Case Report

# A case of lung abscess caused by *Burkholderia cepacia* in healthy child

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*Burkholderia cepacia* is a Gram-negative aerobic bacillus known to cause opportunistic infections in the immune-compromised hosts. This microorganism is strongly virulent and causes a necrotising invasive infection that may lead to death. As *B. cepacia* is highly resistant to various antimicrobials, combination antimicrobial therapy must be used instead of monotherapy. We report a successful treatment of lung abscess that was naturally caused by *B. cepacia* in a healthy child, through combination antimicrobial therapy of meropenem and trimethoprim/sulfamethoxazole and operative management. **(Korean J Pediatr 2007;50:89-92)**

**Key Words :** *Burkholderia cepacia*, Lung abscess

## Introduction

*B. cepacia*, formerly known as Pseudomonas cepacia as it was belonged to the Pseudomonas genus, is a Gram-negative aerobic bacillus. *B. cepacia* is known to cause opportunistic infections in immune-compromised hosts. It is highly virulent and often causes a necrotising invasive infection. It is also highly resistant to various antimicrobials. Two cases of lung abscess caused by *B. cepacia* have been reported from all over the world: one occurring in a diabetic adult with pneumonia, after treatment by a nebulizer[1] and the other occurring in a patient with cystic fibrosis, after a lung transplant[2]. In Korea, an adult patient with benign thyroid nodules was infected by *B. cepacia* after undergoing an invasive procedure, eventually giving rise to suppurative thyroiditis[3], but no case of a lung abscess caused by *B. cepacia* has been reported in a healthy child. We report a lung abscess caused by *B. cepacia* in a healthy child with successful treatment.

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## Case Report

A four-year-old boy was hospitalized due to fever and productive cough that had persisted for seven days. He also complained of tachypnea and dyspnea that had been present for a day. The patient had been treated in a local pediatric clinic for seven days for his fever and cough, but his symptoms worsened. His oral intake and activity gradually decreased within three days prior to hospitalization. His medical records and those of his family do not reflect a history of any specific disease.

In the physical examination, he was found to have achieved normal growth and development. He was observed to be mentally alert but he looked acutely ill. Upon chest examination, he was found to have subcostal retraction with short and shallow respiration. His breathing sound was decreased in the right lung field and was coarse with moist rale in both lung fields. According to the laboratory examination, a complete blood count indicated white blood cell (WBC) count of 16,900/$mm^3$ (neutrophil 84%, lymphocyte 13%), hemoglobin of 11.5 mg/dL, platelet count of 181,000/$mm^3$, and erythrocyte sedimentation rate (ESR) of 60 mm/hr. C-reactive protein (CRP) titer was shown to be 6.31 mg/dL, and the cold agglutinin test and Mycoplasma antibody test both yielded negative results in the serologic tests. On the day he

was hospitalized, a chest radiography showed mottled infiltration and pleural effusion in the right lung field (Fig. 1A). As there was not enough pleural fluid, defined by the chest ultrasonography, thoracentesis was not performed, but combination antimicrobial therapy with ceftriaxone and cephalothin was started. During five days of hospitalization, the fever and cough persisted, and dyspnea was aggravated. Through the follow up chest radiographic examination at 5th day of hospitalization, these were showed a marked increase of pleural fluid and mass shadow in the right lung field and left deviation of trachea (Fig. 1B). Thus, thoracentesis and closed thoracostomy were performed. The obtained pleural fluid was orange in color, with a pH of 6.800, a WBC count of 2,940/$mm^3$ (neutrophil 30%, lymphocyte 70%), and a red blood cell (RBC) count of 70,000/$mm^3$. It contained protein of 3.5 g/dL, glucose of 34 mg/dL, lactic dehydrogenase (LDH) of 2,040 IU/L, and adenosine deaminase (ADA) of 94.7 IU/L as exudate. But, it yielded negative for Gram stain and acid fast bacillus (AFB) stain. On the 6th day of hospitalization, a chest computed tomography (CT) was performed and showed generalized consolidation and atelectasis in the right lung field as well as an abscess formation in the superior segment of the right lower lobe (Fig. 2). Thus, thoracotomy was performed. During the operation, multiple small loculated abscesses were found in the right thoracic cavity, as well as an abscess, which was 7- to 8- cm in diameter in the superior segment of the right lower lobe. Therefore, an incision and wedge resection of the abscess were performed, followed by pleural decortication after the reconstruction of the abscess cavity. The blood culture was negative but culture of pleural fluid was positive for *B. cepacia*. An antimicrobial susceptibility test on *B. cepacia* showed that it was susceptible to cefoperazone, ceftazidime, imipenem, meropenem, piperacilin, and ticarcillin, and was resistant to aztreonam, cefotaxime, cephalothin, ciprofloxacin, gentamicin, tetracyclin, and tobramycin. Therefore, combination antimicrobial therapy was altered with meropenem and trimethoprim/sulfamethoxazole (TMP/SMX), and it lasted for 10 days. The patient showed a positive response to this regimen, and was discharged from the hospital after 20 days. After discharge, he was given TMP-SMX medication as an outpatient after eight more days. He completely recovered from mild pleural thickening as shown by the chest radiography (Fig. 1C).

