Case Report

Balanitis circinata as the sole clinical presentation of underlying sexually acquired reactive arthritis: A case report

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ABSTRACT

A 35-year-old male was diagnosed with a case of HLA27-positive reactive arthritis post-evaluation. He presented with the only symptoms of recurrent genital ulcers on glans penis of 4 months duration. The patient was treated with nonsteroidal anti-inflammatory drugs, a short course of systemic steroids, and a local immunomodulator cream on the penile lesion. The patient responded well to the treatment and returned to his usual life activities with no occurrence so far.

Key words: Circinate balanitis, Pimecrolimus, Reiter's disease

Reactive arthritis (ReA) also known as Fiessinger-Leroy disease is a genetically determined syndrome of post-genitourinary or gastroenterological infections. It presents as the classical triad of peripheral or axial arthritis, conjunctivitis, and genitourinary symptoms such as urethritis and circinate balanitis [1,2]. As a pentad, the condition includes circinate balanitis and keratoderma blenorrhagicum as characteristic lesions [3]. It was first described by Hans Conrad Julius Reiter, a German physician during the First World War in 1916. Fiessinger and Leroy published similar findings as "oculo-urethro-synovial syndrome" in the same year. Commonly seen in young Caucasian males with HLA-B27 positivity associated in 80% of cases; however, other age groups including pediatric patients cannot be excluded [4]. Its pathogenesis has been studied extensively and exogenous pathogen-associated molecular patterns, derived from microbes such as shigella, salmonella, yersinia, campylobacter, and Chlamydia have been proposed behind disseminating infection upward through the pelvic and spinal lymphatic pathways. These molecules activate toll-like receptors (TLRs) which trigger signaling pathways resulting in the expression of immune response genes and cytokine production. Initially, TLR-4 expression by neutrophil was thought to be responsible for host clearance. However, recent studies from human data suggest that TLR-2 is important in determining the susceptibility of ReA after infection by microbes [5].

We report a case with circinate balanitis as the only initial clinical finding which was later diagnosed as a case of sexually acquired ReA. Circinate balanitis is rarely seen as a preceding finding although it is the most common finding of SARA. Hence, this case is being reported for its rare presentation.

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CASE REPORT

A 27-year-old unmarried male presented with chief complaints of asymptomatic multiple superficial lesions over the glans penis for 4 months. The lesions appeared as single raw areas that progressed in size and number over a period of these 4 months. History revealed mild lower back pain on and off for 2 months which was not associated with early morning stiffness. It was not significant according to the patient as it did not interfere with the day-to-day activities. History of diarrhea, conjunctivitis, and lesions in the mouth, palms, soles, or elsewhere was negative. Sexual history was not significant and the patient denied any sexual contact with a known, unknown, or amateur sexual partner. He was treated elsewhere with oral and topical antibiotics and oral antiviral drugs on two occasions suspecting it to be herpes genitalis infection but there was no relief and the lesions never healed on any of the treatments given.

Examination revealed multiple well-defined round- to oval-shaped superficial erosions of size 0.5–2 cm, with irregular margins, coalescing at places to form a typical circinate pattern over the glans penis (Fig. 1). From the above findings, differential diagnoses of herpes genitalis, ReA, candidal balanitis, and genital psoriasis were considered.

Laboratory investigations showed normal results. On his complete blood count, hemoglobin was 14.8 g/dL, total leukocyte

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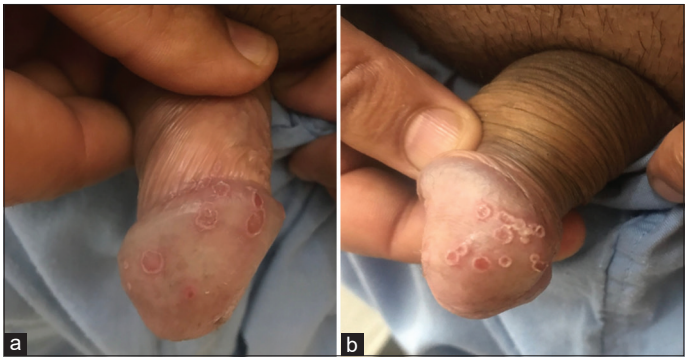

