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Surgical Emphysema Following Third Molar Extraction in a Patient with Gilles de la Tourette's Syndrome

Abstract: Surgical emphysema is a rare complication of dental surgery, usually associated with the inappropriate use of an air turbine drill. We present a case of extensive subcutaneous emphysema, pneumomediastinum and bilateral pneumothorax following surgical removal of a lower third molar tooth. This appears to have arisen due to motor tics associated with Gilles de la Tourette's syndrome. The clinical features and management are outlined and the current literature pertaining to surgical emphysema related to oral surgery is reviewed.

CPD/Clinical Relevance: It is important that clinicians are made aware of the potential complications that can occur following surgical removal of teeth and the appropriate management should they arise.

Dent Update 2017; 44: 669–671

Surgical emphysema is a well-documented, rare complication of dental procedures ranging from dental extractions, to crown preparations and endodontic treatment.¹ For this to occur, air must be forced under pressure through a breach in the oral mucosa into the submucosal tissue. Most commonly this follows raising of a mucoperiosteal flap. Air can also travel more widely via deep fascial planes to the

pleural and mediastinal spaces.²

Tourette's syndrome is a neurological condition characterized by repetitive involuntary noises and movements called tics, that occur for at least one year.³ These may be simple or complex, depending on the types of movement and sounds made. They are thought to increase in frequency when the individual experiences anxiety or excitement. It was originally described by French neurologist Gilles de la Tourette in 1885. The cause of this syndrome is unknown but thought to be due to misfiring in the basal ganglia. Approximately 1 in 100 people are affected by Tourette's.⁴ It is typically first noted in childhood at around 7 years of age.⁵ It usually resolves spontaneously, but can be a life-long condition in around one third of patients. Treatment includes behavioural therapy to reverse tic habits

and pharmacological therapy. Despite these treatments, some cases remain refractory.³

In a review of the literature, only one other case of subcutaneous emphysema associated with Tourette's syndrome has been reported, in which the motor tics led to extensive cervico-facial tissue emphysema, including the supraclavicular region.⁶ In this case report, we describe more extensive surgical emphysema leading to pneumomediastinum and bilateral pneumothorax caused by motor tics of Tourette's syndrome following surgical extraction of a mandibular third molar tooth.

Case report

A 54-year-old man was referred to the Oral Surgery department of Bristol Dental Hospital by his General Dental Practitioner for extraction of an

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unrestorable carious lower right third molar. His past medical history included Tourette's syndrome and an allergy to penicillin. Radiographic examination demonstrated a disto-angularly impacted, carious LR8 with bulbous roots. The apex of the distal root terminated close to the inferior alveolar nerve canal (Figure 1). Following discussion of treatment options, it was decided to extract under local anaesthesia with intravenous sedation. He attended for surgical extraction of the tooth with a member of staff at the dental



Figure 1. Sectional dental panoramic tomograph showing the lower right third molar.



Figure 2. Frontal view showing swelling right side of face and neck.

hospital, during which a mucoperiosteal flap was raised to afford osteotomy and tooth-sectioning using a slow-speed surgical drill with irrigation. The flap was re-approximated with 4–0 resorbable sutures.

A repetitive motor tic was noted after the procedure whereby the patient forced air from the oral cavity through pursed lips with the tongue interposed, effectively 'blowing a raspberry'.

Five hours later, the patient attended the Emergency Department as a result of increasing cervicofacial swelling and hoarseness of voice, which had occurred over the preceding two hours. He reported having some difficulty in swallowing food. Initially it was thought by the emergency department staff that he was having an anaphylactic reaction. Vital signs were unremarkable, however. Clinical examination revealed extensive bilateral erythematous, tender swelling, palpation of which elicited crepitation. The swelling was more extensive on the right side, from the temporal and inferior orbital region through the neck to the mid-sternum (Figures 2 and 3). Intra-orally, the surgical site appeared healthy and the sutures remained *in situ* (Figure 4). Throughout the examination the patient repeatedly performed the motor tic as previously described.

A computerized tomography scan of the neck and thorax showed extensive subcutaneous emphysema involving the face, neck and chest wall. Pneumomediastinum and small bilateral pneumothoraces with mild atelectasis were also noted (Figures 5 and 6).

The patient was reviewed by the Cardiothoracic Surgery team and admitted for close monitoring, and prophylactic intravenous clindamycin 600 mg QDS was prescribed. No surgical intervention was indicated.

His swelling resolved spontaneously over the next few days and he was discharged after 3 days with a course of oral clindamycin. At review, 10 days later, the surgical emphysema had completely resolved and nothing abnormal was detected at the intra-oral surgical site.

Discussion

Review of the literature reveals that the aetiology of subcutaneous emphysema can be iatrogenic, traumatic, infectious or spontaneous in origin.⁷ The majority of cases of subcutaneous emphysema are iatrogenic and in dentistry are most commonly caused by the inappropriate use of the air turbine dental drill. In a recent review,¹ this was implicated

