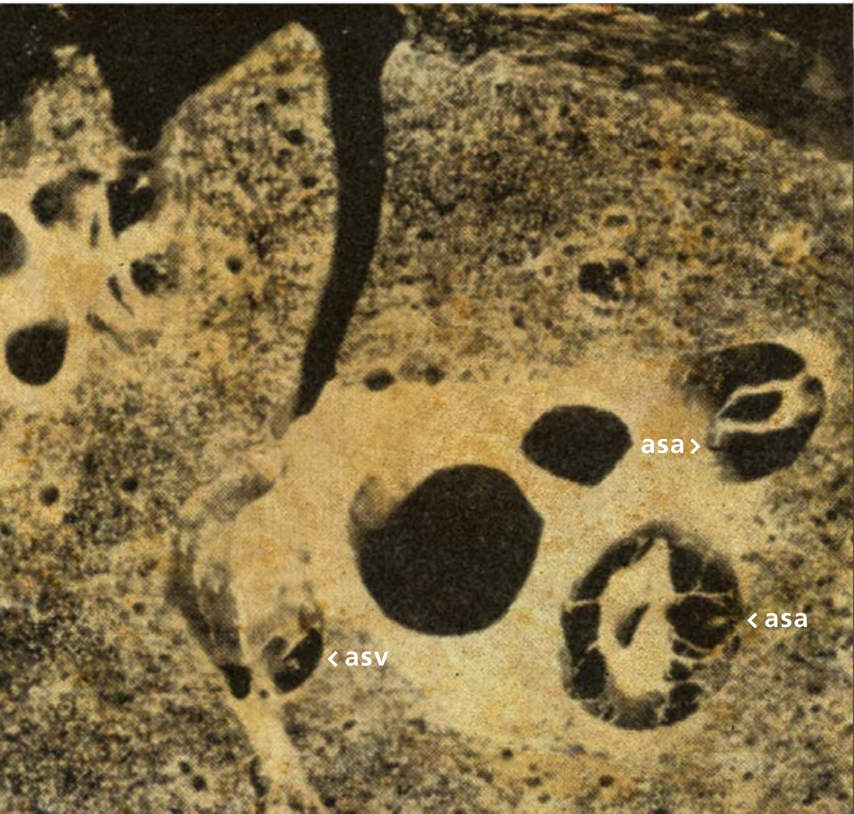
SWISS SOCIETY OF NEONATOLOGY

Challenges of air leak complications in a resource-limited country

February 2018



Berger TM, Berger S, Kamara I, Naurenge E, Ndepavali C,
NEO FOR NAMIBIA – Helping Babies Survive (BTM, BS),
Lucerne, Switzerland, Rundu State Hospital (KI, NE, NC),
Rundu, Namibia

Title figure:
The Macklin effect (asa: air surrounding artery, asv:
air surrounding vein (Source: Arch Intern Med (Chic.)
1939;64:913–926)

Respiratory distress is a common, yet non-specific symptom in sick neonates. Determination of the underlying cause relies on the patient's history, clinical examination, laboratory examinations and, last but not least, chest X-ray. Importantly, respiratory failure is a frequent cause of death. Improving support of neonates with respiratory failure thus has the potential to substantially improve survival rates (1). In low income countries, both diagnostic and therapeutic options for patients with respiratory distress are frequently limited. The following two cases illustrate some of the challenging aspects of neonatal respiratory care in a resource-limited setting.

FIRST CASE REPORT

This infant was a male twin A born to a 17-year-old non-married mother. This had been the 4th pregnancy of this teenager, two of which had resulted in preterm deliveries and early neonatal deaths. The mother has one healthy 3-year-old child.

Delivery of the twins had occurred in a taxi on the way from the village to the hospital. On admission to the neonatal unit, the infant was severely hypothermic (rectal temperature of 33.2 °C) and profoundly hypoxemic (SpO_2 45%) while breathing room air. His birth weight was 1280 g and his gestational age was estimated at 30 weeks. A nasogastric (NG) tube had been inserted into the umbilical vein in the emergency department of the hospital.

