Successful outcome of early surgical debridement for pulmonary gangrene in a child
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Citation

Abstract
Necrotizing lung disease and gangrene is rare in children. Different factors predispose to this problem, but most commonly it occurs as a complication of lung infection. Surgical debridement and lung resection are both considered in the treatment of this disease. Our experience in this report suggested that early lung debridement of pulmonary gangrene might limit progression of necrosis of the lungs and should run concurrently with medical management.

INTRODUCTION
The earliest known clinical and pathologic description of pulmonary gangrene was made by Laennec in 1808, while the first reported case was in 1921 by Lenoble and Jegat, who used the term “spontaneous amputation” referring to this phenomenon.

Pulmonary gangrene is extremely rare in children; only one case was reported in 2006 by Yu-chia et al.

Other terms used to describe pulmonary gangrene include spontaneous lobectomy, lobar or pulmonary sloughing and massive sequestration of lungs. Similarly, Eren et al. used the term destroyed lung disease to describe end stage irreversible parenchymal destruction that warrants surgical intervention.

Different factors have been listed as causes of this disease including pulmonary infections, congenital bronchial malformations, bronchiectasis, and hemolytic uremic syndrome. Treatment of pulmonary gangrene is usually surgical. Our experience in this report suggested that early lung debridement of pulmonary gangrene might limit progression of necrosis of the lungs and should run concurrently with medical management.

CASE REPORT
A 21-month-old male child, delivered at full term with a birth weight of 3.9kg, presented with recurrent chest infection and otitis media for which he had been admitted three times since birth. Only three weeks prior to admission, he developed fever (39-40°C) with hypoactivity and poor feeding, as well as cough (but no hemoptysis) and runny nose. He was admitted in another hospital as a case of pneumonia before he was referred to us for further management. On examination, he looked generally weak, was pale and grunting. He had congested throat and otitis media; in the left hemithorax there was dullness with decreased air entry. His spleen and liver were slightly enlarged at palpation.

Chest X-ray revealed left-sided lobar infiltration with effusion. A chest tube was inserted which drained a moderate amount of turbid exudate. He was started on intravenous antibiotics, including vancomycin and ceftriaxone. Both were continued for about 20 days.

Chest computerized tomographic (CT) scan demonstrated left-sided pleural effusion and multiloculated fluid collections with minimally enhancing parietal pleura almost close to the lingular segment, suggestive of lung abscesses. (Figures 1 & 2)

Hematologic work-up confirmed that the patient was anemic (HB: 7.2g/dl) and had neutrophilic leukocytosis (WBC: 36x10^3, 70% neutrophils). Other tests conducted to rule out immunodeficiency (including burst test and flowcytometry) and hypercoagulability statuses (including factor V Leiden, protein C and S) were all negative.

In an attempt to drain the abscess, a left posterolateral thoracotomy was done which revealed a black middle part of the lung (part of upper and lower lobes: gangrene) with small area of empyema and thickened pleura. Full
mobilization of the lung was done and all dead lung tissues were debrided. Repair of the remaining part was effected by continuous suturing. He made a remarkable improvement with no further respiratory distress after surgery.

The patient was discharged home 5 days after the surgery and followed up for 2 months in outpatient clinic with repeated chest x-rays that proved satisfactory expansion of remaining lung tissue.

Lung biopsy showed a large area of necrosis with exudative changes in alveoli, consistent with necrotizing lung tissue and abscess formation with no evidence of malignancy or bronchiectasis (Figure 3); there was associated neutrophilic infiltrate with fibrin thrombi formation (Figure 4). Sample cultures of blood, sputum, cerebrospinal fluid (CSF) and lung tissue were all negative for any growth of aerobic or anaerobic bacteria, acid-fast bacilli or any fungal species.

**Figure 1**
Fig. 1: Necrotizing lung disease (lung window)

**Figure 2**
Fig. 2: Lung abscess

**Figure 3**
Fig. 3: Lung necrosis with abscess formation
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DISCUSSION

Complicated lung infection is the major predisposing factor for lung gangrene. About 3-5% of community-acquired pneumonia cases will progress to irreversible respiratory failure and death in adults\textsuperscript{12}. The incidence of lung gangrene following pneumonia in children might be underestimated due to lack of case reporting and the intention to treat similar cases conservatively unless an abscess is proved to be present. Yu-Chia reported that one out of 15 children (6.5%) with necrotizing pneumococcal pneumonia was found to have gangrene by autopsy\textsuperscript{4}. The infecting agent in all cases was Streptococcus pneumoniae. Penner and colleagues\textsuperscript{5} have ordered the sequence of pathologic events in this disease, starting by necrosis that causes crescents to be seen on radiographs, followed by abscess formation and thrombosis of bronchial and pulmonary circulation. Finally, tissue integrity will be lost and spontaneous sloughing occurs.

In most cases, including ours, the left lung is predominantly involved. Eren and colleagues\textsuperscript{6} attributed this finding to the fact that the left main bronchus is longer and 15% narrower than the right one and has a limited peri-bronchial space, which makes it prone to stretching by lymph node (LN) enlargement. In addition, the course of the left main bronchus is more horizontal, which might impede drainage of secretions. In contrast, the ratio of right to left lung involvement was 12:7 in one of the reported series\textsuperscript{15}.

Infection is the main cause of necrotizing lung disease; unexpectedly, no evidence of infection was present in our case. Causative agents reported in different cases including Mycoplasma pneumoniae, Mycobacterium and Adenoviruses\textsuperscript{14,15} were ruled out, too.

The presence of fibrin thrombi raises the possibility that pulmonary necrosis may be secondary to vascular thrombosis or thrombo-embolization, although the radiologic findings as well as biochemical analysis were inconsistent with this etiology. In 85% of patients, thrombosis develops while the patient is in the hospital\textsuperscript{16,17}. In general, infants older than 3 months and teenagers are the largest groups developing thrombo-embolization with the most important triggering risk factors being the presence of central venous lines, cancer and chemotherapy\textsuperscript{18,19}.

The treatment options for lung necrosis in children include one-stage lung resection, drainage of necrotic tissue followed by resection and drainage only. Pneumonectomy for destroyed lung disease was found by Eren and colleagues to have a mortality rate of 11.7% and a morbidity rate of 23.5\%\textsuperscript{6}.

Treatment of necrotizing pneumonia by lung debridment has been shown to yield excellent long term outcome, without proceeding to lung gangrene. The outcome of the debridement (conservative surgery) of lung necrosis was acceptable on short-term follow-up in our patient, with almost no morbidity. However, more evidence is needed before this can be considered the standard treatment in all cases. Similarly, primary operative therapy for pediatric empyema was found to be associated with a lower in-hospital mortality rate, re-intervention rate, length of stay, time with tube thoracostomy, and time of antibiotic therapy, compared with nonoperative treatment\textsuperscript{20,21}.

Many surgeons would be concerned about the development of a broncho-pleural fistula after this type of treatment, but this was not evident in our case.

In conclusion, necrotizing lung disease and gangrene seem to be extremely rare in children, but should not be underestimated in patients at risk with progressive respiratory distress and lung destruction. Conservative surgical treatment (debridement) has good outcome in selected cases.

References

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