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Case Report

Isolated epididymal tuberculosis: case report and review of literature

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ABSTRACT

Ethiopia is one of the countries with high burden of pulmonary tuberculosis and the number of cases of extra pulmonary is increasing. However, diagnostic evidence to initiate anti-tuberculosis treatment among extra pulmonary tuberculosis (EPTB) cases is not well known. We presented here a case of isolated tuberculosis epididymitis with scrotal sinus after developing nonspecific testicular symptoms for more than 2 months and review the literature. Tuberculous epididymitis is a form of urogenital tuberculosis that is particularly rare and has the same clinical manifestation that mimics other more common infectious causes of epididymitis, leading to delay in diagnosis. So, one should maintain a high-index of suspicion to accurately diagnose tuberculous epididymitis in individuals presenting with suggestive signs and symptoms.

Keywords: Epididymitis, Orchitis, Fistula, Tuberculosis, Genitourinary tuberculosis, Scrotum

INTRODUCTION

Mycobacterium tuberculosis is one of the leading causes of morbidity and mortality worldwide. Despite advances in diagnostics, treatment, and prevention, tuberculosis (TB) remains a global problem, infecting one-third of the world's population. Approximately 90% of those who fall sick with TB each year live in 30 countries.[1]

Ethiopia is among the countries on the high burden TB list 2022 (in 2019, the estimated incidence rate for TB was 140 per 100,000 population) and reports one of the highest incidence rates of EPTB. Despite advances in anti-mycobacterial therapy and strict implementation of well-known TB control measures, the prevalence and incidence of TB remains high worldwide. Most of new cases of TB are attributable to five risk factors: undernutrition, HIV infection, alcohol use, smoking and diabetes.[1]

M. tuberculosis is an etiologic agent for tuberculosis. TB typically affects the lungs but can also affect other parts of the body.[2]

Genitourinary tuberculosis (GUTB), which accounts for 20-40% of EPTB cases, is the second most common site in developing and third most common site in developed nations.[3] It occurs in 2 to 20% of individual with pulmonary tuberculosis.[2] In case of genital tuberculosis, it can affect the entire male genital tract (including the prostate, seminal vesicles, vas-deference, epididymis, testicles, cooper glands and penis).[2]

Genital TB occurs via hematogenous spread to the prostate and epididymis or via the urinary tract to the prostate with a spread from ejaculatory duct to the seminal vesicles, vas deferens and epididymis.[2-5]

Isolated tuberculous epidymitis (ITE), which is defined as TB epididymitis without clinical and laboratory evidence of renal involvement, is a rare EPTB that occurs in young adults. Progression of disease is slow and have clinical manifestation that mimic other more common infectious causes of epididymitis, leading to delay in diagnosis.

The pathological features of tuberculosis of the epididymis are characterized by extensive tissue

destruction and fibrosis, which eventually lead to the destruction of the epididymis and the surrounding genital tissues leading to complications such as infertility and other male reproductive system malfunction. Thus, one should maintain a high-index of suspicion to diagnose TB epididymitis accurately in individuals presenting with suggestive signs and symptoms.

In this report, we briefly reviewed and presented a case of ITE that initially presented with scrotal pain that later progressed to scrotal sinus formation, diagnosed by ultrasound and histopathological examination and subsequently treated with anti-tuberculosis drugs according to WHO guideline.

CASE REPORT

A 35-year old businessmen presented with a history of non-specific discomfort and ulcer over the right scrotum of 2-month duration. He did not have associated fever, weight loss or night sweet. A month after noticing the swelling, he visited nearby health center and was given ceftriaxone and doxycycline for 7 days. On the fifth day of treatment, the swelling ruptured and started draining a thin, odorless, whitish discharge. After completing the 7-day course of antibiotics, he was started on ciprofloxacin and cephalexin at another health center, and after failing to respond he was referred to our center for better evaluation and management.

He had no recent history of dysuria, frequent urination, hematuria, back or flank pain. He had never been treated for tuberculosis, had no chronic cough or significant weight loss and claimed to have received a bacillus calmette guerin (BCG) vaccine in infancy. There was no prior history of contact with a chronic cougher or a known TB patient. He was married but had recently been having unprotected sex with a new partner.

