

therapeutic decision making. Sclerosing agents, steroids, radiotherapy, lasers, cryotherapy, embolization, and even continued observation represent reasonable approaches to the vascular aspect of the lesion.<sup>6,7</sup> Nevertheless, when feasible, surgical excision is the treatment method of choice and will also succeed in eliminating the phlebolith.

Our patient with the IMH opted for no treatment because she stated that other than a moderate cosmetic asymmetry, she had no subjective problems. She did agree to return every 6 months for evaluation, but she failed to present for further follow-up.

In our patient with the VM, significant symptoms had developed that prompted her to seek therapy. Normal masticatory function inevitably caused trauma with associated hemorrhage. Furthermore, she became increasingly embarrassed about her speech impediment. She was referred to an interventional radiologist who plans to embolize the lesion.

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68:1976-1982, 2010

# Cervicofacial Subcutaneous Emphysema: Case Report and Review of Literature

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Subcutaneous emphysema is by no means a contemporary discovery, having been mentioned more than 180 years ago. Previously described as pneumomediastinum, Laenec, in 1827, termed the disease “interlobar emphysema.”<sup>1</sup> Reports described subcutaneous emphysema in a patient with a violent coughing episode in 1850.<sup>2</sup> The first official report by an anonymous investigator of the phenomenon stemmed from a musician playing the bugle after a dental extraction at the beginning of the 20th century.<sup>3</sup> Macklin in 1939 and 1944 proposed a pathophysiologic clarification,<sup>2</sup> which led to the rudimentary 4-tier classification according to the etiology of cervicofacial emphysema in 1957, derived by Shovelton. The various etiologies of

cervicofacial emphysema have included increased intraoral pressure at the site of a mucosal injury or the allowance of compressed air into an intraoral wound. Additional causes were theorized, such as trauma from a challenging extraction, especially a mandibular third molar, and, finally, no apparent cause.<sup>3</sup> Non-head and neck injuries, such as penetrating trauma to the gastrointestinal or respiratory systems, can also result in the disease. Gases trapped under the tissue can lead to infection, such as gas gangrene.<sup>4</sup> Iatrogenically, improper placement of, or a nonfunctioning, chest tube for treatment of a pneumothorax can also cause it.<sup>2</sup> This can lead to the pressure of the alveoli being greater than the surrounding tissues.<sup>4</sup> Cervicofacial emphysema can be associated with a positive Hamman's sign, which is a crunching sound heard over the pericardium, and is associated with mediastinal emphysema. The etiology of subcutaneous emphysema, whether traumatic, iatrogenic, incidental, or pathologic, all stems from the introduction of gas between the tissue layers (Table 1).<sup>2,4-21</sup>

## Report of a Case

On August 5, 2007, an 18-year-old black man who had sustained blunt trauma from fists to his head, but no other bodily trauma, presented to the Kings County Hospital Center Emergency Department. He was admitted to the surgical intensive care unit by the trauma service for airway monitoring owing to the substantial cervicofacial edema. On consultation, the patient complained primarily of left mandibular pain and swelling but not of dyspnea or orthopnea. He reported a medical history of uncomplicated childhood asthma with no hospitalizations, intubations, or attacks during the previous 10 years. The patient had a surgical history of an appendectomy but no history of facial trauma. He denied known drug or food allergies, denied any medication use, intravenous drug abuse, or tobacco consumption; however, he was intoxicated and uncooperative.

On admission, the patient was hemodynamically stable and had a low-grade fever. During the head and neck examination, the patient had significant left facial swelling inconsistent with the severity of the left mandible fracture. The occlusion was stable and reproducible without any step deformities or mobile segments. Although the patient complained of left inferior alveolar nerve paresthesia, this was attributed to the severe left facial edema. The neck demonstrated bilateral crepitus that extended medially from both sternocleidomastoid muscles to the thyroid cartilage and inferiorly from the border of the mandible to the clavicle and suprasternal notch. This crepitus was most notable along the clavicle, zone 1 region, of the neck owing to his slim frame. The remaining physical examination findings were nonsignificant.

