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Case Report

Burkholderia cepacia cervical spondylodiscitis in a 39-year-old immunocompetent woman after a rhinoplasty: A case report and review of literature

Francisco Perona-Fajardo[1,2], Arturo Morales-Díaz[3], Oscar Malpartida-Tabuchi[4], Ricardo Manuel Salcedo[2]

[1]Research Centre of the Faculty of Health Sciences, Peruvian University of Applied Sciences, Departments of [2]Internal Medicine, [3]Neurosurgery, [4]Infectious Diseases, "SANNA El Golf" Clinic, Lima, Perú.

E-mail: *Francisco Perona-Fajardo - francisco.perona.f@gmail.com; Arturo Morales-Díaz - mordi1074@yahoo.es; Oscar Malpartida-Tabuchi - osc.malta@gmail.com; Ricardo Manuel Salcedo - risales1983@gmail.com

***Corresponding author:**
Francisco Perona-Fajardo, Research Centre of the Faculty of Health Sciences, Peruvian University of Applied Sciences, Lima, Perú.

francisco.perona.f@gmail.com

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ABSTRACT

Background: *Burkholderia cepacia* has been described as a rare etiology for spondylodiscitis (SD) after surgical procedures.

Case Description: We report a rare case of cervical SD caused by *B. cepacia* in a 39-year-old immunocompetent woman after a rhinoplasty treated with complex cervical surgery and prolonged antibiotic therapy. The follow-up visits showed no recurrence of symptoms.

Conclusion: Diagnostic and therapeutic of rare spinal infections, particularly those caused by atypical pathogens such as *B. cepacia,* are challenging. Identification and personalized therapy are crucial.

Keywords: *Burkholderia cepacia*, Cervical vertebrae, Spondylitis, Spondylodiscitis

INTRODUCTION

Burkholderia cepacia is a Gram-negative, aerobic, nonspore-forming motile bacilli, first described by William Burkholder in 1954 as the agent of onion rot. It belongs to a group of 24 distinct species now called *B. cepacia complex* (Bcc).[5,24] It was found to be a contaminant of respiratory mucus in cystic fibrosis and was responsible for cepacia syndrome, a lethal complication in those patients. It was also identified as a pathogen in immunocompromised patients with chronic granulomatous disease. Later, it was reported in immunocompetent patients, but its finding in infections other than pulmonary remains rare.[20] Although *B. cepacia* is one of the least associated with respiratory diseases of the whole Bcc, it has been described as the etiology for osteomyelitis, discitis, skin abscesses, or bacteremia.[16,25]

Spondylodiscitis (SD), an infection of an intervertebral disc and the adjacent bone, is etiologically classified as tuberculous SD or pyogenic SD (PSD). *Staphylococcus aureus* is the most prevalent in PSD, followed by *Escherichia coli* and other Gram-negative bacteria. The reported incidence of PSD is 2.2–5.8 cases/100,000 person-years and is far more frequent in males, especially those over 70 years old.[14] Few cases of SD due to *B. cepacia* have been reported.

We report a case about a 39-year-old female dentist with 2 months of severe neck pain after a rhinoplasty, with an unsuccessful response to analgesics. Subsequent magnetic resonance imaging (MRI) showed cervical SD of C5–C6 and a paravertebral abscess positive to *B. cepacia* after vertebral corpectomy. Specific antibiotic treatment consisted of ceftazidime, which resulted in clinical and radiological improvement.

This case highlights the diagnostic and therapeutic challenges associated with a rare spinal infection and emphasizes the importance of bacterial cultures and tailored treatment based on microbiological sensitivity.

CASE PRESENTATION

A 39-year-old dentist woman with no previous medical history or comorbidities presented to the clinic with a 2-month history of severe neck pain that began 2 days after a rhinoplasty. Initial emergency treatment included diclofenac and tramadol, followed by a consultation with a traumatologist who requested a cervical spine MRI, which was reported as normal. The pain was not well controlled, and she experienced paresthesias in the upper limbs, requiring further emergency care. Acupuncture provided some relief, but the symptoms persisted. She denied fever or diaphoresis. Physical examination at that point demonstrated cervical tenderness upon palpation, with marked limitation on range of motion, and Spurling's foraminal compression test was positive.

A follow-up MRI revealed SD with spondylotic changes in C5 and C6, associated paravertebral abscess measuring 58 × 9 mm extending from C3 to C7, and a probable epidural abscess at the same level with severe spinal canal stenosis [Figure 1a-c]. Laboratory tests showed hemoglobin 11.9 mg/dL, leukocytes 4300/mm^3, procalcitonin 0.020 ng/mL, C-reactive protein (CRP) 0.52 mg/L, erythrocyte sedimentation rate (ESR) 20 mm/h, positive interferon-gamma release assay tuberculosis (TB)-spot, normal serum proteinogram, negative blood cultures, and negative human immunodeficiency virus rapid test. She denied respiratory symptoms, and a chest X-ray showed no abnormalities.

