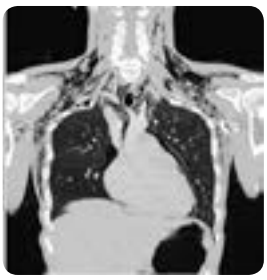

SPONTANEOUS PNEUMOMEDIASTINUM: CASE SERIES

Neumomediastino espontáneo: una serie de casos

Tatiana Suárez Poveda[1]
Johan Sebastián Lopera Valle[2]
Antonella Arrieta Rojano[2]
Vanessa García Gómez[3]

Key words (MeSH)
Pneumomediastinum
Diagnostic
Mediastinal emphysema
Tomography, X-ray computed

Palabras clave (DeCS)
Neumomediastino
Enfisema mediastínico
Tomografía computarizada por rayos X

Summary

Spontaneous pneumomediastinum, a clinical condition with low incidence, is characterized by the presence of free air in the mediastinum without associated trauma, and it is considered a benign and self-limiting disorder. The diagnosis is made by clinical suspicion and is confirmed with diagnostic images. This entity has been little reported in the national literature, considering that the probable underdiagnosis is due to lack of knowledge of its clinical and epidemiological characteristics in our environment. A series of 11 cases of spontaneous pneumomediastinum in patients admitted to two institutions of high complexity in Medellin-Colombia are presented.

Resumen

El neumomediastino espontáneo es una condición clínica de baja incidencia. Se caracteriza por tener aire libre en el mediastino sin trauma asociado, y se considera un trastorno benigno y autolimitado. El diagnóstico se realiza por sospecha clínica y se confirma con imágenes diagnósticas. Esta entidad ha sido poco informada en la literatura nacional, y se considera que el probable subdiagnóstico se debe al poco conocimiento de sus características clínicas y epidemiológicas en nuestro medio. Se presenta una serie de 11 casos de neumomediastino espontáneo estudiados en dos instituciones de alta complejidad de Medellín, Colombia.

1. Introduction

Spontaneous pneumomediastinum (SPM) or Hamman syndrome, initially described by Louis Hamman in 1939 (1), is a rare clinical condition, with an incidence of 1 in 30,000 (2) to 1 in 102,000 (3,4) hospital admissions and 0.3 to 5 % of paediatric patients hospitalized for asthma attacks (5,6). It is characterized by the presence of open air in the mediastinum without associated trauma and is considered a benign and self-limited disorder (5).

Physiopathologically, it is characterized by overdistension and an increase in intra-alveolar pressure, with or without a decrease in perivascular pressure (5). This favours the rupture of the marginal alveoli and the passage of air towards the mediastinum along the interstitium and vascular support tissues during the respiratory cycle, as a compensatory mechanism for pressure gradients (5-8).

It is more frequent in young males, with a history of smoking, recent respiratory infection and asthmatic crisis. The commonly described triggering events are intense physical exercise, severe paroxysmal cough and narcotic inhalation. The diagnosis is made on clinical suspicion and is confirmed with diagnostic images, once potentially fatal causes of pneumomediastinum such as esophageal perforation, airway injury or mediastinal infection have been ruled out (3). Although chest radiography represents the initial diagnostic aid, approximately 30 % of cases are subdiagnosed by this imaging method (5,9), and up to 50 % of cases are subdiagnosed when only the fromtal projection is performed (10,11).

[1]Radiologist, subspecialist in Trauma and Emergency Radiology. Universidad de Antioquia. Medellín, Colombia.

[2]Resident Radiologist. Universidad de Antioquia. Medellín, Colombia.

[3]Radiologist, subspecialist in Body Image. Universidad de Antioquia. Medellín, Colombia.

Name of the department and institution to which the work is attributed: Hospital Universitario San Vicente Fundación and Hospital Pablo Tobón Uribe.

This entity has been little described in the national literature, and it is the probable sub-diagnosis that significantly limits the knowledge of its clinical and epidemiological characteristics in our environment (11). For this reason, the following is a series of cases admitted to two highly complex institutions in Medellín, Colombia.

