

CASE REPORT

Pneumoparotid and Subcutaneous Emphysema in a Man with Psychiatric Disorders

Cosimo Galletti¹, Francesco Ciodaro², Francesco Freni³, Galletti Bruno⁴

Received on: 08 January 2022; Accepted on: 05 February 2022; Published on: 20 March 2023

ABSTRACT

Pneumoparotid is an uncommon clinical presentation of parotid swelling. An incompetent Stensen's duct may cause pneumoparotid. Infectious complications and subcutaneous emphysema are mentioned in the literature. We present a 45-year-old man with numerous episodes of distressing distension of the left parotid region. Bedside inspection is carried out and fluctuant left-sided sore parotid enlargement with crepitation at the palpation is noted. A head-neck computed tomography (CT) study with oral medium contrast is performed marking heavy emphysema of subcutaneous layer that is extension gone from temporal region to the upper thoracic outlet, also is noted an important ectasia of Stensen's duct, salivary glands, and left parotid. Medical treatment with antibiotic therapy and support measures are administered with a complete resolution of subcutaneous emphysema. Pneumoparotid could be correlated with auto-insufflation behavior common in psychiatric conditions and some professions, it is essential besides medical treatment, a psychosocial follow-up in addition.

Keywords: Computed tomography scan, Head and neck region, Parotid gland, Parotid swellings, Parotitis, Salivary gland.

Otorhinolaryngology Clinics: An International Journal (2022): 10.5005/jp-journals-10003-1411

BACKGROUND

The persistence of the air in the parotid parenchyma is called pneumoparotid. The pneumoparotid is first described in 1865 which indicates the presence of air within the parotid gland and Stensen's duct.¹

It also was reported at the beginning of 1915 during an infection of mumps affected the soldiers of the French Foreign Legion based in North Africa. The soldiers intentionally blow into a small bottle or perform Valsalva's maneuver, provoking an increased intraoral pressure that leads to a condition of self-inducing pneumoparotid.²⁻⁵ It is an intentionally self-inducing attitude to avoid duty.² When inflammation or infection of the gland occurred is called pneumoparotitis. In general, the patient was referred to pain in the region and swelling at the same time. One of the most important complications is subcutaneous emphysema.^{6,7} It is just more frequent in adults but it is a very unknown clinical condition in children.⁸ The pneumoparotid is more frequent in young adults and adults with psychiatric disorders.^{2-6,9-12} Appropriate clinical history and imaging studies like echography, sialendoscopy, and CT are essential to perform a correct diagnosis.^{4,5,12} The medical therapeutic approach is generally implicated, reserving surgery in severe cases.¹⁰⁻¹³ We present a case of recurrent pneumoparotid in a male middle-age patient with psychosocial issues and an exhaustive review of the literature.

CASE DESCRIPTION

We present a case of a male patient of 45-year-old with a family history of cardiopathy, diabetes, and obesity. He suffered from cataracts and retinal detachment and received multiple dental treatments,¹⁴ the last in September 2019. Treated in November 2015 for a periorbital fat herniation,¹⁵ partial parotidectomy for pleomorphic adenoma in the right parotid.¹⁶ The patient presents multiple attacks of intermittent, painful, monolateral left facial swelling in the last years. The clinical exam showed a fluctuant left-sided parotid swelling, associated with pain, with crepitus.

^{1,2}Department of ENT, Azienda Ospedaliera Universitaria "G Martino", Università di Messina, Messina, Italy

^{3,4}Department of Adult and Development Age Human Pathology, "Gaetano Barresi", Unit of Otorhinolaryngology, University of Messina, Messina, Italy

