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Case report

Ocular syphilis masquerading as bilateral peripheral ulcerative keratitis

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ABSTRACT

Ocular syphilis has varied manifestations in the eye. Peripheral ulcerative keratitis (PUK) is a crescent-shaped ulcer involving the peripheral cornea and associated with thinning. PUK is caused by both autoimmune and infectious diseases, such as rheumatoid arthritis, tuberculosis, and herpes. Here, we report a rare case of bilateral PUK caused by syphilis. A 55-year-old man presented with recurrent pain and redness in both eyes for 2 months. The cornea of both eyes had bilateral peripheral crescent-shaped ulcers suggestive of PUK. The patient was started on topical steroids elsewhere, but the lesion was not showing any signs of healing. A series of investigations were performed, with positive venereal disease research laboratory and fluorescent treponemal antibody absorption tests. The patient was then started on systemic penicillin, as well as topical steroids. The response to the treatment was good and the ulcer began to heal. PUK as the presenting feature of acquired syphilis is a rare scenario. Such infective causes should be managed with systemic antimicrobials for optimal outcomes.

1. Introduction

Peripheral ulcerative keratitis (PUK) is a form of peripheral crescent-shaped corneal ulcer involving the area adjacent to the limbus and characterized by sectoral thinning of the cornea. It is usually unilateral and is caused by both autoimmune and infectious diseases. PUK due to infectious diseases is rare, with syphilis as a cause of PUK rarely reported in the literature. Here, we describe a rare case of bilateral symmetric PUK as the presenting feature in a patient with syphilis.

2. Case Report

A 55-year-old man from India presented at the outpatient department with recurrent pain and redness in both eyes for 2 months. On examination, his vision in the right eye was 20/80 and in the left eye was 20/40. Circumcorneal congestion was present in both eyes. The cornea of both eyes had bilateral crescent-shaped ulcers in the periphery, with stromal infiltration, thinning, and overlying epithelial defects suggestive of PUK (Figures 1 and 2). The ulcer had a circumferential spread over time, and corneal sensation was normal. The right eye exhibited nuclear sclerosis with a Grade 2 cataract, and the left eye was pseudophakic. Fundus examination was normal, with intraocular pressure of 12 mmHg in both eyes. Cultures from the peripheral ulcer were negative for any infectious organism. The patient had been treated elsewhere with topical prednisolone acetate every 4 hours without improvement in the signs and symptoms. A series of investigations were undertaken to determine the underlying cause. The results of the laboratory investigations were as follows: hemoglobin, blood count, and erythrocyte sedimentation rates were normal; serum rheumatoid factor and serum C-reactive protein were negative; antinuclear antibody by enzyme-linked immunosorbent assay was negative; serum antineutrophil cytoplasmic antibodies (ANCAs), cytoplasmic ANCAs, and perinuclear ANCAs were both negative; serum angiotensin converting enzyme levels were within normal limits; and serum anticyclic citrullinated peptide antibody levels were normal.

Mantoux test and chest X-ray were both unremarkable. The patient's venereal disease research laboratory and fluorescent treponemal antibody absorption tests were positive. Tests for other infective agents, such as hepatitis B and C and human immunodeficiency virus, were negative. The patient reported a history of sexual promiscuity, but there were no signs of active syphilis present on examination. The patient was then started on 2.4 million units of benzathine penicillin G intramuscularly once weekly for 3

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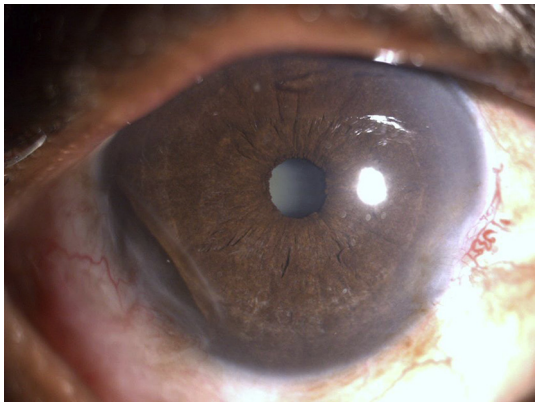

Figure 1. Peripheral ulcerative keratitis in the right eye.

