Open Forum Infectious Diseases
NOVEL ID CASES

Nocardia beijingensis Isolated From an Adrenal Abscess in a Diabetic Host

Melissa Pender,[1] Ninad Mehta,[2,3] Blake D. Hamilton,[4] and Sankar Swaminathan[1,5]

[1]Division of Infectious Diseases, Department of Medicine, University of Utah School of Medicine, Salt Lake City, Utah, USA, [2]Department of Pathology, University of Utah School of Medicine, Salt Lake City, Utah, USA, [3]Department of Pathology and Laboratory Medicine, University of Saskatchewan, Saskatoon, Saskatchewan, Canada, [4]Division of Urology, Department of Surgery, University of Utah School of Medicine, Salt Lake City, Utah, USA, and [5]Department of Medicine, George E. Wahlen Veterans Affairs Medical Center, Salt Lake City, Utah, USA

We describe the case of a 57-year-old man with poorly controlled type 2 diabetes mellitus who presented with 30 days of left-sided abdominal pain. He was found to have a left adrenal abscess and underwent adrenalectomy. Intraoperative cultures grew *Nocardia beijingensis*, which is an uncommonly identified *Nocardia* species rarely affecting immunocompetent patients. We review the published literature on cases of *N beijingensis* among immunocompetent patients. This is the first report summarizing the diagnosis and management of *N beijingensis* isolated from an adrenal abscess.

Keywords. adrenal abscess; diabetes; *Nocardia beijingensis*.

Nocardia species are aerobic actinomycetes found in the soil; these organisms are pyogenic bacteria with a predilection for abscess formation and extension into surrounding tissues [1–3]. To date, 54 of the 92 identified *Nocardia* spp have been documented to cause human disease [4], with infections occurring primarily among immunocompromised patients [1, 3]. Species are identified and differentiated using 16S ribosomal ribonucleic acid (rRNA) gene sequencing [5, 6].

Wang et al first isolated *Nocardia beijingensis* from a soil sample in China in 2001 [5]. *Nocardia beijingensis* has subsequently been identified as a rare cause of infection primarily affecting immunocompromised hosts [7–12]. Our case represents the first published report of *N beijingensis* isolated from an adrenal gland.

CASE REPORT

A 57-year-old man was admitted to the hospital for evaluation of progressive left upper quadrant abdominal pain over the past month. Additional symptoms included subjective fevers, diaphoresis, tachycardia, anorexia, nausea, a 6.8-kg (15-lb) weight loss, and several episodes of nonbloody, nonbilious vomiting. Prior to onset of abdominal pain, the patient was asymptomatic and physically active. His medical history included hypertension and diabetes mellitus type 2 with a hemoglobin A1c of 10%. He had no history of trauma or intra-abdominal procedures. He was a former smoker with a 25 pack-year smoking history and had no other relevant substance use history. The patient lived in a medium-sized town within a largely rural Wyoming community and had no international travel history. He had no animal exposures. He reported riding his motorcycle in the Rocky Mountain area and described associated dust inhalation.

At the time of initial evaluation, vital signs demonstrated a temperature of 96.6°C, heart rate of 123 beats per minute, blood pressure of 170/115 mm Hg, and normal oxygenation on room air. He described 9/10 left upper quadrant abdominal pain but lacked evidence of peritonitis, and his abdomen was not tender. An abdominal computed tomographic (CT) scan revealed a 9 × 4.2-cm heterogenous left adrenal mass (Figure 1). Laboratory evaluation demonstrated leukocytosis of 18 000 cells/μL. Normal levels of plasma free metanephrines ruled out pheochromocytoma. Chest radiographs showed a possible left-sided infiltrate with parapneumonic effusion, and cefuroxime was administered for 5 days to treat presumed community-acquired pneumonia.

The patient underwent robot-assisted left adrenalectomy on day 10 of admission with concern for malignant disease. Purulent drainage from the left adrenal gland was observed during surgery. Perinephric fat was sent for tissue culture. The patient remained clinically stable by postoperative day 2, and he was discharged home with a 5-day course of amoxicillin-clavulanic acid.

