Iatrogenic cervicofacial emphysema after dental procedures: a case series and radiographic review

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Cervicofacial emphysema (CE) occurs as a result of traumatic soft tissue injuries or open fractures, in which air is forced into the surrounding soft tissues and potential spaces of the head and neck.[1-9] Traumatic intubations and positive pressure ventilation aggravate mucosal or tracheal injuries, and air propagates through the injury, resulting in the formation of CE.[5,6] Cervicofacial surgeries create wounds adjacent to the aerodigestive tract, which can become inlets for air.[4,8] High-speed dental handpieces have a forward air exhaust port used to cool the bur, which can inject air into injured structures and cause CE.[3,8]

CE is dangerous because it can progress rapidly and cause massive swelling that leads to airway compromise.[10] Air has the potential to cause a mass effect and pressure on adjacent vital structures. As a result, patients typically require hospitalization for observation.[2,3,8,10,11] In this report, we present 2 cases of CE, their unique radiographic findings, and their management. Approval by the institutional review board was not required for this project.

CASE 1

A 30-year-old man was referred to the Emory University Hospital emergency department (ED) by his dentist for anaphylaxis. The patient's past medical history was significant for a traumatic brain injury, as a result of a motorcycle accident, and moderate residual lower extremity motor deficits. Before his presentation at the ED, his dentist was performing a crown preparation on the permanent left mandibular second molar (tooth #18) with an air-driven restorative handpiece. The dentist lacerated the gingiva on the left floor of the mouth, but he continued the procedure using the same dental handpiece. After the mucosal injury, the dentist noticed a gradual painless swelling of the patient's neck. The dentist thought this was an early sign of anaphylaxis and sent the patient to the ED.

Clinical examination revealed a healthy-looking man, who was resting comfortably on a stretcher but complaining of dysphagia. He was tolerating his secretions, was not posturing, and was able to communicate freely. His blood pressure, heart rate, and oxygen saturation levels were unremarkable. He was afebrile. Head and neck examination revealed prominent, painless bilateral lower facial and neck swelling, with crepitus that was worse on the left side. Intraoral examination revealed a maximal interincisal opening of 3.5 cm, bilateral floor of mouth elevation with crepitus, a 5-mm laceration along the left floor of mouth, and a prepared but unrestored tooth #18. The remainder of the examination was unremarkable.

A computed tomography (CT) scan revealed air dissecting into the subcutaneous tissues and multiple compartments in the neck, including the left parotid, bilateral masticator, bilateral carotid, left buccal, and retropharyngeal spaces. The air within the retropharyngeal space presumably dissected into the potential danger space and gained access to the anterior and posterior mediastinum (Figures 1-5).

The patient was subsequently admitted to the hospital medicine service. He received intravenous ampicillin-sulbactam (3 g every 6 hours) and had serial examinations performed by the oral and maxillofacial surgery service every 24 hours to monitor progression of the swelling. He remained NPO (nothing by mouth) until a modified barium swallow evaluation was completed the next day. Swallow evaluation showed no extravasation of contrast into the trachea, so the patient was advanced to a PO diet. After 2 days, his neck swelling improved, and he was able to tolerate a general diet. He was discharged home with a prescription for amoxicillin-clavulanate (875 mg every 12

Statement of Clinical Relevance

Cervicofacial emphysema is a rare but worrisome complication of head and neck procedures that can invade potential spaces. Distension of these potential spaces can reveal anatomic anomalies that can have clinical relevance for treatment.

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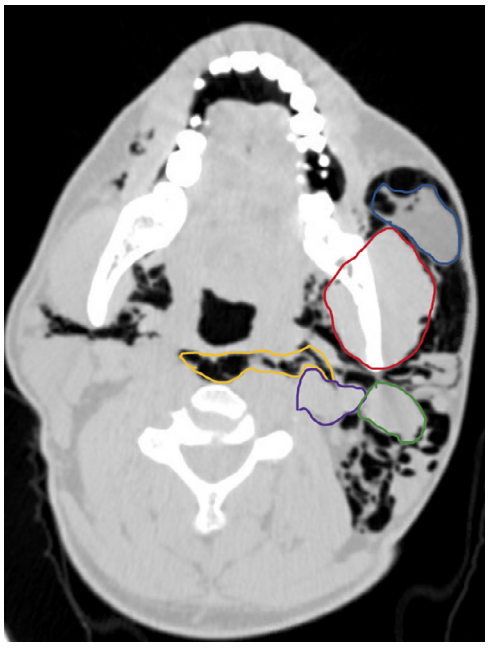

