# Pneumoparotid Related to Obstructive Sleep Apnea Syndrome Treated by Oral Appliance with Anterior Opening to Reduce Intraoral Pressure

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**Abstract**

An oral appliance with anterior opening to reduce intraoral pressure was effective for a 57-year-old man with obstructive sleep apnea syndrome accompanying pneumoparotid, which refers to the retrograde insufflation of air inside the parotid gland or Stensen's duct.

## Introduction

Swelling or pain in the parotid region is a relatively common complaint in clinical practice of internal medicine, otolaryngology, and oral and maxillofacial surgery. There are many causes of parotid gland swelling, such as inflammation, viral infection, sialoliths, benign or malignant tumors, autoimmune diseases, and endocrine disorders. The pneumoparotid is one of the causes of parotid gland swelling, which refers to the retrograde insufflation of air inside the parotid gland or Stensen's duct. The pneumoparotid was first described by Hyrtl[1]in 1865 in wind instrument players. Moreover, until the first half of the 20th century, the pneumoparotid has been recognized as a typical occupational disease in glass blowers.[2-4] However, the other etiologies of the pneumoparotid are variable and include self-induction, [5-11] abnormal habit,[12-14] coughing attack,[15-17]nervous tic,[5, 18, 19] iatrogenic causes such as dental treatment,[19-21] continuous positive pressure[23, 24] and spirometry.[25]

Obstructive sleep apnea is a disorder characterized by repetitive, complete, or partial closure of the upper airway during sleep.[26] Sleep apnea is defined as 30 or more apneic episodes (cessation of airflow at the mouth and nose for more than 10 s).[26-29] Typical subjective complaints of sleep apnea are excessive daytime sleepiness, loud and irregular snoring, disturbed nighttime sleep, mental deterioration and depression.[30, 31] A recent systematic review highlighted that the overall prevalence of obstructive sleep apnea ranged from 9% to 38% in the general adult population.[32] Patients with this condition are at an increased risk of automobile accidents.[33, 34] It is associated with an increased prevalence of cardiovascular complications, such as arterial hypertension,[35, 36] coronary artery disease,[37] and nocturnal angina.[38]

Continuous positive airway pressure is an effective treatment option. However, problems associated with its use leading to non-compliance include nasal congestion, discomfort secondary to pressure sensation and air leak, and mask intolerance.[30, 31] In 1984, Meier-Ewert et al.[39] first described an oral appliance for obstructive sleep apnea syndrome. Oral appliances are an important treatment choice and may be the preferred initial treatment for mild to moderate obstructive sleep apnea syndrome or snoring.

In this study, we describe a patient with pneumoparotid disease related to obstructive sleep apnea syndrome and oral appliance therapy. Plausible relationship between pneumoparotid and sleep apnea syndrome is discussed.

## Case Presentation

A 57-year-old man was referred to the Department of Oral and Maxillofacial, Kyoto Medical Center with the chief complaint of recurrent swelling of the right parotid region. Four years before the first visit, he developed bilateral parotid gland swelling and was prescribed antibiotics (cefditoren pivoxil) by his otolaryngologist, and the symptoms disappeared. Three years later, the patient experienced rapid swelling and pain in the right parotid region during sleep when he boarded an airplane and landed. He was prescribed antibiotics in the landed country. The symptoms disappeared gradually and spontaneously, but 3 weeks before the first visit to our department, severe symptoms recurred in the same region, and he visited an otolaryngologist. He took antibiotics (cefditoren pivoxil) for 2 weeks without improvement; he was advised to visit a dentist, following which the dentist introduced him to visit our department.

