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Case Report

Necrotizing pneumonia with bronchopleural fistula as an uncommon complication of pneumonia in children: a case report*

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ABSTRACT

Necrotizing pneumonia is an uncommon but severe complication of community acquired pneumonia characterized by the development of necrosis, liquefaction, and cavitation of the lung parenchyma. It occurs infrequently in children, ranging from 0.8% to 7% of community acquired pneumonia cases. We reported a case of 28-month-old female infant with a history of severe dyspnea and fever 5 days before admission. After administration of appropriate antibiotics for pneumonia, the patient's condition was still unresolved. Then, contrast CT scan showed cavitary lesions within consolidated lungs with loss of volume and lack of contrast enhancement that confirmed the diagnosis as necrotizing pneumonia. The presence of pneumothorax in the patient depicts a possible bronchopleural fistula which significantly increase morbidity and mortality risk. Surgical management could not be implemented due to worsening condition of the patient. It is suggested that patients with suspicion of necrotizing pneumonia are subjected to chest CT scan to avoid delay in diagnosis and appropriate management.

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Introduction

Necrotizing pneumonia (NP) is a process of necrosis, liquefaction and cavitation of the lung parenchyma caused by infectious pathogen [1]. It is characterized by a condition of progressive pneumonia in previously healthy children despite administration of adequate antibiotic treatment [2,3]. Occurrence of bronchopleural fistula can further cause deterioration of the patient. NP occurs in 3.7% of community pneumonia. However, retrospective studies show an increasing incidence in the last 20 years [1].

The diagnosis of NP is determined by using imaging modalities that show multiple thin-walled cavities within the con-

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Fig. 1 – Chest radiography performed on the 1st day of hospitalization shows homogeneous opacity in the right hemithorax with infiltrates in the upper-middle field of the left lung.

solidation area of the lung. Pathological examination of autopsy or resection of lung specimens reveals lung inflammation, alveolar consolidation and intrapulmonary vascular thrombosis with necrosis and multiple small cavities [3].

CT scan with contrast remains as the gold standard for the diagnosis of NP. Chest radiography have a lower sensitivity compared to CT scan because consolidation and effusion can conceal small radiolucent lesions. Early diagnosis of NP will affect the intensity of patient surveillance, choice of therapy and duration of hospitalization.

Case report

A 28-month-old girl came to the Emergency Room in Dr. Cipto Mangunkusumo National Hospital (RSCM) with complaints of severe shortness of breath and fever 5 days before admission. This patient was admitted to the hospital before the COVID-19 pandemic. Ten days before being admitted to the hospital, the patient had complaints of cough, runny nose, and constipation. On initial physical examination, her vital signs were blood pressure 107/60 mmHg, pulse 170 bpm, temperature 37.2°C, and respiratory rate 40 breaths per minute. There are ronchi and wheezing in both lungs. Laboratory examination showed anemia with Hb 5.4 g/dL. The number of leukocytes increased (32,380 cells/mm) with a Procalcitonin value of 0.77 ng/mL which indicate sepsis.

The chest radiography examination revealed a massive right pleural effusion, possibly accompanied by right lung atelectasis, and left lung infiltrates (Fig. 1). Furthermore, the patient was subjected to a thoracic ultrasound which showed a complex right pleural effusion with pleural thickening and suspicion of localized pleural effusion with right lung consolidation in the medial side (Fig. 2).

Pleural fluid tapping and water sealed drainage (WSD) in the patient revealed a cloudy yellow, seropurulent pleural fluid. Cytology analysis showed that the pleural fluid consisted of mesothelial cells, macrophages, and lymphocytes. There were no malignant tumor cells found in the analysis. Contrast chest CT scan was performed and showed heterogeneous consolidation with multiple cavities in 1 to 5 segments of the right lung, narrowing of the right superior bronchial branch which suggests NP, paraaortic window and sub-carina lymphadenopathy, right hydropneumothorax, and left pleural effusion (Fig. 3).

The patient received a blood transfusion due to anemia and was administered ampicillin sulbactam 250 mg/6 hours, and meropenem 400 mg/8 hours. Follow-up chest radiography examination was performed a week later which showed right lung consolidation with multiple bullae, right middle and lower lung atelectasis, right hydropneumothorax, and left pleural effusion (Fig. 4). WSD production had increased to 30 to 50 ml. The patient was initially planned for thoracotomy resection of the bullae and lobectomy. However, since the general condition of the patient continued to deteriorate, the surgery was delayed.