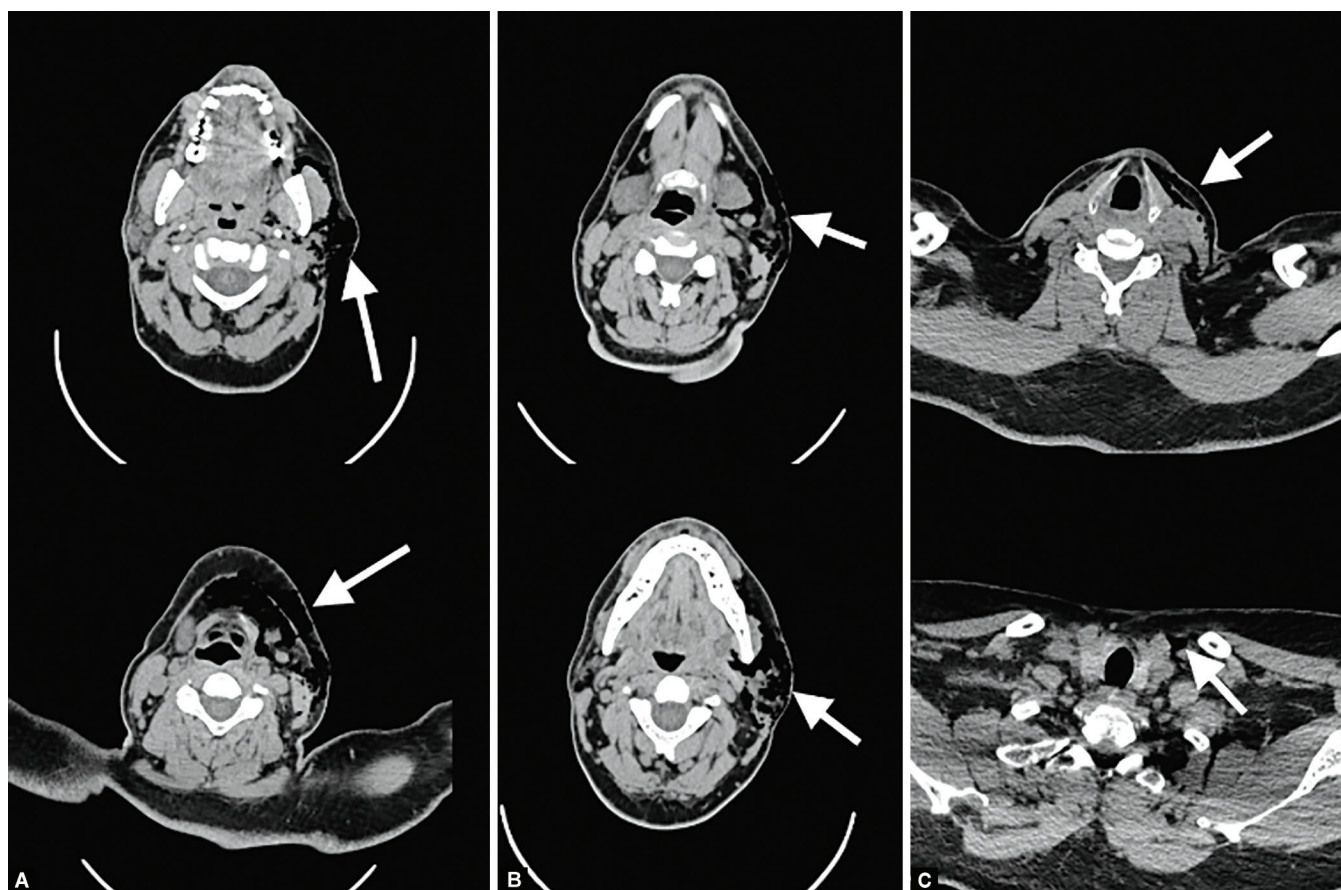
Corresponding Author: Cosimo Galletti, Department of ENT, Azienda Ospedaliera Universitaria "G Martino", Università di Messina, Messina, Italy, e-mail: cosimogalletti92@gmail.com

How to cite this article: Galletti C, Ciodaro F, Freni F, *et al.* Pneumoparotid and Subcutaneous Emphysema in a Man with Psychiatric Disorders. *Int J Otorhinolaryngol Clin* 2022;14(3):110-113.

