CASE REPORT

Conservative management of bilateral pneumoparotitis with sialendoscopy and steroid irrigation

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SUMMARY

Pneumoparotitis is a rare condition related to retrograde airflow into the ductal system of the gland and secondary infections. Although counselling is enough in the majority of cases, persistent problems require surgery. Sialendoscopy and ductal irrigation with steroids have never been described as a treatment option. We present the case of a 61-year-old man with recurrent bilateral parotid swellings who had three episodes of sialadenitis on the right side within 2 years. Massage of the glands revealed air bubbles coming out from both papillae. A previous CT scan confirmed the presence of air in both parotid glands. The patient underwent sialendoscopy and irrigation of the ductal system with prednisolone. Ductal irrigation with steroids in three additional sessions led to a significant improvement of symptoms 6 months later. Sialendoscopy and irrigation with steroids could be another treatment modality in cases of recurrent pneumoparotitis avoiding major surgery.

BACKGROUND

The term pneumoparotid describes the presence of air in the parotid gland and/or its ductal system.¹ This condition can be complicated with ascending infections, in which case the term pneumoparotitis is used. The pathophysiology of this condition includes an incompetent valve system in the papilla of Stensen's duct, resulting in the entrance of air in the parotid gland in case of increased intraoral pressure.²

In the majority of cases, pneumoparotid is self-induced due to psychiatric disorders³ or behavioural problems.⁴ Children often inflate parotids in order to avoid school⁵ and adolescents develop pneumoparotid as an adjustment reaction.⁶ However, there are cases where the onset of pneumoparotid is spontaneous,⁷ and when this is complicated with an ascending infection, it can cause problems in differential diagnosis, for example, anaerobic infection of the parotid gland.

Sialendoscopy has become a routine diagnostic and minimally invasive therapeutic procedure of the parotid gland within the past two decades.^{8–10} Its primary goal is the evaluation and management of the salivary ductal system disorders including chronic inflammatory conditions.^{8–9} The current management of pneumoparotitis cases includes counselling and surgery in persistent cases. As recurrent parotid infections due to autoinsufflation can lead to chronic parotitis and permanent structural changes, we suggest that sialendoscopy could be an additional minimal invasive option before the decision for a surgical intervention.

CASE PRESENTATION

A 61-year-old man presented with a 2-year history of frequent recurrent episodes of right parotid swelling (2–3 every month) and only a few mild swellings on the left side, unrelated to meals, having a quick resolution in the vast majority of cases. During this period of time, the patient developed sialadenitis three times on the right side requiring antibiotics. He did not report awareness of air entering the salivary duct. The patient was seen by another otorhinolaryngologist (ORL) specialist 2 weeks earlier who ordered a CT scan. Imaging showed the presence of air in both ductal systems with more prominent findings in the right parotid gland ([figure 1A, B](#)). However, no cystic changes or gross distortion of the parotid parenchyma were noted. Clinical examination was unremarkable with a mild parotid swelling on the right side, no crepitation on palpation of both glands and no dilated orifice of Stensen's ducts. During massage of the glands, saliva with air bubbles came out of both Stensen's duct papillae. The patient denied self-insufflation and his occupation was not related to increased oral pressure.

INVESTIGATIONS

All blood investigations of the patient including inflammatory markers, viral serology (HIV, parvovirus, herpes virus) and studies for autoimmune disorders (eg, Sjogren's syndrome, sarcoidosis) were negative.

TREATMENT

To complete the assessment and remove any epithelial and inflammatory debris from the ductal system, a sialendoscopy under local anaesthesia was performed in both glands. The lumen of the right duct had some epithelial inflammatory debris causing partial obstruction and numerous air bubbles ([figure 2A, B](#)). The left duct had only a few air bubbles without evidence of inflammation. No stenosis or remarkable dilated areas were identified on both sides. As sialendoscopy requires continuous irrigation with normal saline, the air was removed from the ductal system in a few minutes along with the epithelial debris and then a final irrigation followed with 250 mg of prednisolone in 50 mL of Ringer's solution.

The same irrigation with prednisolone was repeated once a week for 3 weeks using a 22-gauge (0.9 mm) blunt-tipped needle on a syringe for the intraductal administration of steroids without performing sialendoscopy.

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Figure 1 Axial sections of a CT scan shows air in both parotid glands. (A) In this section, the presence of air is clear in the duct orifice and the proximal part of Stensen's duct on both sides. The right parotid gland presents numerous air pockets within its parenchyma. (B) Presence of air in the left parotid parenchyma.

OUTCOME AND FOLLOW-UP

The patient reported significant resolution of his symptoms with only two mild episodes of swellings on the right side at a follow-up period of 6 months. No persistent swelling for more than 2 days and/or sialadenitis was reported. The patient has an ordinary follow-up in sialendoscopy clinic every 6 months.

