was pregnant. In addition they reported 1 cases of Krukenberg tumour in pregnancy associated with virilization. A similar case operated recently by one of the authors is presented below:

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CASE REPORT

Mrs. S. 20 years, para 1, second gravida, was admitted to Cantonment General Hospital, Poona on 13th September 1976 with symptoms of early labour following amenorrhoea of 8 months' duration and cough and low grade fever of 2 days duration.

Her first pregnancy and labour were uneventful and she had a 2 years old alive. normal female child. Her periods had returned 6 months after this delivery and continued regularly till middle of January 1976 when she conceived again.

During the present pregnancy she did not have any antenatal care, nor did she consult any physician for any illness and she took no medicines during this pregnancy. She had not been suffering from any illness and had no symptoms pertaining to gastrointestinal tract and breasts.

The patient was of average built and nourished. She had mild pallor, acne on face and slight oedema of feet, no jaundice or cervical lymphadenopathy was detected, blood pressure was 160/110 mm Hg, pulse 92 min., respiration 22 per minute and temperature 99°F; signs of upper respiratory tract infection were present. Breasts were normal clinically, liver and spleen were not palpable. Uterus was o 34 weeks' size. Foetal head was engaged, foetal heart rate was within normal range. No other mass was felt. Vaginal examination revealed a taken up cervix and half dilated os, head was at 'O' station, and no mass was felt in the pelvis.

Haemoglobin was 9 gms%, leucocytic count within normal range. Urine examination—sugar and protein not detected.

She delivered normally on the same day a alive female infant weighing 2,200 gms. Baby's clitoris was slightly enlarged and labia were hypertrophied, vagina was located with probe. About 36 hours after delivery she felt pain in the lower abdomen. She passed normal stool. Temperature was 100°F, pulse-126 min. and respiration 26 per minute. Mild signs of bronchitis were detected. Abdominal examination revealed a firm, slightly tender swelling occupying whole of the lower abdomen extending upto the umbilicus and partly to the right lumbar region. Uterus was not felt separately from the mass. No free fluid was detected in the peritoneal cavity. No mass was felt in pelvis per vaginal examination.

X-Ray chest—N.A.D. X-Ray abdomen (standing) revealed a soft tissue shadow covering whole of lower abdomen.

A provisional diagnosis of twisted ovarian tumour was made. Laparotomy was done. There was about 100 ml of clear free fluid in the peritoneal cavity. Two solid, firm and freely mobile masses with smooth surfaces replacing both ovaries were seen in the lower abdomen. The right mass was 19 x 10 x 6 cm and left one 9 x 6 x 4 cm (Fig. 1) . The tumours were greyish-white in colour. There was no haemorrhage, excessive vascularity and softening anywhere on the tumour. Exploration of other organs of abdomen and pelvis revealed no abnormality. Total hysterectomy, bilateral salpingo-oophorectomy and omentectomy were performed. The patient required blood transfusion at the time of operation. Postoperative recovery was uneventful and she was discharged two weeks after operation.

Laboratory Reports

1. Female infant sex chromatin study of buccal mucosal scraping showed presence of Barr bodies.

2. Peritoneal fluid: cytology did not show malignant cells.

3. Histopathology report: Cut surface of both masses were homogenous, greyish white in colour and greasy. The tumour on the right side was oedematous. There was no haemorrhage and necrosis. Multiple sections from both ovarian tumours showed almost identical picture. There were areas of proliferated compressed ovarrian stroma with areas of myxomatous degeneration. There were areas of large number of focal collections of signet ring cells with nuclear atypism and they showed strongly mucin positive cytoplasm (Fig. 2). In addition there were areas of adenocarcinoma and focal collections of cells with eosinophilic cytoplasm and vesicular nuclei giving an appearance of Leydig type of cells. No abnormality was detected in the uterus and fallopian tubes.

