



СЛУЧАИ ИЗ ПРАКТИКИ CASE REPORT

Случай из практики

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Необычная причина серпигиозоподобного хориоидита. Связь с инфекцией *Mycobacterium Fortuitum* в моче?

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РЕФЕРАТ

Цель исследования – представить клинический случай развития двустороннего серпигиозоподобного хориоидита у пациента с урогенитальной инфекцией, вызванной *Mycobacterium fortuitum*. У 49-летней женщины, страдавшей потерей зрения, был диагностирован двусторонний серпигиозный хориоидит. Результаты обследования на наличие возбудителя были отрицательными, за исключением положительного результата кожной туберкулиновой пробы. Учитывая полученные данные и проживание пациентки в районе, эндемичном по *M. tuberculosis*, было начато эмпирическое лечение туберкулеза. Позже, в ходе заболевания, цитологический анализ мочи дал положительный результат на ARB, в посевах мочи был выделен *M. fortuitum*. Лечение атипичной микобактериальной инфекции привело к клиническому улучшению. После лечения рецидивов серпигиозоподобного хориоидита не наблюдалось. Это первый задокументированный в литературе случай серпигиозного хориоидита, связанный с инфекцией *Mycobacterium fortuitum*. Нетуберкулезные микобактерии также могут вызывать серпигиозный хориоидит.

Ключевые слова: *Mycobacterium fortuitum*, серпигиозоподобный хориоидит, увеит, инфекция мочевыводящих путей

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Case report

An Unusual Cause of Serpiginous-Like Choroiditis; Association With Urinary *Mycobacterium Fortuitum* Infection?

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ABSTRACT

The purpose of these case reports is to present a case who developed bilateral serpiginous like choroiditis in association with urogenital infection of *Mycobacterium Fortuitum*. A 49-year-old female who presented with visual loss was diagnosed with bilateral serpiginous like choroiditis. Uveitis workup findings were negative except for a positive tuberculin skin test positivity. Considering the findings and her residence in an endemic area for *m. tuberculosis*, empirical treatment for tuberculosis was started. Later in the course of the disease, her urine cytology came back positive for ARB, and *M. Fortuitum* was isolated in the urine cultures. Treatment for atypical mycobacteria resulted in clinical improvement. No relapses of serpiginous like choroiditis occurred following the treatment. This is the first documented case of serpiginous choroiditis related to mycobacterium fortuitum infection in the literature. Non-tuberculous mycobacteria may also cause serpiginous like choroiditis.

Keywords: *Mycobacterium Fortuitum*, Serpiginous-like choroiditis, Uveitis, Urinary tract infection

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INTRODUCTION

Non-tuberculous mycobacteria (NTM) are defined as mycobacteria other than *Mycobacterium tuberculosis* and *Mycobacterium leprae*. NTM are

present mainly in water and soil. They are occasionally responsible for trauma and/or surgery associated infections in humans [1]. *Mycobacterium fortuitum* is a rapidly growing NTM and associated with various ocular infections [1].

Serpiginous choroiditis (SC) is a progressive, bilateral, relapsing, and inflammatory disease that causes

chorioretinal atrophy. Syphilis, Herpesviruses, *Candida* spp, and *Toxoplasma gondii* have been blamed for playing a role in the pathophysiology of SC [2].

The term serpiginous-like choroiditis (SLC) was used to differentiate TB associated SC from classic SC [2]. Idiopathic noninfectious choroiditis that extends from the peripapillary area to the posterior pole with a geographic pattern is named as SC. Similar morphological involvement with a known etiologic infectious agent is SLC. Differentiation of these entities is vital for the management of disease [3].

To our knowledge, no cases of SLC have been reported in association with NTM infection. Herein, we report a case of SLC in association with *M. fortuitum*.

CASE PRESENTATION

A 49-year-old female presented with acute onset visual loss in both eyes. Her medical history was unremarkable. She had the history of being a former penitentiary prisoner. Her ophthalmological examination revealed that her visual acuity was counting fingers in both eyes. Intraocular pressures were within normal limits. Patient was phakic. There were +2

cells in the anterior chamber, seclusio pupilla and Busacca nodules, mild vitritis, and chorioretinal scars at the posterior pole and peripapillary area in both eyes. Fundus fluorescein angiography and optical coherence tomography clearly showed signs of chorioretinal scars and atrophy along with cystoid macular edema (Fig. 1, 2).