After the appropriate workup, consisting of a complete blood count, comprehensive chemistry panel, and coagulation studies, only the white blood cell count of 21,000 was notable. Head and facial computed tomography (CT) scans with respective reconstructions demonstrated air tracking from the face caudally. A barium swallow test was negative. The neck CT scan demonstrated a large amount of air in the neck and retropharyngeal space (Figs 1, 2). Chest radiography and noncontrast chest CT scan were obtained (Fig 3). No pneumothorax was present, but the subcutaneous emphysema extended through the superior mediastinum to the level of the carina. Otolaryngology and cardiothoracic surgery staff were consulted, and they wished to follow-up the patient closely in a monitored setting. Regarding the left mandible fracture, the facial CT scan showed a nondisplaced left body of the mandible fracture. Dental panoramic studies were later obtained with the patient in a more stable condition to better study the fractures and the dental relationships (Fig 4).

The treatment plan for the left angle mandible fracture consisted of a closed reduction of the left angle owing to the nondisplaced nature of the left angle fracture to be performed in the clinic with the patient under local anesthesia. After optimization and clearance from the trauma and cardiothoracic service, the patient underwent the procedure without issue and was then treated directly by the oral and maxillofacial service. The patient, since admission, had been receiving intravenous clindamycin 600 mg every 6 hours and dexamethasone 8 mg every 8 hours. For his

**Table 1. ETIOLOGY OF SUBCUTANEOUS EMPHYSEMA**

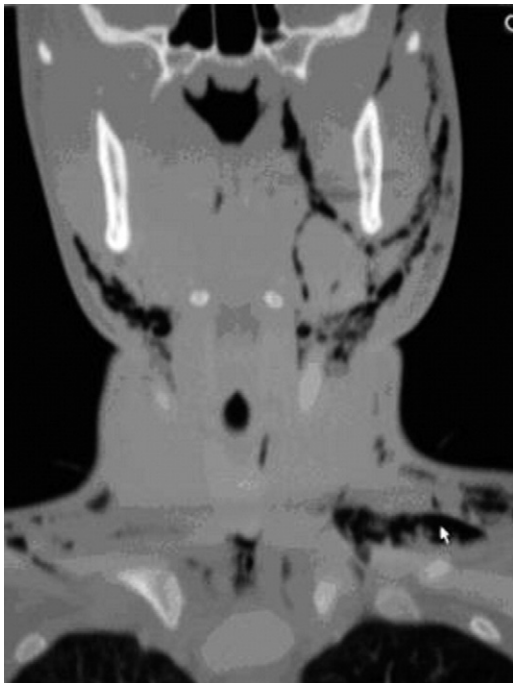
Trauma	Iatrogenic	Incidental	Pathologic
Blunt chest <sup>2,10,19</sup> (27% of rib fractures, tension pneumothorax)	Anesthesia <sup>8,9,17</sup> (endotracheal/nasotracheal intubation, laparoscopy)	Barotrauma <sup>2,6,13,14,18</sup> (diving, positive pressure ventilation)	Infections <sup>5</sup> (anaerobic, clostridia, mixed, gas-forming facultative aerobic)
Penetrating chest <sup>9,21</sup> (stabbing, gunshot)	Chest surgery <sup>7,20</sup> (improper chest tube placement, pneumonectomy, thoracotomy)	Labor <sup>2,11</sup> (1 in 2000-100,000 deliveries)	Neoplasia <sup>5</sup>
Facial bones <sup>5</sup> (sinus fractures)	Neck surgery <sup>8</sup> (esophageal, cricothyrotomy)	Foreign body <sup>12</sup>	
Neck <sup>12,16</sup> (tracheal/laryngeal tears, esophageal rupture)	Pneumatic tools <sup>15</sup> (dental high-speed handpieces)	Heimlich maneuver <sup>2</sup>	

