## Mini-Review

### Cervicofacial emphysema: A systematic review

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**SUMMARY** Cervicofacial Emphysema (CFE) is a self-limiting condition, defined by the presence of air in face and neck. The purpose of the manuscript is to systematically review the existing literature on CFE evaluation and management for updated clinical understanding of this condition. A literature search was conducted of publications about CFE on PubMed and Google Scholar by identifying all the articles with key search terms "Cervicofacial Emphysema" and "Sub Cutaneous Emphysema". Inclusion criteria were case series published in English between 1980 and 2024. In total, 241 case series were selected and reviewed to determine presenting symptoms, clinical signs and predisposing factors associated with CFE. Average age at diagnosis was 38.1 years, male and female are almost equally affected. The most common presenting symptoms were face and neck swelling. The most common finding was crepitus. The condition was most commonly reported in patients undergoing dental procedures, otorhinolaryngology procedures, or in patients who experienced transient change in intra nasal/thoracic pressure. The management includes clinical monitoring, reassurance of the patient, antibiotic prophylaxis and monitoring to rule out pneumomediastinum. The odds of concurrent pneumomediastinum is highest in patients with abdominal procedures as an etiology of CFE.

Keywords cervicofacial emphysema, crepitus, face, neck swelling

#### 1. Introduction

Cervicofacial Emphysema (CFE) is defined as the presence of air within the facial planes (1). It often occurs in the setting of trauma such as injury to face, dental procedures, recent surgery, or conditions causing changes in intranasal/intrathoracic pressure (2). The most common reported presenting symptom is face and neck swelling (3). The most common clinical exam finding is crepitus (4). Clasically, for patients with cervicofacial emphysema, workup includes evaluation of mediastinal involvement (pneumomediastinum/pneumothorax) (5) The condition is non-fatal and self-limited. Therefore management is conservative and includes reassurance, clinical observation, and antibiotics. The majority of patients with cervicofacial emphysema make an uneventful recovery (3).

While previous studies enumerate the presenting clinical characteristics of CFE, there is a paucity of work describing the various triggering events / predisposing risk factors of this interesting clinical entity as well as risk factors for progression to pneumomediastinum. Thus, the purpose of the present manuscript is to systematically review the existing literature on various causes of CFE, evaluation, and management to create an up-to-date understanding of this condition with a relevant review of the literature.

#### 2. Methodology and Literature Search Strategy

We systematically searched clinical literature databases including pub Med and Google Scholar for case series on CFE published between 1980 and 2024 following PRISMA guidelines. Key search terms included "cervicofacial emphysema" and "subcutaneous emphysema". Inclusion criteria were articles that described clinicodemographic information for patients with CFE, articles that detailed patients older than 18 years of age, and articles published in the English language. Abstracts of all articles were independently screened by authors to assess eligibility. Articles with insufficient clinical details (on symptomatology, workup, and treatment) were excluded from review with discrepancies resolved by senior author. Clinical information of interest included patient age, sex, presenting symptoms, clinical exam findings, triggering events, comorbidities, and management. The full text of selected articles was then reviewed, and reference lists were examined for additional relevant studies. All data was accessed between May to June 2024.

#### 3. Results

Our initial search strategy led to abstracts of 363 publications which were screened for inclusion criteria eligibility (Figure 1). 122 records were excluded as they were not relevant to our current study. It was determined that 241 articles (detailing a total of 267 cases) had sufficient detail for full text review.

#### 3.1. Demographics

Among 241 articles from 1980-2024, we found 267 reported cases of CFE. This population contained a similar number of male (136, 50. 9%) and female (131, 49%) patients. The mean age of this cohort was 38.1 years, with an age range spanning from 19 to 85 years. The age distribution was as follows: 3.7% (n = 10) age < 20, 24.7% (n = 66) age 20-30, 15.7% (n = 42) age 31-40, 18% (n = 49) age 41-50, 13% (n = 36) age 51-60, 9.3% (n = 25) age 61-70, 13%(n = 35) age > 70. Age was not mentioned for 4 patients.

#### 3.2. Features of presentation

The most common presenting symptom in the analyzed

cohort of patients was face swelling, which occurred in 47% of patients (n = 125). Other common symptoms included neck swelling (18%, n = 48), and eyelid swelling (14%, n = 37). There were also sporadic complaints of facial pain (9%, n = 24), dyspnea (5%, n = 15), chest pain (4.8%, n = 13), dysphagia (2.6%, n = 7), and dysphonia (2%, n = 6). Clinical examination revealed the presence of facial crepitus in 55% (n = 146) of the patients. Pneumomediastinum was observed on radiological investigation in 28% (n = 74) of the patients. Other less commonly reported findings were pneumoperitoneum (1.4%, n = 4) and pneumopericardium (1%, n = 3).

