

# Male Wolffian adnexal tumor: The first report of long-term follow-up after radical surgical treatment

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## Abstract

The male Wolffian tumor is an extremely rare case in male patients. Here, we report a patient with such malignancy and successful radical surgical treatment at 15-year follow-up. The clinicopathological, immunohistochemical, and ultrastructural features are described. The differential diagnosis of this tumor in a male patient is discussed.

**Keywords:** Male; Prostate; Wolffian adnexal tumor

## 1. Introduction

Female adnexal tumor of probable Wolffian origin (ATWO) was first described in 1973 by Kariminejad and Scully.<sup>[1]</sup> Less than 100 cases of this rare gynecologic tumor have been documented in the English literature, with up to 20% developing recurrence or metastases.<sup>[2]</sup> To the best of our knowledge, only 3 cases of male ATWO (one of them developing metastasis later on) have been described.<sup>[3,4]</sup> Location, histology ultrastructural analyses<sup>[5,6]</sup> and immunohistochemical studies<sup>[7-9]</sup> have supported its derivation from mesonephric (Wolffian) duct remnants. We report a male ATWO arising at the apex of the prostate around the prostatic and membranous urethra and successful surgical treatment with 15 years follow-up.

## 2. Case report

A-57-year-old man sought medical attention due to difficulty in initiating urination, decreased urinary stream and a slight elevation of the prostatic-specific antigen serum level (7.08 ng/mL). Transrectal ultrasonography revealed a localized nodular mass between the apex of the prostate and the pubic bone. Transrectal ultrasonography-guided biopsies of the prostate and the lesion were initially reported as a metastasis possibly mesothelial in origin. A complete clinical evaluation, bone scan and computed tomography of the abdomen and thorax ruled out other lesions. After multidisciplinary oncologic consultation, and with informed

consent of the patient, the lesion was regarded as a single lesion (Fig. 1A), and an extended radical treatment was suggested. The patient underwent an open radical retropubic prostatectomy with resection of the urogenital diaphragm and Mitrofanoff catheterizable vesicostomy.

The en bloc surgical resection specimen of the prostate, seminal vesicles, proximal prostatic, and membranous urethra, showed a partially cystic and solid mass measuring 2.7 cm × 3.1 cm, located near the apex of the prostate around the membranous urethra. The prostate itself and both seminal vesicles were unremarkable. Grossly, the tumor was a solid, rubbery, pale yellow encapsulated mass with focal cystic degeneration and hemorrhage (Fig. 1B).

On microscopic examination, the tumor was well-circumscribed and showed a characteristic mix of distinct histopathologic features. The tumor was composed of varying proportions of diffuse and tubular (Fig. 2A), retiform (Fig. 2B) and multicystic patterns. In addition, papillary features with fibrovascular cores were focally present. The retiform pattern was characterized by a network of elongated and branching tubules, occasionally in a sieve-like pattern, morphologically similar to the rete ovarii. Tubular structures of varying size and shapes, were lined by cuboidal to columnar epithelium, with basally located nuclei and clear cytoplasm. The diffuse or solid pattern was composed of a more spindled cell population. Overall, the nuclei were cytologically bland with fine, evenly dispersed chromatin, prominent folds and clefts, an absence of discernible nucleoli, and a low mitotic index (2 mitoses/10 HPF). Abnormal mitotic figures were not observed. Immunohistochemical analysis showed the tumor cells to be reactive for keratin, vimentin, calretinin, and cytokeratin 7 (CK7). There was no immunoreactivity for prostatic-specific antigen, inhibin, epithelial membrane antigen (EMA), monoclonal carcinoembryonic antigen (mCEA), cytokeratin 20 (CK20), estrogen receptor (ER), and progesterone receptor (PR).

Ultrastructural findings showed groups of cells forming tubular structures surrounded by a well-developed basal lamina (Fig. 2C). The luminal aspect of the tumor cells showed short

