Microbes, Infection and Chemotherapy

CLINICAL CASES

https://doi.org/10.54034/mic.e1859

Burkholderia cepacia Vertebral Osteomyelitis Following Cesarean Section: Case Report and Review of the Literature

Authors: Suha Kalash[1], Zeina A. Kanafani[1]

Abstract

The Burkholderia cepacia complex (Bcc) initially emerged as an important opportunistic pathogen in patients with predisposing lung disease. Bcc infections outside the respiratory tract are less common, although their incidence has been increasing particularly in immunocompromised patients. We herein describe the case of a healthy young woman who started experiencing low back pain 2 weeks after an uncomplicated cesarean section. She was found to have vertebral osteomyelitis at T12-L1. CT guided biopsy was performed, and cultures grew Bcc. She was treated with 3 weeks of meropenem and 5 weeks of oral trimethoprim-sulfamethoxazole with good response. No surgical intervention was warranted. We also performed a review of the literature on other existing reports. To the best of our knowledge, this is the first case of Bcc vertebral osteomyelitis in an immunocompetent patient following an obstetric procedure.

Key word: burkholderia cepacia complex, vertebral osteomyelitis, spondylodiscitis, postpartum, cesarean section.

[1]Division of Infectious Diseases, Department of Internal Medicine, American University of Beirut Medical Center, Beirut, Lebanon.

Corresponding author:
Zeina A. Kanafani, MD, MS, CIC, FIDSA
Address: Associate Professor of Medicine. American University of Beirut Medical Center. Cairo Street, PO Box 11-0236/110. Riad El Solh 1107 2020, Beirut, Lebanon. Phone: +961-1-350000 ext 4747; Fax: +961-1-744489.
E-mail: zk10@aub.edu.lb

Submitted: april 18, 2023
Reviewed : may 24, 2023
Approved: june 17, 2023

How to cite: Kalash S, Kanafani ZA. Burkholderia cepacia Vertebral Osteomyelitis Following Cesarean Section: Case Report and Review of the Literature. Microbes Infect Chemother. 2023; 3: e1859

Introduction

The *Burkholderia cepacia* complex (BCC) comprises more than 20 different species. It was previously known as *Pseudomonas cepacia* and was first described in 1949 by Walter Burkholder of Cornell University (1). These Gram-negative rods are ubiquitous in the environment, especially in association with soil, water, and plants (2). In the hospital setting, they have been isolated from water taps, distilled water, water baths, nebulizers, dialysis setups, contaminated disinfectants, intravenous fluids, catheters, and thermometers (3). BCC initially emerged as an important opportunistic pathogen in patients with predisposing lung disease, specifically cystic fibrosis, leading to poor outcomes, including accelerated pulmonary decline, necrotizing pneumonia known as "cepacia syndrome," and a high mortality rate (4). BCC infections outside the respiratory tract are less common, although their incidence has been increasing, particularly in immunocompromised patients, those who have serious underlying diseases, have received intensive care, or have undergone invasive procedures (5).

Acute vertebral osteomyelitis is usually a monomicrobial infection, most commonly due to Staphylococcus aureus in more than 50% of cases. Other causative agents include other staphylococci, streptococci, enteric Gram-negative bacilli, and *Pseudomonas aeruginosa*. There have been limited reports of BCC vertebral osteomyelitis in the literature. Herein we report a case of acute vertebral osteomyelitis caused by *Burkholderia cepacia* in a healthy young woman following cesarean section and we review the literature on other existing reports.

Case report

A 22-year-old previously healthy Iraqi woman presented to the orthopedic surgery clinic at the American University of Beirut for evaluation of low back pain. The patient underwent an uncomplicated cesarean section two months before presentation under spinal anesthesia. Two weeks after surgery, she started experiencing low back pain accompanied by chills but no documented fever. She did not report any focal neurological symptoms or changes in her mobility. Her pregnancy had been uneventful without any infections, including urinary tract infections, and she had no immediate postoperative complications. She received no intramuscular or intravenous injections during or before her pregnancy. She denied illicit drug use or the use of immunosuppressive medications. The patient had undergone work-up for her back pain in Iraq, and she was told that she had a vertebral infection. She received ceftazidime, vancomycin, metronidazole, and fluconazole for 5-7 days,

then was switched to teicoplanin, clindamycin, and ceftriaxone, which she took for five days. She then decided to stop all antibiotics and pursue her treatment in Lebanon. Upon evaluation in the orthopedics clinic, a physical examination revealed mild tenderness to palpation of the lower spine. She had a well-healed cesarian section scar in the lower abdomen. The remainder of the examination, including the neurological examination, was non-contributory.

