Community-acquired *Burkholderia cepacia* pneumonia: a report of two immunocompetent patients

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ÖZET

Toplum kökenli Burkholderia cepacia pnömonisi: Bağışıklığı sağlam iki olgu sunumu


Anahtar Kelimeler: Burkholderia cepacia, bronş lavajı, kinolon, pnömoni, toplum kökenli infeksyonlar.

SUMMARY

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Burkholderia cepacia is a gram-negative bacilli leading to pneumonia with poor prognosis and usually seen in patients with immunosupression or with structural lung diseases. This report is about two patients with no underlying disease diagnosed as B. cepacia pneumonia mimicking malignancy and tuberculosis. Bronchoscopy was applied on both patients since no response to treatment with wide spectrum antibiotics and negative sputum smears for acid-fast bacilli. B. cepacia was isolated from bronchial lavage culture. Antibiogram revealed sensitivity to quinolones in both cases. Radiological and clinical complete remission was seen in patients by quinolones. The current cases showed that community-acquired Burkholderia pneumonia is possible in healthy patients. Bronchial washing is important in diagnosis.

**Key Words:** Burkholderia cepacia, bronchial lavage, quinolone, pneumonia, community-acquired infections.

**CASE REPORT**

Case 1, a 60-year-old male was evaluated with malaise and productive cough lasting for 10 days. Case 2, 66-year-old male was admitted because of a 5-days history of cough and expectoration of purulent sputum. The patients had a smoking history of 30 and 25 packs-year, respectively. Both had previously been healthy and reported no history of hospitalization for the last year. They declared no family history of any pulmonary illness or chronic disease like diabetes mellitus. Case 1 has been worked as a servant in an office and got retired 6 years ago. Case 2 was formerly a farmer but has not been worked for approximately ten years.

Their physical examinations were normal, as were the oxygen saturations. Laboratory studies showed on admission a total WBC count of 13,100/mm³ and 7,200 mm³, an erythrocyte sedimentation rate of 96 mm/hour and 90 mm/hour, a C-reactive protein of 174 mg/dL and 8.85 mg/dL for Case 1 and Case 2, respectively. Both had normal levels of glucose and electrolytes, and normal renal and hepatic function tests. Case 1 had an irregular cavitary lesion, about 3 cm in diameter, with air-fluid level in the upper zone of right hemithorax on chest X-ray. Chest X-ray of case 2 revealed heterogeneous density on left lower zone. Both had negative sputum smears for acid-fast bacilli (AFB). Gram-positive diplococci were seen on sputum Gram-stain of case 1 and abundance of PNL and gram-positive cocci of case 2. No growth was detected on both sputum cultures. Case 1 was started on ceftriaxone 2 g/day intravenous (IV) and clarithromycin 1 g/day PO, case 2 was started on ampicillin/sulbactam 4 g/day IV and clarithromycin 1 g/day PO. A thorax computed tomography (CT) was performed when no improvement was achieved with empiric antibiotherapy. A thick-walled cavity with spicular contours was observed on chest X-ray. Chest X-ray of case 2 revealed heterogeneous density on left lower zone. Both had negative sputum smears for acid-fast bacilli (AFB). Gram-positive diplococci were seen on sputum Gram-stain of case 1 and abundance of PNL and gram-positive cocci of case 2. No growth was detected on both sputum cultures. Case 1 was started on ceftriaxone 2 g/day intravenous (IV) and clarithromycin 1 g/day PO, case 2 was started on ampicillin/sulbactam 4 g/day IV and clarithromycin 1 g/day PO. A thorax computed tomography (CT) was performed when no improvement was achieved with empiric antibiotherapy. A thick-walled cavity with spicular contours was observed on thorax CT of case 1 (Figure 1). Case 2 had a triangular consolidation in the lingular segment of the left upper lobe on CT (Figure 2). Fiberoptic bronchoscopy was performed to case 1 with a suspicion of bronchial carcinoma and to case 2 with a suspicion of obstructive pneumonia. Both had no endobronchial pathology and no malignant cells were detected on cytological examination of their bronchial lavage and post-bronchoscopic sputums. Lavage and post-bronchoscopic sputum smear results were negative for AFB. B. cepacia grew on bronchial lavage cultures of the cases. Antibiogram was reported to be resistant to amoxicillin/clavulanic acid, ticarcillin,
ceftriaxone, cefuroxime, ureidopen, cefoxitin, and co-
trimoxazole while sensitive to ceftazidime, imipenem,
amikacin, gentamicin, ciprofloxacin, piperacillin/tazo-
bactam, meropenem and less sensitive to cefepime in
Case 1 and it was reported to be resistant to gentami-
cin, imipenem, amikacin, aztreonam, cefepime, and
piperacillin whereas sensitive to ceftazidime, ciproflo-
xicin, and levofloxacin in Case 2. Case 1 received cip-
rofloxacin 750 mg PO bid and case 2 levofloxacin 500
mg/day IV significant radiological improvement was
observed (Figure 3). Final laboratory findings were;
WBC count 7200/mm³ and 6040 mm ³, erythrocyte
sedimentation rate 12 mm/hour and 34 mm/hour, and
C-reactive protein 2.6 mg/dL and 0.4 mg/dL for Case
1 and Case 2, respectively. All sputum and bronchial
lavage cultures of the patients remained negative for
Mycobacterium tuberculosis. Both patients are cur-
rently under follow-up and have no complaint.

