# CASE REPORT

# Otomastoiditis Caused by Rhodococcus equi in a Patient With AIDS

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ABSTRACT. Rhodococcus equi is a well-recognized pathogen in veterinary medicine and a rare but well-documented cause of cavitary pneumonia in immunocompromised patients. Most cases of Rhodococcus equi infections in these patients involve the lungs. Otomastoiditis due to Rhodococcus equi is

rare, and disseminated *Rhodococcus equi* with otomastoiditis has never been reported. We report a case of otomastoiditis with systemic dissemination due to *Rhodococcus equi* in a patient with AIDS. *Key words: Otomastoiditis, Rhodococcus equi, AIDS* 

hodococcus equi is a well-documented veterinary pathogen causing granulomatous pneumonia in foals (1-3). Most cases of *Rhodococcus equi* have occurred in immunocompromised patients, especially those infected with human immunodeficiency virus (HIV) (1-4). The majority of cases have involved patients with advanced HIV infections (1). Clinical illness is usually manifested by chronic or relapsing cavitary pneumonia; extrapulmonary disease is unusual and otomastoiditis is rare. To our knowledge, we report the first case of otomastoiditis with pulmonary *Rhodococcus equi* and systemic dissemination in a patient with AIDS.

## **Case Report**

A 31-year old hispanic male, HIV seropositive since 1995 came to our hospital on August 27 1998 with persistent purulent discharge from the right ear of 3 months duration and right facial weakness and pain. He had right facial paralysis since one week prior to admission

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and productive cough, with blood tinged yellow/green phlegm for several months. He also complained weight loss and anorexia for several weeks, fever, chills and occasional headaches, pleuritic chest pain, in the back and shoulders of 3-4 day duration. He denied blurred vision, neck stiffness, nausea, vomiting, diarrhea or abdominal pain.

The patient had esophageal candidiasis in the past and he denied recent travel or exposure to tuberculosis or close contact with animals. Upon admission he was receiving fluconazole and clindamycin and had not been on antiretroviral therapy for the previous two years.

On physical examination, he was in moderate respiratory distress. His vital signs included temperature of 101° F, blood pressure 122/66 mmHg, heart rate 93 per minute, respiratory rate 30 per minute. A right facial palsy was present and had a purulent discharge from the right ear with perforated tympanic membrane. Decreased breath sounds on the right lung and diffuse bilateral crackles were heard, bilateral inguinal lymphadenopathies were noted. Initial laboratory values showed hemoglobin of 8 g/dl, hematocrit of 24.4%, and white blood cell count of 8,900/mm with 88% neutrophils, 7% lymphocytes, 4% bands and 1% monocytes. Liver and renal function tests were normal. The absolute T cell count was 94/mm and the viral load was 285,000 copies/ml.

Chest radiography showed a right pneumothorax with patchy opacities and mild pleural effusion within the right lung base, with cavitary lesions in the right middle lobe. On head computed tomography the right mastoid air cells were full with soft tissue, with multiple bony destruction in the right middle ear (figure 1, 2). Purulent material from the right ear yielded *Rhodococcus equi*. Three of 4 blood culture specimens showed

Rhodococcus equi. The culture from sputum also



**Figure 1.** Nonenhanced axial CT scan in bone windows, demonstrates extensive destruction of the petrous portion of the right temporal bone with associated soft tissue density. Note the opacification of the mastoid air cells. Ethmoid disease is also identified



**Figure 2.** Nonenhanced coronal CT scan shows destruction of the right temporal bone with soft tissue and erosion of the middle ear. The mastoid air cells are opacified and sclerotic.

yielded *Rhodococcus equi*. One sputum specimen was positive by flourochrome stain. The soft tissue from right ear biopsy showed acute inflammation, necrosis, fibrinous exudate and collection of granular macrophages with intracytoplasmic bacterial organisms consistent with malakoplakia. Cytology from this soft tissue was negative for malignant cells. The organism was susceptible to vancomycin, levofloxacin, erythromycin and gentamicin.