## Discussion

*B. cepacia*, is a gram-negative aerobic bacteria, formerly called *P. cepacia*. Since *B. cepacia* was found to have genetic characteristics different from those of the other bacteria in

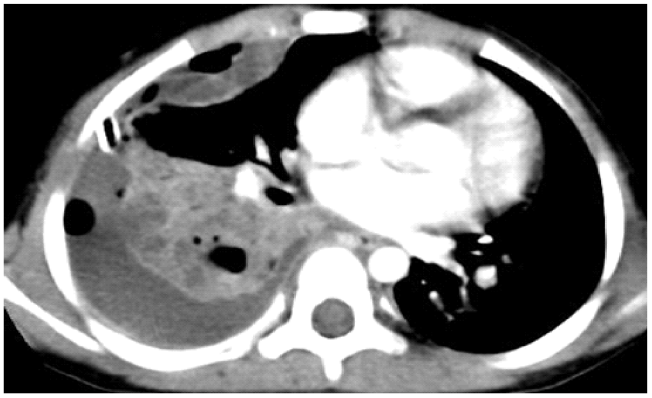

**Fig. 2.** Chest CT on the 6th days of hospitalization : there were pneumonic consolidations and multiple abscesses in right lower lobe and free, loculated pleural effusions and chest tube in effusion.

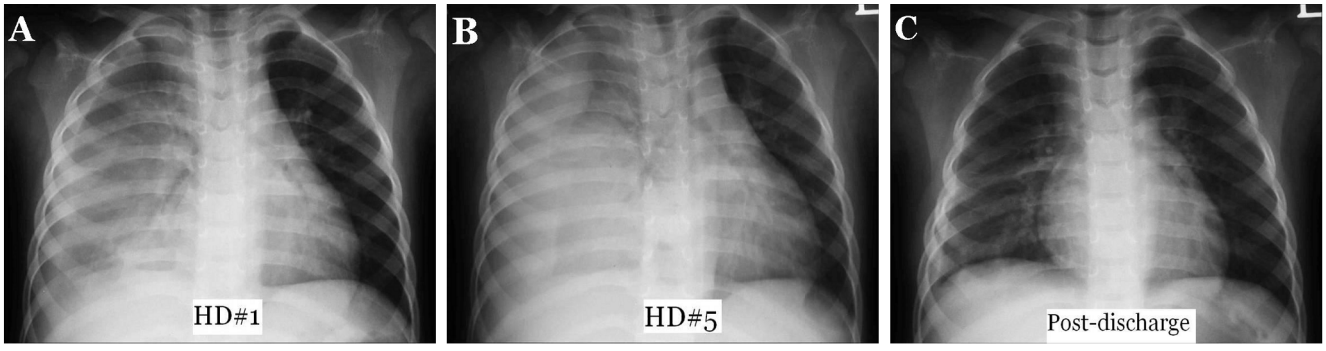


**Fig. 1.** Serial chest AP. (A) On admission, consolidation was shown in right middle to lower lung zone. Also there was pleural effusion of small amount in right lung field. (B) On the 5th days of hospitalization, it was shown the marked increased pneumonic consolidation, pleural effusion in right lung field, and tracheal deviation toward left side. (C) After treatment of outpatient, there was disappeared consolidation and remained only mild pleural thickening in right lung field.

the Pseudomonas genus, it was recategorized into the *Burkholderia genus*[4]. *B. cepacia* was found to consist of at least nine heterogeneous groups, and is thus called *B. cepacia* complex. As it is impossible to classify the elements of the *B. cepacia* complex into groups phenotypically, the classification of such elements is therefore based on their genetic differences[4, 5]. The *B. cepacia* complex was initially described as a pathogen of plants, causing sour skin in onion. However, *B. cepacia* complex has been recently recognized as an important opportunistic pathogen in immune-compromised hosts.

