The Seoul Journal of Medicine
Vol. 26, No. 1: 15-21, March 1985

Female Adnexal Tumor of Probable Wolffian Origin†

Geung Hwan Ahn and Hyo Sook Park*

Department of Pathology, College of Medicine, Seoul National University and Department of Pathology, National Medical Center*

= 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 Kariminejad 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.

†Presented at the annual meeting of the Korean Cancer Research Association, Seoul, Korea, September 21, 1981.

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 coallesced. 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 tubules 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 fibrous stroma around blood vessels. Ovary was separated 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 borders were straight with focal interdigitations. Around the cell nests multilayered basement membranes were noted. Tight junctions were noted between the tumor cells. Microvilli were noted on the cell surface. The nuclei were indented; the chromatin was marginated along the nuclear membrane and occasional nucleoli were noted. The cytoplasm contained abundant endoplasmic reticulum, which was markedly dilated and predominantly 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 cytoplams. 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 membrane 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; Sivathondan 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 characteristic histologic appearance suggest its origin from wolffian remnants. The prominent tubule formations and basement membrane are the features of female adnexal tumors of wolffian origin(Kariminejad and Scully 1973; Sivathondan et al. 1979). and their ovarian counterparts(Young and Scully 1983) and wolffian remnants (Gardner et al. 1948; Gardner et al. 1957; Lamb et a:. 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 wolffian tumor are incompatible with that diagnosis. The tubular pattern resembles well-differentiated endometrioid 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; absence of solid aggregates of clear cells, papillary areas, well developed hobnail cells, and intraluminal mucin or vacuoles containing "targe toid" eosinophilic 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 Sertoli-Leydig cell tumor (Scully 1980), but are uniformly absent from the wolffian tumor. Extragonadal location has hot been encountered in Sertoli-Leydig cell tumor. Virilization which is one of the characteristic features of Sertoli-Leydig cell tumor (Scully 1980) has never been reported in wolffian tumor.

The electron microscopic findings observed in this case and previous report (Dernopoulos et al. 1980) are not specific for either müllerian or wolffian structure. In our case cilia were absent. The secretory granules and golgi apparatus were infrequently seen. Glycogen was not apparent. Charcot-Böttcher crystal seen in Sertoli cell or crystalloid of Reinke in Leydig cells was not identified. These findings are compatible with those seen in wolffian duct remnants (Riva 1967; Bransilver 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 Battifora (1976) suggested low grade malignant potential. One of the 9 cases of ovarian counterpart also showed low grade malignant potential (Young and Scully 1983).

(Authors thank Miss Chung, Y.H. for her assistance in the preparation of the manuscript and Mr. Kim, S.S. for the preparation of electron microscopic pictures).

REFERENCES

Bransilver BR, Ferenczy A., Richart R. Female genital tract remnants: An ultrastructural comparison of hydatid of Morgagni and mesonephric ducts and tubules. Arch. Pathol. 1973, 96:255-261.

Demopoulos RI, Sitelman A., Flotte T, Bigelow B. Ultrastructural study of a Female Adnexal Tumor of Probable Wolffian Origin. Cancer 1980; 46:2273-2280.

Gardner GH, Greene RR, Peckham BM. Normal and cystic structures of the broad ligament. Am. J. Obstet. Gynecol. 1948, 55:915-937.

Gardner GH, Greene RR, Peckham BM. Tumors of the broad ligament. Am. J. Obstet. Gynecol. 1957, 73:536-55.

Kariminejad MH, Scully RE. Female Adnexal Tumor of Probable Wolffian Origin. Cancer 1973, 31:671-677.

Lamb EJ, Fucilla I, Greene RR. Basement membranes in the female genital tract. Am. J. Obstet. Gyncol. 1960, 79:79-85.

McGee CT, Cromer DW, Greene RR. Mesonephric carcinoma of the cervix. Differentiation from endocervical adenocarcinoma. Am. J. Obstet. Gynecol. 1962, 84:358-366.

Riva A. Fine structure of human seminal vesicle epithelium. J. Anat. 1967, 102:71-86.

Scully RE. Tumors of the Ovary and Maldeveloped Gonads. (Atlas of Tumor Pathology, 2nd series, fasc. 16) Armed Forces Institute of Pathology, Washington, D.C. 1980. pp. 190-208.

