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Case Report

Tubulosquamous Polyp of The Vagina: A Report of Three Cases and Review of The Literature

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Abstract:

Background: Tubulosquamous polyp (TSP) is a rare benign lesion predominantly occurring in the vaginal wall, most commonly affecting postmenopausal women and rarely observed in women of late reproductive age. The lesion typically arises in the upper vaginal wall. Its histogenesis remains unclear, although it is widely believed to originate from Skene's glands, which are homologous to the male prostate.

Case: We report three cases of TSP, all occurring in postmenopausal women. Each case underwent detailed clinicopathological evaluation, including histological and immunohistochemical analysis, to characterize the features of this uncommon lesion.

Conclusion: This case series highlights the importance of recognizing TSP as a distinct benign entity to enhance awareness and further improve the understanding of this rare disease, thereby helping to reduce the risk of missed or incorrect diagnoses.

Keywords: tubulosquamous polyp; clinicopathological analysis; case report

1.Introduction

Neoplasms of the vaginal wall are predominantly benign, with fibroepithelial polyps being the most common type [1]. In addition, some stromal tumors, such as leiomyomas and angiomyofibroblastomas, can present as neoplasm-like growths [2]. However, tubulosquamous polyps (TSPs) have rarely been reported, and their origin remains unclear. Most experts believe that TSPs originate from Skene's glands, which are similar to the male prostate in terms of physiological functions [3]. In the cervical region, these lesions are referred to as ectopic prostatic tissue [4, 5]. At present, there are relatively few reports on TSPs in the literature. Herein, we present three cases of TSP, describe their clinicopathological characteristics, and aim to enhance the awareness and understanding of this rare disease among pathologists and clinicians.

2. Case presentation

We selected three cases diagnosed at the Department of Pathology, Jiading Maternity and Child Healthcare Hospital in Shanghai, and conducted a clinicopathological analysis to enhance the understanding of this rare disease. All patients were postmenopausal women, aged 54, 64, and 68 years old. The lesions were located in the vaginal fornix, the upper part of the vaginal wall, and the lower part of the labia majora, respectively. None of the patients had a history of gynecological disease, prior treatments such as radiotherapy or chemotherapy, or a family history of hereditary tumors.

All three cases presented as grayish-white polypoid lesions measuring 0.5 to 1.5 cm in diameter. The surface was grayish-white and smooth, with a relatively soft consistency and no roughness or nodular protrusions. Upon sectioning, the cut surface appeared grayish-white with a medium texture, and there were no signs of hemorrhage or necrosis.

Microscopic examination revealed squamous epithelial cell nests embedded within the fibrous stroma of the nodular lesions. These nests varied in size and morphology, and most of them lacked mature squamous epithelial cells. Surrounding the nests were relatively flattened basaloid cells arranged in a compact pattern. The cell nuclei were round or oval, with evenly distributed chromatin and no obvious nucleoli. Central areas of the nests occasionally contained necrotic nuclear debris or calcification. The necrotic nuclear debris appeared as unstructured eosinophilic substances, while the calcifications

presented as blue-stained granular or confluent deposits. The surface of the nodules was covered by squamous epithelium; however, there was no direct connection between this surface epithelium and the underlying squamous epithelial nests, and the boundary between them was clear (Figure 1A). Tubular structures could be seen around the cell nests, composed of single-layered or double-layered epithelial cells. The luminal surface was composed of cuboidal cells with relatively regular morphology and centrally located round nuclei containing fine chromatin. The basal layer consisted of small, flat cells that were neatly arranged. The lumens often contained homogeneous eosinophilic secretions, which appeared as uniformly red substances that filled the glandular spaces under light microscopy. The fibrous stromal cells appeared bland, without mitotic figures or necrosis. Stromal cells exhibited regular morphology, uniform nuclear size, and no cytologic atypia (Figure 1B).