2. Case Series

This series consists of 11 cases, seven men and four women, with a median age of 15 years (range 3 months-32 years). Most of the patients had a history of asthma or recurrent broncho-obstructive syndrome, and three of them referred to the use of tobacco, marijuana or cocaine at the time of consultation (Table 1).

The mean number of days of evolution of the clinical picture prior to hospital admission was 1.8 (range 1-5), the most frequently reported symptoms being dyspnea and cough. Five patients had a general clinical aspect of regular to bad at the time of admission, with decreased oxygen saturation, tachycardia and tachypnea in most of them. Pulmonary auscultation was found hypoventilation and wheezing in seven of the patients, and in only three was emphysema documented subcutaneous to physical examination (Table 1).

With respect to blood chemistry studies, three patients had altered arterial gases, four had leukocytosis with neutrophilia, with a median C-reactive protein of 2 mg/dl (range 0.3-6.6). The median hospital stay was 6.5 days (range 2-38) and two of the 11 patients required transfer to an intensive care or special care unit. In terms of management, all patients received conservative (medical) treatment, with the exception of one who required bilateral thoracostomy and ventilatory support due to critical clinical condition.

The most frequent etiologic diagnosis was broncho-obstructive crisis in eight cases, three of which had associated pneumonia. The remaining three patients were diagnosed with parotid pneumocele, spontaneous pneumomediastinum secondary to emesis and associated with cocaine use.

All patients underwent radiography and computed tomography (CT), in which air was evidenced dissecting the mediastinum, mucosal pharyngeal spaces, carotid, for and prevertebral, even with involvement of the parotid space and the spinal canal, with varying degrees of extension and severity (Figures 1-3).

3. Discussion

Spontaneous pneumomediastinum is a rare condition with a frequency of 0.001 % to 0.01 % of all adult hospitalized patients; however, because it usually presents with mild symptoms and is self-limiting, it is likely to be subdiagnosed (12-14). Simmons first reported a case of postpartum subcutaneous emphysema in 1783 (15); however, until 1939 Hamman published the first cases of spontaneous pneumomediastinum in previously healthy young men (1,11). In 1944, Macklin and Macklin described the increase in the alveolo-interstitial pressure gradient precipitating air escape to the pulmonary interstitium, with progression to the mediastinum, as a physiopathological mechanism of this entity (Macklin effect) (16).

This effect occurs more frequently in young patients due to the elasticity and laxity of the interstitium, which is why spontaneous pneumomediastinum is not common in people older than 60 years (11,16).

Risk factors include young people (18-25 years) (12), males (17), with a history of smoking, recent respiratory infection (18), asthma and bronchial hyperreactivity, and interstitial lung disease (3). In studies conducted in Korea (19) and Portugal (3), the most frequent factors found were in order: active smoking, respiratory infection and asthma. A review that included 600 patients with SPM and lung disease. previously described asthma as the main associated factor, followed by chronic interstitial or obstructive pulmonary disease (20).

Table 1. Clinical characteristics of patients

Background	*n*
Asthma	7
Marijuana/tobacco use	2
Cocaine use	1
Bronchopulmonary dysplasia	1
Cystic fibrosis	1
Eating disorder	1
Symptoms	*n*
Dyspnea	10
Cough	10
Chest pain	8
Cervical pain	2
Signs	
General	*n*
Good.	6
Regular	4
Bad	1
Pulse	*n*
Tachycardia	10
Normal	1
Respiratory frequency	*n*
Tachypnea	9
Normal	2
Oxygen saturation	*n*
Desaturation	9
Normal	2
Pulmonary Auscultation	*n*
Hypoventilation	7
Wheezing	7
Roncus	3

