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CASE REPORT

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Necrotizing pneumonia-a rare but complex complication in bronchiectasis patients

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Abstract

Pneumonia occurs commonly in bronchiectasis patients with exacerbation, though necrotizing pneumonia remains rare. This report presents two cases of bronchiectasis complicated by necrotizing pneumonia. The first case involves a 39-year-old female with bronchiectasis due to primary ciliary dyskinesia. She presented with severe chest pain and persistent fever unresponsive to oral antibiotics. Chest computed tomography (CT) revealed necrotizing pneumonia with associated empyema, necessitating prolonged antibiotic therapy, pleural drainage and ultimately surgical decortication. The second case is a 39-year-old male with bronchiectasis following ammonia inhalation injury, presenting with pleuritic chest pain and productive cough, with a CT scan showing consolidation with low attenuation areas. Intensive antibiotic treatment was required, alongside corticosteroids and inhaled antibiotics afterward. These cases demonstrate the importance of identifying risk factors for necrotizing pneumonia in bronchiectasis patients. Tailoring treatment strategies, including extending antibiotic regimens, using inhaled antibiotics, and administering corticosteroids, is essential to mitigate poor outcomes.

KEYWORDS
antibiotic, bronchiectasis, necrotizing pneumonia, pneumonia

INTRODUCTION

To date, the concept of the 'vicious vortex' is widely accepted in the pathophysiology of bronchiectasis. This spiral can be described as an ongoing interaction of factors such as chronic bacterial infection, elevated airway inflammation, reduced airway clearance, and the destruction of airway structures. The progression of these factors, especially infection and airway inflammation, is associated with exacerbation of bronchiectasis.[1] During exacerbations, if the infection spreads from the airways to the parenchymal, it can lead to pneumonia. Polverino et al. found that pneumonia occurs in 33% of bronchiectasis exacerbations.[2] The question of whether pneumonia in bronchiectasis patients differs from those without bronchiectasis remains open. Polverino et al. showed similar outcomes,[3] while a recent study by Seo et al.[4] reported a higher frequency of dyspnea, complicated parapneumonic pleural effusion, or empyema. Patients with extensive radiological airway dilation, especially involving three or more lobes or presenting cystic bronchiectasis, appear to be at increased risk of developing more severe pneumonia.[4] Here we present two bronchiectasis cases diagnosed with necrotizing pneumonia, a rare but complex complication of lung parenchymal infection.

Quoc-Khanh Tran-Le and Nam Vu-Hoai have contributed equally as first author.

CASE REPORT

Case 1

A 39-year-old female patient presented with fever, left-sided pleuritic chest pain radiating to the left shoulder, and

productive cough with thickened yellow phlegm for 2 weeks. Oral antibiotic therapy was initiated immediately after symptom onset; however, there was no improvement. She subsequently experienced worsening chest pain, persistent fever with chills, dyspnea, and fatigue, leading to her hospital admission. Her medical history included chronic sinusitis, otitis media with complications of a perforated eardrum and partial hearing loss, and bronchiectasis diagnosed at age 17, with daily productive cough and 3–4 exacerbations annually. She lacked official birth records but recalled being told she experienced respiratory failure at birth, nearly resulting in cardiac arrest and requiring neonatal care. Table 1 presents her blood test results conducted previously to determine the aetiology of her bronchiectasis. Due to her medical history and symptoms, nasal nitric oxide (nNO) was indicated, and the result was 67 nL/min, supporting primary ciliary dyskinesia as the cause of her bronchiectasis. Baseline chest computed tomography (CT) scan revealed cylindrical bronchiectasis in the middle lobe, lingula, and left lower lobe, with a FACED (forced expiratory volume in 1 s (FEV1), age, chronic colonization, extension, and dyspnea) score of 1 and a bronchiectasis severity index (BSI) of 4. Her therapeutic management before this episode included airway clearance techniques (active cycle breathing and postural drainage), long-term azithromycin therapy, along with streptococcal and influenza vaccinations.

TABLE 1 Blood test results of two bronchiectasis patients.

