ANDROBLASTOMA OF THE OVARY

(A Case Report)

by

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Introduction

Virilising tumours of the ovary are very rare, constituting less than 0.4% of all the primary ovarian tumours. Inspite of their rarity, these neoplasms are surrounded with a sort of scientific glamour not possessed by other ovarian tumors, because of their capacity to produce striking sex changes.

In 1965 Novak and Long reviewed all the authentic cases of arrhenoblastoma from the Ovarian Tumour Registry and found the number of reported cases to be 321. Review of literature in our country reveals only 9 reported cases of ovarian arrhenoblastoma (Paranjpe, 1959; Parekh, 1963; Iype and Mukherjee 1966; Banerjee, 1967; Sexena and Srivasthva, 1970; Deshmukh and Deshpande, 1971; Daruwala, 1973 and Kochhar and Ghosh, 1974).

CASE REPORT

J.D., a 28 years old married woman was admitted in the Gynaecology wards of the Rajendra Hospital, Patiala, on September 16, 1974, with complaints of amenorrhoea since 2 years, hirsutism since 1½ years, hoarseness of voice since 9 months and enlargement of abdomen since 6 months.

History of present illness: After her last confinement 5 years ago the patient had lactational amenorrhoea for 2 years when she weaned her child. One month after weaning, she had normal menstruation followed by amenorrhoea of 2½ months duration. For this some injections were given and she started bleeding 12-13 days after the injections. The bleeding continued off and on for about 3 months and the patient thought it to be due to threatened abortion. She was admitted to the local Lady Dufferin Hospital with the diagnosis of missed abortion and curettage was done. Still the bleeding continued off and on for another two months. Then the patient got admitted in the Rajendra Hospital in August 1972, where another curettage was done. Bleeding stopped four days after the curettage and the patient never menstruated after that.

She noticed excessive growth of hair over the face and body-1½ years back. The growth over the face became so thick that it required regular shaving. The patient was not very sure as to when exactly she started noticing atrophy of the breasts but the breasts had become much smaller by the time of admission (Her undergarment size was originally 36" but only 30" on admission).

Her relatives noticed the hoarseness of her voice about 10 months back. She got treatment in the department of medicine for her hirsutism and hoarseness of voice without any relief. Six months back she noticed enlargement of the abdomen and then she came to gynaecology outpatient department, where she was diagnosed as a case of virilising tumour of the ovary and was admitted in the hospital on 16th September, 1974.

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Menstrual History

Menarche at the age of 14 years, previous menstrual pattern was 5-6/30 days, regular, painless, normal flow.

Obstetrical History: She was married at the age of 13 years and had 3 full term normal deliveries, all females, the youngest child being 5 years old.

Examination: Moderately built, 151 cms. tall, weight 49 kgm. with thick growth of hair all over the body. The growth of hair was completely masculine in distribution (Fig. 1).

Pulse 88/mt.. B.P. 110/70 mm. Hg., Heart and lungs—N.A.D.

Abdominal Examination: A mass was felt in the lower abdomen reaching upto the umbilicus. It had well defined upper and lateral margins but the lower border could not be reached. Surface was smooth but lobulated and consistency variable—solid to cystic. The mass was mobile from side to side and some tenderness was present on deep palpation.

Vaginal Examination: Thick curly growth of typical male distribution type was present over the external genitalia with slight hypertrophy of the clitoris as compared to atrophy of labia majora and minora (Fig. 2). Uterus retroverted, normal size (later confirmed by passing a sound upto 7 cms). A semisolid mass about 20 x 20 cms. size was palpable in the anterior and lateral fornices. It was mobile and separate from uterus.

Investigations: Hb. 10.8 gms. per cent. Total and differential leucocyte count normal. Fasting blood sugar 84 mg/100 ml. Blood urea 18 mgm. per cent. Plain X-ray abdomen, skull and chest—N.A.D., Urine N.A.D. Urinary 17-Ketosteroids 13.4 mgm./24 hrs.

Operative findings: On 20 September, 1974 a lower paramedian laparotomy was done and a tumor of about 20 x 20 cm. size was found to be arising from the left ovary. The surface of the growth was smooth and glistening like that of a mucinous cystadenoma. There were no adhesions. The other ovary and uterus were normal. Left ovariotomy was done. Postoperative period was uneventful. The patient was discharged on October 2, 1974.

Follow-up: Exactly one month and one day after the operation on 21st October, 1974, she had her first menstruation after about 2¼ years of amenorrhoea. It lasted for 5 days and the flow was good. After having regular periods for 2 months, she again missed her period. Pregnancy test was found to be positive. She delivered a live full-term healthy male child on 27th August 1975. On follow-up, inspite of complete reversal of defeminising symptoms, hirsutism was still marked. Urinary 17-Ketosteroids done 3 months later were 12.4 mgm./24 hrs.

