Pneumoparotitis: A case report

Angela E. McGreevy, MB, Anna M. O’Kane, MB, David McCaul, MB, Shaik I. Basha, MD

ENT Department, Craigavon Area Hospital, Portadown, Northern Ireland.

Accepted 25 May 2011
Published online 20 March 2012 in Wiley Online Library (wileyonlinelibrary.com). DOI 10.1002/hed.21873

ABSTRACT: Background. Pneumoparotid is a rare cause of parotid enlargement. Pneumatic enlargement of the gland is caused by retrograde insufflation via Stensen’s duct. Most reported cases have been managed conservatively, thus running a short course.

Methods and Results. We report on a case of a 48-year-old man with a chronic painful right facial swelling and symptoms of repeated infection. Clinical examination found a parotid swelling with surgical emphysema and a dilated Stensen’s duct. Pneumoparotitis with cystic changes and dilated intraparotid ducts was confirmed by CT. The patient proceeded to undergo excision of the gland, with subsequent resolution of symptoms.

Conclusion. Repeated retrograde movement of air and contaminated saliva leads to chronic infection and sialectasis. Management is aimed at preventing these sequelae by identifying and addressing the insult early; however, repeated pneumoparotid leads to chronic pneumoparotitis, the management of which is excision of the gland.

KEY WORDS: Pneumoparotid, pneumoparotitis, parotitis, stensen’s duct.

Pneumoparotid is a rare cause of parotid enlargement, which occurs air within the parotid gland. When it coexists with inflammation or infection, it is called pneumoparotitis. Terms such as pneumosialadenitis, pneumatocele glandiae parotitis, wind parotitis, and anaesthesia mumps have been used previously in the literature. This pathologic state is usually associated with retrograde pneumatic insufflation via Stensen’s duct into the more distal ducts and glandular acini. The differential diagnosis in the acute setting would include a gas-producing anaerobic infection of the parotid.

The condition was first recognized in 1915 when a strange epidemic of mumps occurred in the French Foreign Legion in North Africa. After an in-depth investigation, it was discovered that the soldiers were deliberately self-inducing the condition by blowing into a small bottle to avoid duty. This behavior was swiftly curtailed because the investigation for mumps at that time was serial lumbar punctures.

CASE REPORT

A 48-year-old man presented with a protracted history of intermittent, painful, bilateral facial swelling, with the right side more marked. The swelling and change in size was not related to eating. The patient had been seen by an ear nose and throat specialist 25 years earlier, and imaging at that time had shown a bilateral air sialogram. The patient was not a wind instrumentalist or glass blower. No obvious cause was identified other than self-insufflation. The patient underwent dilation of Stensen’s duct at the time and was lost to follow-up.

Over the years he had repeated infections of the gland that were becoming increasingly problematic. He had mastered the technique of milking air out of the gland. He had no other relevant medical history.

Clinical examination demonstrated a 7-cm × 6-cm fluctuant right-sided polycystic parotid swelling with crepitus (Figure 1). The patient was able to self insufflate both parotid glands by expiring against a closed mouth (thus increasing intraoral pressure). Intraoral inspection of Stensen’s duct orifices revealed that they were dilated (Figure 2), and the patient was able to manually express frothy saliva from the parotid duct. He had undergone dental treatment on the upper molar teeth adjacent to the opening of Stensen’s duct, as seen. The facial nerve function was intact, and there were no palpable cervical lymph nodes.

CT scanning showed evidence of air within both parotid glands. In the right gland there was also a fluid level with cystic changes and dilated air-filled intraparotid ducts (Figure 3). The patient was referred to an interventional radiologist with a view to identify strictures in the ducts that may have caused proximal dilation that were amenable to stenting. However, the parotid duct architecture was so distorted that no intervention could be carried out. The patient subsequently underwent a right parotidectomy. Pathologic examination of the gland confirmed chronic sialadenitis, chronic inflammatory cell infiltrate...
within the parenchyma with an increase in fibrous and adipose tissue, and dilated ducts. If there is repeated insult to the left parotid gland and duct, the patient may require a left-sided parotidectomy in the future.

**DISCUSSION**

Pneumoparotid may be categorized as occupational or self-induced; however, regardless of the cause, it is usually caused by an increase in intraoral pressure and subsequent retrograde flow of air through Stenson’s duct and into the parotid acini. Both unilateral\(^6-14\) and bilateral\(^2,6,15-24\) pneumoparotid have been reported in the literature. Bilateral cases may be asymmetrical, with 1 side being more severe than the other,\(^24\) as was the case with our patient.

