Necrotizing Pneumonia Complicated by Pulmonary Artery and Middle Cerebral Artery Pseudoaneurysms in A Healthy Young Child in Saudi Arabia

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ABSTRACT

Background: Necrotizing pneumonia (NP) is a common complication of community-acquired pneumonia. However, pulmonary artery pseudoaneurysm (PAP) is a rare but serious complication that could follow the infection including NP even in previously healthy children. The usual presentation of PAP are fever, cough with hemoptysis, which is commonly encountered in significant numbers of bronchopneumonia.

Objective: Prompt diagnosis and early intervention to prevent a fatality, identification of the underlying cause, prognosis and outcome.

Material and Methods: A case report of PAP and cerebral pseudoaneurysm complicating NP with residual central nervous system sequelae.

Results: The patient stayed in the hospital for 6 weeks, received vancomycin and meropenem, antifungal therapy, levetiracetam for seizure and prophylactic enoxaparin. His level of consciousness was gradually improving. However, he had left hemiparesis. His speech and swallowing was markedly improved. His follow up chest x-ray 6 months after the vent was normal.

Conclusion: Many causes for PAP exist including lung infection. PAP need to be considered in patients with pneumonia who develop hemoptysis. Cerebral artery pseudoaneurysm can rarely complicate NP with PAP, especially in patients with neurological manifestations.

Keywords: Necrotizing pneumonia (NP), Pulmonary artery pseudoaneurysm (PAP), cerebral artery pseudoaneurysm, endovascular embolization.

INTRODUCTION

Necrotizing pneumonia (NP) is increasingly common complication of chest infection in children for different reasons. Such infection could be serious and leads to significant morbidity and mortality [(1)].

Pulmonary artery pseudoaneurysm (PAP) is a rare, potentially life-threatening condition. It might occur secondary to trauma, pulmonary artery interventions, or infections [(2)].

Here, we reported PAP secondary to NP where we passed through multiple challenges:

1- Intubating a coded patient while doing the CT scan of the brain.
2- Unexpected CT scan finding with massive intraparenchymal hemorrhage.
3- Arranging un-urgent CNS intervention to control the massive CNS bleeding.
4- Knowing what is the underline cause.
5- What is the prognosis and the outcome for such case?
6- Talking to a very anxious parent.

CASE REPORT

Patient information:

Our patient is a 5 years old previously healthy boy presented to our emergency department with high-grade fever, wet cough, progressive dyspnea and post-tussive vomiting containing fresh blood for the last 10 days. He received oral antibiotics without improvement. His family denied a history of a similar condition or chronic illness in the family.

Clinical examination:

The patient was tachypneic with a respiratory rate of 40/m, tachycardic with a heart rate of 150/m, febrile 38.8 °C with normal
oxygen saturation 98% in room air and normal blood pressure. Chest auscultation revealed decreased breath sounds on the right lower region with no added sounds. Otherwise, he had a normal growth parameter and a normal systemic examination. He was diagnosed with right-sided bronchopneumonia, for this admitted to our institute (Prince Sultan Military Medical City) and started on IV ceftriaxone and IV fluid.

Diagnostic assessment:

The initial blood workup showed marked leukocytosis of 27,000 x 10^9/L (normal *4-10 x 10^9/l)* with high neutrophils level 23,000, Hb was low 10.3 g/dl, platelet count was 591 x 10^9/L normal (*150-450x 10^9/L)*, elevated inflammatory markers (CRP 147 mg/l, ESR 61 mm/hr) and normal coagulation profile. Viral screening for respiratory syncytial virus and Influenza A and B were negative. Blood and nasopharyngeal cultures showed no growth.

Chest X-ray (Figure-1) showed patchy opacity in the right middle and lower lung zones with rounded lucencies represent a cavitary lung lesion. Chest

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computed tomography with contrast (Figure-2) showed a large cavitary lung lesion in the right lower lobe with decreased parenchymal enhancement going with NP and small focally dilated vessels represent PAP arising from right pulmonary artery branches.

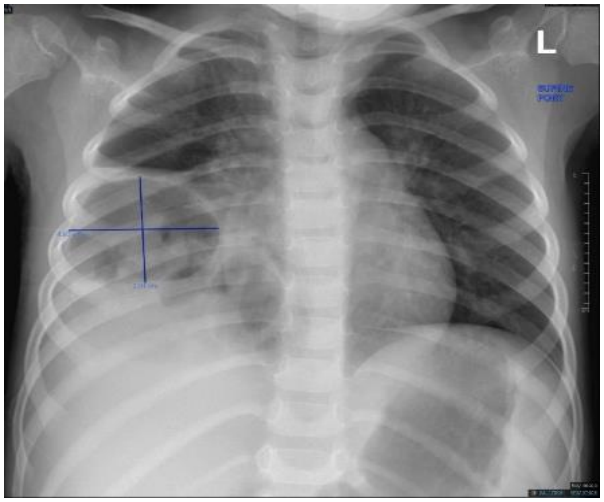

Figure (1): CxR–AP view: showed air-filled cyst (3.9 x 4.9 cm) involving the right middle and lower lung on the background of air space disease.

