Cervicofacial and mediastinal emphysema following minor dental procedure: a case report and review of the literature

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Abstract
Background: Subcutaneous cervical emphysema is a clinical sign associated with many conditions, including laryngotracheal trauma, pneumothorax and necrotizing deep tissue infections.

Case presentation: We discuss a case of a 76-year-old man presenting with extensive cervical emphysema a few hours after a minor dental filling procedure. The CT-scan revealed a significant amount of air within the cervical and mediastinal spaces, reaching lobar bronchi. Vitals were within normal values. Bloodwork demonstrated an elevation of creatinine kinase (3718; normal < 150) and mild leukocytosis (WBC = 11.6). We decided to proceed to an urgent cervical exploration to exclude necrotizing fasciitis. This revealed air but no tissue necrosis nor abnormal fluid. The patient improved clinically and was discharged two days later with oral antibiotics. Although cervicofacial subcutaneous emphysema following dental procedures has been reported, it is usually less extensive and involving more invasive procedures using air-driven handpieces.

Conclusion: As an otolaryngologist confronted with extensive subcutaneous emphysema following a potential entry route for an aggressive infection, given the seriousness of this diagnosis, the decision of whether or not to perform a diagnostic surgical exploration should remain.

Keywords: Pneumomediastinum, Subcutaneous emphysema, Dental restoration, Necrotizing fasciitis

Introduction
Subcutaneous cervicofacial emphysema is a relatively frequent clinical entity and has a large differential diagnosis including, among others: angioedema and/or anaphylactic reaction, deep neck space infections, necrotizing fasciitis, airway trauma, dental or surgical procedures, pneumothorax or pneumomediastinum.

Iatrogenic subcutaneous emphysema can be diagnosed through history and physical examination, combined with the right radiological and laboratory tests, after exclusion of life-threatening pathologies.

The first case of subcutaneous emphysema caused by a dental procedure has been reported in 1900 by Turnbull et al. [1] So far, two reviews have been published in dentistry journals, respectively in 1995 by Heyman et al. [2] and in 2006 by McKenzie et al. [3]. Our objective is to report a severe case of subcutaneous emphysema, to review the last 10 years of literature on the topic and to discuss the management of those patients from an otolaryngologist’s point of view.

Case report
A 76-year-old male presented to the emergency department in our tertiary care center with left-sided cervicofacial subcutaneous emphysema. The questionnaire revealed he had sustained a routine dental filling of tooth #34 a few hours before. A small retraction
cord (#00) was used without an air-driven high-speed hand piece. However, an air syringe was used to do the filling. In that case, a rubber dam could not be placed due to the presence of an old subgingival defective restora tion in place. The procedure was done under local anesthesia without any ventilation, positive pressure event or CPAP use. About an hour after, cervical swelling and tenderness progressed. There were no other complaints. He had the same filling with the same procedure on the tooth #44 two weeks before.

He was otherwise known for hypertension, dyslipidemia and moderate chronic renal failure (baseline serum creatinine: 130 μmol/L). He had no history of head and neck pathologies or surgeries. He had known mild allergies to sulfamethoxazole/Trimethoprim and to amoxicillin, but no to penicillin. The patient was on simvastatin and had no recent change to his medication.

Physical examination revealed extensive, mainly left sided cervicofacial subcutaneous emphysema with associated erythema and tenderness on palpation. Vitals were: blood pressure 195 over 97 mmHg, heart rate 60 bpm and body temperature 37.5 °C. Oral cavity and teeth were unremarkable. There was no evidence of airway obstruction or respiratory distress. The remainder of the physical examination was within normal limits.

Blood tests showed a mild neutrophil-driven leucocytosis (white blood cells count of $11.6 \times 10^6$ (normal $3.8-10.6 \times 10^6$/mm$^3$) with $7.8 \times 10^6$ neutrophils) along with a marked elevation of creatinine kinase at 3714 (normal < 185 units/L) and patient’s baseline at 216). C-reactive protein was within normal limits. A chest x-ray (CXR) confirmed diffuse cervical emphysema and pneumomediastinum (Fig. 1).

