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HEAD AND NECK

Severe subcutaneous and deep cervicofacial emphysema of unusual etiology

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Abstract Subcutaneous and deep cervical emphysema (SCE) in the head and neck are found in a wide spectrum of conditions. Most of them are seen in patients with midfacial trauma or oropharyngeal infections. Subcutaneous and deep cervical emphysema can also be a symptom of life-threatening mediastinitis and/or necrotizing fasciitis, both of which need immediate surgery. Rarely however does SCE occur in isolation as a consequence of elevated intraoral pressure in combination with or without visible lacerations of the oral mucosa. As a consequence, air penetrates the mucosal tears and results in subcutaneous emphysema even extending down to the mediastinum in severe cases. This article describes a series of five cases of isolated SCE. It discusses the diagnosis, the pathomechanism, the differential diagnosis and the treatment. It underlines the importance of anamnesis and careful physical and laboratory examinations in order to differentiate isolated SCE from more severe conditions such as necrotizing fasciitis or mediastinitis, which necessitate immediate surgery.

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Introduction

Subcutaneous emphysema is a relatively common clinical and radiological finding in the head and neck area [1–6]. Post-traumatic facial subcutaneous emphysema is classically associated with midfacial fractures following nose blowing, and it is usually a benign and self-limited condition. By contrast, subcutaneous and deep cervicofacial emphysema (SCE) complicating oropharyngeal infection is a serious phenomenon that can be associated with potentially lethal complications such as necrotizing fasciitis (NF) and mediastinitis if not promptly recognized and aggressively treated [7–9].

But what if there is no infection and no obvious history of major facial trauma? Do we have to act quickly, perform wide fasciotomy or administer antibiotics intravenously (i.v.)? Alternatively, can we simply observe and act should deterioration occur? The purpose of this article is to describe five consecutive cases of SCE of unusual etiology, their treatment and differential diagnosis.

Patients and methods

We retrospectively searched the medical notes of our department between 2008 and 2010 for patients who presented with SCE. Only patients without infections or midfacial trauma, but palpable emphysema and visible free subcutaneous air on computed tomography were included. Anamnesis, clinical appearance and treatment modalities are described for each patient. The different pathophysiologic mechanisms are explained and discussed.

Table 1 Summary of patient data

Patient	Age (yr)/sex	Hospital stay	Diagnosis—pathomechanism	Leuc/shift CRP/ProC	Antibiotics during hospitalization	Antibiotics after hospitalization	Blood cultures
1	48/f	4 days	Tooth extraction 28 with air-driven drill—direct air penetration through mucosal tear or Valsalva	10.6/89% 53/–	Augmentin® 1,200 mg 3 × 1 i.v. for 4 days	Augmentin® 625 mg 3 × 1 for 7 days	Sterile
2	50/f	2 days	Tooth extraction 13 with air-driven drill—direct air penetration through mucosal tear	9.5/– 8.9/–	Augmentin® 1,200 mg 3 × 1 i.v. for 2 days	Augmentin® 625 mg 3 × 1 for 10 days	Sterile
3	51/m	3 days	Excessive nose blowing—air penetration through mucosal tear and Valsalva	9.8/67% <1/–	Augmentin® 1,200 mg 3 × 1 i.v. for 3 days	Augmentin® 625 mg 3 × 1 for 5 days	Sterile
4	7/f	3 days	Excessive Valsalva for pain relief—air penetration through aphtha in the left cheek	6.2/52% –/<0.18	i.v. Augmentin® 1,200 mg 3 × 1 i.v. for 3 days	Augmentin® 312.5 mg 3 × 1 for 7 days	Sterile
5	29/m	½ and 1 day	2 episodes: self-inflicted oral wound—excessive Valsalva	8.6 and 13.3/– – and 2/–	Augmentin® 625 mg 1× p.o.	Augmentin® 625 mg 3 × 1 for 7 days	–

