

Sigmoid-Urachal-Vesical Fistula Associated with Urothelial Carcinoma in an Adult Male: Case Report¹

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We report a case of a sigmoid-urachal-vesical fistula associated with urothelial carcinoma that resulted from complications of an infected urachal remnant with a brief review of the most relevant clinical and radiological findings. The lesion presented as a supravescical enhancing mass that communicated with the sigmoid colon. In this case, the lesion was suggestive of malignant transformation of the infected urachal diverticulum in an adult male. Radiological findings are considered as helpful for the preoperative diagnosis and proper management of unusual complications of an urachal remnant.

Index words : Urachus

Diverticulum

Carcinoma, transitional cell

Urinary bladder diseases

Urachal remnants that abnormally remain patent are often subject to infection, spontaneous rupture, fistula formation or a malignant change. Sigmoid-urachal fistulas have been reported in cases of colonic diverticulitis or an infected urachal cyst (1-3); however, for these cases, there is no communication with the urinary bladder or any association with urothelial carcinoma. Although the normal urachus is most commonly lined by the transitional epithelium, an urachal carcinoma predominantly manifests as an adenocarcinoma. Urothelial carcinoma is extremely rare (4-6). We report an unusual case of

a sigmoid-urachal-vesical fistula associated with urothelial carcinoma in an adult male. For accurate diagnosis and treatment planning, radiologists should consider the presence of a coexistent malignant lesion in a complicated urachal remnant.

Case Report

A 45-year-old man presented to our hospital with pneumaturia and fecouria on voiding of two months duration. The patient had no prior abdominal surgeries. The vital signs were within normal limits. On a physical examination, there was focal percussion tenderness in the suprapubic region. The patient reported occasional dysuria and urgency but experienced no urinary frequency, incontinence or diminished urine stream. The patient denied having diarrhea, constipation, hematochezia, melena, and abdominal cramping. The leukocyte count was elevated at 18,600 per millimeter. Urinalysis revealed 2+ leukocyte esterase and a blood and microscopic examination demonstrated 3-5 red

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blood cells and many white blood cells per high-power field. The serum electrolyte level and creatinine level were within normal limits.

Contrast-enhanced computed tomography (CT) scans of the abdomen and pelvis demonstrated the presence of a 2.5 cm sized heterogeneously enhanced urachal le-