When oxygen saturation did not improve with nasal cannula oxygen at 2 l/min, the infant was put on bubble CPAP using a device that had only recently been introduced in this hospital (Pumani®, Hadleigh Health Technologies, San Rafael, CA, USA). Initial settings were a CPAP pressure of 6 cmH_2O, an oxygen flow of 5 l/min and a total flow of 6 l/min, resulting in an FiO_2 of 80%. SpO_2 values rapidly increased and the FiO_2 could be reduced to 45% within 30 minutes. At this point, a chest X-ray was obtained and was felt to be consistent with hyaline membrane disease (Fig. 1).

On day of life (DOL) 2, following a stable 24-hour-period, the infant suddenly desaturated and developed signs of poor perfusion. On auscultation, an unusual rhythmic crunching sound was heard that could not be classified. Findings on a second chest X-ray, which became available two hours later, were consistent with pneumopericardium (Fig. 2). SpO_2 was 80% on an FiO_2 of 90% and capillary refill time remained prolonged despite the administration of a fluid bolus. At this point, it was decided to attempt pericardiocentesis. A 24 G venous cannula with a 2-ml syringe attached was used. The pericardial space was entered from a subxiphoid approach at a 30-degree-angle with the needle directed against the left shoulder. Immediately, 8 ml of air could be aspirated. SpO_2 increased to 91% and skin perfusion improved. The venous cannula was left in place for intermittent aspiration (Fig. 3). Unfortunately, the patient died during the following night.

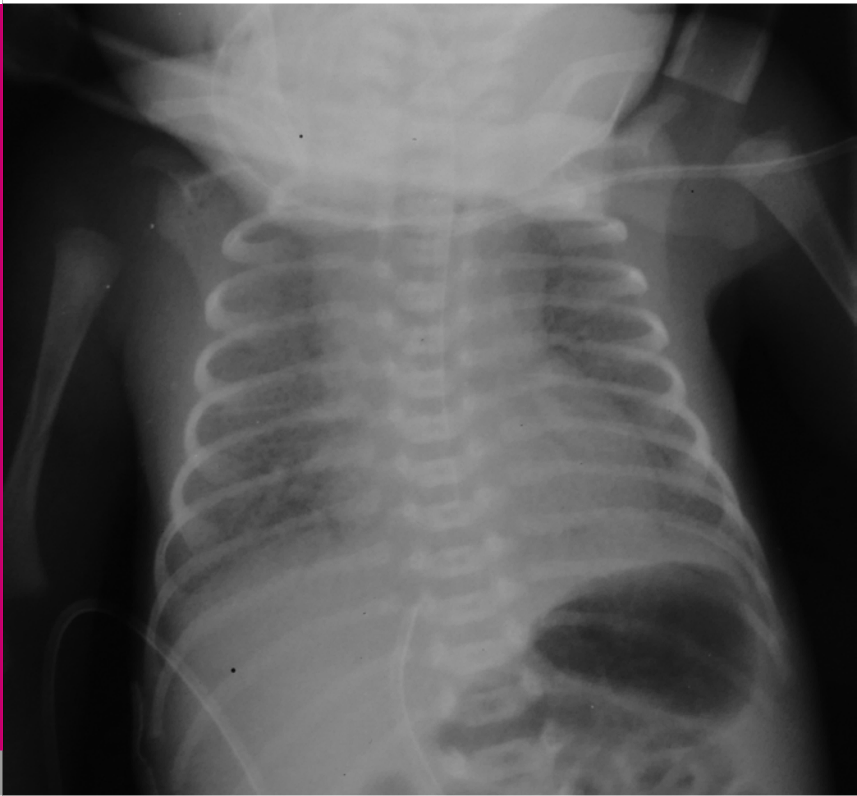


Fig. 1

Twin A: Chest X-ray following stabilization with bubble CPAP on DOL 1: low lung volumes, reticulo-granular pattern and air bronchograms, consistent with hyaline membrane disease (note intrahepatic position of UVC and malpositioned nasogastric tube).