Source of support: Nil

Conflict of interest: None

Lymph-nodal cervical stations are negatives. Intraoral examination showed that is partial edentulous, the oro-buccal mucosa is lesioned by chronic traumatizes and Stensen's duct orifice appears dilated. Dentistry treatment on the lower left side molar teeth near the opening of Stensen's duct is done in the last year. The facial nerve function is preserved. Head and neck computed tomography with oral contrast medium study documents a heavy quote marking heavy emphysema of subcutaneous layer, that is extension gone from temporal region to the upper thoracic outlet, and an important ectasia of Stensen's duct, salivary glands, and left parotid is also noted. The study is carried out after the ingestion of the oral contrast medium showing reflux in the Stensen's duct on the left and some homolateral salivary glandular ducts (Fig. 1). The patient is admitted to the maxillofacial department. After performing the CT exam, the patient was suffering from subjective vertigo with instability sensation. At the otorhinolaryngology counseling, spontaneous nystagmus was absent and maneuvers for benign positional vertigo are negative.¹⁷ Conscious of the patient's psychiatric disorder, a psychiatric consultant is required, reporting that the man suffered from a minor cognitive disability with a tendency to somatization,



Figs 1A to C: Head and neck computed tomography with oral contrast medium study documented a very important presence of subcutaneous emphysema that affected caudocranial left soft tissues from temporal region to the upper thoracic outlet, severe ectasia of Stensen's duct, ducts of salivary glands and left parotid. The study is carried out after ingestion of oral contrast medium showing the presence of a reflux in the Stensen's duct on the left and some homolateral salivary glandular ducts. (A) Subcutaneous emphysema in temporal region, severe ectasia of Stensen's duct, ducts of salivary glands and left parotid; (B) The study is carried out after ingestion of oral contrast medium showing the presence of a reflux in the Stensen's duct on the left and some homolateral salivary glandular ducts; (C) Subcutaneous emphysema that affected caudo-cranial left soft tissues from temporal region to the upper thoracic outlet

underlying an important state of anxiety, and insomnia. Medical therapy with venlafaxine tablets 75 mg one administration daily after lunch, quetiapine tablets 25 mg one administration daily after dinner, and alprazolam six drops are prescribed. To avoid possible infective complications during the hospital stay, antibiotic therapy with one vial of amoxicillin and clavulanic acid 1 g three times daily is performed and a vial of acetaminophen 1 g if needed.¹⁸ No incidences occurred during the stay in the hospital, day by day facial swelling and subcutaneous emphysema passed, vital parameters are stables, and the patient is discharged on the 7th day of admission. We recommended avoiding Valsalva's maneuvers and a psychiatric follow-up is required.

Outcome and Follow-up

After 6 months from the discharge, the first control follow-up visit is performed, the patient does not report having suffered from the further episodes.

We have scheduled a second follow-up visit at 12 months. He is currently being followed by a psychiatric specialist.

DISCUSSION

Pneumoparotid is a singular clinical presentation of parotid swallowing, usually caused by stasis of the air in the parenchyma.

It is related to a retrograde insufflation of the air and the saliva via Stensen's duct,⁵ also anatomic predisposition related to a hypotonicity of buccinator muscle and/or hypertrophy of the master muscle and/or stenosis of Stensen's duct are reported in the literature.¹

The literature considers a most important mechanism of prevention of air and saliva reflux, the anatomic dimension of the duct orifice, the mucosal layer that protects the duct aperture when the intraoral pressure increases, and the actions of the masseter muscle and buccinator muscle that compress in its lateral course the Stensen's duct.

Frequently it is associated with glassblowing, playing wind instruments, exercising, and self-induced conduct many times associated with psychiatric disorders. Normally, the intraoral pressure is averagely 2 mm Hg, in glassblowing, and trumpet playing a value of 150 mm Hg is registered to predispose the onset of the disease.

