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A case of urachal villous adenoma with high grade dysplasia focally bordering on adenocarcinoma in situ

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1. Introduction

The urachus, or the median umbilical ligament, is a vestigial structure of the allantois that connects the urinary bladder and the umbilicus. Urachal carcinoma, originally described by Hue and Jacquin in 1963, is a rare non-urothelial carcinoma. The incidence is estimated to account for 0.01% of all malignancies, 0.5% of all bladder tumors, and compose 20–40% of bladder adenocarcinomas. Most urachal cancer is associated with poor prognosis because of advanced stage at diagnosis.[1]

Urachal villous adenomas, which may coexist with in situ and invasive adenocarcinoma, is even more rare than urachal adenocarcinoma. Kato et al. found that there are fewer than 40 cases of villous adenoma with or without other types of carcinoma reported in the literature.[2] It is hypothesized that there is a biological progression of villous adenoma to adenocarcinoma, but only one case has been reported thus far.[3]

We present a case of urachal villous adenoma with high-grade dysplasia focally bordering on adenocarcinoma in situ.

2. Case presentation

A 51-year-old female presented with complaints of suprapubic discomfort, urgency, and passage of mucous in her urine of one-year duration. Urine dipstick showed microscopic hematuria for the previous 6 months with no gross hematuria. Notable medical and history included 10-pack-year smoking history, cholecystectomy, and colon cancer with colon resection. There is no significant family history of cancer. On physical examination, there were no positive physical findings. The laboratory results (CBC, CMP, urine culture, TSH) were within normal limits with the exception of small blood found on urine dipstick. Renal ultrasonography revealed a complex heterogeneous mass was found superiorly along the anterior wall of the bladder. Computed tomography of the abdomen confirmed a 3.5 cm complex solid and cystic mass anterior and superior in the bladder. Cystoscopy showed a 3 cm lesion contiguous with the mucosa on the dome of the bladder. Following the doctors' recommendation, the patient consented to laparotomy for radical resection of the urachal mass and partial cystectomy. Post-operatively, the patient recovered well.

3. Pathology

3.1. Gross pathology

The resection specimen consisted of a portion of soft tissue with a portion of skin and soft tissue at one end and a portion of soft tissue with a patent 0.6 cm lumen at the opposite end. The shaggy resection margins were previously shaved for intraoperative consultation, were negative for tumor, and inked blue subsequently. The specimen is opened through the previously described patent opening to reveal a friable tan-pink papillary mass (Fig. 1) partially surfaced by clear mucoid material measuring 3.5 × 3.0 cm. Further sectioning reveals the mass to extends to a maximum depth of 1.5 cm and was 1.1 cm from any resection margin.

3.2. Light microscopy

The mass that protrudes from the surface of the mucosa is a villous adenoma, similar to that seen in the colon (Fig. 2). There are features of high-grade dysplasia, bordering on adenocarcinoma in situ (Fig. 3). No invasive component was identified in the entirely submitted mass. The adjacent mucosa is lined by transitional type epithelium. A curious component of the lesion is focal Paneth cell metaplasia, which are found in the normal gastrointestinal tract, typically confined to the small bowel; however, can appear in anomalous gastrointestinal sites in various disease states.

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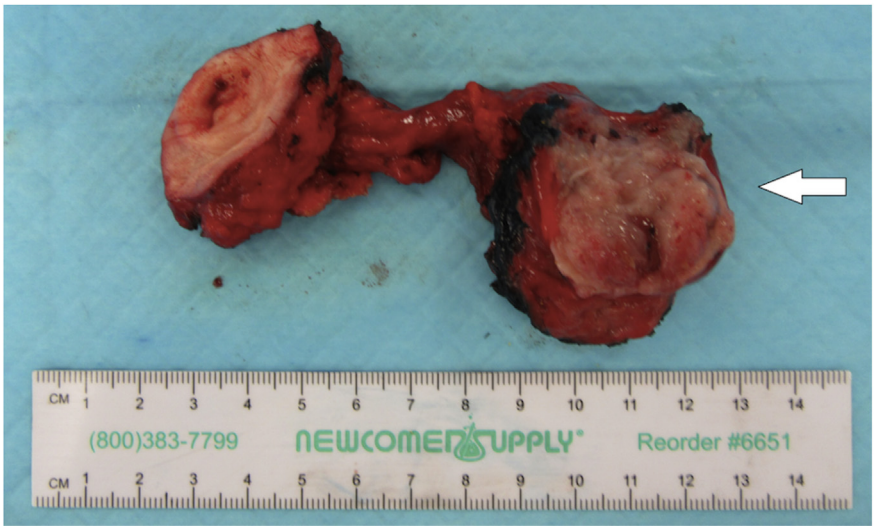

Fig. 1. Macroscopic appearance of the opened resection specimen showing the mucosal surface involved by a lesion protruding from the mucosa covered with mucin.