With the pre-diagnosis of SLC, diagnostic tests were performed. A review of systems revealed back pain with inflammatory characteristics. The patient was referred to the rheumatology and pulmonology departments, but they were unremarkable. The blood count was normal. Blood anti-neutrophil antibody, angiotensin-converting enzyme, rheumatoid factor, and anti-citrullinated protein antibodies testing were negative. Serologies for human immunodeficiency virus and syphilis were negative. Thorax computerized tomography, sacroiliac joint, and chest radiograph were unremarkable. The tuberculin skin test (TST) was 20 mm at 48 h, while the QuantiFERON TB gold test was negative. Further diagnostic work-up revealed microhematuria. Urine cytology was used to investigate urogenital TB. Acid-fast-bacillus was detected in one of the samples; however, *M. tuberculosis* polymerized chain reaction was negative. Mycobac-

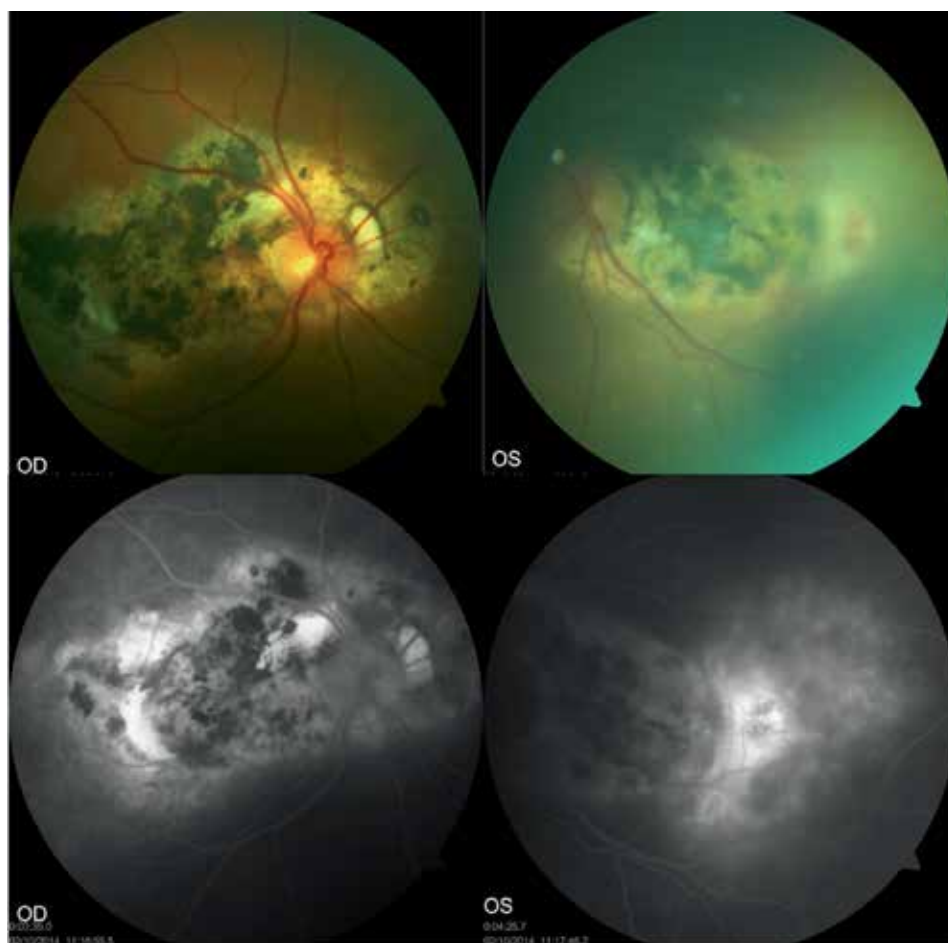


Fig. 1. Fundus fluorescein angiographies (left column) and colored fundus images (right column) of the patient's right eye (upper) and left eye (lower). Note the extensive scar formation in the macula. Active choroiditis focus was noted in the juxta-temporal part of the macula

Рис. 1. Флуоресцентная ангиография глазного дна (левая колонка) и цветные изображения глазного дна (правая колонка) правого глаза пациента (верхний рисунок) и левого глаза (нижний). Обратите внимание на обширное рубцовое образование в макуле. Активный очаг хориоидита был отмечен в южно-височной части желтого пятна

