

# A RARE CASE OF A PNEUMOPAROTID

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A Study design/planning  
B Data collection/entry  
C Data analysis/statistics  
D Data interpretation  
E Preparation of manuscript  
F Literature analysis/search  
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## Abstract

**Introduction:** There are many reasons for an increase in volume of the salivary glands, including inflammatory changes due to infection, auto-immune and allergic reactions, endocrine disorder, tumor, or injury.

**Aim:** Presentation of a rare case of a pneumoparotid.

**Case report:** A 54-year-old patient was admitted to the department for diagnosis of a nodular swelling in the area of the left parotid gland. The swelling had appeared about 3 weeks earlier, initially with pain, and had gradually increased in size. He denied head or neck injury, problems with chewing or swallowing food, or a dry mouth. Laryngological examination revealed swelling of the cheek in the left preauricular area, palpably soft, oval-shaped (about 20 x 15 mm), slightly painful when pressed, non-fluctuant, and with no associated skin changes. Fine-needle aspiration biopsy and contrast CT scan of the neck revealed a pneumoparotid on the left side. After decompression of the pneumoparotid, conservative treatment (a pressure dressing) was applied in an outpatient mode. Due to lack of therapeutic effect, the patient was qualified for surgical excision of the left pneumoparotid.

**Conclusions:** A pneumoparotid of the salivary glands is a rare pathology affecting the parotid glands and is associated with an increase in intraoral pressure. Treatment is primarily avoidance of the triggering factor and pressure dressings, but sometimes surgery is necessary. The pathology in most cases does not cause permanent damage to the salivary glands and the effects of treatment are good.

**Key words:** parotid gland • pneumoparotid • fine-needle aspiration biopsy • computed tomography

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## UN CASO RARO DE NEUMOPAROTIDITIS

### Resumen

**Introducción:** Hay muchas causas de agrandamiento de las glándulas salivales. En la mayoría de los casos el aumento de su volumen es causado por enfermedades de etiología bacteriana, autoinmune, alérgica, endocrina, así como por traumatismos o tumores.

**Objetivo:** El objetivo de este estudio es presentar el caso de un paciente con una poco frecuente neumoparotiditis izquierda.

**Descripción del caso:** Un paciente de 54 años ingresó en la clínica de otorrinolaringología para diagnosticar el agrandamiento del área de la glándula parótida izquierda - la tumefacción apareció unas tres semanas antes, desde el principio causó dolor, que iba aumentando gradualmente. El paciente no informó de ninguna lesión de la cabeza ni del cuello, ni tampoco de los problemas de deglución, trastornos de la masticación o sequedad en la boca. El examen laringológico reveló una hinchazón de la mejilla izquierda en la parte anterior al oído. La hinchazón era blanda, de forma ovalada y de unos 20 mm x 15 mm de diámetro, ligeramente dolorosa a la hora del examen físico. La piel de esta zona permaneció sin cambiar. La biopsia por aspiración con aguja fina y la tomografía computarizada [TC] del cuello permitieron diagnosticar la neumoparotiditis derecha. Después de la descompresión del quiste, se aplicó un tratamiento conservador (vendaje compresivo) en régimen de consulta externa. Debido a la falta de efecto terapéutico, el paciente fue calificado para la cirugía de extirpación de neumoparotiditis.

**Conclusiones:** La neumoparotiditis es una enfermedad poco frecuente que se presenta en la mayoría de los casos en las glándulas parótidas y se asocia con el aumento de la presión intraoral. El método de tratamiento consiste principalmente en evitar el factor desencadenante, el uso de apósitos de compresión y, a veces, el tratamiento quirúrgico. La patología en la mayoría de los casos no causa daño permanente a las glándulas salivales y los resultados del tratamiento son buenos.

**Palabras clave:** glándula parótida • neumoparotiditis • biopsia por aspiración con aguja fina • tomografía computarizada

## РЕДКИЙ СЛУЧАЙ ПНЕВМОФИБРОЗА

### Аннотация

**Введение:** Существует множество причин увеличения слюнных желез. Чаще всего воспалительные изменения вызваны инфекцией, аутоиммунными и аллергическими реакциями, эндокринными нарушениями, опухолью или травмой.