#### 3.3. Predisposing factors

The most common triggering event / predisposing risk factor for development of cervicofacial emphysema was sudden nasal/intrathoracic pressure changes, which was identified in 31% (n = 83) of patients. This was followed by dental procedures (*i.e.* extraction (6), endodontic/restorative procedures (7)), which triggered CFE in 27.7% (n = 74) of the patients. Otherwise, the etiology of CFE was facial injuries in 15% (n = 41) of patients, otorhinolaryngology procedures in 8.2% (n = 22) of patients, thoracic in 5.6% (n = 15) and abdominal procedures in 2.9% (n = 8). Interestingly, spontaneous occurrence was found in 5% (n = 14) of patients. These triggering factors are detailed in Figure 2. Only 8.6% (n = 23) of patients reported having a smoking history.

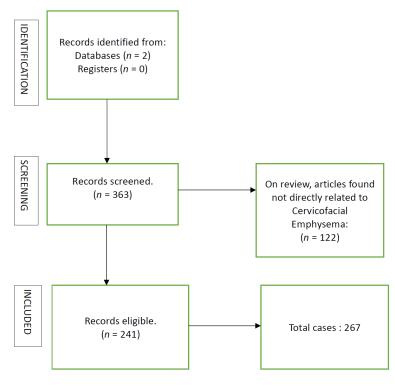


Figure 1. Identification of studies via databases and registers.

#### 3.4. Clinical outcomes

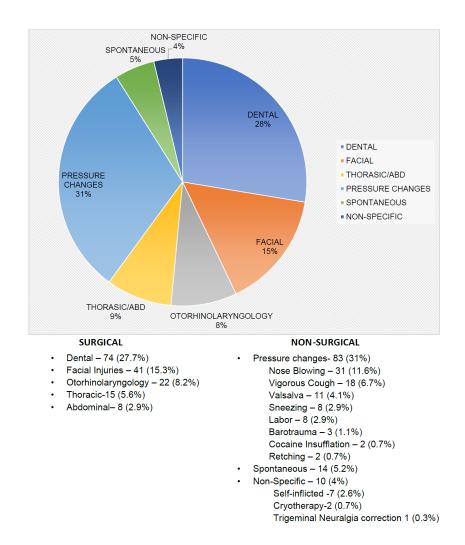
In our review, 74 cases developed pneumomediastinum either after or concurrently with CFE. The most common triggering events for CFE in this population were dental procedures (32%), abdominal procedures (20%), and facial injuries (20%). The odds of pneumomediastinum developing with CFE were 1.37 (95% CI: 0.77 - 2.46) for patients with dental procedures, 5.88 (95% CI: 2.37 -14.56) for patients with abdominal procedures, and 1.63 (95% CI: 0.81 - 3.29) for patients with facial injuries.

In our review, the average time from presentation to resolution of CFE was noticed to be around 1 week. 208 (77.9%) cases resolved completely in 7 days and 44 (16.4%) cases resolved completely in 8-14 days. There was no follow-up reported in 15 (5.6%) cases. The average number of days from presentation to CFE resolution was 2.4 days for dental procedures, 2.3 days for otorhinolaryngology procedures ,3.1 days for thoracic procedures, and 4.4 days for abdominal procedures.

In our review of CFE, the most common treatment was antibiotics, which was given in 165 (61.7%) cases. Surgical aspiration was required due to eyelid closure in 6 (2.2%) patients, and canthotomy was required for 2 (0.7%) patients. Oxygen was given in 10 (3.7%) patients due to desaturation. 2.2% (n = 6) of patients passed away due to other pre-existing conditions. 10 out of 22 (45%) cases of pneumothorax required placement of chest tube.

#### 4. Discussion

Cervicofacial emphysema is an uncommon clinical entity which is often unrecognized and may be misdiagnosed as anaphylaxis, angioedema, or internal hemorrhage( $\delta$ ). The pathophysiology of CFE is inclusion of air, typically under pressure, into subcutaneous tissues. This air has the potential to spread along fascial planes in the neck and mediastinum(9). Air typically gains access into these subcutaneous planes when the integrity of the oral mucosa is interrupted or and intraoral pressure is increased (1). The clinical presentation is characterized by the sudden onset of facial swelling. Crepitus, pain, and tenderness may be noted as well (10). Retrosternal pain, dyspnea and Hamman sign may indicate concurrent pneumomediastinum. Diagnosis is suspected on clinical examination and may be confirmed/monitored with



radiologic imaging (X-Ray/CT) which will demonstrate the presence of subcutaneous air (11).

