



Cholesteatoma in ectopic kidney

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ABSTRACT

Cholesteatoma in the urinary system is a rarely seen benign condition. Rosina firstly defined this condition in the year 1953. Histopathologically it is characterized with keratinization, and squamous metaplasia of urothelial epithelium associated with desquamation of keratinized layers. Flank pain is the most common symptom that is caused by elimination of keratinous material. In our case we will discuss cholesteatoma developed in an ectopic kidney which has not been described in the literature before.

Keywords: Ectopic kidney; cholesteatoma; renal pelvis.

Introduction

Cholesteatoma of the urinary system is a rarely seen benign condition.^[1] Firstly it was reported by Rosina in the year 1953.^[2] Histopathologically it is characterized with keratinization, and squamous metaplasia of urothelial epithelium associated with desquamation of keratinized layers. Flank pain is the most common symptom that is caused by elimination of keratinous material. In our case we will discuss cholesteatoma developed in an ectopic kidney which has not been described in the literature before.

Case presentation

Physical examination of the patient who applied to urology outpatient clinic of our hospital with complaints of colicky pain, and flank pain revealed costovertebral angle tenderness which suggested the presence of urinary system stone disease. Intravenous pyelography, and computed tomography (CT) aided in the diagnosis of ectopic kidney with manifestations of bilateral nephrolithiasis, right kidney was situated more inferiorly than its normal location in the superior part of the pelvis with its hilus facing anterior abdominal wall (Figure 1). Besides, on CT, increase in nodular density within right renal collecting system was a remarkable finding (Figure 2). The patient was scheduled for renal stone surgery. Dur-

ing surgery plenty of yellow-colored material with soft consistency between right pelvis stones of the ectopic kidney were noticed, and allocated for histopathological analysis. Macroscopically, creamish-yellow-colored tissue particles with irregular contours measuring 4 x 3 x 1.5 cm were sampled, and subjected to histopathological analysis. Specimens appeared as glycogenated, and keratinized multi-layered squamous epithelium (Figures 3a, b). Any sign of atypia was not encountered. Histopathology was reported in consistent with cholesteatoma.

Discussion

Desquamative, keratinized, squamous metaplasia (cholesteatoma) of the urinary system is most frequently localized in the renal pelvis, and adjacent calyces. Most often its cystic form is observed.^[1] More frequent involvement of the lower urinary system has been also indicated.^[3-5] It is more frequently seen in adults with a slight male predominancy (male/female: 3/2). In children a few cases have been reported.^[5,6]

Squamous metaplasia of the renal pelvis can occur secondary to irritative etiologies as chronic urothelial inflammation, and renal stone and embryologic anomalies. Radiological diagnosis is difficult to make. For diagnosis radiolucent filamentous defect in the renal pelvis, cornified epithelium or keratinized scaly material should be observed.^[5,6]

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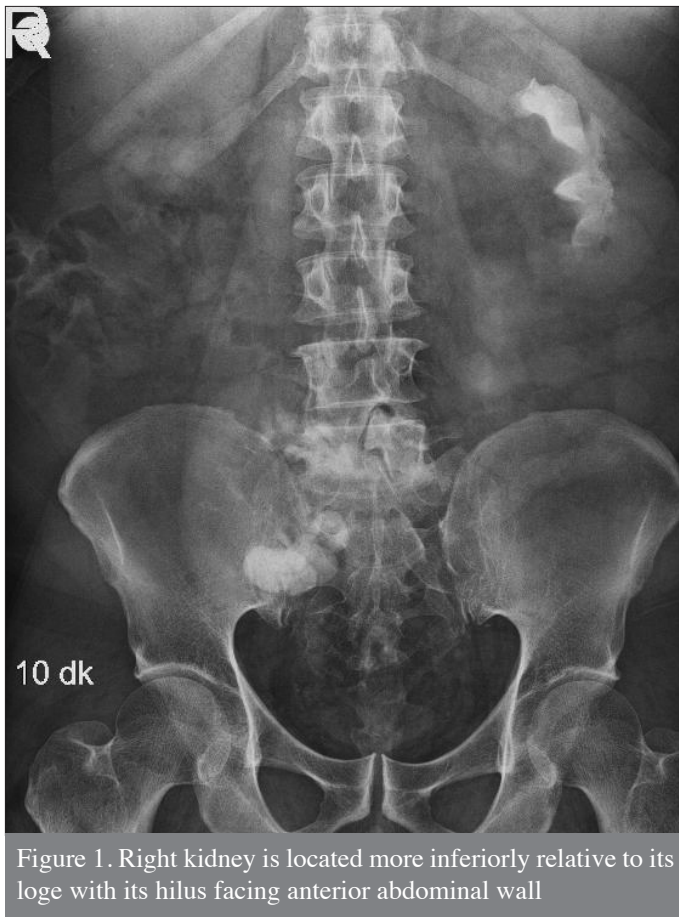


Figure 1. Right kidney is located more inferiorly relative to its loge with its hilus facing anterior abdominal wall



Figure 2. On computed tomogram, increases in nodular density in the collecting system of the right kidney are observed

Since it is a rarely seen entity, a standard treatment protocol has not been defined. Since it is rarely seen, preoperative diagnosis is generally challenging. Nephrolithiasis, inflammatory events, and urothelial carcinoma are considered in the differential diagnosis. Although any malignant transformation has not been reported so far, nephrectomy is an aggressive procedure among

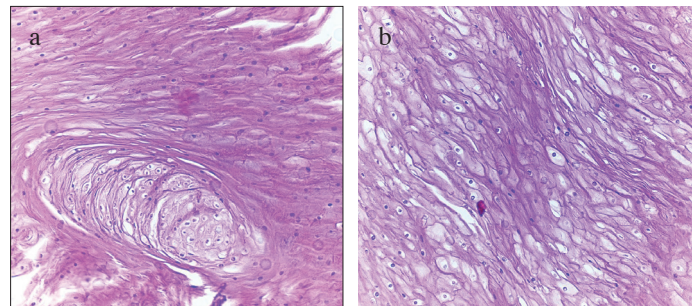


Figure 3. a, b. Specimens of glycogenated, and keratinized multilayered squamous epithelium (H&E x200)

treatment alternatives. In addition, alternatives as nephron-sparing surgery, partial nephrectomy, and watchful waiting are also available.

Recurrence was reported in a patient who had been treated with percutaneous implantation of nephrostomy tube 15 years ago.^[3] In 8-12% of upper urinary tract squamous cell carcinomas coincidentally squamous metaplasia was seen, however squamous cell carcinoma which developed from squamous metaplasia has not been described so far. Lupovitch et al.^[6] claimed that long-term use of supportive vitamin A supplements prevented recurrences in non-recurrent cases during 56 years of follow-up, and demonstrated presence of recurrence when treatment was discontinued. Therefore as is the case with oral leukoplakia, it has been emphasized that similar leukoplastic lesions can be prevented with vitamin A supplementation.^[6]

In the literature nearly 90 cases with urinary system cholesteatomas have been reported.^[2] However as is seen in our patient, a case with cholesteatoma developed in an ectopic kidney has not been reported in the literature, so far.

Informed Consent: Written informed consent was obtained from patient who participated in this case.

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