Leuc leucocytes in G/l (normal range 4–11 G/l), *Shift* leftward shift (normal range 33–80%), *CRP* C-reactive protein (normal range 0–10 mg/l), *ProC* procalcitonin (normal range <0.25 µg/l), *p.o.* peroral, *i.v.* intravenous, *Augmentin® 1,200 mg* 1,000 mg amoxicillin + 200 mg clavulanic acid, *Augmentin® 625 mg* 500 mg amoxicillin + 125 mg clavulanic acid, *Augmentin® 312.5 mg*: 250 mg amoxicillin + 62.5 mg clavulanic acid

Results

Patient histories are described below. Relevant patient data, details of treatments and results from blood examinations are summarized in Table 1.

Patient 1

A 49-year-old female patient suffered from toothache from her upper left wisdom tooth. Because it was partially impacted, the patient was sent to a dental surgeon for the necessary extraction under local anesthesia. To facilitate the intervention, ostectomy with an air-driven dental handpiece was carried out. The wounds were surgically closed, and the patient left. Some 30 min later, she realized that her midface was swollen as well as both sides of her neck. Within an hour, she came to our emergency department. While the swelling was relatively minor, we found extensive crepitation in almost all of the head and neck. Blood tests revealed partially elevated infection parameters. Emergency CT evaluation (Figs. 1, 2) revealed massive subcutaneous emphysema and pneumomediastinum. The air was present in the submandibular and masticator spaces on both sides and extended to the retropharyngeal space and in the visceral space of the infrahyoid neck along the deep and superficial fascias without evidence of infection. The patient was hospitalized and administered i.v. antibiotics. Four days later, the swelling had decreased considerably, infectious blood parameters were almost normal, and the patient was discharged. We continued peroral antibiotics for another 7 days and instructed the patient not to blow her nose and to sneeze with her mouth wide open. During re-examination 11 days after onset, all symptoms had resolved.

Patient 2

A 50-year-old female patient underwent an extraction of her upper right canine by means of an air-driven handpiece. Immediately after the procedure, there was a diffuse swelling of her right face extending from the lower eyelid down to the clavicle. Twelve hours later, she presented in the emergency department. We found small vestibular lacerations close to the socket of the right upper canine and the aforementioned swelling with crepitation. Blood tests were normal. Computerized tomography scans showed emphysema involving the right orbit, the right masticator and buccal space, Bichat's fat and the right submandibular space. The patient was hospitalized and administered i.v. antibiotics. Two days later, findings were unremarkable. We discharged the patient prescribing peroral antibiotics for another 10 days and gave the same instructions as described for Patient 1. A re-examination 3 days later was unremarkable.

