ADRENAL TUMOUR (MASCULINOVOBLASTOMA) OF OVARY

(A Case Report)

by

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The histogenesis of many ovarian tumours remains an unsettled problem. This is especially true of the group of the functioning tumours, both feminizing and masculinizing. With reference to the latter subgroup a further complication is the "nomenclature muddle" (Willis, R. A., 1960) due to hasty introduction of new names based upon postulated histogenesis. Without connoting that they are distinct species, the ovarian tumours accompanied by masculinization may be listed as follows: arrhenoblastoma, gynandroblastoma, virilising hilus cell tumour, masculinizing luteoma and adrenal tumour of the ovary.

Adrenal tumour of the ovary has paraded in the literature under several names: adrenal-like tumour, adrenal rest tumour, adrenocorticoid tumour, inter-renaloma, hypernephroma or hypernephroid tumour of the ovary, virilising lipoid cell tumour, luteoma (luteinoma), androblastoma diffusum, Leydig cell tumour and masculinovoblastoma (Hertig and Gore, 1961; Gadd et al., 1960); of these, adrenal tumour of the ovary and masculinovoblastoma are the most popular. Up to 1960 only 40 authentic cases of this tumour have been described in the world literature (Gadd et al., 1960). We have not been able to discover a single report in the Indian literature. Gadd et al., while describing their case, have succintly reviewed the various aspects of this tumour from which are gleaned the following important features: (1) The tumour can occur at any age from menarche to menopause, or later. (2) There is a single instance of a bilateral tumour. (3) Four to five cases of the malignant variety of the tumour are on record. (4) The clinical features consist of virilism and Cushing's syndrome. (5) Though it cannot be said to be definitely established, the consensus of

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opinion is that it is derived from adrenal cell rests (ectopic adrenal) in the ovary. (6) Hormonal studies show increased urinary excretion of ketosteroids; a rise in the alpha fraction is characteristic, though this has also been observed in a single case of the hilus cell tumour of the ovary. (7) In the present state of knowledge the diagnosis of this tumour can only be histological.

Case Report

Mrs. H.K., Hindu aged 33 years, was admitted to the gynaecological wards of the Sassoon Hospitals, Poona, on 12-3-1962, with the following history. Menarche was at the age of 15; the periods were regular but slightly painful. There was a single normal delivery 5 years ago. Three years ago, the menstruation had ceased abruptly. At about the same time she noticed that her voice was becoming thicker and hoarse. A little later hair had developed on the upper lip, chin, chest and legs; simultaneously hair from the scalp had started falling off. About 3 weeks ago she had an attack of severe abdominal pain accompanied by vomiting; along with this she noticed a small lump in the lower abdomen on the right side. The pain had persisted as a dull ache with exacerbations, which also brought vomiting, and the lump had steadily become bigger. Since the onset of pain she had also experienced difficulty in passing urine and faeces.

Examination showed a person with average build. She was poorly nourished and looked ill. The nails and conjunctivae were pale. Temperature, 97; pulse, 90; respirations, 22; B.P. 110/60. A growth of thick hair was seen on the upper lip, chin, chest and legs. The scalp showed patches of alopecia. The breasts were atrophied (Fig. 1). The voice was hoarse and masculine. There was no enlargement of the superficial lymph nodes. The cardio-vascular and respiratory systems were normal. Abdominal examination showed a lump in the suprapubic region, more on the right side; it was arising from the pelvis and had well-defined upper, medial and lateral borders. It was somewhat fixed to the deeper structures, firm and slightly tender. Vaginal examination located the lump in the right fornix; the uterus was retroverted and retroflexed and was separate from the lump. The cervix was healthy. The clitoris was enlarged to 4 cm. in length (Fig. 2) and there was male distribution of hair over the pubis.

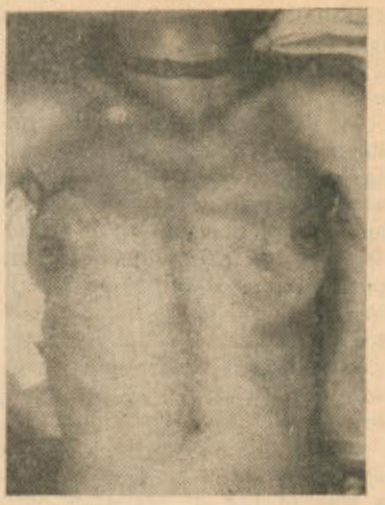

Fig. 1
Atrophy of the breasts and male distribution of hair are apparent.

Fig. 2
Hypertrophy of the clitoris is well seen.

The relevant data from the laboratory investigations were: Hb., 10 g.; blood urea, 22 mg.; fasting blood sugar, 74 mg.; and urinary 17-ketosteroids 16 mg./24 hours.

The clinical diagnos s was arrhenoblastoma. At the exploratory laparotomy done five days after admission (17-3-1962) the findings were: a pedunculated growth arising from the right ovary with twisting of the pedicle and fine adhesions between the growth and the fallopian tube, intestines and peritoneum; slight enlargement of the left ovary; a uterus of normal size; and absence of ascites. The mass in the right ovary was excised and a biopsy was taken from the left ovary. The convalescence was uneventful. After the histological diagnosis was made a curettage was done—11 days after the operation; the endometrium was in the proliferative phase but not hyperplastic. On 15-4-1962 she had her first post-operation period lasting for 4 days. The second period came on 15-5-1962 and lasted for 3 days. By this time she had developed some hair on the scalp on the patches of baldness, the voice had become less hoarse and the clitoris was significantly reduced in size. She did not come for a check-up after discharge from the hospital.