Figure 1: Thickened right lower scrotum pole with a purulent discharging sinus.

On physical examination, he appeared healthy looking with normal vital signs and there were no pertinent positive findings except in the urogenital system.

Examination of the external genitalia revealed a normal urethral meatus and a slightly enlarged right testicle over the lower pole with purulent discharging sinus. Inguinal and femoral lymph nodes were not enlarged (Figure 1). Digital rectal examination revealed a non-tender, firm and rubbery prostate.

He was seronegative for syphilis and HIV antibodies and his kidney function and liver enzymes were within normal range. Total WBC count was 11.2×10^3, with total neutrophil count of 6.5×10^3 and ESR was 10 mm/hr. Urinalysis (simple microscopy and deep stick) was non-revealing and urine was negative for the GeneXpert MTB/RIF (*Mycobacterium* TB/ resistance to rifampin) assay. Chest X-ray was clear (Figure 2).

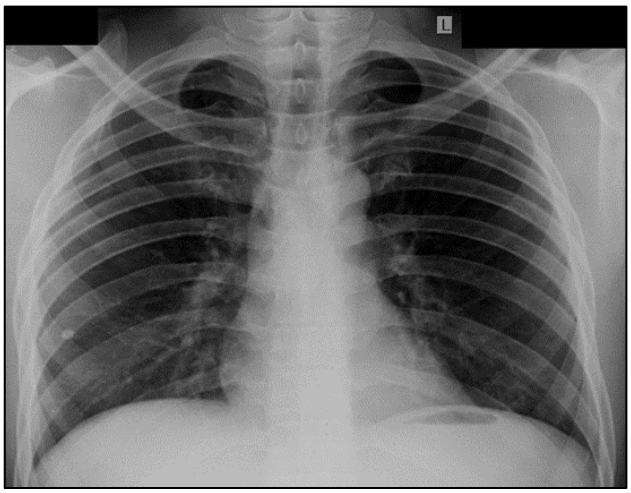

Figure 2: PA X-ray of the chest showing a clear chest.

Scrotal ultrasonography showed a gross 2.5×2.3 cm heterogeneous enlargement over tail of the right epididymis, with lesions continuation into the scrotal skin, with normal echotexture of right epididymal body and head, and normal right and left testis (Figure 3).

Fine needle aspiration cytology (FNAC) was done and revealed sheets of mixed inflammatory cells with small mature lymphocytes, neutrophils, epithelioid granuloma and multinucleated langerhans type giant cells, suggestive of TB.

The presence of a sinus tract, with characteristic ultrasonography and histopathology findings helped us in reaching to our diagnoses.

The patient started on a four-drug combination (isoniazid, rifampicin, ethambutol, pyrazinamide) of anti-TB for 2 months, to be followed by two-drug (isoniazid, rifampicin) therapy for 4 months.

