ScientificScholar®
Knowledge is power
Publisher of Scientific Journals

https://ijmsweb.com

Indian Journal of Medical Sciences

Case Report

Primary vesical actinomycosis with changes of cystitis cystica presenting as bladder mass

Nagendra Mishra[1], Henil Upadhyay[1], Charmy Parikh[1]

[1]Department of Urology, Pramukhswami Medical College, Anand, Gujarat, India.

ABSTRACT

Actinomycosis is a rare chronic granulomatous suppurative infection caused by Gram-positive bacteria. The occurrence of primary vesical actinomycosis is extremely rare and only a few cases have been reported. Pre-operative diagnosis of vesical actinomycosis is challenging as the clinical and radiological features usually point towards bladder malignancy. Therefore, in most cases, definitive diagnosis is usually made after histopathological examination of the involved tissue. A 60-year-old male presented with complaints of hematuria, burning micturition, irritative, and obstructive urinary symptoms for 15 days. USG revealed a large soft-tissue mass having a polypoidal intraluminal and extraluminal component and involving the right posterolateral urinary bladder wall. CT scan showed a large irregular soft-tissue mass with multiple cystic lesions involving the right lateral wall of the urinary bladder. Transurethral resection of bladder mass biopsy was performed and the histopathological examination showed bacterial colonies of Actinomyces with changes of cystitis cystica. The patient was treated with amoxicillin and potassium clavulanate for 3 months. Actinomycosis should be kept as a rare differential diagnosis in cases presenting as bladder mass. The diagnosis is most commonly made by histopathology and may need a repeat biopsy to arrive at the correct diagnosis. The patient should be treated by penicillin group of antibiotics for 2–3 months and followed up for years to detect any recurrence.

Keywords: Actinomycosis, Bladder mass, Cystitis cystica, Infectious disease

INTRODUCTION

Actinomycosis is a rare bacterial infection caused by *Actinomyces* which are a group of Gram-positive anaerobic bacteria. These bacteria measure up to 1 μm diameter and form filamentous microcolonies without any spores. These are opportunistic pathogens endogenous to the gastrointestinal, respiratory, and genitourinary tracts that cause infection only when the normal mucosal barriers are damaged thus making the deeper tissues accessible to the bacteria. The infection has a chronic course with a suppurative and granulomatous nature. The most common clinical form of actinomycosis is cervicofacial followed by pelvic actinomycosis with a greater female predominance. The urinary bladder is usually involved secondary to gynecological involvement in females. Less than 100 cases of pelvic actinomycosis have been reported worldwide.[1-4]

Actinomycosis of the urinary bladder is a rare disease.[5] The most common etiology of vesical actinomycosis is a direct extension from a pre-existing focus of infection in the pelvic organs. The occurrence of primary vesical actinomycosis, that is, without a primary focus of infection elsewhere is extremely rare.[6,7]

Pre-operative diagnosis of vesical actinomycosis is challenging as the clinical and radiological features usually point toward bladder malignancy. Therefore, in most cases, a definitive diagnosis is usually made after histopathological examination of involved tissue.[7]

We present a case of vesical actinomycosis with changes of cystitis cystica presenting with features similar to bladder malignancy.

CASE REPORT

A 60-year-old male presented to the urology department with complaints of painless total hematuria with clots and burning micturition for 15 days. Burning was associated with increased frequency of urination, nocturia, hesitancy, and feeling of incomplete emptying of the bladder, intermittent, and weak urinary stream. There was associated low-grade fever for the past 5 days. There were no complaints of weight loss, loss of appetite, lower abdominal pain, or backache. There was no significant medical, surgical, or drug history.

His vitals on admission were as follows: Afebrile, pulse 76/min, blood pressure 130/80 mmHg, respiratory rate 16/min, and SpO_2 98% on room air. General head-to-toe examination

***Corresponding author:** Nagendra Mishra, Department of Urology, Pramukhswami Medical College, Anand, Gujarat, India. nagendraad1@yahoo.com

Received: 29 January 2021 **Accepted:** 15 April 2021 **Published:** 11 May 2022 **DOI** 10.25259/IJMS_47_2021

and systemic examination did not reveal any abnormality. Routine laboratory investigations were normal.