Sivathondan Y., Salm R., Hughesdon PE, Paccini JM. Female adnexal tumor of probable wolffian origin. J. Clin. Path. 1979, 32:616-624.

Taxy JB, Battifora H. Female adnexal tumor of probable wolffian origin. Evidence for a low grade malignancy. Cancer 1976, 37: 2349-2354.

Thrasher TV, Richart RM. An ultrastructural comparison of endometrial adenocarcinoma and normal endometrium. Cancer 1972, 29:1713-1723.

Young RH, Scully RE. Ovarian tumors of probable wolffian origin. A report of 11 cases. Am. J. Surg. Pathol. 1983, 7:125-135.

= 국문초록 =

中腎管起源 子宮附屬器 腫瘍

서울대학교 의과대학 병리학교실 및 국립의료원 병리과*

安亘煥 · 朴孝淑*

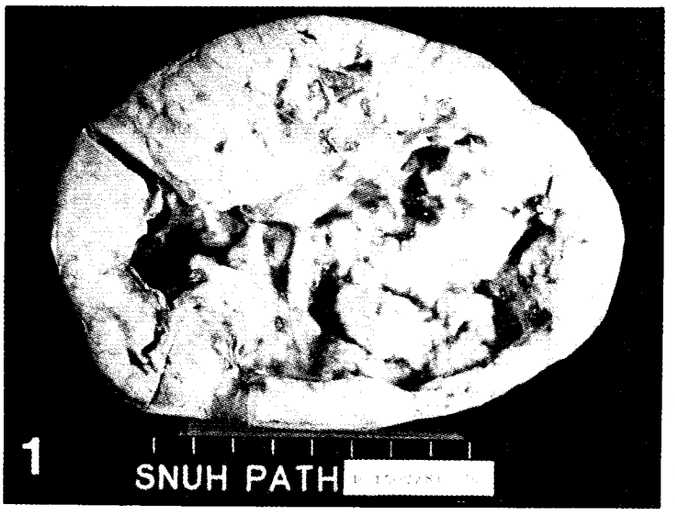
6개월간 간헐성 우측 옆구리동통 및 입원 1주일 전에 촉지된 하복부 腫塊를 主訴로 入院한 經産婦에서 우측 子宮附屬器에 15×11×9cm 크기의 腫塊가 수술결과 발견되었다. 종괴의 변연부에 卵巢 및 卵管이 부착되어 있었다. 종괴의 割面은 淡黃褐色 充實性 分葉狀 組織으로 중앙부에 황색의 혼탁액으로 충만된 낭성 퇴행성병소가 보였다.

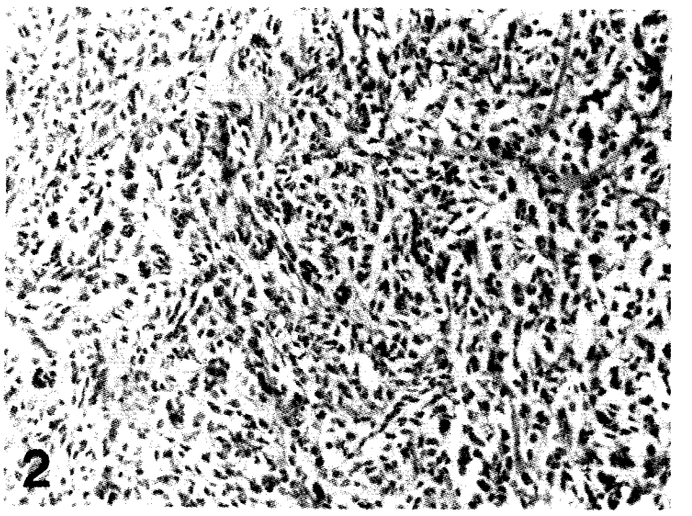
현미경적으로 종양세포가 瀰漫性, 內腔이 없는 充實性 細管 혹은 內腔을 가진 細管狀으로 증식하는 양상을 보였고 細管 혹은 종양세포집단 주위로 잘 발달된 基底膜이 관찰되었다.

