Unexplained Recurrent Subcutaneous Emphysema of the Face, Neck and Mediastinum in an Otherwise Healthy Adult
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Abstract

Objectives: Understand the presentation and work-up of a rare cause of recurrent, diffuse subcutaneous emphysema in the head and neck.

Methods: We describe a case of a 23 year-old man who presented to the Emergency Department at a tertiary care center complaining of an acute onset of diffuse swelling throughout his face, neck, and chest. In our case we describe the work-up, diagnosis, and review of the current literature.

Results: A full diagnostic work-up for this patient was employed. Thoracic Surgery and Otolaryngology consultations were obtained. The patient had severe, diffuse subcutaneous emphysema throughout the face, neck, and chest on CT imaging. The remaining work-up was negative for organic causes. Upon further examination of the patient, several small, inconspicuous wounds were noticed on the patient’s right cheek and it was later determined that the patient had intentionally created puncture marks on the patient’s right cheek and buccal mucosa (Figure 4). After questioning the patient about the nature of his symptoms and percutaneous wounds, self harm was suspected and Psychiatry was consulted. Psychiatric evaluation revealed a history of self-mutilation and recurrent factitious subcutaneous emphysema was revealed as the working diagnosis for this patient. The patient was later discharged to his correctional institution with psychiatric follow-up. He has not re-presented to our institution.

Case Report

Our patient is a 23 year old inmate with a history of bipolar disease and depression who presented to our institution with a history of recurrent facial swelling and pain. Per his report, he had experienced severe swelling, pain and erythema twice in the previous month and had been evaluated at various hospitals without receiving a definitive diagnosis. He endorsed no other pertinent symptoms or history. He denied any recent history of trauma or surgery. The rest of his past medical history, family history, and social history were otherwise non-contributory. In the Emergency Room, he was placed empirically on broad spectrum IV antibiotics and evaluated by the Thoracic Surgery service. He was subsequently admitted to the hospital with suspected aerodigestive tract rupture. On exam, he was noted to have significant swelling and crepitus throughout his face, neck, and superior chest. The patient was found to have a mild leukocytosis but otherwise did not demonstrate any signs of distress, infection, or toxicity. Routine laboratory studies were within normal limits. Contrast CT demonstrated diffuse subcutaneous emphysema throughout the soft tissues of the face, neck, and upper chest without obvious aerodigestive tract defect or signs of infection (Figures 1-3).

Case Report (Continued)

A fluoroscopic esophagram was performed and was positive for esophageal leak and stricture. An operative flexible esophagoscopy and bronchoscopy were performed by the Thoracic Surgery service and also negative. At this point, an Otolaryngology consultation was ordered by the primary team. Flexible laryngoscopy was performed and also negative for obvious injury. On close physical examination, small needle-sized puncture marks were noted on the patient’s right cheek and buccal mucosa (Figure 4). After questioning the patient about the nature of his symptoms and percutaneous wounds, self harm was suspected and Psychiatry was consulted. Psychiatric evaluation revealed a history of self-mutilation and recurrent factitious subcutaneous emphysema was revealed as the working diagnosis for this patient. The patient was later discharged to his correctional institution with psychiatric follow-up. He has not re-presented to our institution.

Discussion

Subcutaneous emphysema of the head and neck can be caused by numerous etiologies which range from harmless to life threatening. Trauma, recent surgery, damage to the laryngotracheal complex, esophageal rupture, or infection with gas-forming organisms have all been well described in the literature and comprise the vast majority of cases. Self-inflicted subcutaneous emphysema, however, is exceedingly rare and has been reported only a handful of times in the literature since it was first described in 1971. Patients presenting with factitious subcutaneous emphysema are thought to introduce air into the subcutaneous tissues with needles and syringes, or by performing a valsala maneuver after creating defects in the oral or pharyngeal mucosa. This disorder may be of particular significance in the incarcerated population. Lopez-Peal et al. described a case of four prisoners in South America who presented with similar sudden onset of cervicofacial subcutaneous air. The patients had negative work-ups, and eventually admitted to creating puncture defects in their oral mucosa and expiring against a closed glottis, creating massive facial swelling, crepitus, and prompting their transfer to a medical facility with generally better conditions than their institutions. The DSM-V describes factitious disorder as “falsification of physical or psychological signs or symptoms” and states that resolution generally depends on successful behavior modifications and/or psychotherapy. When factitious causes of facial subcutaneous air are undetected, sequelae can be significant. A reported case of self-induced orbital emphysema undetected for 10 years resulted in blindness and eventual orbital exenteration.

Conclusion

While rare, factitious subcutaneous emphysema does occur in the general population and should be suspected when patients with prior psychiatric history present with physical findings inconsistent with the reported history. Because the work-up for this disease can be rigorous, invasive, and expensive if the true etiology is not uncovered early, a detailed initial physical examination is crucial and may help the consulting otolaryngologist discover the key (e.g. small puncture marks) to an otherwise puzzling case.

References


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