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Progressive subcutaneous emphysema of unknown origin: a surgical dilemma.

OBJECTIVE: Subcutaneous emphysema can be an alarming sign of a necrotizing soft tissue infection. However, non-infectious etiologies exist that can be treated conservatively. This case report describes a subcutaneous emphysema of unknown origin and highlights the importance of distinguishing these clinical entities.

METHODS: We present a 17-year old female with pain and subcutaneous emphysema of the left arm. There were no systemic symptoms. Inflammatory parameters were slightly elevated. CT scan of the chest excluded intrathoracic abnormalities. Despite antibiotic treatment, the pain increased and the emphysema extended. Necrotizing fasciitis was feared. A surgical exploration was performed and hyperbaric oxygen therapy was started.

RESULTS: Intraoperatively, puncture marks were identified on the left arm. Air noticeably escaped, but normal, unaffected tissues were identified and microbiological cultures remained negative. We observed a good clinical evolution.

CONCLUSIONS: The lack of apparent causes, the unexplained puncture marks and psychiatric comorbidity suggests the possibility of subcutaneous emphysema due to factitious manipulations.

Patients with subcutaneous emphysema who remain clinically stable, have minimal pain and no significant inflammatory changes could be treated conservatively. Close clinical monitoring is essential to avoid delayed intervention in case of a necrotizing soft tissue infection.

Keywords;

Subcutaneous emphysema; necrotizing fasciitis; factitious subcutaneous emphysema; hyperbaric oxygen therapy; plastic surgery

Introduction

Subcutaneous emphysema (SE) of the upper extremity is an uncommon clinical feature. It can be caused by a necrotizing soft tissue infection (NSTI) or gas gangrene. A NSTI is a severe, rapidly progressing infection of the subcutaneous tissue and deep fascia with a high morbidity and mortality. Early empiric antibiotic treatment and aggressive surgical debridement is necessary [1-3]. However, non-infectious etiologies of SE exist that can be treated conservatively with good clinical outcomes. They must be promptly recognized to avoid extensive surgery [4].

We describe the case of a progressive SE of unknown origin in a young patient and highlight the importance of early recognition of NSTI and a low threshold for surgical exploration.

Case Report

We present the case of a 17-year-old Caucasian woman who was admitted at the Emergency Department with pain and SE of the left arm and shoulder. The pain started two days earlier with a swelling of the wrist. The patient denied a history of trauma. Since a cellulitis was suspected, Azitromycine was prescribed by the general practitioner. Despite the antibiotic treatment, the pain increased and the swelling spread towards the shoulder. The past medical history revealed coeliac disease and an Intensive Care Unit admission two years earlier for bilateral pneumonia and pericarditis. The patient is suffering from obesity and has a request for gender reassignment surgery. She is under care of a mental health care team.

Clinical examination showed subcutaneous crepitations and tenderness all over the left arm and axilla. There was no discoloration of the skin or development of blisters. Sensation of the limb was normal and there was no loss of function. Pulsations were clearly palpable and capillary refill time was normal. The patient was clinically stable and underwent a series of

investigations. Inflammatory parameters were slightly elevated with a leukocytosis of $11,6 \times 10^9/L$ and C-reactive protein of 37,2 mg/L. Blood cultures were taken and intravenous broad-spectrum antibiotics (Amoxicillin/Clavulanic Acid and Clindamycin) were started. Radiography showed overt SE spreading from the left arm towards the axilla, without any signs of pneumothorax, fracture or osteolysis (Figure 1). On magnetic resonance imaging (MRI) of the arm, SE and inflammation of the superficial and deep fascia were noted, without muscle involvement. An additional computed tomography (CT) scan was performed and excluded pneumothorax and pneumomediastinum. Progression of the SE towards the base of the neck and anterior thorax was noted (Figure 2).

Since the inflammation of the deep fascia on MRI and the spreading of the SE to the chest wall, a NSTI was feared. A surgical exploration was performed. With the bright lights of the operating theatre shining on the patient's arm, multiple puncture wounds became visible. Explorative incisions were made over the areas of maximal crepitation: the dorsum of the hand, the lower and upper left arm. Air noticeably escaped, nevertheless healthy tissues were identified without any signs of infection or tissue necrosis. Deep tissue samples were taken. Wounds were rinsed and subsequently wrapped in sterile gauzes. In addition, the patient received two sessions of hyperbaric oxygen therapy (HBOT).

Because of the identification of puncture marks on the non-dominant left arm, we performed a toxicological screening on blood and urine samples. The patient denied intravenous drug abuse and claimed the puncture points to be mosquito bites.

All microbiological cultures remained negative. The toxicology screen was positive for cannabinoids and opiates, although the samples were taken after general anaesthesia. We observed a good clinical evolution with improvement of the pain and crepitations and a resolution of the leukocytosis. Two days after the initial surgery, the wounds were closed. The postoperative recovery was uneventful. The patient was discharged two days later.