We started empiric vancomycin and ceftriaxone for 10 days without relief. After that, she underwent a C5 and C6 corpectomy plus anterior fixation with plate and screws plus posterior cervical fixation to C4–C7 lateral masses with screws. Cultures taken during cervical surgery were positive for *B. cepacia*, and the antibiogram reported meropenem and ceftazidime sensitivity as susceptible, while cotrimoxazole and levofloxacin were intermediate. Due to its importance in the treatment, we ordered an ETEST® for levofloxacin, which confirmed intermediate sensitivity. Culture for fungus, bacilloscopy, and GeneXpert-Rifampin/*Mycobacterium tuberculosis*® resulted in negative. Finally, the pathology report showed nonspecific necrosis and degenerative changes with minimal inflammatory infiltration and no TB granulomas or neoplasm.

We decided to initiate ceftazidime 2 g 3 times a day (TID) for a total of 4 weeks, and radiological findings [Figure 1d] and pain improved successfully. Inflammatory markers showed mildly elevated values with progressive normalization after the initiation of antibiotic treatment. After discharge, oral cotrimoxazole 160/800 mg 2 times a day plus levofloxacin 750 mg once daily was prescribed for one more week. The follow-up visits showed no recurrence of symptoms.

DISCUSSION

We reviewed similar cases published on PubMed and Google Scholar. We found 14 case reports of SD due to *B. cepacia*, including this case. Only 4 were not related to previous surgery. Only 3 of these cases occurred after spinal surgery. Of the other 7 related to nonspinal surgery, three were related to nasal surgery (two after rhinoplasty and one after nasal submucosal resection), two cases were after laparoscopic cholecystectomy, one after cesarean section, and one after bariatric surgery. Three cases after nasal surgery occurred at the cervical spine. All of them reported mildly elevated CRP or ESR. Eighth of the 14 cases required spine surgery. The antibiotics frequently indicated after the *B. cepacia* isolation were oral ciprofloxacin, oral clindamycin, meropenem, or ceftazidime for 4–12 weeks [Table 1].[6,10,11,13,14,19,22,23,27,28] Kwayess *et al*. reported another 8 cases, but no complete information was available.[16]

Our case has the particularity to start with cervical pain a few days after a rhinoplasty; this surgical risk factor has been described only once by Weinstein *et al*., in association with a C5–C6 osteomyelitis and treated only with meropenem 1 g TID for 6 weeks.[27] Hammoud *et al*. also reported a case with a previous history of a submucosal resection of the nose 1 month before a C5–C6 discitis treated with the same antibiotic and without surgery.[10]

Bcc has inherent resistance to antiseptics and antibiotics commonly used in health care, could survive under nutrient-limited conditions, and even use important pollutants or certain antimicrobials as carbon sources, so it is a feared contaminant of in-hospital aqueous pharmaceuticals and personal care cosmetic products.[24] Furthermore, it has been linked to human disease and even to hospital outbreaks where it was found as a contaminant of intravenous fluids, distilled water, disinfectants, contaminated chlorhexidine mouthwash solution, moisturizing body milk, dialysis fluids, and machines, blood gas analyzers, ultrasound gels, contaminated anesthetic gel, nebulizers, catheters, thermometers, ventilator temperature sensors, and even tap water.[2,4,7,8,13,14,26]