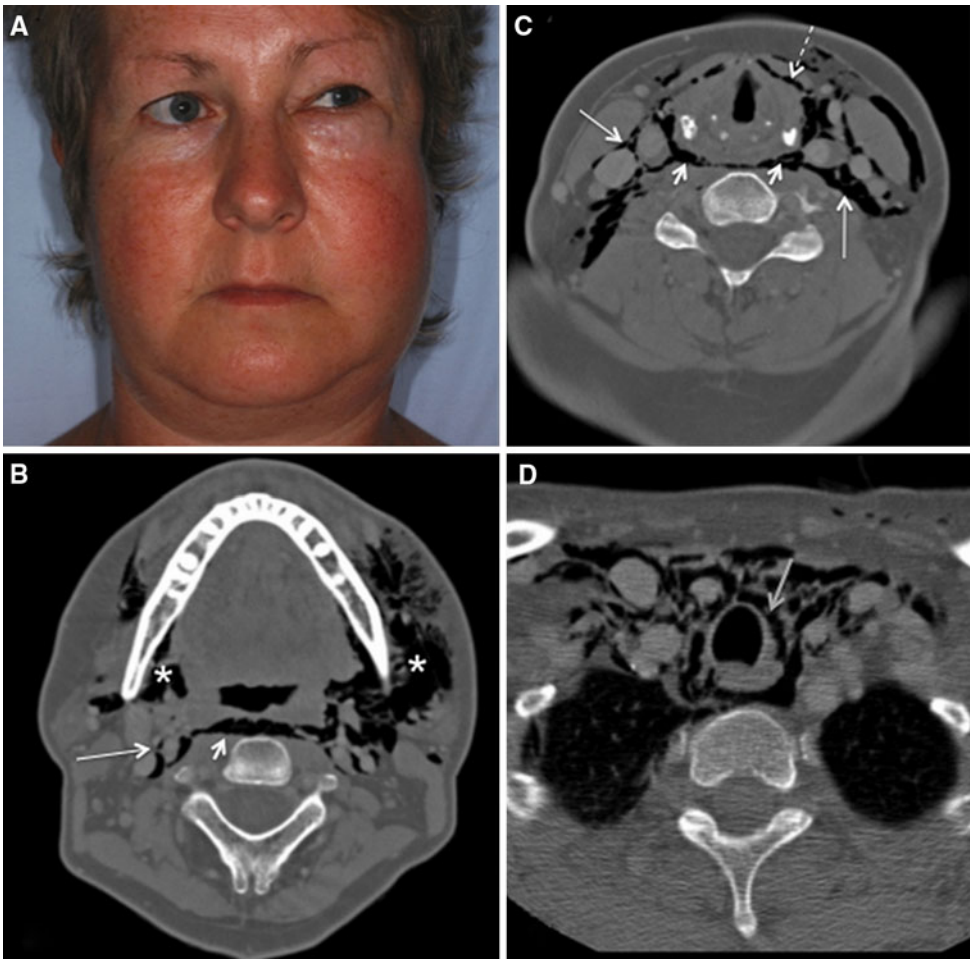

Fig. 1 **a** Photograph of patient 1 as she presented in the emergency department. Note bilateral facial swelling, more pronounced on the left, especially around the inferior left eyelid. **b–d** Axial images (bone window settings for better assessment of fractures and air) of a contrast-enhanced CT show massive emphysema involving both masticator spaces (*asterisks*), the retropharyngeal space (*white short arrows*), the carotid spaces (*white thin arrows*) and extending along the fasciae of the prelaryngeal muscles (*dashed arrows*) into the mediastinum (*gray arrows*)

