

# Obstructive Sleep Apnea Syndrome Associated with Soft Palate Pleomorphic Adenoma: Case Report and Review of Literature

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## Abstract

**Context:** Pleomorphic adenoma is the most common benign tumor arising in the salivary gland. Soft palatine localisation is rare but can cause an obstructive syndrome. We report a case of pleomorphic adenoma of the soft palate causing obstructive sleep apnea (OSA).

**Case report:** A 62-year-old woman was addressed to ENT consultation because of obstructive sleep-disorder breathing. On physical exam we discovered a submucosal mass of the right soft palate of 3 x 2 cm. Therapy consisted of a tumor excision. Histopathology spoke in favour of a pleomorphic adenoma. Excision of the local mass led to sleep exam normalization and symptoms resolution.

**Conclusion:** Oropharyngeal and oral cavity tumors may cause obstructive sleep-disorder breathing. All patients with sleep disordered should undergo a thorough ENT clinical examination.

**Keywords:** Sleep-disorder breathing ; Palatine mass; Pleomorphic adenoma

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## Introduction

Pleomorphic adenoma is a common salivary benign tumor mostly affecting parotid glands. It draws its name from its histological architecture which can be subdivided in three types: classical, myxoid and hypercellular. The myxoid type accounts for more than half of cases. In the major salivary glands, the tumor is enclosed by a layer of fibrous tissue termed a capsule. Incomplete capsule is observed in 33-49% of pleomorphic adenomas [1]. A small minority of tumors are located in the oral and nasal cavity [2]. They appear as slowly growing mass usually without symptoms. The differential diagnosis includes abscess, cysts, fibroma, lipoma, neurofibroma or squamous cell carcinoma [3,4]. Tumour spillage increases the risk of recurrence. Meticulous surgical excision with capsular integrity is the treatment of first choice. Recurrence is described in about 46% of cases and risk of malignant transformation in 3.3 to 13% [1]. It is rarely associated with obstructive sleep-disordered breathing. Obstructive sleep apnea (OSA) is a common well recognised entity mainly caused by obesity. The Apnea-Hypopnea Index (AHI) is defined as the number of apneas or hypopneas per hour of sleep and is mapped

to disease severity [5]. Patients are considered to have OSA if their AHI is greater than 15 or if they have minimum oxygen saturation below 90 (American Academy of Sleep Medicine). In 1981, Moses et al. first reported a patient with OSA caused by nasopharyngeal carcinoma [6]. Since then 30 articles about tumors of the upper aerodigestive tract causing sleep disorder have been published [7,8]. Rada et al. studied the relationships between OSA and head and neck tumors [9]. The most frequent localisations were salivary gland, larynx, nasopharynx and oropharynx. There were 16 benign and 8 malignant neoplasms. The most frequent benign neoplasm related to OSA were lipomas. Among malignant lesions the most common were squamous cell carcinoma and non-Hodgkins lymphoma. We report a case of pleomorphic adenoma of the soft palate causing OSA and make a literature review.

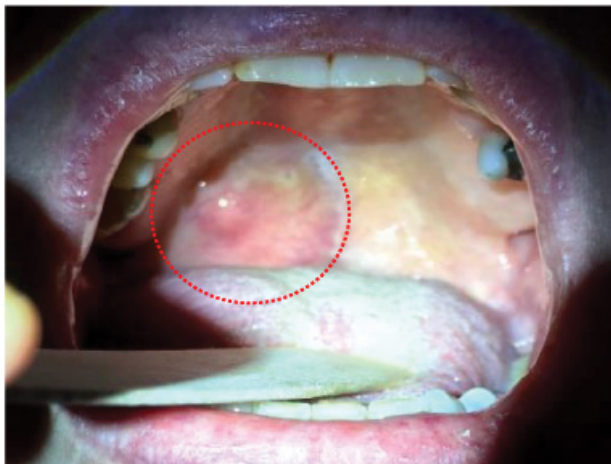
## Case Report

A 62-year-old woman was addressed to Ear Nose and Throat (ENT) consultation because of sleep obstructive breathing disordered symptoms including snoring and daytime fatigue.

