Journal of Kerman University of Medical Sciences

JKMU

Journal of Kerman University of Medical Sciences, 2020; 27 (3): 270-276

Neonatal Pneumomediastinum, Spinnaker-Sail Sign: A Case Report and Review of the Literature

Yazdan ghandi, M.D. [1], Shima zargar, M.D.[2], Danial habibi, M.Sc. [3]

1- Associate Professor, Amir Kabir Hospital, School of Medicine, Department of Pediatrics, Pediatric Cardiology, Arak University of Medical Sciences, Arak, Iran (Corresponding author; E-mail: y.ghandi@arakmu.ac.ir)

2- Amir Kabir Hospital, School of Medicine, Arak University of Medical Sciences, Arak, Iran

3- PhD student, Department of biostatistics, Arak University of Medical Sciences

Received: 10 October, 2018 Accepted: 27 May, 2020

ARTICLE INFO

Article type:
Case Report

Keywords:
Air leak syndrome
Chest Radiograph
Neonate
Preterm
Pneumomediastinum
Spinnaker-Sail sign

Abstract

Background: Pneumomediastinum (PM) occurring in approximately 0.1% of neonates is an asymptomatic accumulation of free air in the mediastinum. In this paper, we report on a preterm newborn experiencing a significant spontaneous pneumomediastinum subsequent to an uneventful vaginal delivery in the absence of resuscitation maneuvers.

Case presentation: A-2560 g-male newborn was delivered vaginally to a 30-year-old mother at 34 weeks' gestation following an uneventful twin pregnancy. Physical examination was unremarkable except for the presence of respiratory symptoms including dyspnea and oxygen desaturation (O_2Sat<85%) which required nasal continuous positive airway pressure (nCPAP). Over the ensuing day, RDS was suspected and considering the possibility of secondary surfactant insufficiency, surfactant therapy was performed. A chest X-ray obtained after 24 hours was compatible with a pneumomediastinum without subcutaneous emphysema. By 96 hours of age, his respiratory distress began to resolve. Chest x-ray films obtained at the 5th day of birth showed complete reabsorption of the pneumomediastinum. Arterial blood gas measurements showed an acceptable level of gas exchange. Blood cultures taken at the referral hospital were sterile after 48 hours of incubation. He was eventually discharged at good condition after 13 days. This paper outlines the fundamentals of diagnosis and management of a pneumomediastinum in the neonate.

Conclusion: In conclusion, spontaneous pneumomediastinum is a rare, benign condition which is usually over-diagnosed and over-treated. Conservative therapy with respiratory support and careful observation in the NICU are recommended until spontaneous resolution of the condition.

Introduction

Pneumomediastinum (PM), aka mediastinal emphysema, is the accumulation of free air in the mediastinum primarily described by Laennec in 1819 subsequent to trauma (1). Pneumomediastinum (PM) occurring in approximately 0.1% of neonates is an asymptomatic accumulation of free air in the mediastinum which is usually underestimated (2). Pulmonary air leak syndrome, mostly prevalent on the third day of life, has been reported to affect 1-2% of all neonates; however, it roughly affects half of newborns on mechanical ventilation (3,4).

Pneumomediastinum succeeding birth in the absence of traumatic injury or resuscitation maneuvers is an infrequent condition that may present with grunting, deafening of cardiac sounds, and bulging of the hemi-thorax. The majority of pneumomediastinums in the neonatal population will resolve spontaneously without medical intervention, but despite that,

close monitoring is essential to provide prompt recognition of possible complications. In this study, a case will be presented in order to outline the clinical and radiographic features of pneumomediastinum and yield a better understanding.