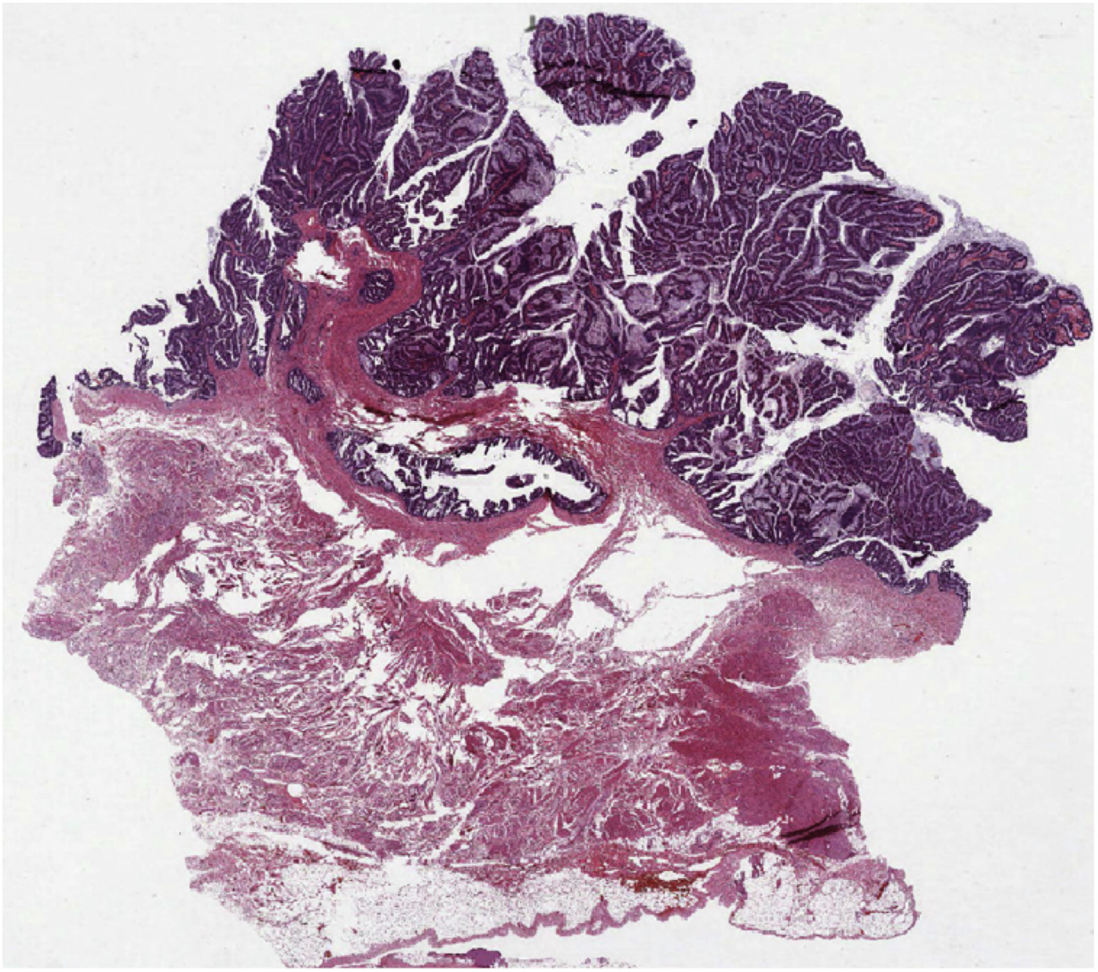

Fig. 2. Fronds of villous adenoma, H&E 10X.

