CASE REPORT

# Inflammatory pseudotumour of the urachus: Case report and literature review

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## Abstract

A 52 year old woman presented to the emergency department of Affiliated Hospital of Zunyi Medical University, Zunyi, China in May 2022, complaining of a palpable lower abdominal mass since two days. She denied haematuria, umbilical drainage, or any other urinary symptoms. Previous health record indicated that the patient was diagnosed with urachal inflammatory pseudotumour. Inflammatory pseudotumourous masses of the urachal canal are rare chronic inflammatory disorders with only a few case reports. Ultrasonography is the preferred method for diagnosing urachal lesions. Contrast-enhanced ultrasonography (CEUS) allows real-time visualization of the microvascular blood flow within the solid lesion, reducing the probability of misdiagnosis of the disease. We have reported a case of urachal inflammatory pseudotumour and analyzed its ultrasonographic findings from two-dimensional conventional ultrasonography and CEUS to provide support for the diagnosis of urachal inflammatory pseudotumour in the clinic and to assist clinical selection of effective treatment modalities.

**Keywords:** Urachus, Inflammatory pseudotumor, Ultrasonography, CEUS.

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## Introduction

The urachus is a tubular structure that connects the bladder to the umbilicus and is the remnant of the cloaca and allantoic duct during embryonic development. According to literature studies, urachal masses are mostly malignant, among which mucinous adenocarcinoma is the most common; however, the masses can also be benign lesions, wherein urachal inflammatory pseudotumour (IPT) is an extremely rare benign lesion. No specific clinical symptoms are often observed, and the detection of the disease is not easy in its early stages. The imaging manifestations of IPT of the urachal are similar to those of urachal carcinoma, and differentiating between the two is difficult. At present, only a few cases have reported the imaging features of IPT of the urachus, particularly in contrast-enhanced ultrasonography (CEUS); however, there is no relevant literature report. This case study is a report of CEUS manifestations of urachal IPT.

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## Case Report

A 52-year-old female patient was presented to the emergency department of Affiliated Hospital of Zunyi Medical University of China in May 2022 complaining of a lower abdominal mass since two days. The patient denied haematuria, umbilical drainage, or any other urinary symptoms. On physical examination, a firm mass in the lower abdomen was palpated, and the position was fixed. After admission, the patient's vital signs were stable; blood routine, liver function, and renal function were normal; and urine culture showed no obvious abnormality. Pelvic computed tomography examination showed uneven thickening and density of the bladder wall, and a soft tissue mass was observed below the anterior abdominal wall, which seemed to be connected to the urachal lesion. The boundary was unclear, and the largest cross-sectional area was approximately 44 × 41 mm (Figure-1). The enhanced scan was clearly unevenly enhanced, indicating that presence of possible urachal tumour or infection. The ultrasound examination of the patient showed an uneven mass of approximately 60 × 35mm (Figure-2A) in size in the abdominal wall. The boundary was unclear, the shape was still regular, and a little irregular liquid dark area was visible inside. The mass was connected to the anterior wall of the bladder that showed irregular thickening with uneven echogenicity, the medial wall margin was regular and clear, and the lateral wall margin was less clear and regular. Colour Doppler flow imaging (CDFI) (Figure-2B) showed

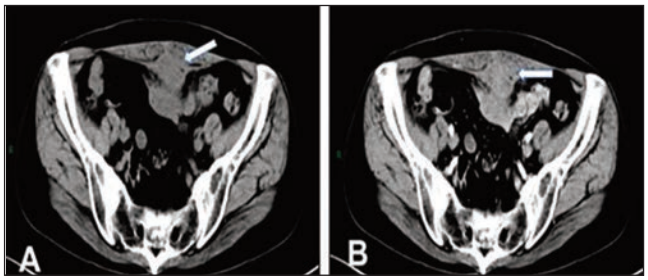


**Figure-1:** A) CT image of the pelvic shows a soft-tissue space occupying mass (Headed arrow) measuring 44×41 mm (AP x TR ) with seemingly connected cord urachal is seen below the anterior abdominal wall, ,and the mass is not clearly demarcated. Contrast-enhanced scan B) shows that the mass is markedly heterogeneously enhanced. AP, anteroposterior; TR, transverse.

punctate and linear blood flow signals inside the mass, and the blood flow signal was more abundant in the periphery of the mass.