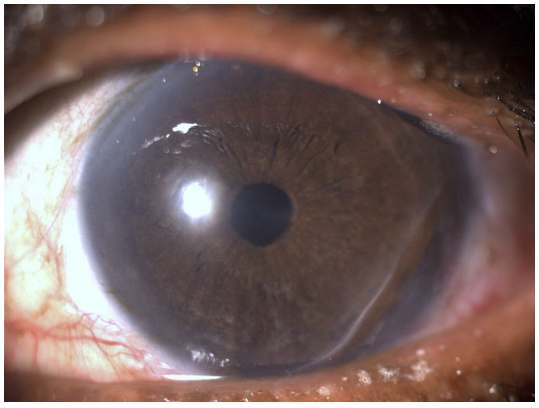

Figure 2. Peripheral ulcerative keratitis in the left eye.

consecutive weeks in addition to topical steroids for PUK treatment. The response to the treatment was good, and the ulcer started to heal. The patient was followed up regularly, and the vision improved in the right eye to 20/40 and in the left eye to 20/20.

3. Discussion

Syphilis is a sexually transmitted disease caused by *Treponema pallidum* that has been classified into primary, secondary, latent, and tertiary forms. Ocular syphilis usually manifests in secondary and late syphilis, with ocular manifestations involving almost every structure in the eye, including interstitial keratitis, anterior, intermediate, and posterior uveitis, chorioretinitis, retinitis, retinal vasculitis, and cranial and optic neuropathies.[1] Corneal involvement in acquired syphilis usually constitutes interstitial keratitis and is usually unilateral. PUK is an autoimmune disorder of the cornea in response to exogenous antigens.[2] Various etiologies were attributed to PUK, with the most common being rheumatoid arthritis and other autoimmune disorders, such as systemic lupus erythematosus and polyarteritis nodosa. The infectious causes implicated in its etiology include tuberculosis and herpes. It is usually unilateral. Bilateral PUK has been described in cases of sarcoidosis,[3] acute myeloid leukemia,[4] cat-scratch disease,[5] and treatment with rituximab.[6] Syphilitic involvement of the cornea is usually characterized by stromal keratitis; however, PUK in acquired syphilis is a rare entity. The pathogenesis of keratitis in ocular syphilis involves its cause by direct spirochete invasion or by an immune reaction against spirochaetal antigens. Merchant et al[7] suggested that antibodies against phospholipid epitopes of the treponemal outer membrane might cross-react with corneal lipids, such as phosphatidylglycerol and phosphatidylcholine, leading to keratitis. Radolf et al[8] suggested that *T. pallidum* eludes host clearance mechanisms, and its immune-evasiveness allows stealthy treponemes to circumvent phagocytosis and to persist in extracellular niches, pointing to keratitis caused by direct invasion. In this case, direct involvement of the spirochete may be the predominant factor, given that the keratitis responded to penicillin therapy. In our case, the presenting feature of the patient was bilateral symmetric PUK. This was a rare case where bilateral PUK was the presenting feature of acquired syphilis, and revealed the importance of initiating systemic penicillin in such scenarios.

In conclusion, syphilis, known as “The Great Imitator”, has varied clinical manifestations not only systemically, but also, in this case, in its ocular manifestations. Here, it presented as bilateral PUK, which is rarely reported in the literature. This case also showed the importance of initiating systemic antibiotics in treating infective causes of PUK. Our results indicate that syphilis should be kept in mind as a differential diagnosis in nonhealing cases of PUK.

Patient consent

Appropriate consent was obtained from the patient for the publishing of this article.

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