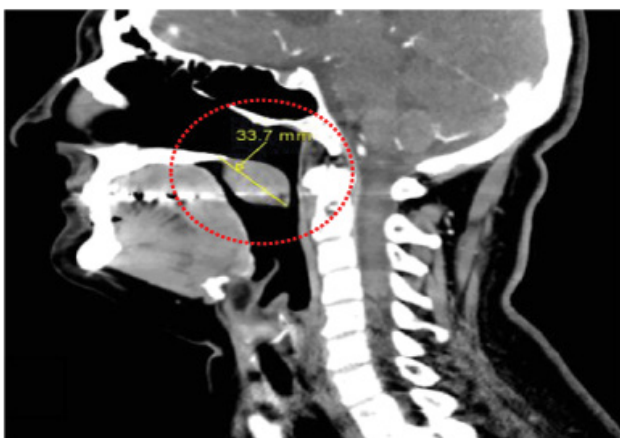
She was non-smoker and had a social alcohol consumption. The ENT work-up included clinical exam, head and neck computed tomographic scan and sleeping test. On physical exam we discovered a submucosal mass of the right soft palate crossing the midline (**Figure 1**). The patient had a good oral care and satisfactory dental condition. There was no mucous lesion in the oral cavity and no cervical lymph node. The rest of the ENT exam was normal. The computed tomographic scan identified a mass of 3 × 2.4 × 2.3 cm in the soft palate without invasion of the adjacent tissues (**Figure 2**). Preoperative polygraphy confirmed OSA with AHI of 17, 2 per hour (**Table 1**). Therapy consisted of surgical tumor removal preserving the soft palate (**Figure 3**). No OSA surgery was performed. The defect of the soft palate was treated by primary closure without reconstruction. Histopathology concluded to entirely resected pleomorphic adenoma (**Figure 4**). Soft palate healed without complication (**Figure 5**). Snoring during night disappeared. Postoperative polygraphy realised 6 months later confirmed normal sleep with AHI of 5, 9 per hour (**Figure 6 and Table 1**).

**Table 1:** Pre- and postoperative polygraphy, AHI=Apnea-Hypopnea Index, ODI=Oxygen Desaturation Index.

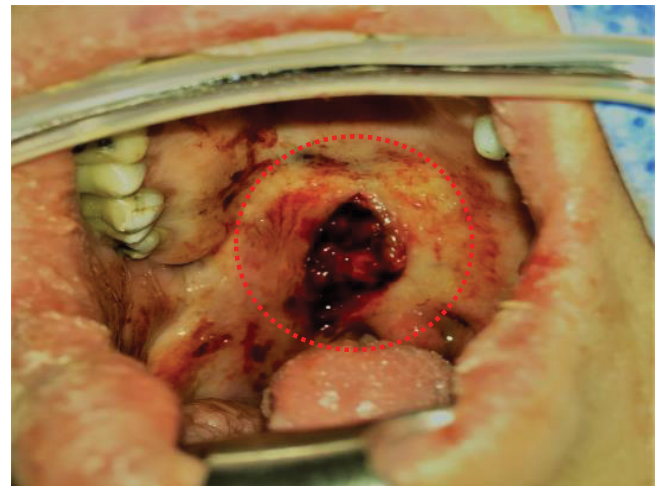
	Pre operative (29.11.17)	Post operative (01.06.18)
IAH	17.2 (<5/h)	5.9 (<5/h)
ODI	21.4 (<5/h)	11.1 (<5/h)
Period of O <sub>2</sub> <90%	349 min (66%)	50 min (15%)



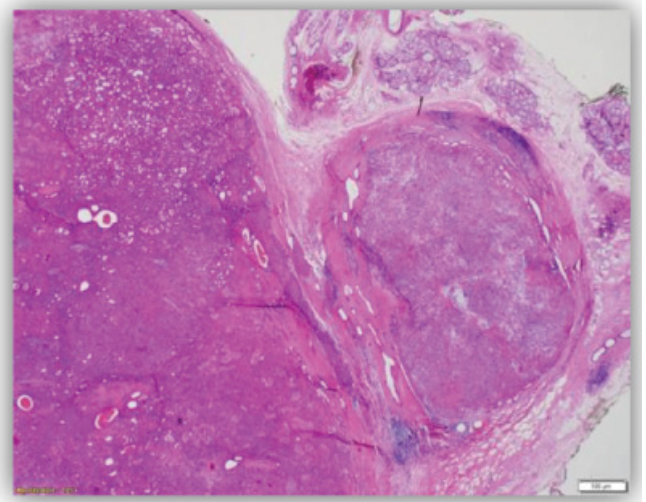
**Figure 1** Oropharyngeal examination: submucosal mass of the right soft palate crossing the midline.



