Abstract

**Aim:** To analyse the aetiology, pathophysiology and prognosis in a series of patients who developed clinically significant subcutaneous emphysema in a pediatric intensive care unit.

**Method:** Retrospective investigational study, recording and diagnosing all cases of clinically significant subcutaneous emphysema observed in the pediatric intensive care unit of a Spanish hospital during the period from January 2007 to December 2017.

**Results:** 19 patients (7 girls and 12 boys), aged from 4 months to 14 years, were diagnosed with clinically significant subcutaneous emphysema during the study period.

Regarding the aetiology of the condition, the origin of the emphysema was traumatic in 31.5% of patients (6 cases); associated with an invasive procedure or technique in 37% of patients (7 cases); and emphysema occurring in the context of a medical illness in 31.5% of patients (6 cases).

The emphysema occurred associated with pneumothorax in the 42% of patients but when the emphysema was due to traumatic event the association appeared in the 83% of cases.

Three patients required specific treatment for emphysema (15.8%) and one died due to its severity (5%).

**Conclusions:** Small subcutaneous emphysemas are asymptomatic and occur very frequently, associated with medical illnesses, traumas or technical/surgical procedures that are performed daily in intensive care units. Clinically significant, symptomatic emphysemas, however, are exceptional and can aggravate the clinical state of a patient who is already in a critical condition and may even be life threatening. In our serial, emphysema occurred most frequently associated to pneumothorax and it appeared isolated in the 16% of cases. Unfortunately we have not found any published data to compare our results. Despite the low mortality in our serial, emphysemas should be diagnosed as early as possible, and their evolution and clinical impact closely monitored.

Introduction

Subcutaneous or tissue emphysema is provoked when air enters the subcutaneous tissues. It can appear in various clinical situations in which the mucous skin barrier is disrupted, affecting the oropharyngeal, digestive or respiratory systems [1]. It occurs relatively frequently in pediatric patients, sometimes even spontaneously when performing Valsalva manoeuvres such as coughing, or as the result of prolonged crying.

In the case of critically ill patients, it is relatively frequent, occurring in the course of invasive procedures and manoeuvres such as the insertion of a pleural drainage tube or with positive pressure ventilation. It is also associated with certain surgical techniques or in potentially serious pathologies such as closed or penetrating trauma (wounds caused by firearms or stabbing) and soft tissue infections. In young children, it can even be a clinical sign of ill treatment [2].

The term spontaneous subcutaneous emphysema is used when the underlying cause is unknown; in these cases, and especially in young children, a more exhaustive and detailed clinical history is required in order to rule out the suspicion of mistreatment [3]. The fundamental pathophysiological mechanism by which air penetrates the tissues consists in the perforation at any level of some element of the oropharyngeal, respiratory or digestive mucous membrane barrier. A continuum of fascial planes connects the soft cervical tissues with the mediastinum and the retroperitoneum, allowing the aberrant air that penetrates any of these areas to spread elsewhere.

In most cases, the symptoms are minimal and the treatment is conservative (small subcutaneous emphysemas). Subcutaneous emphysema usually presents as a sudden, non-painful swelling that appears in the upper chest, neck and/or face, since the air usually comes from the chest cavity (it is either pulmonary or of intestinal origin). The condition is characterized by the presence of crepitus, described as “walking in the snow”, which is pathognomonic.

The compartment syndrome is a rare complication, which can provoke tissue vascular compromise and ischaemia. In these cases, surgical decompression may be necessary through “blowhole incision” and the placement of a subcutaneous drain [4]. If emphysema in the neck and/or face is very large, it can produce compression of the upper airway and severe respiratory insufficiency. In the most severe cases, it can extend to the abdomen or the limbs, given the
A continuum of fascial planes that connects the cervical soft tissues with the mediastinum and the retroperitoneum.

Diagnosis is fundamentally clinical, with the detection of the characteristic crepitus by palpation of the swollen area. It can be confirmed by simple radiography, where the condition appears as radiolucent striations that traverse the subcutaneous tissue and muscle and can dissect pulmonary and mediastinum structures.

With respect to other radiological techniques, it is important to know that when emphysema is present, thoracic ultrasound is less effective. On the other hand, computerized tomography is a very sensitive technique and can be used to locate the exact site at which the air enters the soft tissues.

Method

This retrospective study was conducted to analyse all the cases of subcutaneous emphysema diagnosed at the pediatric intensive care unit of a third-level Spanish hospital during the period from January 2007 to December 2017.

Results

19 patients (7 girls and 12 boys), aged from 4 months to 14 years, were diagnosed with clinically significant subcutaneous emphysema during the study period. The patients were classified into three groups, according to the aetiology of the condition: the first included emphysema of traumatic origin in 31.5% of cases (6 cases); the second, the cases associated with an invasive procedure or technique in 37% of cases (7 cases); and the third, the cases occurring in the context of a medical illness in 31.5% (6 cases). Three patients required specific treatment for emphysema and one died due to its severity.

