



## A Fatal Infection due to *Gordonia Terrae*

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Mycetoma, Actinomycetoma, *Gordonia terrae*, Surgery, rRNA sequencing

Mycetoma is a chronic granulomatous, non-contagious disease of the subcutaneous tissues, this most often caused by *Nocardia*, *Actinomycetes* or fungi [1]. It is endemic in tropical and subtropical areas but not in temperate climates such as South Korea [2]. Among many causative pathogens for mycetoma, *Gordonia* species, rare nocardioform actinomycetes, are difficult to be identified by microbiologic and serologic tests [3,4], because the pathogen requires a precise identification using genomic sequencing [1]. In addition, *Gordonia* species have been reported in just a few numbers of case series, and its epidemiology and clinical importance have been underestimated [5]. Herein, we report a rare case of mycetoma caused by *Gordonia terrae* in an immunocompetent patient in a temperate climates area.

A 53-year-old man with a history of poliomyelitis underwent left total hip arthroplasty in December 2011. His past medical history was not significant except the medical conditions described disease. The patient had developed a sore on right perianal area since March 2013 (Figure 1A), and infected sore lesion was extended to abscess on a right buttock abscess one year later, it was managed with incision and drainage in right buttock abscess since November 2014. However, a few months later, a multifocal erosions and a sinus tract were presented in the same lesion (Figure 1B). The patient was again admitted for extensive surgical treatment. On admission, his body temperature remained within a normal range during hospitalization. Laboratory data showed white blood cell count  $14,200 \times 10^3/\mu\text{L}$ , hemoglobin 8.8 mg/dL, high sensitivity C-reactive protein concentration 11.21 mg/dL, and erythrocyte sedimentation rate 120 mm/hr. Other laboratory results were normal. An initial computed tomography of the pelvis revealed an enhancement of skin and soft tissues on the right buttock

area. Consequently, intravenous antibiotics treatment with daily 2 gram of ceftriaxone was initiated and continued for 7 days along with drainage. An initial blood culture result showed no pathogens, but a tissue culture of the wound site identified a penicillin susceptible *Streptococcus anginosus* and methicillin susceptible *Staphylococcus aureus*. All laboratory examinations for *Mycobacterium tuberculosis*, nontuberculous mycobacteria, and fungi were negative. Therefore, intravenous ceftriaxone was switched to intravenous Cefazolin with a daily dose of 6 grams (2 grams three times per day) according to antibiotic susceptibility test. However, the skin lesion continued to deteriorate despite adequate antibiotics trials. Following the result for gram stain of the wound site revealed a gram-positive bacilli, and the colonial morphology showed small and white-colored convex colonies on blood agar (Figure not shown). Furthermore, *Corynebacterium* species were identified in blood culture using the Vitek II system (bioMerieux Vitek Inc., Durham, NC). Consequently, vancomycin, ceftriaxone and clindamycin combination, and piperacillin/tazobactam were tried to target *Corynebacterium* species for a total of 4 months during hospitalization due to lack of response. In addition, the patient underwent incision and drainage procedures more than twenty times over the course of hospitalization. Despite medical and surgical treatments, his medical condition did not improve, and the extent of lesion became deeper and wider. Therefore, we decided to identify by 16S rRNA gene sequencing for gram-positive bacilli from blood and wound specimens at the another microbiology and immunology laboratory, the gram-positive bacilli was identified as *Gordonia terrae* by 16S rRNA gene sequencing (96% match with type strain) using a pair 27FLP (5'-AGAGTTTGATCMTGGCTCAG-3') and 1492RPL (5'-GGTTACCTTGTTACGACTT-3') [5,6]. Unfortunately, an antibiotics susceptibility test was not performed due to lack of technological availability at our institution. As reported in previous articles, an antibiotic regimen was changed to a combination of intravenous amoxicillin/sulbactam and ciprofloxacin for 7 days. Despite changes in antibiotics regimen, his medical condition did not show any improvement, which made us consider

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**Figure 1:** Clinical photo showing the changed morphologies of a patient's lesion and histopathology of mycetoma.

(A) There are no signs of active inflammation until October 2014; (B) He had a pus discharge and multifocal sinus tract in the right hip in July 2015 (white arrowhead; sinus tract); (C) The bulk tumorous lesion was showed in the subcutaneous tissue of right hip in July 2015 (after 7 months) (black arrow, granulation tissues); (D) Tissue section of a biopsy of the lesion showed a proliferations of vascular tissue with reactive endothelial cells, plump fibroblasts, and a mixed inflammatory infiltrate (granulation tissue). Focally, a dense cluster of neutrophils is seen in the vascular lumen. (H & E,  $\times 100$ ).

a wider surgical resection rather than an incision and drainage procedure. However, the patient refused both blood transfusion for anemia of chronic disease and surgical treatment causing intraoperative blood loss, owing to the beliefs of Jehovah's Witnesses. Then, we alternatively switched an antibiotic regimen to 500 mg of intravenous imipenem per 8 hours and 500 mg of levofloxacin per 24 hours. Despite multiple alterations in antibiotic regimens, the wound still continued to worsen for the following two weeks. At last, the patient was transferred to another hospital where surgical resection was possible without blood transfusion. However, he had refused the operation after all and was transferred back to our hospital three months later. While delaying in treatment, the wound developed to painless mycetoma in the subcutaneous tissue of right hip (Figure 1C and Figure 1D). Unfortunately, the patient died due to septic shock and multiple organ failure.

Mycetoma which is reported 97% were actinomycetoma and 3.5% were eumycetoma is a major problem in tropical and subtropical areas [1]. The trial of a painless firm subcutaneous mass, multiple sinus formation, and a purulent discharge that contains grains is pathognomic of mycetoma [1]. Our case fulfilled the above signs. We identified *Gordonia terrae* from blood and gram positive bacilli from the wound by the 16S ribosomal RNA gene sequencing. To our knowledge, mycetoma caused by *Gordonia terrae* is a rare in temperate climate, although a case series has been just reported in worldwide.

*Gordonia terrae* has been reported in central venous catheter related infection [5,7-12], primary bacteremia [5], skin and soft tissue infection [5,13], brain abscess [11,14], traumatic wound infection [5], acute cholecystitis [15], suppurative mastitis [16], and abdominal wall abscess [5]. Previous cases were common in indwelling catheter-related blood

stream infection and primary bacteremia in immunocompromised hosts, followed by skin and soft tissue infection. Among these reports, only few mycetoma related reports were found. And most of the cases were improved with antibiotic combination treatment with/without surgery. However, in the current case, the patient's condition led to death despite antibiotic combination treatment and multiple procedures of incision and drainage because firstly antibiotic susceptibility test for *Gordonia* species was not available at our institution, secondly extensive surgical resection was not performed, and lastly *Gordonia* species are frequently misidentified as *Rhodococcus*, *Corynebacterium*, and *Nocardia* by conventional microbiological culture, which may lead to delay in accurate diagnosis [5,12]. Thus, mycetoma caused by *Gordonia terrae* would be consider as one of fatal pathogens causing skin and soft tissue infection.

As stated above, fatal infections by *Gordonia* species are rare disease in infected sore disease and in temperate climate areas. Because it is time consuming to identify *Gordonia terrae* in clinical settings, physicians would be consider a molecular sequencing methods for chronic tumorous granulation tissues and extensive surgical treatments for bulky mycetoma.

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### Conflict of Interest

No potential conflict of interest relevant to this article.

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