

Rev. Inst. Med. Trop. Sao Paulo
53(5):291-294, September-October, 2011
doi: 10.1590/S0036-46652011000500009

CASE REPORT

***Rhodococcus equi* INFECTION AFTER REDUCTION MAMMAPLASTY IN AN IMMUNOCOMPETENT PATIENT**

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SUMMARY

The majority of infections caused by *R. equi* occur in hosts with some degree of cell-mediated immunodeficiency. Immunocompetent individuals are infrequently affected and usually present with localized disease. Infections of the skin or related structures are uncommon and are usually related to environmental contamination. The microbiology laboratory plays a key role in the identification of the organism since it may be mistaken for common skin flora. We describe a 31 year-old woman without medical problems who presented nine weeks after breast reduction with right breast cellulitis and purulent drainage from the surgical wound. She underwent incision and drainage, and cultures of the wound yielded *Rhodococcus equi*. The patient completed six weeks of antimicrobial therapy with moxifloxacin and rifampin with complete resolution.

KEYWORDS: *Rhodococcus equi*; Skin infection; Wound infection; Mammoplasty.

INTRODUCTION

R. equi is a pleomorphic facultative, intracellular, nonmotile, non-spore forming, gram-positive coccobacillus that can be weakly acid-fast. It appears coccoid on solid media and in clinical specimens, but in liquid media it forms long rods or short filaments. Although it is named for its production of red pigment, cultures need 4 - 7 days to demonstrate the salmon pink colonies. It is an intracellular pathogen and the basis for its pathogenicity is its ability to survive inside macrophages, causing inflammation, cell destruction, and purulent granulomas with progression to caseating necrosis. Histopathology shows necrotizing granulomas with macrophages, or an unusual chronic granulomatous inflammatory process that is associated with an impaired ability to process microorganisms within histiocytes called malacoplakia^{9,17}. The majority of the *Rhodococcus* infections occur in immunocompromised individuals, most commonly in patients with impaired cell mediated immunity (e.g. leukemia, lymphoma, HIV infection, solid organ transplant recipients, etc); two-thirds of reported case have occurred in patients with HIV infection^{9,11,17}. For reasons that are not clear, the male to female ratio is 3:1 for both immunocompetent and immunocompromised patients⁷. Skin manifestations are rare even in immunocompromised hosts¹, and only a few cases of skin infections, including abscesses, caused by *Rhodococcus* among immunocompetent patients have been reported. Soil contamination was determined to be the predisposing factor in two cases (one cured with surgical debridement alone and the other with

antimicrobial therapy), in one case the predisposing source was unknown (cure was achieved with antimicrobials and surgical debridement), and in one case, scalp infection was determined to be caused by self-inoculation^{3,5,10,12}. We present the first report of skin infection with *Rhodococcus equi* after breast reduction surgery.

CASE REPORT

A 31 year old Hispanic female patient without previous medical problems presented to St. Luke's-Roosevelt Hospital Center with a 10 day history of erythema, swelling and induration over her right breast, accompanied by purulent discharge. Although she recalled having chills and subjective fever; no temperature was recorded. She denied other systemic symptoms including, chest pain, and shortness of breath, abdominal pain, nausea, vomiting, or diarrhea. She had traveled to the Dominican Republic three months earlier in order to have bilateral breast reduction mammoplasty and abdominoplasty. She stayed in the city and did not visit any rural areas during the whole stay. She and her sister went together and both had the same surgical procedure performed without immediate postoperative complications. The patient recalled performing the indicated postoperative care (which she said only consisted of dry dressing changes until stitches were removed) and denied insect bites or any contact with animals. Six weeks after the procedure, she noticed purulent discharge from the abdominoplasty scar. She was seen by a local physician, who diagnosed a postsurgical wound infection and prescribed

Potential conflicts/disclosures: the authors have no conflicts of interest or disclosures.

