

CASE REPORT

A young healthy female with idiopathic extensive subcutaneous emphysema occurring in neck, mediastinum, left arm, peritoneal and retroperitoneal spaces: A case report

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Abstract

This is a case of a young healthy female patient with idiopathic extensive subcutaneous emphysema in neck, mediastinum, left arm, peritoneal and retroperitoneal spaces. The patient presented to the emergency department and then admitted to the ward for monitoring of vitals, evaluation and to manage her accordingly. The patient had a surgical intervention to release emphysema. She improved significantly post-surgery. She stayed in hospital for a total of 10 days. Then, the patient was followed in outpatient clinic weekly for a total of 4 weeks. She was completely asymptomatic with no new issues since discharge.

Introduction

Emphysema (Greek-derived word) medical terminology simply means 'inflation'. This term is frequently used to describe chronic lung disease where air is trapped in the lungs. Whereas, subcutaneous means underneath the dermal layer of the skin. Hence, the term 'subcutaneous emphysema' refers a collection/infiltration of air in a limited space in the body [1]. In other words, it is swelling of the tissues under the skin because of air tracking along tissue planes [2]. Several risk factors can contribute to the formation of subcutaneous emphysema, including but are not limited to surgical or endoscopic operations, trauma, pneumothorax, infections and spontaneous [2,3]. Injury to the thoracic cavity, sinus cavities, facial bones, barotrauma, bowel perforation or pulmonary blebs are some common causes [3]. Iatrogenic causes may occur due to malfunction or disruption of the ventilator circuit [3]. Additionally, injury to esophagus (as in gastric tube placement) can form communicating entry points for air passage [4]. Moreover, it could be a complication of Coronavirus disease of 2019 (COVID-19) infection [5].

The incidence of subcutaneous emphysema is anywhere from 0.43% to 2.34% [6]. In terms of management, subcutaneous emphysema will usually resolve conservatively without serious complications, however, concerted effort with careful observation should be made [6]. Severe cases of subcutaneous emphysema may require surgical interventions such as subcutaneous incisions, needles, drains or even mediastinotomy and this is determined mainly by the amount and sites of trapped air, presenting symptoms, haemodynamic instability and possible complications accordingly (compartment syndrome, for example) [2,6]. Herein, we will present a case of extensive subcutaneous emphysema that needed a surgical intervention.

Case presentation

A previously healthy 16-year-old Kuwaiti female brought by her mother to the emergency department in Farwaniya Hospital, Kuwait with complaints of spontaneous progressive diffuse neck swelling and tightness, dysphagia, mild breathing difficulty and swollen left arm and forearm started 7 days before presentation. She denied history of blunt or penetrating trauma, asthma, smoking, recent fish eating (fishbone), taking any medications. No history of foreign body aspiration or choking. Normal bowel habits and no urinary symptoms were observed. No similar attacks happened before. No medical/surgical history or relevant family history.

Her weight is 36 kg (recent intended weight loss) with body mass index (BMI) at 14.5 (underweight). On examination, the patient was alert and oriented. All vital signs were stable. She is maintaining 100% oxygen saturation on room air. On palpating the affected areas (neck, chest and left arm) crepitus were elicited. No obvious abdominal distension or bloating noted with no masses noted. On neck examination, no palpable masses, lymph nodes or thyroid abnormalities are found.

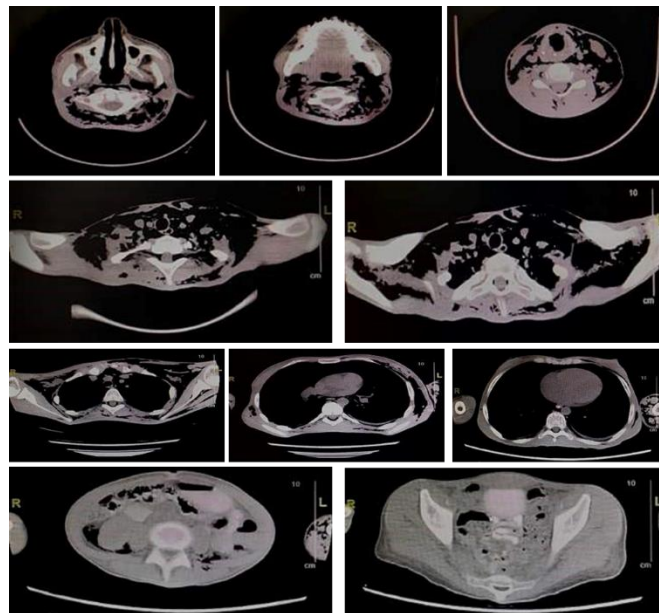


Figure 1: Axial slices of CT scan (Neck, Chest, Abdomen and pelvis) shown in a horizontal from left to right.

