TWO CASES OF PNEUMOMEDIASTINUM IN FULL-TERM NEWBORNS AFTER UNCOMPLICATED VAGINAL DELIVERIES: VARIOUS ETIOLOGIES AND RADIOGRAPHIC APPEARANCES

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Pnemomediastinum is defined as the presence of aberrant air in the mediastinum, also known as mediastinal emphysema. Significant pneumomediastinum is rare, particularly in term neonates. It is usually related to the different predisposing factors such as birth injury, meconium aspiration, assisted ventilation or infectious diseases although it may occur idiopathically. Pneumomediastinum is one of the important causes of respiratory distress syndrome that should not be overlook in term neonates. The elevation of thymic lobes off the heart on plain view and the presence of retrosternal air collection on lateral view are crucial signs on chest x-rays that need to be assessed cautiously by radiologists and neonatologists. We report two cases of pneumomediastinum in term infants who presented with respiratory distress. One case was diagnosed as pneumomediastinum and assisted ventilation was supposed as a predisposing factor, the other case was diagnosed as spontaneous pneumomediastinum without underlying lung disease or predisposing factors. The definitive diagnosis was made by combination plain and lateral chest x-rays without further imaging method was required. Both patients received conservative treatment with close observation clinically and radiologically and were discharged after several days in good condition.

Keywords: Pneumomediastinum, newborn, complication, vaginal delivery.

I. INTRODUCTION

Pneumomediastinum (PM) is a condition defined as the presence of aberrant air in the mediastinum, also known as mediastinal emphysema. It is relatively uncommon in the neonatal practice and most cases occur asymptomatically.[1] Significant pneumomediastinum causing respiratory distress in the term newborns is scarce. The incidence of PM is in the order of 2.5 per 1000 live births and of 1-2% of infants with respiratory distress.[2,3] The appearance of pneumomediastinum is associated with certain predisposing factors such as birth injury, meconium aspiration, assisted ventilation or infectious diseases.[4] However, it also may occur without underlying lung diseases or trauma, also known as spontaneous pneumomediastinum.[1,5] Chest radiography is a valuable method used for the definitive diagnosis as well as monitoring in neonatal cases. In this article, we report two cases of term neonates appearing respiratory distress resulting from pneumomediastinum in different clinical circumstances. Both newborns received conservative treatment with close monitoring and pneumomediastinum resolved after few days.

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Received: 12/08/2022
Accepted: 27/09/2022

II. CASE PRESENTATION

Our first reported patient is a 40-week gestational age male newborn delivered by uncomplicated vaginal delivery with a birth weight of 3200 gram. APGAR score was 6 at

the first minute which is compatible with the presence of weak cry, pale skin, and a reduction of muscle tone. Following a neutral response to the initial stimulation, he required positive-pressure ventilation immediately in the first several minutes. APGAR score at 10th minute improved to 8. However, he was still presenting marked xiphoid retraction and nasal flaring with the respiration rate of 60 breaths per minute. Therefore, he commenced on oxygen by face mask at rate of 5 L/min, followed by continuous positive airway pressure (CPAP) at a pressure of 5 cm H_2O, FiO_2 60% after one hour of delivery. The capillary blood gas at the second hour of age on CPAP 5cm, FiO_2 0.6 showed: pH 7.29; pCO_2 41; pO_2 94; BE -6, HCO_3 20.3. Bedside chest x-ray at three hours of age revealed a pneumomediastinum with the features of a "wind- blown spinnaker sail" of both thymic lobes being elevated off the heart, which is compatible with the retrosternal translucency seen on the lateral view (Figure 1A, 1B). Surfactant replacement therapy was indicated since his respiratory distress syndrome was unimproved. Close observation and repeat radiography were performed to evaluate his pneumomediastinum. Following the improvement of his clinical condition, it was agreed to discontinue the CPAP and transfer to mask oxygen therapy at 10L/min at 20 hours of age. His blood gas at 24 hours of life revealed: pH 7.497; pCO_2 29.1; pO_2 105; BE -1, HCO_3 22.5. Oxygenation was decreased during the following days and was ceased on day three. There was a favorable clinical evolution, with minor tachypnea and desaturation during breastfeeding. There was no sign of pneumomediastinum revealed on the repeat x-ray on day three (Figure 1C). On day seven, his condition was back to normal.

