ADRENAL TUMOUR OF OVARY

(Case Report)

by

SHASHI MALHOTRA, M.S.

The histogenesis and function of the cells associated with ovarian tumours possessing an endocrine potential are largely ill-understood. Remnants of the embryological vestiges are supposed to manifest themselves as some of the functioning tumours, both feminizing and masculinizing. Commenting on the unsatisfactory state of our knowledge in this respect, Novak and Woodruff (1962) feel inclined to state "To say that confusion reigns is a masterpiece of understatement".

The virilising tumours of the ovary are now being recognised more often, and hence are no more rare, although the frequency is much less as compared to the feminizing tumours. Different types of masculinizing tumours have been recognized in the form of "arrhenoblastoma", 'gynandroblastoma', 'virilizing hilus cell tumour', 'masculinizing luteoma' and 'adrenal tumour' of the ovary. The tumours of the adrenal type are very rare indeed, only 40 authentic cases having been described in the world literature till 1960 (Gadd *et al.* 1960). Aberrant adrenal tissue is quite frequently seen along the ovarian and spermatic vessels, although less commonly in the ovary itself. Large tumours arising from this tissue can be capable of producing virilising effects. Since the tumour cells show a close resemblance to corpus luteal cells, it was formerly thought to be a luteoma or luteinised granulosa cell tumour. Its origin from the adrenal cortical cells seems to be more feasible (Masani, 1960). The adrenal tumours have been given different names from time to time e.g. 'adrenal rest tumour', 'adrenal corticoid tumour', 'hypernephroma', 'hypernephroid tumour', 'virilising lipoid cell tumour,' 'luteoma,' 'adrenal like tumour' or 'masculinovoblastoma.'

The adrenal tumours are usually unilateral, solid in consistency and have a smooth surface which may get adherent to the adjacent structures in late stages. The tumour produces both male and female hormones (Masani, 1960), the former being much in excess of the latter. The virilizing effects of the tumour are perhaps due to over-production of the male hormone, and the clinical characteristics are indistinguishable from arrhenoblastoma (Novak and Woodruff, 1962). The tumour can occur at any age and 4-5 cases of malignant variety have also been reported (Gadd *et al.* 1960). An increase in the urinary excretion of ketosteroids with a rise in the alpha fraction is usually considered charac-

**Lecturer in Gynec. & Obst. S.M.S. Medical College, Jaipur.*

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teristic, although this has also been observed in a single case of hilus-cell tumour of the ovary (Gadd *et al*, 1960). In the present state of our knowledge, the histopathological appearance of the tumour is the key to diagnosis. The sections show large, polyhedral cells with extremely vacuolated or clear cytoplasm, and small pyknotic nuclei arranged in small and large solid groups with scanty stroma containing sinusoidal blood vessels in between. Anaplastic change may be seen in some parts of the tumour. Presence of fat may be demonstrated in the cells in frozen sections. The uterus is usually enlarged with a thickned and hyperplastic endometrium. The patient presents a history of masculinisation with cessation of menstruation. An exploratory laparotomy with removal of the affected ovary is indicated. The prognosis is good after the operation with an early recession of masculinizing signs and return of menstrual function.

A typical case of adrenal tumour is being presented with the clinical history, laboratory findings, histopathological data and operative results.

Case Report

A Hindu, aged 35 years, was admitted to the gynaecological wards of the Zenana Hospital, Jaipur, on 7th January, 1965. She had menarche at the age of 14 years and subsequently the periods were regular, though slightly painful. She had six full-term normal deliveries, the last one 7 years ago. The menstruation ceased abruptly 2 years ago and she started feeling that her voice was becoming thicker and hoarse. A little later hair started appearing on the upper lip, chin, chest and legs. She also developed acne over the face (Fig. I).

General examination showed a person with average build and height, conjuctivae normal, pulse 80 minute, temp. 98.4, °F B.P. 120/80, breasts not atrophied, voice hoarse and masculine and no enlargement of the superficial lymph glands Cardiovascular and respiratory systems were normal. A growth of thick hair was seen on the upper lip and chin, chest and legs. Abdominal examination revealed nothing abnormal Bimanual pelvic examination showed an enlarged clitoris (Fig. 2) normal consis-

Fig. 1
Masculinizing changes associated with "Adrenal tumour of the Ovary".

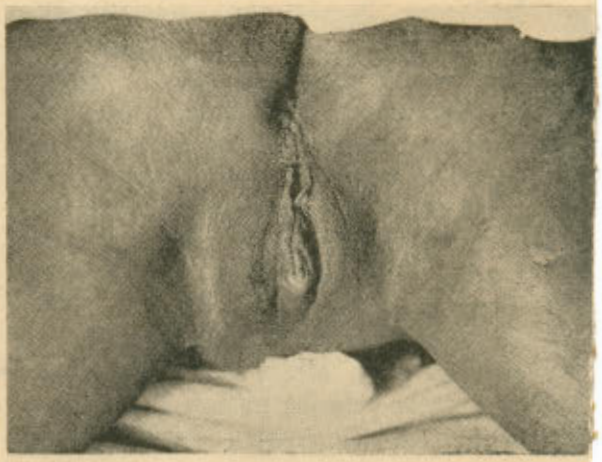

Fig. 2
Hypertrophy of clitoris.

tency of the cervix and an anteverted, anteflexed uterus, which was of parous size, mobile, and firm. The left fornix was clear. In the right fornix a freely mobile mass, — 3″ x 4″ in size was felt. Per speculum, the cervix was found eroded and congested.