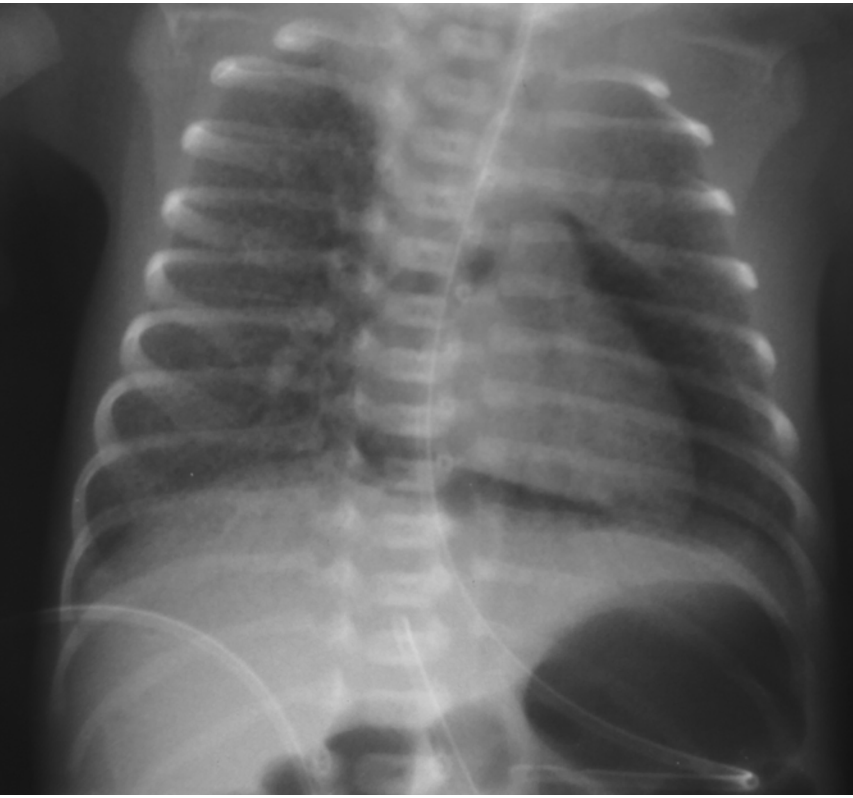


Fig. 2

Twin A: Chest X-ray following sudden deterioration on DOL 2: the heart is almost completely surrounded by air, with the pericardium sharply outlined by air density on either side.

Fig. 3

Twin A on DOL 2: A 24 G venous catheter was used to drain the pneumopericardium: despite initial aspiration of 8 ml of air, the patient's condition improved only transiently (note poor skin perfusion).

Like his brother, twin B arrived in a critical condition with hypothermia (rectal temperature 33.3 °C) and significant respiratory distress and an SpO_2 of 48%. His birth weight was 1150 g. He was put on nasal cannula oxygen at 2 l/min and his color improved. After stabilization of his twin brother, twin B was also put on CPAP with a pressure of 6 cmH_2O, an oxygen flow of 4 l/min and a total flow of 6 l/min, resulting in an FiO_2 of 60%. A chest X-ray revealed findings consistent with hyaline membrane disease. An NG tube had been inserted into the umbilical vein, but was malpositioned in the portal vein (Fig. 4). Another NG tube was inserted next to the first one, hoping that it would enter the inferior vena cava. The infant responded well to CPAP and the FiO_2 could be weaned to 45%.

On DOL 2, the patient appeared stable with decreasing signs of respiratory distress and a lower oxygen requirement. However, on DOL 3, his course was complicated by a right-sided pneumothorax (Fig. 5). A 24 G venous cannula was used to aspirate air (Fig. 6). Unfortunately, there appeared to be a large air leak and almost continuous aspiration was required. This was done manually since no vacuum system was available. A nurse was called to duty, sat at the bedside and continued to intermittently aspirate air. Despite these efforts, the patient died 6 hours later.