A cervicothoracic computed tomography (CT) was ordered and showed a significant quantity of air in the superficial and deep spaces of the neck and mediastinum reaching the lobar bronchi bilaterally, suspicious of an aggressive infectious process according to the radiologist report (Fig. 2).

A developing necrotizing fasciitis could not be ruled out considering the extensive clinical and radiological subcutaneous emphysema associated with the leukocytosis and the significant rise in CK levels. Antibiotic therapy consisting of piperacillin/tazobactam, vancomycin and clindamycin was administered and an urgent surgical cervical exploration was performed, revealing air bubbles that had dissected the involved deep spaces but no evidence of tissue necrosis nor exudative fluid. Hemocultures and surgical wound cultures eventually came back negative. A CXR on postoperative day 2 showed a marked decrease of the cervical subcutaneous emphysema (Fig. 3).

The patient was discharged two days later with moxifloxacin for a total of 7 days. Under infectious disease specialist’s advice, moxifloxacin was chosen because of the patient’s allergy to amoxicillin and of its daily dosage. The patient was seen for follow-up at 3 months and was doing well without any sequelae except the well-healed scar.

**Literature review**

A comprehensive review of the English and French literature from 2009 to 2018 was conducted through the
PubMed database, using the research terms “dental”, “cervical emphysema” and “dental procedure”, in January 2020. Thirty-eight articles were selected based on their abstract and full text and were analyzed by 2 separate authors (AB and MP). The articles are summarized in Table 1. All patients presented with fascial and/or cervical swelling, and 37 (90.2%) presented within 24 h of the dental procedure. Thirty patients (73.2%) had a procedure involving molar teeth, of which twenty-two (73.3%) were mandibular. Twenty-eight patients (68.3%) had their procedure performed with a dental high-speed handpiece and five (12.2%) with an air-syringe. Thirty-eight patients (92.6%) had thoracic imaging (CXR or CT-scan), of which 27 (65.6%) had intrathoracic air or pneumomediastinum. Thirty-eight (92.6%) also received prophylactic antibiotics. Antibiotic regimen was heterogenous and was not detailed in 25 cases (61%). No complications were noted. No surgeries were performed, and all patients evolved well with resolution of the subcutaneous emphysema.

**Fig. 2** Cervicothoracic CT on arrival. a through f) Extensive emphysema, involving almost every deep neck and mediastinal spaces.

