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

Burkholderia cepacia immünsüpresif ve yapısal akciğer hastalığı olan hastalarda kötü prognozlu pnömoniye neden olan gram-negatif bir basil ailesidir. Bu yazıda, altta yatan hastalığı olmayan, bağışıklığı sağlam olan iki hastada tüberküloz ve malignite ile karışan B. cepacia pnömonisi sunulmuştur. Her iki hastada da geniş spektrumlu antibiyotiklere yanıt alınamaması balgam aside dirençli basil yaymalarının negatif olması nedeniyle bronkoskopi uygulandı. Alınan bronşiyal lavaj örneklerinde B. cepacia üredi. Antibiyogramda her iki hastada da kinolonlara duyarlılık saptandı. Kinolon tedavisi ile her iki hastada da klinik ve radyolojik tam düzelme saptandı. Bu iki hasta sağlıklı bireylerde de Burkholderia pnömonisi görülebileceğini ve tanıda bronş lavajının önemini gösterdi.

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

SUMMARY

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

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Burkholderia cepacia is a gram-negative bacilli leading to pneumonia with poor prognosis and usually seen in patients with immunsupression 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 bacili. 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.*

Burkholderia (previously known as *Pseudomonas*) *cepacia* was first described in 1949 by Walter Burkholder, as the phytopathogen responsible for a bacterial rot of onions (1). It was first reported as a human pathogen causing endocarditis in the 1950s (2). *B. cepacia* is quite widespread in the environment and resistant to many antibiotics and antiseptic solutions (3-5). These agents are factors of potentially fatal pneumonia in patients with suppressed immunity, especially cystic fibrosis or chronic granulomatous disease (6). *B. cepacia* is also reported to cause epidemics of nosocomial respiratory infections through contamination of medical devices (7,8).

It has been shown that the organism have antifungal and degradative properties which have created interest in its potential use as a biological control agent to improve crop yields and its use for the bioremediation of contaminated soils (9). In contrast to its potential agricultural benefits, *B. cepacia* has also emerged as a multiresistant opportunist human pathogen, leading to concern about the relationship between environmental and clinical isolates and the potential hazards of releasing *B. cepacia* as a biological control agent (9). This report is about two patients with no underlying disease diagnosed as community-acquired *B. cepacia* pneumonia mimicking malignancy and tuberculosis, which resolved completely with quinolone treatment.

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/mm^3 and 7.200 mm^3, 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 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,

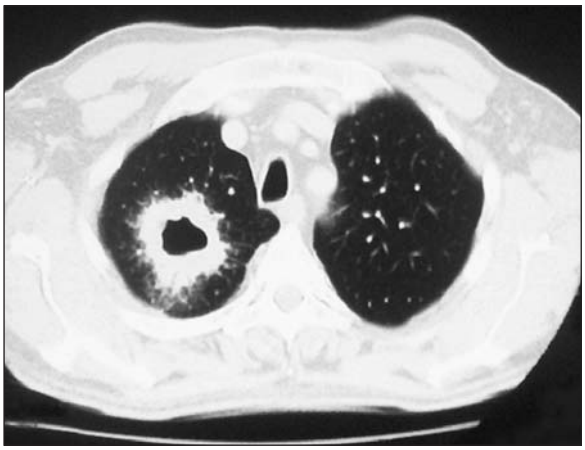

Figure 1. A thick-walled cavity with spicular contours was observed in the right upper lobe on thorax computed tomography.

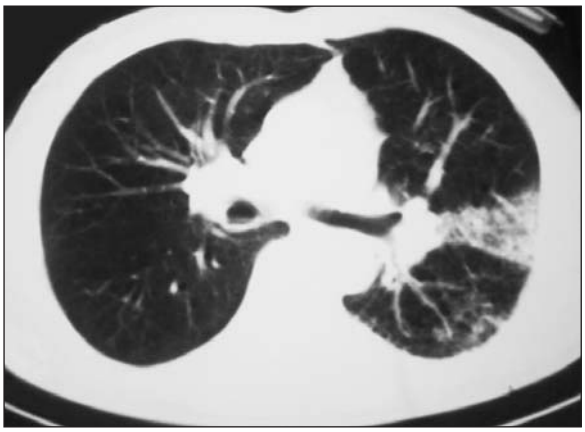

Figure 2. Triangular consolidation on left upper lobe limited by fissure and enlarged pulmonary artery mimicking a mass lesion.

ceftriaxone, cefuroxime, ureidopen, cefoxitin, and cotrimoxazole while sensitive to ceftazidime, imipenem, amikacin, gentamicin, ciprofloxacin, piperacillin/tazobactam, meropenem and less sensitive to cefepime in Case 1 and it was reported to be resistant to gentamicin, imipenem, amikacin, aztreonam, cefepime, and piperacillin whereas sensitive to ceftazidime, ciprofloxacin, and levofloxacin in Case 2. Case 1 received ciprofloxacin 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/mm^3 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 currently under follow-up and have no complaint.

DISCUSSION

B. cepacia complex is a family of catalase-producing, non-lactose-fermenting gram-negative bacteria comprising 9 sub-groups. They cause pneumonia in immunocompromised persons, particularly in those with cystic fibrosis and chronic granulomatous disease. It is rarely encountered in individuals with a normal immune system and without a structural lung disease. To date, community-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 hospitalization eliminates the possibility of nosocomial infection 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 presented with a cavitary lesion in the right upper zone, tuberculosis was considered as diagnosis in the first place. Due to negative sputum smears for AFB, thick cavity wall along with his smoking history and advanced age, malignity was also strongly considered. The triangular consolidation of lingular segment and a suspicious central mass appearance as well as the patient's smoking history and advanced age of case 2 suggested also malignity. The noteworthy aspects of both cases include the fact that they are not immunocompromised patients, 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 absence of hospitalization.

Bacterial culture of bronchoscopic lavage has been the method of diagnosis in both cases. Demir et al. have 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 treatment. To note, he was a diabetic hemodialysis patient and had a central catheter. Cultivation of bronchial lavage seems to be important in the diagnosis of *B. cepacia* 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 antibiotherapy resulted with complete resolution. Waterer and Demir also treated successfully their patients with quinolone antibiotics (10,11). The unusual susceptibility to antibiotics suggested that it is not transmitted

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

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.

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