LUNG BULLAE DUE TO SEPTIC PULMONARY EMBOLISM IN A 4-YEAR-OLD CHILD: A CASE REPORT

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Abstract
Bullous lung disease in pediatric patients is a rare case. The etiology of lung bullae is cigarette smoking history, pulmonary sarcoidosis, alpha 1-antitrypsin deficiency and many more. One of the treatments for lung bulla is surgical approach. We present a case of 4-year-old girl with lung bulla due to septic pulmonary embolism. Primarily the patient was admited with a decrease in consciousness, fever, cough and dyspnea. A CT scan revealed bilateral bullae, multiple nodules in various size, mostly with internal cavities and feeding vessel sign suggestive of a septic pulmonary embolism accompanied by a pneumothorax. We did thoracotomy, wedge resection and pleurodesis, the outcome was clinical improvement and no postoperative complication occurred. Thoracotomy, wedge resection and pleurodesis can be considered as a treatment for lung bulla in pediatric population, so this case report may provide guidance on management of these cases for clinicians.

Keywords: Case Report, Lung Bullae, Septic Pulmonary Embolism, Pediatric, Thoracotomy

1. INTRODUCTION
Bulla is a subpleural air filled space within the lung with the diameter of greater than 1 cm in the distended state, with well demarcated thin wall ≤1 mm. This air containing lesion can be single or may have multiple septations and lies within the lung parenchyma (Negussie et al., 2020). Most patients with bullae have a significant cigarette smoking history, although cocaine smoking, pulmonary sarcoidosis, alpha 1-antitrypsin deficiency, 1-antichymotrypsin deficiency, Marfan’s syndrome, Ehlers-Danlos syndrome and inhaled fiberglass exposure have all been implicated (Goldberg & Carey, 2013).

The most common primary source of infection Septic pulmonary embolism (SPE) is an uncommon disorder in which a microorganism-containing thrombus causes an inflammatory reaction and a mechanical obstruction in the pulmonary vasculature was liver abscess, followed by pneumonia, tricuspid valve infective endocarditis, renal abscess, deep neck infection, and soft tissue abscess.3 SPE can induce formation of lung bullae due to lung inflammation. Here, we report a case of lung bulla due to septic pulmonary embolism in 4-year-old child and treated surgically with thoracotomy, wedge resection, and pleurodesis.

2. RESEARCH METHOD
A 4-year-old girl came to the hospital with a decreased level of consciousness, fever in the last 4 days prior to hospital admission, coupled with dyspnea and cough since 3 days before the admission. On the initial physical examination, there were hyperthermia, tachycardia, tachypnea, and a furuncle in dorsum nasi. No abnormalities were found in other general examinations. In complete blood count, microcytic anemia and thrombocytopenia
were found. The results of blood gas analysis are partial compensated respiratory alkalosis. The patient also had hypokalemia and hypocloremia. The patient was diagnosed with metabolic encephalopathy with thrombocytopenia and nasal abscess.

On the first day since admission, hypoalbuminemia and alkalosis were corrected. A brain CT scan was performed with normal results and a chest CT scan showed multiple nodular opacities and consolidation in all lobes of both lungs, partly accompanied by intralesional cavities and bilateral pleural effusions with differential diagnosis of septic pulmonary embolism.

![Figure 1 CT-scan Thorax 1st day (multiple nodular opacities and consolidation in all lobes of both lungs, partly accompanied by intralesional cavities)](image1)

On the eighth day, she had pneumothorax in the left lung which was treated with water sealed drainage (WSD). In laboratory evaluation, there were anemia, leukocytosis, thrombocytosis, increased levels of CRP and D-dimer. HIV test, IGRA, and Covid-19 swab test were negative and blood cultures were sterile. In the chest CT scan, there were multiple nodules in various size, mostly with internal cavities and feeding vessel sign suggestive of a septic pulmonary embolism accompanied by a pneumothorax (compared to the CT scan on the first day, it’s worse).

![Figure 2 CT-scan Thorax 8th day (multiple nodules in various size, mostly with internal cavities and feeding vessel sign and pneumothorax)](image2)

On the fourteenth day of hospitalization, she had pneumothorax in the right lung and planned on getting a surgery. Thoracotomy, wedge resection and pleurodesis were done in the right lung. Multiple pus and abscesses were found. The anatomical pathology
examination results were chronic xanthogranulomatous inflammation accompanied by an abscess. During hospitalization, patients received meropenem, vancomycin and metronidazole. On the postoperative CT scan results, the lung bullae were reduced and the clinical state in this patient was improved.