Most commonly, pneumoparotid is self-induced via forced retrograde pneumatic auto-inflation,\(^2,6,7,10,11,13,15-19,21-23,25-28\) often by expiring against a closed mouth. There are a number of reports describing self-induced pneumoparotid and pneumoparotitis in children and adolescents,\(^6,7,10,11,13,15-19,22,25-27,29\) most of which have attributed the disorder to psychiatric, psychological,\(^12\) and behavioral issues including nervous tics,\(^2,6,9,15,25\) stress,\(^13\) after a traumatic event,\(^19\) adolescent adjustment reactions,\(^7,16,17\) sibling rivalry,\(^16\) and to avoid school.\(^6,15,16,18\) One child had been seen by his parents standing in front of a mirror and forcefully blowing against a closed mouth.\(^16\) It may also manifest as a result of an unconscious habit.\(^11,30\) One case described a man who had developed an unintentional habit of blowing out his cheeks 300 to 400 times a day.\(^30\) An adult female with Munchausen’s syndrome, who had presented to multiple centers with multiple self-inflicted injuries, also presented with pneumoparotid caused by self-induced auto-inflation of the gland.\(^28\)

Pneumoparotitis is a recognized occupational hazard of glass blowers\(^31\) and wind instrumentalists,\(^3,27\) divers, and watch keepers working under high pressures.\(^12,18\) It has been described after orthodontic interventions, such as the application of braces\(^19,32\) or dental instrumentation.\(^33-35\) Pneumoparotitis has also been associated with general anesthesia with positive pressure ventilation, when a patient coughs during extubation (known as anesthesia mumps).\(^4\) It has been reported in patients in association with cough suppression with chronic obstructive pulmonary disease,\(^1,36,37\) with cystic fibrosis,\(^9,34\) and in an infant with bronchiolitis,\(^10\) after spirometry testing,\(^7\) forceful nose
blowing,\textsuperscript{38} and in association with sneezing in hay fever while pinching the nostrils.\textsuperscript{39} Another person with sensorineural hearing loss had development of it after repeated Eustachian tube autoinflation to "clear his ears."\textsuperscript{30} Pneumoparotid in association with blowing bubbles with chewing gum, blowing up balloons, and blowing bubbles in water has also been reported.\textsuperscript{27} Sometimes a cause for pneumoparotid is not identified,\textsuperscript{10,40} however, it may be the case that the patient is withholding the information, either because of embarrassment, personal gain, or fear of parental ridicule.

The opening of the parotid duct (Stensen’s duct) lies adjacent to the second upper molar tooth bilaterally. The normal anatomic features of Stensen’s duct preventing the reflux of air and saliva into the parotid gland are 3-fold: (1) the diameter of the duct orifice is smaller than that of the duct itself; (2) the duct opening, which is slitlike, is covered by redundant mucosal folds, which cover the duct orifice when there is increased intraoral pressure; and (3) the duct is compressed in its lateral course along the masseter muscle and its passage through the buccinator muscle with increased oral pressure.\textsuperscript{6,7,12,16–18} As stated previously, pneumoparotid is usually caused by increased oral pressure and the retrograde flow of air and saliva through Stensen’s duct. This has been demonstrated by viewing water-soluble contrast in the mouth enter the duct with air bubbles under fluoroscopy with increased intraoral pressure.\textsuperscript{7} Another study has described the patient filling their mouth with contrast and blowing out their cheeks, and subsequent CT examination of the parotid gland showed reflux of air and contrast into Stensen’s duct.\textsuperscript{15} The pathophysiologic condition of pneumoparotid has also been demonstrated by Lasboo et al\textsuperscript{24} using a “puffed-cheek” technique. They performed a CT examination after sialography, which highlighted filling defects, air in the parotid ductal system, and sialoliths. The patient then massaged both parotid glands, and a further CT scan confirmed a reduced amount of air and absence of contrast. The patient was then instructed to puff out his cheeks to increase intraoral pressure, and a third CT scan confirmed that air had refluxed through Stensen’s and intraglandular ducts.

Normal intraoral pressure is 2 to 3 mm Hg; however, glassblowing and trumpet playing can increase intraoral pressure to 150 mm Hg.\textsuperscript{41} It is not fully understood why some people experience pneumatic insufflation with high intraoral pressures whereas others do not. Anatomic abnormalities, which are believed to contribute to pneumoparotid, include an insufficiency or hypotonia of the buccinators muscle fibers surrounding the papilla,\textsuperscript{42} hypertrophy of the masseter muscle,\textsuperscript{37} transient mucous plugging causes decreased salivary flow,\textsuperscript{6,17,20} and abnormal dilatation of the duct orifice or patulous duct.\textsuperscript{1,15,23} Pneumoparotid has been described after facial trauma,\textsuperscript{1,8} which may have damaged the normal protective mechanisms as described above.

