

Urachal remnant with heterotopic sinus in an adult male

A case report

Zhao-Hui Sun, MB, Xiang-Hui Kong, MB, Wen-Jie Huang, MD, PhD, Gang Chen, MM, Xiao-Jun Huang, MM*

Abstract

Introduction: Urachal remnant with heterotopic sinus is an extremely rare congenital anomaly, and usually coexists with other congenital anomalies. We report the case of a 32-year-old adult male with urachal remnant with heterotopic sinus.

Patient concerns: A 32-year-old adult male presented with purulent secretion in the heterotopic sinus on the dorsal side of the normal external urethral orifice and pain in the balanus since 5 months.

Diagnosis: The computed tomography scan demonstrated a 4 cm cystic mass next to the anterior wall of the urinary bladder. Retrograde urethrography was performed, which demonstrated that this mass communicated with the heterotopic sinus on the dorsal side of the normal external urethral orifice. Cystoscopy showed that there was no communication between the mass and the bladder. Pathology results confirmed that this mass was urachal tissue.

Interventions: The patient underwent a laparoscopy surgery to undertake the cystic mass, part of the anterior wall of urinary bladder and the epithelium of channel which communicated with the cystic mass.

Outcomes: The patient was discharged without any complications after 6 months and follow-up was continued in the clinic.

Conclusions: Urachal remnant with heterotopic sinus is rare, and we recommend that urachal remnant should be considered when a patient presents with a mass in the retropubic space.

Abbreviation: CT = computed tomography.

Keywords: heterotopic sinus, urachal anomaly, urachal remnant

1. Introduction

Urachal remnant is a rare congenital anomaly of the urinary system. It will cause many complications including becoming cancerous and is harmful to people's health. The main clinical manifestation of urachal remnant is umbilical drainage according to the relevant published data. However, there are only 2 cases reported on the urachal remnant with heterotopic sinus on the dorsum of the penis.^[1] Here we report a case of urachal remnant with heterotopic sinus on the dorsal side of the normal external urethral orifice.

Editor: N/A.

The authors have no conflicts of interest to disclose.

The 2nd Clinical Medical College of Zhejiang Chinese Medical University, Hangzhou City, Zhejiang Province, China.

* Correspondence: Xiao-Jun Huang, Department of Urology, The Second Affiliated Hospital of Zhejiang Chinese Medical University, No.318 Chaowang Road, Gongshu District, Hangzhou 310005, P.R. China, Zhejiang Provincial Key Laboratory of Traditional Chinese Medicine, Hangzhou, Zhejiang, China (e-mail: hxj258111@163.com).

Copyright © 2019 the Author(s). Published by Wolters Kluwer Health, Inc. This is an open access article distributed under the terms of the Creative Commons Attribution-Non Commercial License 4.0 (CCBY-NC), where it is permissible to download, share, remix, transform, and buildup the work provided it is properly cited. The work cannot be used commercially without permission from the journal.

Medicine (2019) 98:18(e15430)

Received: 30 November 2018 / Received in final form: 26 March 2019 / Accepted: 2 April 2019

<http://dx.doi.org/10.1097/MD.00000000000015430>

2. Case report

The written informed consent was obtained from the patient for publication of this case report and accompanying images. A 32-year-old adult male presented with purulent secretion in the heterotopic sinus on the dorsal side of the normal external urethral orifice and pain in the balanus since 5 months. The patient had no prior history of urinary tract infection. He had undergone surgery for congenital malformations of his feet more than 20 years ago. On physical examination, there was a heterotopic sinus on the dorsal side of the normal external urethral orifice (Fig. 1A). Urine culture revealed the presence of *Pseudomonas fluorescens* in the secretion from the heterotopic sinus. The computed tomography (CT) scan demonstrated a 4 cm cystic mass next to the anterior wall of the urinary bladder (Fig. 1B). Retrograde urethrography was performed to understand the nature of this mass further, which revealed a communication between this mass and the heterotopic sinus on the dorsal side of the normal external urethral orifice (Fig. 2A). Cystoscopy showed that there were no communication between the mass and the bladder. Urodynamic study demonstrated the function of bladder was normal.