The right parotid gland of the patient was slightly swollen and tender. He had no fever, and no abnormalities, such as redness, were observed on the skin of the cheek. Blood tests showed no evidence of inflammation. Crepitus was generated by light compression of the right parotid gland, and frothy saliva with fine air bubbles was discharged from the papilla of the right parotid gland (Fig 1). The crepitus and swelling in the parotid region were prominent during waking up. Computed tomography showed air predominantly on the right side of both parotid glands and ducts, and no sialoliths (Fig 2). He was on oral medication because of hypertension. The history of mumps remains unclear. For 20 years from the age of 7 years, he played a traditional Japanese wind instrument, hichiriki (Fig 3). He did not snore. He has gained 4 kg in the last 5 years. His body mass index was 24.1. Since the patient complained of daytime sleepiness, sleep apnea syndrome was suspected. A sleep study was performed on three nights at his home using a pulse oximeter (Pulsox-300, Konica Minolta, Tokyo, Japan). The examination revealed a mean apnea-hypopnea index of 10.3 and a mean minimal oxygen saturation of 81.0%. The patient was diagnosed with mild sleep apnea syndrome. The minimal oxygen saturation was considerably reduced.

It was postulated that desaturation events could be related to a rapid increase in intraoral pressure. Therefore, an oral appliance to prevent obstruction of the upper airway and reduce intraoral pressure (Fig 4) was fabricated as described previously.[29-31] The mandibular position was 6 mm, protruding from the intercuspal position. An anterior opening was made between the upper and lower incisors (Fig 4). After insertion of the oral appliance, the sleep variables improved considerably (mean apnea-hypopnea index, 2.9; mean minimal oxygen saturation, 90.8%). One month later, the symptoms of pneumoparotid disappeared completely. The patient was followed up with the appliance. After a few years, the patient discontinued the appliance as the symptoms disappeared. After 4 years, the patient had bilateral parotid gland swelling, and antibiotics were prescribed for 1 week by an otolaryngologist. After 9 years, the patient had tenderness and swelling of the left parotid gland. He revisited our department for re-examination. The left parotid gland was tender. No abnormalities were observed on the skin of the parotid region. Blood tests showed no evidence of inflammation. Crepitus was detected by compression of the left parotid gland, and frothy saliva with fine air bubbles was discharged from the papilla of the left parotid gland. Computed tomography showed the presence of air in both ductal systems, with more prominent findings in the left parotid gland (Fig 2B). A sleep study was conducted again. The test revealed a mean apnea-hypopnea index of 10.1 and mean minimal oxygen saturation of 79.8%. The results were remarkably similar to those 9 years ago. Oral appliance was inserted again, and sleep variables improved (mean apnea-hypopnea index: 3.1, mean minimal oxygen saturation: 90.3%). The symptoms related to sleep apnea syndrome and pneumoparotid disappeared completely. The patient was followed up for 10 years from the first visit. Relapse of the entities has not been observed until now.

## Discussion

This study is the first to report a patient with obstructive sleep apnea syndrome accompanying pneumoparotid, who was treated effectively with an oral appliance and was followed up for 10 years.

Pneumoparotid describes the presence of air within the duct system and/or parenchyma of the parotid gland secondary to its reflux through Stensen's duct. Parotitis associated with pneumoparotide is referred to as

pneumoparotitis. The pneumoparotid has been observed in glass blowers until the first half of the 20th century.[2-4] In those days, 6%–10% of glass blowers had symptoms of suspected pneumoparotid.[4] More pressure is required to produce larger glassware than delicate works and is more likely to cause pneumoparotid.[3, 4] Fortunately, subsequent innovations and mechanization have dramatically reduced the pneumoparotid as an occupational disease in glass workers.

Previous studies indicated an association between playing wind instruments, such as the trumpet,[1, 40, 41]horn,[1, 8, 42] tuba,[43]clarinet,[8] flute,[44] or recorder.[45] The Stensen's duct valve prevents reflux into the parotid gland by the smaller diameter of the orifice, which is covered by redundant mucosal layers. The duct is laterally compressed by the masseter muscle and the buccinator muscle contraction with an increase in oral pressure. If the intraoral pressure exceeds the protective mechanism, pneumoparotid can occur. Hyrtl[1]stated that when pressure in the oral cavity increases when playing a wind instrument, air can enter retrogradely from the orifice of Stensen's duct. He added that it is easier for beginners to pull in the air when they blow with their cheeks full, and less likely to occur when they learn the appropriate embouchure technique.