Also, the literature described a type of iatrogenic pneumoparotid consequently a spirometry test performing, oral surgery, fine-needle aspiration test for cytology of the parotid gland, and positive pressure ventilation used preoperatively or in the intensive care setting^{2-12,19-21} also drug-induced is described. In the literature is reported a case of pneumoparotid in patients who undergone

a chronic use of oronasal continuous positive airway pressure. Infectious agents, like viruses and bacteria, and autoimmune diseases, like Sjögren syndrome, sarcoidosis, hypothyroidism, liver disease, Cushing disease, Wegener's vasculitis, and diabetes, are reported in the literature as possible etiologic agents of pneumoparotid or pneumoparotitis.²²

Numerous events provoke chronic inflammation processes or reinfection or sialectasias. The use of "puffed-cheek" technique leads us to explain the pathogenesis of the disease's onset.¹³

The literature shows that repeating exercises of autoinsufflation may cause the acini's rupture. The glandular capsule is open in the superomedial side at the posterior border of mandible bone, the emphysema may be caused by the reaching of the parapharyngeal and retropharyngeal space.² In our case, the patient's CT scan revealed the presence of emphysema in the left side neck region, probably due to the frequent auto-induced Valsalva's maneuvers that the patient still recreated during his daytime, counseling about change in his habits is performed.

Radiologic studies are imprescindibile to perform a good diagnosis; multiple techniques are indicated, in particular, ultrasonography and nuclear magnetic resonance (NMR) are the most suggested.²³

The echography is strongly recommended in the diagnosis of superficial tissue of the cervical region and to study the parotid diseases associated. It delineates the air in the parotid gland, ducts, and soft tissue. Sialography study is less sensitive however is useful to establish the presence of calculi.^{2,4}

The gold standard technique CT result allows us to define the anatomy and is a noninvasive technique. The CT study describing an enlargement of Stensen's duct, distension of glandular acini, free air into the glandular parenchyma, and a good representation of the glandular duct tree, also delimited the extension of the air volume in the nearest cervical areas.^{2,24} Puffed-cheek CT is a technique describing an enlargement in intraductal and intraglandular parotid due to the airflow compared to the simple CT.¹³

Acute situation handling embraces the use of antibiotic therapy, also intravenous; the role of steroid therapy is well defined when the swelling and pain are massive and also analgesia is considered if the pain increases.

The therapeutical approach is based on apport hydrique, mouthwashes, frottage of the gland, sialogogues, and warm compresses. A psychotherapeutical approach is needed in self-induced pneumoparotid cases.

The surgical approach is mandatory when the recurrences are associated with infection or complication like pneumomediastinum; in these cases, the gold standard technique is the parotid duct ligation. Parotidectomy is the endpoint line treatment, its implication is destined to the noncompliant patient or when medical treatment fails or the patient suffered from recurrent infections.²⁵⁻²⁹ The parotidectomy may lead to important complications like partial or complete facial nerve lesion, Frey's syndrome,³⁰ salivary fistula, auricular Magnus nerve lesion, and keloid cicatrization of surgical incision.

To prevent pneumoparotid is already fundamental the medical support to explain which activities that increase intraoral pressure must have to stop.

Our patient referred to several cases of facial swelling during last years that suggested a particular disposition. Focusing on his medical history, are reported depression, a mild cognitive impairment with a propensity to somatization due to anxiety and

insomnia. Probably our patient provoked his condition performing autoinflation and Valsalva's maneuvers due to his psychological disorders. Despite the cause of pneumoparotid was not ascertained was in this man, the only intervention that we performed was antibiotic therapy as prophylaxis from a possible superinfection of subcutaneous emphysema and supportive therapy. It is essential to consider psychotherapeutic counseling to support our patient in his follow-up.

