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Benign subcutaneous emphysema: A case report with bite

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ABSTRACT

INTRODUCTION: Subcutaneous emphysema is the presence of air/gas within the subcutaneous tissue plane. Though there are numerous causative factors, benign subcutaneous emphysema is rare.
CASE REPORT: We report a very rare case of benign subcutaneous emphysema following an insect bite on the forearm of a 48-year old man. The puncture area was erythematous and the emphysema extended to the whole arm, axilla and superior mediastinum. Despite, conservative management, the patient had residual discomfort and erythema around the puncture site. Surgical debridement resulted in resolution of symptoms.
DISCUSSION: Subcutaneous emphysema affecting an isolated limb is extremely rare. It is vital to differentiate it from life-threatening soft-tissue infections secondary to a gas-forming organism. Immunodeficiency states are key factors in the development and the outcomes of patients with subcutaneous emphysema. Medical management successfully resolves symptoms in the majority of cases. However, surgical debridement of the puncture site and surrounding fibrotic tissue has been advocated for those with persistent symptoms.
CONCLUSION: This case highlights a rare cause of benign subcutaneous emphysema, highlighting key issues surrounding its management for a successful outcome.

1. Introduction

Subcutaneous emphysema is defined as the presence of gas or air in the subcutaneous tissue plane. There are numerous causes for this condition. However, benign subcutaneous emphysema is a rare condition that commonly occurs secondary to trauma [1]. It is distinguished from necrotizing fasciitis, a life-threatening systemic condition caused by gas forming organisms, which involves infection of the subcutaneous plane, with extension into the deeper tissues [2]. Its aggressive nature means that early and prolonged antibiotic therapy with extensive surgical debridement is typically required. In contrast, benign subcutaneous emphysema is limited to the subcutaneous tissue, without systemic symptoms, and is more common in the upper extremities [3]. It is rarely progressive and generally does not require surgical intervention, unlike necrotizing fasciitis.

2. Case report

We present the case of a 48-year-old male farmer who presented to the emergency department with a 10-day history of insect bite to the extensor aspect of the left forearm. Initially the swelling was localized to the forearm, around the puncture site (Image 1). However, over the following three days the swelling became more diffuse involving the whole left arm with extension into the axilla. In addition, the patient was significantly concerned with a "crackling" sound of the involved area and therefore sought medical attention. The patient was otherwise well. Background medical history was significant for non-insulin dependent diabetes mellitus (poorly controlled) and non-Hodgkin's lymphoma which had been successfully treated with chemotherapy ten-years ago.

Examination revealed a diffuse swelling with associated subcutaneous emphysema involving the left upper limb and inner aspect of the axilla and hemi-thorax. Subsequent radiography also confirmed the presence of emphysema in the subcutaneous tissues of the forearm (Image 2). Computed tomography scanning of the thorax demonstrated subcutaneous emphysema tracking into the left axilla and superior mediastinum with no associated pneumothorax (Image 3). Interestingly, the patient's blood tests did not reveal a significant inflammatory process (white cell count 7.7, C-reactive protein 4.3, haemoglobin 14.1 g/dL). Despite this, blood cultures were sent for microbiology assessment, but were negative for any organismal growth.

The patient was admitted for supportive care and intra-venous antibiotics (cefuroxime) were administered over a forty-eight hour period due to the presence of extensive erythema at the puncture site. The patient was subsequently discharged home on oral antibiotics.

