

A Case of Female Adnexal Tumor of Probable Wolffian Origin: Histologically, Tumor Cells Showed Three Different Patterns

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WAGATSUMA, S., YAEGASHI, N., NAMIKI, T., SATO, S. and YAJIMA, A. *A Case of Female Adnexal Tumor of Probable Wolffian Origin: Histologically, Tumor Cells Showed Three Different Patterns.* Tohoku J. Exp. Med., 1997, **181** (3), 371-377 — We report a 63-year-old woman with a female adnexal tumor of probable Wolffian origin, which arose within the leaves of the broad ligament, connected to the left ovary and the fallopian tube. She remains alive without evidence of disease 2 years after left salpingo-oophorectomy without subsequent adjuvant therapy. Microscopically, tumor cells were arranged in three different patterns; closely packed tubules, solid, and sieve-like. Cells had uniform round or oval nuclei and mitoses were rare. Although the majority of these tumors are benign, a few cases have shown malignant potential. Therefore, such tumors should receive careful follow-up for possible recurrence and/or metastasis. ——— adnexal tumor; Wolffian origin

Kariminejad and Scully (1973) described nine cases of a distinctive extraovarian tumor, which they designated “female adnexal tumor of probable Wolffian origin (FATPWO)”. These tumors, which arise within the leaves of the broad ligament or hang from it or the fallopian tube, are characterized microscopically by epithelial cells growing in tubular, diffuse, and sieve-like patterns. Since then, about 60 cases, including 9 malignant tumors, have been described in the literature. The distinctive and extremely rare pathological features of one additional case are presented herein.

CASE REPORT

The patient is a 63-year-old multiparous woman who demonstrated a 7 × 6 × 6-cm tumor in the pouch of Douglas on physical examination. She had entered menopause at 50 years of age and no genital bleeding had been noted since then.

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She had no noteworthy past history. On ultrasonography, the tumor had a hypoechoic and homogeneous component surrounded by a thick capsule. An abdominal computed tomography revealed monolocular tumor in the left adnexal region, of which capsule was enhanced irregularly indicating malignant potential (Fig. 1). The tumor markers, CA125, CA19-9, CA72-4, SLX, CEA, and AFP were all normal. The clinical diagnosis was left ovarian cystic tumor, it could not

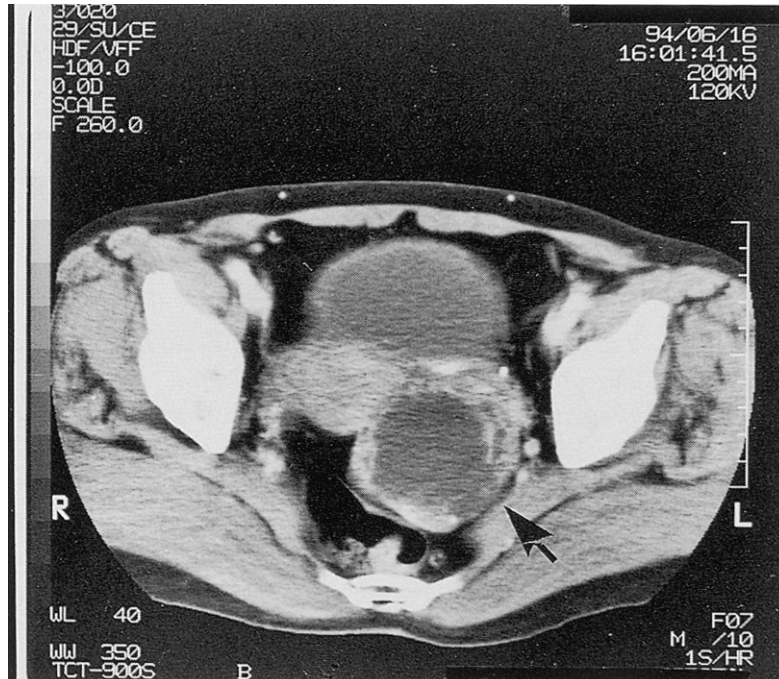


Fig. 1. A computed tomography showing a left adnexal mass (arrow) with a thick capsule enhanced irregularly.

