Infectious Diseases Diagnosis & Treatment
Cellier O, et al. Infect Dis Diag Treat 9: 272.
www.doi.org/ 10.29011/2577- 1515.100272
www.gavinpublishers.com

OPEN ACCESS

GAVIN PUBLISHERS

Case Report

Burkholderia cepacia Vertebral Osteomyelitis in an Immunocompetent Patient: A Case Report and Literature Review

Oriane Cellier[1*], Alexis Roditis[2], Hejer Herrabi[3], Frederique Bouchand[3], Karim Jaffal[3], Véronique Perronne[3], Christel Mamona-Kilu[3], Hanane Mehawej[3], Latifa Noussair[4], Aurélien Dinh[3, 5]

[1]Internal Medicine Resident, Paris-Est Créteil University, France

[2]Radiology Department, Raymond-Poincaré University Hospital, AP-HP, Paris Saclay University, Garches, France

[3]Infectious Disease Department, Raymond-Poincaré University Hospital, AP-HP, Paris Saclay University, Garches, France

[4]Microbiological Laboratory, Raymond-Poincaré University Hospital, AP-HP, Paris Saclay University, Garches, France

[5]Université Versailles saint Quentin Paris Saclay, France

***Corresponding author:** Oriane Cellier, Internal Medicine Resident, Paris-Est Créteil University, France.

Citation: Cellier O, Roditis A, Herrabi H, Bouchand F, Jaffal K, et al. (2025) *Burkholderia cepacia* Vertebral Osteomyelitis in an Immunocompetent Patient: A Case Report and Literature Review. Infect Dis Diag Treat 9: 272. DOI: 10.29011/2577-1515.100272.

Received Date: 26 March 2025; **Accepted Date:** 3 April 2025; **Published Date:** 7 April 2025

Abstract

Burkholderia cepacia is a Gram-negative, environmental, and saprophytic bacterium that can act as an opportunistic pathogen, most notably causing necrotizing pneumonia in immunocompromised patients, with granulomatous disease or cystic fibrosis.

We report a rare case of vertebral osteomyelitis caused by *B. cepacia* in a 45-year-old immunocompetent patient with a history of intravenous drug use and prior residence in Southeast Asia. This diagnosis was challenging due to the absence of typical risk factors such as immunosuppression, recent hospitalizations, or exposures. Remarkably, this case is one of the few described in association with Scheuermann's disease, expanding the differential diagnosis for such infections.

A literature review identified 38 cases of *B. cepacia* vertebral osteomyelitis, primarily affecting immunocompromised individuals around 50 years with intravenous drug use or hospital exposure. Treatment typically spans 10 weeks but carries a poor prognosis. This case emphasizes the need to consider *B. cepacia* in atypical presentations of bone infections.

Citation: Cellier O, Roditis A, Herrabi H, Bouchand F, Jaffal K, et al. (2025) Burkholderia cepacia Vertebral Osteomyelitis in an Immunocompetent Patient: A Case Report and Literature Review. Infect Dis Diag Treat 9: 272. DOI: 10.29011/2577-1515.100272.

Keywords: Vertebral osteomyelitis; Scheuermann disease; *Burkholderia cepacia;* Immunocompetent disease; Intravenous drug

Highlights

- Vertebral osteomyelitis caused by Burkholderia cepacia is a rare condition.
- It predominantly affects immunocompromised patients or those with specific risk factors.
- Key risk factors include prior exposure in Asia and intravenous drug use.
- The prognosis is generally poor.
- A 10-week course of antibiotic therapy appears to be effective.

Background

Burkholderia cepacia is a rare Gram-negative, oxidase-positive, non-fermenting bacillus that belongs to the *B. cepacia* complex (BCC), a group of closely related bacteria with high phenotypic diversity [1]. These bacteria are widely found in the environment, and healthcare settings. *B. cepacia* is endemic in tropical regions, especially Southeast Asia, and is transmitted via airborne droplets or direct contact.

Primarily an opportunistic pathogen, *B. cepacia* is best known for causing necrotizing pneumonia in immunocompromised patients. Its high morbidity and mortality rates are linked to its virulence factors and natural resistance to antibiotics.