LEARNING POINTS/TAKE-HOME MESSAGES

- Pneumoparotid is a singular clinical presentation of parotid swelling. Usually caused by stasis of the air in parotid parenchyma. In general, the patient presents multiple attacks of intermittent, painful, facial swelling. One of the most important complications is subcutaneous emphysema. Literature shows that repeating exercises of autoinsufflation may cause the glandular acini's rupture.
- For a good diagnosis an exhaustive anamnesis and radiological study like CT, actually the gold standard technique, are essential.
- The medical therapeutic approach is normally actuated, reserving surgery in severe cases.
- Psychotherapeutical approach is already fundamental to prevent the maneuvers that may increase intraoral pressure avoiding the recurrent pneumoparotid.

ORCID

Cosimo Galletti  <https://orcid.org/0000-0002-7226-4596>

REFERENCES

1. Zuchi D, Da Silveira P, Cardoso C, et al. Pneumoparotitis. *Braz J Otorhinolaryngol* 2011;77(6):806. PMID: 22183290.
2. McGreevy A, O'Kane A, McCaul D, et al. Pneumoparotitis: a case report. *Head Neck* 2013;35(2):E55–E59. DOI: 10.1002/hed.21873.
3. Markowitz-Spence L, Brodsky L, Seidell G, et al. Self-Induced pneumoparotitis in an adolescent. Report of a case and review of the literature. *Int J Pediatr Otorhinolaryngol* 1987;14:113–121. DOI: 10.1016/0165-5876(87)90021-8.
4. Ghanem M, Brown J, McGurk M. Pneumoparotitis: a diagnostic challenge. *Int J Oral Maxillofac Surg* 2012;41:774–776. DOI: 10.1016/j.ijom.2011.12.013.
5. Kostantinidis I, Chatziavramidis A, Constantinidis J. Conservative management of bilateral pneumoparotitis with sialendoscopy and steroid irrigation. *BMJ Case Rep* 2014;2014:bcr2013201429. DOI: 10.1136/bcr-2013-201429.
6. Birzgalis A, Curley J, Camphor I. Pneumoparotitis, subcutaneous emphysema and pleomorphic adenoma. *J Laryngol Otol* 1993;107(4):349–351. DOI: 10.1017/S0022215100123011.
7. Lee K, James V, Ong G. Emergency department diagnosis of idiopathic pneumoparotitis with cervicofacial subcutaneous emphysema in a pediatric patient. *Clin Pract Cases Emerg Med* 2017;1(4):339–402. DOI: 10.5811/cpcem.2017.7.34879.
8. Balasubramanian S, Srinivas S, Aparna K. Pneumoparotitis with subcutaneous emphysema. *Indian Pediatr* 2008;45(1):58–60. PMID: 18250510.
9. House L, Lewis A. Pneumoparotitis. *Clin Exp Emerg Med* 2018;5(4):282–285. DOI: 10.15441/ceem.17.291.
10. McCormick M, Bawa G, Shah R. Idiopathic recurrent pneumoparotitis. *Am J Otolaryngol Head Neck Med Surg* 2013;34(2):180–182. DOI: 10.1016/j.amjoto.2012.11.005.
11. Prabhu S, Tran B. Pneumoparotitis. *Pediatr Radiol* 2008;38(10):1144. DOI: 10.1007/s00247-008-0930-x.