**Цель:** Целью данной работы является описание редкого случая пневмофиброза с левой стороны.

**Описание случая:** Пациент в возрасте 54 лет поступил в отоларингологическое отделение с целью диагностики узелкового отека в области левой околоушной железы. Отек появился примерно 3 недели ранее, с самого начала он сопровождался болью и постепенно увеличивался в размерах. Пациент отрицал травму головы или шеи, проблемы с пережевыванием и глотанием пищи, сухость во рту. Ларингологическое исследование выявило отек щеки в левой преаурикулярной области, ошутимо мягкий, овальной формы (диаметр около 20 мм x 15 мм), слегка болезненный при пальпации, без изменений кожи. Тонкоигольная аспирационная биопсия и контрастная компьютерная томография шеи выявили пневмофиброз с левой стороны. После декомпрессии кисты в амбулаторном режиме было применено консервативное лечение (перевязка под давлением). Из-за отсутствия терапевтического эффекта пациент был направлен на хирургическое удаление пневмофиброза с левой стороны.

**Выводы:** Пневмофиброз слюнных желез является редкой патологией, поражающей околоушные железы, и связан с повышением внутриушного давления. Лечение, прежде всего, основано на избегании пускового фактора и использовании утягивающих повязок, но иногда требуется хирургическое вмешательство. Патология в большинстве случаев не вызывает необратимого повреждения слюнных желез, а результаты лечения эффективны.

**Ключевые слова:** околоушная железа • пневмофиброз • тонкоигольная аспирационная биопсия • компьютерная томография

## RZADKI PRZYPADKOWY TORBIELI POWIETRZNEJ ŚLINIANKI PRZYUSZNEJ

### Streszczenie

**Wstęp:** Istnieje wiele przyczyn powiększenia gruczołów ślinowych. Najczęściej wzrost ich objętości jest spowodowany chorobami o etiologii bakteryjnej, autoimmunologicznej, alergicznej, endokrynologicznej, a także urazami lub pojawieniem się guzów.

**Cel:** Celem pracy jest przedstawienie przypadku pacjenta z rzadko występującą torbielą powietrzną ślinianki przyusznej lewej.

**Opis przypadku:** Pacjent w wieku 54 lat przyjęty do kliniki otolaryngologii w celu zdiagnozowania powiększenia okolicy lewej ślinianki przyusznej – obrzęk pojawił się około trzy tygodnie wcześniej, od początku powodował ból, który stopniowo narastał. Pacjent nie zgłaszał urazów w obrębie głowy i szyi, problemów z połykaniem pokarmów, zaburzeń żucia czy suchości w jamie ustnej. Badanie laryngologiczne ujawniło obrzęk lewego policzka w części przedusznej. Obrzęk był miękki, owalny o średnicy około 20 mm x 15 mm, lekko bolesny w trakcie badania palpacyjnego. Skóra w tym obszarze była niezmienną. Biopsja aspiracyjna cienkoigłowa i tomografia komputerowa szyi pozwoliła zdiagnozować torbiel powietrzną ślinianki przyusznej prawej. Po odbarczeniu torbieli zastosowano leczenie zachowawcze (opatrunek uciskowy) w trybie ambulatoryjnym. Z powodu braku efektu terapeutycznego chory został zakwalifikowany do operacji usunięcia torbieli powietrznej ślinianki.

**Wnioski:** Torbiel powietrzna gruczołów ślinowych jest rzadką chorobą, która występuje głównie w śliniankach przyusznych i związana jest ze wzrostem ciśnienia wewnątrzustnego. Metodą leczenia jest przede wszystkim unikanie czynnika wywołującego, stosowanie opatrunków uciskowych, a niekiedy leczenie chirurgiczne. Torbiel powietrzna w większości przypadków nie powoduje trwałego uszkodzenia gruczołów ślinowych, a efekty leczenia są dobre.

**Słowa kluczowe:** ślinianka przyuszna, torbiel powietrzna ślinianki, biopsja aspiracyjna cienkoigłowa, tomografia komputerowa.

