CASE REPORT

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Open pleural decortication in a 12-day-old neonate with empyema thoracis

Otvorena dekortikacija pleure kod novorođenčeta uzrasta 12 dana sa empijemom

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Abstract

Introduction. Empyema thoracis, defined as the accumulation of pus in the pleural space, is rare in the neonatal population. Limited data are reported in the medical literature, and still, no treatment guidelines are available for this age. **Case report.** We present a term 12-day-old neonate (born healthy) who developed sepsis caused by methicillin-resistant *Staphylococcus aureus* (MRSA) and pneumonia associated with advanced-stage empyema. The child was admitted to our hospital with a few-hours history of difficulty breathing and lethargy. On admission, the child was cyanotic with desaturation and in severe respiratory distress; therefore, the child was intubated, and mechanical ventilation was started. Imaging tests were performed in an emergency, hence chest computed tomography (CT) scan was done without contrast. Suspected congenital pulmonary airway malformation with trapped air collections, significant mediastinal shift on CT scan, and deterioration of the patient's condition indicated urgent surgery. Intraoperatively, the diagnosis of stage II empyema was established, and decortication of thickened parietal and visceral pleura was performed. Afterward, the baby showed quick and progressive clinical improvement. **Conclusion.** The diagnosis and management of empyema in neonates may be challenging, especially in the case of unremarkable history, fulminant progression of the disease, and incomplete imaging tests.

Key words:
diagnosis; infant, newborn; empyema, pleural; sepsis; thoracic surgical procedures; tomography, x-ray computed.

Apstrakt

Uvod. Empijem pleure, definisan kao prisustvo gnoja u pleuralnom prostoru, retko se javlja u neonatalnom uzrastu. Ne postoji mnogo podataka u medicinskoj literaturi, kao ni smernica za lečenje u toj uzrasnoj grupi. **Prikaz bolesnika.** Prikazujemo zdravo terminsko novorođenče uzrasta 12 dana, koje je razvilo sepsu izazvanu meticilin rezistentnom bakterijom *Staphylococcus aureus* (MRSA) i pneumoniju, udruženu sa kasnim stadijumom empijema pleure. Dete je primljeno u bolnicu zbog otežanog disanja i letargije, koji su se javili nekoliko sati pre prijema u bolnicu. Na prijemu u bolnicu novorođenče je bilo cijanotično, u teškom respiratornom distresu, zbog čega je odmah intubirano i započeta je mehanička ventilacija. Hitno su sprovedena radiološka ispitivanja i urađena je kompjuterizovana tomografija (KT) grudnog koša bez kontrasta. Zbog sumnje na kongenitalnu malformaciju disajnih puteva sa „zarobljenom" kolekcijom vazduha, značajnog pomeranja medijastinuma viđenog na snimku KT, kao i zbog pogoršanja stanja deteta, sprovedena je hitna hiruška intervencija. Intraoperativno je dijagnostikovan empijem pleure drugog stadijuma i učinjena je dekortikacija parijetalne i visceralne pleure. Novorođenče se brzo oporavilo nakon intervencije. **Zaključak.** Dijagnoza i lečenje empijema kod novorođenčeta mogu predstavljati izazov, posebno u slučaju nejasne anamneze, fulminantne progresije bolesti i nekompletnog radiološkog ispitivanja.

Ključne reči:
dijagnoza; novorođenče; empijem, pleuralni; sepsa; hirurgija, torakalna, procedure; tomografija, kompjuterizovana, rendgenska.

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Introduction

Empyema thoracis is defined as a pyogenic infection of the pleural cavity with an accumulation of pus in the pleural space [1]. It is frequently seen in children; however, it is very uncommon in the neonatal population. Only a few reports of neonatal empyema thoracis are described in the medical literature [2–5]. Due to the paucity of cases and their differences, the predisposing factors and etiopathogenesis in the neonate are still uncertain. Moreover, there are still no treatment guidelines for managing empyema in neonates. Various modalities of treatment, from antibiotics, chest tube drainage, intrapleural fibrinolytic agent instillation, video-assisted thoracoscopic surgery (VATS) to surgical decortication, have been suggested for treating different stages of empyema in children [6]; however, there are no data available for neonatal age. Although pleural empyema is rare in neonates, it is a life-threatening emergency with rapid progression and high mortality.

We present a term 12-day-old neonate (born healthy) who developed sepsis caused by methicillin-resistant *Staphylococcus aureus* (MRSA) and pneumonia associated with stage II pleural empyema.

