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Squamous and glandular metaplasia related to a renal pelvis stone leading to secondary pyeloureteral junction syndrome

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ABSTRACT

Keratinizing squamous metaplasia of the renal pelvis is a rare lesion of the upper urinary tract that can occur in the context of chronic aggression to the urothelium, potentially leading to a secondary pyeloureteral junction syndrome.

We report the case of a 43-year-old patient discovered intraoperatively in relation to a renal pelvis stone causing a pyeloureteral junction syndrome. The extemporaneous histological examination ruled out a malignant process, and we performed a pyeloplasty according to KUSS-ANDERSON technique. This pathology should be recognized by the urologist for appropriate management. Treatment is conservative, with extended follow-up to detect recurrences or carcinomatous degenerations.

1. Introduction

The pyeloureteral junction syndrome, characterized by dilation of the renal pelvis and calices upstream of a normal-sized ureter, is primarily a congenital syndrome, but secondary forms also exist. We present the case of a 43-year-old patient who exhibited squamous and glandular metaplasia in the context of pyelic lithiasis, leading to a stenosis of the pyeloureteral junction. Intraoperative discovery of suspicious lesions with potential malignancy at the pyeloureteral junction necessitated an extemporaneous histological examination, which ruled out malignancy, and subsequently, a pyeloplasty was performed using the KUSS-ANDERSON technique.

2. Case presentation

F. M, a 43-year-old patient, presented to our department with left-sided lower back pain that had been evolving for several months, without any other associated symptoms. The clinical examination was unremarkable. Serum creatinine levels and urine cytobacteriological examination were within normal limits. An abdominal CT scan revealed a left pyeloureteral junction syndrome, with a renal pelvic dilatation measuring 50 mm and the presence of a 35 mm long axis renal pelvic stone (Fig. 1). Renal scintigraphy demonstrated delayed left renal excretion and incomplete drainage. A pyelolithotomy combined with pyeloplasty was indicated. Under general anesthesia, we performed a lumbar incision between the 11th and 12th ribs and proceeded with a pyelotomy, extracting a rounded, dark, and firm stone. Subsequently, we resected the pyeloureteral junction and the redundant portion of the renal pelvis (Fig. 2). Exploration revealed a complete junctional stenosis and irregular whitish patches lining budding lesions on the inner wall of the resected renal pelvis. An extemporaneous histological examination of the specimen concluded that squamous and glandular metaplasia, without signs of malignancy, was responsible for the stenosis (Fig. 3). We performed a KUSS-ANDERSON pyeloplasty using a JJ stent. The postoperative course was uneventful, and the patient was discharged on the 5th postoperative day. The JJ stent was removed after 3 weeks. We are at a follow-up duration of 3 months after the procedure, and the patient does not report any specific symptoms.

3. Discussion

Acquired pyeloureteral junction syndrome can result from various etiologies, such as obstruction caused by a lodged urinary stone at the junction, inflammatory stenosis, upper urinary tract tumors, fibroepithelial polyps, or extrinsic compression.[1] Another less-documented etiology in the literature is squamous metaplasia of the renal pelvis, which involves a transformation of the renal pelvis urothelium into a skin-like epithelium without cellular atypia.

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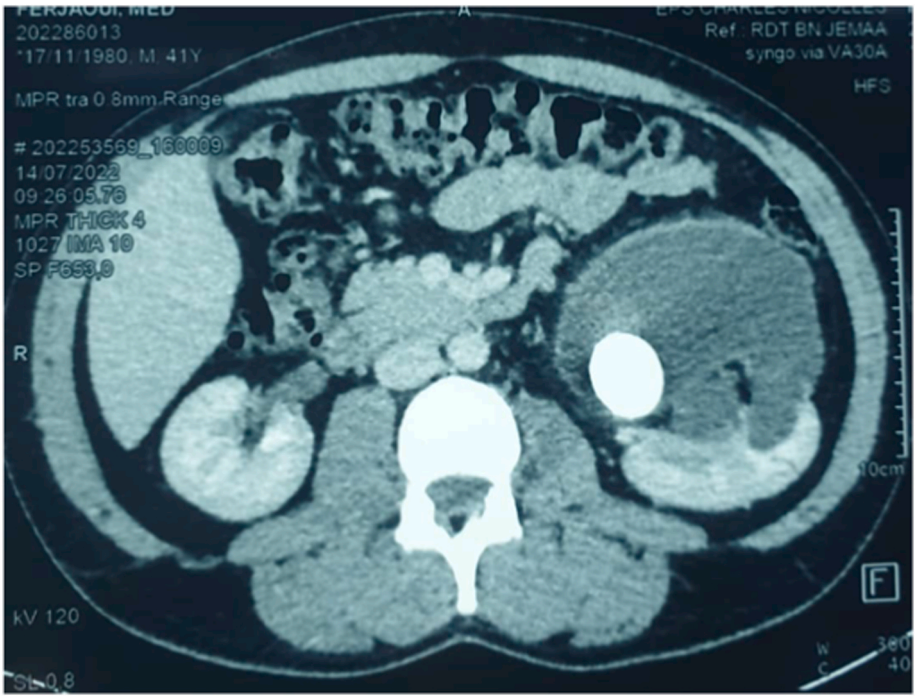