Laboratory results demonstrated a white blood cell count of 7,000 cells/mm^3 with a normal differential, a C-reactive protein (CRP) of 37 mg/L, and an erythrocyte sedimentation rate (ESR) of 14 mm/hr. Serological testing for Brucella was negative. She underwent magnetic resonance imaging (MRI) of the lumber spine with gadolinium which revealed vertebral osteomyelitis at T12-L1.

Since she was clinically stable with no fever or neurological abnormalities, and to optimize the microbiological yield, antibiotics were held for one week, then she underwent computed tomography (CT) guided biopsy of the spine at T12-L1. A tissue exam of the intervertebral disc biopsy showed fragments of fibrocartilage with multifocal acute inflammation consistent with the clinical diagnosis of acute osteomyelitis. The Gram stain was non-revealing. However, bacterial cultures grew *Burkholderia cepacia* from the thioglycollate broth. The organism was identified using Matrix Assisted Laser Desorption/Ionization–Time of Flight (MALDI–TOF) mass spectrometry. The susceptibilities were determined by the Kirby–Bauer method, which revealed susceptibility to ceftazidime, meropenem, minocycline, and trimethoprim-sulfamethoxazole. The patient was assessed in the infectious diseases clinic, and she was started on meropenem intravenously for three weeks. She traveled back to Iraq and was then switched to oral trimethoprim-sulfamethoxazole. Follow-up measurements of ESR and CRP indicated a favorable response to treatment. She completed eight weeks of total treatment with a good pain response. As of the writing of this manuscript, the patient had been off antibiotics for four weeks. She continued to complain of some low back pain and was advised to repeat MRI imaging to evaluate the therapeutic response.

Discussion

Osteomyelitis in the peri-postpartum period has been reported in the literature, including a case series of osteomyelitis of the symphysis pubis. Causative organisms include various organisms, including Staphylococcus aureus and *Pseudomonas aeruginosa* (6,7). Postpartum osteomyelitis involving the femoral head and tibia has also been described (8, 9). Septic sacroiliitis can occur during pregnancy and, if left untreated, can lead to osteomyelitis. In a retrospective study between 1995 and 2011, 5 out of 39 patients with septic sacroiliitis were postpartum, with infection occurring on average nine days after delivery (10). Nutcharoen et al. reported a 30-year-old female with septic sacroiliitis nine days after an uncomplicated C-section under spinal anesthesia. The initial source of infection was suspected to be a gluteal cleft dermal piercing that was forcibly removed prior to symptoms (11).

BCC vertebral osteomyelitis is not a commonly described entity in the literature, even less so in immunocompetent individuals. Pathogens are thought to reach the spine via one of three possible routes: direct inoculation after a trauma or vertebral surgery, hematogenous spread after skin, oral, urinary, digestive, or pulmonary infections, or contiguous spread from surrounding infected tissues (12).

Our patient developed osteomyelitis within two weeks postoperatively after the cesarian section in Iraq. We postulate that the infection was acquired in the hospital during surgery. Possibilities include contaminated intravenous fluids or urinary catheters, which could have led to transient bacteremia and seeding of the vertebral spine. Alternatively, direct inoculation during spinal anesthesia could have caused vertebral osteomyelitis.

The organism was identified using the MALDI–TOF, which has been shown to have 100% sensitivity in identifying BCC at the genus level. However, in one study, 23.1% of BCC isolates tested were not correctly identified at the species level (13). An alternative molecular diagnostic test would be a PCR-based assay with a sensitivity of 96% and specificity of 100% (14). On the other hand, phenotypic microbial identification methods often lack sensitivity and are rarely relied on for diagnosis (13).