During laparoscopy, methylene blue was injected into the cystic mass using a 6F urethral catheter through the heterotopic sinus. The dissection was difficult because of severe adhesions between the cystic mass and the bladder; hence, we removed the cystic mass and part of the anterior wall of urinary bladder. We also closed the heterotopic sinus. Histopathological examination confirmed that this mass was urachal tissue (Fig. 2B). After the

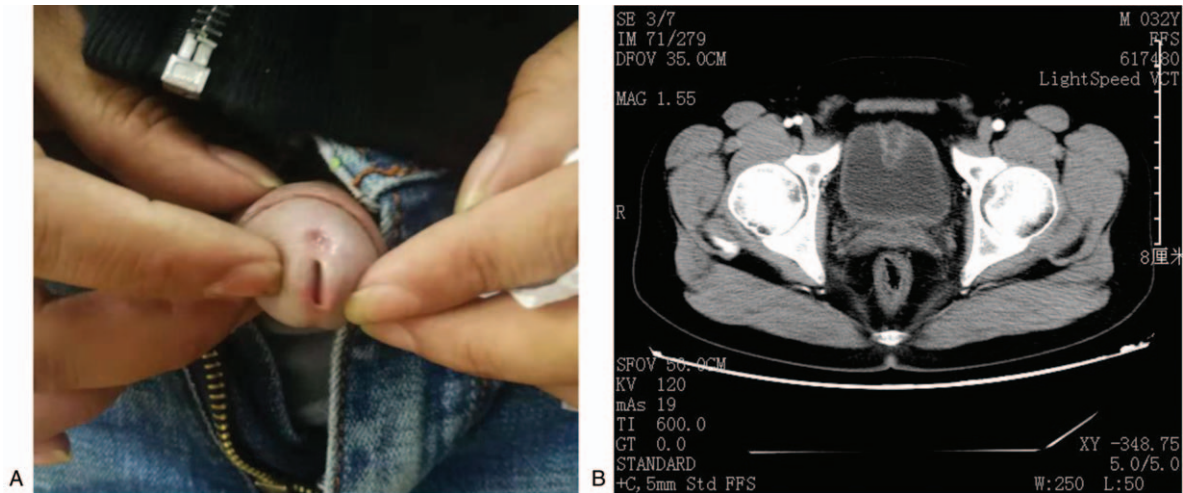


Figure 1. (A) Heterotopic sinus on the dorsal side of the normal external urethral orifice. (B) CT scan demonstrates a cystic mass next to the anterior wall of urinary bladder. CT=computed tomography.

patient was discharged from the hospital, we conducted a 6-month follow-up to him by telephone and the recent results showed that the patient was discharged without any complications. We will follow-up by telephone every 6 months until 5 years after surgery.

3. Discussion

The urachus is located in the center of a pyramid-shaped space outside the peritoneum. This space has its base on the anterior dome of urinary bladder, and the tip is directed toward the umbilicus. The length of the urachus ranges from 3 to 10 cm, and the diameter ranges from 8 to 10 mm.^[2] It is connected by 1 or 2 closed umbilical arteries. The urachus is surrounded by the umbilicovesical fascia, which means that a disease of the urachus usually occurs inside the pyramid-shaped space. If the urachus remains either open or partially obliterated, different urachal anomalies are seen.

The Fox's classification, which identifies 4 types of urachal anomalies, is most commonly used; patent urachus (50%): it might be caused by retubularization, rather than as a primary patency. This type is more common in newborn babies and presents with continuous or intermittent drainage of fluid from the umbilicus. Umbilical-urachus sinus (15%): In this type, the urachus remains open at the umbilical site and closed at the bladder level. It has a similar manifestation to the patent urachus. It can occur at any age and presents with a purulent secretion. Urachal cyst (30%): The cyst has no communication with the bladder or umbilicus. If there are no complications, there will be no symptoms for life. Once infected, it can manifest as lower abdominal pain, voiding symptoms, or a painful and palpable mass. Vesicourachal diverticulum (3–5%): Except at the level of the bladder apex, the urachus obliterates completely. This type usually has no symptoms because the diverticulum communicates with the bladder and drains into the bladder well. If the neck is narrow, it may cause stone formation and urinary tract