Case report

A previously healthy term 12-day-old male neonate was admitted to our hospital due to difficulty breathing and lethargy. The mother noticed these symptoms the same morning the child was admitted to the hospital. The infant was born by spontaneous delivery at term gestation with an unremarkable antenatal history.

On physical examination on admission, the baby was cyanotic with desaturation and in severe respiratory distress. Blood gas analysis revealed hypoxia, hypercapnia, and acidosis, so the baby was immediately intubated, and mechanical ventilation was started. However, despite mechanical ventilation and administration of 100% oxygen, the baby had low oxygen saturation (Sat O_2 = 77%).

A supine babygram following intubation showed hyperlucent right hemithorax with triangular soft tissue opacity in the lateral aspect, increased intercostal spaces, depression of the diaphragm, and mediastinal shift to the left (Figure 1). On the chest ultrasound examination, an absence of lung sliding and comet tail artifacts was evident in most of the right hemithorax, with some pleural effusion and debris in its posterior aspect, without the possibility of differentiating pneumatoceles and pneumothorax. An emergency computed tomography (CT) scan was then performed but without contrast medium administration due to the patient's bad clinical condition in order to further characterize the lung pathology. On noncontrast emergency chest CT, a large septated "coffee bean-shaped" air collection in the anterior aspect of the hemithorax was visualized, extending from the lung apex to the diaphragm, with interposed compressed lung parenchyma (possibly part of the middle lobe) between these air collections and numerous cystic, air-filled spaces in the rest of the compressed lung parenchyma. The ipsilateral increased intercostal spaces, depression of the diaphragm, and contralateral mediastinal shift were confirmed. Differential diagnoses included right-sided congenital pulmonary airway malformation (congenital cystic adenomatoid malformation) vs. pneumothorax (Figure 2).

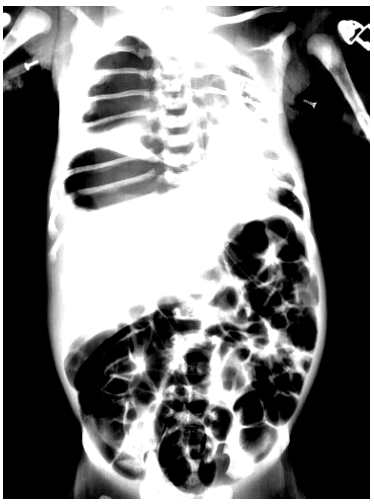

Fig. 1 – Babygram on admission showing hyperlucent right hemithorax with triangular soft tissue opacity in the lateral aspect, ipsilateral increased intercostal spaces, depression of the diaphragm, and mediastinal shift to the left.

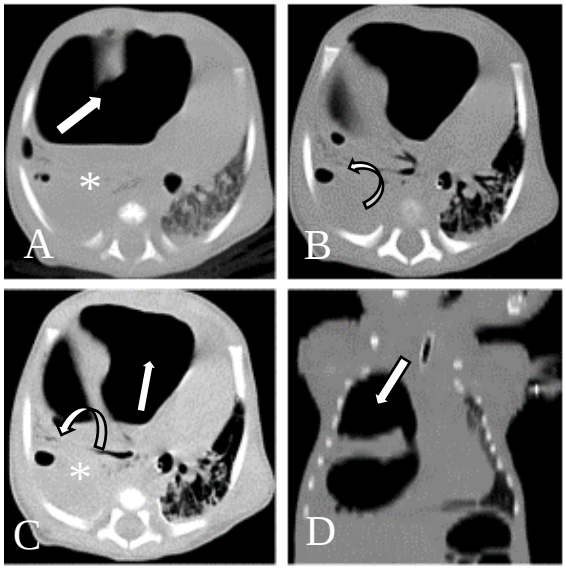

Fig. 2 – Noncontrast chest computed tomography scans in axial (A, B and C) and coronal (D) views demonstrate a large septated "coffee bean-shaped" air collection in the anterior aspect of the hemithorax (white arrow on A, C and D), extending from the lung apex to the diaphragm, with interposed compressed lung parenchyma (possibly part of the middle lobe – asterisk on A and C) between these air collections and numerous cystic, air-filled spaces in the rest of the compressed lung parenchyma (curved arrow on B and C). The ipsilateral increased intercostal spaces, depression of the diaphragm, and contralateral mediastinal shift were evident.