Gram stain of the tissue sent for culture showed 1+ polymorphonuclear leukocytes without organisms. On postoperative day 3, blood and chocolate agar plates grew *Nocardia* spp in

Received 09 May 2022; editorial decision 27 June 2022; accepted 30 June 2022; published online 4 July 2022

Correspondence: Sankar Swaminathan, MD, Division of Infectious Diseases, Department of Medicine, University of Utah School of Medicine, Salt Lake City, Utah, USA (sankar.swaminathan@hsc.utah.edu).

Open Forum Infectious Diseases®

https://doi.org/10.1093/ofid/ofac328

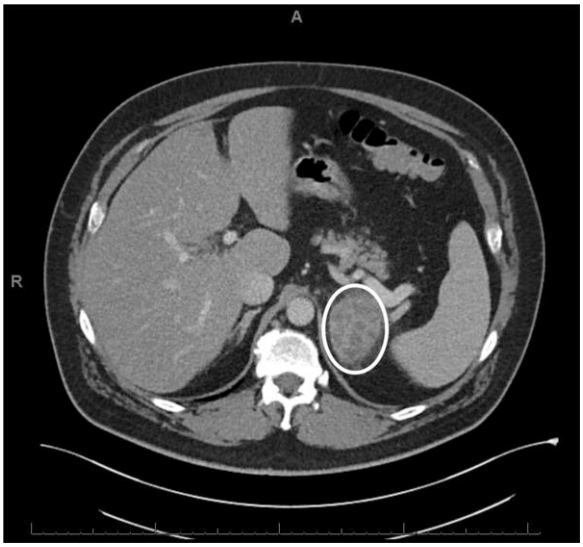

Figure 1. Abdominal computed tomographic scan highlighting a 9 × 4.2-cm heterogenous left adrenal mass (circled in white).

aerobic culture subsequently identified by 16s rRNA sequencing as *N beijingensis* (Figure 2). Sequencing was performed at a reference laboratory (ARUP Laboratories, Salt Lake City, Utah) and analyzed using the RipSeq database [13] with a 98.45% identification match to *N beijingensis*. The organism demonstrated susceptibility to trimethoprim-sulfamethoxazole (TMP-SMX), linezolid, carbapenems, tetracyclines, amoxicillin-clavulanic acid, and third-/fourth-generation cephalosporins with resistance only to fluoroquinolones. Pathology of the left adrenal mass was negative for neoplasm and demonstrated acute and chronic inflammation including prominent neutrophils and macrophages. Gram stain demonstrated filamentous micro-organisms on routine hematoxylin-eosin stain and modified acid-fast stain.

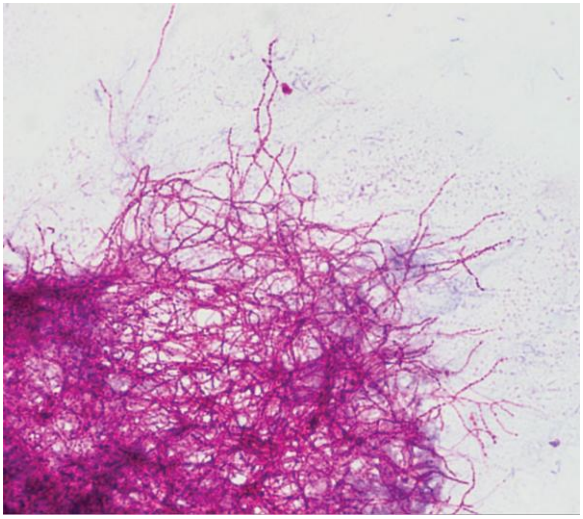

Figure 2. Partially acid-fast gram-positive bacilli grew from resected adrenal tissue on chocolate agar after 48 hours and is pictured at ×1000 magnification. The typical beaded appearance is evident at the periphery of the field.

After confirming organism susceptibilities, the patient began treatment with TMP-SMX 320 mg/1600 mg twice daily and linezolid 600 mg twice daily. Given the propensity of *Nocardia* to invade the brain and pulmonary systems [3], magnetic resonance imaging of his brain and a CT scan of his chest were performed. His brain imaging did not show intracranial pathology. Chest imaging demonstrated multiple small calcified pulmonary granulomas with several subcentimeter noncalcified nodules that were unchanged on repeat imaging 4 months later. He received linezolid for 3 months before this was discontinued due to thrombocytopenia, and he received TMP-SMX for a total of 7 months targeting visceral and suspected pulmonary nocardiosis. The patient recovered and remained without signs of recurrent disease at time of publication, >7 months from his last treatment dose.