Case presentation

A-2560 g-male newborn was delivered vaginally to a 30-year-old mother at 34 weeks' gestation following an uneventful twin pregnancy. No resuscitation was necessary at the delivery room, however, the baby presented with progressive respiratory distress and symptoms of tachypnea, grunting and subcostal retractions developed in the neonatal intensive care unit due to RDS. Physical examination was unremarkable except for the presence of respiratory symptoms including dyspnea and oxygen desaturation (O_2Sat<85%) which required nasal continuous positive airway pressure (nCPAP). An initial chest x-ray revealed bilateral reticulonodular densities and the arterial blood gas analysis revealed mild respiratory and decompensated metabolic acidosis. On admission, his vital signs showed a heart rate of 160 beats per minute and respiratory rate of 80 per minute. His blood pressure was 45/22mmHg with an oxygen saturation of 85%. A white lung and air-bronchogram pattern were also found on the chest x-ray. Initially, arterial blood gases showed a PH of 7.22, a PCo2 of 59 mmHg, a Po2 of 34 mmHg and a bicarbonate of 13.8. Over the ensuing day, RDS was suspected and considering the increased work of breathing under treatment with nCPAP and possibility of secondary surfactant insufficiency, surfactant therapy was performed by the intubation-surfactant-extubation (INSURE) technique. A second chest X-ray obtained after 24 hours was compatible with a pneumomediastinum without subcutaneous emphysema (Figure 1). O2 support therapy continued with NIPPV and hood box. The patient was monitored closely over the next 24 hours. By 96 hours of age, his respiratory distress began to resolve. Chest x-ray films obtained at the 5th day of birth showed complete resorption of the pneumomediastinum. Arterial blood gas measurements showed an acceptable level of gas exchange. Blood cultures taken at the referring hospital were sterile after 48 hours of incubation. He was discharged at good condition after 13 days (Figure 2). On echocardiography, left ventricular ejection function (EF=60%) was preserved and no evidence of inferior vena cava distention or systolic or diastolic ventricular failure was found. There was no evidence of cardiovascular compromise. Therefore, in this paper we report on a preterm neonate suffering from isolated pneumomediastinum. Despite coincidence with RDS, PM was asymptomatic and eventually resolved without necessitating mechanical ventilation or mediastinal drains.

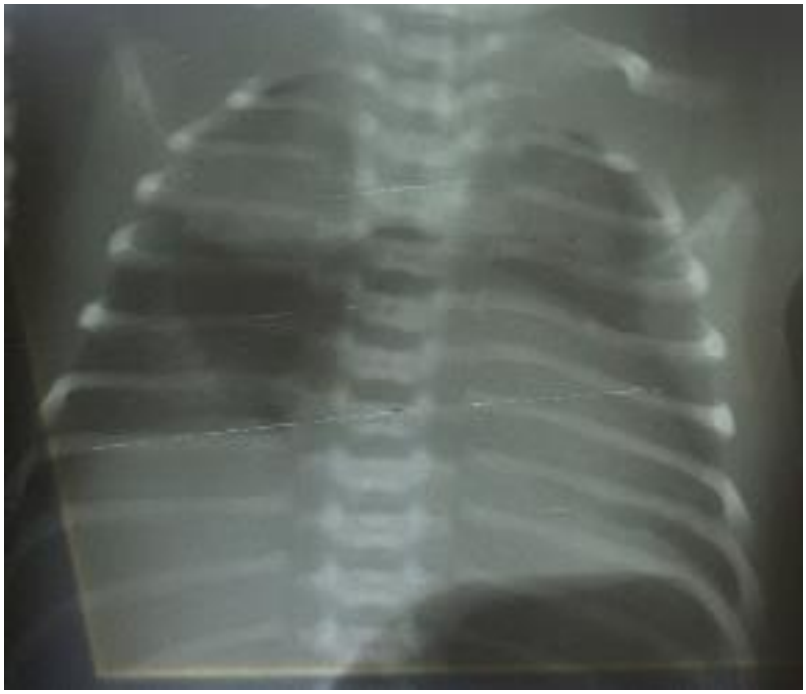

Figure 1. Anteroposterior chest radiograph consistent with a pneumomediastinum and Spinnaker-Sail sign

