Female Adnexal Tumor of Probable Wolffian Origin†

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Abstract—A peculiar right adnexal tumor was removed from a 53-year old multiparous woman. The tumor had distinctive diffuse, tubular and trabecular patterns. Ultrastructural study suggested derivation from wolffian duct.

Key Words: Broad ligament, Wolffian tumor, Ultrastructural study

INTRODUCTION

In 1973 Karimnejad and Scully reported 9 cases of a distinctive female adnexal tumor. The tumor was characterized microscopically by epithelial cells growing in diffuse, trabecular and tubular patterns. They proposed the term female adnexal tumor of probable wolffian origin for the tumor. We performed an ultrastructural study to identify the features suggesting its origin.

CASE REPORT

The patient was a 53-year old multiparous woman (4-0-3-1) who was admitted to the hospital for the evaluation of intermittent right flank pain for 6 months. She noticed palpable mass in the lower abdomen 1 week prior to the admission.

She was in one year postmenopausal. Her past history was noncontributory. A total abdominal hysterectomy and bilateral salpingo-oophorectomy were performed under the clinical impression of leiomyoma of the uterus and pelvic mass.

In the right broad ligament a well encapsulated tumor mass was noted. The tumor measured 15×11×9 cm. The outer surface was smooth and glistening; focally it was bosselated. The cut surface was solid and cystic. The solid tissue was pale yellow brown lobulated and firm. The cyst measured 9 cm in diameter and contained yellow brown turbid fluid. The right ovary and salpinx were noted on the outer surface of the tumor mass. The uterus and left adnexa were unremarkable.

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MATERIALS AND METHODS

For light microscopy the specimen was fixed in 10% neutral formalin solution. PAS and reticulum stains were performed besides routine hematoxylin-eosin stain. Samples of tissue were removed from formalin, fixed in glutaraldehyde, postfixed in 2% buffered osmium tetroxide. The samples were embedded in epon after dehydration in graded alcohols and propylene oxide. Ultrathin sections were cut on LKB ultramicrotome and stained with lead citrate and uranyl acetate, and examined in a Hitachi 500 electron microscope.

RESULTS

Light microscopy: The tumor had various patterns of growth. The predominant patterns were the tubular and diffuse or solid patterns. In the diffuse areas the tumor cells were closely packed into a massive non-lobular architecture with scanty stroma. Reticulum staining showed fibrillar material among individual tumor cells or elongated groups of the tumor cells. Periodic acid–schiff (PAS) stain revealed distinct basement membrane around the groups of the tumor cells. In areas diffuse or solid area contained oval vacuoles, some of which coalesced. The tubular pattern consisted of closely packed, winding, branching and anastomosing tubular structures which were slender and solid, and had distinct basement membrane. The solid tubes had peripheral nuclei and central cytoplasm. Scattered hollow tubules were noted among the solid tubules. The tumor cells in the diffuse area were oval or elongated with small amount of cytoplasm and were spindle-shaped in areas, simu-
lating mesenchymal tumor. About one third of the
tumor mass consisted of true-tubular pattern with
closely packed hollow tubules varying in size and
shape presenting sieve-like appearance. The
tubules were lined by single or stratified epithelial
cells with clear or faintly eosinophilic cytoplasm.
Some of the tubules were cystically dilated with
flattened epithelial cells; some tubules contained
eosinophilic secretion.

The nuclei of the tumor cells were oval, elongated,
or round. The chromatin granules were fine,
somewhat uneven and the nucleoli were indistinct.
Mitosis was not encountered. The stroma of the
tumor consisted of bands of varying thickness of
hyalinized fibrous tissue, which encircled nests or
cords of tumor cells. In areas the stroma consisted
of delicate reticulum, especially in diffuse areas.
Focal calcification was noted in the hyalinized fib-
rous stroma around blood vessels. Ovary was sepa-
rated from the tumor mass by loose vascularized
fibrous tissue; salpinx was demarcated from the
tumor mass by the thick fibrous capsule.

**Electron microscopy:** The tumor cells formed
tubular structures with central lumen. The cell bor-
ders were straight with focal interdigitations.
Around the cell nests multilayered basement mem-
branes were noted. Tight junctions were noted be-
tween the tumor cells. Microvilli were noted on the
surface of the nuclei were indented; the chromat-
in was marginalized along the nuclear mem-
brane and occasional nucleoli were noted. The
cytoplasm contained abundant endoplasmic reticu-
ulum, which was markedly dilated and predominant-
ly rough. Golgi apparatus was infrequently seen.
Mitochondria were moderate in numbers. Small
numbers of lysosomes were noted. Polyribosomes
were moderate in amount and free in the cyto-
plasm. Secretory granules were rarely encountered.
Microfilaments were abundant and dispersed
throughout the cytoplasm and formed curvilinear
entangled aggregation in area. A rare lipid droplets
were encountered in some cells but glycogen was
not identified.