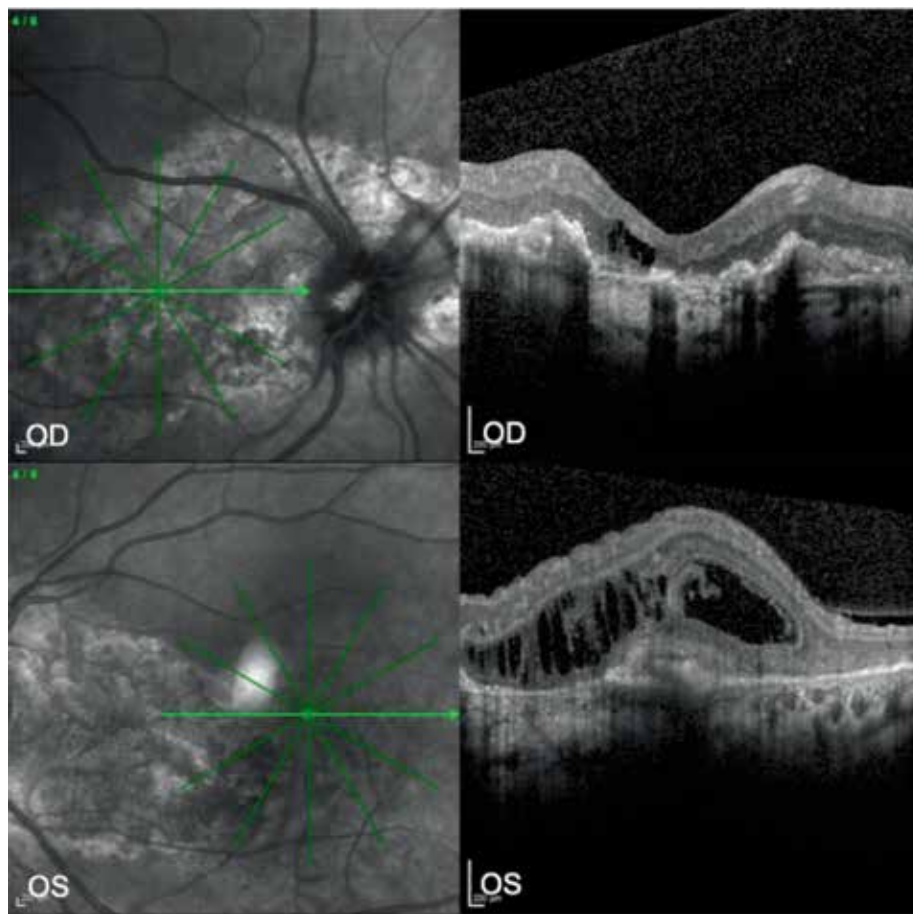


Fig. 2. Infrared images (left column) and optical coherence tomography images (right column) of the patient's right eye (upper) and left eye (lower). Note the intraretinal edema in the active focus of choroiditis

Рис. 2. Инфракрасные изображения (левая колонка) и изображения оптической когерентной томографии (правая колонка) правого глаза пациента (верхний рисунок) и левого глаза (нижний). Обратите внимание на интратретинальный отек в активном очаге хориоидита

terium culture was requested. While pending identification and sensitivity of test results, a four-drug regimen (isoniazid, ethambutol, pyrazinamide, and rifampin) was started. The patient had also received 1 mg/kg oral methylprednisolone treatment for a week, and then it was tapered slowly.

Urine mycobacterium culture was reported positive for *M. fortuitum* group (Mycobacterium Other Than Tuberculosis, MOTT). A combination of ciprofloxacin, clarithromycin, and trimethoprim/sulfamethoxazole was swapped for MOTT. Antimicrobial susceptibility was found as 100 %, 80 %, and 100 % for these antibiotics, respectively. During treatment, urine cytology turned out to be negative. The patient is still under close follow-up with no activation of choroiditis since then (Fig. 3).

DISCUSSION

The pathogenesis of SC is accepted as an "idiopathic" intraocular inflammation with a possible organ-specific autoimmune process. There are some clinical features belonging to SLC rather than SC like similar to our patient. [4] In our case, moderate ocular inflammation with excessive posterior pole atrophy and scar formation suggested that

an infectious agent could be a cause. Since TST results came back positive for an endemic region, ATT had been started with pre-diagnosis of latent TB associated SLC.

There are mainly two mechanisms that TB bacilli blamed for choroiditis. The first hypothesis is direct tissue invasion by *M. tuberculosis*, and supportive pieces of evidence have shown in different studies [4, 5]. The second hypothesis is the immune reaction to tubercular antigens. There is also evidence of immune-related disease [6].