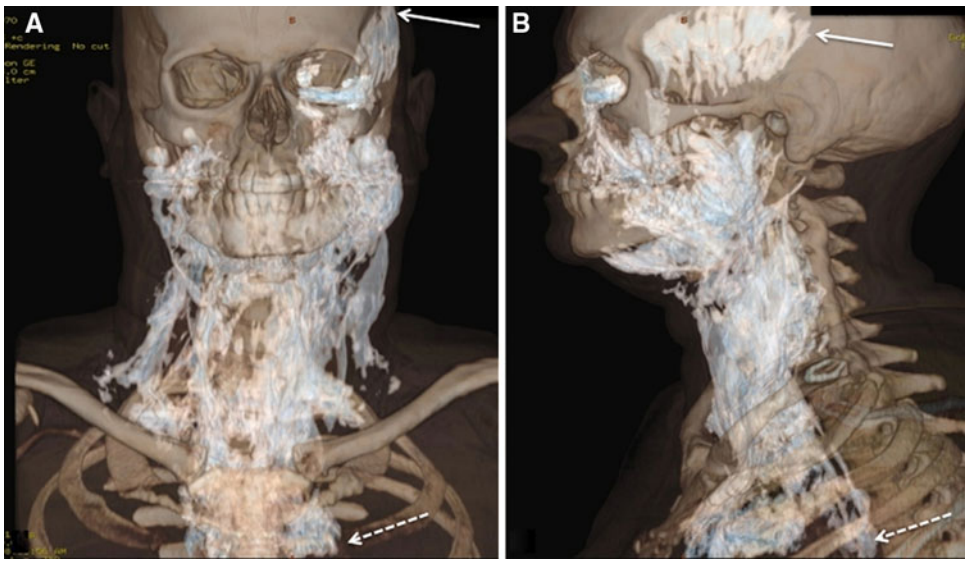

Fig. 2 3D reconstructions in frontal (**a**) and lateral (**b**) view of patient 1 with extensive emphysema (*white areas*) extending from the cranial portion of the temporalis muscle (*arrow*) to the upper mediastinum (*dashed arrow*)

Patient 3

After a night out drinking, a 51-year-old male patient woke up with swollen upper and lower left eyelids and pain behind his eye. Apart from the eyelid swelling, an ophthalmologic consultation in our emergency department was normal, and he was referred to us for further examination. Detailed anamnesis revealed good general health, no

trauma, no amnesia (despite the drinking) and a slight cold by self-report and observation. He also reported excessive nose blowing since childhood and especially since having contracted the current cold. Clinical examination showed unremarkable oral findings and no oral lacerations. Extraorally, we found a wide-ranging left hemi-facial swelling extending from the supra-orbital region down to the angle of the mandible. Crepitation was palpable in all of these areas as well as in the left temporal region. Blood examinations for infectious parameters and cultures were normal. Thin-slice CT scans showed air situated within the subcutaneous tissue of the left face, in the infratemporal fossa, in the orbit, extending along the facial planes of the masseter and medial pterygoid muscles, and around the mandible. Air was also present in the neck in the posterior and anterior cervical space, along the fascia of the sternocleidomastoid muscle, in the carotid space, in the retropharyngeal space and along the esophagus. In order to exclude a potential perforation of the esophagus, investigations were completed with a gastroduodenoscopy, which was unsuspicious. The patient was given i.v. antibiotics and hospitalized. Symptoms disappeared after 3 days, and he was discharged. Peroral antibiotics were continued for another 5 days. He was also given the usual instructions. Re-examination 8 days after onset revealed that the patient was free of symptoms, and crepitation was no longer present.

Patient 4

A 6-year-old girl was urgently transferred from a local general hospital to our pediatric colleagues with a suspicion of NF of the left face and neck. She had been ill for 2 days with a rhino-pharyngitis with a febrile peak of 39 and 37.4°C at presentation. Furthermore, she suffered from a large painful aphtha in the left cheek after having bitten herself one day before admission. She had been frequently inflating her cheeks to relieve the pain from repeatedly biting the 2.5 cm in diameter measuring aphtha in the left molar region (Fig. 3). Clinically, we found a left-sided crepitating swelling of the upper and lower eyelids, the cheek, the mandible and the neck as well as subcutaneous crepitation in the temporal region. Blood examinations were normal, and blood cultures remained sterile. A CT scan demonstrated a significant amount of air within the subcutaneous tissues and the masticator space of the left side of the face but no abnormal fascial and muscular enhancement and no fluid collections. She was hospitalized and treated with i.v. antibiotics. During the next days, symptoms diminished. The aphtha healed visibly, however residual crepitation was still palpable in the neck region. She was discharged 72 h after admission and given peroral antibiotics for another 7 days. She was instructed to stop blowing her cheeks, not to blow her nose and to sneeze with her mouth open. For insurance reasons, she could not be followed-up at our hospital.