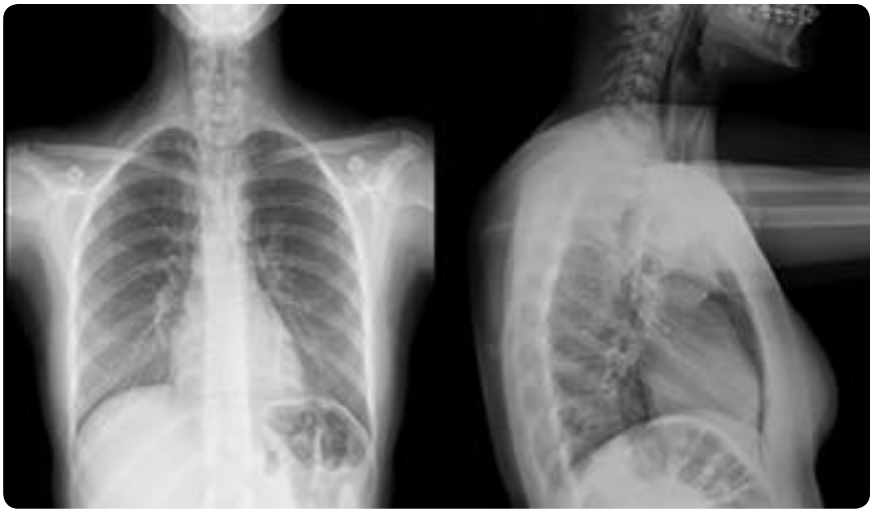

Figure 1. 15-year-old patient with pneumomediastinum secondary to episode of emesis. Frontal and lateral chest radiography, in which pneumomediastinum, air dissecting the retrotracheal space and right subcutaneous cervical emphysema are evidenced.

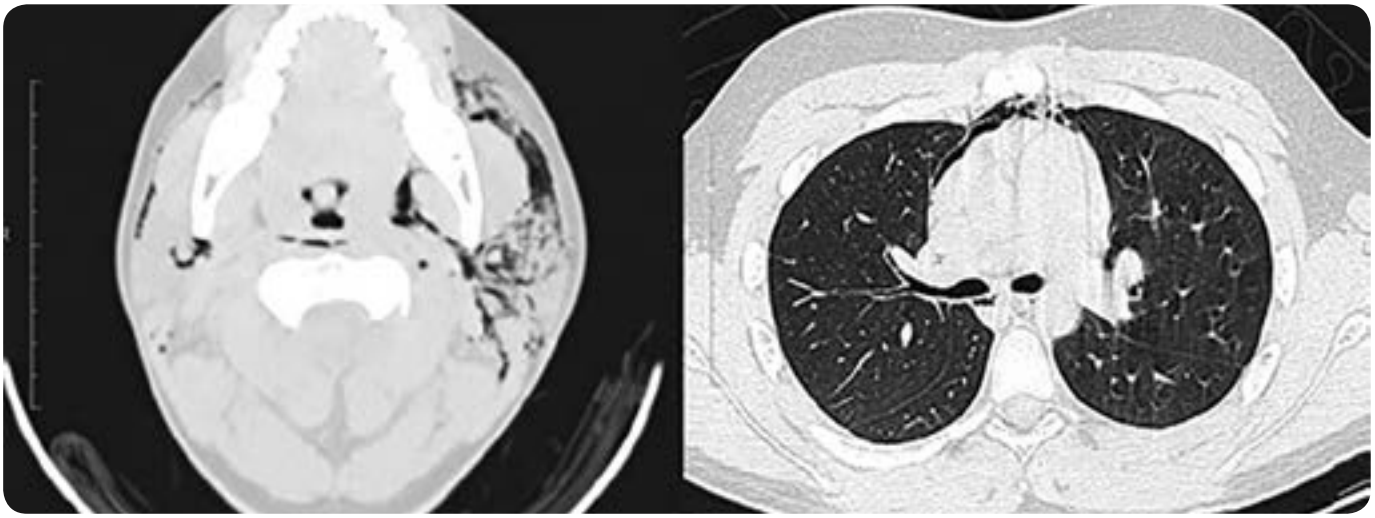

Figure 2. 14-year-old patient with parotid pneumocele and associated pneumomediastinum. Axial CT cuts of neck and thorax: subcutaneous emphysema and air dissecting the parotid, parapharyngeal, mucosal and prevertebral pharyngeal spaces are observed, as well as air in the anterior and middle mediastinum.