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Fig. 1 - Right breast showing erythema on the lower quadrants and a crusted area over the surgical scar which drained purulent material.

ciprofloxacin 500 mg orally twice daily for 10 days long with local wound care. Routine aerobic wound cultures taken at that time revealed no pathogens, and her symptoms resolved after the administration of antimicrobial therapy. Approximately nine weeks after the surgery, the patient developed tenderness and purulent discharge from the right breast incision and came to our institution for further evaluation. The patient was not in any distress, her vital signs were unremarkable and temperature was normal. Examination revealed a 3 x 1 cm erythematous and indurated area with purulent discharge over the surgical scar on her right breast (Fig. 1). The area was warm and tender to touch. The abdominal scar showed no erythema or drainage. Initial laboratory results showed a white blood cell count of 8.4 K/ μ L (70% neutrophils); the rest of the complete blood count as well as blood chemistries were normal. The patient underwent incision and drainage. Gram stain of the operative specimen showed gram-positive coccobacilli that were partially acid-fast bacteria on modified Kinyoun stain. Small colonies were appreciated on solid media (blood and chocolate agars) at 24 hours, and after four days developed into salmon-pink color colonies. Growth was also observed

on Middlebrook 7H10 agar although no mycobacteria were isolated. Gram stain performed on the Middlebrook 7H10 agar growth, showed branching filamentous gram-positive rods that were also modified acid-fast positive. The organism, which was catalase-positive and nonmotile, was identified as *Rhodococcus equi* using a RapID™ CB Plus system (Remel Products, Thermo Fisher Scientific, Kansas, USA), used to identify medically important coryneform bacteria and gram positive bacilli. The organism was susceptible to ciprofloxacin, moxifloxacin, erythromycin, rifampin, imipenem, vancomycin and gentamicin. A combination of oral moxifloxacin 400 mg and rifampin 600 mg, both daily, was begun. Serologic evaluation for anti-human immunodeficiency virus (HIV) antibody test was negative by ELISA testing. The patient's wound infection resolved without complications and antimicrobials were stopped after a total of six weeks. She has been without recurrence for more than four years, and continues to follow up periodically in clinic.

DISCUSSION

R. equi lives in soil and it is found in 50-95% of farms. Concentrations are high in horse manure and it is carried in the gut of many herbivores. Infection is primarily acquired through the respiratory tract, but it can also be acquired by the oral route and by traumatic inoculation or superinfection of wounds^{3,10,12,17}. Nosocomial cases have been only reported in immunocompromised patients and one case of occupational acquisition by a healthy laboratory worker who developed pneumonia has also been described^{4,16}. Our patient did not recall any other exposure other than the surgical intervention, she denied having visited farms or having contact with animals.

The incidence of infections due to *R. equi* increased markedly in the early eighties, probably related to the HIV epidemic, advances in chemotherapy for patients with cancer, organ transplantation and also improvements in microbiology laboratory identification techniques^{9,17}.

Immunocompetent patients tend to have localized disease whereas infections can be disseminated in patients with underlying immunodeficiency. Pneumonia is the most common manifestation with most patients showing radiological evidence of cavitation. Other manifestations include lymphadenitis, endophthalmitis, musculoskeletal

Table 1
Summary of the reported cases of skin infections caused by *Rhodococcus* in immunocompetent patients

Authors	Age (years) and gender	Possible predisposing factors	Localization	Treatment	Outcome
CASTOR, <i>et al.</i> ³	35, female	Self inoculation	Leg ulcer	Multiple antimicrobial courses	Relapsed with repeated infections and multiple pathogens
GARCIA MORILLO, <i>et al.</i> ⁵	76, female	Unknown	Left thigh, abscesses	Surgical debridement plus Ciprofloxacin and rifampin	Cured
MULLER, <i>et al.</i> ¹⁰	13, male	Soil contamination	Knee and foot	Surgical debridement and irrigation. No antimicrobials	Cured
NASSER, <i>et al.</i> ¹²	16, male	Soil contamination	Scalp	Erythromycin and rifampin	Cured

and central nervous system infections. Indwelling catheter infections and a liver abscess have also been described. Bacteremia occurs in 30% of normal hosts but in up to 80% of immunocompromised patients. Relapses are a well-known feature manifesting at the initial site of the disease or at distant locations^{6,7,9,16,17}.

Since *R. equi* is not a common pathogen, a high index of suspicion is required or the diagnosis may be easily missed. Identification of the organism from microbiological specimens is the method of choice. The organism grows easily but may be dismissed as a contaminant, so laboratory personnel should be notified if *R. equi* is suspected (e.g., immunocompromised patients with cavitary pneumonia). The organism has been isolated from virtually any site¹⁷.