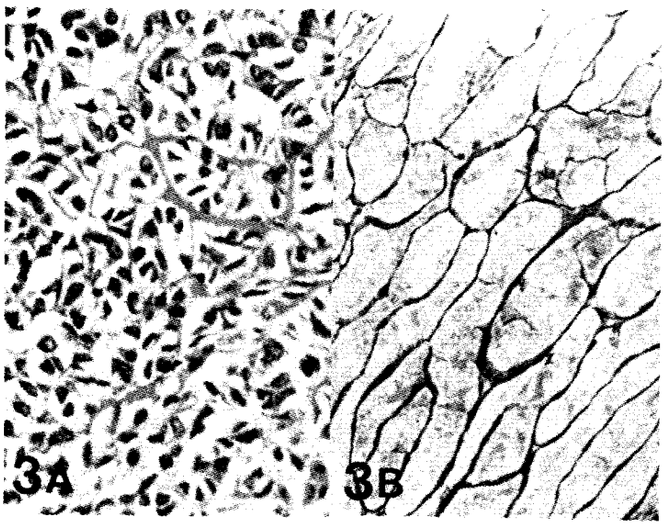
전자현미경적으로 微細융모, 중등도의 顆粒性內形質細網, 극소수의 분비과립이 관찰되었으나 그리코겐 및 纖毛는 없었다. 이는 이 종양이 中腎管에서 기원하였을 가능성을 시사한다고 생각되었다.

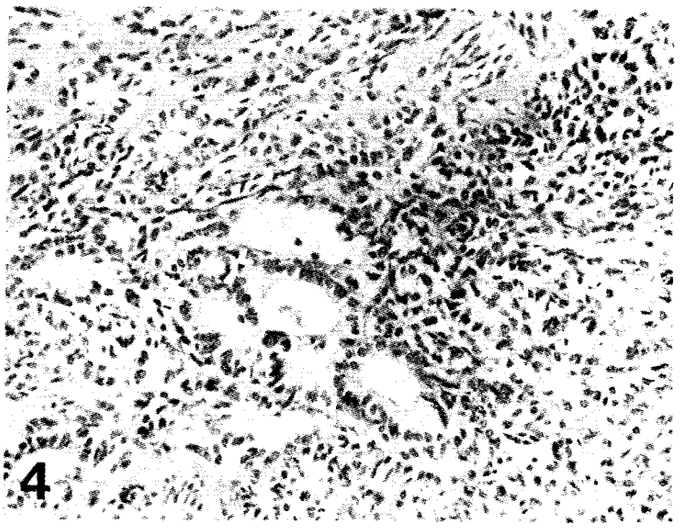
LEGENDS FOR FIGURES

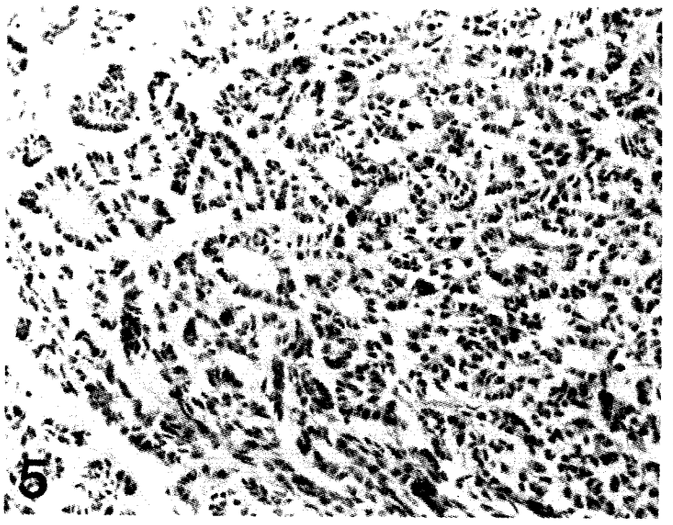
Fig. 1. Gross appearance of the tumor. Note lobulated tumor tissue with cystic degeneration.
Fig. 2. Diffuse pattern with focal tubule formation. H & E, X100.
Fig. 3. Closely packed solid tubules separated by delicate basement membrane. 3A: H&E, X200 and 3B: reticulum X100.
Fig. 4. Tubular pattern, Note 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.