Fig. 1. Axial view of a computed tomography (CT) neck scan without contrast in the suprahyoid neck at the level of the mandibular ramus. Extensive air emphysema outlines the buccal space and buccal fat *(blue)*, masticator space *(red)*, parotid space *(green)*, retropharyngeal space *(yellow)*, and carotid space *(purple)*. There is also air around the masticator space on the contralateral side.

hours) for 5 days. He did not present for routine follow-up. However, he presented to the same ED 5 months later for an unrelated illness, and no residual findings were documented.

CASE 2

A 3-year-old girl was brought to the Children's Hospital of Atlanta ED by emergency medical services from her dentist's office for anaphylaxis. Her medical history was noncontributory. Before presentation, the pediatric dentist was performing complete dental rehabilitation with intravenous sedation (propofol and oral benzodiazepine) provided by an anesthesiologist. During sedation, the child became agitated and developed significant facial swelling. The dentist stopped, and the patient was given flumazenil, diphenhydramine, and intramuscular epinephrine. She remained agitated without improvement in swelling and was taken to the ED.

Upon presentation, the patient was found to be diaphoretic, with periorbital and lip edema and a proptotic tongue, and it was not possible to visualize the posterior oropharynx. The patient also had multiple carious teeth. Her respiratory examination was significant for wheezing, but she did not have stridor, rhonchi, or rales. She was afebrile, tachypneic, and agitated but had a normal blood pressure and heart rate. The patient's oxygen saturation was 94%. Venous blood gas was notable for a respiratory acidosis, which was attributed to her agitation. Once stabilized, the patient was admitted to the pediatric intensive care unit for continued observation. Approximately 8 hours after admission, she had a tonic-clonic seizure and developed respiratory distress. Lorazepam (0.5 mg/kg) was provided and positive pressure ventilation with a bag valve mask was administered. The patient remained in respiratory distress and was

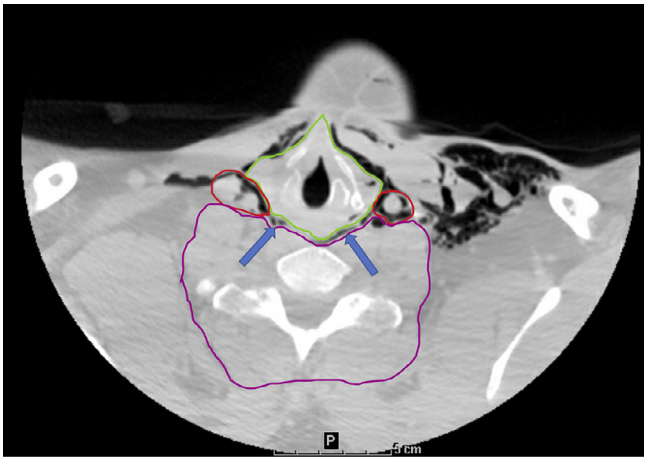

Fig. 2. Axial view from the computed tomography (CT) neck scan without contrast at the level of the infrahyoid neck. Extensive air emphysema outlines several compartments of the neck. The bilateral carotid spaces *(red)*, visceral compartment of the neck *(green)*, and prevertebral compartment *(purple)* are well defined. There is also air emphysema between the prevertebral and visceral compartments in the retropharyngeal and danger space *(blue arrows)*.