**Fig. 3** Postoperative day 2 CXR. Postero-anterior view showing significant improvement of subcutaneous emphysem
| Reference | Age (years) / Sex | Procedure / tooth (#) | Suspected cause | Timing of SC emphysema | Imaging modality/ air in mediastinum or intrathoracic | Labs | Hospitalization (#days)/Treatment |
|-----------|------------------|------------------------|----------------|------------------------|-----------------------------------------------------|------|----------------------------------|
| Arai & al. (2009) [4] | 40/F | Extraction/ 48 HS | 1 day | CT / - | N | 5 days / Ampicillin |
| Parkar & al. (2009) [5] | 55/F | Endodontic treatment/ left upper molar HS | 1 h | XR/ - | N | 1 day / corticosteroids + anti-histaminic + antibiotics |
| Samuels (2009) [6] | 20/F | Extraction/ left lower molar HS | Immediate | XR/ + | N | admitted / corticosteroids + analgesia + antibiotics |
| Kim & al. (2010) [7] | 40/M | Endodontic treatment / 36 HS | Immediate | CT / + | N | 5 days / O2 + antibiotics |
| Kim & al. (2010) [7] | 52/F | Endodontic treatment / 16 HS | Per-procedure | CT /+ | N | 8 days / antibiotics |
| Sainsbury & Jaiganesh (2010) [8] | 40/M | Endodontic treatment / 27 HS | Per-procedure | XR / - | N | < 1 day (14 h) / O2 + Amoxiclavin |
| Afzali & al. (2010) [9] | 16/M | Extraction / 37 HS | 1 day | XR + CT / + | WBC 21000 | 5 days / IV Clinda and cefazidime |
| Hsu (2010) [10] | 59/F | Endodontic treatment / 38 + 48 HS | 1 h | CT /- | N | Antibiotics |
| Bilecenoglu & al. (2012) [11] | 39/F | Extraction / 46 HS | 1 day | - | N | N/A / analgesia + antibiotics |
| Durukan & al. (2012) [12] | 45/F | Endodontic treatment / 16 AS + HS | Immediate | XR+ CT/ + | N | 3 days / O2 + metronidazole+ ampicillin |
| Bergen (2013) [13] | 72/F | RDP / molar HS | Per-procedure | XR / - | N | N/A / Amoxi Clav |
| Elia & al.(2013) [14] | 41/F | Extraction/ 47 HS | Per-procedure | CT /+ | N | 7 days/ analgesia + antibiotics |
| Khandelwal & al. (2013) [15] | 4,5/F | Crown preparation / 16 AS + HS | 1 h | - | - | - / Amoxicillin |
| Mitsunaga & al.(2013) [16] | 76/F | Laser treatment / 26 Laser | Immediate | CT / + | - | 5 days / antibiotics |
| Olate & al. (2013) [17] | 23/F | Extraction / 48 HS | 4 h | CT/ - | - | admitted / analgesia + chlorhexidine mouth wash + Cefazolin |
| An & al. (2014) [18] | 33/F | Endodontic treatment/ 44 AS | Per-procedure | CT/+ | N | 5 days / steroids + IV fluids + O2 + clindamycin and switch to ampicillin + metronidazole |
| Fleischman & al. (2014) [19] | 15/F | Extraction / 28 ? HS | Immediate | CT/ - | - | N/A / attempt to decompress the eyelid (30G needle) + antibiotics |
| Kün-Darbois & al. (2014) [20] | 41/F | Extraction / 38 HS | Per-procedure | CT / + | WBC 10370 | 2 days / - |
| Paik & al. (2014) [21] | 13/M | RDP / 36 HS | Immediate | CT /+ | - | 1 day / - |
| Nishimura & al. (2015) [22] | 68/M | RDP /? HS | 1 day | XR + CT / + | N | N/A / antibiotics |
| Picard & al. (2015) [23] | 27/M | Extraction / 48 HS | 4 days | CT / + | - | 4 days / antibiotics |
| Ocakcioglu & al. (2015) [24] | 23/M | Extraction / 48 HS | 7 days | CT / + | - | 4 days / O2 + antibiotics |
| Alonso & al(2017) [25] | 73/F | Peri-implant cleaning / 47 AP | Immediate | CT / - | - | N/A / corticosteroids + antibiotics |
| Alonso & al(2017) [25] | 43/M | Dental cleaning / 42-43 AP | Immediate | XR / + | - | < 1 day (12 h) / - |
| Alonso & 62/F | Dental cleaning / AP | Immediate | CT / - | - | N/A / Ibuprofen + antibiotics |
Discussion

The association between dental procedures and cervicofacial emphysema has been described in the dental literature. Even though every tooth may be implicated, mandibular molars are more frequently involved, for they have a closer relationship with head and neck deep spaces. The buccal, sublingual and submandibular spaces are intimately connected with the roots of these molars. The supra-hyoid spaces are contiguous with infra-hyoid spaces, notably the parapharyngeal and retropharyngeal spaces, which can lead to the mediastinal compartment. Different procedures have been associated with cervicofacial emphysema, ranging from endodontal treatment to teeth extractions, even hygiene procedures [42]. Use of air syringes, or more frequently air-driven dental handpieces, which inject air at high pressure, are prominent risk factors [3]. With their use, air can dissect in the soft tissues exposed around the tooth and spread through the deep neck spaces. Dentists can use special equipment, like rubber dams, to isolate the tooth in order to prevent such complications.

Given the eventuality of a dental iatrogenic cause being most probable based on history, surveillance in the emergency department or admission can be considered based on the clinician’s judgment. However, invasive infections, such as necrotizing fasciitis or mediastinitis, should be considered as they are infrequent but potentially catastrophic if not diagnosed.