USG of the abdomen and pelvis revealed a large soft-tissue mass involving the right posterolateral urinary bladder wall with size 6.2 × 4.9 cm and having a polypoidal intraluminal and extraluminal component. There was increased vascularity in the Doppler study. Both kidneys were WNL on USG.

CT scan showed a large irregular shaped contrast-enhancing soft-tissue mass sized 6.2 × 5.4 × 5.1 cm involving the right lateral wall of the urinary bladder. The mass had multiple variable sized thick-walled cystic lesions with irregular frong-like margins and pockets of fluid collection attached to the right lateral wall and base of the urinary bladder. A marked perivesical fat stranding was noted with preserved fat planes with rectus muscle anteriorly, iliac vessels, and obturator muscle laterally and prostate inferiorly [Figure 1].

Based on clinical features and radiological investigations, we suspected urinary bladder malignancy and decided to perform transurethral resection of bladder tumor biopsy with a definitive diagnosis and further management depending on histopathological reports of the resected specimen. Cystoscopy showed a large mass projecting into the bladder with an edematous hyperemic wide base.

Multiple superficial biopsies of the mass were taken and sent for histopathological examination. Gross examination of superficial bladder chips showed multiple grayish-white soft-tissue bits aggregately measuring 5 × 4 × 1.5 cm and microscopic examination showed focally hyperplastic and ulcerated urothelial lining, squamous metaplasia, keratin pearls, and Brunn nests. The lamina propria was edematous with few congested blood vessels and reactive lymphoid follicles. Changes of cystitis cystica were seen with the urothelium invaginating into the lamina propria. There was no evidence of malignancy on H.P. examination.

However, keeping in mind the strong suspicion for urothelial malignancy, we took repeat biopsies from deeper levels which revealed a muddy thick pus-filled cavity within the mass. The mass on resection had a yellow hue. Pus was aspirated and sent for microbiological examination.

Gross examination of deep bladder chips showed multiple grayish-white soft-tissue bits aggregately measuring 2 × 1.8 × 0.7 cm and microscopic examination showed muscle fibers with patchy lymphoid follicles. Gram staining of pus showed plenty of bacterial colonies consistent with actinomycosis with areas of calcification and neutrophilic exudates. There is no evidence of malignancy in both sections [Figure 2].

Based on H.P. findings, the patient was diagnosed with primary vesical actinomycosis with changes of cystitis cystica.

The patient was treated with intravenous cefoperazone + sulbactam for 2 weeks and intravenous ampicillin for 1 week. On discharge, the patient was prescribed oral amoxicillin + clavulanic acid for 3 months.

On a follow-up after 3 months, the patient was asymptomatic and a CT scan showed a reduction in the size of the bladder mass. This improvement is persistent even after 18 months on a recent follow-up.

DISCUSSION

Genitourinary actinomycosis may be primary (without a known focus of infection) or secondary to trauma or surgery. The incidence of primary vesical actinomycosis is very rare. Secondary actinomycosis is more common as the bacteria require an altered mucosal function for infection. Actinomycosis may present with varied clinical features but with a common underlying pathology of purulent focus surrounded by compact fibrotic reaction.

Common clinical features of vesical actinomycosis include suprapubic pain, abdominal mass, burning micturition, increased frequency, hematuria, fever, and weight loss.[9] These can be accompanied by constitutional symptoms and mass effect, such as obstruction or visceral pressure sensation.[10] In our case, the patient presented with hematuria with clots and irritative urinary symptoms.

Routine laboratory examinations were not very helpful in the case of actinomycosis as they only show an elevation in inflammatory markers which are non-specific and hence do not aid in diagnosis.[1]

CT scan usually reveals a solid mass infiltrating locally (pseudotumor), with cystic lesions and dense contrast enhancement on the walls or solid components of the masses. Due to its infiltrative nature and proliferative pseudotumor pattern, actinomycosis is often misdiagnosed as a malignant disease. Hence, histopathological examination is the gold standard method for diagnosing actinomycosis. It shows central suppurative necrosis with sulfur granules surrounded by granulomatous tissue and intense fibrosis. *Actinomyces* spread locally through the tissue planes instead of the hematological or lymphatic route and give a pseudotumor appearance due to the proteolytic enzymes produced by *Actinomyces*.[11]

In our case, the histopathological report also showed changes of cystitis cystica. According to our literature search, this is the first reported case of primary vesical actinomycosis with changes of cystitis cystica.