Immunohistochemical analysis showed that the squamous epithelial cell nests were positive for P63, AE1/AE3, and cytokeratin (CK)5/6. P63 exhibited strong brownish-yellow staining in the cell nuclei, indicating squamous epithelial differentiation. AE1/AE3 showed positive staining in the cytoplasm, further confirming the epithelial nature of the cells. CK5/6 was also expressed in the cytoplasm, suggesting that these cells belonged to the stratified squamous epithelial cell type. The tubular structures were positive for prostatic markers, including prostate-specific acid phosphatase and prostate-specific antigen (PSA), both of which showed brownish-yellow cytoplasmic staining, implying the differentiation of the prostate lineage in the tubular structures (Figures 1C and 1D). Additionally, the flattened cell layer surrounding the tubular structures expressed S100, calponin, and smooth muscle actin (SMA). S100 showed cytoplasmic positivity, while calponin and SMA also showed positive staining in the cytoplasm, indicating myoepithelial differentiation in these flattened cells.

Follow-up data were available for all three patients. None of them received additional treatment after surgical excision. The follow-up period ranged from 1 to 4 years, during which patients were monitored through regular gynecological examinations, vaginal ultrasound, and other assessments. No signs of recurrence or metastasis were observed during the follow-up period. All patients had a good quality of life, with no reports of abnormal vaginal bleeding, pain, or other discomfort.

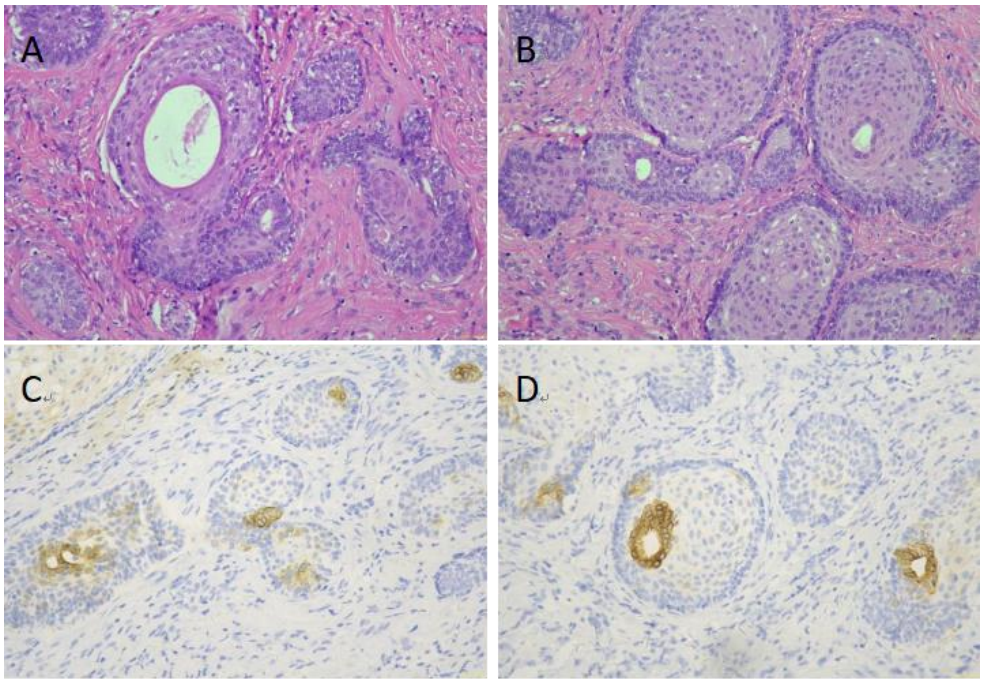

Figure 1: Histological and immunohistochemical features of tubulosquamous polyp (TSP)

A: Low-power view (20×) showing squamous epithelial cell nests with central keratinized material; B: Tubular structures with mucinous differentiation within the squamous epithelial nests (20×); C: Immunohistochemical staining showing cytoplasmic positivity for prostate-specific antigen in the tubular structures (EnVision method); D: Cytoplasmic positivity for prostatic acid phosphatase in the tubular structures (EnVision method)

