Case of caverno-cutaneous sinus and subcutaneous emphysema

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DESCRIPTION

We present the case of a man in his 50s, a smoker with 30 pack-year smoking history, with no known comorbidities, with a history of adequately treated pulmonary tuberculosis 5 years ago, now presented to the emergency department with sudden onset of shortness of breath, bilateral chest pain (more on the left side with radiation towards the back) and fever for the last 3 days. The patient also complained of gradual-onset swelling over the chest, which progressed to the face, abdomen and arms over 1 day. The patient denied any history of trauma, vomiting or surgical intervention. The patient visited a nearby centre for similar complaints where an intercostal drain on the right side was placed. He was subsequently referred to our centre due to ongoing respiratory distress.

On arrival, the patient was conscious and oriented but was tachypnoeic with a respiratory rate of 24 breaths per minute, a saturation of 90% on room air and a pulse rate of 100/min. The rest of the general physical examination was normal. On inspection, there was generalised swelling over the face, neck and chest. On palpation, there was crepitus over the chest, neck, face and arms suggestive of subcutaneous emphysema, more marked on the left side. ECG, renal and hepatic function tests were normal. On the chest radiograph (figure 1A), a ‘gingko leaf’ sign was seen with striations of gas along the pectoralis major muscle, suggestive of subcutaneous emphysema outlining pectoralis major muscle fibres (red arrows), giving rise to classical ‘Gingko-leaf sign’ with a right-sided intercostal chest tube in situ (yellow arrow). Subcutaneous emphysema extends up to the neck (green arrow). (B) Axial CT chest (lung window) showed communication (red arrow) between the left parenchymal cavity and subcutaneous space along with subcutaneous emphysema in the left chest wall (blue arrows). (C) Axial CT chest (mediastinal window) showed a pigtail catheter in situ in the left lung cavitary lesion (red arrow).
emphysema extending up to the neck and intercostal chest tube drain in situ. Axial CT chest (lung window, figure 1B) showed communication (red arrow) between the left parenchymal cavity and subcutaneous space along with subcutaneous emphysema in the left chest wall (blue arrows). Complete blood counts showed leucocytosis (total leucocyte count—17 x 10^9/L), so blood, sputum and urine cultures were sent, and the patient was started on injection meropenem and linezolid. The patient continued to deteriorate with worsening dyspnoea and septic shock. His left upper chest subcutaneous swelling increased in size, especially with coughing. He was given inferior vena cava (IVC)-guided fluids for shock and started on vasoressors. Because of respiratory distress and increased subcutaneous swelling, a pigtail catheter (figure 1C) was inserted to decompress the left chest subcutaneous swelling. A repeat high-resolution computed tomography (HRCT) chest showed no significant pleural collection of air.

Meanwhile, sputum culture showed *Escherichia coli*, sensitive to colistin, tigecycline and cotrimoxazole; urine culture isolated *E. coli* sensitive to cotrimoxazole and fosfomycin while the blood culture was sterile. Sputum acid-fast bacillus (AFB) smear and sputum cartridge-based nucleic acid amplification test (CBNAAT) were unremarkable. Despite giving sensitivity-based antibiotics and supportive care, the patient’s condition worsened. Cardiothoracic and vascular surgeon (CTVS) opinion was sought and surgical intervention was deferred until the patient stabilised, but the patient died on the day fifth of admission.

Caverno-cutaneous sinuses are rarely encountered clinical entities. The most common cause of subcutaneous emphysema encountered by a respiratory physician in pneumothorax or pneumomediastinum is either spontaneous or due to trauma. In our case, we saw extensive subcutaneous emphysema due to a caverno-cutaneous sinus without significant pleural collection. Grossly asymmetrical subcutaneous emphysema, both clinically and radiologically, with an increase in the subcutaneous swelling on coughing, were the pointers towards the presence of caverno-cutaneous sinus, while HRCT chest was confirmatory in this case. Contrast instillation has been used to demonstrate communication in a few case reports.¹

Most cases reported in the literature are spontaneous due to tuberculosis aetiology.² Cases of posttraumatic caverno-cutaneous communication have also been reported.³ On the contrary, in our case, both AFB smear and sputum CBNAAT were negative. Bacterial aetiology leading to caverno-cutaneous sinuses has been rarely reported in the literature. Lack of significantly associated pneumothorax can be due to adhesions with a history of pulmonary tuberculosis.

In the majority of case reports, the administration of anti-tubercular treatment has yielded favourable outcomes in cases of tubercular aetiology. However, in instances where medical therapy and pigtail insertion prove ineffective, early surgical intervention tailored to the patient’s baseline status can be lifesaving in caverno-cutaneous sinuses of non-tubercular aetiologies.

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**Learning points**

- Subcutaneous emphysema can result from pneumothorax, pneumomediastinum or the rare caverno-cutaneous sinus. Asymmetrical emphysema and increased swelling on coughing can suggest caverno-cutaneous sinus, which can be confirmed through CT scans.
- There are no established treatment protocols due to limited literature. Possible management options include addressing the underlying cause, pigtail insertion for distressed patients and surgical intervention.

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Case reports provide a valuable learning resource for the scientific community and can indicate areas of interest for future research. They should not be used in isolation to guide treatment choices or public health policy.

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