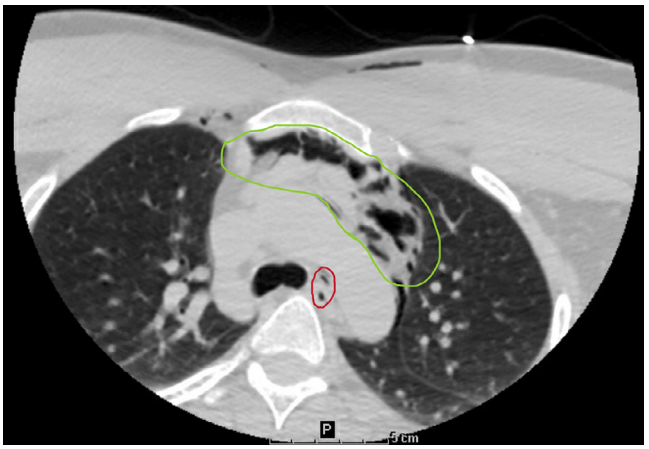

Fig. 3. Axial view of a computed tomography (CT) neck scan without contrast at the level of the fifth thoracic vertebrae. There is air in the anterior *(green)* and posterior *(red)* mediastinum.

intubated on the first attempt by using a Miller 1 blade and cricoid pressure.

After intubation, the patient was taken for computed tomography (CT) of the head to determine the cause of her seizure. CT showed diffuse nonvisualization of cerebral sulci, raising suspicions of cerebral edema vs meningeal process but no pneumocephalus. CT also showed air in the suprahyoid spaces of the neck, including the infratemporal, buccal, masticator, parotid, and lateral parapharyngeal spaces bilaterally (Figure 6).

The patient received weight-based intravenous vancomycin (20 mg/kg every 6 hours), ceftriaxone (100 mg/kg daily), and metronidazole (10 mg/kg every 8 hours) for 7 days to empirically treat meningitis and CE. A lumbar puncture was performed, and analysis of the specimen showed no bacterial growth and no herpes simplex virus type 1, so the antibiotic treatment was not altered. Four primary teeth, which were previously planned for extractions, were removed. The patient was successfully extubated 2 days later. Eight days after admission, she was discharged home with her neurologic baseline not showing any residual deficiency. Upon follow-up 5 days later, the

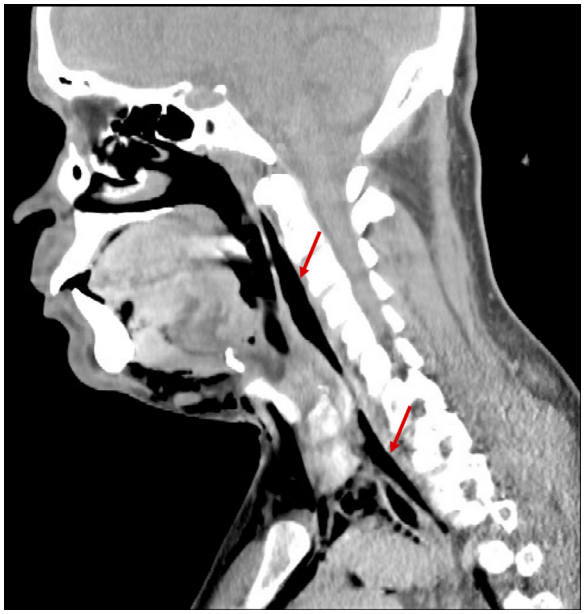

Fig. 4. Sagittal view of a computed tomography (CT) neck scan without contrast. There is air emphysema tracking from the posterior oropharynx to the mediastinum *(red arrows)* through the retropharyngeal/danger space. There is also prominent submandibular and superficial air emphysema.

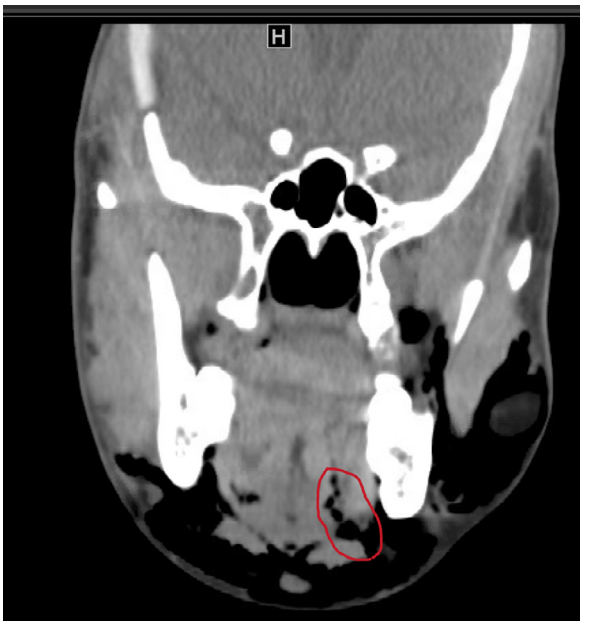

Fig. 5. Coronal view of a computed tomography (CT) neck scan without contrast at the level of the mandibular ramus. There is prominent air emphysema in the left masticator, buccal, and bilateral submandibular spaces with a boutonniere defect of the left mylohyoid *(red)*.

patient was found to have fully recovered, with return to her normal activities.