Dental procedures are considered to be the most common source of CFE (9). High speed dental air turbine drills(12,13) are used in endodontic treatment(14) and surgical tooth extraction(7). During these interventions, compressed air may be forced into subcutaneous layers at a high pressure. Hydrogen peroxide usage has also been implicated in the development of CFE during endodontic treatment(7,15). Our study, confirms these findings and reports on relative incidences of other triggering events within the literature – the most common etiologies of CFE in our study were dental procedures (27.7%) as well as activities leading to sudden change in intrathoracic pressure (*i.e.* from repeated nose blowing, sneezing, vigorous coughing, retching, or valsalva) (31%).

Treatment of cervicofacial emphysema is usually conservative which includes bed rest, observation and reassurance, as the condition is benign and selflimiting (11). In fact, most patients with cervicofacial emphysema recover within 7 days (16). This was true for patients in our cohort, as most recovered within 1 week on average. It has been reported that supplemental oxygen can hasten the resolution of subcutaneous emphysema because oxygen, which replaces the air, is more readily absorbed (14). Surprisingly, oxygen was given to only 10 (3.7%) patients included in our study. The use of supplemental oxygen to improve time to recovery in patients with CFE needs to be the subject of future studies.

Complications of CFE, such as sepsis, can develop when microorganisms from oral cavity migrate to the mediastinum. Generally, broad-spectrum antibiotics are administered to avoid development of such severe infections (17). In our review, antibiotics were given in 165(61.7%) patients. The data collected in this study were insufficiently granular to understand whether or not the administration of antibiotics helped decrease incidence of sepsis. However, we support this treatment approach, as there is a potential for spread of bacterial organisms from subcutaneous tissue to mediastinum which may lead to significant adverse events. The authors also feel that most patients with CFE can be managed conservatively at home with supportive measures. In our review, supportive measures provided to patients included techniques to decrease airway pressure (*i.e.* telling patients to avoid blowing their nose or bearing down for a few days) (18).

Life threatening complications of CFE include pneumothorax(5), pneumomediastinum(5,8,19), and pneumopericardium(20). Urgent surgical decompression may be required if cardiovascular collapse or large airway obstruction occurs (8). Patients may be observed in hospital if there is any suspicion of impending respiratory compromise. In our review, a majority of the cases of pneumothorax cases were mild and selflimited, however 10 out of 22 cases of pneumothorax required chest tube drainage. Pneumomediastinum was seen in 74 patients included in our study (28%). The odds of pneumomediastinum in patients with CFE were significantly higher in those who had an abdominal procedure as a trigger for CFE (OR 5.88, 95% CI: 2.37 - 14.56). These finding is easily conceivable as subcutaneous air in the head and neck originating from the abdomen must pass through the mediastinum. Still, this is the first publication to indicate that CFE secondary to recent abdominal procedure is significantly associated with comorbid pneumomediastinum, and should therefore prompt chest imaging for further interrogation. The importance of chest imaging in these patients is underscored by the fact that resolution of CFE for patients with recent abdominal procedures takes a longer period of time (4.4 days) as compared to CFE from other etiologies (~2 days).

CFE from dental procedures is known to sometimes cause pneumomediastinum/pneumothorax secondary to alveolar rupture and escape of air into perivascular planes (5,6). Additionally, the roots of the lower posterior molar teeth communicate directly with sublingual and submandibular space. The sublingual space communicates with the pterygomandibular, parapharyngeal and retropharyngeal spaces, a main route of communication from oral cavity to mediastinum(16,21). However, interestingly, while a majority of patients with pneumomediastinum in our review had dental procedures as an etiology of CFE (32%), CFE from dental procedure was not significantly associated with the development of pneumomediastinum.

In the present analysis, 6 out of 267 patients passed away, all due to complications of underlying comorbidities such as tension pnuemomediastinum(22), massive cerebrovascular accident(23), or acute respiratory distress syndrome (24). Most of these patients were also within the elderly age group (mean 56 years + 28.2 years).

#### 5. Conclusion

In conclusion CFE is generally a self-limiting condition that occurs due to dental, thoracic, abdominal, or otorhinolaryngology procedures. It may also occur sporadically where there are transient changes in intranasal/ thoracic pressure. Diagnosis is clinical and may be confirmed with radiological examination to determine the extent of gas dissection into the subcutaneous space. CFE from abdominal procedures have a significant association with comorbid pneumomediastinum. Management is conservative and includes reassurance, supplemental oxygen, clinical observation and antibiotics. The condition is non-fatal and self-limiting. The majority make an uneventful recovery.

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*Conflict of Interest*: The authors have no conflicts of interest to disclose.

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