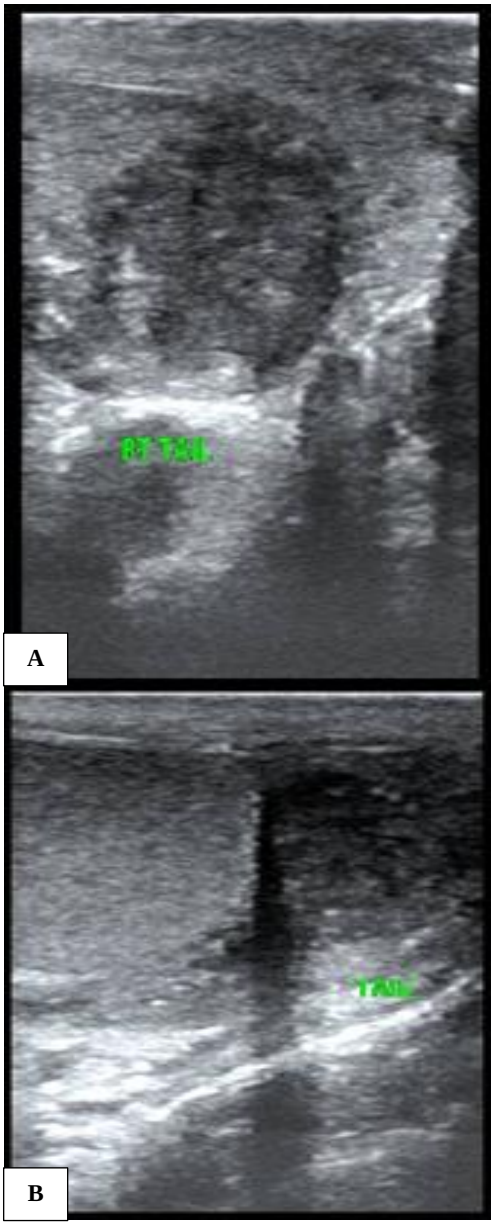

Figure 3 (A and B): Testicular ultrasound shows enlarged heterogeneous right epididymis.

DISCUSSION

TB is a global epidemic with an estimated 10 million cases and 1.5 million deaths in 2020 (including 214,000 people living with HIV worldwide). The epidemiology of TB varies substantially around the world. The highest rate (100 per 100,000 or higher) is observed in Sub-Saharan Africa, India, and islands of Southeast Asia and Micronesia.[6]

Poverty, HIV and drug resistance are major contributors to the resurgence of the global epidemic.[6,7] Roughly 1 in 9 new TB cases occur in individuals who are infected with HIV: 74% of these cases occur in Africa. An estimated 600,000 cases are of multidrug resistant (MDR-TB) or rifampin- resistant.[6]

PTB, caused by aerosolized *M. tuberculosis bacilli* invading the pulmonary alveoli, accounted for 85% of reported TB cases worldwide.[7] This pulmonary infection led to hematogenous and lymphatic dissemination and Seeding of the bacilli in other organ systems resulting in EPTB. Currently, EPTB is a growing problem. This re-emergence had been attributed to the AIDS pandemic and the increasing prevalence of comorbid conditions.[3]

The commonest EPTB reported sites were lymph node, pleural and GUTB. GUTB, which accounts for 20-40% of EPTB cases, is the second most common site in developing and the third most common site in developed nations.[3]

Jessica et al reported a case series of genitourinary TB at a single center that identified kidney disease as the most common site of infection (64.9%), followed by urethral (27.4%), bladder (17%), epididymis (5.2%) and prostate (3.4%) involvement.[8]

Although urogenital organ involvement is usually secondary to TB in other parts of the body, isolated genital organ involvement has been described in 5-30% of cases, with higher rates in epidemic situations. However, some authors had questioned the existence of a true ITE because initial imaging studies or microscopic urine examinations may not reveal kidney damage.[9]

Following bacteria implantation in more vascular regions of the genitourinary system during the primary bacillaemia, these lesions cicatrize in about 6 months, and a latent phase ensues. Reactivation of the latent foci results from a decrease in the host immunity, usually after a long period of latency.

The reactivation is usually of a single focus which accounts for the greater frequency of a unilateral clinical disease, but can be multifocal, either concurrent or sequential. Chung et al reported in their case series of ITE patients that 14 patients (78%) had unilateral and four (22%) patients had bilateral involvement.[4]

Siddharth et al reported that 45% of TB epididymo-orchitis patients have involvement of epididymis alone, with testicular involvement developing later as the disease progresses.[3] The cauda of the epididymis being affected more than the caput, possibly owing to the increased vascularity of the cauda, as was the case in our patient. The lesion of epididymis tuberculosis gradually invades the body and the head, eventually affecting the entire epididymis.

A study of 29 patients with scrotal TB showed that, 25% had evidence of active pulmonary TB. In these patients, the mean interval from emergence of symptoms to making a clinical diagnosis was 142 days, reflecting the long delay in making a diagnosis of TB.[10] In another case series, Jae et al reported that **s**eventy percent of patients

with tuberculous epididymitis have a previous history of tuberculosis.[4]

Patients with tuberculous epidymitis may present with different signs and symptoms. In a Chinese ITE case series with 47 patients: it is reported that painless swelling of the scrotum occurs in 21 patients (44.7%); scrotal pain in 21 patients (44.7%); urinary tract irritation such as frequency, dysuria and hematuria in four patients (8.5%); and ulceration of the scrotal skin happen in one patient (2.1%). Six patients (12.8%) showed systemic symptoms such as low-grade fever, fatigue and night sweats. Of the 47 cases, eight (17.0%) had a history of tuberculosis, including three cases of pulmonary tuberculosis, four cases of renal tuberculosis, and one case of prostate tuberculosis.[11] And in another case series, it is reported that eighty percent of ITE patients present with scrotal mass.[3]