Extrapulmonary infections are rarely reported and typically occur in immunocompromised individuals or healthcare-associated infections [2-6].

In our literature review, we identified 38 cases of vertebral osteomyelitis due *B. cepacia.*

Here, we report a case of *B. cepacia* vertebral osteomyelitis in an immunocompetent patient with Scheuermann's disease, a history of intravenous drug use (IDU), and prior exposure in Southeast Asia, along with a review of the literature on bone and joint infections caused by *B. cepacia*.

Case description

A 45-year-old man with a history of intravenous drug use (IDU) presented to the infectious disease ward with a prolonged fever of unknown origin. He had lived in Cambodia, five years earlier and had ceased intravenous drug use since then. For six months, he experienced intermittent fever with chills and excessive sweating but no other symptoms.

Physical examination revealed poor oral and dental health but no cardiac, pulmonary, or injection site abnormalities. Routine laboratory tests, including liver and kidney function, were normal, with no inflammatory response.

Comprehensive infectious disease testing included negative urine cultures, hepatitis B and HIV serology, Dengue and arbovirus tests. Hepatitis C serology was positive with active infection confirmed by PCR. QuantiFERON testing was negative. Both transthoracic and transesophageal echocardiograms revealed no valvular abnormalities, and CT imaging ruled out tuberculosis, neoplasms, and deep-seated infections but identified severe pulmonary emphysema.

A blood culture was positive later to *B. cepacia*. The isolate was susceptible to piperacillin-tazobactam, ceftazidime, aztreonam, and cefotaxime, but resistant to carbapenems, aminoglycosides, fluoroquinolones, tetracyclines, fosfomycin, and colistin. Based on his IDU history, prior Southeast Asia residence, and absence of alternative etiologies, he was treated with intravenous piperacillin-tazobactam (4g qid IV) for 15 days, resulting in rapid defervescence.

Two months later, he presented with recurrent fever and intermittent thoracic pain but no dyspnea or identifiable triggers. Examination was unremarkable, and he was afebrile. Laboratory tests, including C-reactive protein, blood counts, and liver/kidney function, were normal, while blood cultures remained negative. CT-scan revealed T6-T7 vertebral endplate erosions suggestive of erosive disc disease. MRI confirmed T6-T7 infectious spondylodiscitis, showing edema of the vertebral bodies of T6 and T7, centered on the intervertebral disc and erosions enhancement of the disc, and paravertebral soft tissue enhancement without epiduritis. The MRI showed also signs of Scheuermann's disease.

On vertebral biopsy, *B. cepacia*, was identified by mass spectrometry (Biotyper on a Microflex LT mass spectrometer, Bruker Daltonics, Bremen, Germany) and antimicrobial susceptibility tested by disk diffusion. Confirmation of the minimal inhibitory concentration (MIC) was performed by ellipsometry or broth microdilution (Etest, BioMérieux, Marcy l'Etoile, France or UMIC, Biocentric, Bandol, France) on available isolates and reinterpreted according to the EUCAST 2018 guidelines.

Histological analysis revealed mild inflammatory fibrosis with no organisms detected by Grocott staining, consistent with *B. cepacia* vertebral osteomyelitis secondary to bacteremia.

The patient received intravenous piperacillin-tazobactam (4 g every six hours) for six weeks. On Day 5, trimethoprim-sulfamethoxazole (1600 mg of sulfamethoxazole and 320 mg of trimethoprim, administered orally twice daily) was added to

improve bone penetration. The patient responded favorably, with no recurrence observed at 36-month follow-up.

Discussion

Vertebral osteomyelitis is an infection involving adjacent vertebrae and the intervertebral disc. It is rare, with a prevalence of approximately 40 cases per million annually, accounting for 4% of all bone infections. In most cases (60–80%), the infection spreads hematogenously, though direct inoculation following trauma or surgery, or local spread from nearby infections, is also possible.