During hospitalization, the patient still had complaints of persistent cough, shortness of breath, and fever. On physical examination, the breath sound in the right lung is weakened and wheezing is heard. The patient was given additional treatment of Ventolin and Combivent nebulization. The results of blood culture, IgM and IgG of rubella, NS1, sputum AFB and GeneXpert examinations were negative.

On the 16th day of treatment, the patient clinical condition had worsened with complaints of severe dyspnea and high fever up to 40°C. Previous antibiotic treatments were replaced with Cefotaxime-Sulbactam 250 g/6 hours and Amikacin with loading dose of 250 mg/12 hours, followed by 180 mg/24 hours. At that time, the patient was advised to be treated at the PICU. The family decided to withdraw cardiopulmonary resuscitation on the patient. On the 20th day of treatment, the patient began to appear somnolent with gasping breath, acral coldness, and weak pulse. Then, the patient experienced cardiac arrest and was pronounced dead.

Discussion

Necrotizing pneumonia is defined as multiple cavities without marginal enhancement in the necrotic areas of lung parenchyma [4]. Necrosis of the lung parenchyma occurs due to thrombotic occlusion of the alveolar capillaries because of inflammation that leads to ischemia [5]. This condition is rarely seen in children [6,7]. Pediatric patients with pneumonia symptoms, such as persistent fever and dyspnea, that do not improve despite administration of adequate antibiotic treatment need to be evaluated for the diagnosis of NP [8].

CT scan remains as the gold standard for the diagnosis of NP. Chest radiography is less sensitive than CT scans because consolidation and effusion can conceal small radiolucent lesions [4]. Furthermore, CT scan is superior in assessing further complications of the lung parenchyma and pleura [9]. Diagnostic findings on CT scan include loss of normal parenchy-

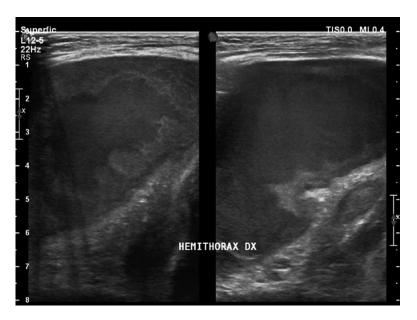


Fig. 2 – Ultrasonography performed on the 1st day of hospitalization reveal pleural effusion with internal echo, pleural thickening across the right hemithorax with collapsed lung, and consolidation in the upper lobe of the right lung.

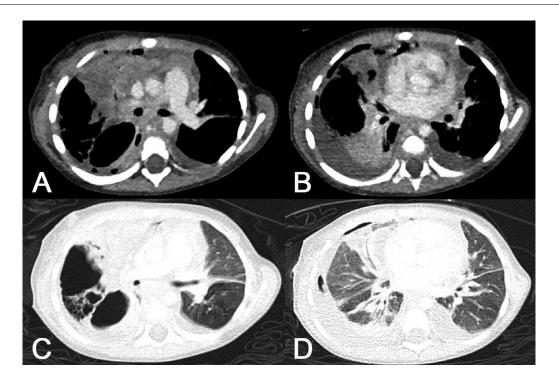


Fig. 3 – CT scan performed on the 1st day of hospitalization shows heterogeneous consolidation in the right lung (A), bilateral pleural effusion with air in the pleural space of the right hemithorax with partial collapse of the inferior lobe of the right lung (B and C). Narrowing of the right superior bronchial branch is noted (B). There is also fibrosis and thickening of the multiple interceptions in the medial and inferior lobes of the right lung (D).

mal pattern, decreased parenchymal enhancement, and multiple thin-walled cavities [4,10].

In this patient, consolidation and cavities without contrast enhancement on chest CT scan were consistent with NP. In addition, multiple bullae, pleural effusions, and localized pneumothorax were shown in CT scan, indicating complications of NP. The presence of a pneumothorax depicts a possible bronchopleural fistula. In another study, bronchopleural fistula occurred in 63% of patients and 80% of patients required surgical therapy [10].