**DISCUSSION**

The distinctive light microscopic findings of this
adnexal tumor include diffuse, tubular and trabecular patterns of ovoid or elongated tumor
cells with clear cytoplasm. In addition to the tubule
formation, prominent peritubular basement mem-
brane is noted. But Leydig cells or Call–Exner
bodies are absent. These microscopic findings are
compatible with those described in the previous
reports (Kariminejad and Scully 1973; Sivalthondan
et al. 1979). Recently Young and Scully (1983)
described 11 tumors with a similar histologic
appearance that arose in the ovary. The peculiar
location of tumor in broad ligament and character-
istic histologic appearance suggest its origin from
wolfian remnants. The prominent tubule forma-
tions and basement membrane are the features of
female adnexal tumors of wolfian origin (Karimine-
jad and Scully 1973; Sivalthondan et al. 1979),
and their ovarian counterparts (Young and Scully
1983) and wolfian remnants (Gardner et al. 1948;
Gardner et al. 1957; Lamb et al. 1960; McGee
1962). The diffuse areas may suggest the diagnosis
of granulosa cell tumors. Absence of characteristic
nuclear morphology including pale angular nuclei
with grooves and other patterns seen in the wolfian
tumor are incompatible with that diagnosis. The
luminal pattern resembles well-differentiated en-
dometrioid adenocarcinoma, but sieve-like area
and other patterns are unusual for endometrioid
adenocarcinoma. Dilated tubules with flattened
hobnail like cells and epithelial cells with clear
cytoplasm can simulate clear cell carcinoma; ab-

cence of solid aggregates of clear cells, papillary

areas, well developed hobnail cells, and intratumor-

an mucin or vacuoles containing “targe tolloid” eosi-

nophilic material is not compatible with clear cell
carcinoma. The tubules of the adnexal tumor may
bear a resemblance to Sertoli–Leydig cell tumor.
The Leydig cells are commonly present in Serto-
li–Leydig cell tumor (Scully 1980), but are uni-
formly absent from the wolfian tumor. Extragonad-
al location has been encountered in Serto-
li–Leydig cell tumor. Virilization which is one of the
characteristic features of Sertoli–Leydig cell tumor
(Scully 1980) has never been reported in wolfian
tumor.

The electron microscopic findings observed in
this case and previous report (Demopoulos et al.
1980) are not specific for either Müllerian or wolf-

fian structure. In our case cilia were absent. The
secretory granules and golgi apparatus were fre-
frequently seen. Glycogen was not apparent. Char-

cot–Böttcher crystal seen in Sertoli cell or crystal-
lloid of Reinke in Leydig cells was not identified.
These findings are compatible with those seen in
wolfian duct remnants (Riva 1967; Bransil et
al. 1973; Demopoulos et al. 1980) rather than
those seen in Müllerian epithelia (Thrasher and
Richart 1972).
Although previous reports (Kariminejad and Scully 1973; Sivathondan et al. 1979) indicated benign clinical course, that of Taxy and Battilora (1976) suggested low grade malignant potential. One of the 9 cases of ovarian counterpart also showed low grade malignant potential (Young and Scully 1983).

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REFERENCES


= 국문초록 =

중장관起源 子宮附屬器 腫瘍

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6개월간 간헐성 우측 열구리동통 및 압원 1주일 전에 축지한 하복부 흉증을 주로로 인한

중장관에서 우측 子宮附屬器에 15×11×9cm 크기의 寒腫이 수술후 발견되었다. 중장의 변

화부위에 寒腫 및 寒증이 부착되어 있었다. 중장의 設面은 深黃褐色 充実性 分葉状 組織으로 중

장부위 환색의 혼탁으로 중앙된 냉정성병소가 보였다.

寒性경직으로 중장세포가 略円形, 内腔이 없는 充実性 細胞은 寒腫를 가진 細胞이로

중장이 있는 양상이 보였고 寒腫 혹은 중장세포단 주위로 잘 발달된 基底膜이 관찰되었다.

寒性寒象으로 寒細胞, 중장의 順粒性가세포절막, 극소수의 분비조직이 관찰되었으나

내로병의 寒細胞이 없었다. 이는 이 중장이 寒관에서 가져들었을 가능성을 시사한다고

생각되었다.
LEGENDS FOR FIGURES

Fig. 1. Gross appearance of the tumor. Note lobulated tumor tissue with cystic degeneration.

Fig. 2. Diffuse pattern with focal tubule formation. H&E, X100.

Fig. 3. Closely packed solid tubules separated by delicate basement membrane. 3A: H&E, X200 and 3B: reticulum X100.

Fig. 4. Tubular pattern, Note both solid and hollow tubules. H&E, X100.

Fig. 5. True-tubular pattern. H&E, X100.

Fig. 6. Tubular pattern presenting sieve-like appearance. H&E, X100.

Fig. 7. Electron micrograph of neoplastic cells with thick basement membrane. Uranium acetate and lead citrate. X11, 200.

Fig. 8. Tumor cells with lysosome and lipid droplets. Many microfilaments are apparent. Uranium acetate and lead citrate. X14,000.

Fig. 9. Microvilli along the luminal border. Uranium acetate and lead citrate. X47,600.

Fig. 10. Dilated rough endoplasmic reticulum and mitochondria. Uranium acetate and lead citrate. X28,000

Fig. 11. Microfilaments forming focal curvilinear aggregation. Uranium acetate and lead citrate. X84,000.