Cystitis cystica is a chronic inflammatory hyperproliferative condition, wherein the submucosal masses of epithelial cells known as Brunn nests undergo cavitation to form fluid-filled cystic structures. The underlying cause is postulated to be the local immune response to any chronic irritative stimulus to the urothelium such as infection, calculi, outlet obstruction, or tumor. Cystitis cystica commonly presents with irritative

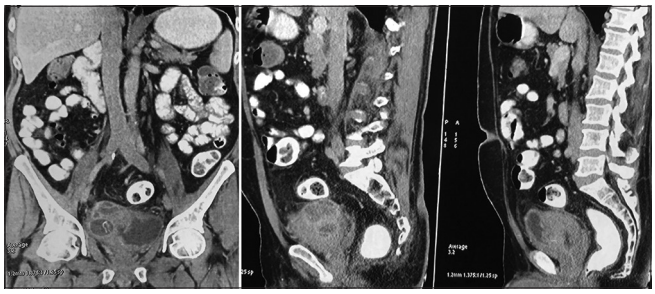

Figure 1: Coronal and sagittal plain CT scan images showing multiple cystic lesions with round to oval pockets of fluid collection attached to the right lateral wall of the urinary bladder.

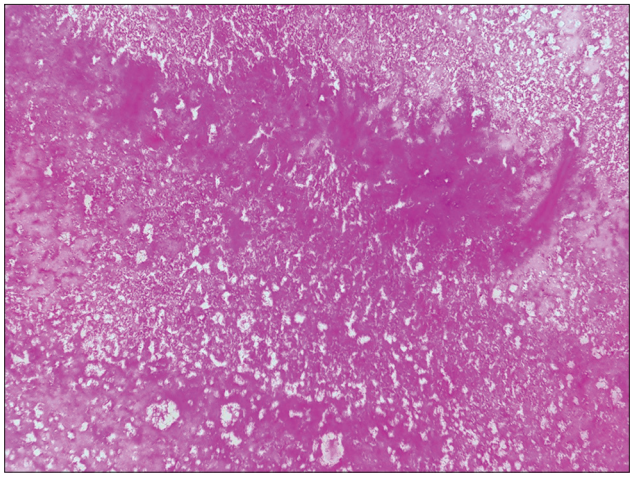

Figure 2: H&E staining of the pus sample showing colonies of *Actinomyces* arranged in a radiating pattern giving a sunray appearance.

lower urinary tract symptoms and hematuria. It may occur at any age with a slight male predominance. CT imaging classically shows the appearance of multiple small filling defects of 2–5 mm in the bladder wall which is consistent with findings in our case. Histopathological examination after cystoscopy is mandatory in a case of cystitis cystica as it can be easily misdiagnosed as a malignancy on cystoscopy. The treatment approach consists of removing the source of irritation and further management depending on the etiology.[12,13] We do not know if there is any significance of such an association or it is simply a coincidence.

Antibiotic therapy is the gold standard for the treatment of actinomycosis. Actinomyces are susceptible to several antimicrobials though high-dose penicillin administered over a prolonged duration of several weeks to months is the cornerstone treatment for actinomycosis. Other beta-lactam antibiotics including amoxicillin, amoxicillin-clavulanic acid, and ceftriaxone have been used with good outcomes. In patients allergic to penicillin, doxycycline and clindamycin can be used as alternatives. In patients with adequate surgical resection outcomes, shorter courses of 6–8 weeks could be adequate. The dosing and duration of antibiotics should be tailored according to individual needs and depend on the severity of infection, size of the lesion, and the compliance of the patient. Surgical intervention is usually limited to excision of abscesses and fistulas, decompression of closed space infections, and establishing the correct diagnosis of infection. Although surgery can be effective, it is not curative without antimicrobial treatment.[6-8,14] In this case, we treated the patient for 4 months only.