3. Discussion

TSP is most commonly found the upper vaginal wall but can also occur in the cervix and vulva. It is a benign lesion with an unclear origin. Currently, it is believed that TSP originates from Skene's glands, which are similar to the male prostate [3, 6]. In 2007, McCluggage and Young first reported and described 10 cases of tubulosquamous epithelial polyps. They also believed that some previously reported cases of vaginal Brenner tumors were likely misdiagnosed TSPs [7]. This highlights the possibility of diagnostic confusion due to insufficient understanding of TSPs among clinicians and pathologists. Improving recognition of this entity is, therefore, of great clinical significance. TSP typically occurs in postmenopausal women, possibly related to hormonal changes, particularly decreased estrogen levels, which may affect the local microenvironment of the vaginal wall, thus promoting the occurrence of this lesion. However, TSP can occasionally occur in older women of childbearing age. Most patients are diagnosed either because of symptoms such as vaginal bleeding or incidentally during routine gynecological examinations [7]. In this report, all three patients sought medical treatment after growths were identified during physical examinations. One lesion was located in the vaginal fornix, another in the upper part of the vagina, and the third—in the youngest patient (54 years old)—was found below the labia majora. Clinically, TSPs usually present as polyps or nodules and often cause vaginal bleeding. This bleeding is likely attributed to the thin mucosal surface of the lesion, which is easily affected by factors such as friction. TSP is considered biologically inert, with no reports of recurrence to date. In our series, the longest follow-up period was 4 years, during which no recurrence was observed in any of the three patients, further confirming the benign and non-aggressive nature of this lesion.

TSP should be differentiated from the following six lesions: ① Fibroepithelial polyp: This is a more commonly encountered lesion in clinical practice. Histologically, it exhibits a fibrovascular core in the central fibrous stroma, covered by stratified squamous epithelium of varying thickness. There are no squamous epithelial cell nests or tubular structures in the stroma, and the stromal cells are sparse and bland, though mild atypia might be present, distinguishing them from TSPs [1, 8]. Clinically, fibroepithelial polyps are soft and mobile, often shifting with changes in body position, while TSPs are relatively fixed and have a firmer texture. ② Benign mixed tumor of the vagina (vaginal spindle cell epithelioma): It is more commonly found in the posterior vaginal wall near the hymenal ring in premenopausal women. Histologically, it contains both epithelial (squamous and glandular) and spindle-shaped stromal cells. The stromal component is rich in cellularity and expresses CK, with WT-1 and calretinin also being helpful for differentiation [9]. The intermingled architecture of epithelial and stromal components in this tumor contrasts with the distinct squamous nests and tubular structures seen in TSP. ③ Vaginal Brenner tumor: Typically located in the lower part of the vagina or around the hymen, this tumor is composed of bland transitional epithelium in dense fibrous stroma. A characteristic feature is the presence of nuclear grooves within the epithelial cells [10]. The transitional epithelial morphology and grooved nuclei are important differential features that differ from the squamous nests in TSP. ④ Ectopic prostate tissue (EPT): Histologically, EPT mimics the structure of normal prostate acini. In typical cases, the epithelial lining forms inverted papillae or cribriform structures, similar to the central zone of the normal prostate. These lesions are located beneath the surface epithelium of the urogenital tract [11]. Although both EPT and TSP contain glandular elements suggestive of prostatic differentiation, the tissue structure of EPT is closer to that of normal prostate acini, with unique papillae or cribriform structures, unlike the simpler tubular structures of TSP.

4. Conclusion

In summary, although TSP is a benign lesion often occurring in the vagina, it is extremely rare and prone to missed diagnosis, misdiagnosis, or overdiagnosis. When vaginal polyps are observed in postmenopausal women, especially those exhibiting biphasic differentiation with both squamous and glandular components, TSP should be considered in the differential diagnosis. To aid in the recognition of this uncommon entity, we reported three cases of TSP and conducted a literature review to enhance the understanding of its clinicopathological features among clinicians and pathologists. Improved awareness is essential for establishing an accurate and definitive diagnosis.

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Conflict of interest

The authors declare no competing interests.

Ethical Approval

Not applicable. This article does not contain any studies with human participants or animal subjects.

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