Risk factors for vertebral osteomyelitis include older age, pre-existing spinal conditions, and immunosuppression, such as in diabetes, chronic kidney disease, intravenous drug use, HIV, or immunosuppressive therapy. Lumbar involvement is most common due to mechanical stress contributing to degenerative changes. Symptoms typically include vertebral pain with an inflammatory pattern (90–100% of cases). Fever is variable (15–75%) and less common presentations, such as neurological compression, urinary symptoms, or isolated fever, can occur [7].

Diagnosis relies on radiological and microbiological findings. Blood cultures are essential. MRI is the gold standard imaging modality, though CT and PET scans may assist in complex cases or contraindications. Identifying the causative bacteria is crucial; when blood cultures are negative, percutaneous biopsy techniques (CT- or fluoroscopy-guided) are safe and minimally invasive diagnostic options [8].

Scheuermann's Kyphosis (SK) is a common spinal deformity in adolescents, affecting around 5% of the population, with a slight male predominance [9]**.** It is caused by osteochondrosis and leads to rigid spinal deformity.

SK can mimic vertebral osteomyelitis, making diagnosis challenging. Radiologic findings like subchondral erosion and adjacent vertebral plate destruction suggest infection but can overlap with SK features, such as Schmorl's nodes. In our case, MRI findings like erosive changes and T2 hyperintense signals were more consistent with infection [10, 11]. A vertebral biopsy is essential to confirm infection. Bone scintigraphy may help distinguish SK from vertebral osteomyelitis, but more research is needed.

In vertebral osteomyelitis, *Staphylococcus aureus* is implicated in 50% of cases, and Gram-negative bacilli are more common in elderly patients and those with neoplasia. Vertebral osteomyelitis caused by *B. cepacia* is rare patients. Our literature review identified 38 cases, including infections after surgeries (e.g., rhinoplasty, discectomy, cholecystectomy), in intravenous drug users, and in nosocomial settings (table 1). *B. cepacia* is known for surviving in environments, such as distilled water, nebulizers, and disinfectants, contributing to outbreaks in hospitals. Since 1971, 111 hospital outbreaks worldwide have involved 2,390 patients and 240 deaths [1,12,13].

In our literature review (Table 1), the median age of patients was 50.05 years, and nearly half (14/34) presented with neurological involvement. Risk factors for *B. cepacia* vertebral osteomyelitis included intravenous drug use (IDU), immunosuppression (12/24), and hospital exposure. Blood cultures were positive in 8 out of 35 cases.

Study : Authors, Journal, year	N cases	Localisation	Neurological involvment	Age in years (median, IQR)	Immunocompromised	Risk Factors	Positive Blood culture	Antibiotic	Treatment duration (weeks)	Surgery	Cure
Luk Ks *et al.* Emerging infectious diseases 2022	7	Cervical (1) Thorax (1) Lombar (5)	5/7	62 (51-66)	6/7	IDU (7/7)	1/7	Amoxicillin/clavulanate (EMP) Trimethoprim/sulfamethoxazole (6/7); Cephalosporin (5/7); Fluoroquinolones (5/7); Carbapenem (2/7)	6 (1–12)	4/7	6/7
Hammoud M *et al.* Spine Surg. 2019	4	Cervical (1) Lombar (3)	0/4	52 (34-64)	0/4	Previous surgery (3/4)	0/4	Glycopeptide/Cephalosporin (EMP) Cephalosporin (1/4) Carbapenem (3/4)	5 (4-6)	0/4	4/4
Jaafar D *et al.* Case Rep Orthop. 2017	1	Lombar (1/1)	0/1	43	0/1	Previous surgery	0/1	Glycopeptide/Carbapenem (EMP) Cephalosporin/Fluoroquinolone	12	0/1	1/1
Subramanin R. *et al.* Case Rep Infect Dis. 2022	1	Lombar (1/1	0/1	34	0/1	Previous surgery	0/1	Glycopeptide/Cephalosporin (EMP) Cephalosporin Fluoroquinolone	12	0/1	1/1
Miryala R *et al.* Surg Neurol Int. 2021	1	Lombar (1/1)	1/1	35	0/1	Previous surgery	1/1	Carbapenem (EMP) Clindamycin	15	1/1	1/1
Li SK. *et al*. Case Rep Infect Dis, 2018	1	Cervical (1/1)	1/1	68	0/1	IDU	0/1	Glycopeptide + Piperacillin/ tazobactam (EMP) Cephalosporin	6	1/1	1/1
Smith MA. *et al.* J Clin Microb, 1985	1	Cervical (1/1)	UNK	59	1/1	IDU	0/1	Trimethoprim/sulfamethoxazole Cephalosporin	5	0/1	1/1
Weinstein *et al* J Infect Developing Countries 2008	1	Cervical (1/1)	0/1	49	1/1	Previous surgery	0/1	Carbapenem	6	0/1	1/1
Yang B.H *et al* Infect Chemother 2008	1	Lombar (1/1)	1/1	73	0/1	Previous surgery	0/1	Glycopeptide+ Cephalosporin+Aminoglycoside (EMP) Fluoroquinolone	6	1/1	1/1
Candido RCSR *et al.* J Brazilian Soc Trop Med 2024	1	Lombar (1/1	1/1	68	1/1	Dialysis catheter infection	1/1	Carbapenem + Fluoroquinolone	UNK	UNK	1/1