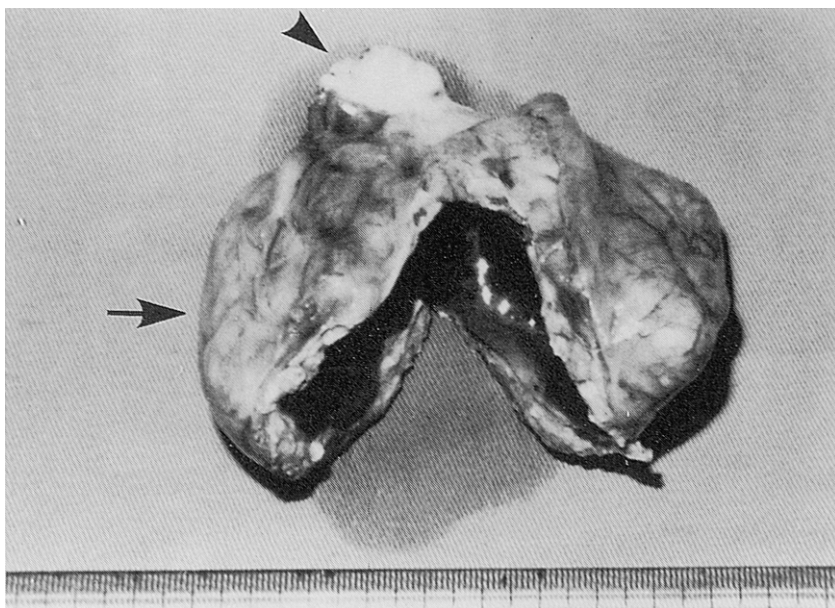


Fig. 2. Cross section of the cystic tumor (arrow) which connected to the left ovary (arrowhead).

be denied malignancy.

At surgery, the cystic tumor mass was connected to the left ovary and fallopian tube as well as the posterior broad ligament. The uterus, bilateral fallopian tubes, and ovaries were unremarkable macroscopically. There was no ascites. Left salpingo-oophorectomy with tumor resection was performed uneventfully. On gross examination the surface of the 7×6×6-cm cystic mass was intact, smooth, and glistening. On cut section, the cystic mass contained brownish and mucinous fluid, and its capsule was thick and had no papillary component as expected by ultrasonography and computed tomography (Fig. 2). Postoperatively, no adjuvant treatment was proposed. The patient remains alive