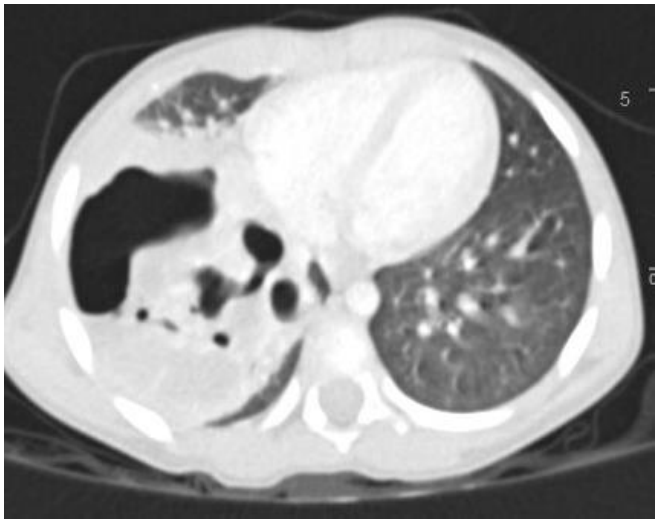

Figure (2): Chest CT scan, axial window showing a large cavity lesion almost occupying the right lower lobe measuring about 63.6 x 76.4 x 46.5 mm with decreased parenchymal enhancement suggestive of necrotizing pneumonia.

Multiple consultations were conducted for this patient. **Immunology**: immunoglobulin level Ig A, M, G, E, antibody to tetanus, diphtheria, pneumococcal, total T, B cell and subset analysis and natural killer cells all were within normal limit.

Rheumatology: Rheumatoid factor, ANA, c-ANCA, p-ANCA were with normal limit.

Cardiology: electrocardiogram ECG and Echocardiograph were normal with no evidence of valvular nor vegetation. **Hematology:** apart of initial marked leukocytosis with left sided shift, coagulation, d-dimer, von Willbrand factor, factor XIII, sickling test were all within normal limit**.**

On the ninth day of admission, the patient was not improving as spiking high-grade fever. He developed generalized seizure with altered level of consciousness.

In addition, he had headache for the last week but was given good attention from the primary team. Meningitis or disseminated brain infection was suspected so, urgent brain CT was arranged before proceeding to the lumber puncture. While the patient in the radiology department, he had bradycardia and desaturation. So, CPR was initiated and the patient, intubated and transferred to the PICU.

Brain CT showed unexpected large acute cerebral hemorrhage in frontal and temporal lobe surrounded by vasogenic edema causing mass effect and midline shift (Figure-3). Neurology was involved for the seizure event and to evaluate the radiological finding for this patient hence started on evetiracetam.

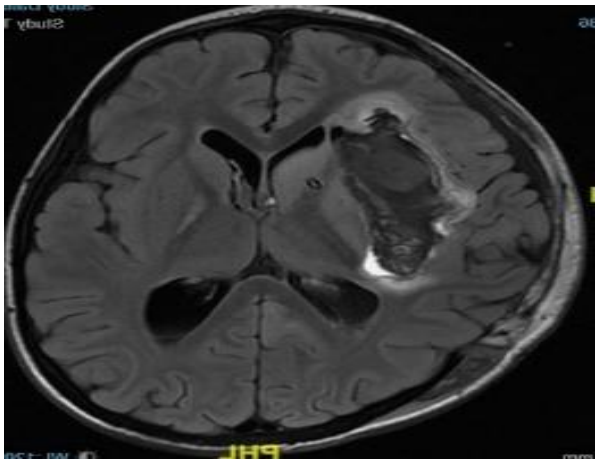

Figure (3): CT brain, axial view Showed large cerebral hemisphere acute parenchymal hemorrhage 6.6 x 3 cm with optimizing most of the frontal and the temporal lobe surrounded by vasogenic edema causing mass effect and midline shift, as well as interventricular hemorrhage bilateral extended to the fourth ventricle.

Genetic team also involved, Whole Exome Sequence (WES) did not detect any mutation related to PAP or cerebral artery aneurysm.

