Case Report

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Subcutaneous Emphysema and Pneumomediastinum in 2 Years Old Children with Pierre Robin Syndrome: Multi-Disciplinary Management in an Emergency Department

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Abstract

Spontaneous pneumomediastinum (SPM) is an uncommon disease defined by the presence of free air in the mediastinal cavity. Its pathogenesis is characterized by all the processes that cause a sudden increase of intrathoracic pressure. Triggering factors are: lower airway infections, asthma, esophageal rupture, foreign body aspiration and diabetic ketoacidosis. When a cause is not found, SPM is defined idiopathic. The abnormal presence of air in mediastinum can cause subcutaneous emphysema (SE) in the neck and upper part of the thorax. The aim of this report was to describe a patient with an underlying genetic disorder (Pierre Robin syndrome) and SPM with consequent SE. A multidisciplinary approach in our Accident and Emergency department was important for the right management.

Abbreviations: SPM: Spontaneous Pneumomediastinum; SE: Subcutaneous Emphysema; A&E: Accident and Emergency Department; ENT: Ear, Nose and Throat; CT: Computerized Tomography

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Introduction

Spontaneous pneumomediastinum (SPM) is an uncommon disease defined by the presence of free air in the mediastinal cavity [1-3]. Reported for the first time by Hammam [4], SPM is an uncommon disorder with an incidence less than 1:44.000 [2,5]; among emergency room visits the frequency calculated is 0.0025% [1,6,7].

Pneumomediastinum is considered spontaneous when there is no defined etiology; it is called secondary when consequent by a specific causative event, for example a trauma (post-traumatic pneumomediastinum), that is estimated as the most common known etiologic cause of pneumomediastinum [2]. The pathogenesis of SPM is characterized by all the processes that cause a sudden increase of intrathoracic pressure. Triggering factors described in the literature are: lower airway infections, asthma, esophageal rupture, foreign body aspiration and diabetic ketoacidosis. When a cause is not found, SPM is defined idiopathic [1,8-11].

In pediatric age, SPM is rare and reported as consequence of forced Valsalva's maneuver due to cough, emesis, wheezing or asthma exacerbations [1,12].

Clinical presentations of SPM include acute chest pain, dyspnea with variable entity and subcutaneous emphysema (SE). The last sign is characterized by the abnormal presence of air in the subcutaneous tissue. It occurs in 90% of patients with SPM [13] and it is generally secondary to the abnormal presence

of air in mediastinum or in the pleural cavity (pneumothorax). When massive, SE can be life threatening due to compression of respiratory system, with consequent hypoxia and hypercapnia. Clinically, SE is characterized by swallowing and crackling during skin palpation. In a case report, SE has been described as consequence of pneumothorax in respiratory syncytial virus bronchiolitis [14].

SPM is usually described with a benign course, and treated with oxygen, monitoring and analgesic, in association with antibiotics when necessary. The necessity of invasive treatments (incision and drainage or chest surgery approaches) is reserved to severe patients with, for example, tension pneumomediastinum [1,14,15].

Case Report

A 2 year old boy with Pierre Robin Syndrome referred to our Accident and Emergency (A&E) department due to the sudden insurgence, when woke up, of a voluminous neck and jugular bulge. The patient had a history of an upper respiratory infection since two days, with an irritable cough on worsening from the preview evening. An empiric antibiotic treatment was started two days before (clarithromycin).

Triaged as immediate emergency (red code) due to the suffering aspect and the important neck swelling with sudden insurgence, he was immediately transferred to the intensive care room into the department. Monitored (cardiac pulse, blood saturation, respiratory frequency), oxygen administration was necessary to blood oxygen desaturation (91%). The patient was managed in collaboration with the ear-nose-throat (ENT) and anesthesiologist consultants. Chest and neck X-ray **(Figure 1)** and a consequent Computerized Tomography (CT) scan showed an important SE with involvement of all the neck, extension until skull basis and upper part of the thorax, and a concomitant important and voluminous pneumomediastinum. The CT scan revealed also an initial laryngeal stenosis. A nasal fibroscopy was performed by the ENT consultant and did not show any upper airway alterations until the glottis.

Blood test done immediately after triage showed a mild leukocytosis and C-reactive protein increase with no other particular alterations. Intravenous hydration was started (to avoid oral liquid or food intake) and, in agreement with the consultants ENT and anesthesiologist the patient did not receive the intubation, but only oxygen administration. To carry on appropriate monitoring, observation and treatment the patient was transferred to a specialist pediatric intensive care unit in another hospital, where thoracic surgery consultant was available in case of worsening.

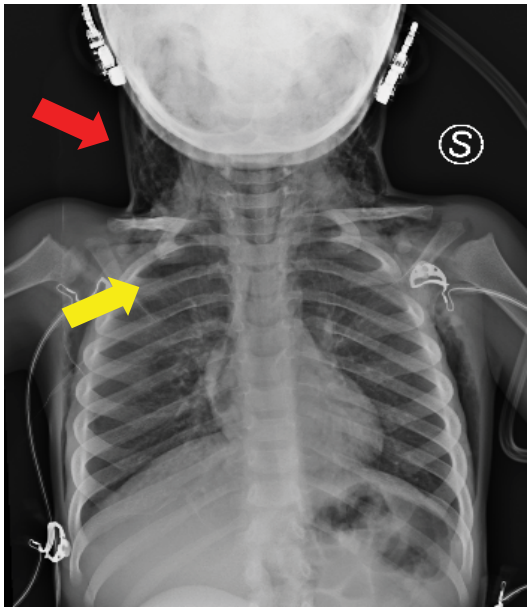

Figure 1 Pneumomediastinum (yellow arrow) and subcutaneous emphysema (red arrow) showed on patient X-ray.

Discussion

SPM and SE are uncommon diseases, in particular in children, and always represent an emergency in A&E departments. The presence of SE needs to look for a cause in thorax and mediastinum, due to the abnormal presence of air in these cavities and subsequent local diffusion through soft tissues. A multidisciplinary approach on patients with this diagnosis is very important to establish the entity of the disease, the risks and evaluate the better management of them.

In our case report, the patient was evaluated by a pediatrician, an emergency department doctor, an ENT consultant and an anesthesiologist, who collaborated to manage and agreed how to treat him. At the end, the patient was transferred to another hospital due to the lack of a pediatric intensive care unit and thoracic surgery consultants.

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