Patients with scrotal TB are sometimes diagnosed incidentally during investigation for male infertility. These patients have oligozoospermia or azoospermia due to granulomatous destruction and obstruction in the epididymis or vas deference.[10]

It can be difficult to distinguish ITE clinical features from those secondary to malignancy, but scrotal fistula with discharging sinus is strongly suggestive of TB.[10,12,13] Such sinus fistular tracts are known to occur as a result of caseous abscess reaching the scrotal skin; thus, the chronic draining sinus should be regarded as having a tuberculous origin until proven otherwise.

The differential diagnosis of tuberculosis of the epididymis includes bacterial epididymitis, epididymal sperm granuloma, epididymal tumor and primary testicular tumor or metastasis.

The choice of the optimal microbiological or molecular diagnostic method for TB depends on the clinical context, available laboratory capacity and resources. The gold standard for diagnosing TB is the isolation and culture of *M. tuberculosis* from the lesion. The presence of characteristic clinical findings combined with; positive urinary acid-fast bacilli (AFB) and/or positive urine culture for *M. tuberculosis* and/or positive urinary PCR for *M. tuberculosis* and/or typical granulomatous inflammation with caseous necrosis on histopathology strongly support the diagnosis of TB.

Fine-needle aspiration cytology or an epidydimal biopsy is used to diagnose TB epididymitis, as Mtb is not usually present in the urine. However, if clinical sings are suggestive of malignancy, FNAC is contraindicated considering the risk of possible lymphatics spread of malignant cells during aspiration cytology.[14]

Granulomatous inflammation in Mtb-infected tissues seen on histological examination is a hallmark of TB. The granuloma is a focal compact collection of epithelioid cells, macrophages, lymphocytes, plasma cells, Langham's giant cells, fibroblasts with collagen and a characteristic central caseous necrosis.[10,15]

Doppler ultrasound is the first choice for imaging analysis of epididymal tuberculosis. CT or MRI have little value in diagnosing epididymal TB; however, they are important to diagnose tuberculosis in lung and kidney and provide support for the diagnosis of epididymal tuberculosis.[11,16]

Ultrasonography of Tb epididymitis is characterized by enlarged, heterogeneous and hypoechoic lesion in the body and tail, associated with hypoechoic testicular lesions or discharging sinuse.[10]

The combination of clinical, radiological, histopathological and microbiological finding all play an important role in the diagnosis of TBEO. Recent imaging technology has not improved the overall accuracy of the diagnosis of TBEO.[11,13]

The primary aims of management of UG-TB are to eradicate MTB infection with TB drug therapy, treat complications and manage comorbidities and risk factors. Close follow-up monitoring is required during TB treatment, for tracking adherence and response to therapy, and monitor individual TB drug toxic effects, development of TB drug resistance, TB drug levels in renal failure and drug interactions with antiretroviral therapy in HIV-coinfected patients.

The latest WHO recommended treatment guidelines for drug-sensitive TB, drug-resistant TB, EPTB and LTB (Latent TB) should be followed.

Drug-susceptible genitourinary TB is typically treated with a 4-drug antituberculosis regimen (isoniazid with vitamin B6, rifampin, ethambutol, and pyrazinamide) for 2 months, followed by isoniazid, rifampin, and vitamin B6 for 4 months. The regimen and duration of treatment for TB epididymo-orchitis was the same as for pulmonary tuberculosis.

Medical therapy alone may be sufficient, although surgical management is required in cases of residual abscesses requiring drainage or if there are no signs of resolution within 2 months of medical treatment.[11,14,17]

CONCLUSION

This case highlights the clinical challenges in diagnosing genitourinary TB. Epididymitis that fails to respond to empiric antibiotic therapy-even in the absence of clinical and laboratory markers of pulmonary, renal and urologic tuberculosis-should prompt further tuberculosis workup.

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