![Figure 3 Postoperative CT-scan Thorax day (no pneumothorax and lung bulla were reduced)](image)

3. RESULT AND DISCUSSION

We present a case of lung bullae due to septic pulmonary embolism in a 4-year-old child. At the beginning, the patient’s consciousness was decreased (apathy), she had fever, shortness of breath and cough. On physical examination, there were tachycardia, tachypnea, furuncle on the nose and pathological findings from laboratory evaluation were anemia, thrombocytopenia, respiratory alkalosis and hypoalbuminemia. Dyspnea and tachypnea cause excess release of pCO2 and result in low pCO2 levels on blood gas analysis resulting respiratory alkalosis. The decreased level of consciousness in these patients can be caused by respiratory alkalosis due to vasoconstriction of cerebral blood vessels so the oxygen supply is reduced. The cause of respiratory alkalosis in this patient may be due to pulmonary disruption, this was supported by CT scan findings of multiple nodular opacities and consolidation in all lobes of both lungs, partly accompanied with intraluminal cavities and bilateral pleural effusions suggestive of septic pulmonary embolism.

On the eighth day of hospitalization, pathological findings in laboratory evaluation were anemia, leukocytosis, thrombocytosis, increased CRP, increased D-dimer and on CT scan thorax, multiple nodules in various sizes were found, mostly with internal cavities and there was feeding vessel sign suggestive of septic pulmonary embolism accompanied by pneumothorax.

Septic pulmonary embolism is a rare but life-threatening infectious disease characterized by the embolization of infected thrombi in the pulmonary vasculature. Septic pulmonary embolism is suspected in febrile patients with pulmonary symptoms such as dyspnea, cough and chest pain. The clinical manifestations are not specific (Kano et al., 2018). In literature, the most common causes of septic pulmonary embolism are liver abscess, pneumonia, endocarditis, septic thrombophlebitis, purulent infections of skin and soft tissue (Chou et al., 2016);(Kapoor et al., 2018). In the present case, the primary infection site was unknown. Infection of soft tissue is possible because the patient had furuncle on the nose.
A bulla is an air-containing space within the lung parenchyma that arises from destruction, dilatation and confluence of air spaces distal to terminal bronchioles and is larger than 1 cm in diameter (Grippi et al., 2015). Bullae is a subpleural air filled space within the lung with the diameter of greater than 1 cm in the distended state, with well demarcated thin wall ≤1 mm. This air containing lesion can be single or may have multiple septations and lies within the lung parenchyma (Negussie et al., 2020). Bullae can be asymptomatic or chest distress (Grippi et al., 2015). However, in this case this patient had spontaneous pneumothorax sinistra which can be caused by rupture of bulla (Devu & Joseph, 2019).

In the literature, the most common etiology of bullae is cigarette smoking (Grippi et al., 2015). Risk factors and etiology of bullous lung disease are older age (>45 y.o), cigarette smoking history, cocaine smoking, heroin/methylfenidat user, pulmonary sarcoidosis, HIV, alpha1-antitrypsin deficiency, 1-antichymotrypsin deficiency, Marfan’s syndrome and Ehlers-Danlos syndrome have all been implicated (Goldberg & Carey, 2013); (Devu & Joseph, 2019). This causes were excluded by alloanamnesis, physical examination, laboratory and radiologic findings. Therefore, based on clinical signs, laboratory and radiologic findings, the possible cause of lung bullae is an inflammatory process in the lungs due to septic pulmonary embolism. Chronic inflammation in the lungs can destroy the terminal bronchioles so it distends the airspace due to delayed emptying/progressive air trapping so the bullae form in the lungs (Grippi et al., 2015).

Treatment typically involves surgery, although a variety of procedures have been proposed, including local excision of the bullae, stapler resection, lobectomy, and videothoracoscopy. Surgical therapy is indicated when patients have incapacitating dyspnea or for patients who have complications related to bullous disease, such as infection or pneumothorax (Goldberg & Carey, 2013). The presence of bronchopleural fistula in the right lung which caused pneumothorax was a consideration for doing the surgery in the right lung. Thoracotomy, wedge resection, pleurodesis were done in this patient. In the literature, British Thoracic Society guidelines on the management of pneumothorax, recommending that a weighted decision be made by clinicians as the video-assisted thoracoscopic surgery (VATS) approach is better tolerated by patients but carries a higher rate of recurrence (5% vs 1% for open surgery) (Joshi et al., 2013). Because of the relatively high recurrence rate after surgery, additional procedures such as mechanical pleurodesis are usually applied to minimize recurrence. VATS has many advantages such as reduced complication rates and reduced number of intensive care unit admissions. At 1 year, the recurrence rate in patient with thoracoscopy wedge resection and mechanical pleurodesis was 10,7% (Lee et al., 2014). In this patient, after the thoracotomy she admitted to Pediatric Intensive Care Unit (PICU) and discharged from hospital within a week. The complication such as postoperative bleeding did not happen, and repeated pneumothorax did not occur after the surgery.
4. CONCLUSION

Lung bullae in pediatric can be caused by septic pulmonary embolism due to the inflammation process that destroy the terminal bronchioles so it distends the airspace due to delayed emptying/progressive air trapping. The treatment was done by thoracotomy and bullae resection with good clinical outcome in this patient.

REFERENCES