### Introduction

There are many reasons for an increase in the volume of the salivary glands. In the initial differential diagnosis, inflammatory changes due to infection, autoimmune or allergic reactions, endocrine disorder, noninfectious granulomatous disease (sarcoidosis), tumor, injury, and other factors such as drugs, alcoholism, malnutrition, and pregnancy should be taken into consideration. One of the rare causes of an increase in the volume of the parotid gland is the formation of a reservoir of air in the parenchyma gland due to air being insufflated through the secretory pathway of the salivary glands via Stensen's duct [1,2]. The main symptoms are painless swelling in the parotid region, crepitus, and frothy saliva on palpation of the gland [1,2,3]. The subcutaneous crepitation can be identified if widespread emphysema occurs [7]. It may occur as a single event or occur periodically, and can affect both adults and children [3,4]. Pathological conditions can affect the parotid glands as well as the mandibular glands [6], although mainly the parotid glands are involved. In the literature, cases have been

called by many names, but the term pneumoparotid is the most frequent [4].

The aim of this study is to present a rare case of a pneumoparotid on the left side.

### Case report

J.S., a 54-year-old man was admitted to the department for diagnosis of a nodular swelling in the area of the left parotid gland. He reported that the swelling appeared about three weeks earlier, initially with pain, and had gradually increased in size. The patient also observed the presence of a small amount of fluid of uncharacteristic taste which appeared in the mouth when pressure was applied to the lesion. He denied head or neck injury, problems with chewing or swallowing food, or dry mouth. The medical history did not indicate chronic increased intraoral pressure associated with e.g. diving, plane flights, recent dental treatment, or playing a wind instrument. The patient reported that from his teenage years he had the habit of pushing his

tongue into the left cheek. He had undergone plastic surgery of the nasal septum 8 years earlier. He was a heavy smoker, smoking about 20 cigarettes a day for the last 30 years.

Laryngological examination revealed swelling of the cheek in the left preauricular area, palpably soft, oval (about 20 × 15 mm), slightly painful when pressed, non-fluctuant, with no associated skin changes (redness or eruptions) and of normal temperature. Lymph nodes not palpable. Body temperature 36.5°C. Laboratory investigations revealed normal values for blood cell count, CRP, and creatinine.

Imaging and fine-needle aspiration biopsy (FNAB) were performed. Ultrasound of the neck showed a thickening of the left parotid parenchyma with a low-echoic focal lesion 12–15 mm in diameter; an acoustic enhancement indicated the presence of calcifications. Lymph node enlargement was not visualised. The result of FNAB: left preauricular area (12 mm diameter) – exudate and multinucleated giant cells.

The result of a contrast neck CT (Figure 1) found a small left parotid gland (2.3 × 1.2 × 2.6 cm), air in the excretory ducts, and an air bubble about 2 cm in diameter in the vicinity of the gland. These findings may be attributed to regressive changes in the parotid gland with damage to the salivary duct opening causing retrograde air flow to the ducts. The right parotid gland was of normal structure, density, and size (3.2 × 2.3 × 3.6 cm). Lymph node enlargement was not found in the neck.

Following these tests, another FNAB was performed. In a smear, indications of a normal salivary gland were detected. Ultrasound and palpation-guided decompression of the recurrent air space gave a shading of about 15 mm in diameter in the left parotid projection.

On the basis of the clinical picture and imaging, an air cyst in the left parotid gland was diagnosed. After its decompression, conservative treatment – a pressure dressing – was applied in an outpatient mode. Due to the lack of therapeutic effect, the patient was then qualified for surgical treatment: excision of the left pneumoparotid.

Under general endotracheal anesthesia, a cutaneous incision was made in the preauricular region in the direction of the mandibular angle on the left side; approaching partially sharp, partially blunt, an air cyst was located within the superficial lobe of the parotid gland penetrating towards the deep lobe. The cyst was approximately 25 × 2 mm, and fused with the salivary gland parenchyma. It was ruptured and the cyst removed in two fragments. The pedicle was stitched up at the bottom of the lodge. Sutures were applied in layers on the subcutaneous tissues and skin. A pressure dressing was placed on the wound area. The surgical procedure was smooth and the postoperative period uneventful.