12. Gazia F, Freni F, Galletti C, et al. Pneumoparotid and pneumoparotitis: a literary review. *Int J Environmental Res Public Health* 2020;17(11):3936. DOI: 10.3390/ijerph17113936.
13. Lasboo A, Nemeth A, Russell E, et al. Use of the “puffed-cheek” computed tomography technique to confirm the diagnosis of pneumoparotitis. *Laryngoscope* 2010;120(5):967–969. DOI: 10.1002/lary.20879.
14. Flores-Orozco EI, Tiznado-Orozco GE, Díaz-Peña R, et al. Effect of a mandibular advancement device on the upper airway in a patient with obstructive sleep apnea. *J Craniofac Surg* 2020;31(1):e32–e35. DOI: 10.1097/SCS.0000000000005838.
15. Galletti B, Gazia F, Galletti C, et al. Endoscopic treatment of a periorbital fat herniation caused by spontaneous solution of continuity of the papyracea lamina. *BMJ Case Rep* 2019;12(4):e229376. DOI: 10.1136/bcr-2019-229376.
16. Galletti B, Freni F, Gazia F, et al. A rare case of a huge pleomorphic adenoma of minor salivary glands in the parapharyngeal space. *EuroMediterranean Biomed J* 2019;14(2):7–10. DOI: 10.3269/1970-5492.2019.14.02.
17. Ciodaro F, Mannella VK, Nicita RA, et al. Therapeutic efficacy of the Galletti-Contrino manoeuvre for benign paroxysmal positional vertigo of vertical semicircular canals in overweight subjects. *Eur Arch Otorhinolaryngol* 2018;275(10):2449–2455. DOI: 10.1007/s00405-018-5086-1.
18. Nicot R, Myon L, Konopnicki S, et al. Pneumoparotid: a rare case of recurrent parotid swelling. *Rev Stomatol Chir Maxillofac Chir Orale* 2014;115(2):111–113. DOI: 10.1016/j.revsto.2013.03.005.
19. Abdullayev R, Saral F, Kucukebe O, et al. Bilateral parotitis in a patient under continuous positive airway pressure treatment. *Rev Brasil Anestesiol* 2014;66(6):661–663. DOI: 10.1016/j.bjan.2014.05.010.
20. Goates AJ, Lee DJ, Maley JE, et al. Parotitis as a complication of long-term oronasal positive airway pressure for sleep apnea. *Head Neck* 2017;00:1–4. DOI: 10.1002/hed.25003.
21. Ambrosino R, Lan R, Romanet I, et al. Severe idiopathic pneumoparotitis: case report and study review. *Int J Pediatr Otorhinolaryngol* 2019;124:196–198. DOI: 10.1016/j.ijporl.2019.07.012.
22. Moenne K, Cordero J, Poli C. Neumoparotiditis o neumoparótida en el niño: Un diagnóstico diferencial a considerar. *Rev Chil Infectivol* 2009;26(6):555–559. DOI: 10.4067/S0716-10182009000700012.
23. Alnaes M, Furevik LL. Pneumoparotitis. *Tidsskr Nor Lægeforen* 2017;137(7):544. DOI: 10.4045/tidsskr.16.0633.
24. Gudlaugsson O, Geirsson AJ, Benediktsdóttir K. Pneumoparotitis: a new diagnostic technique and a case report. *Ann Otol Rhinol Laryngol* 1998;107(4):356–358. DOI: 10.1177/000348949810700416.
25. Golz A, Joachims HZ, Neter A, et al. Pneumoparotitis: diagnosis by computed tomography. *Am J Otolaryngol* 1999;20(1):68–71. DOI: 10.1016/s0196-0709(99)90055-8.
26. Curtin JJ, Ridley NTF, Cumberworth VL, et al. View from within: radiology in focus: pneumoparotitis. *J Laryngol Otol* 1992;106(2):178–179. DOI: 10.1017/S0022215100119024.
27. Meerleer KD, Hermans R. Images in clinical radiology: pneumoparotitis. *JBR-BTR* 2005;88:248. <https://doi.org/10.1002/hed.21873>.
28. Kirsch DDM, Shinn J, Porzio R, et al. Pneumoparotid due to spirometry. *Chest* 1999;116(5):1475–1478. DOI: 10.1378/chest.116.5.1475.
29. Chilla R, Meyforth HO, Arglebe C. Surgical treatment of chronic parotitis. *Arch Otorhinolaryngol* 1982;234(1):53–56. DOI: 10.1007/BF00453538.
30. Freni F, Gazia F, Stagno D’Alcontres F, et al. Use of botulinum toxin in Frey’s syndrome. *Clin Case Rep* 2019;7(3):482–485. DOI: 10.1002/ccr3.2019.