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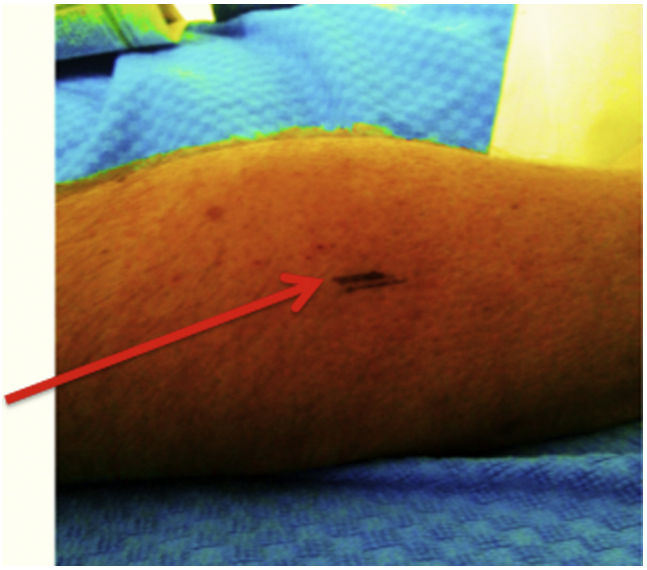

Image 1. Photograph of the left forearm showing puncture site (arrow) with surrounding erythema.

Follow-up review at surgical outpatients revealed persistent induration and emphysema with associated discomfort at the original puncture site. Subsequent magnetic resonance imaging (MRI) of the affected area demonstrated a large loculated collection of air in the subcutaneous plane and minor involvement of the underlying musculature (Image 4). The decision for surgical debridement was taken, as a result of the MRI findings coupled with on-going pain and anxiety expressed by the patient. At surgery, an area of slough and fibrotic tissue was excised (Image 5). No organism was identifiable by microbiology. The patient made an excellent recovery with no complications and is well eight-months post-operatively.

3. Discussion

Subcutaneous emphysema affecting an isolated limb is extremely rare, but is not a life-threating condition [3]. It is important to differentiate from serious soft-tissue infections such as gas gangrene (clostridial myonecrosis) and necrotizing fasciitis [4], especially in the early stages, as the management approaches vary significantly [3].

Subcutaneous emphysema can result from numerous benign causes with the significant majority trauma related [5]. Reported causes to date of benign subcutaneous emphysema include; high pressure injuries from power tools, blast injuries, air-gun injuries, dental extractions, hawthorn injuries, simple abrasive injuries (bike trauma, falls, etc.) and self-harm (factitious emphysema) [3,6–8]. Other rare causes include iatrogenic injuries (post-biopsy

Image 2. Plain (lateral view) radiograph of left forearm demonstrating subcutaneous emphysema (arrow).

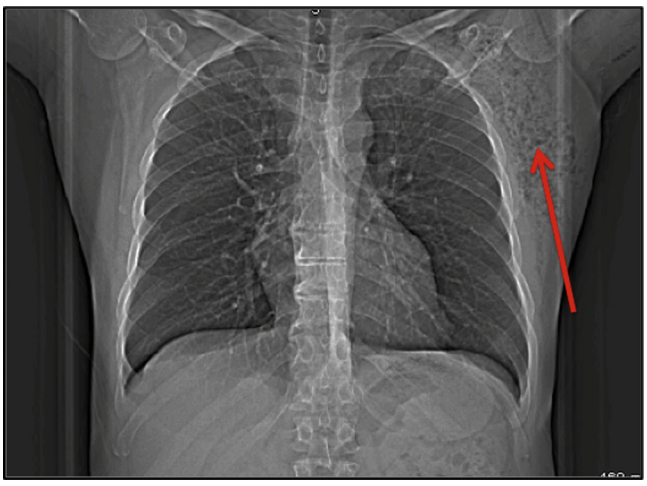

Image 3. Tomogram image from computed tomography scan of thorax – left axillary subcutaneous emphysema evident (arrow).