In our patient, vertebral osteomyelitis was first recognized on MRI. Kulkarni et al. reported the use of bone scintigraphy along with SPECT-CT scan as initial imaging modalities to investigate postpartum low back pain associated with fever with a subsequent MRI. The authors stated that this modality would help establish a differential diagnosis, confirm the presence of polyarthritis, and help localize the initial pathology (15). MRI remains the imaging modality with the greatest sensitivity for diagnosing osteomyelitis; however, if not feasible, appropriate alternative tests are available (16).

Therapeutic options for BCC are limited due to high levels of intrinsic or acquired resistance of the organism to several antibiotics. Trimethoprim-sulfamethoxazole has been classically the drug of choice, barring any allergic reaction (17), and we elected to use it in this case after the patient completed a three-week course of intravenous meropenem due to its excellent oral bioavailability, good concentration in bone (18), and low cost.

Although classically described as an opportunistic pathogen of the respiratory tract in cystic fibrosis patients, BCC extrapulmonary infections, including vertebral osteomyelitis, have been increasingly reported in immunocompetent patients following operative procedures. Weinstein et al. reported a case of a 49-year-old female patient who presented three weeks after a rhinoplasty procedure in Iran with acute BCC cervical osteomyelitis causing neck pain and bilateral upper extremity numbness, for

which she underwent partial corpectomies at C5-C6 (19). The patient was treated with meropenem for six weeks with gradual resolution of her neck pain. It was thought that the patient was exposed to irrigation solution or nasal packing contaminated with BCC leading to subsequent transient bacteremia and then to vertebral osteomyelitis. Jaafar et al. also reported the case of a 43- year-old, previously healthy Syrian woman who was found to have BCC spondylodiscitis after cholecystectomy (12). She received a three-month course of ceftazidime and ciprofloxacin with the successful resolution of her infection. Another report by Subramanian et al. described an immunocompetent female in her mid-30s with BCC lumbar osteomyelitis that developed within a month of elective bariatric surgery in Mexico (20). The infection did not improve with cefepime; however, she was successfully treated with levofloxacin monotherapy. To further illustrate the possibility of acquiring BCC osteomyelitis postoperatively, the case series by Hammoud includes three out of four patients who presented 1-4 weeks following surgery, the procedures being laparoscopic cholecystectomy, laminectomy, and submucous resection of the nose (21). Three patients were treated with a 4-6 week course of meropenem, and one of them was treated with a four weeks course of ceftazidime. Table 1 summarizes the literature on vertebral osteomyelitis due to BCC highlighting the cases that presented postoperatively.

Table 1.
Summary of the published literature on Burkholderia cepacia vertebral osteomyelitis classified by the presence of recent surgery

Reference/ Year	N/Gender	Age (years)	Location of discitis	Possible risk factor or prior surgery	Therapy	Surgery
Postoperative infection						
Weinstein (19)/2007	1/F	49	Cervical	Rhinoplasty	Meropenem for 6 weeks	Partial corpectomies
Jaafar (12)/2017	1/F	43	Lumbosacral	Cholecystectomy	Ciprofloxacin and ceftazidime for 12 weeks	None
Hammoud (21)/2019	4/2 M, 2F	34-64	1 cervical, 2 lumbar, 1 lumbosacral	1 cholecystectomy, 1 submucous resection of the nose, 1 laminectomy, 1 history of laminectomy 2 years prior	1 ceftazidime for four weeks, 3 meropenem for 4-6 weeks	1 hemilaminectomy and removal of instrumentation
Subramanian (20)/2022	1/F	34	Lumbar	Bariatric surgery	Levofloxacin for six weeks (failed cefepime)	None
Present case/2022	1/F	22	Thoracolumbar	Cesarian section	Meropenem for 6 weeks, followed by TMP-SMX for 2 weeks	None
Non-operative infection						
Yang (3)/2008	1/F	73	Lumbosacral	History of partial laminectomy four years prior, recent fall, IM stimulation therapy	Levofloxacin for 6 weeks	Discectomy with fixation followed by corpectomy with bone graft
Rodriguez (22)/2012	1/M	20	Cervical	Cystic fibrosis	Temocillin and doxycycline for 4 months (failed meropenem)	Surgical drainage
Hsieh (23)/2013	1/F	71	Thoracic	None	Levofloxacin for 2 weeks (stopped upon patient's demise)	Laminectomy and corpectomies
Chang (24)/2017	2/NA	NA	NA	NA	NA	NA
Li (25)/2018	1/M	68	Cervical	Minor trauma, IV drug use	Ceftazidime for 6 weeks followed by indefinite oral minocycline	Corpectomy, fusion with allograft placement
Kwayess (26)/2022	8/4 M, 4 F	28-99	4 lumbar, 3 cervical, 1 at multiple levels	3 previous vertebral surgery	Ceftazidime, quinolones, carbapenems, TMP-SMX, for a mean of 23 days; 4 monotherapy, 3 dual therapy, 1 triple therapy	NA