The patient was initially treated with intravenous ciprofloxacin, vancomycin and trimethoprimsulfamethoxazole (TMP-SMX) and continued on levofloxacin and clarithromycin. Two months after being discharged (November 1998), there was no evidence of relapse. He remains with some degree of right facial paralysis and loss of audition in his right ear. At the time of this report his overall condition has improved significantly with antiretroviral therapy. The T-cell count has increased to 196/mm from 94/mm and the viral load had decreased to 2031 copies/ml from 285,000 copies/ml.

## Discussion

This case represents the first report of *Rhodococcus equi* otomastoiditis from systemic dissemination in a patient with AIDS. Previously Lopes Cardoso et al (6) reported a case of mastoiditis caused by *R. equi* in a patient with AIDS. In that case the *R. equi* was isolated only from ear/mastoid abscess drainage. Our report represents a unique case of a patient with AIDS who had a disseminated *R. equi* infection that did involve ear/mastoid, lungs and blood stream with a relatively benign clinical course that has responded well to antimicrobial and local debridement therapy.

Rhodococcus equi (formely Corynebacterium equi) is a facultative intracellular gram-positive, aerobic, nonspore forming bacillus usually found in soil and in the feces of herbivores (2). It is a well-established pathogen in veterinary medicine, usually causing pneumonia and sepsis in horses and cattle. R. equi was first isolated by Magnusson in 1923 from suppurative lung lesions in foals (5), he named it Corynebacterium equi and in 1980 was re-classified as R. equi (9). Although common farm mammals such as horses, swine, cattle and sheep are the usual reservoir, the pathogens has been isolated from other animals including seals, marmosets, koala bears and blood-sucking arthropods (7). It is extremely uncommon in dogs and cats (8). Infection in both humans and grazing animals is thought to be acquired via the respiratory tract (10). Less commonly, infection has been observed to occur via the oral route (11). Although human infections due to R. equi are uncommon, this bacteria is becoming

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an increasingly important opportunistic pathogen in immunosuppresed patients. Before the 1980's, most patients with *R. equi* had either underlying malignancies or were receiving immunosuppresive drugs (12). In 1986, the first case of *R. equi* infection was reported in a patient seropositive to HIV who presented with persistent *R. equi* bacteremia and a lung abscess (14). Since the early 1980's, patients with HIV infections have represented a large and growing proportion of the reported cases (7).

Linder reviewed 115 cases of R. equi infection between 1967 and 1996 and most of them were immunocompromised. Between 1967 and 1976, seven cases were reported and six of them were immunocompromised (13). Between 1977 and 1986, fifteen cases were reported and 7 of them were HIV seropositive individuals, and between 1987-1996, ninety three additional cases were reported, 78 of them were immunocompromised and of those, 67 were HIV seropositive. Pulmonary involvement was present in 80% of the latter. Capdevila et al (15) reviewed 76 cases of R. equi lung infection in patients infected with HIV. The majority (71/78) met the criteria for AIDS at the time of diagnosis: 60 had CD4 lymphocyte count under 200/mm, while the remaining 11 had experienced an opportunistic infection. This suggests that patients with advanced HIV with low T-cell count are at risk of R. equi infection. The most frequent symptoms on admittance to hospital were fever and cough (84%), followed by constitutional syndrome (40%). The most frequent radiological lesion was single cavitary lesion in 57%, mainly in the upper lobes (56%).

In the immunocompromised host, a subacute pulmonary infection is the most common presentation of *R. equi* disease. It typically begins with an insidious onset of symptoms, including fever, malaise, cough, dyspnea and weight loss. Pulmonary relapse is common and generally thought to be due to inadequate duration of therapy or lack of T-cell classes to effectively clear infection. Kanaly et al (17) reported that Th-1 CD4 T-cells are critical for pulmonary clearance of *R. equi*. CD4 T-cells and gamma interferon appear to be required for complete clearance of pulmonary infections with *R. equi*. Pneumonic manifestations are extremely uncommon in immunocompetent persons (7).