RADIOGRAPHIC INTERPRETATION

The noncontrast CT scan in case 1 demonstrates extensive subcutaneous and deep emphysema and highlights the compartments of the neck, their anatomic relationships, and interconnections.

Figure 1 presents an axial CT scan with lung window to emphasize the air in the suprahyoid neck. This scan demonstrates air dissecting into multiple spaces, including the buccal, masticator, parotid, retropharyngeal, and carotid spaces. Specifically, air surrounds the masticator space (outlined in red) and dissects along the lateral border of the masseter muscle, not only toward the buccal fat pad but also deep to the medial pterygoid muscle into the parapharyngeal fat pad. On the left, air dissects posteriorly into the left carotid space and along the jugulodigastric chain in the lateral neck deep to the sternocleidomastoid muscle. Air also highlights the potential retropharyngeal space, immediately posterior to the oropharyngeal space.

Figure 2 is an axial CT scan with lung window at the level of the infrahyoid neck and demonstrates air surrounding the visceral, prevertebral, and carotid compartments. There is also a sliver of air continuing in the retropharyngeal space.

Figure 3 presents an axial CT scan with lung window at the level of the mediastinum and lung apices. It demonstrates air in the anterior and posterior mediastinum. As this air is identified at the level of the fifth thoracic vertebra, below the proposed "trap door" inferior border of the retropharyngeal space, air within the posterior mediastinum likely traveled through the potential danger space.[12,13] Although the existence of the danger space is contested, some anatomists believe that this potential space lies directly posterior to the retropharyngeal space.[12,13]

Figure 4 is a sagittal reformatted neck CT scan demonstrating air within the potential retropharyngeal/danger space. By definition, the "danger space" lies posterior to the retropharyngeal space but anterior to the prevertebral compartment. This space exists because there is a thin layer of alar fascia that lies directly anterior to the prevertebral fascia.[12,13] Thus, if fluid or air penetrates posterior to the visceral compartment but stays anterior to the alar fascia, then it is contained in the retropharyngeal space. If fluid penetrates the alar fascia, then it occupies the danger space.[12,13] The difference in anatomic termination of the retropharyngeal space (T4) above the termination of the danger space (diaphragm) allows for radiographic determination of spaces because the alar fascia is not radiographically visible. The retropharyngeal space, which begins at the cranial base, extends inferiorly to the fourth thoracic vertebra. The danger space, which also begins at the cranium, extends to the diaphragm.[12,13] In this case, because we can identify pneumomediastinum at the level of the fifth thoracic vertebra, it is likely that this air travelled through the danger space.

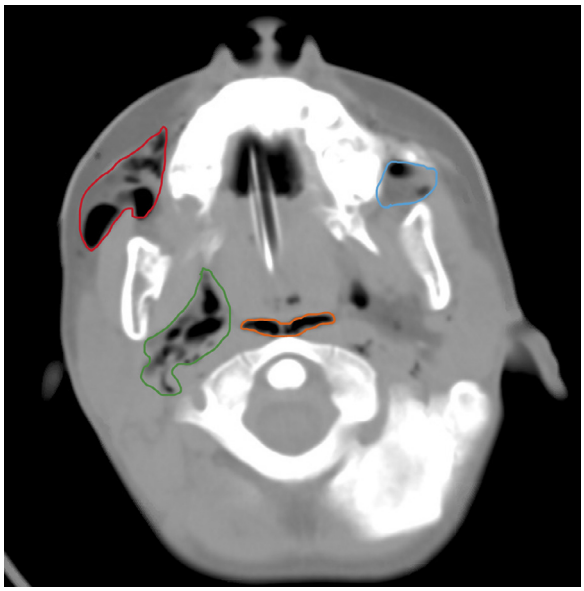

Fig. 6. Axial view of a computed tomography (CT) head scan without contrast at the level of C1. There is prominent air surrounding the masticator, parotid, and lateral pharyngeal spaces *(green)*, the retropharyngeal space *(orange)*, the right buccal space *(red)*, and the left buccal space *(blue)*.