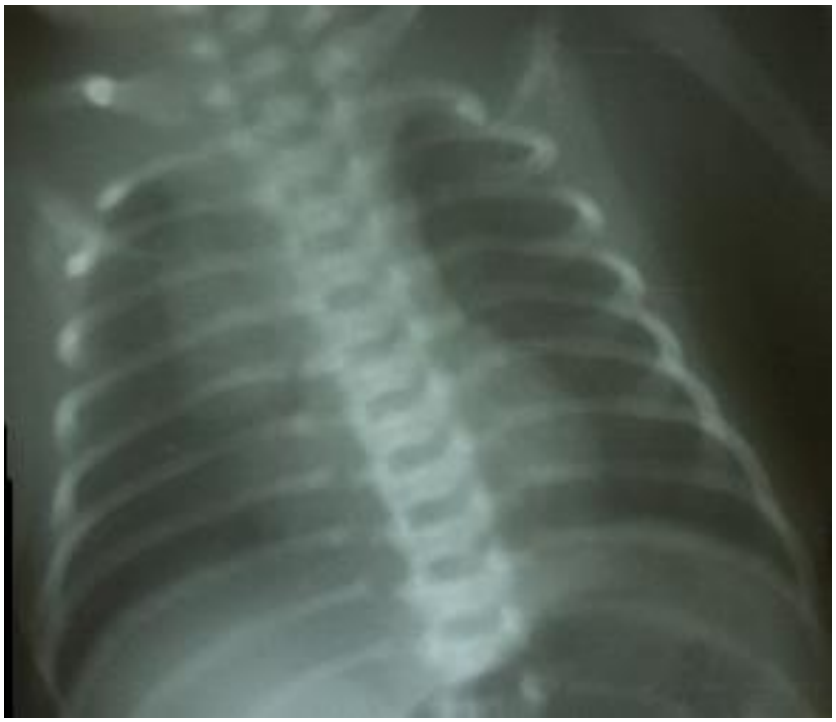

Figure 2. Chest radiograph on day 13 showing resolution of the pneumomediastinum along with residual air leak syndrome

Discussion

Pneumomediastinum is the third most common form of air leak syndrome after pulmonary interstitial emphysema and pneumothorax which occurs as a result of alveolar rupture due to over distention of the alveoli via gas retention or uneven distribution of gas. (5 -8).

Generally, predisposing factors found responsible for pneumomediastinum in neonates are prematurity, low Apgar score, infections, pneumonia or meconium aspiration syndrome, hyaline membrane disease, airway obstruction, Valsalva maneuvers, positive pressure ventilation, mechanical ventilation and traumatic injuries during labor (8). It must be noted that, diminished lung compliance and copious perivascular connective tissue in preterm infants increase the incidence of pulmonary air leak (8). in Previous study, it showed that a sequence of events is known responsible for spontaneous air leak that begin with an aspiration of particles i.e. meconium, followed by vigorous, high-pressured breaths directly after birth increasing alveolar pressure and provoking alveolar rupture (9).

In a study conducted by Hauri-Hohl et al. (10) nine neonates including two premature and seven term infants suffering from PM were evaluated who were all robust at birth, hence, no resuscitation with bag mask ventilation or surfactant was applied. Birth weight ranged from 2,150 g to 4,140 g (mean 3,340 g). Five children were delivered vaginally and four by caesarean section. All neonates presented signs and symptoms of respiratory distress. The diagnosis of PM was confirmed by chest X-ray in the intensive care unit; however, three neonates were also diagnosed with a Pneumopericardium (PP), one with subcutaneous emphysema (SE) and five with a Pneumothorax (PT). All children received oxygen therapy and specific therapy for their underlying disease. One neonate was admitted to the intensive care unit due to convulsions and developed a PM on day six of life. The attending physicians found the Valsalva maneuver occurring during the convulsion responsible for the PM. Overall, four newborns developed PM due to an identifiable mechanical etiology including mechanical ventilation with high inspiratory pressure, CPAP, pulmonary infection and convulsion, in contrast, five neonates were diagnosed with spontaneous PM. Neonates stayed in the intensive care unit for 3–13 days (mean 5.6 day) until clinical and radiographic improvement was established. It was finally concluded that PM in neonates had a good prognosis and no complications due to air trapping was found. The need for further research to identify the etiology of spontaneous PM in robust, term neonates was also emphasized (10).