Hsieh CT *et al.* Surg Infect 2013	1	Thoracic (1/1)	1/1	73	0/1	No	1/1	Cephalosporin + Aminoglycoside (EPM) Cephalosporin Fluoroquinolone	UNK	1/1	0/1
Chang WS. *et al.* J Microbiol Immunol Infect 2018	2	UNK	UNK	UNK	UNK	UNK	UNK	UNK	UNK	UNK	UNK
Ghimire R. *et al.* European J Med Case Rep. 2019.	1	Lombar (1/1)	1/1	50	1/1	Epidural Injection Methylprednisolone	0/1	Carbapenem (EMP) Fluoroquinolone	10	1/1	1/1
Rodriguez M. *et al*. J Ped Infect Dis Society 2013	1	Cervical (1/1)	0/1	20	1/1	Colonization to *B.cepacia*	1/1	Carbapenem + Doxycycline + Piperacillin-tazobactam (EMP) Temocillin + Doxycycline	16	0/1	1/1
Chen Y. *et al.* World Neurosurgery 2024	1	Thoracic (1/1	1/1	49	0/1	Previous surgery	0/1	Cephalosporin + Sulbactam (EMP) + Tigecycline	24	1/1	1/1
Vithiya G. *et al.* Indian J Med Microbiol 2023	2	Lombar (2/2)	2/2	48 (59-37)	1/2	Dialisis (1/2) Previous Surgery (1/2)	0/2	Cephalosporin + Carbapenem (EMP) Fluroquinolone + Cotrimoxazole	12	2/2	2/2
Kwayess R. *et al.* J Epidemiol Glob Health 2022	10	UK	2/10	45,9 (28-99)	UK	Nosocomial (4/10)	2/10	Tetracycline + Rifampin (1/10) Cephalosporin+ Trimethoprim/sulfamethoxazole (1/10) Cephalosporin (1/10) Tigecycline + Fluoroquinolone (1/10) Fluoroquinolone (2/10) Trimethoprim/sulfamethoxazole (1/10) Cephalosporin+ Trimethoprim/sulfamethoxazole+ Fluoroquinolone (1/10) Cephalosporin+Tetracycline+ Fluoroquinolone (1/10) UK (2/10)	7,5 (2-30)	2/10	2/10 UNK
Our case	1	Thoracic (1/1)	0/1	45	0	IDU	1/1	Piperacillin-tazobactam+Trimethoprim/sulfamethoxazole	6	0/1	1/1

Total	38	Cervical (6/26) Thoracic (4/26) Lombar (15/26)	15/34	50.05	12/25	various	8/36	Multiple regimen	9,9	14/35	26/36
n: number of case; EMP: Empiric treatment; UNK: Unknown											

Table 1: Littérature review of vertebral osteomyelitis due to *Burkholderia cepacia,*

Among the cases of spondylodiscitis, the lumbar region was the most commonly affected, followed by the thoracic, cervical, and sacral spine. In our case, the thoracic spine was involved, likely due to kyphosis. In this literature review, the lumbar spine was most frequently affected, while cervical vertebral osteomyelitis (VO) was often associated with IDU in this region. It is speculative that the patient's poor dentition increased the risk of *Burkholderia* infection, as one study identified the presence of *Burkholderia* in the apical portion of teeth during microbiologic characterization [14].