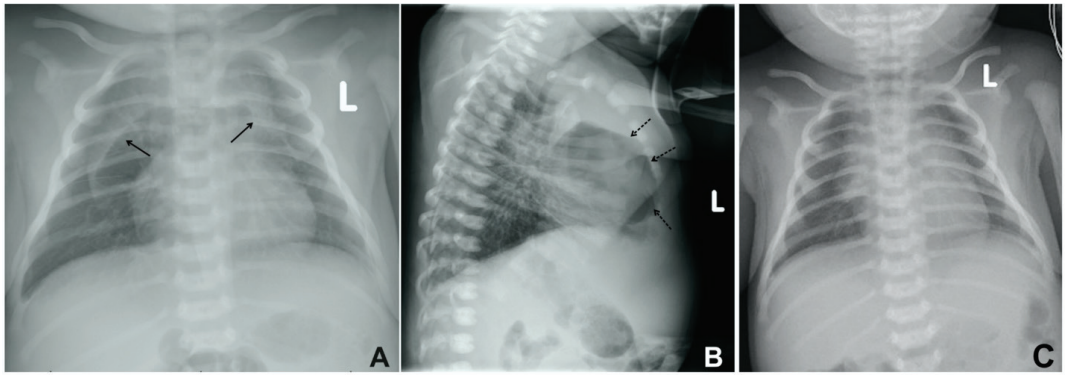

Figure 1. *(A) Anteroposterior (AP) chest radiograph demonstrates pneumomediastinum with the thymic lobes (black narrows) are elevated off the heart due to the underneath air collection. (B) Lateral chest radiograph reveals retrosternal air collection (dashed narrows). (C) AP chest radiograph after 3 days shows the thymic lobes at normal position without displacement.*

The second reported patient is also a male infant born at 40 weeks of gestation by uneventful vaginal delivery. The infant was of full-term size and weighed 2900g. APGAR scores were 8 and 9 after one and five minutes. He was sent to the baby nursery with the mother after the normal first examination. Three hours of age, he developed spontaneously persistent grunting and showed a poor sucking reflex. Clinical examination by neonatologist revealed a mild cyanosis and the reduction of heart sounds. No sign of fever or other systemic symptoms was found. Oxygen saturation remained at 95% on room air. Emergency

bedside chest x-ray and echocardiography were indicated immediately to evaluate lungs and heart condition.

Plain chest radiography at the third hour after birth showed well-defined radiolucent band framing the right border of the heart, extending from superior mediastinum to right hemidiaphragm with a crescent shaped homogeneous opacity peripherally representing displaced thymic lobe and compressed lung parenchyma (Figure 2A). On left field, it clearly showed the thymic lobe lifted off the left margin of heart. To confirm the diagnosis of pneumomediastinum, a lateral projection was obtained and the mediastinal air was seen as a retrosternal translucency (Figure 2B). The definitive diagnosis of spontaneous PM was given.

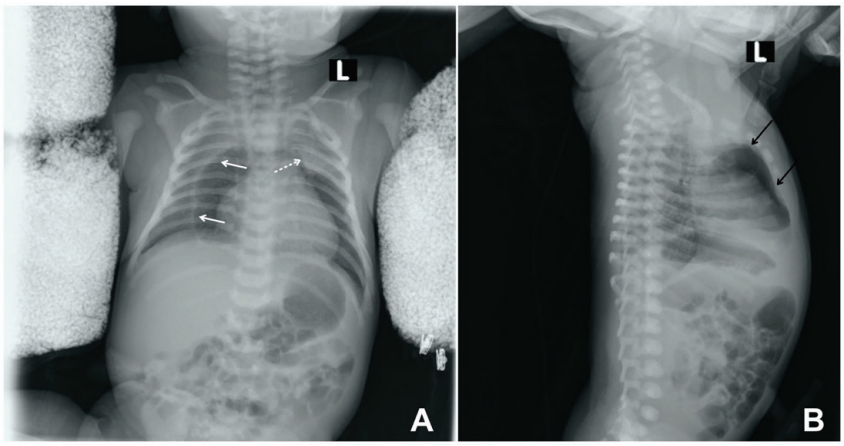

Figure 2. *(A) AP chest radiograph shows well-defined radiolucent band framing the right border of the heart, extending from superior mediastinum to right hemidiaphragm with a crescent shaped homogeneous opacity peripherally representing displaced thymic lobe and compressed lung parenchyma (white narrows), the left lobe of thymus is lifted of the heart (white dashed narrow). (B) Lateral chest radiograph reveals retrosternal air collection (black narrow).*

Emergency cardiac and mediastinal ultrasound showed an anechoic, thin stripe surrounding the heart, representing pericardial effusion without evidence of cardiac tamponade. No sign of air within mediastinum or pericardium was observed in this ultrasound study.

The patient was admitted to neonatal intensive care unit (NICU), received extra oxygenation in an incubator and close observation. His clinical symptoms gradually improved. On the day four of life, his respiratory resolved and chest radiograph showed complete resorption of PM. Neither mechanical ventilation nor invasive procedure required. The infant was discharged after a week in good condition.