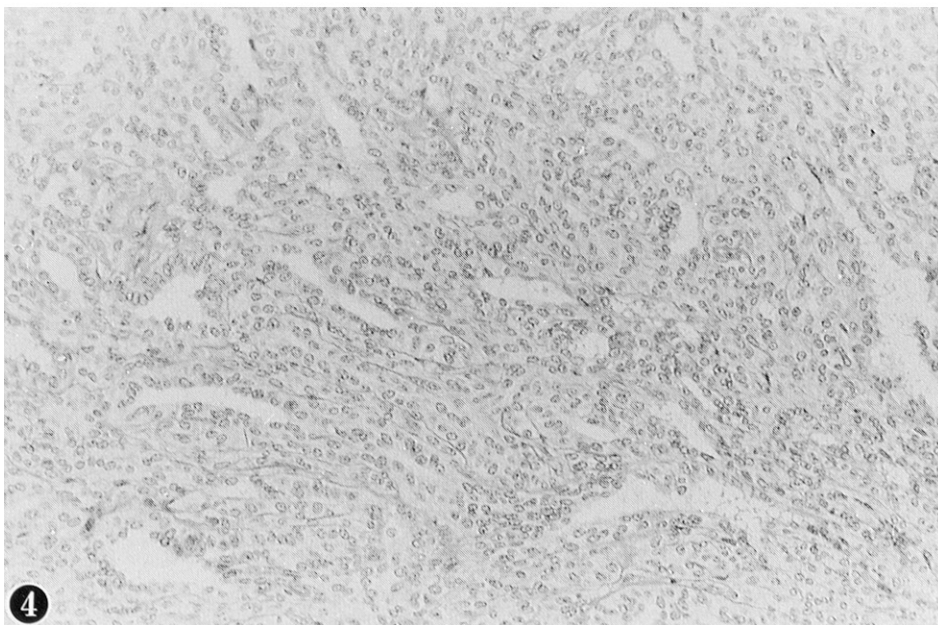
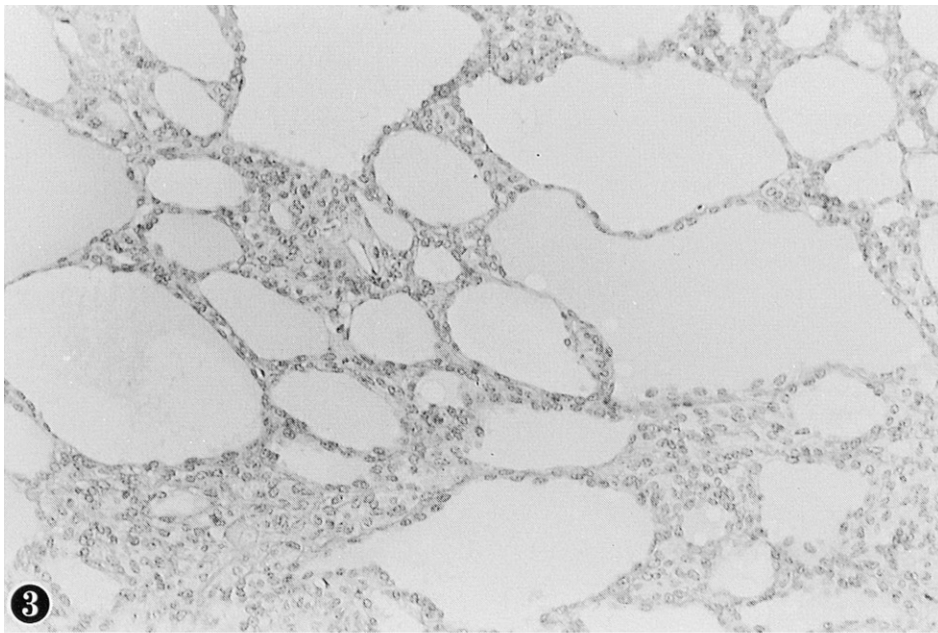


Fig. 3. Cysts of varying sizes creating a sieve-like pattern (hematoxylin and eosin [H & E], ×200).

Fig. 4. Closely packed tubular pattern (H & E, ×200).

without evidence of disease 2 years postoperatively.

MICROSCOPIC FEATURES

Microscopic examination of this tumor revealed many cysts of varying sizes. These cysts were lined by flattened cuboidal epithelial cells, which occasionally showed a hobnail appearance. Solid areas separated the cysts creating a sieve-like pattern on low power examination (Fig. 3). In some other areas the dominant pattern was characterized by a closely packed tubular pattern and a solid pattern with no apparent tubular formation (Figs. 4 and 5). There was an eosinophilic secretion within the lumens of some of the cysts and tubules. Cells