Table 1 Summary of the literature review (Continued)

| Reference | Age (years) / Sex | Procedure / tooth (#) | Suspected cause | Timing of SC emphysema | Imaging modality/ air in mediastinum or intrathoracic | Labs | Hospitalization (#days)/Treatment |
|-----------|-------------------|-----------------------|----------------|------------------------|--------------------------------------------------------|------|----------------------------------|
| Chien (2018) [30] | 59/F | RDP / 44 + 46 HS | Immediate | – | – | N/A | N/A / + antibiotics |
| Jeong & al. (2018) [31] | 60/F | Crown preparation / 15 HS | 1 h | XR + CT/ + | N | 4 days / O2 + antibiotics |
| Lee & al. (2018) [32] | 51/F | Peri-implant cleaning / 12 AS | Per-procedure | XR + CT/ + | N | 13 days / O2 + analgesia + antibiotics |
| Liu & Lin (2018) [33] | 22/M | Extraction / 38 ? | 1 week | XR + CT/ + | Elevated CRP + WBC | Admitted / Amoxi-Clav |
| Tay & Loh (2018) [34] | 18/M | Extraction / 18 + 28 + 38 + 48 HS | 1 day | XR + CT/ + | – | 5 days / O2 + antibiotics |
| Tenore & al. (2017) [35] | 60/F | Endodontic treatment / 22 AS | Per-procedure | CT / - | – | Admitted / corticosteroid + analgesia + antibiotics |
| Cuccia & al. [36]. | 30/F | Extraction / 37 HS | Immediate | CT / + | N | 7 days / corticosteroids + tazocin / cubicin + bed rest |
| Fehrle & al [37]. | 32/M | Extraction / 48 ? | Weeks | CT / + | CRP 75 | Admitted / + antibiotics |
| Mascarenhas & al [38]. | 43/M | RDP / 47 HS | Immediate | XR / - | – | N/A / + Amoxicilin |
| Paschos & al [39]. | 17/F | Extraction / 38 HS | 30 min | CT / + | N | 3 days / antibiotics |
| Rad et & [40]. | 36/M | Extraction / 37 HS | Immediate | XR / + | N | N/A / + antibiotics |
| Rawlinson & al [41]. | 40/F | RDP / upper and lower molar ? | 1 day | CT / + | WBC 12500 | 1 day / + antibiotics |

F Female, M Male, RDP restorative dental procedure, HS High speed handpiece, AP Air polishing, AS air syringe, XR X-Ray, CT computed tomography scan, WBC white blood cell count, CRP C-Reactive protein, HBP High blood pressure, SC subcutaneous, N normal
promptly. Both are also known complications of the dental procedure itself [26]. In our review, most but not all patients received prophylactic antibiotics. The choice of the antibiotic, the route of administration and the duration of the treatment were heterogenous. Because of its adequate coverage of the buccal flora, penicillin is an adequate first choice, and it was chosen in most of the reported cases [1]. We found no case of significant infection, as was the case for McKenzie et al. [3]

Among other treatment modalities that have been reported, steroids have been used empirically in 5 patients to decrease edema and inflammation. Antihistamines were used in only 1 patient to treat empirically for a local anesthetic allergic reaction. However, depending on the clinical context, if an anaphylactic reaction or angio-oedema is suspected, epinephrine, steroids and antihistamines should be administered in a timely fashion [3] Oxygen supplementation was administered in 7 patients. Although no study was done to evaluate the efficacy of 100% O2 supplementation in case of subcutaneous emphysema, its use is extrapolated from pneumothorax cases: using 100% O2 accelerates the resorption of pneumothorax by reducing nitrogen gas pressure in pleural capillaries thus promoting resorption of air (mostly nitrogen) from the pleural space [8, 43, 44]. Despite the patient’s well-being and normal CRP, the choice of performing surgical exploration was not instinctive but made mainly because of the significant CK elevation and the radiologist’s report raising a high suspicion index of necrotizing fasciitis. The dental procedure could have been the entry route for an aggressive infection even if, in retrospect, this was not the case.