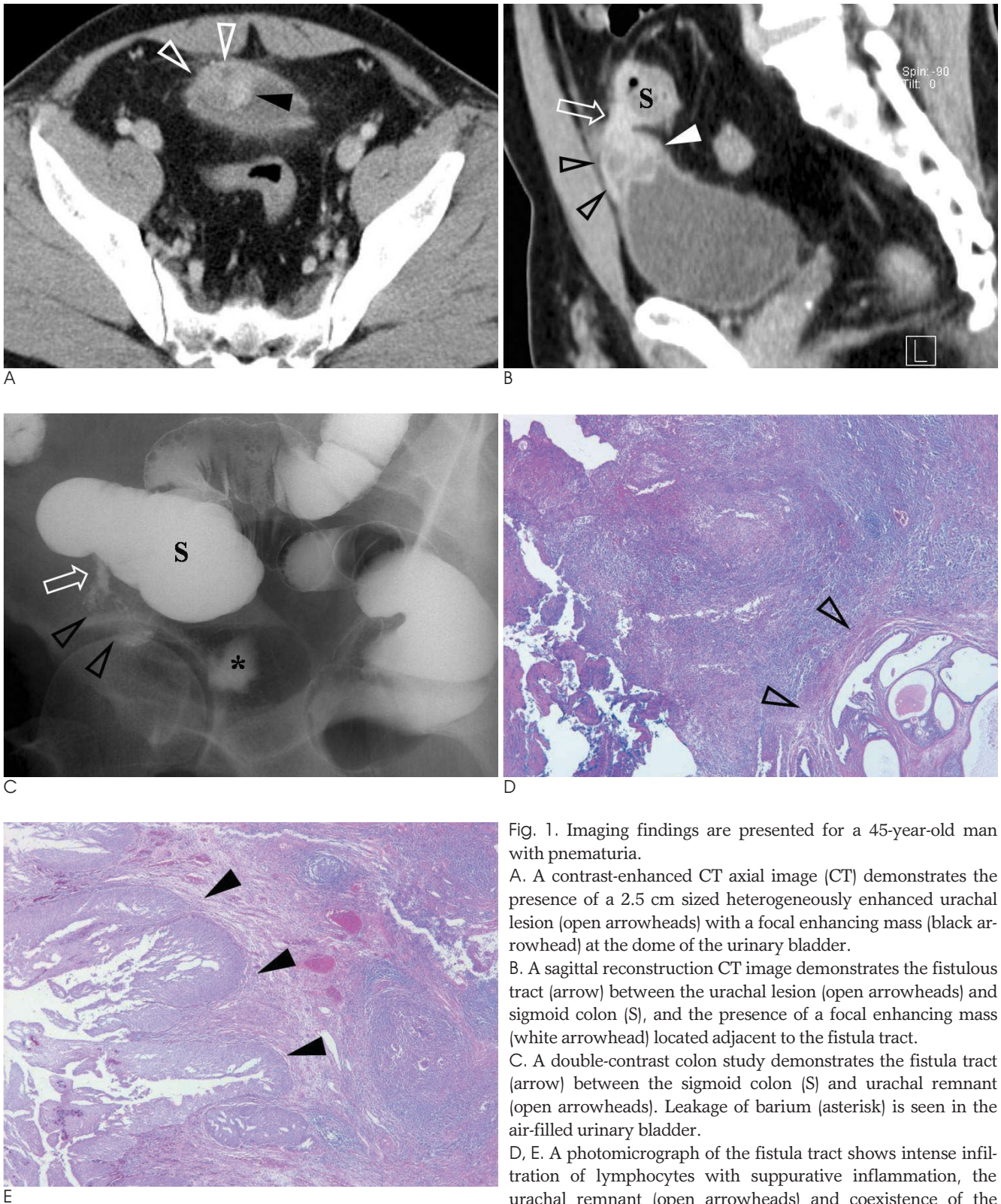


Fig. 1. Imaging findings are presented for a 45-year-old man with pneumaturia.

A. A contrast-enhanced CT axial image (CT) demonstrates the presence of a 2.5 cm sized heterogeneously enhanced urachal lesion (open arrowheads) with a focal enhancing mass (black arrowhead) at the dome of the urinary bladder.

B. A sagittal reconstruction CT image demonstrates the fistulous tract (arrow) between the urachal lesion (open arrowheads) and sigmoid colon (S), and the presence of a focal enhancing mass (white arrowhead) located adjacent to the fistula tract.

C. A double-contrast colon study demonstrates the fistula tract (arrow) between the sigmoid colon (S) and urachal remnant (open arrowheads). Leakage of barium (asterisk) is seen in the air-filled urinary bladder.

D, E. A photomicrograph of the fistula tract shows intense infiltration of lymphocytes with suppurative inflammation, the urachal remnant (open arrowheads) and coexistence of the urothelial carcinoma (black arrowheads) (hematoxylin and eosin staining, $\times 200$).

sion containing a focal enhancing mass at the dome of the urinary bladder (Fig. 1A). A sagittal reformed CT image demonstrated the fistula tract between the urachal lesion and the sigmoid colon (Fig. 1B). A cystogram showed the normal bladder contour without extravascular leakage of contrast media. Cystoscopy demonstrated no pathological findings. Bladder washing cytology was negative for malignancy and showed many neutrophils and a few degenerated urothelial cells. The use of barium enema confirmed the presence of the sigmoid-vesical fistula tract, and leakage of barium and air into the urinary bladder (Fig. 1C).

On the next day, an exploratory laparotomy was performed. As determined from surgery, a fistulous tract from the sigmoid colon to the infected urachal remnant that contained the mass lesion was present, along with severe perivesical adhesion. A partial cystectomy with en bloc tumor resection, adhesiolysis of the mesocolon and segmental resection of the sigmoid colon and excision of the fistula were performed. The resection specimen showed a 1.2 cm sized polypoid tumor adjacent to the fistula tract. A histological examination demonstrated that the transmural fistulous tract consisted of intense lymphocytic infiltration with acute and chronic suppurative inflammation, an omphalomesenteric duct remnant and a coexistent low-grade papillary urothelial carcinoma (Fig. 1D, E). The surgical resection margins were free of carcinoma. The postoperative course was uneventful, and the patient did not display any bowel or bladder dysfunction. At 14 days after surgery, the patient was discharged from the hospital with a chemotherapy plan.

Discussion

The urachal diverticulum communicates only with the bladder dome and is asymptomatic in most cases. This lesion tends to be found in patients with chronic bladder outlet obstruction and may be complicated with a urinary tract infection, intraurachal stone formation and an increased prevalence of carcinoma (7). The route of infection of urachal remnants may be via the lymphatics, hematogenous or vesical. A wide variety of gram-positive and gram-negative microorganisms have been cultured from infected urachal remnants (7). A colo-urachal or colo-vesical fistula is a very rare disease entity, and colonic diverticulitis, inflammatory bowel disease, gastrointestinal or genitourinary neoplasms, post-radiation therapy, pelvic surgery and foreign bodies

have also been implicated as causes for fistula formation (2, 8). In the present case, the fistula tract was detected between an infected urachal diverticulum and the normal colon, and the colonic contents subsequently drained into the urinary bladder.