DISCUSSION

Systemic nocardiosis remains a rare presentation among immunocompetent patients, particularly among those with no prior history of surgery, injection drug use, or trauma. Whereas pulmonary nocardiosis is associated with dust inhalation or aspiration [14], extrapulmonary disease results from traumatic inoculation or hematogenous spread from a primary site of infection [2]. Nocardiosis occurs primarily among immunocompromised hosts with approximately one-third of infections occurring among immunocompetent patients [1, 2]. Diabetes mellitus, while not a primary cause of immunodeficiency, can increase risk of infections through the effect of hyperglycemia on the immune system and is considered a secondary cause of immunodeficiency [15, 16]. While still considered relatively immunocompetent, our patient's poorly controlled diabetes may have increased his risk for systemic nocardiosis through impaired phagocytosis, chemotaxis, and/or decreased mitogen-induced lymphoproliferation [15].

We suspect this patient's *N beijingensis* infection began as primary pulmonary nocardiosis with subsequent hematogenous spread to the left adrenal gland. Chronic lung disease and cigarette smoking have been identified as risk factors for nocardiosis [17, 18]. The nodules on chest imaging likely represent sites of infection, although he remained free of pulmonary symptoms. His only identified exposure risk was dust inhalation while riding his motorcycle. Saubolle and Sussland [14] hypothesize that dry, windy conditions facilitate *Nocardia* aerosolization, leading to inhalation and pulmonary infection.

Nocardia beijingensis was first isolated from a soil sample in China in 2001 [5]. Human infections with this species have been identified as early as 2004 among immunocompromised

Table 1. Published Reports of *Nocardia beijingensis* Infections Among Immunocompetent Hosts

Study [Reference]	Sex, Age	Comorbidities or Potential Risk Factors	Symptoms	Infection Site	Treatment
Crozier et al, 2014 [12]	M, 48 y	Cotton farmer	Cough, fevers, night sweats, weight loss for 1 mo	Pulmonary: hilar mass	Ceftriaxone for 6 wk; TMP-SMX for 6 mo
Rigotti et al, 2015 [25]	M, 75 y	None identified	Lower back pain, abdominal pain, constipation for 10 wk	Paravertebral abscess L3–L5	Imipenem-cilastatin + amikacin for 3 wk; TMP-SMX for 3 mo
Abdel-Rahman et al, 2015 [19]	F, 55 y	Breast cancer treated surgically 9 y earlier	Fever, cough, hemoptysis for 6 mo	Pulmonary: endobronchial mass	Ceftriaxone for 4 wk; TMP-SMX for 3 mo
Gonzalez et al, 2016 [26]	F, 52 y	Contact lens use	Right eye pain, redness, photophobia for 8 wk	Scleritis	TMP-SMX + topical amikacin + topical polymyxin-trimethoprim for 10 mo
Solano-Varela et al, 2019 [22]	M, 58 y	None identified	Headache, right hemiparesis for 6 mo	CNS: left thalamic abscess	Meropenem + amikacin + TMP-SMX
Tanaka et al, 2020 [23]	M, 68 y	Petrochemical plant employee	Anorexia for 1 mo, acute headache and vomiting	CNS: miliary cerebrospinal abscesses	Meropenem + TMP-SMX for 4 wk; TMP-SMX for 7 mo
Roy et al, 2020 [24]	F, 57 y	None identified	Acute seizure	Pulmonary: left upper lobe mass, CNS: right temporal abscess	TMP-SMX for 12 mo
Raslan et al, 2021 [21]	M, 47 y	None identified	Lung mass identified on imaging	Pulmonary: lung mass	TMP-SMX + doxycycline (duration unspecified)
Present case	M, 57 y	Smoker, dust inhalation riding a motorcycle, type 2 diabetes	Abdominal pain, fevers, nausea, weight loss for 1 mo	Abdominal: Left adrenal abscess	Linezolid for 3 mo + TMP-SMX for 7 mo

Abbreviations: CNS, central nervous system; F, female; M, male; TMP-SMX, trimethoprim-sulfamethoxazole.

patients in Japan and Thailand [7]. There have been few documented cases in the Western Hemisphere [9]. In 2014, Crozier et al reported the first case affecting an immunocompetent patient in the United States [12]. To date, there are 8 previously published reports of *N beijingensis* affecting immunocompetent individuals. As in infections with other *Nocardia* spp, pulmonary and central nervous system (CNS) disease are most commonly reported. A literature review of *N beijingensis* infections in immunocompetent hosts yielded 3 documented pulmonary cases [12, 19–21], 2 cases of CNS abscesses [22, 23], 1 overlap case presenting with both pulmonary and CNS disease [24], 1 case of a subfascial paravertebral abscess [25], and 1 case of scleritis [26] (Table 1).