The initial laboratory analyses revealed leukocytosis [30.3 × 10^9/L (reference range – RR 5.0–20.0 × 10^9/L)] with 78% neutrophils (RR 25–55%), hyponatremia [121 mmol/L (RR 131.0–141.0 mmol/L)], and hyperglycemia [32 mmol/L (RR 3.0–6.49 mmol/L)]. C-reactive protein (CRP) was significantly elevated – 196 mg/L (normal values < 3 mg/L), whereas other serum chemistries were normal. The patient was managed with intravenous (iv) fluids, antibiotics (amikacin plus ceftriaxone), various ventilator modes and mechanical ventilation settings, and other supportive therapies.

In the following few hours, the patient's general condition got worse. The child had severe hypoxemia – PaO_2 = 42 mmHg (RR 83–108 mmHg), Sat O_2 = 72% (RR 95–98%), hypercapnia [$PaCO_2$ = 80 mmHg (RR 35–40 mmHg)], and combined respiratory and metabolic acidosis despite optimization of the ventilator settings and FiO_2 of 100%. Considering the deteriorating patient's condition and the high suspicion of progressively enlarging congenital lung malformation, the baby underwent urgent surgery on the same day. Thick fibrous parietal and visceral pleura were found. Frank pus and debris in the pleural cavity were found and all removed. Decortication of thickened parietal and visceral pleura was performed. The right lung expanded completely. Collections of pus in the lung were not found. Three air leaks detected were closed, and chest tube drains were placed. On postoperative babygram, the chest tube was *in situ*, and a small residual apical pneumothorax was evident. The right lung was completely expanded with diffuse, patchy, partially fused reticulonodular airspace opacities, mostly present in basal and central lung projections (Figure 3).

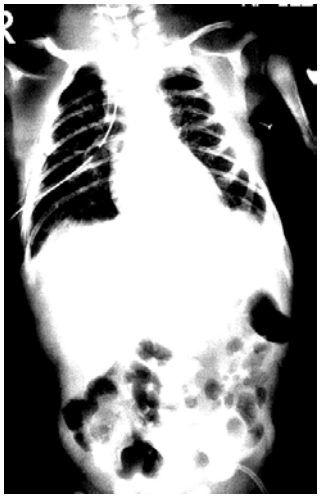

Fig. 3 – Postoperative babygram showing chest tube *in situ* and small residual apical pneumothorax, the right lung completely expanded with diffuse, patchy partially fused reticulonodular airspace opacities, mostly present in basal and central lung projections.

Pus was sent for analysis. Biochemical examination revealed pH = 7.0 (RR 7.60–7.64), glucose 1.9 mmol/L (RR 3.0–6.49 mmol/L), and LDH = 4,550 U/L (normal values < 113 U/L). Pus culture showed growth of MRSA. The blood cultures from admission grew MRSA with the same antibiotic sensitivity. At this point, vancomycin was added to the therapy, and iv immunoglobulin as well**.** The infant's nasal swab was negative for MRSA colonization. Cerebral spinal fluid analysis showed normal glucose and protein level with no pleocytosis, and there was no growth on bacterial culture. The echocardiogram was normal. Investigation for primary immunodeficiency for the infant was also normal.

In the days following surgery, the baby showed quick and progressive clinical improvement, inflammation markers and leukocyte count became normal, and the repeated blood cultures were negative. The intercostal drain was removed on the seventh postoperative day, and the baby was weaned from mechanical ventilation on the eighth postoperative day. Antibiotics were stopped after a total duration of 18 days, and the baby was discharged home.

Chest X-rays performed at hospital discharge and in a four-week follow-up revealed persistent patchy opacity in the central portion of the right lung (Figure 4). The patient, seen at the outpatient clinic at monthly intervals, was asymptomatic and steadily gaining weight.

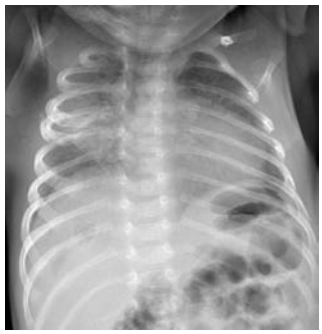

Fig. 4 – Chest X-ray upon discharge showing persistent patchy opacity in the central portion of the right lung.

On the follow-up chest CT scan performed six months after surgery, some pleural thickening and pleural adhesions as well as parenchymal fibrotic bands in the right middle and lower lobe were evident (Figure 5).