Figure 5 is a coronal reformatted neck CT scan at the level of the oral cavity and shows air dissecting through a boutonniere defect of the left mylohyoid. This allows air to travel from the sublingual space to the submandibular space. A boutonniere defect is an embryologic defect, resulting in failure of fusion of the separately formed posterior and anterior portions of the mylohyoid muscle. This defect can be identified in the majority of the population but is not commonly visualized radiographically.[14] In our case, CE highlights the anatomic connection between the sublingual and submandibular spaces via the boutonniere defect.

A noncontrast CT scan of case 2 presents an axial view of a head CT scan with lung windows (see Figure 6). There is air in the suprahyoid neck involving the right buccal fat pad (red), left buccal space (blue), right parapharyngeal space (green), and air in the retropharyngeal space (orange).

DISCUSSION

These cases feature examples of pronounced CE caused by iatrogenic air that was forced into potential spaces in the head and neck, highlighting important anatomic relationships. Cases of CE have been reported previously in the literature, but these reports did not identify in detail the potential spaces to which the air spreads.[1,2,5,6] The 2 cases presented in this report demonstrate the massive infiltration of air into the potential spaces of the lower face and neck. Similar to previous cases, both our patients had undergone dental procedures on the lower molars before presentation to our institution.[7,8] Previous reports have described unilateral CE, but our 2 patients had bilateral CE.[1,2] Also, in contrast to previous reports, the patient in case 1 clearly demonstrated involvement of the danger space and the presence of a boutonniere defect of the mylohyoid.

Although some known cases of CE have been reported in the literature, CE is easily misdiagnosed. The rapid onset of swelling and possible dyspnea is consistent with the diagnosis of anaphylaxis. This is similar to our 2 cases, in which the patients were thought to have anaphylaxis. Regardless of the mechanism, CE can result in significant complications. Although most patients present with a painless swelling, some can have dysphagia with diet limitations, airway compromise requiring intubation, pneumomediastinum, and respiratory collapse, and, in some cases, death.[1,3,7,8,15]

It is important for the clinician to perform a thorough history and physical examination to understand the temporal connection of the swelling to any head and neck procedures and to rule out the possibility of a spreading infection. This will help the clinician discriminate iatrogenic CE from other causes. Additionally, in isolation, the radiographic imaging of CE would show free air in the deep spaces of the face and neck, which should raise concerns of necrotizing fasciitis. Mediastinal involvement is even more worrisome because mediastinitis has a mortality rate of up to 50%.[3]

If infection is high on the clinician's list of differential diagnoses, contrast-enhanced CT must be considered. Although noncontrast CT can identify subcutaneous emphysema, it cannot easily differentiate soft tissues from fluid collections. Missing a fluid collection associated with CE could lead to delayed diagnosis and poor outcomes. Our patients did not require contrast-enhanced CT because the temporal relationship of swellings appearing shortly after oral procedures made infection very unlikely.

Management of CE is often supportive. Broad-spectrum antibiotics are frequently used to cover oral flora that may have contaminated the sterile spaces of the neck.[2-4,6-9] Intubation can be required when airway obstruction is present.[11] Most times, however, air will resolve on its own over the course of a few days or weeks. If air does not resolve, it can be managed with negative pressure wound care.[10]

CONCLUSIONS

Intimate knowledge of the radiographic anatomy presented in these cases can help guide surgical intervention when the swelling is deemed infectious. Presence of a boutonniere defect near a sublingual abscess may lead to surgical opening and exploration of the submandibular space to prevent re-collection of air. Prudent recognition of air in the retropharyngeal or danger spaces may also warrant neck or chest CT to rule out mediastinitis. Radiographic studies are an indispensable tool for the surgeon, and careful review of the radiographs can lead to more efficacious treatment. Although none of these radiographic findings changed the management of our patients, the knowledge gained from identifying radiographic features will prove useful in future patient care.

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