Annals of Medicine and Surgery 76 (2022) 103443

Contents lists available at ScienceDirect

Annals of Medicine and Surgery

journal homepage: www.elsevier.com/locate/amsu

Case Report

Recurrent spontaneous subcutaneous emphysema of unknown origin: A case report with literature review

Kamaran Amin Karadakhy [b,d], Fahmi H. Kakamad [a,b,c,*], Shvan H. Mohammed [c], Abdulwahid M. Salih [a,b], Razhan K. Ali [d], Suhaib H. Kakamad [c], Bnar J. Hama Amin [a], Berwn A. Abdulla [a,c]

[a] Smart Health Tower, Madam Mitterrand Street, Sulaimani, Kurdistan, Iraq
[b] College of Medicine, University of Sulaimani, Madam Mitterrand Street, Sulaimani, Kurdistan, Iraq
[c] Kscien Organization, Hamdi Str, Azadi Mall, Sulaimani, Kurdistan, Iraq
[d] Shar Hospital, Sulaimani, Kurdistan, Iraq

ARTICLE INFO

Keywords:
Subcutaneous emphysema
Surgical emphysema
Diagnostic challenge
Conservative treatment
Pneumomediastinum

ABSTRACT

Introduction: Subcutaneous emphysema caused by a surgical operation is known as surgical emphysema, and if the cause is unknown, it is known as spontaneous subcutaneous emphysema. The current study aims to report a rare case of recurrent spontaneous SE of unknown origin.

Case report: A 27-year-old male patient presented with swelling of the chest, neck, and face that had started 20 days prior. There was crepitation on palpation. Pulmonary function tests were normal. Laryngoscopy showed a normal larynx. Bronchoscopy showed a normal bronchial tree except for some redness in the trachea and left main bronchus. Computed tomography of the chest with contrast showed subcutaneous emphysema in the anterior chest and lower neck.

The patient reported a similar condition 3 years prior resulting in swelling of the upper left chest with an associated pneumothorax that was treated with tube thoracostomy. Workup including VATS was done to find the underlying cause but no cause was found.

Discussion: The pathogenesis is the same as in the most cases. Air that is driven into the interstitial tissues around the pulmonary vasculature gradually moves back toward the lung's hilum, resulting in pneumomediastinum. The air gradually spreads to the soft tissues of the neck, face, chest, and limbs, resulting in widespread subcutaneous emphysema.

Conclusion: Spontaneous subcutaneous emphysema without known origin is a rare condition that may resolve by conservative treatment.

1. Introduction

Subcutaneous emphysema (SE) arises when air enters the tissues under the skin and the soft tissues. Because of the proximity to the airway, this commonly arises in the soft tissues of the chest wall and neck, but it can also occur in other areas of the body [1]. It can spread rapidly and affect the entire face, chest, upper and lower extremities [2]. It can occur as a result of a variety of processes, including trauma, pneumothorax barotrauma, infection, malignancy, or as a complication of surgical interventions [3]. It may occur in conjunction with pneumothorax or pneumomediastinum [4]. SE caused by a surgical operation is known as surgical emphysema, and if the cause is unknown, it is known as spontaneous SE [5]. SE is not a life-threatening condition, and the air will be reabsorbed over time - it might be irritating for the patient. The underlying cause, on the other hand, is a reason for concern and may necessitate both investigation and therapy [5]. Recurrent spontaneous SE of unknown origin is an extremely rare condition.

The current study aims to report a rare case of recurrent spontaneous SE of unknown origin. The report has been arranged in line with SCARE 2020 guidelines and includes a brief literature review [6].

* Corresponding author. Kakamad Doctor City, Building 11, Apartment 50, Zip code:00964, Sulaimani, Iraq.
E-mail address: fahmi.hussein@uniuvsul.edu.iq (F.H. Kakamad).

https://doi.org/10.1016/j.amsu.2022.103443
Received 6 January 2022; Received in revised form 19 February 2022; Accepted 27 February 2022
Available online 2 March 2022

2. Case report

2.1. Patient information

A 27-year-old whiteman presented with swelling of the chest, neck, and face that had started gradually 20 days prior. The patient was a manual worker. There were no associated symptoms such as pain and shortness of breath. There were no known aggravating and relieving factors.

He had a similar condition 3 years prior. After the swelling of the left upper chest, he was later admitted for tube thoracostomy after a pneumothorax was confirmed. He was discharged after three days.