N = number of patients; F = female; M = male; TMP-SMX = trimethoprim-sulfamethoxazole; IM = intramuscular; NA = not available; IV = intravenous

To the best of our knowledge, we herein present the first case of BCC vertebral osteomyelitis in an immuno competent patient following an obstetric procedure.

Burkholderia cepacia should be on the differential in hospital-acquired vertebral osteomyelitis, especially following surgical procedures, even if the procedure is not contiguous to the vertebral column. There is also a general gap in the guidelines for managing BCC infections and BCC vertebral osteomyelitis. Treatment guidelines are needed to lead the clinician in treating this multidrug-resistant pathogen.

Author contribution Statement

All authors participated in study conception, data collection, reviewed the results and approved the final version of the manuscript. All authors agreed to be responsible for all aspects of the work to ensure the accuracy and integrity of the published manuscript.

Ethics statement

The authors declare that the published work reflects an investigation and analysis carried out truthfully and completely. Consent was obtained from the patient for publication of the case.

Conflict of interest

The authors declare no conflict of interest.

Funding

None.

Availability of data

Available from the corresponding author upon request.

References

1. Burkholder W. Sour skin, a bacterial rot of onion bulbs. Phytopathology. 1950;40:115-8.
2. Ranjan R, Chowdhary P, Kamra A. Community Acquired Burkholderia cepacia Bacteraemia Presenting as MODS in an Immunocompetent Individual: An Unusual Case. J Clin Diagn Res. 2017;11(3):DD01-DD2.
3. Yang BH, Lee MS, Lee J-H, Lee H-J. Pyogenic Spondylitis in a Healthy Adult Caused by Burkholderia cepacia. Infect Chemother. 2008;40(4):233-6.
4. Frost F, Shaw M, Nazareth D. Antibiotic therapy for chronic infection with Burkholderia cepacia complex in people with cystic fibrosis. Cochrane Database Syst Rev. 2019;6(6): CD013079.
5. Tavares M, Kozak M, Balola A, Sa-Correia I. Burkholderia cepacia Complex Bacteria: a Feared Contamination Risk in Water-Based Pharmaceutical Products. Clin Microbiol Rev. 2020;33(3).
6. Cosma S, Borella F, Carosso A, Ingala A, Fassio F, Robba T, et al. Osteomyelitis of the pubic symphysis caused by methicillin-resistant Staphylococcus aureus after vaginal delivery: a case report and literature review. BMC Infect Dis. 2019;19(1):952.
7. Lawford AM, Scott K, Lust K. A case of massive vulvar oedema due to septic pubic symphysitis complicating pregnancy. Aust N Z J Obstet Gynaecol. 2010;50(6):576-7.
8. Lee KS, Kong S, Kim J, Kim T, Choi CB, Kim YS, et al. Osteomyelitis of Bilateral Femoral Heads After Childbirth: A Case Report. Ann Rehabil Med. 2015;39(3):498-503.
9. Lee R, How E. Postpartum haematogenous osteomyelitis of the tibia: A case report and review of literature. Internet J Orthop Surg. 2009;18(1).
10. Hermet M, Minichiello E, Flipo RM, Dubost JJ, Allanore Y, Ziza JM, et al. Infectious sacroiliitis: a retrospective, multicentre study of 39 adults. BMC Infect Dis. 2012;12:305.