The emphysema occurred associated with pneumothorax in the 47% of patients but when the emphysema was due to traumatic event the association appeared in the 83% of cases. In 37% occurred associated to pneumomediastinum and 1 patient presented it associated to pneumoperitoneum. The emphysemas were isolated in 16% of the patients.

Three patients required specific treatment for emphysema (15.8%) and one died due to its severity (5%).

Emphysema of traumatic origin

In six of the patients, the emphysema was of traumatic origin: in two of these cases, it was caused by a knife wound in the thorax (Figure’s 1 & 2), in another two it resulted from blockage of the airway by a foreign body during an intense Valsalva manoeuvre, in one following a traffic accident and in the final case, due to insufflation of the nasal cavity by pressurized air (the patient was the victim of mistreatment (Figure 3). In every case except the final one mentioned, the emphysema was associated with a pneumothorax that required urgent evacuation via a drainage catheter. In no case was specific treatment required for emphysema, and all these patients survived.

Emphysema secondary to medical-surgical procedures

Of the seven patients who developed emphysema due to invasive procedures, in two cases, it resulted from positive pressure ventilation: one of these patients was intubated via an orotracheal tube and also developed tension pneumoperitoneum requiring evacuation by puncture-aspiration (Figure 4). The other patient, who received mechanical ventilation through a tracheostomy tube, simultaneously developed tension pneumothorax and was the only case of death among the patients in our series (Figure 5).

Regarding the other patients, one case of emphysema was secondary to adenoidectomy (Figure 6) and another was secondary to thymectomy; in another two, it was secondary to a thoracotomy and in one case it followed an oesophageal dilatation manoeuvre. In none of these cases was specific treatment of emphysema necessary.

Emphysema associated with a medical condition

Six patients developed subcutaneous emphysema with clinical repercussions, in the context of the following diseases: two cases of severe asthma (Figure 7 & 8), one of necrotising pneumonia (Figure 9 & 10), one of laryngeal tumour, one of severe laryngitis (Figure 11) and one of spontaneous pneumothorax.

Only the patient with severe laryngitis required specific treatment. The remaining patients achieved a spontaneous favorable evolution, although the patient with severe asthma and associated pneumothorax needed thoracic drainage.
Discussion

Subcutaneous emphysema is caused by the penetration of air into the subcutaneous tissues. It occurs relatively frequently in pediatric patients, and can even arise spontaneously when performing Valsalva manoeuvres such as coughing, or in cases of prolonged crying.

In critically ill patients, it is frequently associated with invasive procedures and manoeuvres such as the insertion of pleural drainage or the use of positive pressure ventilation. The condition is also associated with certain surgical techniques and with potentially severe pathologies such as closed or penetrating injuries (injuries by firearm or stabbing) and soft tissue infections.

In our study population, subcutaneous emphysema was also observed in a patient who was the victim of mistreatment. This condition was, in fact, the sign that allowed the mistreatment to be detected.

In no case should this condition be underestimated; not only because of the possible clinical repercussions (as commented above, three of these patients required specific treatment), but also to detect the specific mechanisms that led to its appearance.

Its aetiology can be very diverse, and so clinicians should be aware that emphysema can develop in any clinical situation in which there is a disruption of the mucous skin barrier, and may affect the oropharyngeal, digestive or respiratory system.

Subcutaneous emphysema is said to be spontaneous when the underlying cause is unknown; in these cases, and especially in young children, a more exhaustive and detailed clinical history should be obtained in order to discount the possibility of mistreatment. In our series, the patient who had suffered abuse presented emphysema after the insufflation of air into the nasal cavities. This diagnosis was obtained following complementary studies including a CT of the skull and neck and a rhinofibroscopy.

Regarding the location of emphysema, the cases described in this paper show that due to a continuum of fascial planes that connects the cervical soft tissues with the mediastinum and the retroperitoneum, aberrant air can penetrate any of these areas and then be disseminated elsewhere. In our patients, emphysema was most commonly associated with pneumothorax (47% of patients) but when the emphysema was due to traumatic event the association appeared in the 83% of cases. Followed by pneumomediastinum in 37% of patients. The emphysemas were isolated in 16% of the patients. Three patient’s required specific treatment for emphysema (15, 8%) and one died due to its severity (5%). Unfortunately there are no published data to compare our results; in fact in our bibliography revision we could only find few case reports.

In our series, in all the cases the clinical presentation of the condition began as a sudden, non-painful swelling in the upper part of the chest, neck and/or face. When emphysema was associated with

pneumothorax, chest drainage was required. This was also the case of the patient with pneumoperitoneum. In all the cases in which the emphysema was associated with pneumomediastinum, the treatment was conservative.

Conclusion

Although clinically significant and symptomatic emphysema is an exceptional occurrence, it can worsen the situation of a patient who is already in a critical state and may even be life threatening [5,6]. Accordingly, it should be diagnosed at an early stage, its cause identified and its evolution and clinical impact exhaustively monitored.

We aim to investigate and publish the casuistic of other centers to know the real impact of this pathology in the pediatric intensive care units.

References