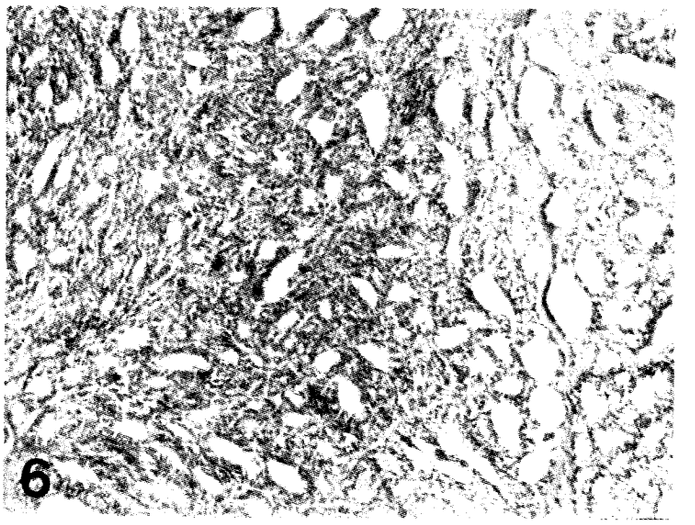

1
SNUH PATH

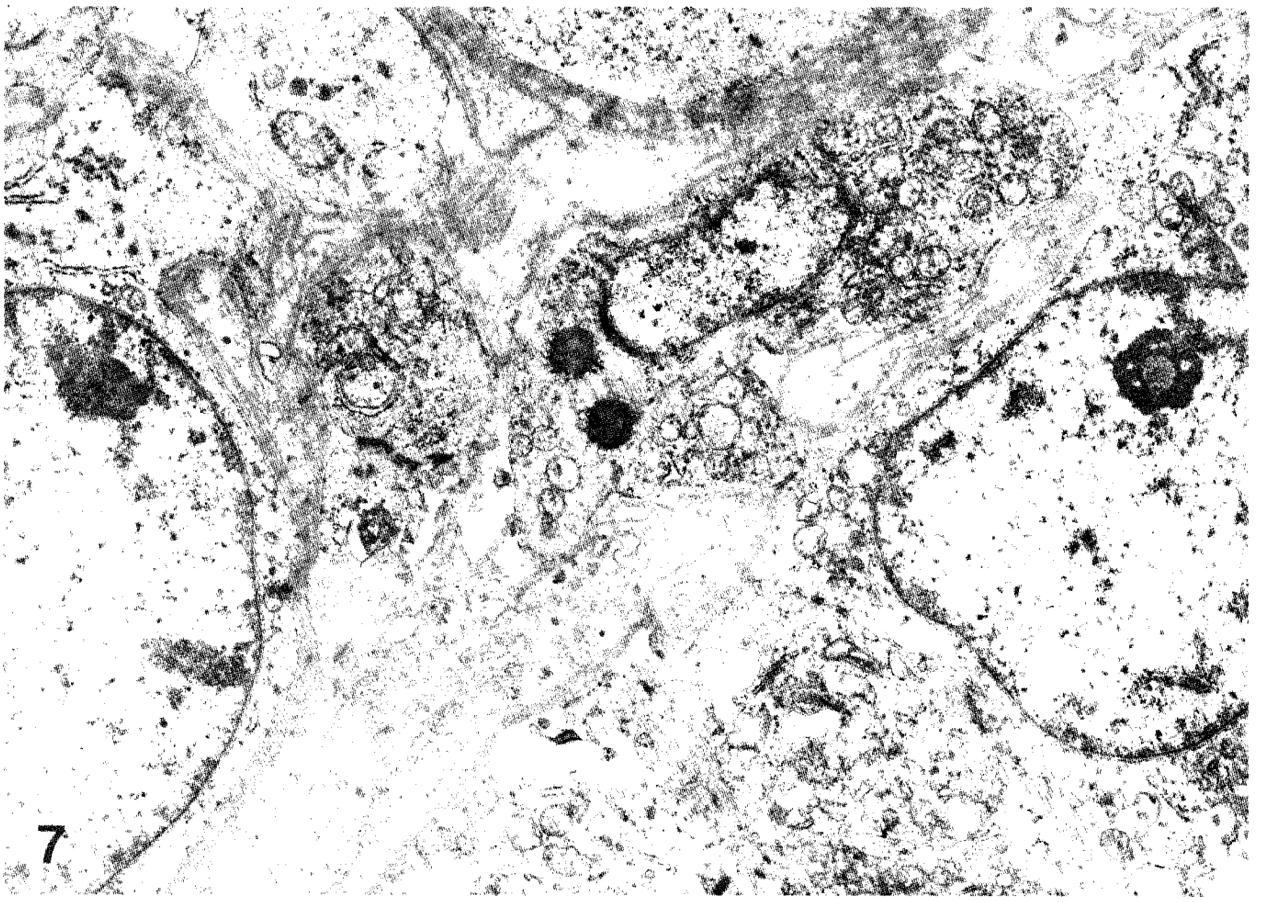