Identification of *Nocardia* spp remains an important step in treatment as antimicrobial susceptibility patterns vary between species. Most documented *N beijingensis* infections have responded to treatment with TMP-SMX, carbapenems, amikacin, and/or third-generation cephalosporins [24], although imipenem nonsusceptibility has been observed in 1 example [26]. As there are no available trials to guide the treatment of *Nocardia* infections, the recommended duration of therapy for systemic nocardiosis is typically 6–12 months based on expert opinion [3, 17, 27]. Given the surgical resection of our patient's abscess and subsequent clinical improvement, we elected to treat him for a 7-month course with TMP-SMX.

In conclusion, we describe a case of adrenal nocardiosis caused by *N beijingensis* masquerading as malignant disease This pathogen has not previously been described from an adrenal site.

Notes

Author contributions. S. S. supervised the patient's care and assisted in manuscript revision. N. M. identified the organism and assisted with manuscript revision. M. P. was the major contributor for writing this manuscript. All authors approved the final manuscript.

Financial support. Financial support was through the University of Utah School of Medicine.

Patient consent. Patient consent was obtained for publication.

Potential conflicts of interest. The authors: No reported conflicts of interest.

All authors have submitted the ICMJE Form for Disclosure of Potential Conflicts of Interest. Conflicts that the editors consider relevant to the content of the manuscript have been disclosed.

References

1. Beaman BL, Beaman L. *Nocardia* species: host-parasite relationships. Clin Microbiol Rev **1994**; 7:213–64. doi:10.1128/CMR.7.2.213
2. Brown-Elliott BA, Brown JM, Conville PS, Wallace RJ. Clinical and laboratory features of the *Nocardia* spp. based on current molecular taxonomy. Clin Microbiol Rev **2006**; 19:259–82. doi:10.1128/CMR.19.2.259-282.2006
3. Lerner PI. Nocardiosis. Clin Infect Dis **1996**; 22:891–905. doi:10.1093/clinids/22.6.891
4. Conville PS, Brown-Elliott BA, Smith T, Zelazny AM. The complexities of *Nocardia* taxonomy and identification. J Clin Microbiol **2018**; 56:e01419-17. doi:10.1128/JCM.01419-17
5. Wang L, Zhang Y, Lu Z, et al. *Nocardia beijingensis* sp. nov., a novel isolate from soil. Int J Syst Evol Micr **2001**; 51:1783–8. doi:10.1099/00207713-51-5-1783
6. Schlaberg R, Fisher MA, Hanson KE. Susceptibility profiles of *Nocardia* isolates based on current taxonomy. Antimicrob Agents Chemother **2014**; 58:795–800. doi:10.1128/AAC.01531-13
7. Kageyama A, Poonwan N., Yazawa K, Mikami Y, Nishimura K. *Nocardia beijingensis*, is a pathogenic bacterium to humans: the first infectious cases in Thailand

