Case report

Hamman's syndrome (spontaneous pneumomediastinum presenting as subcutaneous emphysema): A rare case of the emergency department and review of the literature

Konstantinos Grapatsas, Zoi Tsilogianni, Vasileios Leivaditis, Sotirios Kotoulas, Christoforos Kotoulas, Efstratios Koletsis, Ilias Stylianos Iliadis, Manfred Dahm, Anastasios Kallianos, Haidong Huang, Christoforos Kosmidis, Michael Karanikas, Vasilis Thomaidis, Konstantinos Porpodis, Paul Zarogoulidis

1. Introduction

Pneumomediastinum (PM) is a clinical entity that is characterized as the existence of air in the anatomical space of mediastinum. This clinical entity can be characterized as spontaneous or secondary. Spontaneous pneumomediastinum (SPM) (Hamman's syndrome) is a rare medical case that concerns the emergency department's personnel [1]. We present a case of a young man that presented to our hospital's emergency department with SPM along with subcutaneous emphysema.

2. Case report

2.1. Medical history

A male 24 year old caucasian presented in our hospital's emergency department with a cervical subcutaneous emphysema. He denied having any sort of dyspnea. He reported that the subcutaneous emphysema preexisted 2 hours before. The patient had cough 2 weeks before. In the patient's medical history there was no known disease. The patient was under no medication.

2.2. Diagnostic assessment

In the chest x-ray the subcutaneous emphysema was confirmed. Due
to increased suspicion of pneumothorax a thorax-computed tomography was performed (CT) (Figs. 1 and 2). This revealed the subcutaneous emphysema and the PM while pneumothorax was excluded. During his stay in the emergency department the patient was in stable cardiorespiratory status. A bronchoscopy and an esophagoscopy that were performed showed no pathology. Figs. 3 and 4. The laboratory control showed: Leukocytes 13,000, Hemoglobin: 14.3 g/dl, Hematocrit: 38.0%, Sodium: 141 mmol/L, Potassium: 4.0 mmol/L, Creatinine 1.1 mg/dl, CRP: 46 mg/dl. Heart rate: 87/min, Blood pressure: 97/61 mmHg. The electrocardiography showed no alterations and a sinus rhythm.

2.3. Procedure

The patient was admitted in the cardiothoracic department's intermediate care unit precautionary for 24 hour observation. The next day the subcutaneous emphysema had been greatly reduced and the patient was transferred to the clinic; on the same day he was discharged without medication.

A follow-up was suggested. On the second day after the hospital discharge the subcutaneous emphysema had further subsided and the patient was without symptomatic.

3. Discussion

Spontaneous Pneumomediastinum is a rare clinical entity that concerns the hospital’s emergency department personnel. It is also referred as “Hamman’s syndrome” as Louis Hamman was the first to report case series of this entity [2]. It is defined by a, non-injury-cause, free air existence in the mediastinum [3]. Under increased intrathoracic pressure (as it happens when a Valsalva maneuver is performed) alveolar rupture can happen and air dissects to the interstitial space and bronchovascular sheaths and to mediastinum (The Macklin effect) [4].

Its’ incidence is less than 1:44000, maybe approximately 1/25.000 between 5 and 34 years old; while the majority of patients (70%) are males [1,5]. However, this frequency may be higher as many patients do not appear in the emergency department or are underdiagnosed attributing any mild symptoms to possible muscular pain or anxiety [1,6].