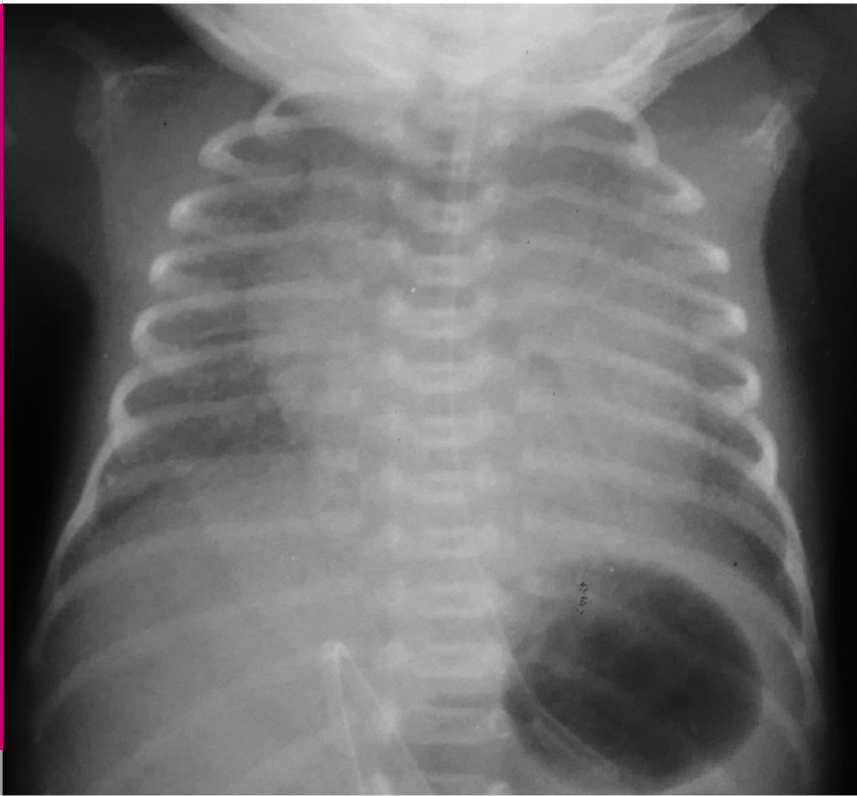


Fig. 4

Twin B: Chest X-ray following stabilization with bubble CPAP on DOL 1: low lung volumes, reticulo-granular pattern and air bronchograms, consistent with hyaline membrane disease (note malposition of the UVC in the portal vein).

Fig. 5

Twin B on DOL 3: chest of X-ray following gradual deterioration on DOL 3: right-sided pneumothorax with slight mediastinal shift to the left (note mal-positioned UVC).

Fig. 6

Twin B on DOL 3: A 24 G venous catheter was used to drain the pneumothorax: because a continuous suction system was lacking, intermittent manual aspiration was performed every 5 minutes over 6 hours (until the patient's death).

The two case reports illustrate multiple challenges encountered in perinatal care in resource-limited countries. Teenage pregnancies, poor antenatal care with late booking, delayed presentation or out-of-hospital deliveries even in high-risk pregnancies are common and put both mothers and babies at risk.

Until very recently, nasal cannula oxygen was the only form of respiratory support available at Rundu State Hospital. Flow rates between 0.5 – 4 l/min are used and the gas is not warmed and marginally humidified since there is only cold bubble humidification. SpO_2 is measured only intermittently due to the lack of adequate pulse oximetry equipment. Very likely, both hypoxic and hyperoxic episodes go largely unnoticed.

The introduction of CPAP devices potentially can have a dramatic impact on the survival rates of neonates with respiratory distress (1, 2). In July 2017, the Pumani© bubble CPAP device (Hadleigh Health Technologies, San Rafael, CA, USA) was introduced at the Rundu State Hospital. This machine is very robust, easy to use and affordable. Initially, the two patients presented in this report responded very nicely: SpO_2 rapidly increased and FiO_2 could be reduced to less than 50%.

While patients with poorly compliant lungs benefit from CPAP therapy, they can still develop air leak complications when spontaneous respiratory efforts lead to (focal) overdistension and lung damage.

First described by Macklin in 1939, air can travel along the vascular sheaths and eventually lead to pulmonary interstitial emphysema and pneumomediastinum (3). It has been speculated that mediastinal air dissecting at the reflection of the parietal to visceral pericardium near the ostia of the pulmonary veins leads to pneumopericardium (4). As the two presented cases illustrate, detection and management of air leak syndromes can be very challenging in resource-limited countries.

Literature on pneumopericardium in neonates is with hyaline membrane disease is scarce. One of the largest series was published from the University of Minnesota Hospitals and St. Paul Children's Hospital in the pre-surfactant era (5). In 23 out of a total of 28 patients, pneumopericardium resulted in clinical pericardial tamponade. Mortality rate was 54%. The rhythmic crunching sound heard in twin A is known as Hamman's sign (after Louis Hamman, Johns Hopkins University clinician), which can also be heard in patients with anterior pneumomediastinum, or «bruit de moulin» (mill-wheel murmur).

