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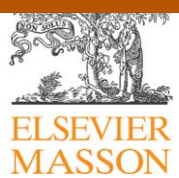

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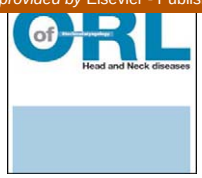

CLINICAL COMMENTARY

Head-neck and mediastinal emphysema caused by playing a wind instrument

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KEYWORDS
Pneumoparotid;
Traumatic head-and-neck emphysema

Summary
Introduction: Etiology of head-and-neck emphysema is mainly infectious (cellulitis) or traumatic (rupture of the aerodigestive tract mucosa, usually in the larynx and trachea).
Case report: We report a case of head-and-neck and mediastinal emphysema due to oral hyperpressure inducing parotid acini rupture. The aim is to highlight the importance of precise interview of patient and family so as to identify this mechanism.
Discussion: Pneumoparotid is a rare cause of swelling induced by insufflation into Stensen's duct. This reflux is caused by intra-oral hyperpressure, and can cause extensive subcutaneous emphysema induced by capsule rupture. The disorder is common in players of wind instruments and glass-blowers. Positive diagnosis is based on focused interview, looking for trigger factors, and on imaging assessment. Evolution is reported to be spontaneously favorable with simple antibiotherapy. Patient education is the best means of limiting recurrence.
Conclusion: In case of cervical emphysema, it is important to determine the mechanism of onset and explore for pneumoparotid on CT, so as to adapt treatment and prevent possible recurrence.

Introduction

Parotid gland tumefaction may be of infectious (viral or bacterial) origin, but also obstructive, inflammatory or cancerous. Pneumoparotid, or air dilatation of the parotids, is a rare cause of parotid hypertrophy. It is defined by presence of air within the gland itself and/or in Stensen's duct. Insufflation is caused by high intra-oral pressure, causing air to

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migrate into the salivary ducts and then into the parotid parenchyma. It may in some cases rupture the acini and break through the capsule, causing facial, cervical or mediastinal emphysema.

Case report

A 7-year-old girl was admitted to Emergency with extensive edema of the face and neck following a game of handball. She had severe subcutaneous facial and cervical emphysema with pain in both parotids. Initial interview disclosed no relevant medical history. The patient was apyretic, without dyspnea. Oropharyngeal examination showed no particularity; the larynx was normal on palpation. No inflammation of the integuments was observed. The two parotid spaces were enlarged and painful on palpation. There was an aspect of cervical emphysema.

AP cervical and thoracic X-ray found severe cervical emphysema (Fig. 1). Cervicothoracic CT confirmed the presence of numerous air bubbles in both parotids and in Stensen's duct (Fig. 2), with gaseous effusion extending to the neck and mediastinum (Fig. 3).

In a later interview, the parents mentioned the recent purchase of a recorder; the girl described her very energetic and unusual way of blowing on it with all holes covered, and recalled a strong resultant pain in both parotids.

Figure 1 Cervical X-ray: subcutaneous emphysema.

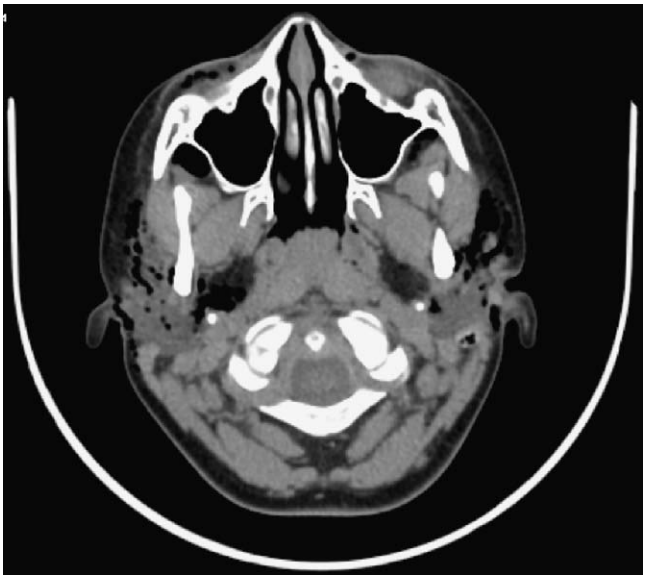

Figure 2 Facial CT: facial and parapharyngeal parotid emphysema.