The PM is classified to two categories: spontaneous and secondary. It can be characterized as spontaneous in seemingly healthy patients as in our case. However there seem to be causes that predispose to SPM (such as asthma, COPD, interstitial diseases, tobacco use, continuous...
drug inhalation) and recent events or conditions that may precipitate its appearance (such as vomiting, cough, upper respiratory infection, constipation during defecation, labor, physical exercise, occasional illegal drug inhalation, neonate, respiratory distress syndrome and rarely balloon filling, use of air instruments and convulsions) [2,3,7]. As far as the secondary PM is concerned its cause can be due to trauma (either blunt or penetrating or due to surgery) or iatrogenic (central line application, endoscopic techniques, endotracheal tube placement or removal) [5]. However, sometimes differentiation between spontaneous and secondary with mild symptoms is difficult. So, always secondary causes should be searched and excluded in order to define an idiopathic pneumomediastinum; even if it is a tendency a benign pneumomediastinum without an obvious cause to be easily categorized as a spontaneous one [1,5,8].

The major symptom of PM is thoracic pain. Dyspnea, vomiting, fever and dysphagia have also been detected. However, this symptomatology can be absent in a SPM. As in our case the subcutaneous emphysema can be detected in 70% of cases; rhinolalia, hoarness and neck swelling can also coexist. Tachycardia, tachypnea and anxiety can also be present in the general clinical image of a patient with PM. Usually the SPM is a benign clinical entity with a good prognosis. However, a large accumulation of air in the mediastinum can lead to developing pressure on the thoracic great vessels and the trachea [1,9-11].

In the diagnostic approach of SPM a plain chest x-ray film can be diagnostic [1,12]. However, in many cases a CT is also performed if a diagnostic dilemma exists. In our case, also a CT was performed to rule out pneumothorax. The CT can also give useful information on the cause of the SPM. The performance of bronchoscopy and esophago-diastinum without an obvious cause to be easily categorized as a spontaneous one [1,9,11].

The diagnostic approach of SPM a plain chest x-ray film can be diagnostic [1,12]. However, in many cases a CT is also performed if a diagnostic dilemma exists. In our case, also a CT was performed to rule out pneumothorax. The CT can also give useful information on the cause of the SPM. The performance of bronchoscopy and esophagoscopy are rarely performed. In our case, were performed in order to exclude an underlying pathology [9,12,13].

Usually the treatment of PM, after excluding any serious causes, is conservative requiring only reassurance of the patient combined with anti-anxiety drugs, as well as providing oxygen and analgesics [1,14,15]. Hospital admission is more frequently applied with a 24-h observation as in our case [1]. Sometimes antibiotics maybe used in suspicion of mediastinitis [2,13,15]. In rare cases of compression of the great vessels or the trachea a video-assisted thoracic surgery or even a thoracotomy are needed. In an extensive subcutaneous emphysema small extent of surgical operation as skin incision, small subcutaneous drainage insertion or even chest drain tubes are needed. A follow-up is in all cases recommended after the patient's discharge [1,13,16].

In conclusion, spontaneous pneumomediastinum is a rare disease that will be treated from the clinician in the emergency department. Despite the lack of specific symptomatology the emergency department clinician should include PM in his differential diagnosis. Usually a conservative treatment is required with routine observation and follow-up. However, sometimes even life-threatening complications can occur, requiring prompt management.

Conflict of interest

None to Declare.