Data from Parker et al,<sup>2</sup> Maunder et al,<sup>4</sup> Wong and McGuire,<sup>5</sup> Papiris and Roussos,<sup>6</sup> Lefor,<sup>7</sup> Brooks,<sup>8</sup> Peart,<sup>9</sup> Wicky et al,<sup>10</sup> Jain and Vanner,<sup>11</sup> Findlay et al,<sup>12</sup> Criner and D'Alonzo,<sup>13</sup> Rankine et al,<sup>14</sup> van der Molen,<sup>15</sup> Kosmas and Polychronopoulos,<sup>16</sup> Pan,<sup>17</sup> Conetta et al,<sup>18</sup> Schnyder and Wintermark,<sup>19</sup> Long et al,<sup>20</sup> and DeGowin.<sup>21</sup>

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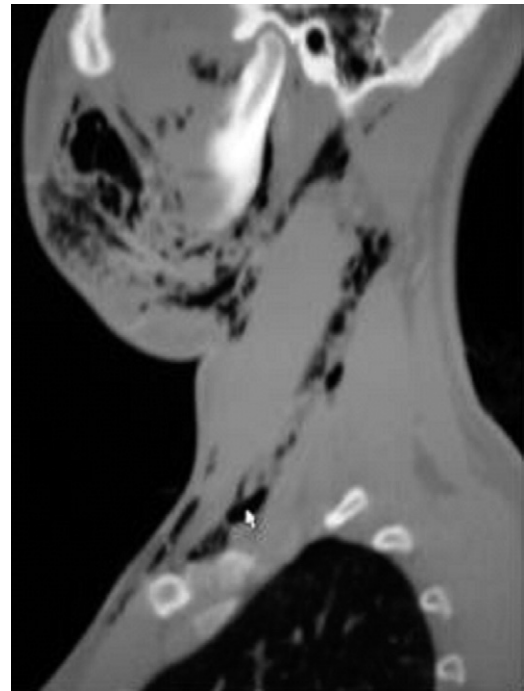
remaining stay, a broader spectrum antibiotic, ampicillin 2 g with sulbactam 1 g, was given every 6 hours. Because of the limited oral intake, the patient's liquid diet was supplemented with intravenous fluid for hydration. Supplementary oxygen using a mask or cannula was not given during the patient's hospitalization.

Throughout his stay, the patient remained afebrile, with his other vital signs within an acceptable range. While in the intensive care unit, blood was taken for serial laboratory tests, and the chest radiography and chest and neck CT scans were repeated (Figs 5-7). The emphysema appeared to be resolving, and the white blood cell count initially decreased to 12,000 but then increased slightly to 15,000, most likely owing to the systemic steroid administration. The patient's general clinical improvement was evident in the resolution of the crepitus of the left neck, decreased edema, and overall disposition. He tolerated the hospital stay, procedure, and diet and returned to normal activities very well by discharge 5 days later. He was discharged with instructions to take oral amoxicillin trihydrate 875 mg and clavulanic acid 125 mg for 7 days. The patient followed up in the dental clinic approximately 7 days after discharge for a panoramic radiograph and evaluation (Fig 7). After approximately 2 weeks, a preliminary chest radiograph was obtained that demonstrated significant resolution of the subcutaneous emphysema. Although mildly evident in the neck, it had disappeared from the mediastinal region. Because of the patient's vast clinical improvement, no additional radiologic studies were performed. He was followed up on successive weekly intervals and had his maxillomandibular fixation released and removed 1 month postoperatively. During follow-up, no additional signs of subcutaneous emphysema were evident.