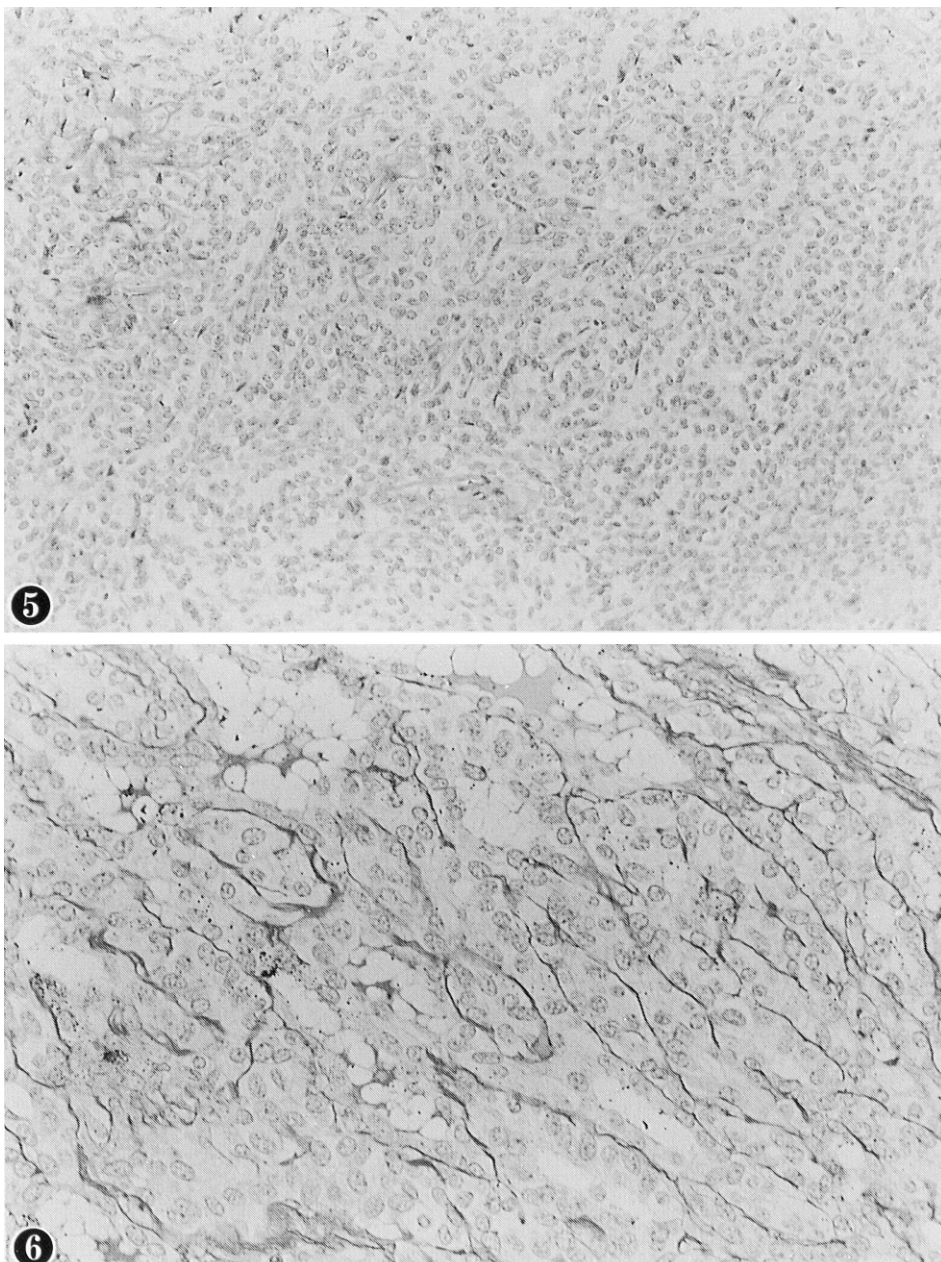


Fig. 5. Solid pattern without apparent tubular formation (H & E, $\times 200$).

Fig. 6. PAS-positive basement membrane surrounding tubular glands (PAS, $\times 400$).

had uniform round or oval nuclei, and mitotic activity was low. PAS stain revealed a PAS-positive basement membrane surrounding many of the sieve-like and tubular glands (Fig. 6). Immunohistochemical stains were negative for thyroglobulin. The left ovary and fallopian tube were unremarkable. The final diagnosis was female adnexal tumor of probable Wolffian origin.

DISCUSSION

The FATPWO is a rare tumor, with about 60 reported cases since the original detailed description (Kariminejad and Scully 1973). The origin of this tumor is most likely from the Wolffian duct (mesonephric) remnants, considering the characteristic location and typical light microscopic appearance. Most patients present with abdominal pain or mass, but some are identified incidentally like our case. The age of diagnosis ranges from 18 to 81 years (average, approximately 50 years). All the tumors were found to be unilateral and varied in size from 1 to 25 cm (average, 12 cm) in diameter. They were solid or solid and cystic and the color was yellow-tan or gray.