This clinical report also highlights the fundamental role of chest radiography as the only imaging modality required to establish the diagnosis of pneumomediastinum. Basically, an anteroposterior (AP) projection is adequate; however, a cross-table lateral x-ray or a lateral decubitus may be obtained if uncertainty remains or a pneumothorax is not yet excluded (11).

PM may have diverse presentations including the continuous diaphragm sign (entrapment of air between the pericardium and the diaphragm becomes evident beneath the cardiac silhouette) and linear collections of air aligning the left side of the heart and the descending aorta extending superiorly beyond the mediastinum along the great vessels into the neck. In infants, the “Spinnaker Sail sign” or “Angel Wing Sign” resembles the headsail of a boat and is a rare, albeit pathognomonic sign of pneumomediastinum; it is an upwards and outwards deviation of one of the two thymic lobes due to

the triangular-shaped collection of air in the mediastinum and the resultant gas pressure (12-14).

A pneumothorax can be distinguished from a pneumomediastinum by the side-lying positions since air elevates in a PT and delineates the lung border outside of the mediastinum; however, in a PM air remains confined to the mediastinal cavity. In the AP view, a halo lucency may be seen surrounding the heart which must be distinguished from a pneumopericardium in which air completely encloses the heart, outlines the inferior cardiac surface and remains confined to the anatomic limits of the pericardium (14,15).

Considering the gradual, imperceptible course of PM, the condition is asymptomatic in a majority of subjects; however, in other patients the condition may be accompanied by symptoms of respiratory distress. An isolated pneumomediastinum is usually self-limited with minimal complications (15). A pneumomediastinum with respiratory distress varies in severity according to the size of the PM and presence of concomitant lung pathologies. Therefore, the therapeutic management of a symptomatic PM depends on clinical findings to a great extent and treatment is centered on respiratory support until it spontaneously resolves.

Generally, conservative therapy with supplemental oxygen at 100 percent FiO_2 and close follow-up with regular exams, careful monitoring, and serial chest x-rays are recommended in the NICU until spontaneous resolution of the PM, because the pneumomediastinum has the potential to advance to pneumothorax, subcutaneous, and interstitial emphysema aggravating the clinical status (16,17). T. A. Lawal et al. reported on a patient with complete resolution of his pneumomediastinum over a two-week period of conservative management (18) which was not compatible with Low et al. who reported on a female neonate with continuous respiratory distress that led to a left thoracotomy probably due to loculation (19).

In hemodynamically stable patients without respiratory distress or sepsis suffering from a stable pneumomediastinum or subcutaneous emphysema along with a subtle tracheal laceration (<1cm), conservative treatment is recommended along with airway stabilization (un-cuffed tubes distal to injury), non-oral feeding, prophylactic antibiotics and prevention of complications (20,21).

Even though no evident etiology was found responsible for the neonate presented in our study, since the pneumomediastinum occurred 18 hours subsequent to intubation, a tear in the tracheal mucosa may have possibly led to the leakage of air. Our patient was also favorably managed with the conservative approach.

Conclusion

Unfortunately, previous literature lacks adequate evidence regarding the best therapeutic approach; therefore, management of PM in the clinical setting is essentially practice-based and experience-based rather than evidence-based. It may be concluded that, spontaneous pneumomediastinum is a rare, benign condition which is usually over-diagnosed and over-treated. It is diagnosed primarily by chest X-ray. Conservative therapy with respiratory support and careful observation in the NICU are recommended until spontaneous resolution of the condition.

Conflicts of Interest

Authors declare there are no conflicts of interest.

Acknowledgments

The authors appreciate the efforts and assistance of the entire NICU staff regarding their kind cooperation.

