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Case Report

Urachal actinomycosis mimicking carcinomatosis: a case report and review of literature

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Abstract: *Actinomyces Israeli* is found to be generally involved with cervicofacial (60%), followed by abdominopelvic (25%) and thoracic (15%) region. However, urachal actinomycosis is an extremely rare occurrence, represented by limited cases. This report described a 41-year-old man infected with *Actinomyces Israeli* who underwent partial cystectomy. Histological examination showed the typical colony which led to the diagnosis of *A.Israeli* infection. Finally, he was discharged home after a 4-month course of intravenous penicillin without significant complications. Review of the English literature revealed seven cases of urachal actinomycosis in the recent two decades. The median age was 37 years (range, 22-70) with a male/female ratio of 1.3:1. The treatment of *A.Israeli* was antibiotics combined with surgery. The duration of antibiotic therapy ranged from three to six months. Urachal actinomycosis is a relatively infrequent disease with nonspecific clinical and radiological features and is easily confused with urachal carcinoma.

Keywords: Urachal actinomycosis, actinomyces israeli, actinomyces, actinomycosis

Introduction

Human actinomycosis is a chronic granulomatous inflammation caused by *Actinomyces Israeli*, a gram-positive anaerobic bacterium [1-3]. *A.Israeli* usually infects men more frequently than women, predominantly individuals aged 20-50 years. *A.Israeli* is histologically characterized by Gram-positive, branching and filamentous anaerobes surrounded by fibrosis and inflammation [4]. Surgical approach remains the mainstay treatment for patient infected with *A.Israeli*. Long-term antibiotic therapy after surgical resection can achieve complete remission of more than 95%.

Precise diagnosis is obtained preoperatively in fewer than 10% of cases, and it is often made in the operation period, which confirmed later by pathologic examination. And early diagnosis is important to minimize the *A.Israeli* invasion and to prevent unnecessary therapeutic approach and inappropriate tissue removal, because actinomycosis usually has a variety of clinical manifestations, which could mimic a tumor or other inflammatory disease.

Herein, we presented an assessment of a 41-year-old man with urachal actinomycosis which was diagnosed by histopathological examination of a specimen obtained by excisional biopsy.

Case report

A 41-year-old man was admitted to our institute who mainly complained of lower abdominal mass with hypogastric pain and frequency-urgency-dysuria syndrome for half a month's duration. There was no history of hemetauria or fever. The symptom gradually improved after anti-inflammatory treatment. A palpable mass about 4×5 cm was founded at suprapubic bladder region during physical examination. Laboratory studies showed normal hematological and biochemical profiles except for RBC (+ + +), WBC (+ + +) during routine urine examination. Meanwhile, Urine culture revealed no abnormality and no tumor cells were found by urine cytology for three times. Ultrasonographic record demonstrated a hypoechoic mass on the anterior wall of the bladder, measuring about 4×6 cm. Abdominal Computed tomogra-

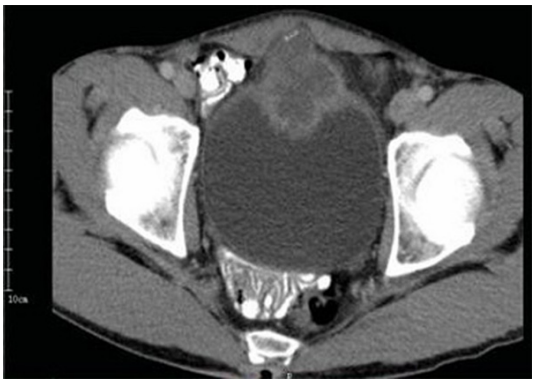

Figure 1. Abdominal CT showing a predominantly mass about 4.5×3.5 cm involving the anterior abdominal wall and dome of the bladder.

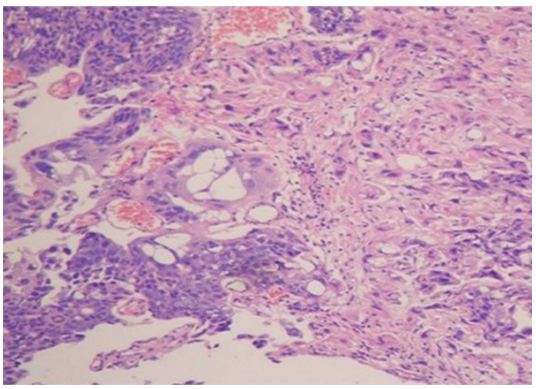

Figure 2. The characteristic filamentous *A.Israeli* colony: dark, dense and filamentous aggregates extended from a central core, along with inflammatory cell infiltration (HE ×400).

phy (CT) showing a predominantly mass about 4.5×3.5 cm involving the anterior abdominal wall and dome of the bladder (**Figure 1**).