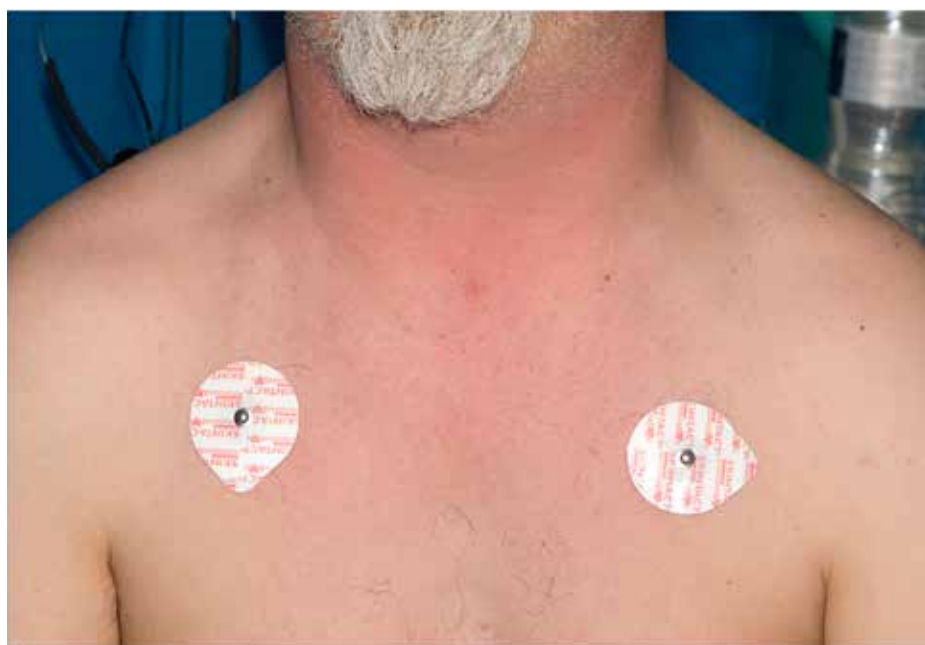


Figure 3. Frontal view showing bilateral swelling and erythema of the neck.



Figure 4. Intra-oral view of the lower right third molar extraction site.

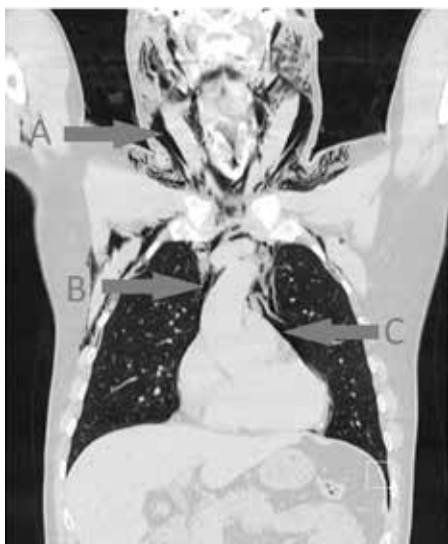


Figure 5. Coronal CT showing air in the neck (A) and mediastinum (B, C).

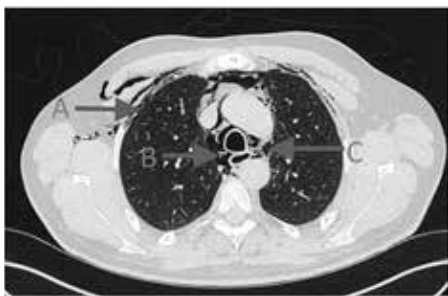


Figure 6. Axial CT showing air in the pleural space (A) and mediastinum (B, C).

in 16 of the 32 cases, with the majority of teeth being mandibular in origin. Other causes of subcutaneous emphysema described include patient activity after a surgical procedure; examples of which are a patient coughing and the use of a peak flow meter, in addition to the case reported due to motor tics in Tourette's syndrome. In these cases, air was forced under pressure from the oral cavity through the surgical site, into the subcutaneous tissues.^{6,8,9}

In this case, the patient's repeated 'raspberry blowing' tic led to an increase in pressure in the oral cavity that forced air into the subcutaneous tissues.

When pressurised air is forced into the subcutaneous tissues, it can dissect along the fascial planes between muscle compartments. From an area such as the mandible this can spread superiorly to the temporal and periorbital regions and inferiorly into the parapharyngeal, retropharyngeal, mediastinal, thoracic and retroperitoneal spaces. When entering the pleural space, air from subcutaneous emphysema can lead to pneumothorax. Three cases from a literature review spanning 1993–2008 describe pneumothorax as a complication, which can potentially lead to respiratory distress.¹ Introduction of air into the pericardial space is another serious complication that can lead to shifts in the cardiac electrical axis.¹ The majority of cases, however, are unlikely to cause long-term morbidity and most resolve with conservative treatment in 2–5 days.

Management can vary depending on the spread of the emphysema. In all cases, the patient requires investigation into the extent of the spread of the emphysema, in the form of clinical and radiographic examination. Subcutaneous crepitus is pathognomonic of surgical emphysema and can help exclude other differential diagnoses, such as anaphylaxis. A full blood count and serum C-reactive protein level can be useful to monitor resolution and infective complications.

In McKenzie's review,¹ the majority of cases were admitted for monitoring of worsening symptoms and administration of a broad-spectrum antibiotic. The rationale for which was prevention of serious infection due to non-sterile air being introduced from the oral cavity into the fascial spaces.

The previously reported case of subcutaneous emphysema related to Tourette's syndrome was managed using a small drainage tube, into the anterior oral cavity, to prevent air pressure building with patient motor tics.⁶ In this case, however, complete resolution of the surgical emphysema occurred without the need for such an intervention.

Conclusion

A number of factors have been implicated as causes of subcutaneous emphysema. Most commonly in dentistry, it is due to the inappropriate use of the air turbine drill. Dentists should be aware of the potential complications that can occur following surgical procedures, their diagnosis and management. In this case, the patient's motor tic led to subcutaneous emphysema, pneumomediastinum and bilateral pneumothorax following surgical lower third molar extraction. The patient made a full recovery following hospital admission and antibiotic treatment, without any further intervention.

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