Surgical intervention was performed in 13/38 cases, and the cure rate was 24/38, despite a median treatment duration of 10 weeks. Our case highlights an infection in a patient with no risk factors other than a history of drug use.

Diagnosing *B. cepacia* can be difficult, as it is a non-fermenting Gram-negative bacillus (NFGNB), and many labs struggle with precise identification. Among NFGNBs, *B. cepacia* complex (Bcc) ranks fourth in clinical significance, following *Pseudomonas aeruginosa, Acinetobacter baumannii*, and *Stenotrophomonas maltophilia*. Accurate identification is critical due to varying resistance profiles. While selective media like *B. cepacia* agar are used, molecular methods such as MALDI-TOF and PCR offer better accuracy.

Treatment of *B. cepacia* vertebral osteomyelitis generally involves a six-week course of antibiotics guided by susceptibility testing due to its high resistance potential [15-17]. *B. cepacia* is intrinsically resistant to carboxypenicillins, aminoglycosides, first- and second-generation cephalosporins, and polymyxins. Effective antibiotics include semisynthetic penicillins (e.g., piperacillin), carbapenems (e.g., meropenem), third-generation cephalosporins (e.g., ceftazidime), fluoroquinolones, tetracyclines, chloramphenicol, and trimethoprim-sulfamethoxazole. Combination therapy, such as trimethoprim-sulfamethoxazole with a β-lactam or fluoroquinolone, is often preferred for severe infections, and triple regimens including meropenem have shown promise [6]. In our patient 5 weeks with betalactam and trimethoprim-sulfamethoxazole were sufficient.

Newer treatments, such as β-lactam–β-lactamase inhibitors (e.g., ceftazidime-avibactam) and experimental agents like cefiderocol, are under investigation but may not overcome resistance mechanisms like efflux pumps [18-20]. Surgical intervention is required in about 9% of cases, especially with neurological deficits, spinal instability, or abscesses. Among reported *B. cepacia* vertebral osteomyelitis cases, only 8 required surgery, with just one documented relapse. While pyogenic vertebral osteomyelitis has a 7% mortality rate within one year, long-term outcomes specific to immunocompetent patients with *B. cepacia* infections remain unclear.