The clinical presentation of a colo-vesical fistula may include recurrent cystitis, pneumaturia, fecouria, fever and abdominal pain. CT is the primary imaging modality to utilize for suspected cases of urachal complications, and intravesical air, focal bladder wall thickening and extraluminal masses are highly suggestive findings for a colo-vesical fistula (2, 8). Furthermore, if an intravenous contrast agent is not used, the presence of an enteric contrast agent in the bladder as seen on CT images is diagnostic of a fistula (8). MR imaging and ultrasonography have been used to delineate colo-urachal fistulas. The use of a barium study helps confirm the nature, location and extent of the fistula tract, but a barium edema study demonstrates only 35% of fistulas (8). The use of cystoscopy may fail to demonstrate a urachal fistula or cancer, and a cystogram is less sensitive.

Malignant urachal neoplasms are also rare, representing less than 0.5% of all bladder cancers; most cases are adenocarcinomas (4-7). Squamous cell carcinomas and urothelial carcinomas each comprise approximately 3% of urachal malignancies, and sarcomas comprise 5 to 10% of urachal malignancies (6, 7). Urachal tumors are typically silent due to an extraperitoneal location and extending superiorly toward the umbilicus; consequently, the majority of patients exhibit local invasion or metastatic disease at the time of presentation (7, 9). Urachal carcinomas may be confused with primary tumors of the bladder dome; unlike vesical tumors, however, urachal tumors have a propensity to grow in the perivesical space toward the umbilicus and are found located in the middle of the urachus or near the umbilical end (7, 8). A primary bladder carcinoma that arises in the mucosa of the bladder apex will usually manifest with less of an extravascular component as compared to an urachal cancer (7).

Due to the lack of specific CT findings of urachal tumors, infected urachal remnants mimic those of urachal carcinoma. Discrete perilesional spiculation or fat stranding is suggestive of tumor infiltration, but is not specific, as many cases of infection have demonstrated perilesional inflammatory spread (7). The presence of an unencapsulated caudal portion of the tumor that involves a portion of the bladder wall and often of a cystic encapsulated supravascular portion with a midline posi-

tion are considered highly characteristic as well (4, 7). In addition, calcification occurs in 50 to 70% of cases and low attenuation components reflecting the mucin content are commonly detected for a urachal carcinoma (4). In our case, the use of CT imaging demonstrated the presence of a complicated urachal abscess containing a contrast-enhanced mass without calcifications or an intravesical component. To the best of our knowledge, no report in the English language literature has described the CT appearance of an unusual urachal fistula associated with urothelial carcinoma. Although cystoscopy is a reliable method for the diagnosis of a bladder lesion, an extravesical mass with a fistula is inaccessible or may be missed, particularly for a fistula with a narrow orifice.

Although the prognosis for an urachal tumor is related to the stage and degree of differentiation, it is generally poor as an urachal tumor arises in a clinically silent area and is discovered only after it has extended into the bladder lumen or has manifested with symptoms related to a large size or extension into adjacent organs (7). Surgery is the treatment of choice for an urachal carcinoma. Although many investigators have advocated the use of a radical cystectomy, it appears that long-term disease specific survival can be achieved with a partial cystectomy with en bloc resection of the bladder dome; removal of urachal ligament and umbilicus is advisable (10).

Complications of the urachal diverticulum are rarely observed clinically and complications give rise to problems such as infection and late malignant changes in

adults. Although preoperative differential diagnosis of an infected urachal lesion or carcinoma is not possible, the presence of a contrast-enhanced focal mass without calcification in a complicated urachal remnant is a useful feature for the diagnosis of an associated urothelial carcinoma.

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성인 남성에서 발생한 요로상피암을 동반한 S자결장-요막관-방광루¹

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잔류요막관의 감염 합병증으로 발생한 S자 결장-요막관-방광루와 동반된 요로상피암의 예에서 의미 있는 임상적, 방사선학적 소견에 대한 간략한 고찰을 보고하고자 한다. 성인 남성에서 발생한 병변은 S자 결장과 연결된 방광 상부 종괴로서, 감염된 요막관계실의 악성 변형을 시사하는 증례이다. 방사선학적 소견들은 잔류요막관의 드문 합병증의 수술 전 진단과 적절한 치료에 유용할 것으로 생각한다.