**FIGURE 1.** Coronal section of neck CT scan showing diffuse subcutaneous emphysema bilaterally (day 1).

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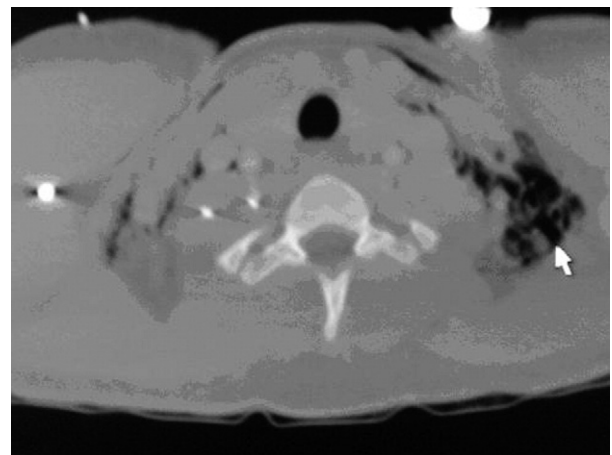


**FIGURE 2.** Sagittal section of neck CT scan showing subcutaneous emphysema extending to clavicle (day 1).

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## Discussion

On examination, a patient with subcutaneous emphysema can present with a nasal voice quality,<sup>22</sup> swelling of the face and neck, a sore throat, neck and chest pain, dysphagia, dyspnea, dysphonia, and wheezing.<sup>2</sup> The trapped air can spread to the abdomen and extremities, because of the lack of separation between the fatty tissues.<sup>19</sup> The palpation of crackling nodules similar to the texture of tissue paper or "Rice



**FIGURE 3.** Axial section of chest CT scan (day 1).

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**FIGURE 4.** Preoperative panoramic radiograph (day 4).

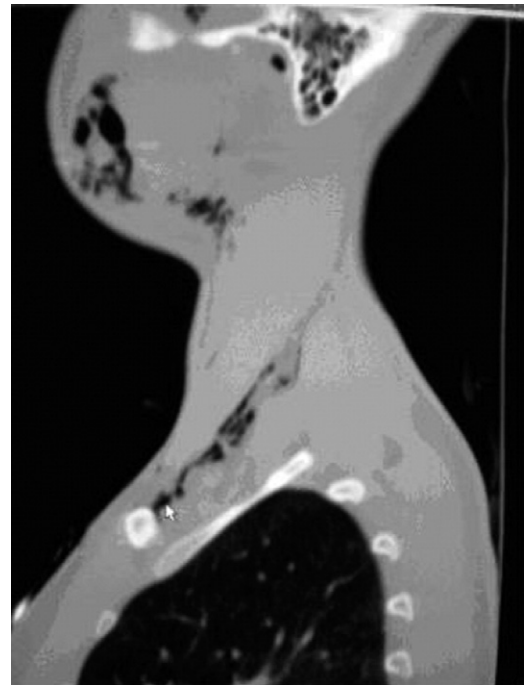
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Krispies” under the skin that can even burst to touch are pathognomonic of subcutaneous emphysema.<sup>20,21</sup> Patients with pneumomediastinum might demonstrate Hamman’s sign, best described as a “crunching” sound that can be auscultated in the retrosternal area or heard during systole and is accentuated on expiration. Approximately 65% of patients with mediastinal emphysema exhibit a positive Hamman’s sign.<sup>23</sup> The etiology of subcutaneous emphysema can be complex. Various mechanisms can cause subcutaneous emphysema, including pneumothorax, dental procedures, laparoscopic procedures, ruptured bronchus, ruptured esophagus, gas gangrene, laryngeal injury, necrotizing enterocolitis, penetrating trauma, percu-



**FIGURE 5.** Coronal section of neck CT scan showing resolving subcutaneous emphysema (day 3).

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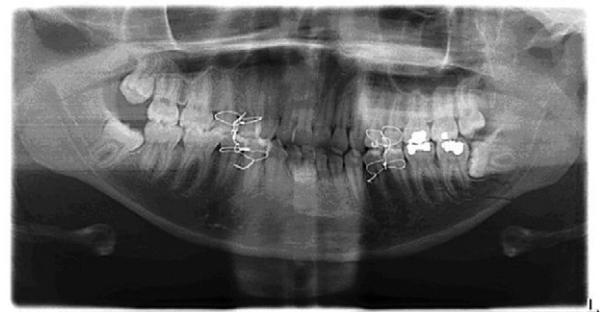