VATS was done to find the cause but no cause was found. The lung parenchyma was normal.

He did not have a previous history of any dental procedures or other operations. He did not have any chronic diseases.

He underwent bronchoscopy, thoracoscopy, and laryngoscopy to identify the underlying cause, but no cause had been identified.

2.2. Clinical examination

On examination, there was crepitation on palpation. Emphysema reaching both eye lids, more on the left side (Fig. 1) with involvement of the chest and neck. Vitals signs were all within normal ranges.

2.3. Diagnostic assessment

Pulmonary function tests were normal. Laryngoscopic examination showed normal vocal cord movement, symmetrical movement, no visible masses, or lesions within the larynx; there was no stenosis. Bronchoscopy showed normal bronchial tree except for some redness in the trachea and left main bronchus; No mass or lesion was found. Bronchial wash revealed normal bronchial and alveolar epithelial cells along with a moderate scatter of foamy macrophages. Computed tomography (CT) of the chest with contrast showed SE in the anterior chest and lower neck. No pulmonary bullae, pneumatocele, pneumothorax, or pneumomediastinum were seen. The lung parenchyma was normal; only a small nonspecific nodule of 3 mm was noted in the middle lobe. Normal pleura with no pleural effusion; there were no enlarged lymph nodes.

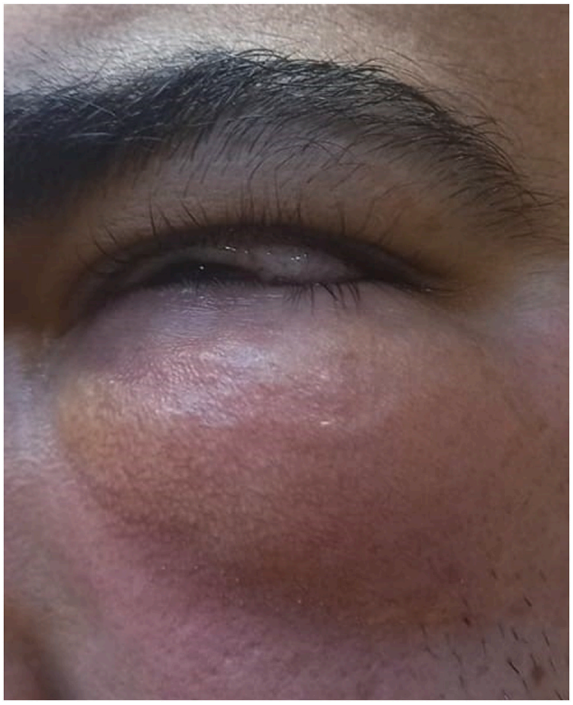

Fig. 1. Photo of the patient during the attacks of the emphysema.

2.4. Therapeutic intervention

With no associated pneumothorax, the patient was treated with a supraclavicular slit incision of the skin on this instance along with antibiotics and daily dressing.

2.5. Follow up

The patient's SE subsided and the patient was discharged several days later.

3. Discussion

Spontaneous SE is described as the presence of free gas or air in the subcutaneous tissue without an evident initiating or pulmonary cause [7]. SE may arise spontaneously as a consequence of rising pressure in the lungs caused by alveolar rupture. Air from the mediastinum and retroperitoneal areas moves via the fascial planes to the subcutaneous region, where it becomes trapped, resulting in subcutaneous emphysema [8].

In most cases, the pathogenesis is the same. Air that is driven into the interstitial tissues around the pulmonary vasculature gradually moves back toward the lung's hilum, resulting in pneumomediastinum. The air gradually spreads to the soft tissues of the neck and face, chest, and limbs, resulting in widespread subcutaneous emphysema [9]. Although SE is not deadly, it can be distressing for individuals and their families. This causes cosmetic defects but rarely causes physiologic complications such as tension pneumomediastinum, pneumothorax, or pneumopericardium [3]. It is a serious, distressing, and potentially fatal disorder if it affects the deeper tissues of the thoracic outlet, chest, and abdominal wall. It can be exacerbated by a restriction of full lung re-expansion, resulting in high airway pressure, severe respiratory acidosis, ventilator failure, pacemaker dysfunction, airway compromise, and tension phenomena [10].

In the current study, the cause of the recurrent spontaneous SE remains unknown after extensive workup examination. During these 3 years after many attacks of SE, only once he developed pneumothorax.