and Japan. Mycopathologia **2004**; 157:155–61. doi:10.1023/B:MYCO.0000020588.60081.37
8. Aragaki-Nakahodo A, Benzaquen S, Kirschner M. Coinfection by *Nocardia beijingensis* and *Nocardia arthritidis* in an immunocompromised patient diagnosed by endobronchial ultrasound guided transbronchial needle aspiration (EBUS-TBNA). Respir Med Case Rep **2014**; 12:22–23.
9. Bertran-Lopez J, Abbott A, Archibald LK, Benninger L, Lascano J, Kalyatanda G. Disseminated *Nocardia beijingensis* masquerading as pulmonary tuberculosis in a patient with human immunodeficiency virus/acquired immune deficiency syndrome. Open Forum Infect Dis **2020**; 7:ofaa186. doi:10.1093/ofid/ofaa186
10. Keenan JG, Mohapatra S. *Nocardia beijingensis* brain abscesses in an HIV-infected individual. IDCases **2017**; 9:65–9. doi:10.1016/j.idcr.2017.03.006
11. Martinaud C, Verdonk C, Bousquet A, et al Isolation of *Nocardia beijingensis* from a pulmonary abscess reveals human immunodeficiency virus infection. J Clin Microbiol **2011**; 49:2748–50. doi:10.1128/JCM.00613-11
12. Crozier JA, Andhavarapu S, Brumble LM, Sher T. First report of *Nocardia beijingensis* infection in an immunocompetent host in the United States. J Clin Microbiol **2014**; 52:2730–2. doi:10.1128/JCM.00588-14
13. Simmon KE, Kommedal Ø, Saebo Ø, Karlsen B, Petti CA. Simultaneous sequence analysis of the 16S rRNA and rpoB genes by use of RipSeq software to identify mycobacterium species. J Clin Microbiol **2010**; 48:3231–5. doi:10.1128/JCM.00362-10
14. Saubolle MA, Sussland D. Nocardiosis. J Clin Microbiol **2003**; 41:4497–4501. doi:10.1128/JCM.41.10.4497-4501.2003
15. Chinen J, Shearer WT. Secondary immunodeficiencies, including HIV infection. J Allergy Clin Immunol **2010**; 125(2 Suppl 2):S195–203.
16. Berbudi A, Rahmadika N, Tjahjadi AI, Ruslami R. Type 2 diabetes and its impact on the immune system. Curr Diabetes Rev **2020**; 16:442–9. doi:10.2174/1573399815666191024085838
17. Steinbrink J, Leavens J, Kauffman CA, Miceli MH. Manifestations and outcomes of *Nocardia* infections: comparison of immunocompromised and nonimmunocompromised adult patients. Medicine (Baltimore) **2018**; 97:e12436. doi:10.1097/MD.0000000000012436
18. Paige EK, Spelman D. Nocardiosis: 7-year experience at an Australian tertiary hospital. Intern Med J **2019**; 49:373–9. doi:10.1111/imj.14068
19. Abdel-Rahman N, Izhakian S, Wasser WG, Fruchter O, Kramer MR. Endobronchial enigma: a clinically rare presentation of *Nocardia beijingensis* in an immunocompetent patient. Case Rep Pulmonol **2015**; 2015:970548.
20. Abdel-Rahman N, Izhakian S, Wasser WG, Fruchter O, Kramer MR. Erratum to "Endobronchial enigma: a clinically rare presentation of *Nocardia beijingensis* in an immunocompetent patient." Case Rep Pulmonol **2016**; 2016:1950463.
21. Raslan R, Bailey P, Sastry S. *Nocardia beijingensis* lung mass in an immunocompetent adult. BMJ Case Rep **2021**; 14:e237454. doi:10.1136/bcr-2020-237454
22. Solano-Varela DM, Barrios-Vidales EM, Plaza DF, et al Immunocompetent patient with a brain abscess caused by *Nocardia beijingensis* in Latin America: a case report. Medicine **2019**; 98:e14879. doi:10.1097/MD.0000000000014879
23. Tanaka H, Kiko K, Watanabe Y, Yaguchi T, Oya S, Shiojiri T. Miliary cerebrospinal lesions caused by *Nocardia beijingensis* in an immunocompetent patient. IDCases **2020**; 20:e00737. doi:10.1016/j.idcr.2020.e00737
24. Roy M, Martial A, Ahmad S. Disseminated *Nocardia beijingensis* infection in an immunocompetent patient. Eur J Case Rep Intern Med **2020**; 7:001904. doi:10.12890/2020_001904
25. Rigotti S, Marocco S, Angheben A, Screpis D, Piovan G, Zorzi C. The first case of *Nocardia beijingensis* isolated infection to lumbar spine. J Neurosci Rural Pract **2015**; 6:462–3. doi:10.4103/0976-3147.158767
26. Gonzalez A, Jennings E, Vaziri S, Yachnis AT, Kubal A. Second report of a *Nocardia beijingensis* infection in the United States: nodular scleritis with in vitro imipenem resistance. Digit J Ophthalmol **2016**; 22:62–6. doi:10.5693/djo.02.2016.01.003
27. Wilson JW. Nocardiosis: updates and clinical overview. Mayo Clin Proc **2012**; 87:403–7. doi:10.1016/j.mayocp.2011.11.016