The isolation of *Rhodococcus* in our patient's wound culture was both striking and unexpected as a cause of wound infection. In a retrospective series, more than half of the skin infections after breast surgery were caused by *Staphylococcus aureus*, followed by *Pseudomonas aeruginosa*, Mixed flora, *Coagulase-negative staphylococci*, Enteric gram negative rods and *Streptococcus viridans* group¹⁴. Apart from common skin pathogens, cases of nontuberculous mycobacterial infections (especially *Mycobacterium fortuitum*) complicating augmentation mammoplasty and breast reconstruction have been reported, but more recently *M. fortuitum* and *Mycobacterium chelonae* were also cultured from patients who underwent reduction mammoplasty^{2,8,15}.

Most cases reported in the literature occurred following contamination of the skin or soft tissues^{1,3,5,10,12,17}. Although we considered soil contamination as the likely source for infection in our patient, we initially saw her several weeks after the surgery was performed so we were unable to prove it. Table 1 summarizes the reported cases of skin infections caused by *Rhodococcus* in immunocompetent patients.

Guidelines or standards for treatment of *R. equi* have not been established due to the small number of reported cases. Single antimicrobial therapy has proven ineffective therefore is not recommended. The use of combination therapy with two to three antibiotics is the mainstay of treatment, along with surgical drainage of large cavities and abscesses. Because *R. equi* is an intracellular organism and can survive inside macrophages some authors have advocated treatment with drugs that achieve high intracellular concentrations; this statement is, however, unclear with others just advocating the use of bactericidal drugs in the initial phase of therapy^{9,17}.

R. equi is usually resistant to penicillins and, even when susceptible, their use is not recommended due to the rapid emergence of resistance⁷. Preferred antimicrobials based on *in-vitro* susceptibilities include imipenem, vancomycin, linezolid, fluoroquinolones, erythromycin, rifampin and the aminoglycosides. Clindamycin, the tetracyclines, chloramphenicol, trimethoprim-sulfamethoxazole and cephalosporins have variable *in vitro* susceptibility and should not be used as first line agents. In an animal model, the most effective agents were vancomycin, imipenem, and rifampin¹³. Our patient was treated with a combination of rifampin and moxifloxacin (both achieve excellent intracellular concentrations), showing resolution of symptoms.

Localized infections can usually be treated with oral antimicrobials and therapy should be continued until clinical improvement occurs with

resolution of signs and symptoms. The prognosis of patients with normal immune function is usually favorable^{9,17}. No data is available regarding prognosis in patients with localized skin infections like ours.

This case illustrates the importance performing appropriate cultures before the administration of antimicrobial therapy and also the key role of the microbiology laboratory in the identification of uncommon pathogens. To our knowledge this is the first case of *R. equi* following reduction mammoplasty described in the literature.

AUTHOR CONTRIBUTIONS

All authors contributed equally.

ACKNOWLEDGEMENT

The authors would like to thank the Microbiology laboratory at St. Luke's Hospital for their hard work and dedication.

RESUMEN

Infección por *Rhodococcus equi* luego de cirugía de reducción mamaria en huésped inmunocompetente

La mayoría de las infecciones causadas por *Rhodococcus equi* ocurren en huéspedes con algún grado de inmunodeficiencia celular. Los individuos inmunocompetentes son afectados con baja frecuencia y suelen presentarse con enfermedad localizada. Las infecciones de la piel o partes blandas son poco frecuentes y están usualmente relacionadas con contaminación ambiental. El laboratorio de microbiología juega un papel clave en la identificación del organismo, ya que este puede confundirse con flora normal de la piel. Se describe una mujer de 31 años sin problemas médicos que consultó nueve semanas después de haber sido sometida a cirugía de reducción mamaria, con celulitis del seno derecho y drenaje purulento de la herida quirúrgica. Se practicó incisión y drenaje quirúrgico y los cultivos de la herida demostraron *R. equi*. La paciente recibió seis semanas de tratamiento antimicrobiano con moxifloxacin y rifampicina demostrando resolución completa.

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Received: 17 May 2011

Accepted: 24 August 2011