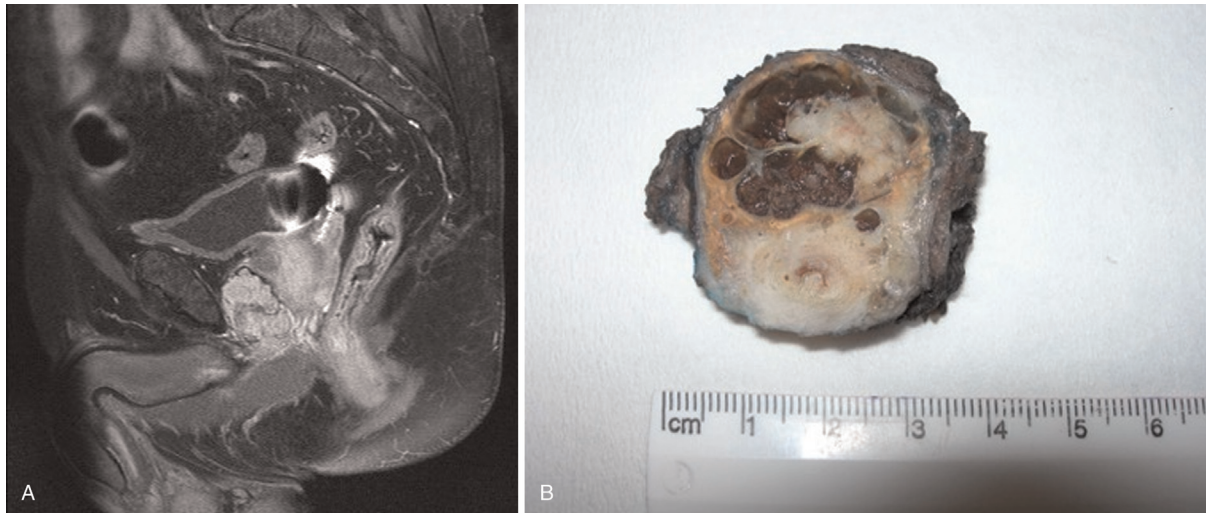
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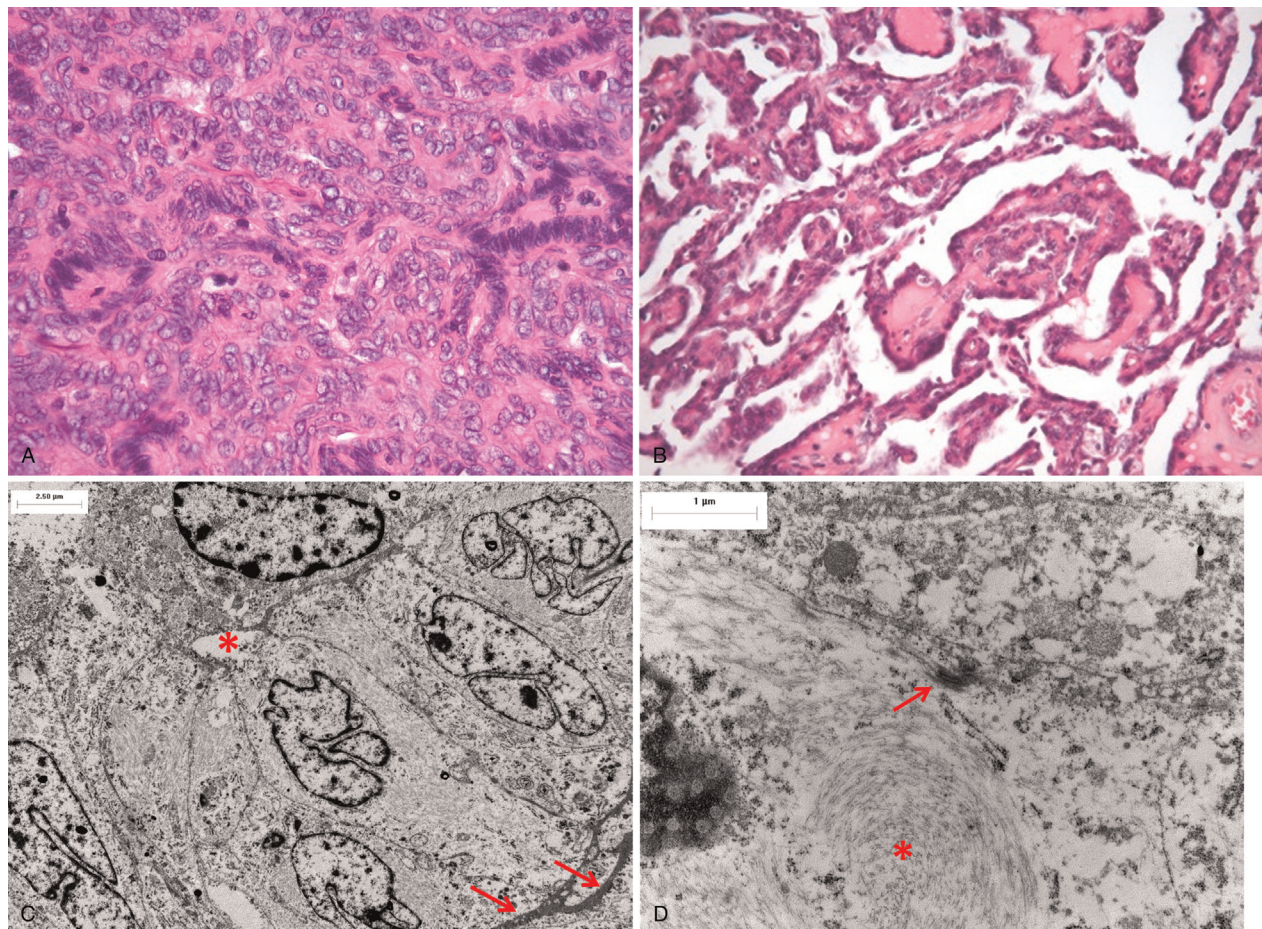
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**Figure 1.** Imaging and pathological tumor features: (A) Computed tomography of pelvis before operation with lesion at between the prostate apex and the pubic bone (B) gross features: para-urethral nodular lobulated mass with focally hemorrhagic cystic degeneration.