The infection of human by *B. cepacia* complex is occurred in immune-compromised hosts, especially in patients with cystic fibrosis or chronic granulomatous disease (CGD). Respiratory tract infection is the most common manifestation of the disease caused by B. cepaica. Bacteremia may also occur[4, 6]. Because it is capable of thriving in aquatic environments with minimal nutritional resources, it can cause an outbreak of nosocomial infections via a common contaminated source, such as contaminated disinfectants, aqueous solutions, liquid reservoirs, and pharmaceutics, particularly contaminated nebulizing solutions[6-8]. In addition, it can be transmitted by one patient to another through droplets[4].

The clinical manifestation and prognosis of *B. cepacia* infection vary according to its strain[4, 5] since each strain has a different cable pilus, lipopolysaccharide, protease, haemolysin, and/or phospholipase C[4]. It has been known that *B. cepacia* is strongly virulent, and is highly resistant to antimicrobials. It is very important for bacterial toxicity and tolerance that a certain concentration of bacteria is maintained to form a biofilm. As *B. cepacia* invades the epithelial cells to be able to live inside infected cells and to survive in the macrophage, it advantageously maintains high concentration in host cells. In addition, *B. cepacia* is resistant to cationic antimicrobial peptide, so it is resistant to non-oxidative defense by phagocytic cells. A patient who relies their natural defense on non-oxidative defense in phagocytic cells, such as CGD, which have defects in the oxidative defense is more likely to grow *B. cepacia* in the phagocytic cells. The higher the possibility of *B. cepacia* survival in cells is, the easier for it to form a biofilm and to exchange signals called *quorum* between individual bacteria. If bacteria can recognize a *quorum*, they can exchange antimicrobial resistance and can elaborate a *quorum* sensing molecule called autoinducer to increase their overall resistance to antimicrobials[4, 5, 8].

It is difficult to select antimicrobials that can effectively treat *B. cepacia* infection because such infection has a naturally high resistance to antimicrobials and can even enhance its resistance during treatment. Lipopolysaccharide, the component of outer membrane of *B. cepacia*, has a very unique chemical composition, and is resistant to aminoglycoside, polymixin, and/or lactam-based antibiotics (cefepime, imipenem, moxalactam, and piperacillin). Many studies have proven that *B. cepacia* is susceptible to semi-synthetic penicillins, carbapenems (especially meropenem), third-generation cephalosporin (especially ceftazidime), quinolones, and TMP/SMX[4, 6, 9-12]. Treatment of infection by *B. cepacia* must be initiated before the results of antimicrobial susceptibility test are available, and therapy should be based on the most recent antimicrobial susceptibility results. Combination therapy should be used with drugs of two different classes to enhance the likelihood of achieving synergic effects. Dual lactam therapy should be avoided whenever possible as it appears to induce expression of lactamases, resulting in apparent antagonism[4]. Many studies have recommended the combination therapy of meropenem and minocycline, amikacin, or ceftazidime to treat infection by *B. cepacia*[4]. A study particularly chose the combination of ceftazidime and amikacin or ciprofloxacin as the best treatment[10]. Other studies recommended the combination of levofloxacin (or travofloxacin) and third-generation cephalosporine (cefoperazone, ceftriaxone, ceftazidime), or imipenem to treat *B. cepacia* infection[11, 12].

We experienced a case of lung abscess caused by *B. cepacia* that naturally occurred in a healthy child. This patient successfully recovered through a combination therapy of meropenem and TMP/SMX, and through a thoracotomy to incise and drain abscess.

## 한 글 요 약

### 건강한 소아에서 발생한 *B. cepacia*에 의한 폐농양 1례

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*B. cepacia*는 그람 음성 호기성 세균으로서 주로 면역 기능 저하 환자들에서 기회 감염의 중요한 원인균으로 알려져 있다. 이 세균은 매우 병원성이 강하여 침습적인 괴사성 감염을 일으키며 사망에까지 이르게 할 수 있다. *B. cepacia*는 여러 항생제에 높은 저항성을 보이므로 한 가지 항생제의 단독 요법보다는

여러 항생제의 병용용법을 사용해야 한다. 저자들은 건강한 소아에서 자연 발생한 *B. cepacia*에 의한 폐농양을 경험하였고 meropenem과 trimethoprim/sulfamethoxazole 병용 요법과 수술적 치료를 통해 성공적으로 치유한 1례를 보고하고자 한다.

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