Conclusion

The use of high-speed dental handpieces and air-syringes during dental procedures can infrequently precipitate extensive subcutaneous emphysema. Clinical history and paraclinical investigation are keys to making the right diagnosis. In cases of iatrogenic subcutaneous emphysema related to dental procedure, conservative treatment has shown to be a safe option. Nevertheless, high clinical suspicion is warranted for an invasive necrotizing infection, given the seriousness of this eventuality but the choice between close observation or surgical exploration should rely on the clinician’s judgement.

Abbreviations

AP: Air-polishing; CT: Computed tomography; CXR: Chest x-ray; F: Female; HBP: High blood pressure; HS: High-speed handpiece; M: Male; RDP: Restorative dental procedure; WBC: White blood cell

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11. Bilecenoglu B, Onul M, Altay OT, Sakul BU. Cervicofacial Emphysema After Dental Treatment With Emphasis on the Anatomy of the Cervical Fascia. J Craniofac Surg. 2012;23(6):e544–8.

12. Durukan P, Salt O, Ozkan S, Durukan B, Kavalci C. Cervicofacial emphysema and pneumomediastinum after a high-speed air drill endodontic treatment procedure. Am J Emerg Med. 2012;30(9):2095–2096.e6.

13. Bergen T. Unusual case of cervicofacial surgical emphysema. Emerg Med Australas. 2013;25(5):473.

14. Ellia F, Lafiace B, Pagnozzi F, Boccazz A, Ferrari G, Perna M, et al. Cervicofacial subcutaneous emphysema and pneumomediastinum complicating a dental procedure. J Emerg Med. 2013;45(5):e179–81.

15. Khandelwal V, Agraval P, Agraval D, Nayak PA. Subcutaneous emphysema of periorbital region after stainless steel crown preparation in a young child. Case Rep. 2013;2013:221519:1.ebc201309952.

16. Mitsuanga S, Iwai T, Aoki N, Yamashita Y, Osumura R, Matsui Y, et al. Cervicofacial subcutaneous and mediastinal emphysema caused by air cooling spray of dental laser. Oral Surg Oral Med Oral Pathol Oral Radiol. 2013;115(6):e13–6.

17. Okate S, Assis A, Freire S, de Moraes M, de Albergaria-Barbosa JR. Facial and cervical emphysema after oral surgery: a rare case. Int J Clin Exp Med. 2013;6(9):840–4.

18. An GK, Zats B, Kunin M. Orbital, Mediastinal, and Cervicofacial subcutaneous emphysema after endodontic retreatment of a mandibular premolar: a case report. J Endod. 2014;40(6):880–3.

19. Fleischman D, Davis RW, Lee LB. Subcutaneous and Periorbital Emphysema Following Dental Procedure. Ophthal Plast Reconstr Surg. 2014;30(2):e43–5.

20. Kün-Darbois JD, Parè A, Chermi H, Daher G, Breheret R. Crépitations cervicales après avulsion dentaire. Rev Stomatol Chir Maxillo-Faciale Chir Orale. 2014;115(2):e17–8.

21. Paik YS, Lollar KW, Chang CWD. Iatrogenic subcutaneous emphysema after dental treatment. Ear Nose Throat J. 2014;93(2):E1–4.

22. Nishimura T, Sawai T, Kadoi K, Yamada T, Yoshie N, Ueda T, et al. Iatrogenic subcutaneous and mediastinal emphysema following a high-speed air drill dental treatment procedure: emphysema following dental treatment. Acute Med Surg. 2015;2(4):253–6.

23. Picard M, Pham Dang N, Mondie JM, Barthelemy I. Cervicotoracic Subcutaneous Emphysema and Pneumomediastinum After Third Molar Extraction. J Oral Maxillofac Surg. 2015;73(12):2286.e1–3.

24. Ocakcioglu I, Koyuncu S, Kupeli M, Bol O. Pneumomediastinum after tooth extraction. Case Rep Surg. 2016;2016:1.ebc2016001.