**Figure 2.** Histological tumor features: (A) Diffuse solid pattern with focal tubular differentiation ( $\times 400$ ) (H.E. stain); (B) Retiform pattern. Note the morphological similarity to the rete ovarii ( $\times 250$ ) (H.E. stain); (C) Tubular arrangement of cells surrounded by a thick basement membrane; remark irregular nuclei with indentations (electron micrograph of tumor cells retrieved from a paraffin-embedded tissue block); (D) Detail of two adjacent tumor cells showing desmosomes; note numerous whorled bundles of intermediate vimentin filaments (electron micrograph of tumor cells retrieved from a paraffin-embedded tissue block).

microvilli. Well-formed desmosomes were present. The nuclei of the cells exhibited deep clefts and indentations. The cytoplasm contained only a few organelles but numerous intermediate vimentin filaments (Fig. 2D).

The patient has been free of disease for 15 years after surgery at pelvic, abdominal and chest imaging.

### 3. Discussion

The herein presented para-urethral tumor, a partially solid and partially cystic mass, shares histological features with female ATWOs.<sup>[1,5,6,8]</sup>

Although the diagnosis of this tumor is mainly based on the exclusion of other possibilities, characteristic histopathological patterns include a solid or diffuse arrangement of spindled epithelial cells, tubular, retiform, and multicystic patterns. The tubules appear to be compressed with slit-like lumens lined by cuboidal epithelium. Another characteristic pattern, also presented in our case, is the retiform pattern characterized by a network of elongated and branching tubules, occasionally in a sieve-like arrangement. A multicystic growth pattern can be seen with variably sized cystic spaces lined by a single layer of flattened cuboidal cells. Nuclei are bland with fine and evenly dispersed chromatin. Nucleoli are small to inconspicuous. The mitotic index is generally low (0–3 mitoses/10 HPFs).

The immunohistochemical analysis of female ATWO was described at its best by Devouassoux-Shisheboran et al.<sup>[9]</sup> In a study of 25 cases, they found that ATWOs were immunoreactive for pancytokeratin (100%), cell adhesion molecule (CAM) 5.2 (100%), calretinin (91%), cytokeratin 7 (88%), inhibin (68%), EMA (12%), estrogen receptor (28%), progesterone receptor (24%). The tumors were not reactive for monoclonal carcinoembryonic antigen (CEA) and cytokeratin 20. The tumor described here shows a similar pattern.

Electron microscopic findings are characteristic but not specific<sup>[3,5,6]</sup> and were first described by Taxy and Battifora.<sup>[10]</sup> The nuclei of the tumor cells are large and irregular in shape due to prominent folds and clefts. Well-formed desmosomes, short microvilli, and well-developed basal laminae surrounding the cell groups are other features. Again, these features were present in the case described.

The differential diagnosis of this tumor of probable Wolffian origin occurring at the apex of the prostate around the prostatic urethra includes nephrogenic adenoma within a urethral diverticulum, adenomatoid tumor, clear cell adenocarcinoma of the urethra, prostatic adenocarcinoma and metastases from mesothelial, testicular or other origin.

Three cases of ATWO in male patients have been published and optimal treatment remains unclear. Whereas malignant potential of ATWO in the male population remains highly possible, radical surgery should be advised with adjuvant radiological follow-up. Awareness of this entity in males is important to prevent histological misdiagnosis.

### 4. Conclusion

We present a neoplasm of probable Wolffian origin at the apex of the prostate in a 57-year-old men with no disease recurrence after

radical surgery at 15-year follow-up. It is important to recognize this distinctive lesion and to separate it from less or more aggressive lesions in the male genitourinary tract.

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### Statement of ethics

The written informed consent to publish their case including images was given by patient.

### Conflict of interest statement

No conflict of interest has been declared by the author.

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### Author contributions

Data acquisition and analysis: E.S, R.V, P.M, S.J.

Manuscript conception and design: D.M, S.J.

Manuscript writing and critical review: D.M, S.J, E.S, R.V, P.M.

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