## Discussion

Pneumoparotid is a unilateral or bilateral enlargement of the salivary gland [21] caused by the presence of air inside the parotid gland. Various terms describing this pathology have been used in the literature: pneumoparotid, pneumoparotiditis, pneumoparotitis, wind parotitis, anesthesia



Figure 1. CT scan (frontal plane) in which a left pneumoparotid is visible

mumps, surgical mumps, pneumo-sialadenitis i pneumatocele glandulae parotitis [5,6].

A pneumoparotid was first described by Hyrtl in 1865. Particularly susceptible to this pathology are persons performing activities involving the production of increased intraoral pressure, including musicians playing wind instruments, glassblowers, and divers [7–9]. Although few cases of air cysts without any apparent cause have been reported, it is believed that predisposing factors relate to facial injury, lung diseases associated with chronic cough (including chronic obstructive pulmonary disease and asthma), nicotine, sleep apnea, weakness of the cheek muscles, bruxism, temporomandibular joint disorders, or psychogenic factors such as stress.

The opening of the parotid duct (Stensen's duct) is located in the cheek near the second upper molar tooth bilaterally. There are three anatomical features of Stensen's duct which protect against reflux of air and saliva into the parotid gland. First, the diameter of the duct's orifice is smaller than the duct section itself. Second, when air pressure increases in the oral cavity, the orifice is covered with several mucosal folds. The slitlike shape of the orifice also helps. Finally, the buccinator and masseter muscles compress the duct in its lateral course, similar to when increased intraoral pressure occurs [17,18]. The anatomical features of Stensen's duct, patulous opening, and mucosal rigidity around the orifice can predispose entry of air into the parotid [12].

Other risk factors are general anesthesia, during which the patient is ventilated via a face mask with gases of elevated pressure [22], spirometry testing [4], manoeuvres to open the Eustachian tube [13], or treatment of sleep apnea (the latter involving a mandibular advancement device which is an alternative for patients who cannot tolerate continuous positive airway pressure [6]).

During physical examinations, a bulged cheek in the parotid area is often observed, and during massage of the lesion foamy saliva or air bubbles escaping from the salivary gland may be seen. Subcutaneous edema may appear and crepitation can occur during salivary gland palpation. The patient sometimes complains of crackling

sounds in the oral cavity and swelling over the parotid region [10–12].

The patient's general condition is also important, although apart from injury it does not usually give cause for concern. Sometimes patients present symptoms such as pain, tenderness, local warmth, or erythema [18]. However, similarly to the patient here, elevated body temperature or other indicators of inflammation, such as elevated CRP values and leukocytosis, are often not observed. Extension of air beyond the parotid can cause subcutaneous emphysema of the face, neck, mediastinum, and subsequent pneumothorax [12]. In the diagnosis of lesions, imaging is important (especially noninvasive ultrasound), sialography, FNAB, and contrast studies (CT or MRI) [12,20].

The lesion can be differentiated with cysts (developmental cysts, mucous cysts or sialoceles, salivary duct cysts, lymphoepithelial cysts), tumors (especially with frequent benign tumors in the parotid glands, i.e., Warthin's tumor, mixed tumor), lymphomas, Kimura's disease, and sialadenitis [12,13].

A pneumoparotid should always be observed. Pressure dressings are the primary treatment option. It is also important to change life-style, reduce exposure to likely triggering factors (e.g. sports, injuries) or start therapy if the cause is for example lung disease. Psychotherapy may be necessary when efforts are needed to change habits or when psychogenic reasons are suspected.

Salivary gland enlargement can resolve itself within several minutes, hours, or days without any treatment [5,12].

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## Conclusions

- Pneumoparotid is a rare disease related to the parotid glands and is associated with an increase in intraoral pressure.
- Treatment is first of all to avoid the triggering factor and apply pressure dressings, but sometimes surgical intervention is necessary.
- In most cases, the pneumoparotid does not cause permanent damage to the salivary glands, and the effects of treatment are usually good.

## Conflict of Interest:

None of the authors have a financial relationship(s) with a commercial interest.

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