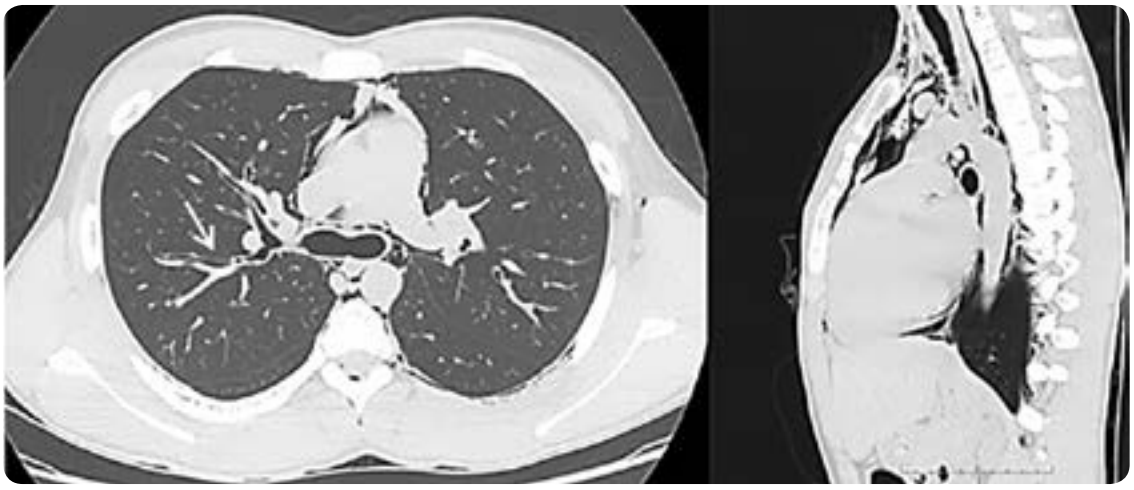

Figure 3. 16-year-old patient with severe asthmatic crisis. Axial and sagittal cut of chest CT: air is identified that dissects the perivascular and peribronchial spaces (Macklin effect), which extends towards the mediastinum and affects the acygoesophageal space, descending aorta, acygos vein, pre-vertebral and paravertebral space, as well as the medullary canal

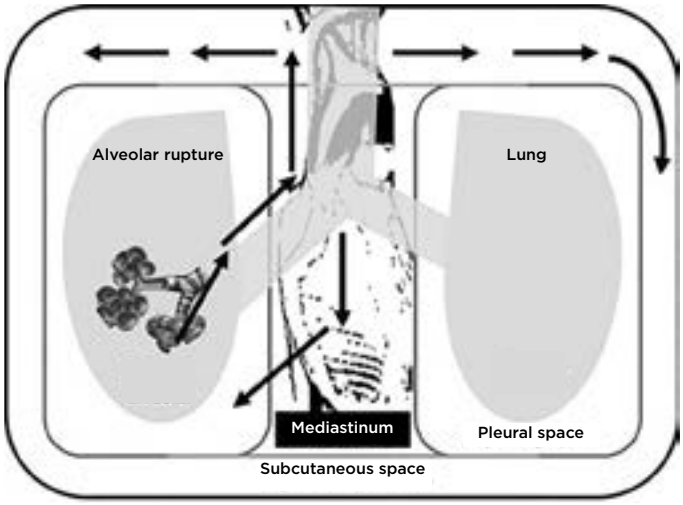

Figure 4. Scheme of the physiopathology of the Macklin effect. Overdistension and increased intra-alveolar pressure favour the rupture of marginal alveoli and the passage of air towards the mediastinum along the interstitial tissues and vascular support during the respiratory cycle, as a compensatory mechanism for pressure gradients.

Source: own elaboration.