Gazia et al.[46] reviewed 49 reports and analyzed 54 patients with pneumoparotid or pneumoparotitis. The most frequent etiology is self-induction by blowing the cheeks, which mainly involves children for conflicts with parents, excuses for not going to school, and nervous tics.[46] Treatment with antibiotics and steroidal anti-inflammatory drugs is the most common treatment. Behavioral therapy is applied to remove bad habits such as blowing cheeks; in some cases, supportive psychotherapy is necessary.[7, 8, 10, 11, 41]

The relationship between pneumoparotid and sleep apnea syndrome remains to be elucidated. This patient played a traditional Japanese wind instrument, hichiriki (Fig 4), for 20 years from the age of 7 years. The hichiriki is made of bamboo and is small compared to other wind instruments (Fig 3), but it requires considerably high pressure to play and emits a very large sound. Kreuter et al.[47]reported intraoral pressure while playing various wind instruments; however, hichiriki was not included. Although the patient had seldom played hichiriki for 30 years after the age of 27, it might be a predisposing factor for enlargement of the orifice of the Stensen's duct. It was suspected that air reflux occurred during sleep, as the crepitus and swelling in the parotid region were prominent when waking up. The patient noticed the first swelling of the right parotid region when he boarded an airplane and slept, and the airplane landed. The sleep test indicated that the patient had a mild obstructive sleep apnea syndrome. Playing hichiriki could be a predisposing factor, and the first occurrence of pneumoparotid was triggered by an increase in barometric pressure on the board. Considering the fact that the patient boarded only once and pneumoparotid recurred, an increase in intraoral pressure could be a plausible cause of pneumoparotid. Cabello et al.[22] reported a case of pneumoparotid, who blew severely overnight. The patient in this study did not snore. He might have been blown during sleep. The oral appliance with anterior opening might have helped prevent the oral cavity from increasing intraoral pressure during sleep. Oxygen saturation was considerably reduced, and during sleep apnea, the oral pressure can be increased, resulting in pneumoparotid.

In 1995, the American Sleep Disorders Association issued guidelines stating that oral appliances are indicated for snoring, mild obstructive sleep apnea, and moderate to severe sleep apnea if continuous positive airway pressure is not accepted or if surgery is not appropriate.[48] Various oral appliances have been increasingly used in the treatment of sleep apnea syndrome. Many randomized trials have confirmed the effects of oral appliances.[49-55] Recent guidelines have extended indications to moderate and severe sleep apnea patients when a patient refuses continuous positive airway pressure therapy after being informed about the risks.[56] All appliances for the treatment of sleep apnea syndrome are constructed with the goal of advancing the position of the mandible and tongue in order to enlarge the airway or reduce its collapsibility. This may lead to an improvement in the upper airway dimensions and possible effects on upper airway muscle tone.[57] It is indispensable to suppress events that increase intraoral pressure for patients in this report. An oral appliance to reduce intraoral pressure with anterior opening was fabricated and inserted during sleep (Fig 4). Some researchers have reported patients with pneumoparotid accompanied by obstructive sleep apnea syndrome.[22-24] Long-term use of oronasal continuous positive airway pressure[23, 24] or mandibular advance-

ment device[22] can be a potential cause of pneumoparotid. In a recent review[46], 24.1% of patients with pneumoparotid disease revealed an unknown etiology. The average age of idiopathic cases was 28.6 years. In some patients, the pneumoparotid gland can be related to sleep apnea syndrome. Further studies confirmed by polysomnography in larger cases may be necessary to clarify this hypothesis.

**Conflict of Interest (COI)**

The author declares no conflict of interest (COI).

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**Informed consent**

The patient described in this manuscript provided his informed consent for publication.

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**Figure Legends**

**Figure 1** Air bubbles from the orifice of the Stensen's duct

**Figure 2** Axial sections of a CT scan show air in both parotid glands and ducts at first visit (A) and 9 years later (B) The arrows indicate air in the parotid glands or ducts.

**Figure 3** Hichiriki (A) is a traditional Japanese wind instrument with 1400 years history. The patient playing the hichiriki (B)

**Figure 4** Oral appliance to reduce intraoral pressure in place