In high income countries, the described patients would have qualified for early surfactant replacement therapy; however, this expensive drug is currently not available at this hospital. It is noteworthy that surfactant preparations were included in the Essential Drug List of WHO in 2008 (6). In its current version (20^{th} List, March 2017, amended August 2017), surfactant is listed in the Complementary List. This list presents essential medicines for «priority diseases, for which specialized diagnostic or monitoring facilities, and/or specialist medical care, and/or specialist training are needed.» (7).

The use of surfactant replacement therapy (SRT) in developing countries is still limited because of a) high cost, which may exceed the per-capita GNP (300 – 500 USD) in some countries, b) lack of skilled personnel to administer SRT, and c) lack of support systems after the SRT. Recent developments may have the potential to address these constraints.

The development of SP-B and SP-C enriched synthetic surfactants (8) may reduce the costs of SRT. In recent years, the INSURE (intubation-surfactant-extubation) procedure and other forms of less invasive surfactant administration (LISA) have become increasingly popular in high income countries (9). The latter procedure does not require mechanical ventilation via an endotracheal tube, since the surfactant preparation is administered intratracheally via a small diameter tube, while the infant is spontaneously breathing (10). Alternatively, surfactant administered through a laryngeal mask airway (LMA) may be a valuable option as it does not require the skills necessary for laryngoscopy and / or intubation (11). Robust and low-cost CPAP devices have been developed and could be used both prior to and after SRT (12).

CONCLUSION

Improving the management of neonates with respiratory distress in developing countries has the potential to greatly improve survival chances for affected neonates. This will require improvements in both diagnosis and therapy, as well as management of complications of various causes of respiratory failure. It is highly desirable that research in neonatology also addresses the needs of newborn infants born in low and middle income countries (10).

REFERENCES

1. Kamath BD, Macguire ER, McClure EM, Goldenberg RL, Jobe AH. Neonatal mortality from respiratory distress syndrome: lessons for low-resource countries. Pediatrics 2011;127:1139 – 1146 _(Abstract)_

2. Kawaza K, Machen HE, Brown J, et al. Efficacy of a low-cost bubble CPAP system in treatment of respiratory distress in a neonatal ward in Malawi. PLoS One 2014;9:e86327 _(Abstract)_

3. Macklin CC. Transport of air along sheaths of pulmonic blood vessels from alveoli to mediastinum – clinical implications. Arch Intern Med (Chic.) 1939;64:913 – 926 (no abstract available)

4. Jokic RR, Kovacevi B, Beserminji M, Tatic M. Surgical approach to tension pneumopericardium in newborns and infants. JPSS 2008;2:32 – 35 _(Abstract)_

5. Emery RW, Landes RG, Lindsay WG, Thompson T, Nicoloff DM. Surgical treatment of pneumopericardium in the neonate. Word J Surg 1978;2:631 – 637 (no abstract)

6. Vidyasagar D, Velaphi S, Bhat VB. Surfactant replacement therapy in developing countries. Neonatology 2011;99:355 – 366 _(Abstract)_

7. WHO Model List of Essential Medicines, 20th List (March 2017) _(Website)_

8. Sweet DG, Turner MA, Straňák Z, et al. A first-in-human clinical study of a new SP-B and SP-C enriched synthetic surfactant (CHF5633) in preterm babies with respiratory distress syndrome. Arch Dis Child Fetal Neonatal Ed 2017;102:F497-F503 _(Abstract)_

9. Gortner L, Schüller SS, Herting E. Review demonstrates that less invasive surfactant administration in preterm neonates leads to fewer complications. Acta Paediatr 2017 Nov 24 [Epub ahead of print] _(Abstract)_

10. Herting E. Less invasive surfactant administration (LISA) – ways to deliver surfactant in spontaneously breathing infants. Early Hum Dev 2013;89:875 – 880 *(Abstract)*

11. Roberts KD, Brown R, Lampland AL, et al. Laryngeal mask airway for surfactant administration in neonates: a randomized, controlled trial. J Pediatr 2017 Nov 21 (Epub ahead of print) *(Abstract)*

12. Berger TM. Neonatal respiratory care: not how, but where and when. Lancet Respir Med 2013;1:280 – 282 (no abstract available)

concept & design by mesch.ch

SUPPORTED BY

VIFOR PHARMA

CONTACT

Swiss Society of Neonatology

www.neonet.ch

webmaster@neonet.ch