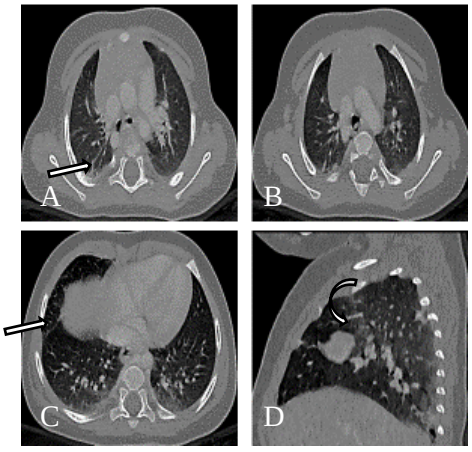

Fig. 5 – Follow-up chest computed tomography scans in axial (A, B and C) and sagittal (D) view six months after surgery showed only pleural thickening and pleural adhesions (straight arrow) as well as parenchymal fibrotic bands in the right middle and lower lobe (curved arrow).

Discussion

In this report, we describe a 12-day-old neonate with MRSA sepsis and pneumonia associated with stage II empyema who was successfully treated with open decortication.

Our patient did not have potential risk factors for MRSA infection as in other reported cases [2, 3]. The patient was not previously hospitalized but was admitted from home. Antenatal history was unremarkable. The parents' nasal swabs were negative for MRSA colonization. The child had no siblings. Primary immunodeficiency was ruled out. The patient suddenly developed signs of severe respiratory distress without prodromal symptoms, so the baby was intubated on admission. This rapid progression of the disease and bad clinical condition on admission are unique from the other reviewed cases [2–5]. In a few case reports of empyema, neonates had stable vital signs, so all the imaging studies could be completely done [3–5].

In terms of diagnosis, chest radiography, ultrasound, and CT scan are utilized for evaluating possible empyema. In the presented case, we performed a CT scan in an emergency without contrast due to the patient's bad clinical condition. The iv contrast allows visualization of pleural inflammation, which is not normally possible with a noncontrast enhanced scan; therefore, split-pleural signs suggestive of pleural empyema could not be shown, which made diagnostics difficult. The literature emphasizes that iv contrast enhancement can be particularly helpful in young children who have poor natural tissue contrast, frequently enabling the differentiation of pneumonia from atelectasis, effusion, empyema, and adenopathy [7].

The management of empyema depends not only on clinical presentation but also on the stage of the disease. Empyema progresses through three stages: exudative, fibropurulent, and organizing stage. According to the American Thoracic Society (ATS), it was the fibropurulent stage (stage II) in our case in which frank pus was present and pleural surfaces became thickened due to fibrin deposition with the formation of loculations [8].

Determination of the empyema stage is important in choosing an appropriate therapeutic option. Symptoms duration has been suggested as one of the criteria for estimating the stage of empyema in children [9], which was challenging in our case due to the rapid progression of the disease. Furthermore, although ultrasound and CT have established roles in the investigation of pleural effusion, previous studies have shown some limitations regarding the determination of the stage of empyema. Kearney et al. [10], studying 50 adult patients with parapneumonic effusion, concluded that neither ultrasound nor CT reliably identifies the stage of pleural infection nor its likelihood of requiring surgical intervention. Additional evidence that CT is unable to distinguish stages of pleural infection in the pediatric population comes from studies by Donnelly and Klosterman [11] and Jaffe et al. [12]. In the presented case, due to a few-hours duration of symptoms and performed CT scan without contrast, it was very difficult to estimate the stage of the disease.

The management of empyema is further influenced by existing practice, surgical experience, availability of appropriate expertise, and local resources. For the advanced stage of empyema, as in our case, therapeutic options included chest tube drainage with or without fibrinolysis, VATS, or open decortication. Considering that our patient was in bad clinical condition and that we could not rule out underlying congenital lung malformation, VATS or open decortication were available treatment options. Even though VATS has been established as one of the standard modalities for the treatment of pleural empyema in older children, it is not routinely used in neonates due to the non-availability of small-sized instruments and other technical limitations of this age [13, 14]. Recently, Sanghvi et al. [5] reported the use of VATS in a 20-day-old newborn with staphylococcal pneumonia and empyema. In our case, due to the non-availability of the equipment for VATS in neonatal age, open decortication was done. Data regarding open decortication in neonates are limited as well since most of the reports were focused on older children beyond neonatal age [9, 15–17]. Open decortication frequently has some perioperative complications, such as persistent air leak from the lung, excessive bleeding, and bronchopleural fistula formation. However, we did not have any of the above complications in the presented case.

According to the literature, if the initial treatment is successful, children usually show complete clinical recovery, and chest radiographs return to normal in 3–6 months [18, 19], which is in accordance with our case.

Conclusion

In conclusion, this case highlights the challenges of diagnostics and management of empyema in neonates, especially in the case of unremarkable history, fulminant and rapid progression of the disease, and incomplete imaging tests.

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