**FIGURE 6.** Sagittal section of neck CT scan showing decreasing subcutaneous emphysema (day 3).

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taneous catheters (central lines), perforated hollow viscus, and pneumatosis intestinalis. These mechanisms can result in subcutaneous emphysema from 4 possible modes, 3 of which involve barrier disruptions, such as cutaneous (ie, trauma, tracheostomy), mucosal (ie, facial fractures, endoscopy), and alveolar membrane (ie, labor, asthma). The fourth includes bacterial infection from organisms that are gas forming, such as *Clostridia* and facultative aerobes.<sup>5</sup>

CT has become the reference standard study in the diagnosis of subcutaneous emphysema. These studies can be obtained with or without a contrast medium. Contrast might assist in differentiating between loculations such as active infection from radiolucencies



**FIGURE 7.** Postoperative panoramic radiograph (day 12).

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**Table 2. HOUNSFIELD UNITS OF COMMON SUBSTANCES**

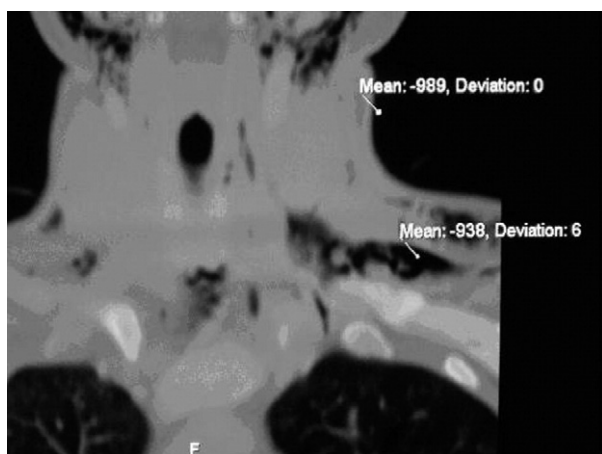
Substance	Hounsfield Units
Air	-1000
Fat	-120
Water	0
Muscle	+40
Bone	≥+400

Data from Hounsfield.<sup>24</sup>

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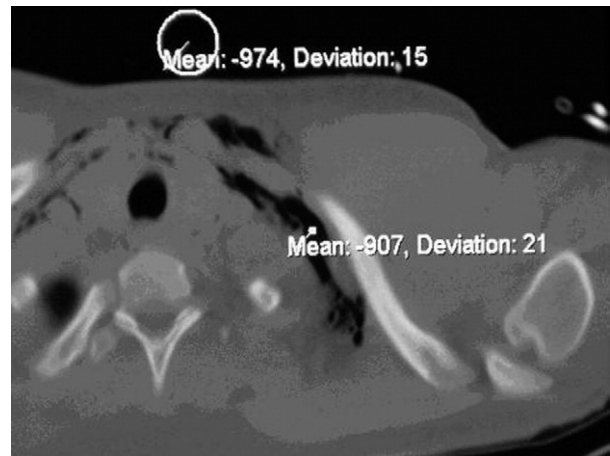
such as subcutaneous emphysema. This has proved quite accurate, especially with the foundation of CT-guided technology and its digital interpretation using Hounsfield units. Sir Godfrey Newbold Hounsfield, a pioneer in CT imagery, centered the development of the scan on radiodensity. The Hounsfield units became the quantitative measuring scale relating the radiodensity of distilled water at a standard pressure and temperature (0 Hounsfield units) to the internal anatomy of water-organized structures existing in spaces of air (Table 2).<sup>24</sup> In the present case, the presence of subcutaneous emphysema was confirmed by evaluating the Hounsfield units of the noted subcutaneous radiolucencies on the CT scan. The Hounsfield units of these areas was consistent with the presence of subcutaneous air compared with extraoral air (Figs 8, 9). This finding confirmed a case of subcutaneous emphysema associated with a mandibular fracture.