**Figure 2** Sagittal view of CT scan. Mass in the soft palate, measuring 3 x 2.4 x 2.3 cm.



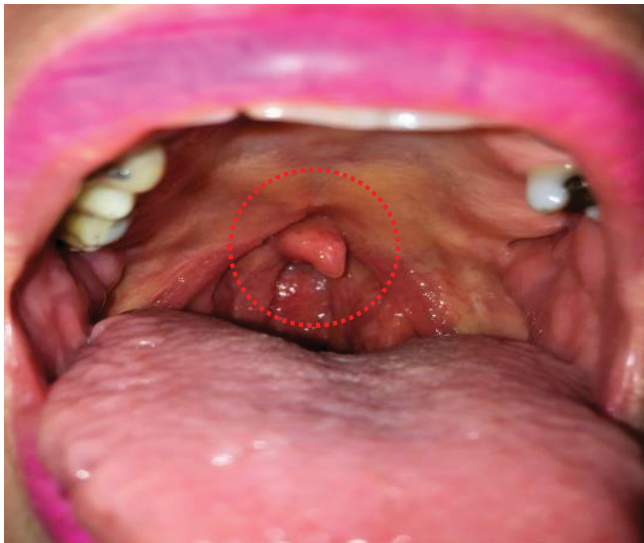
**Figure 3** Endoscopic view during tumor resection.



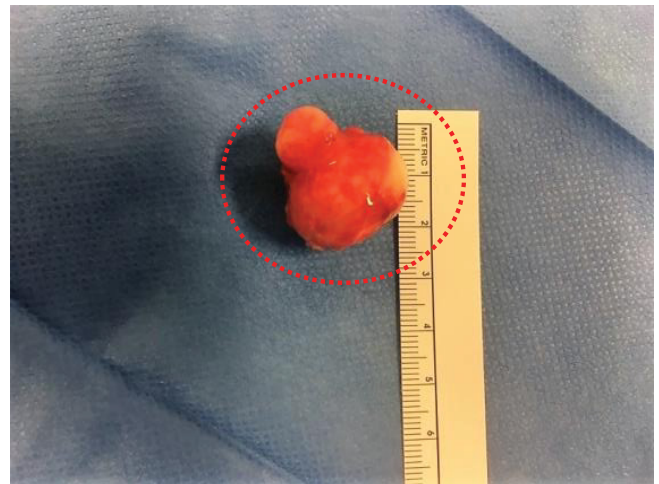
**Figure 4** Microscopy analysis findings confirm pleomorphic adenoma.

## Discussion

Pleomorphic adenoma accounts for approximately 60% of all salivary gland neoplasms [10]. A small minority of tumors are located in the oral cavity, neck and nasal cavity [4,11]. In those localisations it appears as slowly growing painless mass [9] usually in the fourth or fifth decade of life, with a higher frequency in female [4,1]. The diagnosis is established on the basis of history, physical examination and histopathology [12]. Computed tomography scan and MRI can provide information



**Figure 5** Endoscopic view of soft palate at 1 month of follow.



**Figure 6** Macroscopic specimen resected.

on the location and size of the tumor, besides extension to surrounding structures [13,14]. Treatment consists of surgical tumor excision and/or continuous positive airway pressure [9]. Recurrence is rare after complete removal and the prognosis is excellent [15-17]. In the English-language literature, nine articles relate pleomorphic adenomas causing OSA; five cases of deep-lobe parotid tumors extending into the parapharyngeal space [18-22], four cases arising from the minor salivary gland in the oral cavity and pharynx [23-27]. Tumors of the upper aerodigestive tract causing secondary OSA have been rarely reported in large series. Rada et al. [9] make a review finding only 30 articles about OSA caused by head-and-neck

tumors, including 16 benign and 8 malignant neoplasms. Payne et al. [27] conclude that 13 of 17 (76.4%) patients with oral and oropharyngeal cancer have OSA. The most common symptom is sudden aggravation of snoring or gasping during night [17,27].

## Conclusion

All patients with sleep disordered breathing should undergo a thorough ENT clinical examination [12,21]. Oral cavity and oropharyngeal tumors belong to the differential diagnosis of OSA. Sudden aggravation of snoring should raise suspicion of an oropharyngeal tumor.