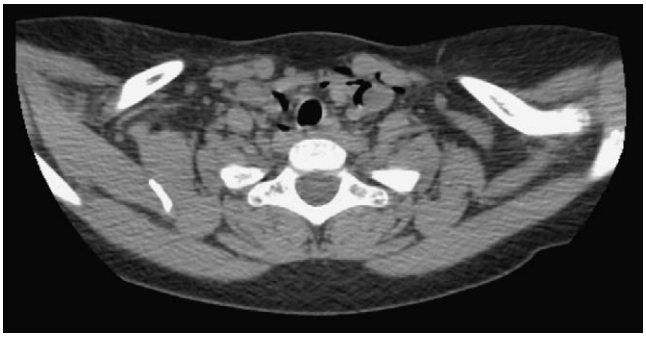

Figure 3 CT: mediastinal emphysema.

In the light of these new interview findings and the imaging data, bilateral pneumoparotid was diagnosed. The effort of blowing into the recorder had created intra-oral pressure leading to air reflux into Stensen's duct and then the parotids, with rupture of the acinar epithelium and gland capsule.

Putting a stop to this trigger factor and administering analgesics and preventive antibiotherapy (Amoxicillin®) secured favorable evolution within a few days. One month later, the girl presented with a recurrent episode of pneumoparotid, managed in the same way. Clinical monitoring found no further recurrence at 2 years' follow-up.

Discussion

Pneumoparotid is a rare pathology, little described in the literature. The swelling may be uni- or bilateral, and is perceptible on palpation. Intra-oral examination finds an excretion of bubbly saliva at the ostium of Stensen's duct. Subcutaneous crepitation is the principal sign of onset of emphysema, indicating rupture of the salivary gland capsule.

The occupations most frequently affected are wind instrument players (trumpeters, clarinetists, etc.) and glass-blowers [1].

The ostium of Stensen's duct has an oval, whistle-like aspect formed by a mucosal fold. This anatomic feature, associated with buccinator muscle contraction, makes the whole duct physiologically airtight [2]. When either of these elements is defective, the result is ostial incompetence. On Valsalva maneuver, insufflation within Stensen's duct can pneumatize the parotid and rupture the acinar epithelium, thus spreading to neighboring subcutaneous tissue and inducing facial, cervical or mediastinal emphysema with respiratory symptoms.

Differential diagnoses with respect to cervicofacial emphysema without associated fever include laryngeal trauma, closed glottis effort and sports trauma.

Pneumoparotid has also been reported following dental treatment using insufflation equipment [3], and associated with orthodontic correction braces, the rings of which can damage the salivary duct ostium.

The disorder may also occur in a psychiatric context: certain patients affected by tics puff their cheeks out more than 300 times a day [4], and children have been reported to self-inflict pneumoparotid for the sake of its collateral benefits [4]. It has also been reported in Münchhausen syndrome and in members of the French Foreign Legion, who blew into bottles to create an effect of viral parotitis with a view to shirking their military duties [5].

More rarely, pneumoparotid may be induced by violent effort in chronic coughing (chronic obstructive bronchopneumopathy [6] or pulmonary fibrosis) or following respiratory functional tests on a spirometer [7].

Any process inducing the same physiopathological mechanism may induce pneumoparotid, including barotrauma in divers, repeated Valsalva maneuver [5,8] or extubation accidents [8].

Diagnosis is thus based on meticulous history-taking in search of any risk factors, and on imaging assessment that can confirm the presence of air within the gland and detect possible complications, cervicothoracic CT being the reference examination.

Etiological management begins by forbidding trigger factors: e.g., stopping playing the implicated wind instrument, sometimes with change of occupation. Psychotherapy can be useful where behavioral disorder is involved. Preventive antibiotherapy is prescribed to avoid cervical and especially mediastinal superinfection by salivary contamination. Surgery may be considered as a last resort in recurrent or complicated pneumoparotid [5,9]: Stensen's duct transposition with reimplantation of a new ostium [10], or even total parotidectomy. Surgery remains, however, the exception, weighing the risk of facial nerve lesion against the benign nature of the disorder.

Conclusion

Pneumoparotid is a rare pathology, but should be considered in case of unexplained recurrent parotiditis. Positive diagnosis is basically clinical. Extension is to be assessed on cervicothoracic CT. Initial treatment being etiological; anamnesis is of prime importance and should look for risk factors and relevant psychological terrain. Analgesic and antibiotic therapy usually allows recovery within two weeks.

Conflict of interest statement

The authors have not declared any conflict of interest.

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