CEUS (Figure-3) examination was performed to observe that the perfusion of the ultrasound contrast agent started to appear within the mass at the eighth second after injection of the contrast medium, and its perfusion volume peaked at the 18th second that began to subside after the second minute and seventh second. The mass showed slightly heterogeneous hyperenhancement in the arterial phase, and a little area of perfusion defect internally and slight hyperenhancement in the venous phase on CEUS were observed.

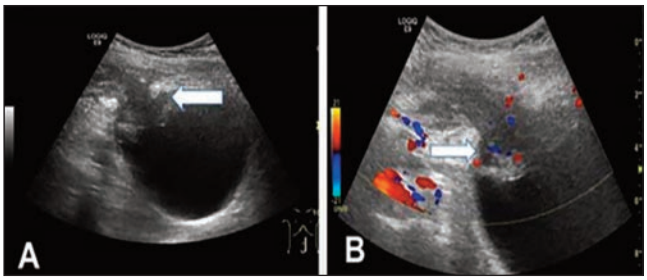


**Figure-2:** A) Two-dimensional ultrasound (2D) shows a hypoechoic heterogeneous mass (Headed arrow) in the anterior and upper part of the bladder, which is connected to the thickened bladder wall, with poorly defined borders, regular shape, and irregular liquid dark areas. B) Colour Doppler flow imaging (CDFI): dot-line blood flow signals are seen in the mass (Headed arrow), and peripheral blood flow signals are abundant.

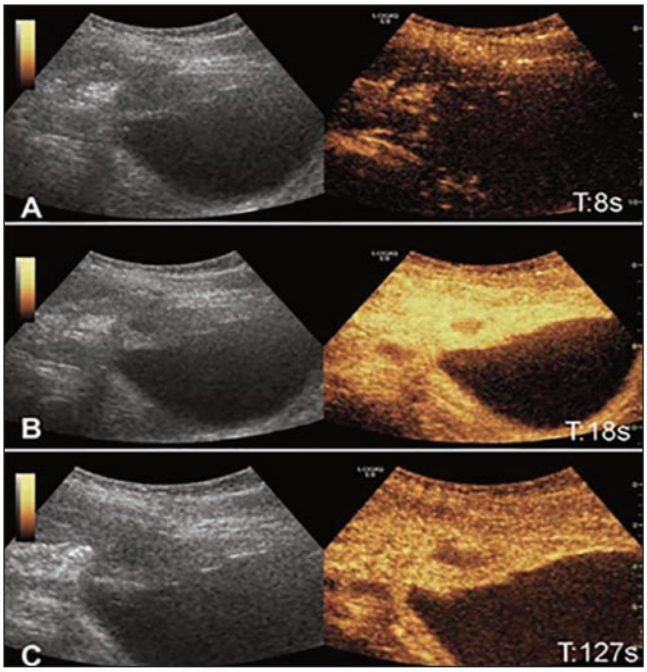


**Figure-3:** Contrast-enhanced ultrasonography (CEUS) examination showed that A) After the injection of the contrast agent, the perfusion of the ultrasound contrast agent began at 8 seconds, slightly earlier than the surrounding normal tissues (bladder wall, cervix). B) The peak was reached at the 18th second, the lesion showed slightly uneven high enhancement, the boundary was not clear, and a few perfusion defect areas were seen inside. C) After 2 minutes and 7 seconds, the contrast agent began to subside, the lesions showed a slightly higher enhancement, and the contrast agent subsided slowly.