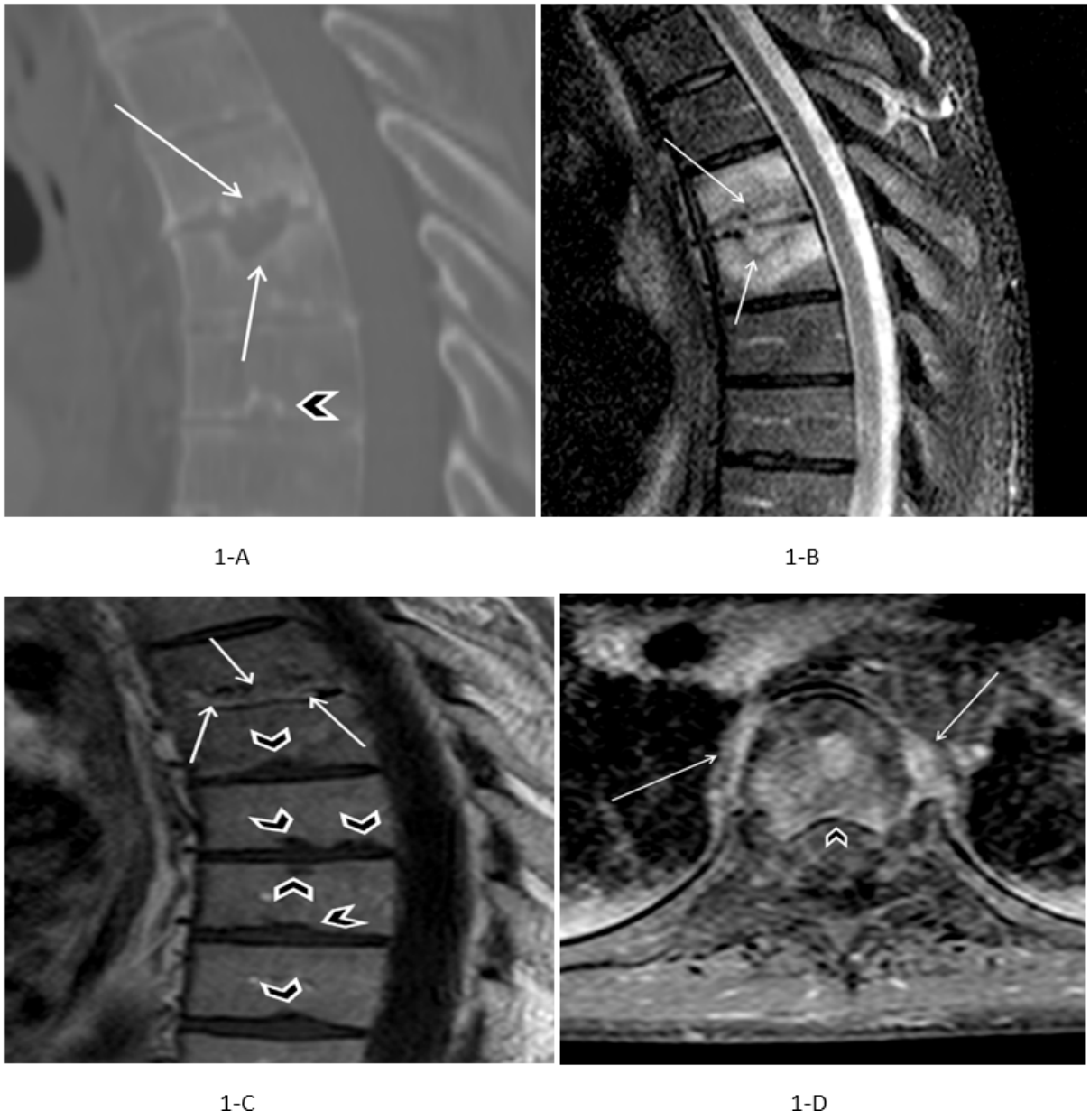

Figure 1: Spine CT and MRI.

Figure 1-A: CT medial sagittal reconstruction: fine erosions of the inferior plateau of T6 and large erosion of the superior plateau of T7 (arrows). The arrowhead shows an irregularity of the inferior plateau of T8 in connection with Scheuermann's disease. **Figure 1-B:** Sagittal section T2-weighted STIR MRI: Extensive edema of the vertebral bodies of T6 and T7, centered on the intervertebral disc and erosions (arrows). **Figure 1-C:** T1-weighted FAT-SAT MRI axial section after contrast injection: paravertebral soft tissue enhancement (arrows). Note the absence of associated epiduritis (arrowhead). **Figure 1-D**: T1 sagittal section after contrast injection: note the diffuse enhancement of the intervertebral disc (arrows). Signs of Scheuermann's disease on the underlying levels (arrowheads).

Conclusion

This case and literature review highlight the potential for *B. cepacia* to cause extrapulmonary infections, including vertebral osteomyelitis, even in immunocompetent individuals and years after initial exposure. Clinicians should remain alert to *B. cepacia* as a possible cause of bone infections, and underlying degenerative conditions.