## References

- 1 De Sousa Lopez ML, Amorim Barroso KM, Gomes Henriques AC, Dos Santos JN, Martins MD, et al. (2016) Pleomorphic adenomas of the salivary glands: retrospective multicentric study of 130 cases with emphasis on histopathological features. *Eur Arch Otorhinolaryngol* 274: 543-551.
- 2 Pires FR, Pringle GA, de Almeida OP, Chen SY (2007) Intra-oral minor salivary gland tumors: a clinicopathological study of 546 cases. *Oral Oncol* 43: 463-470.
- 3 Ansari MH (2007) Salivary gland tumors in an Iranian population: a retrospective study of 130 cases. *J Oral Maxillofac Surg* 65: 2187-2194.
- 4 Toida M, Shimokawa K, Makita H, Kato K, Kobayashi A, et al. (2005) Intraoral minor salivary gland tumors: a clinicopathological study of 82 cases. *Int J Oral Maxillofac Surg* 34: 528-532.
- 5 Young T, Peppard PE, Gottlieb DJ (2002) Epidemiology of obstructive sleep apnea: a population health perspective. *Am J Respir Crit Care Med* 165: 1217-1239.
- 6 Moses FM, Buscemi JH (1981) Obstructive sleep apnea syndrome associated with nasopharyngeal carcinoma. *West J Med* 134: 69-70.
- 7 Stradling JR, Davies RJ (2004) Obstructive sleep apnoea/hypopnoea syndrome: definitions, epidemiology, and natural history. *Thorax* 59: 73-78.
- 8 Koenig S (2001) Pulmonary complications of obesity. *Am J Med Sci* 321: 249-279.
- 9 Rada R (2005) Obstructive sleep apnea and head and neck neoplasms. *Otolaryngol Head Neck Surg* 132: 794-799.
- 10 Pinkston JA, Cole P, Incidence rates of salivary gland tumors: results from a population-based study. *Otolaryngol Head Neck Surg* 120: 834-840.
- 11 Vellios F, WG Shafer (1959) Tumors of the intraoral accessory salivary glands. *Surg Gynecol Obstet* 108: 450-456.
- 12 Casale M, Frari V, Vincenzi B, Bressi F, Quattrocchi CC, et al. (2012) Upper airway study should always come before any sleep study in OSAS evaluation: a giant parapharyngeal lipoma behind OSAS. *Eur Rev Med Pharmacol Sci* 16: 106-109.
- 13 Bullerdiel J, Wobst G, Meyer-Bolte K, Chilla R, Haubrich J, et al. (1993) Cytogenetic subtyping of 220 salivary gland pleomorphic adenomas: correlation to occurrence, histological subtype, and in vitro cellular behavior. *Cancer Genet Cytogenet* 65: 27-31.



- 14 Tuncyurek O, Eyigor H, Ozkul A (2011) What is the importance of radiology in the obstructive sleep apnea?. *J Craniofac Surg* 22: 1971-1973.
- 15 Marioni G, Marino F, Stramare R, Marchese-Ragona R, Staffieri A, et al. (2003) Benign metastasizing pleomorphic adenoma of the parotid gland: a clinicopathologic puzzle. *Head Neck* 25: 1071-1076.
- 16 Bradley PJ (2005) Metastasizing pleomorphic salivary adenoma should now be considered a low-grade malignancy with a lethal potential. *Curr Opin Otolaryngol Head Neck Surg* 13: 123-126.
- 17 Sabesan T, Ramchandani PL, K Hussein (2007) Metastasizing pleomorphic adenoma of the parotid gland. *Br J Oral Maxillofac Surg* 45: 65-67.
- 18 Pang KP, Goh CHK, Tan HM (2002) Parapharyngeal space tumours: an 18 year review. *J Laryngol Otol* 116: 170-175.
- 19 Moriariu I, Dias A, Curran A (2012) Giant parotid pleomorphic adenoma in the parapharyngeal space causing severe obstructive sleep apnoea. *Ir Med J* 105: 184-185.
- 20 Mulla O, Agada F, Dawson D, Sood S (2013) Deep lob parotid pleomorphic adenoma presenting as obstructive sleep apnoea. *BMJ Case Rep* 2013: bcr2013008655.
- 21 Mulla O, Agada F, Dawson D, Sood S (2011) Obstructive sleep apnoea and snoring-is examination necessary? *Aust Fam Physician* 40: 886-888.
- 22 Adams AJ, Patterson AR, Brady G, Whitfield PH (2008) Resolution of obstructive sleep apnoea after resection of a pleomorphic salivary adenoma. *Br J Oral Maxillofac Surg* 46: 53-54.
- 23 Yilmaz AD, Uniu E, Orbay H, Sensoz O (2006) Giant pleomorphic adenoma of soft palate leading to obstruction of the nasopharyngeal pot. *J Craniofac Surg* 17: 1001-1004.
- 24 Giddings CEB, Bray D, Rimmer J, Williamson P (2005) Pleomorphic adenoma and severe obstructive sleep apnoea. *J Laryngol Otol* 119: 226-229.
- 25 Motomura H, Harada T, Muraoka M, Taniguchi T (2000) Elongated uvula with a pleomorphic adenoma: a rare cause of obstructive sleep apnea syndrome. *Ann Plast Surg* 45: 61-63.
- 26 Murthy SV, Murthy NC, Belagavi CS, Munishwara GB (2003) A large pleomorphic adenoma of soft palate causing sleep apnea syndrome-a case report. *Indian J Pathol Microbiol* 46: 466-467.
- 27 Payne RJ, Hier MP, Kost KM, Black MJ, Zeitouni AG, et al. (2005) High prevalence of obstructive sleep apnea among patients with head and neck cancer. *J Otolaryngol* 34: 304-311.