Specimen (Fig. 3). It was a large, ovoid

Fig. 3
Gross appearance of the cut surface of the tumour.

mass, measuring 15 x 10 x 7 cm. and weighing 500 g. Beneath the fibrous capsule were a few, yellow nodular projections. The cut surface showed lobules of yellow, homogeneous tissue separated by pale, greyish white bands of fibrous tissue. Haemorrhage had occurred in some lobules, while necrosis had caused cavitation in others. Normal ovarian tissue could not be recognised anywhere.

Histopathology. The tumour consisted of large, polyhedral cells with extremely vacuolated or clear cytoplasm and small, pyknotic nuclei (Fig. 4). The cells were

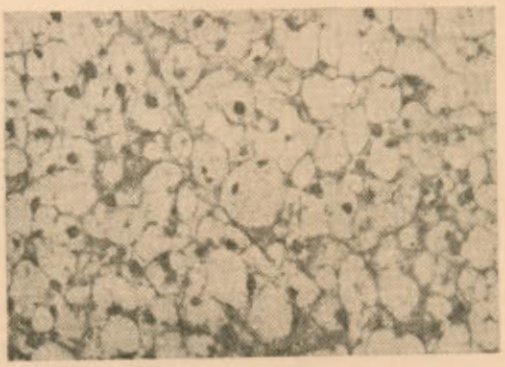

Fig. 4
H. and E. x 400. This illustrates the dominant pattern of the tumour cells — cells with foamy or clear cytoplasm and pyknotic nuclei.

arranged in small and large solid groups with scanty stroma, containing sinusoidal blood vessels, in between. Here the pattern and cytology resembled that of the zona fasciculata of the adrenal. Some cells showed large solitary vacuoles pushing the nucleus peripherally; fusion of such cells had formed large cysts (Fig. 5). These cysts showed fat in frozen sections. A tubular or papillary arrangement was not seen in any of the several sections studied. Foci of cholesterol deposition with foreign body giant cells round them were seen in some of the sections. An intact, fibrous capsule was present all round. There was no anaplasia in the tumour tissue. Frozen sections stained by Sudan III disclosed abundant sudanophilic material in the tumour cells and some even in the giant cells. Sections stained by Best muicarmine technique did not show mucin in the tumour cells. PAS stain disclosed an occasional

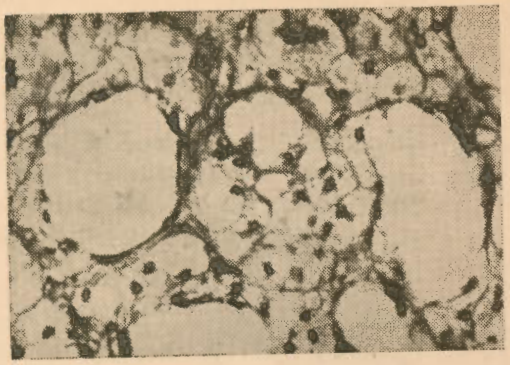

Fig. 5
H. and E. x 600. Large spaces formed by fusion of cells rich in fat are seen.

cell with p nk cytoplasm, but in the sections stained by haematoxylin and eosin such cells did not have cytoplasmic vacuolation.

Biopsy of the left ovary failed to show ovar an tissue. The findings were necrotic tissue, haemorrhages and cholesterol deposits.

Comments

As mentioned earlier the diagnosis of an adrenal tumour of the ovary is mainly histological. Luckily the tumour is always a functioning one (Hertig and Gore, 1961) so that, on the basis of the clinical findings alone, tumours like a clear cell ovarian carcinoma, mesometanephric rest tumour with a clear cell pattern (Hertig and Gore), metastasis of a renal cell carcinoma and metastasis of a clear cell carcinoma of the uterine body (Gadd et al., 1960) can be excluded, and masculinizing ovarian tumours need only be considered in the differential diagnosis. The tumour we are describing has a stereotyped pattern and cellular morphology and this rules out the possibility of an arrhenoblastoma (Willis, 1962) and the so-called gynandroblastoma. The hilar (Leydig) cell tumour is usually a small nodule — less than 5 cm. — with a characteristic morphology: oval or polygonal cells with eosinophilic cytoplasm containing lipoid, lipochrome and Reinke albuminoid crystalloids (Hertig and Gore; Novak and Woodruff). There is no remote resemblance to this structure in our case. For a variety of reasons differentiation between a masculinizing luteoma and an adrenal tumour is most difficult. Novak and Woodruff remark that most of the masculinizing luteomas are really of adrenal origin but concede the possibility that a luteoma, arising from luteinization of a granulosa cell tumour, may function as a masculinizing tumour. Hertig and Gore contend that virilizing luteomas "be regarded as a variety of arrhenoblastomas, because differences between the luteinised theca interna and interstitial cells of the testis are not critical in the absence of Reinke crystalloids". On the other hand, Willis is most critical even of the use of the term "arrhenoblastoma" ("a histogenetically meaningless word"); he believes that the so-called arrhenoblastoma includes three different tumours: (1) granulosa and theca cell tumours with androgenic effects; (2) hilus cell tumours; and (3) virilising adrenal tumours arising close to the ovary, a remote possibility. According to this author, though a heterotopic adrenal cortex in the mesovarium can rarely become neoplastic, almost all of the "adrenal" tumours are either luteinized ovarian tumours or lipoid rich hilar cell tumours.

Summary

A case of adrenal tumour (Masculinovoblastoma) of the ovary is reported.

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