DISCUSSION

The protective mechanism of Stensen's duct valve includes a mucosal flap covering the papilla, a narrow slit-shaped orifice of the duct, and a compressive mechanism by the buccinator muscle during contraction.²

All the conditions that can affect the above described anatomy could be possible predisposing factors of pneumoparotid, like a patulous duct, hypotonia of the buccinator muscle around the papilla, hypertrophy of the masseter muscle and transient obstruction of Stensen's duct by mucus plugs causing decreased salivary flow.^{2 10}

The basic proposed mechanism by which air enters Stensen's duct is increased intraoral pressure. This explains why pneumoparotid occurs as an occupational disorder in glass and balloon blowers, and wind instrument players.¹¹ Pneumoparotitis is also reported during general anaesthesia due to positive pressure¹² and dental and orthodontic treatments.¹³ Numerous reports describe self-induced pneumatic autoinflation of parotids due to psychiatric or psychological problems. Classic examples of behavioural issues are children trying to avoid school and adolescents trying to pay attention.^{5 6} However, there are some cases where a causative factor is not identified like in the present case. In these cases, detailed laboratory tests should especially

be performed to exclude any infectious or autoimmune disease.¹⁴

Regardless, aetiology inflammation and fibrosis of the ducts seem to induce duct dilation and sialectasis, which lead to further ductal incompetence.^{2 7}

There is evidence that irrigation with saline during sialendoscopy and removal of epithelial debris prevent obstructive episodes and sialadenitis in chronic inflammatory conditions such as juvenile recurrent parotitis, Sjogren syndrome, etc.^{15 16} Additional irrigation with steroids seems to prolong the symptom-free intervals in such disorders increasing significantly the patients' quality of life.¹⁷ A similar treatment in pneumoparotitis cases has not yet been described. However, sialendoscopy with steroids irrigation may decrease the possibility of obstructive sialadenitis in a similar way with the aforementioned disorders, especially in cases where a self-induced mechanism is excluded and an infectious or autoimmune cause is suspected. This treatment option may be part of a long-lasting gland preservation strategy.

The combination of sialendoscopy and steroid irrigations seems to extend the improvement period of time, as steroids work not only as a local anti-inflammatory agent in the ductal system but also increase the salivary flow of parotids for a long period of time. In a study by Izumi *et al*,¹⁸ the effectiveness of steroid irrigation of the parotid gland in Sjogren syndrome patients was confirmed in comparison with saline irrigation. The authors presented an improved salivary flow sustained for a mean of 8.4 months. Moreover, the same study showed that steroid irrigation could be repeated being similarly effective, as no refractoriness of the gland to the medication has been

Figure 2 Endoscopic view of the right Stensen's duct (A). An air bubble is coming out during endoscopy and (B). Debris obstructing circumferentially the lumen of the duct.

observed. The precise mechanism is not clear; however, it seems that steroids act inhibiting B and T lymphocytes infiltration of the ductal system.¹⁸

One could argue that systemic steroid absorption from the parotid ducts could increase the salivary flow and decrease the inflammation of the ductal system in a similar way. Although this is a possible mechanism, it is reported to be short-lived.^{19 20}

Regarding the safety of intraductal administration of medications, steroid irrigation has not been associated with any toxicity or side effects in the literature.^{15–18}

In addition, a duct stent may be useful, especially when pneumoparotitis is implicated with ductal stenosis. The presence of a stent could reform the parotid duct preventing stenosis and dilation stabilising the duct with fibrosis. Although not reported in pneumoparotitis cases, its extended use in stenosis and sialolithiasis cases provides enough data for its value to maintain a patent ductal system.^{21 22 16}

Until now in persistent cases of pneumoparotitis, surgical management was required. This includes a variety of procedures such as salivary duct ligation, an operation which leads relatively fast to a non-functional gland and resolution of symptoms.⁴ Another option is the relocation of the parotid duct opening to the tonsillar fossa, the so called Wilkie-Brody procedure.²³ Finally, when severe structural changes exist with recurrent superimposed infections and/or other treatment modalities failed, a parotidectomy is the end point of treatment.^{24 25} Although effective, all the above procedures have a significantly higher morbidity when compared with sialendoscopy. Thus, endoscopic management of persistent pneumoparotitis cases could be the next step of treatment when counselling fails, in order to decrease the number of patients who require major surgery, for example, parotidectomy. However, it should be noted that parotid glands with severe and extended structural changes like cystic degeneration and a long history of recurrent infections do not present good response to conservative options like sialendoscopy and may require, from the beginning, a non-gland-preserving management.

Learning points

- Pneumoparotitis is the presence of air within the parotid gland or duct. When complicated with retrograde infections is called pneumoparotitis.
- Sialendoscopy and steroids irrigation could be an additional treatment option of pneumoparotitis management between counselling and major surgery.
- Some cases of persistent pneumoparotitis could avoid major surgery having conservative management with sialendoscopy and steroid irrigation.

Competing interests None.

Patient consent Obtained.

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