References

1. Laënnec RTH. De l'auscultation médiate ou Traité du Diagnostic des Maladies des Poumon et du Coeur. 1st ed. Paris: Brosson & Chaudé; 1819.
2. Corsini I, Dani C. Clinical management of the neonatal pneumomediastinum. Acta Biomed 2014; 85(1):39-41.
3. Wariki WM, Mori R, Boo NY, Cheah IG, Fujimura M, Lee J, et al. Risk factors associated with outcomes of very low birthweight infants in four Asian countries. J Paediatr Child Health 2013; 49(1):E23-7.
4. Lim HS, Kim H, Jin JY, Shin YL, Park JO, Kim CH, et al. Characteristics of pneumothorax in a neonatal intensive care unit. J Korean Soc Neonatol 2011; 18:257-64.
5. Joseph LJ, Bromiker R, Toker O, Schimmel MS, Goldberg S, Picard E. Unilateral lung intubation for pulmonary air leak syndrome in neonates: a case series and a review of the literature. Am J Perinatol 2011; 28(2):151-6.
6. P S, J K. Preterm neonate with spontaneous pneumopericardium without any other associated air leaks. J Clin Diagn Res 2014; 8(1):181-2.
7. Malek A, Afzali N, Meshkat M, Yazdi NH. Pneumothorax after mechanical ventilation in newborns. Iran J Pediatr 2011; 21(1):45-50.
8. Bas AY, Okumus N, Demirel N, Zenciroglu A. Persistent pulmonary interstitial emphysema in a preterm infant. Indian Pediatr 2008; 45(9):775-7.
9. Kelly S, Hughes S, Nixon S, Paterson-Brown S. Spontaneous pneumomediastinum (Hamman's syndrome). Journal of the Royal Colleges of Surgeons of Edinburgh and Ireland 2010; 8(2):63-6.
10. Hauri-Hohl A, Baenziger O, Frey B. Pneumomediastinum in the neonatal and paediatric intensive care unit. Eur J Pediatr 2008; 167:415-8.
11. Chalumeau M, Le Clainche L, Sayeg N, Sannier N, Michel JL, Marianowski R, et al. Spontaneous pneumomediastinum in children. Pediatr Pulmonol 2001; 31(1):67-75.
12. Correia-Pinto J, Henriques-Coelho T. Images in clinical medicine. Neonatal pneumomediastinum and the spinnaker-sail sign. N Engl J Med 2010; 363(22):2145.
13. Monteiro R, Paulos L, Agro Jd, Winckler L. Neonatal spontaneous pneumomediastinum and the Spinnaker-Sail sign. Einstein (Sao Paulo) 2015; 13(4):642-3.
14. Alves ND, Sousa M. Images in pediatrics: the thymic sail sign and thymic wave sign. Eur J Pediatr 2013; 172(1):133.
15. Silfeler I, Kurnaz H, Acar Y, Arica V, Tutanc M, Pekun F. Spontaneous pneumomediastinum associated with subcutaneous emphysema causing brachial plexus palsy in a term newborn. Pak J Med Sci 2011; 27(1):190-2.
16. Kyle A, Veldtman G, Stanton M, Weeden D, Baral V. Barotrauma-associated posterior tension pneumomediastinum, a rare cause of cardiac tamponade in a ventilated neonate: a case report

and review of the literature. Acta Paediatr 2012; 101(3):e142-4.

17. Rocha G, Guimarães H. Spontaneous pneumomediastinum in a term neonate – case report. Clin Case Rep 2017; 6(2):314-6.

18. Lawal TA, Glüer S, Reismann M, Dördelmann M, Schirg E, Ure B. Spontaneous neonatal pneumomediastinum: the "spinnaker sail" sign. Eur J Pediatr Surg 2009; 19(1):50-2.

19. Low AS, Tan-Kendrick AP, Loh M, Chui CH. Spontaneous multilocu-lated multiseptated pneumomediastinum in a newborn baby: thespinnaker sail is rigged – CT features with pathologic correlation. Pediatr Radiol 2003; 33(10):712-5.

20. Schuman TA, Jacobs B, Walsh W, Goudy SL. Iatrogenic perinatal pharyngoesophageal injury: a disease of prematurity. Int J Pediatr Otorhinolaryngol 2010; 74(4):393-7.

21. Zandvoort HJ, Kroon HM, Schuit SC, Wijnhoven BP. Pneumomediastinum: intervention is not always necessary. Ned Tijdschr Geneeskd 2015; 159:A9379.