Additional studies might be necessary to evaluate the suspected location and extent of the injury. Panendoscopy can be used in the examination of laryngotracheal wounds. For an overall noninvasive exam-



**FIGURE 8.** Comparison of Hounsfield units of coronal neck CT scan (day 1).

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**FIGURE 9.** Comparison of Hounsfield units of axial chest CT scan (day 1).

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ination, fiberoptic nasal pharyngoscopy is suitable. Specifically, the larynx can be evaluated by flexible laryngoscopy. The trachea can be evaluated by bronchoscopy, and esophageal damage can be evaluated with an esophagram, a barium swallow test,<sup>25</sup> or esophagoscopy. Even with routine mandibular molar extractions, these teeth communicate directly with the sublingual and submandibular spaces, which in turn, abut the parapharyngeal and retropharyngeal spaces. Similar to cellulitis leading to Ludwig's angina, subcutaneous emphysema can follow similar planes and track toward the mediastinum and through to the pleural cavity, pericardium, and retroperitoneum, resulting in pneumoperitoneum or cardiac failure from pneumopericardium.<sup>26</sup> As many as 50% of pneumomediastinal cases are involved with pneumothoraces, resulting in decreased venous return to the heart, rapid vascular collapse, and, concluding, with malignant pneumomediastinum.<sup>25</sup> Air between the fascial planes can lead to challenging intubations if airway protection is desired, especially with the presiding neck fullness and resulting airway distortion.<sup>27</sup> Airway management is the greatest priority, sometimes necessitating, especially in instances of significant cervical emphysema, an endotracheal intubation or tracheostomy.<sup>26</sup>

Management consists of a 7-prong approach, of which not all points will necessarily be used, depending on the extent of the affliction. First, monitoring the oxygen saturation is of primary concern for airways at risk of severe subcutaneous emphysema with mediastinal, laryngotracheal trauma, or pneumothoraces. This measure, even limited to just observation, is a basic necessity, whether in a clinical or institutional setting, and can help determine the stability of the patient and viability of additional care. Second, anti-

biotic coverage with a broad-spectrum agent that covers the more common head, neck, sinus, and skin microflora is recommended and should include prophylaxis against necrotizing fasciitis.<sup>28</sup> With knowledge of the location and mechanism, the appropriate antibiotic can be selected: penicillin, or, alternatively, a cephalosporin, because oral/pharyngeal flora are sensitive to both.<sup>27</sup> Of concern is that contaminated air that is forced subcutaneously could lead to gas gangrene with bacteria such as *Clostridium perfringens*, group A streptococci, *Staphylococcus aureus*, and *Vibrio vulnificus*. Certain data have reported that gas gangrene is a major cause of subcutaneous emphysema from gas-producing organisms.<sup>29</sup> Third, the use of 100% oxygen therapy by way of a nonrebreathing mask or even nasal cannula can resolve the emphysema.<sup>3</sup> The composition of air is approximately 78% nitrogen and 21% oxygen, with traces of other gases. Oxygen is more readily absorbed than nitrogen in the soft tissues. Because of oxygen's increased pressure gradient, nitrogen is resorbed from the soft tissue.<sup>25,27</sup> Regarding gas gangrene, hyperbaric oxygen therapy has been suggested, especially for patients with an unknown etiology.<sup>29</sup> Fourth, suppressors of the offending mechanism to reduce or eliminate additional injury; stool softeners to decrease strong Valsalva maneuvers, decongestants as a sinus precaution to reduce the forceful passage of air, and antitussives to suppress coughs can be used.<sup>5</sup> Intravenous fluids are purely for hydration in cases in which the orofacial edema, laryngeal damage, or dysphagia prohibits or hinders oral intake for a short period.<sup>6</sup> Systemic steroids can be used to reduce the soft tissue edema, provided the patient has no contraindications such as poorly controlled diabetes.<sup>7</sup> Serial CT scans are not only important for diagnosis, but also to gauge the progression of treatment and patient improvement.