References

[1] V.K. Kouritita, K. Papagiannopoulos, G. Lazaridis, S. Baka, I. Mpsokovinas, V. Karavasilis, et al., Pneumomediastinum, J. Thorac. Dis. 7 (2015) S44–S49, dx.doi.org/10.3978/j.issn.2072-1439.2015.01.11.
[2] V. Perna, E. Vila, J.J. Guellbenzu, I. Amat, Pneumomediastinum: is this really a benign entity? When it can be considered as spontaneous? Our experience in 47 adult patients, Eur. J. Cardio. Thorac. Surg. Off. J. Eur. Assoc. Cardio Thorac. Surg. 37 (2010) 573–575, dx.doi.org/10.1016/j.ejcts.2009.08.002.
[3] U. Devaraj, P. Ramsachandran, G.A. D'Souza, Recurrent spontaneous pneumomediastinum in a young female: hamman's crunch revisited(dagger), Oxf. Med. Case Rep. 2014 (2014) 18–20, dx.doi.org/10.1093/omcr/omu008.
[4] T.S. Lee, K.T. Liao, Malignant pulmonary interstitial emphysema, Chest 104 (1993) 647–648.
[5] S. Sahni, S. Verma, J. Grillon, A. Esquire, P. Patel, A. Talwar, Spontaneous pneumomediastinum: time for consensus, N. Am. J. Med. Sci. 5 (2013) 460–463, http://dx.doi.org/10.4103/1947-2714.117296.
[6] T. Kaneki, K. Kubo, A. Kawashima, T. Koizumi, M. Sekiguchi, S. Sone, Spontaneous pneumomediastinum in 33 patients: yield of chest computed tomography for the diagnosis of the mild type, Respir. Int. Rev. Thorac. Dis. 67 (2000) 408–411, dx.doi.org/10.1159/000029539.
[7] K. Chouliaras, E. Bench, P. Talving, A. Strumwasser, E. Benjamin, L. Lam, et al., Pneumomediastinum following blunt trauma: worth an exhaustive workup? J. Trauma Acute Care Surg. 79 (2015) 188–192, dx.doi.org/10.1097/TA.0000000000000714 discussion 92-3.
[8] Y. Kohashi, N. Okimoto, T. Matsuhashi, R. Soejima, Comparative study of mediastinal emphysema as determined by etiology, Intern. Med. 41 (2002) 277–282.
[9] K.S. Kim, H.W. Jeon, Y. Moon, Y.D. Kim, M.I. Ahn, J.K. Park, et al., Clinical experience of spontaneous pneumomediastinum: diagnosis and treatment, J. Thorac. Dis. 7 (2015) 1817–1824, dx.doi.org/10.3978/j.issn.2072-1439.2015.10.58.
[10] F. Hogan, C. McCullough, A. Rahman, Spontaneous pneumomediastinum: an important differential in acute chest pain, BMJ Case Rep. (2014) 2014, http://dx.doi. org/10.1136/bcr-2014-207692.
[11] S.J. Park, J.Y. Park, J. Jung, S.Y. Park, Clinical manifestations of spontaneous pneumomediastinum, Kor. J. Thorac. Cardiovasc. Surg. 49 (2016) 287–291, http://dx.doi.org/10.5900/jcts.2016.49.4.287.
[12] V.N. Iyer, A.Y. Joshi, J.H. Ryu, Spontaneous pneumomediastinum: analysis of 62 consecutive adult patients, Mayo Clin. Proc. 84 (2009) 417–421, dx.doi.org/10.1016/S0025-6196(11)60560-0.
[13] K. Takada, S. Matsumoto, T. Hiramatsu, E. Kojima, M. Shizu, S. Okachi, et al., Spontaneous pneumomediastinum: an algorithm for diagnosis and management, Ther. Adv. Respir. Dis. 3 (2009) 301–307, dx.doi.org/10.1177/1753468909350888.
[14] J.B. Jougon, M. Ballester, F. Delcambre, T. Mac Bride, C.E. Drometer, J.F. Velly, Assessment of spontaneous pneumomediastinum: experience with 12 patients, Ann. Thorac. Surg. 75 (2003) 1711–1714.
[15] G.J. Koullias, D.P. Korkolis, X.J. Wang, G.L. Hammond, Current assessment and management of spontaneous pneumomediastinum: experience in 24 adult patients, Eur. J. Cardio. Thorac. Surg. Off. J. Eur. Assoc. Cardio. Thorac. Surg. 25 (2004) 852–855, http://dx.doi.org/10.1016/j.ejcts.2004.01.042.
[16] I.H. Song, S.Y. Lee, S.J. Lee, W.S. Choi, Diagnosis and treatment of spontaneous pneumomediastinum: experience at a single institution for 10 years, Gen. Thorac. Cardio. Surg. 65 (2017) 280–284, dx.doi.org/10.1007/s11748-017-0755-3.