It was suspicious of urachal carcinoma on the basis of imaging features and surrounding tissues involvement. Partial cystectomy was performed subsequently. On operation, it revealed a mass situated on the anterior bladder, and accompanied with the surface of the mass mucosa were hyperemia and edema. Histological examination showed the typical colony which led to the diagnosis of *A.Israeli* infection. As depicted in **Figure 2**, dark, dense and filamentous aggregates extended from a central core, along with inflammatory cell infiltration, which generally revealed marked abscess and fibrosis containing *A.Israeli* organisms. And the final diagnosis was pathologically consistent with *A.Israeli* with sulfur granules.

Following identification of the diagnosis of *A.Israeli*, the patient was treated with a 4-month course of intravenous penicillin without significant complications. Finally, he was discharged home 1 week later without any recurrence.

Discussion

Actinomycosis is a chronic infection caused by Actinomyces family, with *A.Israeli* being the primary etiologic agent. Other species that have been reported in human include *A.naeslundii*, *A.viscosus*, *A.odontolyticus*, *A.gerencseriae*, and *A.meyeri, A.neuii* [5, 6]. *A.Israeli*, a common flora, is found to be generally involved with cervicofacial (55%), followed by abdominopelvic (20%) and thoracic (15%) region [7-12]. However, urachal actinomycosis is an extremely rare occurrence, represented by limited cases.

According to the PubMed database, about seven cases of urachal actinomycosis were recorded in English language in the recent two decades [13-19] (**Table 1**). The median age was 37 years (range, 22-70) with a male/female ratio of 1.3:1. The treatment of *A.Israeli* was antibiotics combined with surgery in the seven cases above. The duration of antibiotic therapy ranged from three to six months. Most patients recovered after a long period of treatment.

Urachal actinomycosis is extremely to diagnose because of unsuccessful cultures with 76% negative rate, and it theoretically requires 5-7 days but may take 2-4 weeks actually. Furthermore, *A.Israeli* could also be morphologically confused with other gram-positive, non-spore forming bacilli and even non-related bacteria including clostridia, corynebacteria, listeria, and leptotrichia. Besides, the organism might also be classified as a fungus for mistake because of its filamentous appearance and indolent growth that mimicked mycotic infection. However, the absence of a nuclear membrane or chitin in the cell membrane as well as reproduction by fission is among the main characteristics which led to its reclassification as a bacterium and not a fungus.

In addition to failures in culture, nonspecific clinical features and imaging characteristics may also lead to indefinite diagnosis of *A.Israeli*. Thus, final identification must be confirmed with histological findings of sulfur granules and

Table 1. Summary of five cases of human urachal actinomycosis due to *A.Israeli*

Publication year	Patient details*	Clinical presentation	Duration of symptom# (days)	Treatment	Antibiotic duration (months)	Outcome
2011	28, male, India	Abdominal mass	20	Penicillin; resection	Unknown	Recovered
2011	57, male, China	Abdominal mass	60	Amoxicillin, clavulanate; resection	5	Recovered
2010	26, male, Korea	Abdominal mass	Unknown	Fluoroquinolone; cystectomy	3	Recovered
2001	70, female, China	Infraumbilical mass	60	Doxycycline; laparotomy	6	Recovered
1999	22, male, Italy	Umbilicalpublic mass	150	Amoxicillin; laparotomy	6	Recovered
1995	31, female, Britain	Suprapublic mass	Unknown	Penicillin, chloramphenicol; laparotomy	Unknown	Unknown
1991	25, female, Africa	Umbilicalpublic mass	Unknown	Penicillin; surgery	Unknown	Recovered

Notes: *(age, gender, country of origin); #before diagnosis.

filamentous gram-positive rods, on the basis of tissue resection, percutaneous needle biopsy or fluid aspiration. Up to now, several risk factors have been found to be associated with urachal actinomycosis, the most common being urachal remnants. Other relevant factors may be involved with renocolic or renoduodenal fistulas. According to sporadic reports, immunocompromised disorder, such as diabetes mellitus, steroid therapy and neoplasm, is additional significant predisposing factors for human actinomycosis.

Diagnosis is seldom possible because of the difficulty in differentiating the inflammation or pseudotumor from urachal malignancy, especially the aggressive nature of it, including abscess formation and pus accumulation followed by partial necrosis and reactive fibrosis. CT, ultrasound and MRI enable confirmation of the presence of an abscess or pseudotumor, but could not distinguish with urachal carcinomatosis, soft tissue sarcoma, Crohn's disease, tuberculosis, and other inflammatory disease.

Removal of infected mass combined with antibiotic treatment could eradicate the inflammatory process [12]. *A.Israeli* is highly susceptible to a wide spectrum of beta-lactam antibiotics with a long-term course. Penicillin is the first drug of choice. A common therapy includes high-dose parenteral penicillin for 2 to 6 weeks followed by oral amoxicillin for 6 months. Besides, tetracycline, erythromycin, and doxycycline are alternatives in the management of patients with penicillin allergy [20]. Moreover, erythromycin administration is recommended for pregnant patients. According to the antimicrobial susceptibility testing of *Actinomyces* species by Smith AJ [21], linezolid has been regarded as a new antibiotic against this kind of organisms apart from the most common used agents above.