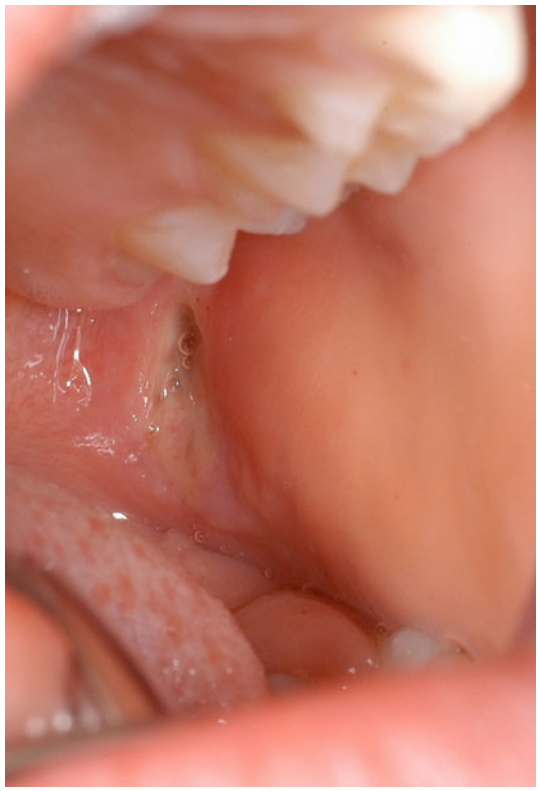

Fig. 3 Patient 5. Aphtha in the left cheek. In partial regression 2 days after admission

Patient 5

In a local prison, a 29-year-old male inmate was found with cut-wounds on his neck. Known for psychiatric problems, he was transferred to our colleagues from the psychiatric department to evaluate suicidal tendencies. Upon examination, our colleagues noticed a diffuse swelling of the face. According to the patient, this had appeared suddenly in the last 24–48 h; therefore, we were asked to see him. Clinically, we found a diffuse swelling of the face and a palpable crepitation from the left periorbital region down to the mandible. Inside the mouth, a razor blade was found hidden between the left cheek and the teeth. Otherwise, all findings were unremarkable. Because anamnesis was incoherent and changing, CT scans were taken to rule out further injuries. The imaging revealed large amounts of air dissecting the soft tissue of the left side of the face, extending from the masticator and submandibular spaces to the infrahyoid neck along the parapharyngeal space. The air crossed the midline to the right side through the submandibular space gaining access to the right masticator space. No air was present in the neck. Meanwhile, suicidal plans were precluded by the

psychiatrists and only hours after admission his return to prison was organized. We prescribed oral antibiotics for 1 week and gave the usual instructions. He was then followed-up by medical staff in the prison. Only 6 weeks later, the patient was re-admitted with a massive crepitating swelling of the left orbit and face, as well as gaping cut-wounds in the neck region. We sutured the lacerations, and our psychiatric colleagues diagnosed a borderline disorder. A CT scan underlined the clinical suspicion of uncomplicated SCE without further lesions. The air extended widely throughout the left face from the eyelid to the carotid space in the suprahyoid neck. We repeated our treatment and instructions, further psychiatric follow-up was organized, and the patient discharged within 24 h. Follow-up was done by medical staff in the prison.

Discussion

In the absence of midfacial fractures and infection, SCE is rare. The oldest description of the condition dates back to 1900. Alexander Turnbull, a retired British Royal Navy surgeon reported a patient he treated in the 1870s [10, 11]. An army bugle player had a "bicuspid tooth…… extracted, and he immediately sounded off on his bugle…… and he swelled up enormously". Other known pathomechanisms are air-driven handpieces used for dental procedures [2, 5, 7], unintentional [12, 13] and voluntary [14, 15] Valsalva maneuvers in combination with injuries of the oral mucosa, subcutaneous injection of air with a syringe [16], accidents with compressed air [17] or intubation [18]. It can also occur in case of mandibular fractures [19] where air penetrates through traumatic mucosal tears under elevated intraoral air pressure.