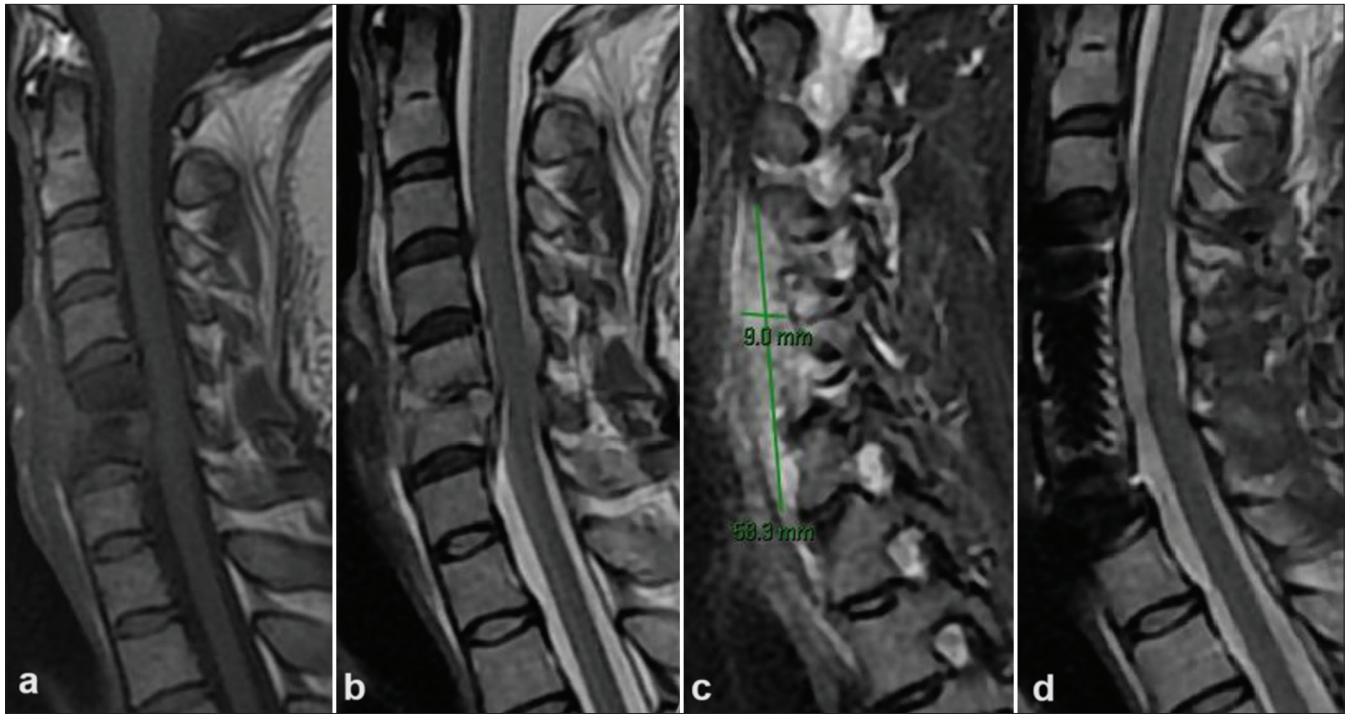

Figure 1: (a-c) Sagittal T1 and T2, and right oblique T2 cervical spine magnetic resonance imaging (MRI). There is decreased T1 and increased T2 signal at the C5-C6 vertebral bodies with a high T2 signal in the disc in between. The disc space is narrowed. The vertebral end plates are irregular and ill-defined. There is a paravertebral abscess extending from C3 to C7. All this corresponds with spondylodiscitis of C5-C6 with moderate spinal stenosis. The green lines mark a paravertebral abscess extending from C3 to C7 that measures 9.0 x 58.3 mm. (d) T2 MRI sequence showing post C5 and C6 corpectomy plus anterior fixation status with improved spinal stenosis, spondylodiscitis, and abscess.

Several mechanisms that explain this innate resistance have been described. For example, it has an outer membrane efflux pump that actively exports chloramphenicol, trimethoprim, and quinolones. Furthermore, it has porin proteins that do not allow the transport of antibiotics into the bacteria. Meanwhile, trimethoprim-resistant dihydrofolate reductase enzymes are found in the cytoplasm. High resistance to penicillins could be explained by the presence of periplasmic β-lactamases that degrade these antibiotics, making them less effective against penicillin-binding proteins.[18] In addition, Bcc lipopolysaccharides have been shown to create antibiotic resistance by undergoing structural changes that reduce the binding of antibiotics and cationic compounds due to lower levels of phosphate and 3-deoxy-D-manno-oct-2-ulosonic acid in its core oligosaccharide, and the presence of 4-amino-4-deoxyarabinose residues on lipid A, which neutralize the cell surface charge and hinder antibiotic binding.[12]

A study found that *S. aureus* biofilm forms on wounded nasal mucosa in mice after bacterial inoculation, particularly with nasal packing, indicating that reducing nasal wounds and packing duration may prevent biofilm formation.[26] Although positive cultures from nasal packing and blood after rhinoplasty have been observed, they usually do not have clinical significance; therefore, prophylactic antibiotics are not routinely recommended.[9,15,17] Thus, our patient could be colonized by nasal irrigation solution or nasal packing contaminated with *Burkholderia cepacia*. Then, it could have disseminated to the vertebral body after biofilm formation or after nasal packing removal.

Inflammatory markers such as CRP may show a better correlation with soft tissue improvement, while ESR with bones. These markers may help detect patients with a potential failure to treatment.[1] Diagnostic accuracy of MRI for PSD is 58% when it is made within 2 weeks after the beginning of symptoms and improves to 82% after that, and its role for follow-up and prognosis is limited. The patients may show no changes or worsening images despite clinical symptom improvements.[29] This may be the reason why the first MRI scan made 10 days after symptoms initiation was negative for PSD in our patient. Otherwise, 18-Fludeoxyglucose-positron emission tomography/computed tomography has a sensitivity of 96% and a specificity of 95%, no matter when symptoms are present, but this test was unavailable.[21] Follow-up images should be considered if the clinical status of the patient shows no improvement or worsens because new abscesses or medullary compression could appear.[3] We decided to order a new MRI before the discharge day to have a radiologic record of the improvement and to be able to compare in case of relapse during the follow-up visits.