(Note: Extensive subcutaneous emphysema seen diffusely. This involves all deep neck spaces, dissecting the muscular planes with marked pneumomediastinum with minimal extra pleural involvement downward intra-abdominal through Peritoneal and retroperitoneal spaces. Also, it is noted in left arm and forearm).

Blood tests including full blood count, thyroid, renal and liver function tests are all acceptable. Computed tomography (CT scan) of the neck, chest, abdomen and pelvis showed extensive subcutaneous emphysema seen diffusely. This involves all deep neck spaces, dissecting the muscular planes with marked pneumomediastinum and minimal extrapleural involvement downward intra-abdominal through peritoneal and retroperitoneal spaces. No other significant findings including nasopharynx, laryngeal skeleton, lymph nodes, parotid and submandibular glands, thyroid gland, trachea and main bronchi,

oesophagus and carotid arteries calibers. No lung masses were detected (Figure 1).

Additionally, x-ray of neck/cervical spine was done which showed air trapped between tissue/muscular planes with no other noted abnormality (Figure 2).



Figure 2: X-ray of cervical spine
(Note: the air trapped in the neck)

Then, she underwent a surgical procedure to release the emphysema and her disturbing presenting symptoms next day of admission in the early morning (details of surgery discussed in the operative part). She remained free of symptoms post operation with resolution of the affected body parts by the emphysema. She stayed for 9 days post operation for observation, monitoring of vital signs and prophylactic antibiotics. During her stay she is doing very well with no episodes of signs deterioration. Before discharging, she had barium swallow studies that showed no narrowing, dilatation or abnormal filling defects with no evidence of leak of the contrast medium along the gastro-intestinal tract and no evidence of surgical emphysema noted in the plain x-ray of the neck region. Visualized oesophagus was normal in appearance and the peristaltic activity was normal. No gastro-oesophageal reflux was noted as well (Figure 3).

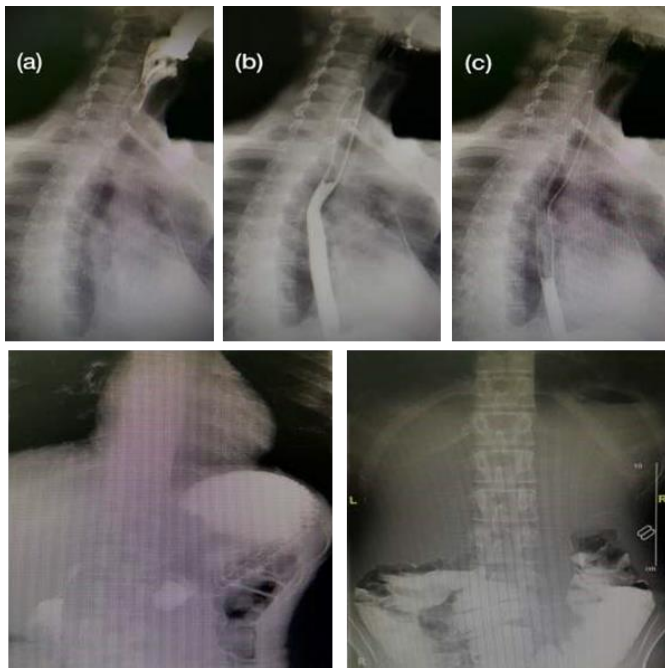


Figure 3: Barium swallow studies. Pictures a, b and c show the upper, middle and lower parts of the oesophagus with barium swallow. Bottom left picture shows the stomach. Bottom right picture shows the large intestine/colon.
(Note: There is no narrowing, dilatation or abnormal filling defects with no evidence of leak along gastrointestinal tract).

In addition, she had neck, chest, abdomen plain x-rays that all came back normal (Figure 4). After discharge she was followed in out-patient clinic weekly for 4 weeks and she was doing very well and asymptomatic. As she is underweight, she was advised to change her lifestyle in terms of nutrition and gain adequate weight and to follow up with nutrition specialists.