Fig. 1. Contrast-enhanced Abdominopelvic CT scan in axial section showing the left pyeloureteral junction syndrome.

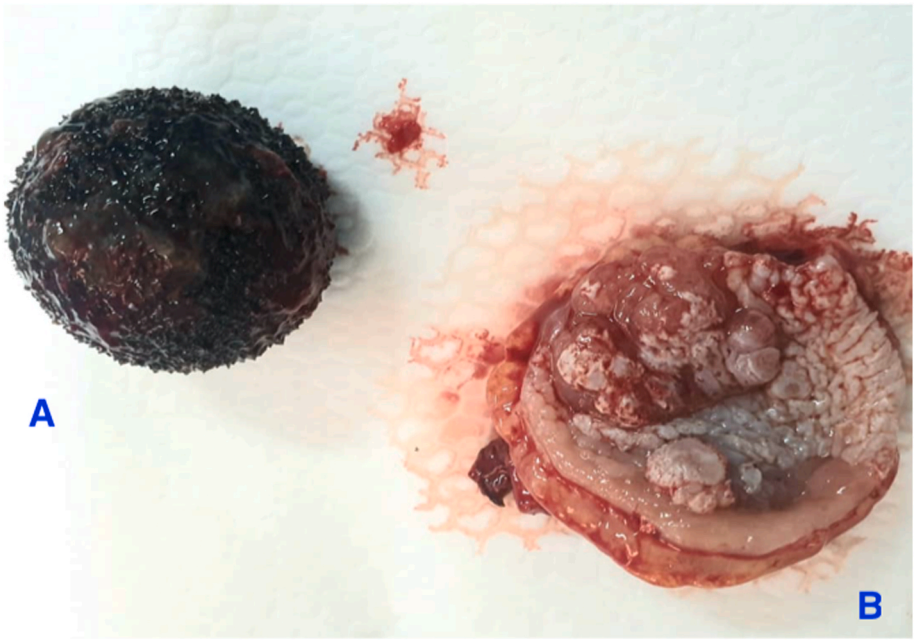

Fig. 2. (A): Extracted renal pelvic stone; (B): Resected specimen including the pyeloureteral junction and the redundant portion of the renal pelvis.

Squamous metaplasia exists in two forms: the non-keratinizing form, which is common and physiological in women, and the keratinizing form. The etiopathogenesis of the keratinizing form remains debated, and three hypotheses have been proposed[2]:

- The first hypothesis is based on the transformation of urothelium exposed to chronic aggressions,[3] whether mechanical, infectious, or generally inflammatory in origin.
- The second hypothesis is a spontaneous transformation of the urothelium, which could occur in 40% of cases in the absence of any pathology.
- The third hypothesis is embryological and suggests that cutaneous ectodermal cells could colonize the Wolffian endoderm during embryogenesis.

In our case, the presence of a renal pelvic stone points to chronic mechanical aggression as the cause. The lodged stone would thus have led to chronic irritation and inflammation, resulting in cellular changes and metaplasia of the epithelial cells at the pyeloureteral junction. This was followed by stenosis, resulting in the pyeloureteral junction syndrome.