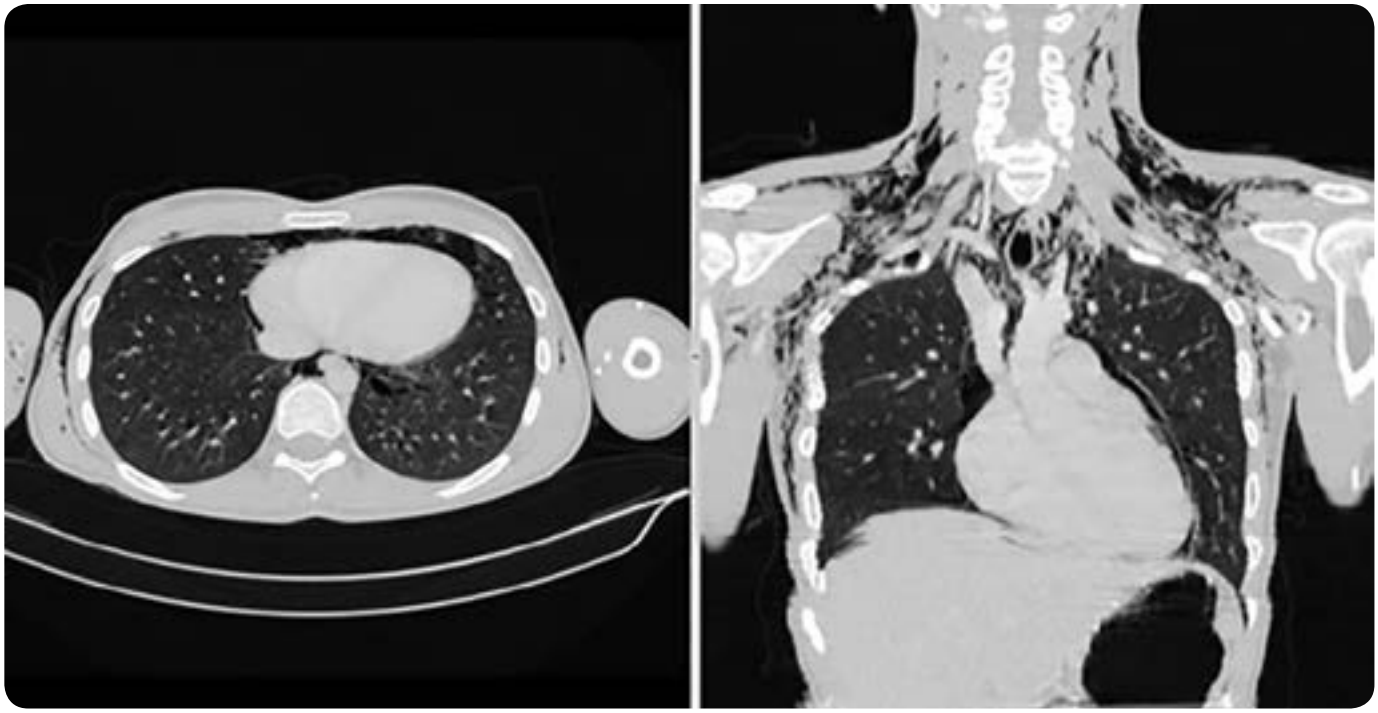

Figure 5. A 32-year-old patient with a history of marijuana and cocaine use. Axial and coronal cut of chest CT in which the Macklin effect is observed, with air extending towards the mediastinum and the cervix-thoracic subcutaneous space.

Thus, and in accordance with what has been reported in the literature, the majority of patients in this series of cases were young men, with a median age of 15 years and a history of recurrent obstructive bronchial syndrome.

The triggering events described in the literature are intense physical exercise (12,21), labor (22), pulmonary barotrauma, deep diving, severe paroxysmal cough, emesis, asthma crisis, narcotic inhalation, singing (12,21), and decompensation of connective tissue diseases (7). The frequency of these varies between the different series and in 11 to 34 % of cases it is not possible to individualize the precipitating factor (20). A systematic review evaluating 27 studies identified physical activity as the most frequently related factor, followed by inhaled drug use, cough episodes, asthma exacerbation and emesis (20). Mondello and collaborators, for their part, described a series of 18 cases in Italy, of which 12 were associated with coughing spells and 6 with physical activity (23), findings that are similar to those recorded in this article, in which 8 of the 11 cases had as a trigger broncho-obstructive crisis clinically characterized by cough and dyspnea.