Tests		Case 1	Case 2	Normal range
Complete blood count	WBC (K/mm^3)	6.99	16.4	4.0–10.0
	NEU (K/mm^3)	3.42	14.0	1.7–7.0
	LYM (K/mm^3)	2.69	1.25	0.6–3.4
	RBC (M/mm^3)	4.05	4.92	3.8–6.3
	PLT (K/mm^3)	261.4	403	130–424
Urea (mmol/L)		3.7	3.2	1.7–7.2
Creatinine (mg/dL)		0.89	0.67	0.5–1.2
AST (U/L)		27.7	20	0–35
ALT (U/L)		18.2	49	0–30
Immunoglobulins	IgG (mg/dL)	1427	1315	552–1631
	IgM (mg/dL)	177.8	44.9	33–293
	IgA (mg/dL)	196.3	227.0	65–421
	IgE (UI/mL)	46.6	< 25.0	<130
Alpha 1 antitrypsin (mg/dL)		158.8	326.3	78–200

Abbreviations: ALT, alanine transaminase; AST, aspartate transaminase; LYM, lymphocyte; NEU, neutrophil; PLT, platelet; RBC: red blood cell; WBC, white blood cell.

At admission, she had acute respiratory failure with peripheral oxygen saturation (SpO_2) 85% on ambient air and decreased breath sounds at the lower left lung. Her chest x-ray (CXR) showed homogeneous opacity at the same area (Figure 1A). Complete blood count (CBC) revealed white blood cells 35,800/mm^3 with 94% neutrophils and the serum level of C-reactive protein (CRP) was 384.0 mg/L. Bronchoscopy showed no obstructions. Cultures of sputum and bronchial lavage fluid detected *Escherichia coli*. Her chest CT, performed after chest drainage for empyema, revealed consolidation with small abscesses and cavitation (Figure 1B,C), leading to a diagnosis of necrotizing pneumonia, complicating her pre-existing bronchiectasis. The patient received a four-week treatment of imipenem/cilastatin and levofloxacin, along with chest tube thoracostomy and surgical decortication. The follow-up chest CT scan 4 months later showed significant improvement but still revealed small areas of partial pleural thickening along with bronchial dilation similar to baseline chest CT scan (Figure 1E–G). Upon stabilization, the patient was prescribed another long-term regimen of azithromycin, chest physiotherapy, annual influenza vaccination, and annual follow-up spirometry.

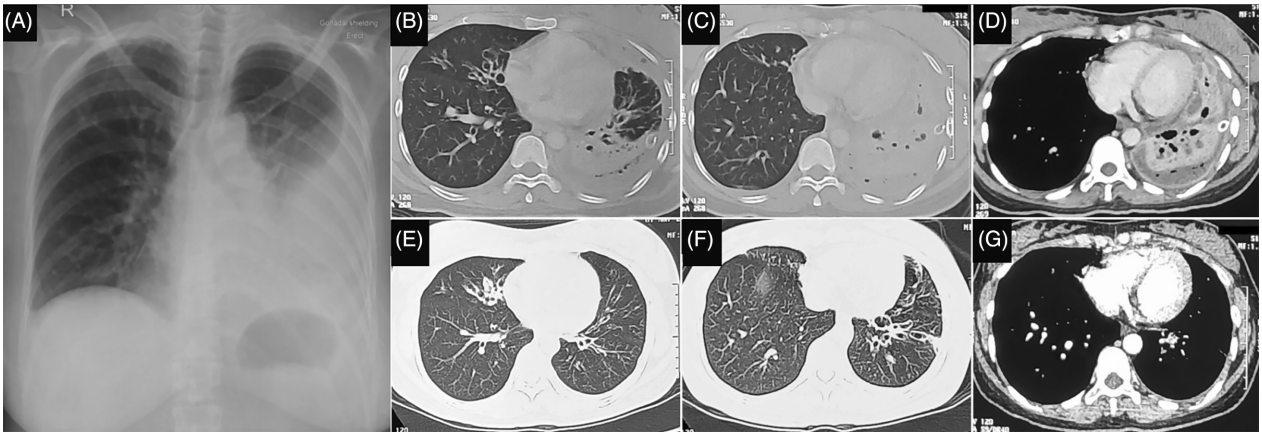

FIGURE 1 Chest x-ray (CXR) and computed tomography (CT) scan of the female patient in case 1. (A) Initial CXR showing homogeneous opacities consistent with pleural effusion in the lower two-thirds of the left lung. (B–D) Initial CT images showing consolidation with small cavitating lesions in the left lower lobe, along with a chest tube for empyema drainage. (E–G) Follow-up CT images demonstrating radiographic improvement and the presence of bronchiectasis in the middle lobe, lingula, and left lower lobe.