2


3A
3B

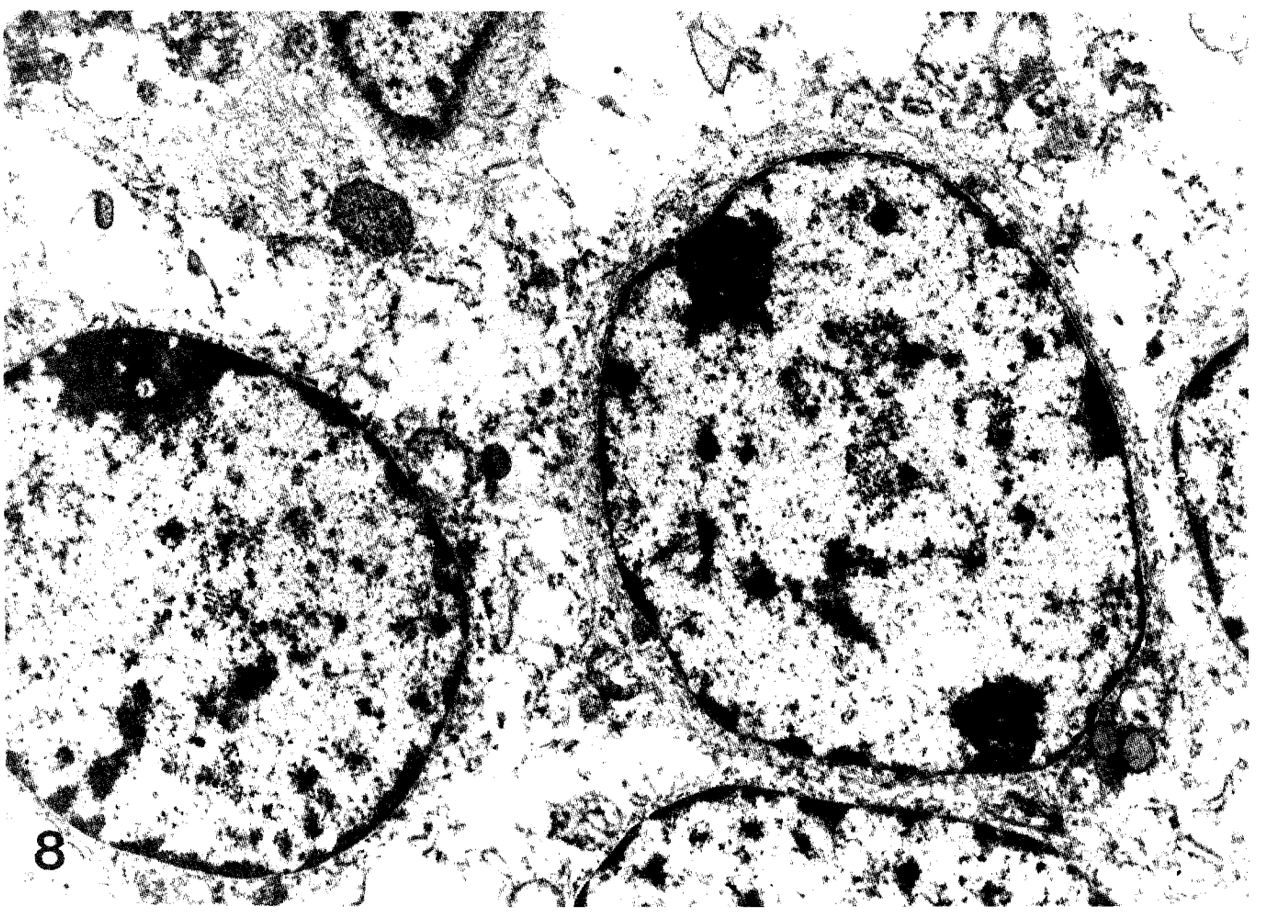

4


5


6