Therapeutic intervention: He had an urgent decompression with craniotomy with intraventricular drain insertion by the **neurosurgeon.** His chest CT was repeated after one week, which showed improvement of the cavitary lesion. However, the size of PAP increased. **Pediatric surgeon** suggested not to intervene as spontaneous resolution is expected and bleeding was stopped.

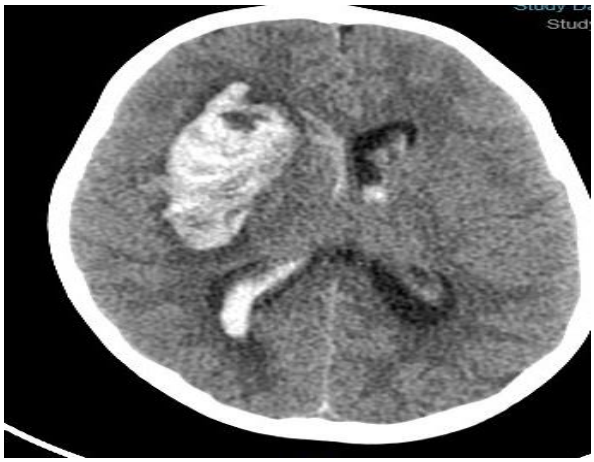

Figure (4): right intraparenchymal and intraventricular hemorrhage

Repeated brain MRI demonstrated right intraparenchymal and intraventricular hemorrhage with an evidence of hypoxic ischemic insult and right middle cerebral artery pseudoaneurysm (Figure 4).

Case conference with all specialties was conducted to decide what the best option to manage for this child is and what to do for such findings. The family were anxious all the time despite updating them on daily bases. Social services were involved, endovascular embolization with coiling of the right middle cerebral artery psudoaneurysm was done successfully by our vascular team.

Follow up and outcome:

The patient stayed in the hospital for 6 weeks, received vancomycin and meropenem, antifungal therapy, levetiracetam for seizure and prophylactic enoxaparin. His level of consciousness was gradually improving. However, he had left hemiparesis. His speech and swallowing was markedly improved. He is being followed in our institute by multi-disciplinary team including neurology, neurosurgery, hematology, pulmonology, occupational therapy and physiotherapy. His follow up chest x-ray 6 months after the vent was normal (Figure- 5).

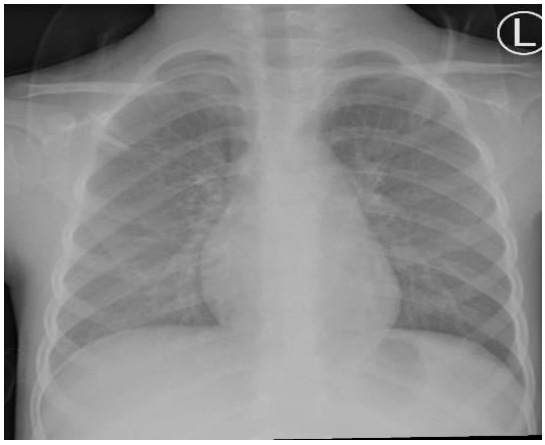

Figure (5): CXR 6 months, which was normal with resolved cavitary lesion in the right lower zone.

DISCUSSION

Pulmonary artery pseudoaneurysm (PAP) is defined as rupture of the vessel wall leading to blood leakage from the vessel. In contrast to true aneurysm, PAP does not involve the three layers of the vessel wall [3]. PAP carries a significant risk of rupture and subsequent mortality [4]. PAP is a rare vascular abnormality, which might occur secondary to penetrating or blunt chest trauma, iatrogenic injury, vasculitis, connective tissue disease, infection and neoplasm [5]. Other causes of central PAP include pulmonary hypertension, congenital heart anomalies, endocarditis, septic emboli and cardiac surgery [3, 6]. Occasionally, no clear cause for PAP is identified [7]. Infectious organisms associated with PAP include S. aureus, S. pyogenes, Klebsiella, Pseudomonas aeruginosa, Actinomyces, Salmonella, Enterococcus, Mycobacterium tuberculosis and various fungi (Mucor, Aspergillus, and Candida) [5, 8, 9]. Our patient had no identifiable organism. The mechanism of PAP secondary to lung inflammation is not completely understood. However, it is thought to result from localized increase in pressure secondary to edema associated with inflammation [3]. Other proposed mechanisms include vasa vasorum embolization, formation of septic emboli obstructing vascular lumen and inflammatory injury of the blood vessel [4, 10]. PAP is rarely complicating necrotizing pneumonia (NP) [6]. (Table 1) represents a collection of cases with PAP secondary to NP.

