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

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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 supravesical 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
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, × 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 cys-
togram showed the normal bladder contour without extravesical leakage of contrast media. Cystoscopy demon-
strated no pathological findings. Bladder washing cytol-
ogy was negative for malignancy and showed many
neutrophils and a few degenerated urothelial cells. The
use of barium enema confirmed the presence of the sig-
omoid-vesical fistula tract, and leakage of barium and air
into the urinary bladder (Fig. 1C).

On the next day, an exploratory laparotomy was per-
formed. 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 exci-
sion of the fistula were performed. The resection speci-
men showed a 1.2 cm sized polypoid tumor adjacent to
the fistula tract. A histological examination demonstrat-
ed that the transmural fistulous tract consisted of in-
tense lymphocytic infiltration with acute and chronic
suppurative inflammation, an ophalmomesenteric 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 pa-
tient was discharged from the hospital with a chemothe-
rapy 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 co-
o-lo-urachal or colo-vesical fistula is a very rare disease en-
tity, and colonic diverticulitis, inflammatory bowel dis-
eease, 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 nor-
mal 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 modali-
ty 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 di-
agnostic of a fistula (8). MR imaging and ultrasonogra-
phy have been used to delineate colo-urachal fistulas.
The use of a barium study helps confirm the nature, lo-
cation and extent of the fistula tract, but a barium ede-
ma 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, represent-
ing 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 ex-
tending 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 tu-
mors of the bladder dome; unlike vesical tumors, how-
ever, urachal tumors have a propensity to grow in the
perivesical space toward the umbilicus and are found lo-
cated 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 extravesical component as compared to
an urachal cancer (7).

Due to the lack of specific CT findings of urachal tu-
mors, 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 in-
volves a portion of the bladder wall and often of a cystic
encapsulated supravesical 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.

References

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대한영상의학회지 2009·60·423-426

성인 남성에서 발생한 요로상피암을 동반한 S자결장-요막관-방광루

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천료요막관의 감염 혈종증으로 발생한 S자결장-요막관-방광루와 동반된 요로상피암의 예에서 의미 있는 임상적, 방사선학적 소견에 대한 간략한 고찰을 보고하고자 한다. 성인 남성에서 발생한 병변은 S자결장과 연결된 방광 상부 중피로서, 감염된 요막관관계의 악성 변형을 시사하는 증례이다. 방사선학적 소견들은 천료요막관의 드문 혈종증의 수술 전 진단에 적절한 치료에 유용할 것으로 생각한다.