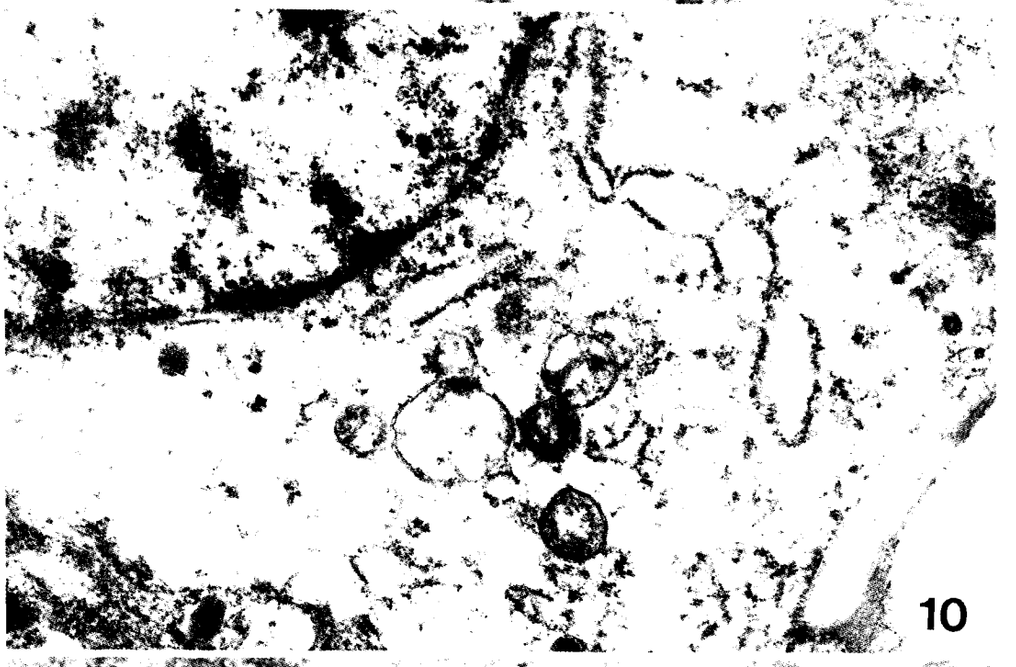


7

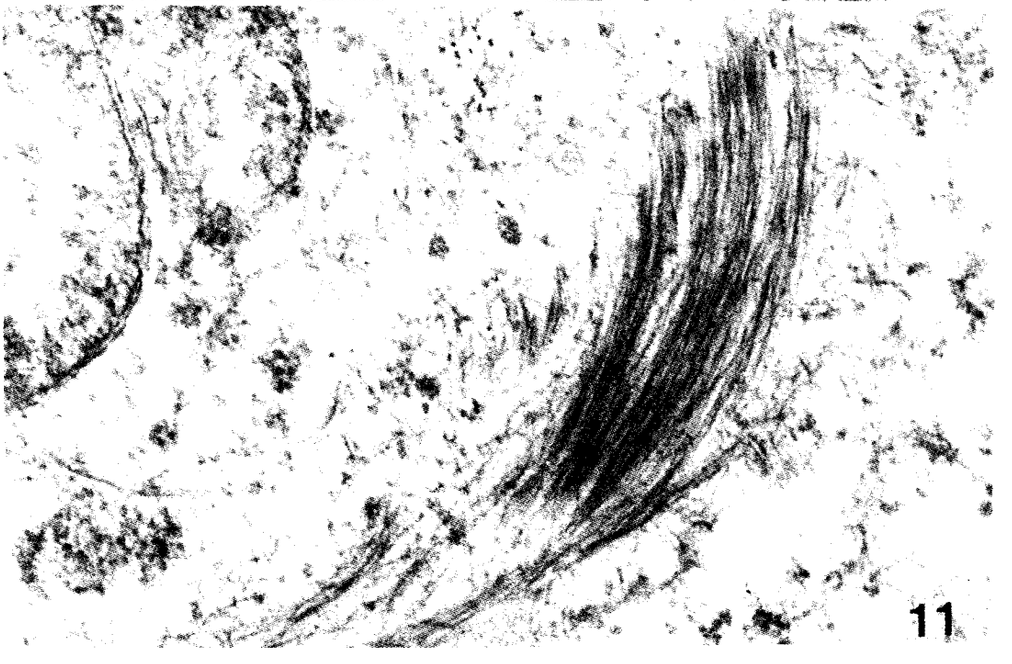


8

9



10



11