Nephrogenic Adenoma in Urethral Diverticulum: an Unusual Finding

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Key Words
Nephrogenic adenoma
Diverticulum urethra

Abstract
The 3rd case of nephrogenic adenoma in the urethral diverticulum of a female is reported herein. A short review of the literature is presented.

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Introduction
Nephrogenic adenoma (NA) is an uncommon lesion of the urinary tract, whose etiology is still debated. Although the lesion can occur throughout the urinary tract, most of the reported cases were located in the bladder. As far as we know, only 2 cases of NA within the urethral diverticulum have been previously reported: 1 by Peterson and Matsumoto [13], and 1 by Berger et al. [2]. The 3rd case report of the world-wide literature is herein described.

Case Report
A 32-year-old woman was admitted for evaluation of dysuria, dispareunia and post-voiding dribbling of 4 years of duration. Vaginal examination revealed a sub-urethral tender mass, producing a scanty yellowish discharge from the urethral meatus when palpated. On urinalysis sterile pyuria was present. History included 2 pregnancies (2 and 4 years before) with normal delivery but no urinary tract infections. A positive pressure urethrogram (fig. 1) confirmed the presumptive diagnosis of urethral diverticulum and a transvaginal diverticulectomy was performed.

Histologic examination showed nephrogenic adenoma (fig. 2A, B). Nine months after surgery the patient is asymptomatic, urinalyses are negative, and vaginal examination reveals a well-healed vaginal wall.

Conclusions
In 1949 David described the first case of NA of the bladder referring to it as ‘hamartoma of the bladder’ [4].
The term ‘nephrogenic adenoma’ was minted by Friedman and Kuhlenbeck [5] in 1950: with this term the authors stressed the histologic pattern of the lesion comprising of epithelial tubules in the lamina propria of the bladder that strictly resembled portions of a nephron.
Fig. 2. A Glandular structures with dilated lumen surrounded by a fibrous stroma and delimited by cuboidal epithelium. The epithelial cells show an eosinophilic cytoplasm, rounded and uniformly dense nucleus with small nucleoli. No evidence of mitoses or cellular atypia can be found HE. × 160. B Glandular structures lined by epithelium with supranuclear vacuoli and endoluminal PAS-positive material. PAS. × 250.

From these first reports, several cases occurring throughout the urinary tract have been described in the literature [1, 2, 6, 8, 12–15]. The etiology of the lesion is still debated. Theories include embryologic [10], inflammatory [14] and immunosuppressive [6] origin.

In our case there was neither a previous history of urinary tract infections nor urethral instrumentation; moreover, the location of the lesion could not be explained by embryologic remnants, and the patient denied immunosuppressive drugs assumption.

In 5 of the reported cases, NA coexisted with a bladder neoplasm: 3 transitional cell carcinomas [2, 11] and 2 adenocarcinomas of the bladder [3, 9]. However, it is still uncertain whether the occurrence of an invasive lesion results from a malignant transformation of NA or the latter simply represents a concomitant lesion.

Although the occurrence of mesonephric carcinoma of the urethra is rare [7], a close clinical observation has to be planned in these patients in order to detect early symptoms or signs of malignancy.

References