Repeated insufflation of Stensen’s duct causes air to travel into the acini, which accounts for the crepitus. Repeated episodes commonly give rise to sialadenitis, sialectasis, and recurrent parotitis, likely because of the retrograde movement of air, contaminated saliva, oral flora, and structural trauma. Superimposed bacterial infection may occur.\textsuperscript{16,17,25,32} Repeated autoinflation with high pressure may cause the acini to rupture. Because the parotid capsule is incomplete superomedially at the posterior border of the mandible, air may travel into the parapharyngeal and retropharyngeal space. There have been cases describing pneumoparotid in association with subcutaneous emphysema of the face (including the infraorbital region) and neck,\textsuperscript{2,15,43} pneumomedianstium,\textsuperscript{15} and pneumothorax.\textsuperscript{7,44}

After consideration of the more common causes of parotid swelling, a diagnosis of pneumoparotid should be borne in mind when assessing a patient with a unilateral or bilateral parotid swelling. An accurate history, which may contain some of the risk factors as outlined above, may provide a clue. The parotid swelling is often recurrent intermittent,\textsuperscript{2,7,13–19,21,38} and often resolves spontaneously.\textsuperscript{6,13,15,18} Clinically the swelling may be unilateral\textsuperscript{6–14} or bilateral.\textsuperscript{2,6,15–24} Important features on clinical examination would include a parotid swelling that may be tender,\textsuperscript{6,8,14,18,21,25} or nontender,\textsuperscript{6} crepitus overlying the gland,\textsuperscript{2,8,11,13,16,20,21,24} a dilated entrance to Stensen’s duct,\textsuperscript{7,8,12,13} and the ability to milk frothy saliva\textsuperscript{6,8,11,13,21,29,30} from Stensen’s duct orifice, with or without exerting pressure over the parotid swelling. Purulent discharge may be present if there is active superimposed infection.\textsuperscript{11,16} A dry mouth has also been reported as a symptom.\textsuperscript{14,15} In some cases these clinical signs are minimal,\textsuperscript{18} usually between episodes of pneumoparotitis.

Hematologic investigations would include inflammatory markers,\textsuperscript{6,10,13,16} and viral serologic study\textsuperscript{5,15,14} for HIV,\textsuperscript{15,16} paramyxovirus,\textsuperscript{15} herpes virus,\textsuperscript{15} antinuclear antibodies,\textsuperscript{6,10,15} and rheumatoid factor,\textsuperscript{6,10,15} as well as studies to rule out Sjögren’s syndrome, sarcoidosis, and immunodeficiency. Some studies have obtained a biopsy specimen of the gland,\textsuperscript{12,14–16} and indeed in one case it was virtually diagnostic because the parotid swelling suddenly decreased when it was punctured.\textsuperscript{14} Needle aspiration has also been performed as a diagnostic aid, with a positive aspiration of air.\textsuperscript{6} However, these are invasive procedures and not without potential complications.

Radiologic investigations that have been described in the literature include ultrasonography,\textsuperscript{13,22,25,26} sialography,\textsuperscript{6,8,10,20,22–25} radionuclide sialography,\textsuperscript{16} salivary gland isotope scanning,\textsuperscript{15} and computed tomography.\textsuperscript{6,8,10,11,13–15,18,19,21,24,25,37,40,45,46} MRI has also been used in 1 case.\textsuperscript{47} Chest radiography should be performed if there is expected breach of the parotid capsule to assess for pneumothorax or pneumomedianstium.\textsuperscript{6}