Case 2

A 39-year-old male patient was admitted to the hospital due to fever and left chest pain. The illness persisted for 20 days, initially presenting with cough producing copious yellow sputum, which then thickened and turned brownish. The patient was treated with oral antibiotics (sultamicillin 750 mg b.i.d. and ciprofloxacin 750 mg b.i.d.) for 2 weeks, but his condition did not improve. Persistent symptoms, including a productive cough, fever, chest pain, and the development of dyspnea, led to his hospital admission. He was otherwise a healthy adult until he sustained an ammonia (NH_3) gas inhalation injury in 2017. Since then, he has experienced persistent coughing on most days with yellowish sputum and 3–4 exacerbations annually. Chest CT scans at baseline showed bilateral cylindrical bronchiectasis, as illustrated in Figure 2E, with a FACED score of 3 and a BSI of 6. Table 1 shows the previous results of his blood tests. Prior to this event, he had prescribed treatment with antibiotics when an exacerbation appeared.

At admission, physical examination showed decreased breath sounds in the lower left lung. His CBC revealed white blood cells 40,200/mm^3 with 90.8% neutrophils and the serum level of CRP was 280.5 mg/L. The sputum culture was negative. The initial CXR revealed opacity in the lower-half left lung (Figure 2A) and a sequent chest CT scan demonstrated consolidation with inner areas of low attenuation, indicating necrosis (Figure 2F,G). Bronchoscopy also showed no obstruction. Empirical treatment for necrotizing pneumonia was initiated with meropenem, amikacin, and vancomycin. Corticosteroids were also indicated as part of the treatment for severe pneumonia. His clinical improvement was documented, and he was subsequently treated with oral sulfamethoxazole/trimethoprim and linezolid for an additional 2 weeks, followed by oral azithromycin 250 mg and nebulized gentamicin 80 mg/2 mL for an extended period. Follow-up CXRs showed consolidation improvement (Figure 2B–D). He was advised to continue chest physiotherapy for drainage, initiate mucolytic medication, and receive pneumococcal and influenza vaccinations once his condition stabilized.

DISCUSSION

Bronchiectasis is a chronic pulmonary condition marked by irreversible dilation of the bronchi. This typically arises from a triad of complications: impaired drainage, obstruction within the airways, and deficiencies in host immune defence. These factors pave the way for persistent infections that destroy the lung's supportive stroma, leading to further dilation of the airways. Once the bronchi are dilated, they lose the ability to generate adequate pressure needed to expel mucus. This further makes the airway more vulnerable to infection, creating a vicious cycle of infection, inflammation, destruction, and further dilation.[1]

The development of lung parenchymal infections in bronchiectasis patients has been described in several studies.[2,4] The risk factors for the development of necrotizing pneumonia, however, are less well understood. Necrotizing pneumonia is a severe complication of pneumonia, marked by intense lung inflammation, alveolar consolidation,