Pathological-observations

Gross-appearance: The specimen comprised of a large cystic mass measuring 23.5 x 14.5 x 9.5 cms. External surface was smooth and translucent. On cut section it was multilocular and filled with straw coloured fluid. There were greyish white homogeneous solid areas at certain places and some of the cysts were filled with dark brown mucinous material (Fig. 3).

Microscopic-appearance: The study of various sections showed a variable picture. The dominant feature was the presence of immature Sertoli cells arranged diffusely in groups or in cords resembling testicular embryonic sex cords (Fig. 4). Well defined tubules in between these immature Sertoli cells were also seen. Some of the Sertoli cells were distended with lipid material. Other areas showed sarcoma-like picture (sarcomoid pattern) containing tumor cells with elongated nuclei. Cystic spaces of variable size and shape dominated the histological picture in certain fields (Fig. 5). The cysts were lined by flattened epithelial cells and contained mucinous material in their lumina. This pattern represents heterologous element present in androblastoma (Serov et al, 1973).

Comments

The hirsute woman with amenorrhoea poses an interesting diagnostic problem regarding the source of excessive androgens. Androblastoma of the ovary is one of the causes of hirsutism with amenorrhoea, others being hilus-cell tumour and polycystic ovarian syndrome. Tumours or hyperplasia of adrenals and pituitary are the extra-ovarian causes of amenorrhoea and hirsutism. Thus, diversity of origin of androgens producing a clinically identical syndrome necessitates that physician and gynaecologist must work together in such cases. The present case had amenorrhoea and hirsutism for more than 1½ years before she reported to gynaecology out-patient department and that

also when she noticed a mass in the lower abdomen. By this time she had marked hirsutism, enlargement of clitoris and deepening of voice. Although, her feminising functions have returned dramatically after the operation as expected, the masculinising effects are still there and may not disappear completely.

Menorrhagia associated with an androblastoma, as in this case, is thought to be due to the effect of weakly acting androgens rather than an oestrogenic influence. Kochhar and Ghosh (1974) also reported menorrhagia as a leading symptom in one of their cases. Other explanation seems to be that the ovulatory function of the pituitary is more sensitive to suppression by androgens than follicular growth and oestrogen function. Thus, in the beginning of an androgen producing arrhenoblastoma the minimal androgenic titre suppresses ovulation but dysfunctional uterine bleeding goes on due to an unopposed action of oestrogens from the follicular growth, till the androgens titre becomes high enough as to suppress the pituitary completely.

Felicissimo and Junqueira (1938) reported the first case of arrhenoblastoma complicating pregnancy. Bretnall (1945) and Falk and Mason (1961) have also reported similar cases. Bretnall (1945) thought that pregnancy occurred whilst the tumor was producing very minute quantities of androgens.

Urinary 17-Ketosteroids are not very helpful in pin-pointing the diagnosis in virilising ovarian tumours though they are markedly increased when adrenals are the cause (Graber et al, 1961). Though Smiley et al (1953) reported that some cases of arrhenoblastomas had raised 17-Ketosteroids, Scully (1964) suggested that small unmeasurable amounts of potentially virilising testosterone may lead to profound masculinisation while other less virilising steroids may have markedly elevated urinary 17-Ketosteroids with no androgenic trends. Novak (1968) also agrees with this view. In the present case as well as in the case reported by Kochhar and Ghosh (1974) 17-Ketosteroids were within normal limits.

Malignancy: Recurrence rate of androblastoma is reported to be 22-23% by Javert and Finn (1951) and Novak and Long (1965). Hobbs (1949) and Flannery (1950) found blood stained free fluid in the abdomen. The tumors in their cases were moderate sized, multilocular and predominantly cystic with solid areas here and there. Henderson (1951) reported recurrence in 78 cases with 15% mortality. The case reported by Daruwala in 1973 had similar characteristics as those of Hobbs (1949) and Flannery in 1950. The tumor of the present case also had the same size and appearance macroscopically and sarcoid pattern microscopically, but so far there is no evidence of recurrence.

Summary

A case of androblastoma is reported. Importance of a thorough gynaecological examination in a hirsute woman for early diagnosis of virilising ovarian tumour is stressed, since the removal of tumour though followed by dramatic reversal of defeminising effects, the masculinising features may not be reversed completely. Follow-up for detection of malignancy is also stressed.

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See Figs. on Art Paper VII-VIII