With subcutaneous emphysema spread past the mediastinum, the risk of pneumothorax is increased with positive pressure ventilation such as a general anesthetic procedure without a chest tube for decompression. Overall, positive pressure ventilation is contraindicated. Avoidance of nitrous oxide is important, because it can exacerbate a pneumomediastinum or pneumothorax. This occurs because the blood/gas partition coefficient of nitrous oxide is 34 times greater than that of nitrogen, leading to an inflation of the gas in the air-filled cavity owing to the inability of the nitrogen to escape.<sup>23</sup>

The etiology is key if subcutaneous emphysema is included in a differential diagnosis. However, the etiology of the present case remains a mystery. The patient denied any loss of consciousness from the attack and claimed to recall the entire event accurately. He also did not admit to any adverse behavior during or after the assault that could have led to the

episode of emphysema. The extent of the subcutaneous emphysema and the point of fracture as the most likely cause of the subcutaneous emphysema did not seem to correlate with each other. Perhaps defensively, after being struck, the patient might have been gasping, and his inspirations and expirations were meeting resistance. The expirations might have mimicked a Valsalva-like maneuver and forced air through the gingival and mucosal lacerations the patient sustained from his left angle of the mandible fracture. From there the air traveled to the floor of the mouth to the sublingual space and posteriorly. Once encountering the pterygomandibular and lateral pharyngeal spaces, the air traveled in the deep fascial spaces of the neck.<sup>23</sup> Regardless of the mechanism of injury, its etiology, or severity, the treatment protocol of subcutaneous emphysema involves airway management, infection prophylaxis, and radiologic assessment. This management, whether medical or surgical, is left to the judgment of the clinician, concerning the extent of the measures to be taken and the overall stability of the patient. Although the treatment of subcutaneous emphysema is conservative and observatory, if not treated, it can lead to a compromised airway, pneumomediastinum, or, even, pneumothorax.

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# Squamous Cell Carcinoma of the Maxillary Sinus Associated With Nevroid Basal Cell Carcinoma Syndrome: Report of a Case With 21-Year Evaluation

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The first recognition of nevroid basal cell carcinoma syndrome (NBCCS) occurred in 1960 by 2 scientists, Gorlin and Goltz.<sup>1</sup> It is an autosomal dominant syndrome distinguished by various signs and symptoms. The major diagnostic features include multiple keratocystic odontogenic tumors (KCOTs) of the maxilla and mandible, multiple cutaneous basal cell carcinomas, skeletal abnormalities, palmar pits, calcification of the falx cerebri, parietal and temporal bossing, and mental retardation.<sup>1-3</sup>

Because the chief complaint of these patients is usually related to the infected jaw cysts, the first

diagnosis is usually performed by a dentist. The presence of accompanying neoplasms is also very critical; they can include medulloblastoma,<sup>4-6</sup> ovarian fibroma,<sup>7</sup> rhabdomyosarcoma, melanoma,<sup>8,9</sup> meningioma,<sup>4</sup> breast tumor, uterine tumor, thyroid tumor, cardiac tumor, and Hodgkin's disease.<sup>8,10,11</sup>

In the oral and maxillofacial area, fibrosarcoma,<sup>12</sup> ameloblastoma, odontogenic myxoma of the upper jaw,<sup>13</sup> squamous cell carcinoma (SCC) of the mandible,<sup>14,15</sup> and 1 case of SCC of the maxillary sinus have been reported.<sup>11</sup> In the present study, we report on the occurrence of SCC of the maxillary sinus in a patient with NBCCS.

## Report of a Case

An 8-year-old boy with a cystic lesion of the left mandible was seen in the Department of Oral and Maxillofacial Surgery of Mashhad Faculty of Dentistry in 1987 (Fig 1).

Under local anesthesia, marsupialization of the cyst was performed. Follow-up radiography (Fig 2) in 1988 revealed the eruption of the lower left premolars. Despite our strong recommendation to continue treatment, he did not return until 11 years later (in 1998). The chief complaint on this occasion was severe infection owing to a mandibular fracture due to a traumatic accident. Large cystic lesions in the body of the left mandible and ramus of the right mandible

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