At the time of discharge the patient was alright. Liver was not enlarged and no mass was felt in abdomen, pelvis or breasts. She then went to her village and returned with jaundice in February 1977 i.e. 4½ months after operation when she had no gastrointestinal symptom excepting loss of appetite of about one week duration. Liver was just palpable and tender. Clinically no evidence of new growth was detected in abdomen, pelvis or breasts. Acne on her face had subsided. Liver function tests of the patient revealed serum bilirubin 15 mg% serum protein within normal range, S.G.O.T. and S.G.P.T. were 620 and 840 I.U. per litre respectively and serum alkaline phosphatase was 18 K.A.U. per 100 ml. Urobilinogen was not increased in the urine. Blood examination for Australia antigen was found positive. Barium meal and enema studies did not show any radiological evidence of new growth or ulcer in the gastrointestinal tract. The patient left the hospital against advice before her jaundice had subsided. She reported in August 1977 for one day with symptom of lump in her abdomen when jaundice was not detected. Pelvis and hypogastric regions were occupied by a fixed, hard and irregular mass. She was still breast feeding her baby, and no abnormality was detected in both breasts. External genitalia of the baby had a normal appearence now. Patient refused operation.

Discussion

Important points in this case were: young age of the patient, rarity of this condition, association of virilisation and absence of primary site.

A very similar case of Krukenburg's tumour with pregnancy was reported by Spadoni *et al* in 1965. The patient was of same age and parity; signs of pre-eclamptic toxaemia and virilization were detected during third trimester and she delivered at 33 weeks a premature female

foetus with enlarged clitoris and hypertrophied labia. She had preoperative high level of urinary ketosteroids and ketogenic steroids which returned to normal 5 days after removal of ovarian tumours. Site for primary lesion could not be detected till 7½ months after operation when she died. Postmortem examination revealed widespread metastases. Breast was probably the primary site of malignancy.

The present patient was very young. None of the 11 cases of Krukenberg tumour collected from Indian literature and 8 cases of Krukenberg tumour associated with virilization collected by Spadoni *et al* (1965) was so young.

Review of recent Indian literature did not show any case of Krukenberg tumour in pregnancy associated with virilization. Development of virilization in the female infant was possibly due to androgen production by the diseased ovaries probably by the Leydig cells which were seen in the histopathological examination of the tumour and the elaboration of androgens might have taken place only after the completion of development of the genital tract of the foetus. History of the case ruled out the possibility of virilization due to hereditary factor and drug therapy during pregnancy. Urinary steroid studies of the patient before removal of the tumour would have confirmed the association of androgen production. Endocrine activity of these tumours have already been reported by a few authors. Turumen (1955) and Scully and Richardson (1961) (quoted by Spadoni *et al*, 1965) reported the oestrogenic activity in menopausal women associated with Krukenberg tumours Spadoni *et al* (1965) collected 9 cases and reported 1 case of Krukenberg tumours associated with virilization.

In this case primary site of malignant growth was not detected. Development of primary Krukenberg tumour has already been reported (Novak and Woodruff, 1974). In many cases signs and symptoms of metastatic growth mask the clinical and laboratory signs of primary site of malignancy in gastrointestinal tract, gall bladder and breasts which clinically manifest occasionally after removal of the ovarian tumours. In this case primary site of malignancy was not detected during laparotomy and during four and half months follow-up period. It is not certain after a lapse of how much period the primary site reveals itself.

It may be possible that a fresh malignant growth would develop de novo on the places of primary sites. Hence it is justified to label this case as of primary Krukenberg tumour of the ovaries. Jaundice that the patient developed was of viral origin.

Summary and Conclusion

A case of Krukenburg tumour of ovaries during pregnancy is presented. Important features in this case are, a rare tumour complicating the pregnancy in a young woman; it was associated with signs of virilization which disappeared after removal of the tumour and the primary site of malignancy was not detected.

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See Figs. on Art Paper VII