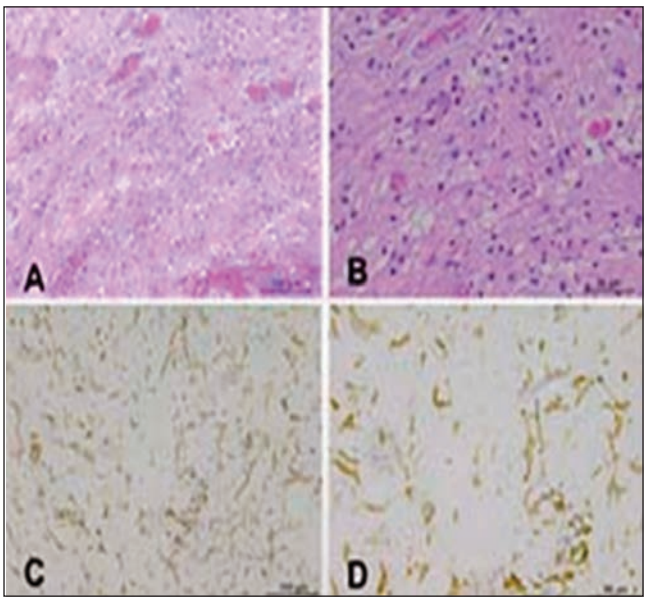


**Figure-4:** A B) Haematoxylin and eosin (H&E) staining image of examined section shows fibrous connective tissue hyperplasia with abundant chronic inflammatory cells, including lymphocytes, plasma cells, and eosinophils, and dilatation of the urachium can be seen, but without atypical cells. Original magnifcations,×20 (A),× 40 (B). C,D) Immunohistochemistry (IHC) image of examined section shows that SMA was present in tissue cells, but Catenin, CD34, CDK4, MDM2, and P16 were not expressed. Original magnifcations,×20(C),× 40 (D).

The ultrasound appearance of the thickened anterior bladder wall was typically consistent with that of this mass. Compared with the surrounding normal tissues (the bladder wall and cervix), the enhancement of the mass was slightly earlier than that of the normal tissue; however, the contrast agent fades from the mass more slowly, the mass after angiography appears less clear, and the shape is still more regular.

Subsequently, the patient underwent surgical resection of the mass, and intraoperatively, the pathology of the tissue sent for review revealed a benign lesion. Postoperative pathology of the mass showed fibroplasia, granulation tissue formation, invasion of chronic inflammatory cells, chronic nonspecific inflammatory changes, and localized abscess formation. Postoperative immunohistochemistry (Figure 4) of the mass detected chronic inflammatory cell infiltration with fibrosis and was catenin (–), CD34(–), CDK4(–), MDM2(–), P16(–), and SMA (+). The results of immunohistochemistry supported it to be an IPT such as hyperplasia.

## Discussion

The urachus is a tube-like structure connecting the bladder and the umbilicus, which is the remnant of cloacal and allantoic catheters from the embryonic period.[2] By 4–5 months of gestation, the bladder is gradually invaginated into the pelvis, and the apical portion is gradually reduced to form a tube-like structure, the umbilical urethra. Lesions in the urachus can occur at any site along the cord, and masses in the cord are rare in clinical practice, reporting for 0.01% of all adult tumors and 0.17-0.34% of all bladder tumours.[1-3] Clinically, gross haematuria was the most crucial predictor of malignancy, increasing the risk for urachal cancer upto 17-fold, according to previously published literature.[4] Therefore, a mass without gross haematuria manifestations tends to be a benign diagnosis. Considering patient's age, the likelihood of malignancy of a mass in the umbilical canal tripled when the patient's age at diagnosis was more than 55 years.[5] In addition, men have a significantly higher risk of developing urachal carcinoma, more than twice that in women.[6] On the basis of the above factors, our patient had no clinical manifestations such as gross haematuria and was a female and not older than 55 years, thus less likely to have malignant urachal lesions, which was in accordance with previously published studies. IPT appears as a very rare lesion in extrapulmonary locations. In 1939, Brun reported the first case of IPT in the lung.[7] IPT has been reported in the literature to be a nonneoplastic inflammatory lesion characterized histopathologically by fibrohistiocytic proliferation and by the presence of myofibroblasts and capillaries, interspersed with more proliferating histiocytes, polyclonal plasma cells and chronic inflammatory cell infiltration such as lymphocytes and eosinophils. Immunohistochemically, smooth muscle antibody expression was positive in the majority of IPT, including urologic IPT and catenin, Ki-67, CD68, etc. were not expressed.[8] In our case study, postoperative histopathology of this patient revealed the abdominal wall mass as an IPT of the urachal carcinoma. Clinically, IPTs of the urachus are rare and the pathogenesis remains unclear. Previous surgery, radiation exposure, trauma, immune disorders, infection, or inflammatory processes are the possible etiological factors for the disease.[9] The mass lacks specific clinical manifestations when it cannot compress or invade the bladder. Ultrasonography, being the first method for diagnosing the urachus, can help in observing the anatomical structure of the urachal throughout the entire process and determining whether the umbilical remnant has sinus tract formation. Its size, course, and other features can be noted on ultrasonography; however, it is often difficult to distinguish it from urachal carcinoma in clinical practice.