Figure 1: (a and b) Multiple well-defined erythematous circinate lesions on the glans penis

count was 8700 cell/mm^3, stool and urine routine and microscopic examinations, serum glutamic oxaloacetic transaminase was 29.6 mg/dL, serum glutamic pyruvic transaminase was 24.5 mg/dL, urea was 34 mg/dL, uric acid was 8.06 g/dL, creatinine was 1 mg/dL, and all biochemistry parameters were within normal limits. General examination revealed no pallor, cyanosis, icterus, clubbing, lymphadenopathy, or pedal edema. He was an averagely built individual with a body mass index of 23.4 and stable vitals.

Tzanck smear, Gram-stain, and potassium hydroxide stain did not show any specific findings. Viral markers such as Hepatitis B surface antigen, anti-hepatitis C antibodies, Human immunodeficiency virus antigen, and venereal disease research laboratory tests were non-reactive. Stool culture and urine culture did not show microbial growth. Fungal culture from the lesion was also negative for any growth. C-reactive protein was raised (11.2 mg/dL, normal <0.6 mg/dL) and HLA 27 was positive for the patient. Magnetic resonance imaging (MRI) findings of lumbosacral spine and sacroiliac joints showed loss of lumbar lordosis and bilateral sacroiliitis.

Histopathology of lesions from the glans resembled psoriasis and ReA with neutrophils forming intracorneal and epidermal pustules, parakeratosis, and acanthosis. Mild lymphocytic inflammation mixed with neutrophils was seen in the submucosa. Based on the above clinical and histopathological findings, a final diagnosis of ReA was made.

The patient was treated for circinate balanitis with pimecrolimus 1% cream twice daily for 3 months. Lesions cleared completely and no recurrence was seen subsequently. The patient was referred to a rheumatologist for the management of arthritis.

DISCUSSION

Circinate balanitis is an often painless lesion seen in up to 40% of men with ReA [4]. In many cases, this may go unnoticed as the lesions are very superficial and painless. The patient might give a history only when asked about it. In our case, circinate balanitis was the only clinical presentation of the patient. The lesions were superficial and painless but were noticed incidentally by the patient as they were progressing in number and size and were persistent for 4 months not responding to any treatment offered by the doctor. He was referred to the dermatology department where suspicion of circinate balanitis was made and he was investigated further. The causative agent of the infection cannot be identified in almost half of the patients [4,6]. The clinical triad of conjunctivitis, urethritis, and arthritis is commonly referred to as Reiter's syndrome, recently and appropriately called sexually acquired ReA (SARA). It rarely presents as such a neat and conclusive diagnosis. Less than one-third of the patients present with a classical triad and in fact, in the majority of cases, the presentation of SARA is incomplete [7,8].

Our patient was treated using topical 1% pimecrolimus cream for 3 months. Various treatment modalities tried for mucosal lesions include less potent topical steroids such as hydrocortisone or triamcinolone. A combination of keratolytic agents like 10% salicylic acid ointment with hydrocortisone 2.5% cream, and oral aspirin has also been reported to clear circinate balanitis in some studies. Topical 0.1% tacrolimus or pimecrolimus 1% cream has been used in refractory cases [9].

This case highlights a rare situation where SARA presented with circinate balanitis as the predominant feature. Early recognition of circinate balanitis as a presenting feature of SARA may be important, particularly as in some patients it may be the only sign. Routine HLAB27 genetic testing of patients presenting with circinate balanitis hereby becomes important as it could help early treatment and predict good future prognosis. One recommendation that can be drawn from our particular case report however is that all dermatologists and medical specialists, rheumatologists, or general physicians should be aware of circinate balanitis, its macroscopic and clinical appearance so as to have a strong clinical suspicion of a more severe underlying illness. This would help in prompt diagnosis and management and overall reduce morbidity associated with the disease in the late stages.

CONCLUSION

The patient described above presented the sole manifestation of ReA as circinate balanitis and later was diagnosed positive for HLA B27 and MRI findings suggested bilateral sacroiliatis. Early recognition and suspicion of circinate balanitis in the absence of any other significant clinical finding enabled prompt multidisciplinary treatment by dermatologists and rheumatologists.

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