The histologic features of our case are similar to those originally reported (Kariminejad and Scully 1973). The tumor was composed of relatively uniform epithelial cells that lined the cysts and tubules. The cells showed uniform round or oval nuclei, and mitoses were rare. The presence of a closely packed tubular pattern, a solid pattern, and a sieve-like pattern is a characteristic histologic finding in these tumors. The homogeneous eosinophilic secretion within the lumens of some of these cysts and tubules, and the PAS-positive basement membrane surrounding cysts and tubules further support the diagnosis of FATPWO.

Tumors of probable Wolffian origin may be confused with struma ovarii, sex cord-stromal tumors, especially Sertoli cell tumors and granulosa cell tumors, and the more common mülerian epithelial tumors (particularly endometrioid and clear cell carcinomas). The absence of Leydig cell differentiation and lack of staining for thyroglobulin exclude the former two differential diagnoses and the absence of significant cytologic atypia excludes carcinomas of the usual surface epithelial type.

It is difficult to assess the malignant potential of this tumor. Tavassoli et al. (1990) reported that none of women in their series of 19 tumors developed recurrences, and that one patient whose tumor was incompletely excised and who was alive with residual tumor 8.5 years after diagnosis attesting to the indolent nature of this tumor. To our knowledge, only 9 cases of recurrences or metastases have been described in the literature (Table 1). Usually, patients developed recurrences or metastases more than 6 years following diagnosis or alived with disease for a long time (Taxy and Battifora 1976; Buntine 1979; Abbot et al. 1981; Young and Scully 1983; Brescia et al. 1985). Hypercellularity, cellular pleomorphism, and an increased number of mitotic figures are good indicators of malig-

TABLE 1. *Malignant tumors of probable wolffian origin*

Source	Patient age (years)	Location	Tumor size (cm)	Mitotic figures	Treatment	Follow-up
Taxy & Battifora (1976)	41	Right broad ligament	8.5×5.5×5	Rare	H, radiotherapy	Hepatic metastases, 6yrs
Buntine (1979)	41	Uterine cervix	—	—	H, BSO	Vaginal vault recurrence, 7yrs
Abbot et al. (1981)	18	Right mesosalpinx	8.5×6×5.5	<1/hpf	RSO	Peritoneal metastases, 6yrs DOD, 2yrs after metastases
Hughesdon (1982)	79	Left ovary	14×9×8	1.4~5.2/hpf	BSO	DOD, 1yr.
Young & Scully (1983)	52	Ovary	8	1.3/hpf	H, BSO	Lung metastases, 8yrs
Brescia et al. (1985)	23	Retroperitoneum	13×8	Rare	Tumor resection, cystectomy, vaginectomy	Local recurrence, 2lmo recurred 2 more times over 16yrs
Prasad et al. (1992)	47	Right broad ligament with peritoneal metastasis	20×25	Rare in the original tumor, more numerous in the peritoneal implants	H, BSO, omentectomy, appendectomy, chemotherapy	NED, 4yrs
Daya et al. (1994)	20	Right paracolicum	10	Rare	Tumor resection	Local recurrence, 2yrs AWD, 3yrs after recurrence
Daya (1994)	81	Right broad ligament with omental metastasis	15	8~10/hpf	H, BSO, omentectomy	DOD, 3mo

hpf, high power field; H, hysterectomy; BSO, bilateral salpingo-oophorectomy; RSO, right salpingo-oophorectomy; DOD, dead of disease; NED, no evidence of disease; AWD, alive with disease.

nancy (Abbot et al. 1981; Hughesdon 1982; Young and Scully 1983; Prasad et al. 1992; Daya 1994). It was not recognized such pathological features in our case, but in rare cases, tumors with minimal nuclear atypia and a very low mitotic rate may also recur (Taxy and Battifora 1976; Brescia et al. 1985; Daya et al. 1994). Therefore, it is best to regard this tumor as potentially malignant.

The most effective therapy is thought to be surgical complete resection. Adjuvant chemotherapy or radiotherapy remain uncertain since so few patients have been treated.

Although the majority of these tumors are benign, the occasional aggressive nature of these tumors, including some that have metastasized, make it imperative that patients with these tumors receive careful follow-up for possible recurrence and/or metastasis.

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