Table (1): Summary of reported patients with PAP secondary to NP

	Age	Gender M=male F=female	Presentation	Duration between NP and hemoptysis onset	Organism	Management	Outcome
Kalina et al. [5]	19y	M	Acute pancreatitis complicated by acute respiratory distress syndrome ARDS	14 days	Pseudomonas aeruginosa	Angiographic coiling	Improved
Koneru et al. [8]	28y	F	Infective endocarditis, with hypoxic respiratory failure.	25 days	Staphylococcus aureus	None (considered high risk for embolization and surgical resection)	Died
Qureshi et al. [6]	14y	M	βthalassemia major, with febrile neutropenia	8 days	Not identified	Angiographic coiling	Improved
Martin et al. [10]	7m	M	Cough, fever, poor feeding with large pleural effusion in previously healthy child	10 days	Methicillin resistant Staphylococcus aureus (MRSA)	Angiographic embolization	Improved
Garnet et al. [13]	9y	M	Septic arthritis with sepsis, develop respiratory distress after surgery	10 days	MRSA	Lobectomy	Persistent air leak which gradually resolved
Deshmuk et al. [12]	6m	F	Cough, fever, shortness of breath in previously healthy child	10 days	Not identified	Angiographic coiling	Improved
Current case	10y	M	Cough, fever, dyspnea, bloody post-tussive vomiting, previously healthy child	10 days	Not identified	Conservative	Improved

The most common symptom of PAP is hemoptysis, which was the presenting symptom in all PAP secondary to NP as in our case. Less commonly observed symptoms include fever, chronic cough, chest pain, dyspnea, recurrent pneumonia, hypoxemia and bloody chest drain [3, 7, 8] . Our patient was the first reported case of cerebral pseudoaneurysm with PAP secondary to NP. The mechanism of cerebral pseudoaneurysm secondary to lung infection is not clear, may be potentially septic emboli. PAP diagnosis requires **high index of suspicion** in patients presenting with hemoptysis. CXR is the initial radiologic investigation. It might detect consolidation, single or multiple pulmonary nodules, cavitary lesion (as in our case) or pleural effusion [8, 11, 12]. CT with contrast confirms the diagnosis, further delineate the location, size of PAP and visualize the feeding vessel [7,8,13]. It may reveal dilated PA as in our case. Other common findings are enhanced nodule, mass with halo sign and thrombus formation within the dilated PA [4, 11, 14]. The management of PAP is either surgical or conservative depending on the size, location, number of the feeding vessels and patient's hemodynamic condition [4, 7, 15]. Endovascular interventions are preferred especially in

hemodynamically unstable patients, several angiographic procedures were previously reported, including angiographic coiling, stenting and embolization [6, 8]. Surgical management of PAP constitute lobectomy with aneurysm resection, wedge resection and surgical ligation [8, 11]. However, as in our case, spontaneous resolution of PAP has been reported [14]. The mortality rate of ruptured PAP is more than 50% [11].Therefore, prompt diagnosis and early management is essential.

CONCLUSION

PAP is a rare vascular anomalies in children. As pulmonary arteries and adjacent bronchi share the same surrounding connective tissues, any rupture in PAP will lead to hemoptysis. Many causes for PAP exist including lung infection. PAP need to be considered in patients with pneumonia who develop hemoptysis. Cerebral artery pseudoaneurysm can rarely complicate NP with PAP, especially in patients with neurological manifestations.

Table (2): Timeline summary of the case

History and Interventions
1- 5 Year old healthy child with fever, cough for 2 weeks.
2- The child received oral antibiotic with no improvement.
3- The patient admitted with right sided pneumonia under general pediatric for 9 days.
4- The patient transferred to pediatric pulmonology service, shortly seized and coded while doing CT scan of the brain prior lumber puncture and transferred to the PICU.
5- The patient had craniotomy and EVD by neurosurgeon within few hours from arrival to the PICU.
6- The patient developed PAP and middle cerebral artery pseudoaneurysm and endovascular embolization with coiling of the right middle cerebral artery aneurysm at 6 weeks of admission.
7- The patient showed some residual CNS sequelae.
8- The patient improved and chest x-ray showed a complete recovery 6 months after admission.
9- The patient still under regular follow up with multidisplinary teams.

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ABBREVIATION

NP: Necrotizing pneumonia
PAP: Pulmonary artery pseudoaneurysm
CXR: chest X ray
AP: Anterior- posterior
CT: computed tomography
ECG: Electrocardiogram
CNS: central nervous system
IV: Intravenous

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