Laboratory investigations showed Hb. 9 gms%, blood urea, 40 mg%, fasting blood sugar, 98 mg.%, serum potassium 8 m.eq. serum sodium 125 m.eq. E.S.R. 15 mm/hr. urinary 17 ketosteroids 5.2 mg/24 hours. Skiagram of the skull showed a normal sized fossa. Endometrial biopsy taken on 30-1-65 showed a thickened and hyperplastic endometrium in the proliferative phase.

A cilinical diagnosis of virilizing tumour of the ovary was made. Exploratory laparotomy done on 9-2-65, showed an enlarged right ovary 4″ x 3″ in size having a smooth external surface with varying consistency. The left ovary was normal in size and appearnce except for a few visible folicles. There were no adhesions and there was no free fluid. The uterus was firm, congested and of parous size. Both adrenal glands were normal in szie. A wedge biopsy was taken from the left ovary and right-sided ovariotomy was done. Convalescence was uneventful. The patient had her first period on 8-3-65 exactly one month after the operation. Since then, she has been having regular periods of average duration of 4-5 days at an interval of 20-25 days with moderate flow. The acne disappeared, hirsutism showed marked regression, (Fig. 3) and the clitoris diminished considerably in size.

Fig. 3
Disappearance of facial hair and acne 3 months after removal of the tumour.

Specimen

(Fig. 4) the mass removed at operation is ovoid is shape and 4″ x 3″ in size. Beneath the fibrous capsule a few yellow nodular projections are seen. The cut surface (Fig. 5) shows lobules of yellow, homogeneous tissue separated by pale greyish white bands of fibrous tissue. Haemorrhagic or necrosed areas not present. Normal ovarian tissue cannot be recognised.

Histopathology (Fig. 6)

The tumour consists of large polyhedral cells with extremely vacuolated or clear cytoplasm and small pyknotic nuclei. The cells are arranged in small and large solid groups with scanty stroma, the pattern and cytology resembling that of the zona fasciculata of the adrenal. Some of the large cells show fat in frozen section. Frozen sections stained by Sudan III disclosed abundant sudanophilic material in the tumour cells. P.A.S. staining disclosed an occasional cell with pink cytoplasm.

Comments

The adrenal tumour of the ovary is always functional and the consequent hormonal effects are a great help in diagnosis. (Hertig and Gore, 1961). It produces virilism by overproduction of the male hormone. The female characteristics start regressing leading to amenorrhea, atrophy of the vulva and breasts. The male characters are evidenced by enlargement of the clitoris, deepening

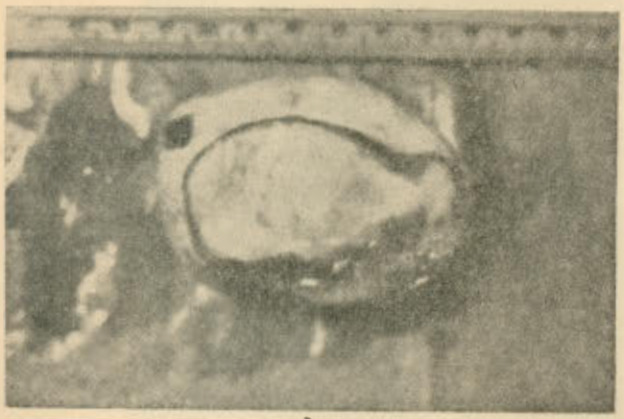

Fig. 4
Gross appearance of the tumour.

Fig. 5
Gross appearance of the cut surface of the tumour.

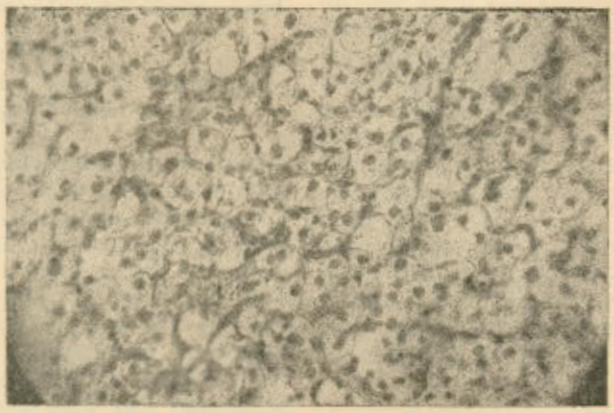

Fig. 6
Microphotograph showing dominant pattern of the tumour cells. Hand E X 430.

of the voice, male type of distribution of suprapubic hair, growth of hair in other parts of the body and hypertrichosis. Thus on the basis of the clinical findings alone, tumours like a clear cell ovarian carcinoma, a clear cell mesonephroma, rest tumour with a clear cell pattern, and metastasis of a clear cell carcinoma of the uterus can be excluded (Hertig and Gore, 1961; Good *et al*, 1960).

The differential diagnosis between the various virilizing ovarian tumours rests largely on the histogical picture. The typical cellular pattern and the cell morphology helps to exclude the possibility of an arrhenoblastoma (Wills, 1962), and

the so-called gynandroblastoma. The hilar (Leydig) cell tumours possess a typical characteristic morphology and show the presence of Reinke albuminoid crystalloids. However, hilus cells with Reinke crystals have recently been described in an adrenal tumour (Scully and Cohen, 1961).

Summary

A case of adrenal tumour of the ovary is reported, which showed complete recession of signs and symptoms after removal of the affected ovary.

Addendum — The patient reported on is now pregnant and is in the sixth month.

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