25. Alonso V, García-Caballero L, Couto I, Diniz M, Diz P, Limeres J. Subcutaneous emphysema related to air-powder tooth polishing: a report of three cases. Aust Dent J. 2017;62(4):505–10.

26. Lee S-W, Huh Y-H, Cha M-S. Iatrogenic subcutaneous cervicofacial emphysema with pneumomediastinum after class V restoration. J Korean Assoc Oral Maxillofac Surg. 2017;43(1):49.

27. Rammarine M, Dubin Z. Cervicofacial and mediastinal emphysema due to a dental procedure. J Emerg Trauma Shock. 2017;10(1):34.

28. Tan S, Nikolakos D. Subcutaneous emphysema secondary to dental extraction: a case report. Aust Dent J. 2017;62(1):95–97.

29. Thompson C, Gohil R. Pneumatic dental extractions: an unusual cause of extensive cervical surgical emphysema. BMJ Case Rep. 2017;2017:9.ebc2017118677.

30. Chien P-H. Iatrogenic subcutaneous facial emphysema secondary to a class V dental restoration: a case report. Aust Dent J. 2019;64(1):43–6.

31. Jeong C-H, Yoon S, Chung S-W, Kim J-Y, Park K-H, Huh J-K. Subcutaneous emphysema related to dental procedures. J Korean Assoc Oral Maxillofac Surg. 2018;44(5):212.

32. Lee S-T, Subu MG, Kwon T-G. Emphysema forming after air-powder abrasive treatment for peri-implantitis. Maxillofac Plast Reconstr Surg. 2018;40(1). doi: https://doi.org/10.1186/s40902-018-0151-7. [cited 2019 Mar 27].

33. Liu C-C, Lin M-Y. Diffuse soft tissue emphysema after dental procedure. CJEOM. 2017(252):338–39.

34. Tay YBE, Loh WS. Extensive subcutaneous emphysea, pneumomediastinum, and pneumonichiasis following third molar surgery. J Oral Maxillofac Surg. 2018;76(12):1609–12.

35. Tenore G, Palaia G, Cillof M, Mohsen M, Battiati A, Romeo U. Subcutaneous emphysema during root canal therapy: endodontic accident by sodium hypochlorite. Ann Stomatol (Roma). 2017;7(3):117–22.

36. Cuccia A, Genaci A. Cervicofacial and mediastinal emphysema after dental extraction. Dent Med Probl. 2019;56(2):203–7.

37. Fehrle C. Mediastinal and cutaneous emphysema following dental extraction. Dtsch Arzteblatt Online; 2019. https://doi.org/10.3238/arztebl.20190212a. cited 2020 Mar 17.

38. Mascarenhas RJ. Management of subcutaneous facial emphysema secondary to a class V dental restoration. Clin Case Rep. 2019;7(5):1025–30.

39. Paschos K, Chatziogiorgiadis A. Cervicofacial emphysema, Pneumomediastinum and pneumothorax caused by a dental procedure. J Coll Physicians Surg Pak. 2019;29(2):191–2.

40. Rad MV, Chan EKY, Ahmed IH. Cervicofacial subcutaneous emphysema and pneumomediastinum secondary to dental treatment in a young man. Respir Med Case Rep. 2019;28:100918.

41. Rawlinson RD, Negmadianov U, Rubay D, Ohanessian L, Waxman J. Pneumomediastinum After Dental Filling: A Rare Case Presentation. Cureus. 2019; Available from: https://www.cureus.com/articles/21620-pneumomediastinum-after-dental-filling-a-rare-case-presentation. [cited 2020 Mar 17].

42. Bocchialini G, Ambrosi S, Castellani A. Massive Cervicothoracic subcutaneous emphysema and Pneumomediastinum developing during a dental hygiene procedure. Case Rep Dent. 2017;2017:1.ebc2017021185.

43. Ali A, Cunliffe DR, Watt-Smith SR. Surgical emphysema and pneumomediastinum complicating dental extraction. Br Dent J. 2000;188(11):589–90.

44. Northern TC. Oxygen therapy for spontaneous pneumothorax. BMJ. 1971; 4(579):86–8.

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