III. DISCUSSION

Neonatal pneumomediastinum is a rare entity and occurs in approximately 0.4 – 2.5 per 1000 live births according to certain studies reported in the 1950s and 1960s.[2,3] In the group of uneventful vaginal deliveries, the incidence of PM was demonstrated about 2.3%.[6] However, the observed prevalence has been probably underestimated compared to the reality as PM is often asymptomatic or shows subtle symptoms and self-limiting. Infrequently, significant PM may cause respiratory distress, occupies about 1-2% of infants with respiratory distress and 0.1% of infants hospitalized in NICU.[3,7] In term neonates, most cases of PM

are associated with certain predisposing factors including labor-related trauma, meconium aspiration syndrome, infectious lung diseases as well as positive pressure during resuscitation or assisted ventilation, although it can be idiopathic and occur without any implicit risk factors.[4–6]

The origin of aberrant air in the mediastinum may be from the upper respiratory tract, intrathoracic airways, gastrointestinal tract, from outside air after trauma or surgery, or can be generated by bacteria in infected conditions.[1] However, pneumomediastinum due to leaking air from microscopic alveolar rupture is the most common clinical situation, particularly in neonates. The underlying pathophysiology of the air leakage from ruptured alveoli in pneumomediastinum firstly introduced by Macklin through the experimental works is owing to an exceeding pressure gradient between the alveoli and the surrounding tissues. The air along the pressure gradient leaks from the ruptured alveoli to the perivascular and peribronchial tissues and subsequently towards the mediastinum.[8] The pressure disparity results from either a sudden increase of intra-alveolar pressure such as positive-pressure ventilation, Valsalva maneuvers or a reduction of interstitial pressure such as in extreme respiratory effort or in rapid reduction of atmospheric pressure.[1] In our first case, although it is hard to confirm if assisted ventilation causes mediastinal emphysema or not, it should be considered as a predisposing factor. In other instances, the clinical setting of pneumomediastinum is probably spontaneous without underlying lung disease as our latter case, which developed a PM without prior assisted ventilation or eventful labor. It is also noteworthy that the apparent definition of spontaneous pneumomediastinum is controversial and used inconsistently among authors. Notably in the instances of pneumomediastinum associated with assisted ventilation, some authors supposed to be a "spontaneous" PM while others supposed to be "traumatic" PM.[1,4,5,9] To our knowledge, PM related to assisted ventilation is a barotrauma and should be considered as a type of traumatic PM rather than spontaneous PM.[1,5]

The diagnosis of neonatal PM is chiefly based on chest radiography and clinical examination. While respiratory distress is a common but not specific symptom, chest x-ray demonstrates typical features of PM. On plain chest x-ray, the classic description is that the presence of a wedge-shaped opacity extending into the superior mediastinum with its inferior pole may attach or not to the hemidiaphragm, representing the thymus lifted off the heart by underlying air, also known as "Spinnaker sail" sign.[10,11] The other signs on anteroposterior projection include the linear bands of mediastinal air paralleling the border of the heart, continuous diaphragm sign representing interposition of gas between the pericardium and the diaphragm which should be distinguished with pneumopericardium. In certain cases, a thin linear opacity connecting the inferior pole of the displaced thymus to the midportion of the heart or to the hemidiaphragm may be seen which is corresponding to connective tissue forming a fascia that envelopes the thymus and merges with the fibrous pericardium. On left lateral projection, mediastinal air may be seen as a radiolucent air collection in retrosternal area. Although detectability of aberrant air of the lateral radiography is more sensitive than the plain radiography, it may be confused with pneumothorax. Therefore, in certain cases, combination anteroposterior and lateral projections is necessary to give a definitive diagnosis.[12,13] In some atypical circumstances, obtaining a computerized tomography is also required. According to the paper of Jung

et al, sonography is feasible in detecting pneumomediastinum however cannot replace chest radiography.[14]

In general, neonatal PM is a benign condition which is self-limiting. Close observation and monitoring are obligatory and may be the only essential measure. Repeat radiography is required to reevaluate the pneumomediastinum. Depending on the severity of clinical symptoms, drainage and/ or mechanical ventilation, including high-frequency oscillatory ventilation, is needed. However, we should avoid, if possible, performing the invasive procedure which could bring more drawbacks rather than benefits. An oxygen-rich environment can be considered for the term infant to attempt nitro washout if the pneumomediastinum is significant.[15]

IV. CONCLUSION

Neonatal pneumomediastinum is a rare entity, particularly in term newborns that were born by uneventful vaginal delivery. Respiratory distress is a common manifestation of pneumomediastinum. Generally, chest radiographs obtained in two projections is adequate to give the definitive diagnosis. Close observation clinically and radiologically is the choice of treatment due to the risk of developing pneumothorax, subcutaneous emphysema or cardiac compromise.

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