In summary, urachal actinomycosis is a relatively infrequent disease with nonspecific clinical and radiological features and is easily confused with urachal carcinoma because of its infiltrative and granulomatous inflammation. The Gram stain of specimen is usually more sensitive than cell culture, especially prior antibiotics were administrated. The treatment is a combination of surgical resection and long-course antibiotherapy. Awareness of this diagnostic entity may enable the urologist to implement therapeutic approach and minimize the unnecessary demand for excessive organ resection.

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Disclosure of conflict of interest

None.

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References

[1] Zaman R, Abbas M, Burd E. Late prosthetic hip joint infection with actinomyces israeli in an intravenous drug user: case report and litera-

ture review. J Clin Microbiol 2002; 40: 4391-932.
[2] Acevedo F, Baudrand R, Letelier LM, Gaete P. Actinomycosis: a great pretender. Case reports of unusual presentations and a review of the literature. Int J Infect Dis 2008; 12: 358-62.
[3] Smego RJ, Foglia G. Actinomycosis. Clin Infect Dis 1998; 26: 1255-61, 1262-3.
[4] Sweis RF, Propes MJ, Hyjek E. Actinomyces-induced inflammatory pseudotumor of the lymph node mimicking scrofula. Ann Intern Med 2011; 155: 66-7.
[5] Fazili T, Blair D, Riddell S, Kiska D, Nagra S. Actinomyces meyeri infection: Case report and review of the literature. J Infect 2012; 65: 357-61.
[6] von Graevenitz A. Actinomyces neuii: review of an unusual infectious agent. Infection 2011; 39: 97-100.
[7] Russo TA. Agents of actinomycosi. In: Mandell GL, Bennett JE, Dolin R, editors. Principles and practice of infectious diseases. Philadelphia: Churchill Livingston; 2000. pp. 2645-54.
[8] Garner JP, Macdonald M, Kumar PK. Abdominal actinomycosis. Int J Surg 2007; 5: 441-8.
[9] Meyer P, Nwariaku O, McClelland RN, Gibbons D, Leach F, Sagalowsky AI, Simmang C, Jeyarajah DR. Rare presentation of actinomycosis as an abdominal mass: report of a case. Dis Colon Rectum 2000; 43: 872-5.
[10] Marella VK, Hakimian O, Wise GJ, Silver DA. Pelvic actinomycosis. Urologic perspective. Int Braz J Urol 2004; 30: 367-76.
[11] Yenarkarn P, Thoeni RF, Hanks D. Case 117: actinomycosis of left kidney with sinus tracts. Radiology 2007; 244: 309-13.
[12] Honda H, Bankowski MJ, Kajioka EH, Chokrungvaranon N, Kim W, Gallacher ST. Thoracic vertebral actinomycosis: Actinomyces israelii and Fusobacterium nucleatum. J Clin Microbiol 2008; 46: 2009-14.
[13] Chaitra V, Rajalakshmi T, Mohanty S, Lahoti NK, George A, Idiculla J. Actinomycosis in urachal remnants: A rare cause of pseudotumor. Indian J Urol 2011; 27: 545-6.
[14] Li MF, Yang TL, Huang JS. Imaging findings of primary urachal actinomycosis. J Chin Med Assoc 2011; 74: 44-7.
[15] Lim KT, Moon SJ, Kwon JS, Son YW, Choi HY, Choi YY, Pyo JY, Park YW, Moon HS. Urachal actinomycosis mimicking a urachal tumor. Korean J Urol 2010; 51: 438-40.
[16] Yeung Y, Cheung MC, Chan GS, Cheung AN, Tam PC. Primary actinomycosis mimicking urachal carcinoma. Urology 2001; 58: 462.
[17] Micheli E, Hurle R, Losa A, Chinaglia D, Ranieri A, Lembo A. Primary actinomycosis of the urachus. BJU Int 1999; 83: 144-5.
[18] Ozyurt C, Yurtseven O, Kocak I, Kandiloglu G, Elmas N. Actinomycosis simulating bladder tumour. Br J Urol 1995; 76: 263-4.
[19] van Wijk FJ, Lodder JV. Actinomycosis of urachal remnants. Eur Urol 1991; 19: 339-40.
[20] Sudhakar SS, Ross JJ. Short-term treatment of actinomycosis: two cases and a review. Clin Infect Dis 2004; 38: 444-7.
[21] Smith AJ, Hall V, Thakker B, Gemmell CG. Antimicrobial susceptibility testing of Actinomyces species with 12 antimicrobial agents. J Antimicrob Chemother 2005; 56: 407-9.