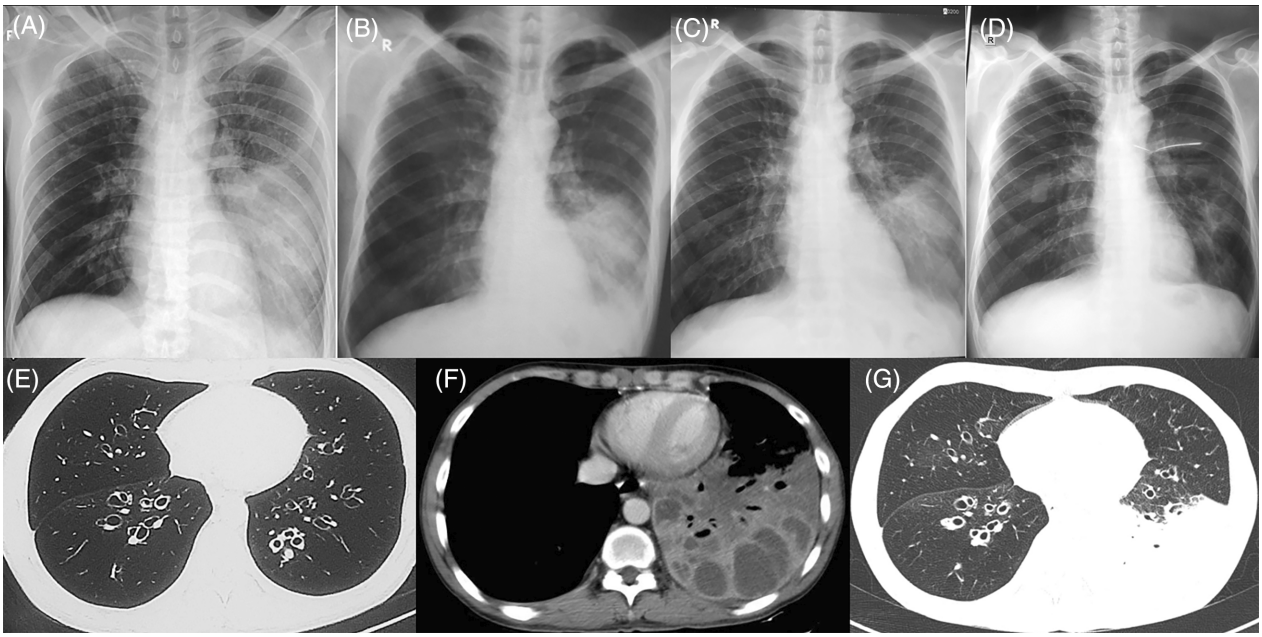

FIGURE 2 Chest x-ray (CXR) and CT scan of the male patient in case 2. (A) Initial CXR with opacity in the lower-half left lung. (B, C) Subsequent CXRs, taken every 14 days, demonstrated gradual improvement in the lower-half left lung. (D) CXR taken 1 month after the previous one revealed a nearly resolved lesion. (E) Baseline chest CT image displaying bilateral cylindrical bronchiectasis. (F) CT image during infection, displaying low-attenuation areas within the consolidated left lower lobe. (G) CT image during infection, showing bilateral bronchiectasis with an opacity in the left lower lobe.

micro-thrombi in small pulmonary vessels, necrosis, and cavitation. The interaction between pathogen virulence and the host defence of airways plays a vital role in the development of necrotizing pneumonia.[5] It is more commonly observed in the paediatric population than in adults. Possible reasons for the higher incidence in children include poor airway clearance due to ineffective coughing, smaller airways prone to obstruction such as bronchiolitis, and underdeveloped immunity. These clinical cases demonstrate that uncontrolled bronchiectasis exacerbations from bacterial infections can lead to severe necrotizing pneumonia, potentially accompanied by empyema. Increased sputum volume and thickening obstruct airways, contributing to this complication.[5] This pathophysiological mechanism may be similar to the compromised airway defences seen in young children.

Early identification of risk factors for necrotizing pneumonia in bronchiectasis patients is essential for optimizing interventions. General risk factors for necrotizing pneumonia include an exaggerated host inflammatory response, pulmonary vascular thrombosis, delayed treatment, smoking, heavy alcohol use, leukopenia, and influenza coinfection.[5] However, no studies have specifically identified these risk factors within bronchiectasis populations. It remains unclear whether these factors apply specifically to bronchiectasis patients or if additional unique factors influence this population. Based on our case experiences, we suggest that bronchiectasis patients presenting with fever and pleuritic chest pain are at increased risk for developing necrotizing pneumonia. Additionally, a poor or delayed response to prolonged antibiotic therapy for exacerbation may indicate this complication.[4]

Management of antibiotic treatment in necrotizing pneumonia presents significant challenges due to vascular obstruction, which reduces the delivery of antibiotics to the affected area.[5] Adjunctive treatments such as corticosteroid anti-inflammatory therapy or surgical intervention are often based on case report experiences. Recent evidence recommended the pivotal role of medical management for necrotizing pneumonia. In the context of treating necrotizing pneumonia in bronchiectasis patients, the following considerations are important: (1) the duration of antibiotic therapy should be extended (in our report, at least 4 weeks), (2) prolonged use of inhaled antibiotics may be effective (achieving high antibiotic concentrations in affected airways), and (3) the use of corticosteroids may be appropriate as per guidelines for treating severe pneumonia to reduce inflammation. Moreover, the empirical antibiotic selection should be tailored to the patient's condition when microbiological results are pending. In cases of necrotizing pneumonia, early coverage for methicillin-resistant *Staphylococcus aureus* (MRSA) and gram-negative bacteria is essential, as these organisms are commonly involved. In addition to pharmacological treatment, physiotherapy for airway clearance is encouraged to promote mucus expulsion, an important factor in both exacerbation and stable stage.[6]