The radiographic findings in *R. equi* pneumonia are similar to those found in normal hosts with tuberculosis or nocardiosis-infiltrate that progress to form thick-walled cavities. Cavitation and effusion are unusual in HIV-infected individuals with tuberculosis (18-20). The differential diagnosis of cavitary pulmonary lesions in HIV patient is broad, especially in patients with advanced disease. It occurs frequently in patients with invasive pulmonary aspergillosis, *Mycobacterium kansasii*,

pneumonia due to *Pseudomona aeruginosa*, *Nocardia asteroides* and *Rhodococcus equi*. It is unusual with pulmonary cryptococcosis, coccidioidomycosis, and histoplasmosis.

Extrapulmonary disease has been reported as a result of either contiguous or hematogenous dissemination of the pathogen. The spectrum of extrapulmonary infections reported with *R.equi* includes bacteremia, endophthalmitis, tenosynovitis/septic arthritis, meningitis, osteomyelitis, peritonitis, bloody diarrhea, colonic polyps, wound infection and granulomatous dermatitis (7, 21). Otomastoiditis due to *R. equi* is unusual, and it has not been previously reported with systemic dissemination.

Treatment of infection with R. equi requires antibiotics with intracellular concentration and activity. The aminoglycosides, especially gentamicin, imipenem and the glycopeptides (vancomycin and teicoplanin) are the most effective antibiotics in the bacteremic phase with low resistance and high bactericidal effect (15). The macrolides, the quinolones and rifampin have high intramacrophage bactericidal activity. The combination of erythromycin with rifampin showed synergy against R. equi (22). Clindamycin, TMP-SMX, chloramphenicol., tetracyclines, azithromycin, clarithromycin and ciprofloxacin also showed activity against Rhodococci. The B-lactam antibiotics (with exception of imipenem) should be avoided because of the rapid development of resistance (20). In large focal lesions, surgical drainage and debridement in combination with antimicrobial therapy may be needed. The duration of the therapy remains uncertain, the treatment is maintained according to the clinical evolution of the patient and the result of surveillance cultures. The presence of intramacrophage bacteria requires prolongation of treatment for long period of time, since relapses are frequent when treatment is interrupted, especially in patients with HIV infection. The length of therapy should be determined on an individual basis. Patients with adequate response to antiretroviral therapy, may recover some degree of immunity with decreased viral load and increased T-cell count which may eradicate the organism without recurrent infection. Some experts suggest the need for long term therapy, generally from 2 months to life-long treatment. The mortality can approach or exceed 50% among patients with HIV infection, with an overall mortality of 26% (16).

In summary, we report a case of disseminated R. equi infection involving the lungs, ear/mastoid in an immunocompromised HIV infected patient. The chest radiographic findings with cavitary lesions characteristic of R. equi pneumonia suggests a respiratory tract acquisition of the infection. In our patient, the intense immunosuppression state was the risk factor of R. equi,

and otomastoiditis probably represent hematogenous dissemination from the pulmonary lesion.

Rhodococcus equi infection should be in the differential diagnosis of cavitary pneumonia and disseminated infection in patients with advanced HIV infection, particularly when cultures fail to identify mycobacteria, nocardia and fungal infections. As the prevalence of HIV infection and organ transplantation increases, R.equi infection may reach greater clinical importance.

### Resumen

Rhodococcus equi es un patógeno bien reconocido en medicina veterinaria y aunque raro, se ha documentado como causa de pneumonias en pacientes inmunocomprometidos. La mayoría de casos de Rhodococcus equi en estos pacientes afecta los pulmones. La otomastoiditis por Rhodococcus equi es rara, y la otomastoiditis con diseminación sistémica no ha sido reportada en la literatura médica. Se informa y discute un caso de otomastoiditis con diseminación sistémica por Rhodococcus equi en un paciente con SIDA

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