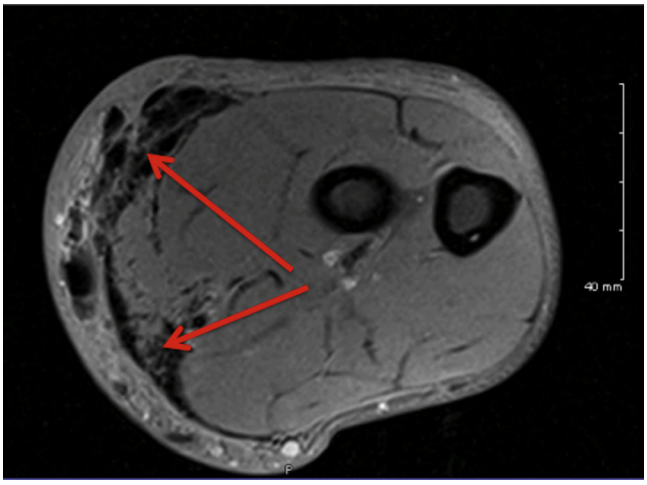

Image 4. Magnetic resonance imaging (coronal view) demonstrating large loculation of emphysema in the subcutaneous plane with minor involvement of the underlying musculature.

Image 5. Intra-operative photography showing slough and fibrotic tissue.

or insertion of peripheral vascular access) [1]. Speculation regarding mechanism of injury is that the trauma/puncture injury results in a one-way ball-valve effect causing accumulation of gas in the subcutaneous plane [3,9].

Predisposing factors for benign subcutaneous emphysema includes: chronic conditions with immunosuppressive states (diabetes, renal failure, congestive cardiac failure). In addition, patients on immunotherapy are also at risk [10]. The patient in our report had two predisposing conditions that placed him at risk (poorly controlled diabetes and history of non-Hodgkin's lymphoma treated with chemotherapy).

Insect bite injuries are known to commonly cause impetigo, folliculitis, cellulitis and lymphangitis. However, benign subcutaneous emphysema following insect bite is extremely rare with only two known cases documented in the literature [11,12]. The first case (De La Fuente Canibano) reported an insect sting on the neck of a forty-seven year old causing cervical subcutaneous emphysema, who was successfully treated with steroids and antihistamines [11]. The other case report (Russell et al.) also was due to an insect sting (bee) on the dorsum of the hand of a twenty-two year-old man. In contrast, he required surgical debridement due to persistent pain and erythema that was refractory to medical therapy (as in our case) [12]. Interestingly, in all cases both white-cell and C-reactive protein counts were all normal and microbiology analysis failed to detect any causative organism. For the majority of patients, current recommendations advocate a conservative management approach with antibiotics prophylactically, despite microbiology analysis commonly not detecting a causative organism. Reasons for prophylactic antibiotics use are due to concerns that the mechanism is commonly due to a traumatic puncture injury and therefore at risk of bacterial infection [12]. Other supportive measures including elevation and physiotherapy are often required [13]. Resolution with supportive care is common. However, protracted inflammation, erythema and/or patient discomfort can persist in some (immune-compromised) patients, with failure of conservative management. For these infrequent cases, surgical debridement may be required for resolution of symptoms. Debridement of the puncture site and any surrounding fibrotic tissue has been advocated, so as to remove the suspected foreign material that has been introduced into the tissue [12].

This case emphasizes the importance of a good clinical suspicion for any patient presenting with clinical evidence of subcutaneous emphysema after minor trauma, insect bites or trivial injuries. Though, often medical management will suffice, we highlight the need and benefits of a more radical management for those refractory to conservative management.

4. Conclusion

Insect bite is an extremely rare cause of benign subcutaneous emphysema. Conservative management may not result in an adequate resolution of symptoms, especially in cases of immunodeficiency. Therefore, in appropriately selected cases, surgical debridement may be required for satisfactory outcomes.

Conflict of interest

All authors declare no conflict of interests.

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Ethical Approval

N/A.

Consent

Written consent was obtained and is available if required.

Author contribution

Onwochei V.E. and Kelly M.E. were involved in the collecting of data, drafting the manuscript and have approved the final draft.

Lyons R. involved in the literature review, drafting of the manuscript and has approved the final version.

Khan W. and Barry K.M. were involved in the conception, design of the report, editing and approved the final version.

Guarantor

Onwochei V.E. and Barry K.M. accept full responsibility for this report.

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