For patients who have stabilized after a severe exacerbation of pneumonic bronchiectasis, implementing a strategy to prevent future exacerbations is crucial. The use of prognostic tools, such as the FACED score or the BSI, can be helpful in evaluating disease severity and aiding clinicians in individualizing management strategies. Long-term antibiotic treatment is essential for patients who experience three or more exacerbations per year, as in our cases above. The choice among inhaled antibiotics, macrolides, or other antibiotics depends on the bacterial pathogen involved and the possible adverse effects on patients.[6] Extended treatment with azithromycin, which serves as both an antimicrobial and anti-inflammatory agent, has been shown to reduce the number of exacerbations and sputum volume, as well as enhance lung function in patients with non-cystic fibrosis bronchiectasis.[6] Although the potential benefits are primarily derived from studies on chronic obstructive pulmonary disease, vaccination for influenza and *Streptococcus pneumoniae* is also recommended for patients with bronchiectasis and other chronic lung diseases, especially for those who have recently recovered from severe acute infections.

In conclusion, necrotizing pneumonia, though a rare complication in patients with bronchiectasis, presents a significant clinical challenge due to its severe nature and the complexities involved in its management. These cases emphasize the importance of promptly addressing infections, particularly in bronchiectasis patients with recurrent exacerbations. Recognizing risk factors and early indicators of necrotizing pneumonia, such as chest pain and persistent symptoms despite antibiotic treatment, is crucial to ensure timely and effective intervention.

AUTHOR CONTRIBUTIONS

The literature search was done by Quoc-Khanh Tran-Le, Lam Nguyen-Ho, Ngoc Duong-Minh, Nam Vu-Hoai and Phung Nguyen-Thi. Data collection was done by Quoc-Khanh Tran-Le, Nam Vu-Hoai, and Lam Nguyen-Ho. All authors contributed equally to analysing and interpreting the data of case reports. Quoc-Khanh Tran-Le and Nam Vu-Hoai drafted the manuscript, with significant contributions by Ngoc Duong-Minh and editing by Lam Nguyen-Ho and Phung Nguyen-Thi. All authors contributed to the article and approved the submitted version.

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CONFLICT OF INTEREST STATEMENT

None declared.

DATA AVAILABILITY STATEMENT

Data sharing not applicable to this article as no datasets were generated or analysed during the current study.

ETHICS STATEMENT

The authors declare that appropriate written informed consent was obtained for the publication of this manuscript and accompanying images.

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REFERENCES

1. Amati F, Simonetta E, Gramegna A, Tarsia P, Contarini M, Blasi F, et al. The biology of pulmonary exacerbations in bronchiectasis. Eur Respir Rev. 2019;28(154):190055. https://doi.org/10.1183/16000617.0055-2019
2. Polverino E, Rosales-Mayor E, Benegas M, Menendez R, Alcaraz-Serrano V, Ansotegui E, et al. Pneumonic and non-pneumonic exacerbations in bronchiectasis: clinical and microbiological differences. J Infect. 2018;77(2):99–106. https://doi.org/10.1016/j.jinf.2018.04.006
3. Polverino E, Cilloniz C, Menendez R, Gabarrus A, Rosales-Mayor E, Alcaraz V, et al. Microbiology and outcomes of community acquired pneumonia in non cystic-fibrosis bronchiectasis patients. J Infect. 2015;71(1):28–36. https://doi.org/10.1016/j.jinf.2015.03.009
4. Seo H, Cha S-I, Park J, Lim JK, Lee WK, Park JE, et al. Clinical relevance of bronchiectasis in patients with community-acquired pneumonia. Am J Med Sci. 2023;365(6):502–9. https://doi.org/10.1016/j.amjms.2023.03.009
5. Krutikov M, Rahman A, Tiberi S. Necrotizing pneumonia (aetiology, clinical features and management). Curr Opin Pulm Med. 2019;25(3):225–32. https://doi.org/10.1097/mcp.0000000000000571
6. Polverino E, Goeminne PC, McDonnell MJ, Aliberti S, Marshall SE, Loebinger MR, et al. European Respiratory Society guidelines for the management of adult bronchiectasis. Eur Respir J. 2017;50(3):1700629. https://doi.org/10.1183/13993003.00629-2017

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