The most common causative microorganisms in NP are Staphylococcus aureus and Streptococcus pneumonia. How-

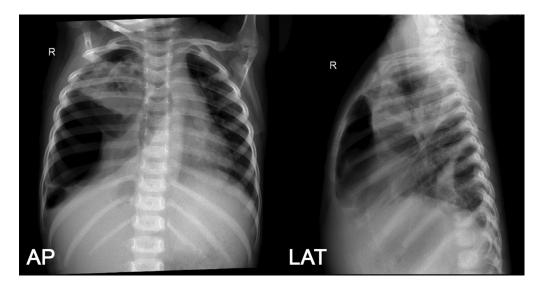


Fig. 4 – Follow up examination of chest radiography with AP and lateral projections were performed on the 8th day of hospitalization, showing multiple bullae and hydropneumothorax with collapsed lung in the right hemithorax, and consolidation with multiple cavities in the upper lobe of the right lung.

ever, causative microorganisms are rarely detected and can only be found in half of the cases [11]. In this patient, there were no microorganisms found on blood culture or pleural fluid analysis. This result can be influenced by the administration of empiric antibiotics to the patient.

The patient's worsening condition of fever, prolonged shortness of breath, and suspected bronchopleural fistula can be an indication for surgical management. However, there is no clear consensus regarding the surgical management of NP. In this patient, surgical management was planned for bullae resection and possibly pneumonectomy, but the patient's condition continued to deteriorate and eventually died before surgical management was implemented. Although the prognosis of NP is generally good, complications such as bronchopleural fistula in the patient can heavily impact the outcome of the patient [7].

Conclusion

Necrotizing pneumonia is one of the rare complications of pneumonia in children that presents severe morbidity. NP is characterized by fever and prolonged shortness of breath that does not respond to adequate antibiotic treatment. The diagnosis is confirmed by finding a consolidation with multiple cavities on chest CT scan examination. When patients are suspected with NP, chest CT scan examination is necessary to avoid delay in diagnosis and appropriate management.

Patient consent

Along with this letter, we would like to confirm that our patient has agreed that her daughter's medical history can be published as a case report paper. In order to protect the patient's privacy, we did not include the physical appearance of

the patient. Furthermore, we focused on imaging and proof of surgical outcomes of the patient, so we did not violate any patient's privacy.

REFERENCES

- [1] Nicolaou EV, Bartlett AH. Necrotizing Pneumonia. Pediatr Ann 2017;46(2):e65–ee8.
- [2] Krenke K, Sanocki M, Urbankowska E, Kraj G, Krawiec M, Urbankowski T, et al. Necrotizing Pneumonia and Its Complications in Children. Adv Exp Med Biol 2015;857:9–17.
- [3] Masters IB, Isles AF, Grimwood K. Necrotizing pneumonia: an emerging problem in children. Pneumonia (Nathan) 2017;9:1–19.
- [4] Sou-Chi S. Necrotizing pneumonia in a young girl: a case report and literature review. Journal of Paediatric Respirology and Critical Care 2007;3(3):8–10.
- [5] Tsai YF, Ku YH. Necrotizing pneumonia: a rare complication of pneumonia requiring special consideration. Curr Opin Pulm Med 2012;18(3):246–52.
- [6] Chatha N, Fortin D, Bosma KJ. Management of necrotizing pneumonia and pulmonary gangrene: a case series and review of the literature. Can Respir J 2014;21(4):239–45.
- [7] Spencer DA, Thomas MF. Necrotising pneumonia in children. Paediatr Respir Rev 2014;15(3):240–5 quiz 5.
- [8] Sawicki GS, Lu FL, Valim C, Cleveland RH, Colin AA. Necrotising pneumonia is an increasingly detected complication of pneumonia in children. Eur Respir J 2008;31(6):1285–91.
- [9] Kosucu P, Ahmetoglu A, Cay A, Imamoglu M, Ozdemir O, Dinc H, et al. Computed tomography evaluation of cavitary necrosis in complicated childhood pneumonia. Australas Radiol 2004;48(3):318–23.
- [10] Hacimustafaoglu M, Celebi S, Sarimehmet H, Gurpinar A, Ercan I. Necrotizing pneumonia in children. Acta Paediatr 2004;93(9):1172–7.
- [11] Lemaitre C, Angoulvant F, Gabor F, Makhoul J, Bonacorsi S, Naudin J, et al. Necrotizing pneumonia in children: report of 41 cases between 2006 and 2011 in a French tertiary care center. Pediatr Infect Dis J 2013;32(10):1146–9.