References

1. Sfeir MM (2018) Burkholderia cepacia complex infections: More complex than the bacterium name suggest. J Infect 77:166-170.
2. Beca FA, Sengillo JD, Robles-Holmes HK, Iyer PG, Miller D, et al. (2023) Endophthalmitis caused by Burkholderia cepacia complex (BCC): Clinical characteristics, antibiotic susceptibilities, and treatment outcomes. Res Sq rs.3.rs-3181158.
3. Gonzalez JM, Lowenhaar G, Ramgopal M, Chalasani P (2024) Burkholderia cepacia: A Rare Source of Endocarditis. R I Med J (2013). 107:23-25.
4. Nnaoma C, Chika-Nwosuh O, Sossou C (2019) A Rare Culprit of Infective Endocarditis in an IV Drug User: Burkholderia cepacia. Case Rep Med 2019:6403943.
5. Nittala R, Behera MK, Panigrahy R, Narayan J, Mishra D, et al. (2023) Burkholderia cepacia causing liver and splenic abscess: Two case reports. Indian J Pathol Microbiol 66:171-173.
6. Siddiqui T, Sahu C, Patel SS, Ghoshal U (2022) Clinical and Microbiological Profile of Patients with Bloodstream Infections Caused by Burkholderia cepacia Complex. J Lab Physicians 14:312-316.
7. Lemaignen A, Bernard L, Marmor S, Ferry T, Grammatico-Guillon L, et al. (2021) Epidemiology of complex bone and joint infections in France using a national registry: The CRIOAc network. J Infect 82:199-206.
8. Chang CY, Pelzl C, Jesse MK, Habibollahi S, Habib U, et al. (2023) Image-Guided Biopsy in Acute Diskitis-Osteomyelitis: A Systematic Review and Meta-Analysis. AJR Am J Roentgenol 220:499-511.
9. Singh M, Bansal M, Singh A, Bharwani N, Bhati M (2023) Atypical Lumbar Scheuermann's Disease: A Rare Entity and Literature Review. J Orthop Case Rep 13:48-50.
10. Bezalel T, Carmeli E, Kalichman L (2019) Scheuermann's Disease: Radiographic Pathomorphology and Association with Clinical Features. Asian Spine J 13:86-95.
11. Ryu S, Kim YJ, Lee S, Ryu J, Park S, et al. (2021) [Pathophysiology and MRI Findings of Infectious Spondylitis and the Differential Diagnosis]. Taehan Yongsang Uihakhoe Chi 82:1413-1440.
12. Häfliger E, Atkinson A, Marschall J (2020) Systematic review of healthcare-associated Burkholderia cepacia complex outbreaks: presentation, causes and outbreak control. Infect Prev Pract 2:100082.
13. Pinilla MI, Cobos-Trigueros N, Soriano Á, Martínez JA, Zboromyrska Y, et al. (2011) [Burkholderia cepacia bacteremia: a prospective analysis of 33 episodes]. Rev Esp Quimioter 24:209-212.
14. Chugal N, Wang JK, Wang R, He X, Kang M, et al. (2011) Molecular characterization of the microbial flora residing at the apical portion of infected root canals of human teeth. J Endod 37:1359-1364.
15. Devanga Ragupathi NK, Veeraraghavan B (2019) Accurate identification and epidemiological characterization of Burkholderia cepacia complex: an update. Ann Clin Microbiol Antimicrob 18:7.
16. Bernard L, Dinh A, Ghout I, Simo D, Zeller V, et al. (2015) Antibiotic treatment for 6 weeks versus 12 weeks in patients with pyogenic vertebral osteomyelitis: an open-label, non-inferiority, randomised, controlled trial. Lancet 385: 875-882.
17. Rhodes KA, Schweizer HP (2016) Antibiotic Resistance in Burkholderia Species. Drug Resist Updat 28:82-90.
18. Avgeri SG, Matthaiou DK, Dimopoulos G, Grammatikos AP, Falagas ME (2009) Therapeutic options for Burkholderia cepacia infections beyond co-trimoxazole: a systematic review of the clinical evidence. Int J Antimicrob Agents 33:394-404.
19. Takemura M, Nakamura R, Ota M, Nakai R, Sahm DF, et al. (2023) In vitro and in vivo activity of cefiderocol against Achromobacter spp. and Burkholderia cepacia complex, including carbapenem-non-susceptible isolates. Antimicrob Agents Chemother 67: e0034623.
20. Narayanaswamy VP, Giatpaiboon S, Baker SM, Wiesmann WP, LiPuma JJ, et al. (2017) Novel glycopolymer sensitizes Burkholderia cepacia complex isolates from cystic fibrosis patients to tobramycin and meropenem. PLoS One 12: e0179776.