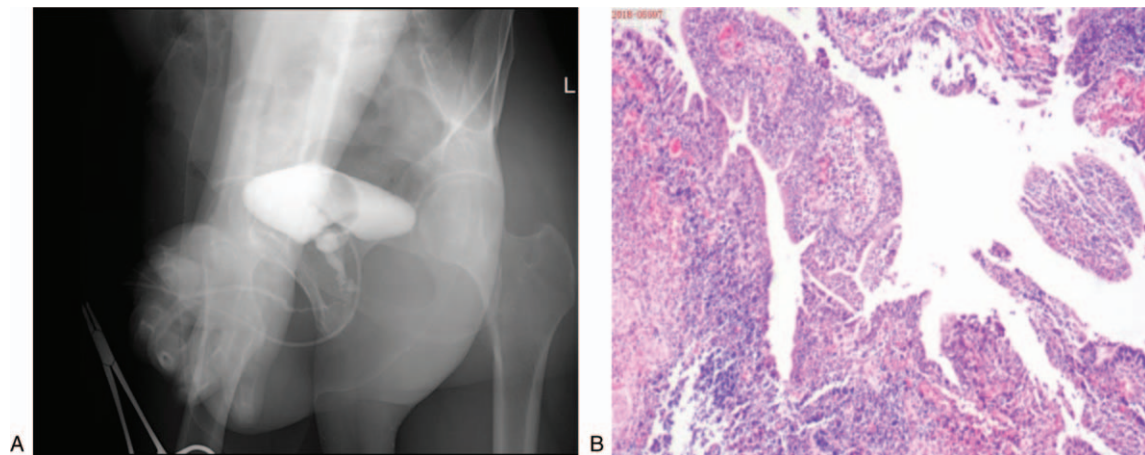


Figure 2. (A) Retrograde urethrography demonstrates communication of the mass with the heterotopic sinus on the dorsal side of the normal external urethral orifice, and no communication between the mass and the bladder.(B) A cystic structure is surrounded by smooth muscle tissue with numerous lymphocytes and neutrophils.

infections. Rich adds another type of urachal anomalies, which is named as the alternating sinus: it is a cyst-like structure that can drain to either the bladder or the umbilicus.^[3] There are only 2 cases reporting the urachal remnant with heterotopic sinus on the dorsum penis. Based on the observations during surgery and the histopathological results, we diagnosed of our case as the alternating sinus.

Diagnosing an urachal anomaly can be complicated. A careful history and physical examination is important. Using the ultrasound, CT scan, retrograde urethrography, and voiding cystourethrogram can be helpful.^[4,5] In our case, the CT scan demonstrated a 4 cm cystic mass next to the anterior wall of the urinary bladder. The retrograde urethrography demonstrated that this mass communicated with the heterotopic sinus on the dorsal side of the normal external urethral orifice, which led us to consider the possibility of an urachal anomaly. The pathology results confirmed our assumption. It is worth mentioning that the possibility of associated genitourinary defects must be considered because they can be more harmful than the urachal anomaly itself. In our case, the patient had undergone a surgery for the congenital malformations of the 2 feet more than 20 years ago. Further examination did not reveal any associated genitourinary defects.

Once the diagnosis of urachal anomaly is confirmed, the urachus and other tissues must be removed immediately because the remnants can be cancerous.^[6–8] If there are some complications such as infection, the surgery should be performed after the infection is controlled.^[9] During the surgery, we can inject methylene blue to identify the direction of the drainage. However, the diseased tissue must be completely removed.^[7,8] The superficial arteriovenous vein and dorsal nerve should be well protected when dissociating and resecting the fistula.

Urachal remnant with heterotopic sinus is rare, and we recommend that urachal remnant should be considered when a patient presents with a mass in the retropubic space.

Author contributions

Data curation: Zhao-Hui Sun, Xiang-Hui Kong, Wen-Jie Huang, Gang Chen.

Writing – original draft: Zhao-Hui Sun, Xiang-Hui Kong, Wen-Jie Huang.

Writing – review & editing: Xiao-Jun Huang.
Zhao-Hui Sun orcid: 0000-0003-2294-7476.

References

- [1] Zhu XW, Shen Y, Zhang BH, Yan JJ. Diagnosis and treatment of urachal remnants with heterotopia sinus: a report of 2 cases. *Zhonghua Nan Ke Xue* 2005;11:142–4.
- [2] Berman SM, Tolia BM, Laor E, et al. Urachal remnants in adults. *Urology* 1998;31:17–21.
- [3] Rich RH, Hardy BE, Filler RM. Surgery for anomalies of the urachus. *J Pediatr Surg* 1983;18:370–2.
- [4] Catanzaro D, Mirk P, Carbone A, et al. Amebic abscess of urachal remnants. *Eur J Radiol* 2001;38:219–24.
- [5] Yu JS, Kim KW, Lee HJ, et al. Urachal remnant diseases: spectrum of CT and US findings. *Radiographics* 2001;21:451–61.
- [6] Gleason JM, Bowlin PR, Bagli DJ, et al. A comprehensive review of pediatric urachal anomalies and predictive analysis for adult urachal adenocarcinoma. *J Urol* 2015;193:632–6.
- [7] Risher WH, Sardi A, Bolton J. Urachal abnormalities in adults: the Ochsner experience. *South Med J* 1990;83:1036–9.
- [8] Jeong HJ, Han DY, Kwon WA. Laparoscopic management of complicated urachal remnants. *Chonnam Med J* 2013;49:43–7.
- [9] Potisek N, Weihe J. Infected urachal cyst. *N Engl J Med* 2016;375:2582.