**DISCUSSION**

*Burkholderia cepacia* complex is a family of catalase-producing, non-lactose-fermenting gram-negative bacteria comprising 9 sub-groups. They cause pneumonia in immuno-
compromised persons, particularly in those with cystic
fibrosis and chronic granulomatous disease. It is rarely
encountered in individuals with a normal immune sys-
tem and without a structural lung disease. To date, com-
munity-acquired pneumonia caused by *B. cepacia* has
been reported in a small number of individuals without
an underlying disease. Firstly Waterer et al. described a
community-acquired pneumonia caused by *B. cepacia*
in healthy adult (10). In present study, absence of hos-
pitalization eliminates the possibility of nosocomial in-
fection in both cases. Microbiology laboratory records
were examined in terms of probability of laboratory
contamination and it was seen that *B. cepacia* was not
produced on any material before. Since case 1 was pre-
sented with a cavitary lesion in the right upper zone, tu-
berculosis was considered as diagnosis in the first pla-
ce. Due to negative sputum smears for AFB, thick ca-
vity wall along with his smoking history and advanced
age, malignity was also strongly considered. The trian-
gular consolidation of lingular segment and a suspicious
central mass appearance as well as the patient’s smo-
kling history and advanced age of case 2 suggested al-
so malignity. The noteworthy aspects of both cases in-
clude the fact that they are not immunocompromised pa-
tients, absence of chronic lung disease as well as no
chronic lung disease patients in their close circles, and
lack of the possibility of nosocomial infection due to ab-
sence of hospitalization.

Bacterial culture of bronchoscopic lavage has been
the method of diagnosis in both cases. Demir et al. ha-
ve reported a hemodialysis patient with cavitary lesion
in the right lung upper zone, similar to case 1 (11). *B.
cepacia* was isolated from his bronchial lavage culture
too and he recovered also after ciprofloxacin treat-
ment. To note, he was a diabetic hemodialysis patient
and had a central catheter. Cultivation of bronchial la-
vage seems to be important in the diagnosis of *B. ce-
 pacia* pneumonia.

Because of its inherent resistance to many antibiotics
the organism can be difficult to treat. Both strains of the
current cases were sensitive to quinolones and the an-
tibiotherapy resulted with complete resolution. Waterer
and Demir also treated successfully their patients with
quinolone antibiotics (10,11). The unusual susceptibi-
ity to antibiotics suggested that it is not transmitted
from someone with chronic lung disease. The anti-tuberculous effects of quinolones, could naturally raise a suspicion of possibility of tuberculosis. However, cultures of all specimens remained negative for *M. tuberculosis*.

It has been shown that the organism have remarkable potential as an agent for both biodegradation and biocontrol, thus it is being considered as a plant-growth-promoting rhizobacterium (9). Used as a pesticide and biofertilizer in agriculture, that *B. cepacia* can be transmitted to humans via contaminated products is generally accepted. Its development in out-of-hospital settings and immunocompetent cases may possibly be attributed to environmental exposure. But we were not able to confirm the use for this purpose in our country.

These two cases have demonstrated that *B. cepacia*, which was previously considered as merely an opportunistic infection factor, may be encountered in individuals that are not in the risk group. Could this also be a sign of we would be likely to encounter this difficult and variable organism as a community-acquired pneumonia agent more frequently in the future?

**CONFLICT of INTEREST**

None declared.

**REFERENCES**


Figure 3. Computed tomography scans show complete improvement in two patients.