Table 1: Cases of spondylodiscitis associated with *Burkholderia cepacia*.

Author	Patient age (years) and gender	Vertebral level	Relevant medical history before the onset of symptoms	Treatment
Smith *et al*., 1985[22]	59, male	C4–C5	IV heroin use, cervical osteomyelitis 10 years before due to *Serratia marcescens*. Neck trauma caused by a motor-vehicle accident 1 month before.	Trimethoprim-sulfamethoxazole 1,200 mg/240 mg QID and cefoperazone 2 g QID for 5 weeks
Yang *et al*., 2008[28]	73, female	L5–S1	L5 partial laminectomy 4 years prior. Back wound due to fall on icy road 3 weeks before.	Discectomy and posterolateral fixation of L5–S1 Levofloxacin 750 mg OD for 5 weeks
Weinstein *et al*., 2008[27]	49, female	C5–C6	Controlled hypertension and type 2 Diabetes mellitus. Rhinoplasty 3 weeks before.	Meropenem 1 g TID for 6 weeks
Hsieh *et al*., 2013[11]	71, female	T5–T6	Farmer	Laminectomy and transthoracic corpectomies of T5–T6 Levofloxacin 250 mg TID Note: 2 weeks after the start of treatment, death due to sepsis and respiratory failure
Jaafar *et al*., 2017[13]	43, female	L5–S1	Pain 1 day after a cholecystectomy in Syria	Ciprofloxacin 400 mg TID + Ceftazidime 2g TID for 12 weeks.
Hammoud *et al*., 2019[10]	60, female	L5–S1	Laparoscopic cholecystectomy 1 month before in Iraq	Laminectomy and debridement of L5–S1 Ceftazidime 2 g TID for 4 weeks.
	64, female	L4–L5	Posterior laminectomy plus fixation 2 months before	Laminectomy and screw removal of L4–L5. Meropenem 2 g TID for 6 weeks.
	34, male	C5–C6	Submucosal resection of the nose 1 month before in Lebanon	Meropenem 2 g TID, duration not described.
	51, male	L4–L5	Laminectomy 4 months before	Laminectomy and debridement of L4–L5. Meropenem 2g TID, duration not described.
Miryala *et al*., 2021[19]	35, male	L5–S1	L4–L5 TLIF + L5–S1 laminectomy 10 days before.	New TLIF L5–S1. Meropenem 1 g TID for 6 weeks+oral, clindamycin for 6 more weeks.
Subramanian and Fitzgibbons, 2022[23]	34, female	L2–L3	Obese, Graves' disease, partial bariatric gastrectomy performed in Mexico 4 days before.	Levofloxacin 750 mg for 6 weeks.
Kalash and Kanafani, 2023[14]	22, female	T12-L1	Cesarean section plus spinal anesthesia 2 months prior	Meropenem for 3 weeks + trimethoprim - sulfamethoxazole for 5 weeks. The dose is not described.
Candido *et al*., 2024[6]	68, female	L4–L5	Chronic kidney disease with hemodialysis and 4 months of lower back pain.	Meropenem 0.5 g q24h + levofloxacin 0.5 g q48h, duration not described.

(Contd...)

Table 1: *(Continued).*

Author	Patient age (years) and gender	Vertebral level	Relevant medical history before the onset of symptoms	Treatment
Perona-Fajardo *et al*., 2024 (Present Study)	39, female	C5–C6	Rhinoplasty 2 months before	C5 and C6 corpectomy plus anterior fixation with plate and screws plus posterior cervical fixation to C4–C7 lateral masses with screws. Ceftazidime 2 g TID for 4 weeks, then oral Trimethoprim-sulfamethoxazole 160/800 mg BID + levofloxacin 750 mg OD for 1 week.

Initial empiric treatment not considered in this table. TID: 3 times a day, QID: Four times a day, BID: 2 times a day, OD: Once daily, TLIF: Transforaminal lumbar interbody fusion

CONCLUSION

This case emphasizes the diagnostic and therapeutic challenges associated with rare spinal infections, particularly those caused by atypical pathogens such as *Burkholderia cepacia*. The successful outcome following tailored antibiotic treatment further highlights the significance of precise microbiological identification and personalized therapy in managing such infections. In addition, the scarcity of reported cases of *B. cepacia*-associated SD underlines the need for continued vigilance and further research into this rare clinical entity.

Ethical approval

Institutional Review Board approval is not required.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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