Two-dimensional (2D) ultrasound findings of IPT are diverse and show them to be mostly irregular in shape, with a portion of the lesion traversed interiorly through small vessel branches and a surrounding silent halo. The borders are sharp and regular, and the interior is mostly hypoechoic with uneven distribution. Sometimes, there may be enhanced light spots in the centre of the hypoechoic area, whereas posterior echogenicity rarely changes[10] In addition, the IPT of the urachus often infiltrates the bladder dome tissue. CDFI showed IPT had mostly no colour blood flow signal, whereas a few strip-shaped blood flow signals could be detected in some of the lesions mostly located in the peripheral part of the lesion. Conventional ultrasonography of our patient revealed a heterogeneous mass with poorly defined borders and irregular morphology. The mass extended to the anterior wall of the bladder, which then thickened and the echo was inhomogeneous whereas the medial wall of the bladder was irregular and clear. CDFI showed punctate and linear blood flow signals inside the mass, and blood flow signal in the periphery was more abundant. This may be associated with the infiltration of the surrounding tissue by a large number of inflammatory cells.

Urachal carcinoma (UC) is a rare malignant epithelial tumour of unknown pathogenesis, arising primarily from the residual tissue of the urachal canal and growing infiltratively into the bladder and its surrounding tissues.[5,11] Ultrasonography of urachal carcinoma revealed that it is mostly hypoechoic, and the echo of the mass is heterogeneous. The mass is generally sessile, with a wide base, and often infiltrates the full thickness of the bladder wall and surrounding tissues. The funnel-shaped upper part of the mass is a characteristic change of urachal carcinoma. Punctate calcifications can be observed in some urachal carcinomas and have been considered as one of the typical features of the disease in the literature.[4,7] The mass in our case study extended into the bladder, making the anterior wall of the bladder thick and more echogenic; however, the medial wall of the bladder was still intact. The irregular thickening of the bladder wall with incomplete bladder wall continuity can be due to possible invasion of the bladder by carcinoma of the umbilical urethra.

CEUS visualizes microbleeds inside solid lesions,[12] improving the diagnostic performance of ultrasonography. IPT has a more characteristic sonographic appearance than cord urachal carcinomas, and Ding et al.[13] reported the sonographic appearance of urachal carcinomas as a pattern of "syngeneic early regression and low enhancement," that is, the mass appears hypo enhanced, the intensity of the developed mass is lower than the peripheral bladder wall, it typically appears synchronous or faster than the

peripheral bladder wall, the regression of the contrast agent is faster, it shows abundant blood supply, and the perfusion of the contrast agent is heterogeneous.[10] In this case study, the sonogram showed hyperenhancement at early perfusion; however, perfusion was more homogeneous. In addition, compared with its own bladder wall, after injection of the contrast medium, the initiation of perfusion of the contrast medium is slightly earlier than the normal bladder wall, the time to reach the peak intensity and the time of clearance are longer than the bladder wall, and the time of contrast medium clearance is more than the bladder wall, and ultrasonography examination showed that the tumour mass is "fast and slow out," which is different from the "same progress or fast out" of cord urinary tract cancer. These manifestations may occur because of the invasion of chronic inflammatory cells of the soft tissue, the formation of granulation tissue leading to an increase in newly formed capillaries, and a relative paucity of free water within fibrotic lesions. In contrast, hypo- and non-enhancing areas in the interior of the mass were observed on ultrasonography, which may be associated with necrotic liquefaction appearing in the interior of the IPT of the umbilical canal. In our case study, a greater possibility of IPT of the urachus was considered. Along with the sonographic findings of our case study and a review of the literature, the aforementioned sonographic changes have some significance in the diagnosis of IPT of the urachus and also facilitate in differentiating them from malignant tumours of the urachus. Ultrasonography can relatively accurately show the size, morphology, and internal echogenicity and can dynamically observe the relationship between the tumour and the bladder and abdominal wall. CDFI can reveal the blood supply of the tumour and combine age, sex, and clinical manifestations of the patients, which along with the ultrasonographic findings, are helpful in differentiating benign from malignant tumours.

## Conclusion

In summary, although it is difficult to differentiate IPT occurring in the urachus from carcinoma of the urachus using conventional ultrasonography. The disease can be better diagnosed by analyzing the findings of conventional ultrasonography combined with CEUS and through a comprehensive analysis of clinical data. Therefore, ultrasonography findings play a crucial role in the differential diagnosis of masses in the urachus to guide further clinical treatment.

**Consent Form:** Patient's consent was obtained for publishing the case for science promotion.

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**Conflict of Interest:** None.

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