Ultrasonography may confirm subcutaneous emphysema with multiple hyperechoic areas corresponding to air within the ducts and glandular parenchyma.\textsuperscript{13,26} Sialography appears to be less sensitive; however, it is useful for excluding calculi and may show air within the ducts or acini,\textsuperscript{10,20} dilations and structuring of Stensen’s duct,\textsuperscript{2,24} beading and narrowing of small ductules,\textsuperscript{6} and other filling defects within the gland ducts and acini.\textsuperscript{24} However, it may still be difficult to distinguish radiolucent calculi from air.\textsuperscript{24} Salivary gland isotope scanning and radionuclide scanning may demonstrate hypofunction of the gland.\textsuperscript{15,16} Although ultrasonography and sialography have been used as imaging modalities in diagnosing and assessing pneumoparotid, they often appear normal\textsuperscript{12,42} or require further clarification of anatomy and disease.
CT is now established as the current gold standard investigation in this condition, primarily because it outlines the anatomy well and is noninvasive. The classical findings on CT are air filled dilatation of Stensen’s duct, acini pneumatic dilations, intraglandular fluid collections, extraductal air, and multiple structures and dilations along Stensen’s duct, as can be seen in Figure 1. It may also demonstrate air extending to the face or the parapharyngeal and retropharyngeal space, if present, and down to the chest. It has been advocated as the best way of detecting air within the gland, through the duct, extending to its buccal orifice, and is useful in distinguishing radiolucent calculi from air. Because pneumoparotid is most commonly intermittent, it is important to obtain images of the gland during an acute exacerbation, because results of investigations in the interim period may appear normal, yielding no diagnosis. In such cases it may be useful to consider the ‘‘puffed-cheek’’ technique as described above.

Numerous management strategies for pneumoparotitis have been cited in the literature. They range from conservative to medical and surgical. Conservative management is centered on avoidance of the causative factor, whether it is self-insufflation or abstaining from wind instrument playing to surgical. In children who have psychological or behavioral problems, often a form of counseling or psychotherapy is required. If this is achieved without multiple repeated insufflations with associated inflammation and infection, then pneumoparotid is often self-limiting when the assaulting action has been removed. For those for whom pneumoparotid is an occupational hazard, such as wind instrument players, the patient should be coached in techniques to decrease the likelihood of air insufflation or advised to use ‘‘cheek compressors.”

Medical treatment of an acute bacterial infection would include analgesia and oral or intravenous antibiotics depending on the severity of symptoms and systemic upset. Massage of the gland, hydration, mouth washes, sialogogues, and warm compresses may also help.

Several surgical interventions have been described. These include salivary duct ligation and clamping of the duct orifices. One surgical option is the Wilkie-Brody procedure, which entails resiting the parotid duct opening to the tonsillar fossa to lengthen the path of the duct deep to the buccal mucosa and, hence, create an intramural valve mechanism, in which the duct is compressed with increasing intraoral pressure. Ductal ligation has been used successfully in 1 case of a 13-year-old boy when conservative measures did not solve the problem. The child had already undergone a superficial parotidectomy on 1 side as treatment for pneumoparotitis. He had bilateral patulous duct orifices. In this case, ductal ligation was successful in prevent further episodes of pneumoparotid on the remaining side. Parotid duct ligation has been condemned as a treatment option for recurrent or chronic parotid infection because ductal ligation in a contaminated field would lead to parotid abscess formation; however, in cases of pneumoparotid with a short history and continued autoinsufflation, it may be effective as described.

Infrequently a patient with pneumoparotitis will require a parotidectomy either because of noncompliance of treatment (ie, stopping offending mechanism), failure of treatment, or chronic infection. The end point of repeated pneumoinsufflations is chronic parotitis; thus the treatment of this would be the same as chronic parotitis of any cause, which is parotidectomy. Our patient had experienced a chronic 25-year history of recurrent pneumoparotitis with recurrent infections, which was becoming increasingly troublesome. There was well-established chronic inflammation with cystic degeneration of the gland, and it could not be guaranteed that the symptoms would respond to other gland-preserving surgical intervention; thus it was more appropriate in this case to proceed to parotidectomy for symptomatic relief and psycho-social reasons.

Pathologic examination of our patient’s parotid gland demonstrated chronic sialadenitis, chronic inflammatory cell infiltrate within the parenchyma with an increase in fibrous and adipose tissue, and dilated ducts. This is in keeping with previous studies that have shown acute and chronic inflammation and ductal destruction.

CONCLUSION

This case demonstrates that persistent pneumoparotid results in recurrent infection and progressively chronic inflammation with cystic degeneration of the parotid gland. On a review of the literature, this appears to be a report of the oldest and most protracted history of pneumoparotitis. The patient denied any of the well-known identified causative factors. This case reinforces that if pneumoparotid does not resolve either by avoidance of autoinsufflation or with interventional procedures, the patient is likely to experience progressive symptoms, predisposing the patient to sialotasia and recurrent parotitis. Although it could not be established what the cause of pneumoparotid was in this man, it was clear that the only intervention for management of his progressive symptoms of pneumoparotitis was to proceed to parotidectomy.

Acknowledgements

Irish Conlon, Consultant Radiologist, Daisy Hill Hospital, Newry, and William Loan, Consultant Interventional Radiologist, Belfast City Hospital, Belfast, Northern Ireland.

REFERENCES