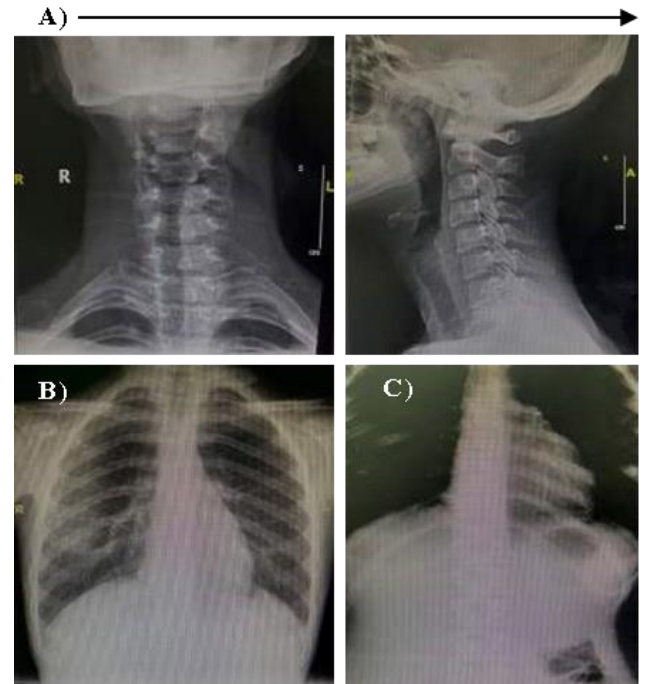


Figure 4: X-rays of A) Neck (Anterior posterior and lateral views); B) Chest C) Abdomen

- No air trapped between muscular/soft tissue planes with patent airway
- No pneumothorax or lung infection
- No air under diaphragm or dilated intestines loops

(Note: the normal findings of each x-ray)

Operative

Under local anaesthesia, bilateral 1 cm incisions were made in the neck anterior to sternocleidomastoid muscle, dissecting down until the level of thyroid lobes. Penrose drains were inserted bilaterally with significant improvement immediately.

Discussion

Subcutaneous was described in the literature when it began in late 1910s by Berkeley and Coffen [7]; then, Meyer and Lucke [8] explained another case in 1920. Years after, Bloomberg reported a speculated mechanism of air leak to cause pneumomediastinum and cervical emphysema (9). The occurrence of spontaneous subcutaneous emphysema is uncommonly described, although there are various etiologies mentioned in the literature [1]. These etiologies can be divided into idiopathic and secondary. The latter is described in several case studies [10-13] whereas just few studies described idiopathic type [14,15].

With regards to grading classifications that help to evaluate the extent of subcutaneous emphysema have received validation in some studies; however it is not globally applied or used on routine basis [6]. For example, Aghajanzadeh *et al.* [6] had a classification for severity of subcutaneous emphysema based on anatomical extension into five stages and manage that (conservatively or surgically) accordingly (Figure 5). The patient mentioned in that study with stage 5 of the classification was managed with two bilateral infraclavicular incisions.

In Kuwait, the incidence of secondary subcutaneous emphysema was reported in two studies; the first entitled "Subcutaneous Emphysema and Pneumomediastinum Following Oro-Facial First-

Degree Burns” by Ahmad Jaafar *et al.* [16]. The second entitled “Valsalva-type maneuver induced cervicofacial subcutaneous emphysema: A case report” by Abbas Al Ramzi *et al.* [17]. No cases of spontaneous mediastinum, cervical, peritoneal and retro-peritoneal subcutaneous emphysema were reported.

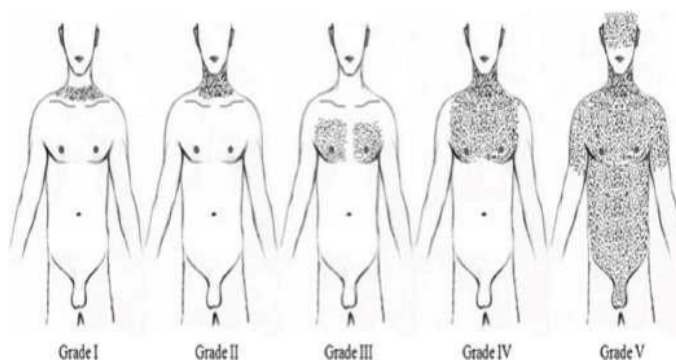


Figure 5: Classification for severity of subcutaneous emphysema based on anatomical extension according to Aghajanzadeh *et al.* [6].

Usually, the course of subcutaneous emphysema is nonfatal and self-limited. However, in cases of rapid and extensive gas expansion it can be life threatening. Massive subcutaneous emphysema can cause compartment syndrome, prevention of thoracic wall expansion, tracheal compression and tissue necrosis and in these dreaded complications, without intervention, respiratory and cardiovascular compromise can occur [3,6]. This is applied to our case which the emphysema was extensive, and the patient was symptomatic. Therefore, the patient needed a surgical intervention and then she was observed, and she left asymptomatic including in the out-patient clinic follow up.

Conclusion

Spontaneous extensive subcutaneous emphysema remains a rare presentation seen in emergency department. Some cases can resolve spontaneously whereas other cases need invasive treatment and the same applied to the investigations required. Therefore, lack of clear guidelines makes the diagnosis and management challenging. More cases need to be reported nationally and internationally in order to establish the protocol and clarify the management accordingly.

Conflicts of Interest: The authors declare that they have no conflict of interest.

Informed Consent: Informed consent was obtained from the patient for publication of this study.

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