It should be suspected, in particular, in individuals with suboptimal response to medical management and persistent respiratory distress with or without hypoxemia (5), after ruling out pneumomediastinum secondary to trauma and infections by gas-producing microorganisms (12,21). The usual symptoms are chest pain (up to 61 % of cases), dyspnea (41 %), cough, cervical pain, dysphagia and odynophagia (3,17,19,20), which are usually accompanied by subcutaneous emphysema (40 %) in the cervical-thoracic region (20). Some authors describe

wheezing associated with cervical pain as key to early diagnosis (24), and neck edema associated with voice changes has been described as unusual clinical signs (25). Similar to what has been found in the literature, the symptoms most frequently reported by the 11 patients in this series were: dyspnea, cough, chest pain and cervical pain, and desaturation, tachycardia and wheezing on physical examination, the most frequently observed signs.

Conventional chest radiography is the initial study in the evaluation of patients with clinical suspicion of SPM (Figure 1). This method has a good correlation with CT in moderate to severe cases, but in mild cases can be normal up to 30 % (9). The most common findings observed in chest radiography are: the thymic candle sign, air surrounding the pulmonary artery or any of its branches (ring sign), air surrounding the main branches of the aorta, double bronchial wall, continuous diaphragm sign, and subcutaneous emphysema (26).

In the tomographic study, the Macklin effect is observed as linear air collections dissecting the peribroncovascular interstitium, which extends towards the hilum and mediastinum, is observed more frequently in the hiliary region than in the periphery, and according to Sakai and collaborators (27), is found in up to 89 % of patients with spontaneous pneumomediastinum (Figures 4 and 5). As mentioned, the tomographic study of the patients in this series showed air dissecting the mediastinum and adjacent structures, with variable degrees of extension and severity.

Spontaneous pneumomediastinum tends to disappear naturally, and outpatient follow-up of stable patients is recommended (28). Hospitalization in an intensive care unit is reserved only for haemodynamically unstable patients and administration of antibiotic therapy is not indicated (29). Similarly, long-term clinical or imaging follow-up is not recommended because of the low rate of reported recurrence (28). All patients in this study were conservatively managed and only two required intensive care unit stay for critical clinical condition.

4. Conclusion

Spontaneous pneumomediastinum is a low-incidence clinical condition and is most common in young males with a history of smoking and asthma attacks. Images are the cornerstone in diagnosis and their course is generally self-limited. The 11 cases described in this article share mostly epidemiological and clinical characteristics previously reported in the literature; however, cases of atypical presentation may represent a diagnostic and therapeutic challenge.