Discussion

Acute widespread SE of the upper extremity is a rare clinical entity and faces the physician with an important surgical dilemma. It can be an alarming clinical feature since it can be caused by a NSTI. However, noninfectious etiologies exist and should be recognized. Benign SE has been described after surgical procedures [5, 6], penetrating wounds [4, 7, 8], pneumothorax [9] or pneumomediastinum [10], an insect bite [11] or due to factitious manipulations [12-15]. The term NSTI describes a group of limb- and potentially life-threatening infections of the skin, soft tissues and muscles, which tend to progress rapidly throughout the fascia planes. The mortality rate is as high as 40%. Early diagnosis is mandatory, since any delay in surgical treatment is associated with higher rates of amputation and higher mortality rates. If a high clinical suspicion exists, no laboratory result or radiological finding should delay surgical intervention [1, 2, 16]. A thorough history and examination is essential to assess timing of possible trauma and crepitations, overall health status, systemic symptoms and signs of an aggressive soft tissue infection such as loss of function. Generally, patients with noninfectious SE are devoid of systemic symptoms. Crepitus will develop within minutes to hours after injury and pain is mild. In contrast, patients with NSTI often appear toxic and show inflammatory changes as swelling and erythema. Disproportionate pain is often the first sign [8, 10].

In this case, we were faced with a young patient, suffering from pain and swelling of the arm, with slightly elevated inflammatory parameters and without a history of trauma. A chest radiography revealed no pneumothorax or pneumomediastinum. In a stable patient with SE of the upper extremity, an additional chest CT may be beneficial [9]. The CT scan of our patient showed no occult pneumothorax or other intrathoracic pathologies. Although the patient remained hemodynamically stable, the lack of a benign cause, the pain and spreading

of the SE towards the chest wall despite antibiotic therapy were alarming symptoms. We decided to intervene surgically and to start HBOT.

Multiple studies have evaluated the role of HBOT in severe soft tissue infections. HBOT creates an oxygen-rich environment in which it augments neutrophil bactericidal activity, kills anaerobes and limits exotoxin and spore production. When implemented early, adjunctive HBOT may decrease mortality and limit the extent of debridement [17, 18].

Since surgical exploration showed healthy tissue and microbiological cultures were negative, an infectious cause of SE was less likely. However, we kept in mind the patient already took antibiotics for two days. The puncture points on the arm and the lack of apparent causes, suggests the possibility of factitious SE. The literature describes some similar cases, in which most patients were adolescents or young adults, often with an underlying psychiatric condition. The patients typically present with unexplained regional subcutaneous crepitus and puncture marks, sometimes with bilateral or recurrent presentation [12-15]. It is critical to recognize warning signs in the history and one should look for puncture marks to support the diagnosis.

Not all SE in the upper extremity signifies a life-threatening infection and benign causes should be considered to avoid an unnecessary surgical intervention. Patients that remain clinically stable and afebrile, who have minimal pain and no significant inflammatory changes can be treated conservatively [4,8]. Close clinical monitoring is essential to avoid unnecessary morbidity that can result from delaying intervention in case of NSTI. In patients with SE, a high clinical suspicion for NSTI with a low threshold for surgical exploration is recommended [19].

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Figure legends

Fig. 1: Radiography of the left wrist and hand shows subcutaneous emphysema without signs of fracture or osteolysis.

Fig. 2: Chest CT shows overt subcutaneous emphysema spreading towards the pectoral muscle and base of the neck. There are no arguments for pneumothorax or pneumomediastinum.

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Figure Radiography of the left wrist and hand

Figure Chest CT

The Editor in Chief

Acta Chirurgica Belgica

December 4, 2017

Dear Editor-in-Chief,

Please find our manuscript entitled “Progressive subcutaneous emphysema of unknown origin: a surgical dilemma”, for review and eventual publication in your journal.

We believe that this paper, addresses a clinically important issue and believe this article merits a place in the Acta Chirurgica Belgica journal. We present the case of a progressive subcutaneous emphysema of unknown origin in a young patient. Since a necrotizing fasciitis was feared, a surgical exploration was performed. However, unexplained puncture marks and psychiatric comorbidity suggested the possibility of subcutaneous emphysema due to factitious manipulations. In this article we discuss the possible etiologies of subcutaneous emphysema and highlight the importance of early recognition of a necrotizing soft tissue infection.

We hereby certify that this paper is not under consideration by any other publication and has not been published elsewhere. All of the co-authors participated in the preparation, have seen the manuscript and agree with its content and the conclusions of the work. There is no conflict of interest in publishing this paper.

Yours sincerely,

L. De Roeck, M.D.

On behalf of the authors