Although we ran up all tests for *M. tuberculosis*, there was not any clue for different organ system involvement. The positivity of the TST results might be related to the history of living in TB endemic area or penitentiary history, NTM cross-reactivity, and previous BCG vaccination [7]. Therefore, SLC is thought to be related to *M. fortuitum* infection. However, it is impossible to rule out *M. tuberculosis* infection with certainty. *M. fortuitum* and *M. tuberculosis* share some common immunodominant antigens [8]. This possible TST cross-reactivity between them might be playing a role in pathogenesis in SLC.

To our knowledge, there are choroiditis, iridocyclitis, and panuveitis associated with NTM in the literature [1]. The majority of these cases had impaired cellular immunity

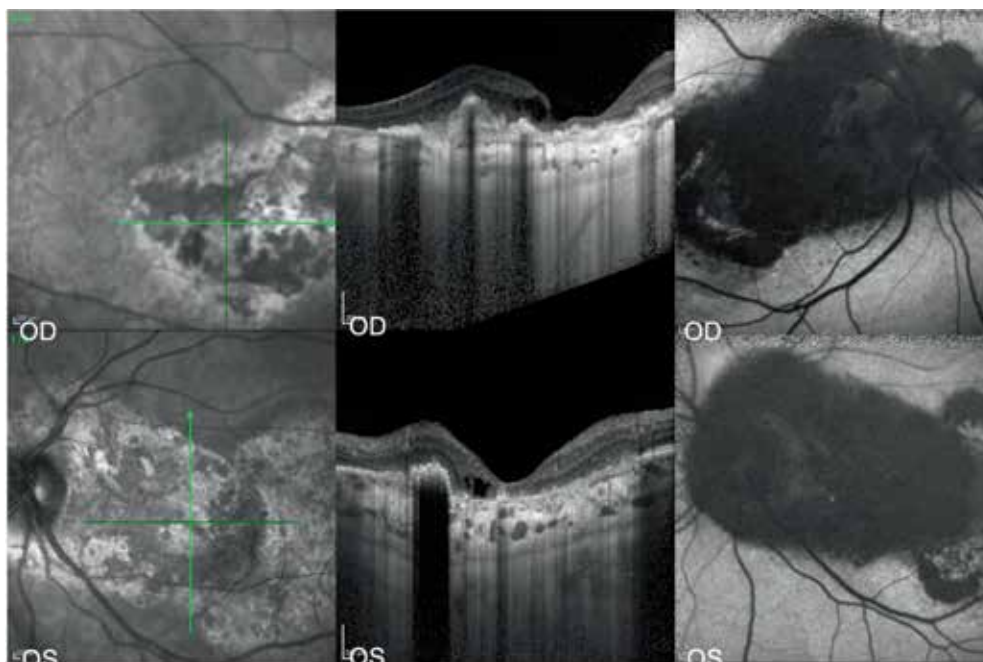


Fig. 3. Infrared images (left column), optical coherence tomography images (middle column) and autofluorescence images (right column) of the patient's right eye (upper) and left eye (lower). Note the regression of intraretinal edema and consequent scar formation

Рис. 3. Инфракрасные изображения (левая колонка), изображения оптической когерентной томографии (средняя колонка) и изображения автофлуоресценции (правая колонка) правого глаза пациента (верхний) и левого глаза (нижний). Обратите внимание на регрессию интратретинального отека и последующее образование рубца

conditions like HIV/AIDS or steroid usage; however, infections can also occur in healthy hosts [9]. A 37-year-old patient by the history of steroid usage was reported with choroidal granuloma associated with *M. fortuitum* [10].

M. fortuitum is susceptible to four-drug anti-TB agents in our antibiogram, so the patient received antimicrobial treatment without delay. However, the treatment was swapped to the newer and safer agents with shorter treatment periods.

This is the first SLC case possibly associated with *M. fortuitum* infection and NTMs. However, urinary *M. fortuitum* infection may be incidental. The exact discrimination of this is only possible with intraocular sampling and molecular testing. However, the ocular inflammation was regressed with antibiotics. We strongly suspect that the same organism as the one found in the urine was causing ocular disease and suggest that *M. fortuitum* could be the causative agent for SLC. However, further case reports and series would be needed to confirm such association.

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Gamze Ucan Gunduz – collected the data and wrote the paper

Ozgur Yalcinbayir – revised the paper

Oner Gelisken – revised the paper

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