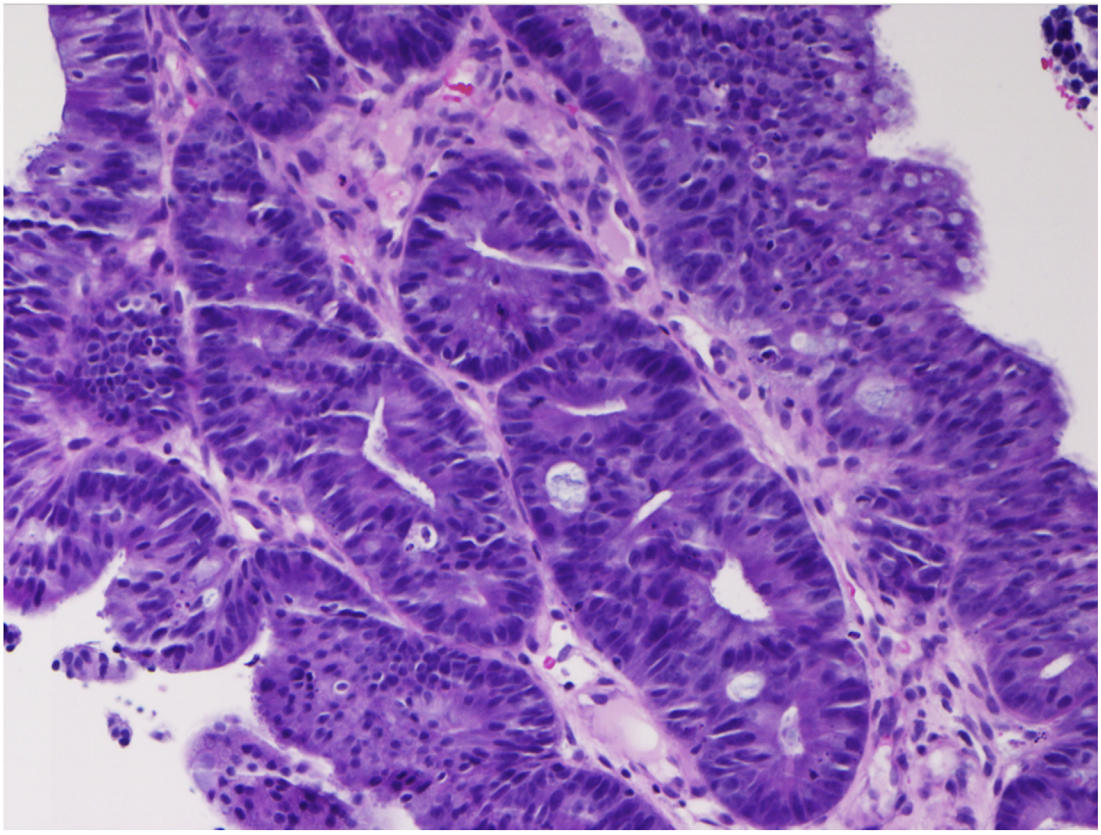

Fig. 3. Villous adenoma with high-grade dysplasia bordering on adenocarcinoma in situ.

4. Discussion

Urachal neoplasms can be differentiated as benign (adenoma) or malignant. The histologic and ultrastructural features of villous adenomas are identical to those of the colon. Seibel et al. suggest that these neoplasms arise in a precursor lesion with broad potential for neoplasia along glandular, urothelial, or sarcomatous pathways, which differ from the single pathway of differentiation in colonic adenocarcinoma.[4] Villous neoplasms arising from the uroepithelium of the urinary tract, especially from the urachus are exceedingly rare. These neoplasms predominantly occur in men over 50 years of age, with an average age of 70 years.[5] Our patient was a 51-year-old female, which is somewhat unusual for these kinds of neoplasms. The usual symptoms are hematuria, irritative voiding symptoms, and mucosuria. Villous adenomas are primarily a histological diagnosis and investigative workup including findings on renal ultrasound, computerized tomography, and magnetic resonance imaging are usually nonspecific. There are also no specific findings on cystoscopic examination.[5]

There is still uncertainty regarding the origin of villous adenoma in the urinary tract. Embryological differentiation of cloaca may give some insights into the origin of these tumors. It is postulated that cloacal remnants may remain in the urogenital sinus and may give rise to villous adenomas. Another possible theory suggested is that these tumors are the result of chronic irritation of urothelium resulting in glandular metaplasia, which in turn has potential for neoplastic transformation.[5]

Differential diagnoses to be considered include gastrointestinal tract adenocarcinoma with secondary involvement, papillary urothelial carcinoma with villoglandular differentiation, cystic glandularis with intestinal metaplasia, and papillary variant of nephrogenic adenoma. Histological examination, immunohistochemistry, immunophenotypic features, and cytogenetics can reasonably exclude all these conditions.[5]

Villous adenomas generally carry an excellent prognosis after local excision and recurrences are extremely rare. Villous adenomas with carcinoma in situ carry a good prognosis after complete surgical resection and require surveillance. Villous adenomas with coexistent adenocarcinoma usually have a worse prognosis as local recurrence and distant metastasis are possible, requiring an aggressive management and follow-up. En bloc excision of the umbilicus, urachus, and surrounding soft tissue coupled with extended partial cystectomy is recommended for urachal carcinomas.[5]

5. Conclusion

While there have been cases of reported of villous adenoma coexistent with adenocarcinoma, there has only been one report of progression of villous adenoma to adenocarcinoma.[3] Here, we present the case of a 51-year-old woman with villous adenoma with high grade dysplasia appearing as adenocarcinoma in situ, suggesting malignant transformation. Therefore, while patients with villous adenomas have an excellent prognosis, close follow up is recommended as development of infiltrating adenocarcinoma suggests a more aggressive course of disease and treatment.

Consent

The Institutional Review Board at Geisinger Medical Center reviewed and approved this study.

Conflict of interest statement

None of the contributing authors have conflicts of interest, including specific financial interests or relationships and affiliations relevant to the subject matter or materials discussed in this article.

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