In a recent review concerning subcutaneous emphysema of dental origin, McKenzie describes a series of 32 patients [5]. In 50%, emphysema was due to air-driven handpieces during dental treatment. Patients 1 and 2 of our study belong to this group. In Patient 1, it is rather unclear if the emphysema was because of direct air intrusion or secondary due to later Valsalva maneuvers. From a pragmatic point of view, no matter what the precise etiology is, the clinical picture and treatment remain the same. In Patients 3 and 4, the etiology is clearly linked to repeated forceful Valsalva maneuvers. In combination with or without detectable lesions (Fig. 3) of the mucosa, this can occasionally lead to massive SCE. Patient 5 belongs to an etiological group that is seldom seen. Self-induced subcutaneous emphysema was described previously in 1971 by Gershwin et al. [20] but identified as a typical condition in the oral cavity only in 1996 [14]. Shortly thereafter, López-Peláez et al. [15] reported four prisoners with self-inflicted oral injuries and forceful Valsalva maneuvers resulting in deep cervical emphysema, pneumomediastinum and occasionally a secondary pneumothorax.

When diagnosing SCE, detailed anamnesis, careful physical and laboratory examinations are of paramount importance. In contrast to advanced NF or mediastinitis, patients with pure SCE have few complaints and feel relatively well. Blood examinations are normal or show only slight elevation of infectious parameters. The physical examination may vary widely from discrete to grotesque swelling plus SCE with or without crepitation [3]. By no means does the aspect of the patient reflect the extension of the air collection. Patient 1 (Fig. 1) had a rather minor swelling of her left eyelid but a complete air dissection of most cervical fasciae down to the mediastinum. The differential diagnosis must include all neighboring sites such as the larynx, pharynx, trachea, esophagus, and the lungs. Subcutaneous air, for example, is found in cases of pneumomediastinum, which classically presents with stabbing pericardial chest pain in 80–90% of patients [1], after blunt trauma to the upper aerodigestive tract [3] or after pneumothorax [21]. If anamnesis is not very clear and doubts remain, then the etiologies we described should be searched for and excluded such as for Patient 4.

In the early stages, both SCE and NF have rather nonspecific symptoms so that clinically the differentiation is difficult. A contrast-enhanced CT scan is the additional examination method of choice. [22]. While uninjected CT may be sufficient to detect air in the soft tissues of the neck, the administration of intravenous contrast material is mandatory, as it allows detection of NF and mediastinitis as well as differentiation of NF from benign soft tissue emphysema. The characteristic CT signs of NF include diffuse thickening and infiltration of the (sub)cutis, contrast material enhancement of the fasciae, the platysma, the sternocleidomastoid muscle as well as prelaryngeal strap muscles and multiple fluid collections in multiple neck compartments. In as many as 50% of the patients there are clinically unsuspected pleural or pericardial effusions and reticulated enhancement of the mediastinal fat or mediastinal fluid collections suggesting mediastinitis [23]. In isolated SCE without NF with or without mediastinitis, these signs are absent. No enhancement of the fasciae, muscles or fat is seen, and there are no fluid collections.

While NF and mediastinitis require immediate and aggressive surgery with extensive debridement and hospitalization in the intensive care unit [7–9, 24], pure SCE is usually a benign, self-limited condition. Typically, no surgical treatment is needed, and the air collections resorb within a few days [2, 5, 16, 21, 25]. To prevent superinfection, conservative management with antibiotic prophylaxis is recommended [5, 13, 16]. Pure observation without antibiotic treatment [6] should be avoided in order to prevent secondary infections.

Conclusion

The rare diagnosis of isolated SCE in the absence of trauma or infection can be established with a suggestive anamnesis, unremarkable blood examinations and in the absence of clinical and radiological features consistent with fasciitis, mediastinitis, pneumomediastinum or pneumothorax. Treatment consists of immediate hospitalization, antibiotic prophylaxis, avoidance of Valsalva maneuvers and patient information. The air collections will resorb within a few days, and patients can be discharged when asymptomatic. Antibiotic prophylaxis should be continued arbitrarily for a few days.

Conflict of interest The authors report no conflict of interest.

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