11. Nutcharoen AA, Salih AA, Volio A, Skolaris A, Ayad S. Suspected Septic Sacroiliitis Following Recent Cesarean Section Under Spinal Anesthesia. Cureus. 2020;12(6): e8891.
12. Jaafar D, Rizkallah M, Atallah F, Bachour F, Barakat A, Maalouf G, et al. Lumbar Spondylodiscitis Caused by Burkholderia cepacia in a Previously Healthy Patient. Case Rep Orthop. 2017;2017:1396950.
13. Sfeir MM. Burkholderia cepacia complex infections: More complex than the bacterium name suggest. J Infect. 2018;77(3):166-70.
14. Whitby PW, Pope LC, Carter KB, LiPuma JJ, Stull TL. Species-specific PCR as a tool for the identification of Burkholderia gladioli. J Clin Microbiol. 2000;38(1):282-5.
15. Kulkarni P, Elangoven IM, Jaykanth A, Simon S. Incremental Value of Three-Phase Bone Scintigraphy and Single-Photon Emission Computed Tomography-Computed Tomography in a Case of Postpartum PUO in the Wake of The Antibiotic-Resistance Era. Indian J Nucl Med. 2021;36(1):62-5.
16. Simpfendorfer CS. Radiologic Approach to Musculoskeletal Infections. Infect Dis Clin North Am. 2017;31(2):299-324.
17. Avgeri SG, Matthaiou DK, Dimopoulos G, Grammatikos AP, Falagas ME. Therapeutic options for Burkholderia cepacia infections beyond co-trimoxazole: a systematic review of the clinical evidence. Int J Antimicrob Agents. 2009;33(5):394-404.
18. Thabit AK, Fatani DF, Bamakhrama MS, Barnawi OA, Basudan LO, Alhejaili SF. Antibiotic penetration into bone and joints: An updated review. Int J Infect Dis. 2019;81:128-36.
19. Weinstein L, Knowlton CA, Smith MA. Cervical osteomyelitis caused by Burkholderia cepacia after rhinoplasty. J Infect Dev Ctries. 2008;2(1):76-7.
20. Subramanian R, Fitzgibbons L. Burkholderia cepacia Complex Lumbar Spondylodiscitis: A Rare Nosocomial Infection. Case Rep Infect Dis. 2022;2022:4378442.
21. Hammoud M, Fares Y, Atoui R, Dabboucy B. Burkholderia cepacia as a cause of pyogenic spondylodiscitis in immunocompetent patients: a single-institution case series and literature review. J Spine Surg. 2019;5(3):372-7.
22. Rodriguez M, Nelson M, Kelly JE, Elward A, Morley SC. Successful Use of Temocillin as Salvage Therapy for Cervical Osteomyelitis Secondary to Multidrug-Resistant

Burkholderia cepacia. J Pediatric Infect Dis Soc. 2014;3(1):77-80.
23. Hsieh CT, Hsu SK, Chang CJ. Thoracic vertebral osteomyelitis caused by Burkholderia cepacia in an immunocompetent adult. Surg Infect (Larchmt). 2013;14(5):476-9.
24. Chang WS, Ho MW, Lin PC, Ho CM, Chou CH, Lu MC, et al. Clinical characteristics, treatments, and outcomes of hematogenous pyogenic vertebral osteomyelitis, 12-year experience from a tertiary hospital in central Taiwan. J Microbiol Immunol Infect. 2018;51(2):235-42.
25. Li SK, Messer WB. Burkholderia cepacia complex Cervical Osteomyelitis in an Intravenous Drug User. Case Rep Infect Dis. 2018;2018:7638639.
26. Kwayess R, Al Hariri HE, Hindy JR, Youssef N, Haddad SF, Kanj SS. Burkholderia cepacia Infections at Sites Other than the Respiratory Tract: A Large Case Series from a Tertiary Referral Hospital in Lebanon. J Epidemiol Glob Health. 2022;12(3):274-80.