Keratinizing squamous metaplasia of the renal pelvis, also known as

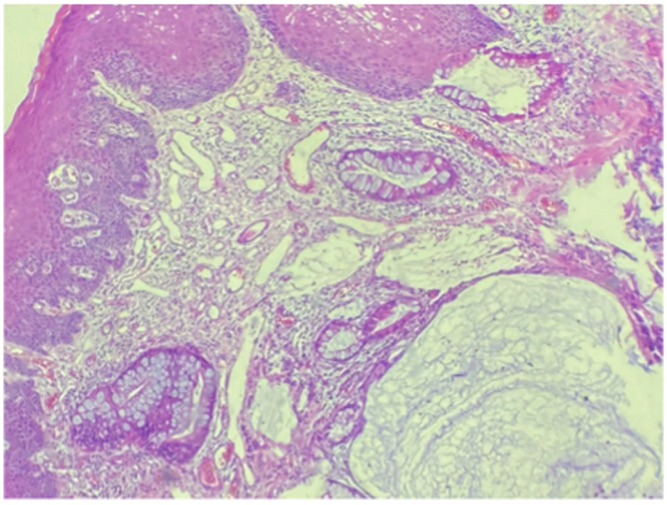

Fig. 3. Histological image showing a presentation of squamous and glandular metaplasia without signs of malignancy.

pyelic leukoplakia, is endoscopically characterized by irregular whitish patches. Described as a preneoplastic urothelial lesion,[3] it presents a challenging and rare differential diagnosis from urinary tract tumors.[2]

These lesions often require tissue biopsy for definitive diagnosis.[2,4]

Awareness of these lesions and their morphological appearances is crucial to avoid overdiagnosis and inappropriate aggressive treatment.

Given the intraoperative frozen section diagnosis of squamous and glandular metaplasia without signs of malignancy, we opted for conservative treatment using the KUSS-ANDERSON technique.

In addition to typical lithiasic complications, squamous metaplasia should not be overlooked, and early treatment for any renal pelvic stone is crucial to prevent long-term induction of squamous metaplasia.

While not all squamous metaplasias progress to cancer, squamous metaplasia increases the risk of certain cancers, including squamous cell carcinoma. Long-term follow-up studies have shown that nearly a quarter of patients eventually develop squamous carcinoma.[5]

Hence, regular monitoring of these patients is necessary to detect any atypical lesion that could lead to recurrence[2] or carcinoma transformation. Carcinomas arising from these lesions are often highly orthoplastic, and any slight atypia should prompt multiple biopsies to search for an infiltrating component.[4]

Given the rarity of this condition, we did not find any recommendations in the literature regarding surveillance modalities. For our patient, we are planning an CT urography (CTU) and ureterorenoscopy with in situ cytology annually for 5 years, considering the benign nature of the initial lesion. We are referring to the surveillance of low-risk upper urinary tract tumors that have undergone conservative treatment, which require even closer monitoring according to the recommendations of the French Association of Urology for the years 2022–2024. Nonetheless, we believe that specific recommendations from professional societies would help standardize practices concerning this condition.

4. Conclusion

Keratinizing squamous metaplasia of the renal pelvis is a rare lesion of the upper urinary tract that can occur in the context of chronic aggression to the urothelium, potentially leading to a secondary pyeloureteral junction syndrome. In some cases, it presents as a rare and challenging differential diagnosis from upper urinary tract tumors. The treatment approach remains conservative, and extended follow-up is essential to detect any potential recurrence or carcinoma degeneration.

Consent

Signed consent was obtained from the patient.

Declaration of competing interest

The authors declare that there are no conflicts of interest regarding the publication of this article.

References

1. Masson E. Syndrome de la jonction pyélo-urétérale. EM-Consulte, Disponible sur : https://www.em-consulte.com/article/1092021/syndrome-de-la-jonction-pyelo-uretherale.
2. Pedron P, Desgrandchamps F, Hoffmann P, Lesourd A, Billebaud T, Teillac P, et al. Place de l'endoscopie dans le diagnostic et le traitement de la métaplasie malpighienne kératinisée et desquamante du bassinet.
3. Mazerolles C. Lésions urothéliales pseudonéoplasiques et prénéoplasiques de la vessie.
4. Jones EC. Urinary bladder. Mimics of neoplasia and new pathologic entities. *Urol Clin North Am. août.* 1999;26(3):509–534. vi.
5. Benson RC, Swanson SK, Farrow GM. Relationship of leukoplakia to urothelial malignancy. *J Urol [Internet].* 1984;131(3):507–511.