Actinomycosis usually shows a good prognosis with low mortality and morbidity if a timely diagnosis is made and appropriate treatment is given.[15] However, long-term follow-up after treatment is essential since relapse is common.[3]

CONCLUSION

Pre-operative diagnosis of actinomycosis is a clinical challenge as there are no clinical, laboratory, or radiological findings specific to the infection. Hence, we have to rely on histopathological examination or culture reports for a confirmed diagnosis.[1] In our case, clinical examination and radiological findings suggested urinary bladder malignancy. We took multiple biopsies from the lesion and the H.P. report showed features of chronic granulomatous inflammation with features of cystitis cystica without any evidence of malignancy. There was no evidence of actinomycosis on superficial biopsies as the lesions were deep seated and did not involve the bladder mucosa.[11] On the second biopsy at a deeper level, we found a pus-filled cavity which on Gram staining showed colonies of *Actinomyces*, thus confirming the diagnosis of actinomycosis.

There are very few reported cases of vesical actinomycosis and with one published literature on primary vesical actinomycosis in India.[16] Hence, we need to consider actinomycosis as a differential diagnosis in cases presenting as bladder malignancy because both diseases have different treatment modalities and prognosis. Early diagnosis of actinomycosis will help in improving prognosis and prevent the patient from being misdiagnosed as a case of urinary bladder malignancy.

Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Acquaro P, Tagliabue F, Confalonieri G, Faccioli P, Costa M. Abdominal wall actinomycosis simulating a malignant neoplasm: Case report and review of the literature. World J Gastrointest Surg 2010;2:247-50.
2. García-García A, Ramírez-Durán N, Sandoval-Trujillo H, Romero-Figueroa MD. Pelvic actinomycosis. Can J Infect Dis Med Microbiol 2017;2017:9428650.
3. Ziypak T, Adanur S, Ozkaya F, Calık M, Polat O, Aksoy Y, *et al*. Primary vesical actinomycosis in a 23-year-old man. Case Rep Urol 2014;2014:691360.
4. Yiğiter M, Kiyici H, Arda İS, Hiçsönmez A. Actinomycosis: A differential diagnosis for appendicitis. A case report and review of the literature. J Pediatr Surg 2007;42:e23-6.
5. Maddah G, Kerigh BF, Mohamadian N, Bagheri V. Primary urachal actinomycosis: Case report and literature review. Nephrourol Mon 2013;5:997-1000.
6. Al-Kadhi S, Venkiteswaran KP, Al-Ansari A, Shamsudini A, Al-Bozom I, Kiliyanni AS. Primary vesical actinomycosis: A case report and literature review. Int J Urol 2007;14:969-71.
7. Lee KR, Ko YS, Yu JW, Yoon CY, Kim CH, Yoon DK. Primary vesical actinomycosis: A case diagnosed by multiple transabdominal needle biopsies. J Korean Med Sci 2002;17:121-4.
8. Bianchini MA, Bigi E, Repetto P, Ceccarelli P, Durante V, Biondini D, *et al*. A case of frozen pelvis: Primary actinomycosis of urinary bladder in a young boy. J Pediatr Surg 2012;47:e9-11.
9. 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.
10. Huang C, Al-Essawi T. Actinomycosis of the urinary bladder. Can Urol Assoc J 2013;7:E502-4.
11. Fujiwara H, Yamada Y, Banno H, Hikami K, Nakashima M, Tamaki M, *et al*. Primary urachal actinomycosis diagnosed by transurethral resection. J Clin Urol 2020;13:195-7.
12. Bastianpillai C, Warner R, Beltran L, Green J. Cystitis cystica and glandularis producing large bladder masses in a 16-year-old boy. JRSM Open 2018;9:2054270417746060.
13. Potts S, Calleary J. Cystitis cystica as a large solitary bladder cyst. J Endourol Case Rep 2017;3:34-8.
14. Maxová K, Menzlová E, Kolařík D, Dundr P, Halaška M. Case report: Pelvic actinomycosis. Prague Med Rep 2012;113:44-8.
15. Elzein F, Kharraz R, Arab N, Alotaibi F, Almohaya A, Almutairy A. A case series of actinomycosis from a single tertiary care center in Saudi Arabia. IDCases 2019;15:e00521.
16. Gupta CL, Gupta R. Actinomycosis of urinary bladder-a rare entity. Indian J Urol 2003;19:159-60.

How to cite this article: Mishra N, Upadhyay H, Parikh C. Primary vesical actinomycosis with changes of cystitis cystica presenting as bladder mass. Indian J Med Sci 2022;74:44-7.