Because of the distinctive signs and symptoms, significant cases of SE are easily identified. The most frequent and prominent sign and symptom of SE is swelling around the neck, which is accompanied by chest discomfort. Other symptoms include a painful sore throat, an aching neck, trouble swallowing, shortness of breath, wheezing, and distension [3]. Periorbital edema might cause visual problems [10]. Srinivas et al. reported that SE can be more severe, resulting in cutaneous tension, dysphonia, and pneumoperitoneum [11]. Sometimes it may lead to cranial nerve weakness, pneumothorax, and infection secondary to contaminated air diffusion [12]. It is seldom reported that it can cause respiratory failure and upper airway obstruction [2]. The current case presented with swelling of the chest, neck, and face and associated crepitation on palpation.

The condition's common differential diagnoses include allergic response, hematoma, angioedema, esophageal rupture, and infectious necrotizing fasciitis [13]. The presence of crepitus on palpation of the swollen area is a prominent clinical feature of subcutaneous emphysema that distinguishes it from other disease types [14]. The diagnosis of subcutaneous emphysema is clear. It is obvious clinically and verified by a chest radiograph. The presence of air in the subcutaneous tissue and occasionally radiolucent striations surrounding the individual fibers of the pectoralis major muscles, mimicking the venous system of Ginkgo leaf, are seen on chest radiographs and are referred to as the 'ginkgo leaf sign' [15]. A chest CT scan is essential to evaluate or confirm the underlying lung abnormalities [15]. The CT scan, bronchoscopy, and

laryngoscopy conducted in this case were all normal.

When SE occurs, it must be detected early and handled carefully to limit the likelihood of subsequent consequences. SE treatment included airway management, and infection prevention.

The treatment of SE is mostly conservative and observational. Sometimes if left untreated, the disease can develop into a compromised airway, pneumomediastinum, or even pneumothorax [13]. Simple subcutaneous emphysema that does not impair breathing can be treated conservatively (bed rest, oxygen, analgesia, antibiotic prophylaxis, reassurance, and close monitoring) [16]. Many procedures have been used to manage severe subcutaneous emphysema, many of which are invasive or painful, and some of which may induce subcutaneous emphysema. Techniques commonly used include inserting chest tubes, opening the skin on the anterior chest, and introducing wide bore subcutaneous drains [17,18]. skin opening often clots, leading to treatment failure and scarring. A chest tube is necessary for the presence of pneumothorax. However, for pneumothorax of less than 20%, therapy isn't indicated unless the patient is having breathing difficulty [17].

Beck et al. reported the first subcutaneous use of micro-drainage by inserting bilateral fenestrated 14-gauge angio-catheters [10]. Matsushita et al. and Cesaria et al. suggested using Penrose drains and colostomy bags to treat severe life-threatening subcutaneous emphysema [19]. Micro-drainage with compressive massage has also been described by Srinivas et al. and Ozdogan et al. Both experienced a quick remission of SE in 12 hours with no complications [11,17]. Sherif et al. described the use of Jackson-Pratt drain, a closed suction drain with a bulb reservoir [20].

In conclusion, recurrent spontaneous SE of unknown etiology is a very rare condition. Reassurance of the patient and careful management are very important in the therapeutic management lines.

Ethical approval

The manuscript approved by ethical committee of Kscien.

Sources of funding

None is found.

Author contribution

Kamaran Amin Karadakhy: physician managing the case. final approval of the manuscript.

Abdulwahid M. Salh: major contribution of the idea, literature review, final approval of the manuscript.

Fahmi H. Kakamad, Razhan K. Ali: literature review, writing the manuscript, final approval of the manuscript.

Shvan H. Mohammed, Suhaib H. Kakamad, Bnar J. Hama Amin, Berwn A. Abdulla: literature review, final approval of the manuscript.

Registration of research studies

It is not applicable for case report.

Guarantor

Fahmi Hussein Kakamad is Guarantor of this submission.

Consent

Written informed consent was obtained from the patient's family for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Declaration of competing interest

None to be declared.