References

1. Hamman L. Spontaneous mediastinal emphysema. Bull Johns Hopkins Hosp. 1939;64(1):1-21.
2. Lopes F, Marchiori E, Zanetti G, et al. Pneumomediastino espontâneo após esforço vocal: relato de caso. Radiol Bras. 2010;43:137-9.
3. Dionísio P, Martins L, Moreira S, et al. Spontaneous pneumomediastinum: Experience in 18 patients during the last 12 years. J Bras Pneumol. 2017;43(2):101-5.
4. Bedolla-Pulido TR, Bedolla-Barajas M. Spontaneous pneumomediastinum and subcutaneous emphysema associated with bronchospasm in a woman with no history of asthma. Rev Alerg Mex. 2017;64(3):386-9.
5. Vianello A, Caminati M, Chieco-Bianchi F, et al. Spontaneous pneumomediastinum complicating severe acute asthma exacerbation in adult patients. J Asthma. 2017:1-7.
6. Tortajada-Girbés M, Moreno-Prat M, Ainsa-Laguna D, et al. Spontaneous pneumomediastinum and subcutaneous emphysema as a complication of asthma in children: case report and literature review. Ther Adv Respir Dis. 2016;10:402-9.
7. Allaoui A, Aboudib F, Bouissar W, et al. Spontaneous pneumomediastinum: A rare complication of dermatomyositis. Rev Pneumol Clin. 2017;73(5):258-62.
8. Carzolio-Trujillo HA, Navarro-Tovar F, Padilla-Gómez CI, et al. Trauma contuso de tórax con neumomediastino y neumoperitoneo secundario a efecto Macklin. Reporte de un caso. Cir Cir. 2016;84(5):409-14.
9. Kaneki T, Kubo K, Kawashima A, et al. Spontaneous pneumomediastinum in 33 patients: yield of chest computed tomography for the diagnosis of the mild type. Respiration. 2000;67:408-11.
10. Iyer VN, Joshi AY, Ryu JH. Spontaneous pneumomediastinum: analysis of 62 consecutive adult patients. Mayo Clin Proc. 2009; 84:417-21.
11. Campbell S, Vargas S, Gómez J, et al. Síndrome de Hamman. Acta Med Colomb. 2016;41(3):206-10.
12. Spotts PH. Spontaneous pneumomediastinum: Case presentation to a college student health clinic. J Am Coll Health. 2017;65(8):575-8.
13. Takada K, Matsumoto S, Hiramatsu T, et al. Spontaneous pneumomediastinum: an algorithm for diagnosis and management. Ther Adv Respir Dis. 2009;3(6):301-7.
14. Kim SH, Huh J, Song J, et al. Spontaneous pneumomediastinum: A rare disease associated with chest pain in adolescents. Yonsei Med J. 2015;56(5):1437-42.
15. Simmons ST. A case of emphysema brought on by severe labor pains. Lond Med Commun. 1783;1:176.
16. Macklin MT, Macklin CC. Malignant interstitial emphysema of the lungs and mediastinum as an important occult complication in many respiratory diseases and other conditions: an interpretation of the clinical literature in the light of laboratory experiment. Medicine. 1944;23:281-358.
17. Kim KS, Jeon HW, Moon Y, et al. Clinical experience of spontaneous pneumomediastinum: diagnosis and treatment. J Thorac Dis. 2015;7(10):1817-24.
18. Huang L, Chen H, Peng S. Spontaneous pneumomediastinum, emphysema, and pulmonary bullae associated with refractory Mycoplasma pneumoniae pneumonia in a child. Pediatr Pulmonol. 2017;52(10):E77-E80.
19. Park SJ, Park JY, Jung J, et al. Clinical manifestations of spontaneous pneumomediastinum. Korean J Thorac Cardiovasc Surg. 2016;49(4):287-91.
20. Dajer-Fadel WL, Argüero-Sánchez R, Ibarra-Pérez C, et al. Systematic review of spontaneous pneumomediastinum: A survey of 22 years data. Asian Cardiovasc Thorac Ann. 2014;22(8):997-1002.
21. Okada M, Adachi H, Shibuya Y, et al. Diagnosis and treatment of patients with spontaneous pneumomediastinum. Respir Investig. 2014;52(1):36-40.
22. Nagarajan DB, Ratwatte MD, Mathews J, et al. Intrapartum spontaneous pneumomediastinum and surgical emphysema (Hamman's syndrome) in a 30-year-old woman with asthma. BMJ Case Rep. 2017;2017.
23. Mondello B, Pavia R, Ruggeri P, et al. Spontaneous pneumomediastinum: experience in 18 adult patients. Lung. 2007;185(1):9-14.
24. Yurtseven A, Saz EU. Red flag; wheezing with neck pain may be a clue to the early diagnosis of spontaneous pneumomediastinum. Tuberk Toraks. 2017;65(2):146-9.
25. Potu KC, Gedela M, Shaikh KA, et al. An unusual presentation of an unusual disease: Spontaneous pneumomediastinum. S D Med. 2016;69(11):495-7.
26. Zylak CM, Standen JR, Barnes GR, et al. Pneumomediastinum revisited. RadioGraphics. 2000;20(4):1043–57.
27. Sakai M, Murayama S, Gibo M, et al. Frequent cause of the Macklin effect in spontaneous pneumomediastinum: demonstration by multidetector-row computed tomography. J Comput Assist Tomogr. 2006;30(1):92–4.
28. Freixinet J, García F, Rodríguez PM, et al. Spontaneous pneumomediastinum long-term follow-up. Respir Med. 2005;99(9):1160–3.
29. Al-Mufarrej F, Badar J, Gharagozloo F, et al. Spontaneous pneumomediastinum: diagnostic and therapeutic interventions. J Cardiothorac Surg. 2008;3:59–62.

Correspondence

Tatiana Suárez Poveda
Calle 64 # 51D-154
Bloque 12, Departamento de Radiología
Medellín, Colombia
tatisuarez@hotmail.com

Received for assessment: November 18, 2018
Accepted for publication: May 30, 2019