References

[1] M.A. Gajardo Rojas, P.E. Gajardo Rojas, C.G. Zúñiga Toro, D. Sepúlveda Pinto, C. A. López Cifuentes, I.J. Roa Henríquez, et al., Subcutaneous emphysema after ultrasonic treatment: a case report, Int. J. Odontostomatol.(Print) (2009) 67–70.
[2] R.K. Ali, F.H. Kakamad, S.H. ali Abdalla, S.I. Hussein, A.M. Salih, R.Q. Salih, et al., Management of post lobectomy subcutaneous emphysema; a case report with literature review, Ann. Med. Surg. 69 (2021) 102610.
[3] M. Aghajanzadeh, A. Dehnadi, H. Ebrahimi, M.F. Karkan, S.K. Jahromi, A.A. Maafi, et al., Classification and management of subcutaneous emphysema: a 10-year experience, Indian J. Surg. 77 (2) (2015) 673–677.
[4] S.O. Bello, S. Umejiaku, T.O. Ogunkunle, O.F. Afolabi, A.A. Yakubu, Spontaneous subcutaneous emphysema in a male toddler in a health facility in Nasarawa state: a case report, J. Pan Af. Thorac. Soc. 2 (1) (2021) 53–55.
[5] Z.N. Maan, A.R. D'Souza, Spontaneous subcutaneous emphysema associated with mephedrone usage, Ann. R. Coll. Surg. Engl. 94 (1) (2012) e38–40.
[6] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, A. Kerwan, A. Thoma, et al., The SCARE 2020 guideline: updating consensus surgical CAse REport (SCARE) guidelines, Int. J. Surg. 84 (2020) 226–230.
[7] Y.J. Lee, S.W. Jin, S.H. Jang, Y.S. Jang, E.K. Lee, Y.J. Kim, et al., A case of spontaneous pneumomediastinum and pneumopericardium in a young adult, Kor. J. Intern. Med. 16 (3) (2001) 205.
[8] A.K. Dhawan, A. Singal, K. Bisherwal, D. Pandhi, Subcutaneous emphysema mimicking angioedema, Indian dermatol. Online J. 7 (1) (2016) 55.
[9] D.B. Herlan, R.J. Landreneau, P.F. Ferson, Massive spontaneous subcutaneous emphysema: acute management with infraclavicular "blow holes", Chest 102 (2) (1992) 503–505.
[10] P.L. Beck, S.J. Heitman, C.H. Mody, Simple construction of a subcutaneous catheter for treatment of severe subcutaneous emphysema, Chest 121 (2) (2002) 647–649.
[11] R. Srinivas, N. Singh, R. Agarwal, A.N. Aggarwal, Management of extensive subcutaneous emphysema and pneumomediastinum by micro-drainage: time for a re-think? Singap. Med. J. 48 (12) (2007) 1149.
[12] B.F. Brasileiro, A.L. Cortez, L. Asprino, L.A. Passeri, M. De Moraes, R. Mazzonetto, et al., Traumatic subcutaneous emphysema of the face associated with paranasal sinus fractures: a prospective study, J. Oral Maxillofac. Surg. 63 (8) (2005) 1080–1087.
[13] S.M. Balaji, Subcutaneous emphysema, J. Maxillofac. Oral Surg. 14 (2) (2015) 515–517.
[14] C.H. Jeong, S. Yoon, S.W. Chung, J.Y. Kim, K.H. Park, J.K. Huh, Subcutaneous emphysema related to dental procedures, J. Kor. Assoc. Oral Maxillofac. Surg. 44 (5) (2018) 212–219.
[15] S. Pandey, A.K. Sahu, R. Sreenivasan, M. Ekka, Spontaneous subcutaneous emphysema: an uncommon presentation of a common disease, Am. J. Emerg. Med. 38 (9) (2020) 1990–e1.
[16] S.W. Dumont, A. Farag, Life threatening subcutaneous emphysema, Anaesthesia 63 (2) (2008) 212–213.
[17] M. Ozdogan, A. Gurer, A.K. Gokakin, S. Gogkus, I. Gomceli, R. Aydin, Treatment of severe subcutaneous emphysema by fenestrated angiocatheter, Intensive Care Med. 31 (1) (2005) 168.
[18] F. Leo, P. Solli, G. Veronesi, L. Spaggiari, Efficacy of microdrainage in severe subcutaneous emphysema, Chest 122 (4) (2002) 1498.
[19] T. Matsushita, A.T. Huynh, T. Singh, D. Thomson, Management of life-threatening subcutaneous emphysema using subcutaneous penrose drains and colostomy bags, Heart Lung Circ. 16 (6) (2007) 469–471.
[20] H.M. Sherif, D.A. Ott, The use of subcutaneous drains to manage subcutaneous emphysema, Tex. Heart Inst. J. 26 (2) (1999) 129.