

A Thyroglossal Duct Cyst Causing Obstructive Sleep Apnea in Adult

Hahn Jin Jung¹ · Jeong-Whun Kim¹ · Chul Hee Lee¹ · Young Jun Chung² · Ji-Hun Mo²

¹Department of Otorhinolaryngology-Head and Neck Surgery, Seoul National University Bundang Hospital, Seoul National University College of Medicine, Seongnam; ²Department of Otorhinolaryngology-Head and Neck Surgery, Dankook University College of Medicine, Cheonan, Korea

Obstructive sleep apnea (OSA) is a common disorder. It usually results from the structural compromise of the upper airway. In patients with OSA, the obstruction predominantly occurs along the pharyngeal airway, and also a variety of tumors have been reported to cause such a condition. We present here the case of a thyroglossal duct cyst causing OSA in adult. This case demonstrates that thyroglossal duct cyst or some kind of mass lesions in the airway lesions should be considered in the differential diagnosis of OSA patients.

Keywords. Thyroglossal duct cyst, Sleep apnea syndromes, Sleep disorders

INTRODUCTION

Obstructive sleep apnea (OSA) is a highly prevalent disease that is characterized by recurrent episodes of upper airway obstruction and it usually results from the structural compromise of the upper airway accompanied with decrease in muscle tone. In patients with OSA, the obstruction predominantly occurs along the pharyngeal airway, and a variety of tumors have been reported to cause such a condition. Here we describe an interesting case of OSA with a thyroglossal duct cyst (TGDC) with preoperative and postoperative sleep videofluoroscopic findings.

CASE REPORT

A 51-year-old man visited the Sleep Center with a 2-year history of snoring and voice change. He had complaints about loud snoring and had been witnessed to have obstructive sleep apnea with excessive daytime somnolence. His Epworth sleepiness

scale (ESS) score was 13. He denied any history of dyspnea. His medical history included mild hypertension and diabetes. His height was 160 cm, his weight was 62 kg and his body mass index was 24.2 kg/m².

On physical examination, his Friedman tongue position [1] was grade III (only the soft palate is visible) and the tonsils were hidden in the tonsillar fossa. On flexible laryngoscopy, about a 20×20 mm sized round cystic mass at the base of tongue was identified. The cyst bended the epiglottis posteriorly, which made the airway narrow. The vocal cords could not be observed because of the lesion (Fig. 1A).

He underwent a full-night laboratory nocturnal polysomnography (PSG). PSG revealed that the respiratory disturbance index (RDI) was 32.2 events per hour of sleep with the supine RDI of 105.8 events/h. The longest apnea duration was 39.7 seconds. The lowest SaO₂ was 89%, and the average SaO₂ saturation was 97.2%.

The computed tomograms showed a 33×31×27 mm sized well-defined cystic lesion located at the midline of the tongue base, compressing the oropharyngeal airway (Fig. 1B, D). The epiglottis was displaced posteriorly by the cystic lesion.

To evaluate the upper airway obstruction and to find any other obstructive lesion, sleep videofluoroscopy (SVF) was performed with administration of 0.05 mg/kg of midazolam as described previously [2]. SVF showed that the pharyngeal airway was completely obstructed during both inspiratory and expiratory phase when the mass pulled the epiglottis down and back-

• Received August 11, 2010
Revised October 17, 2010
Accepted November 1, 2010

• Corresponding author: **Ji-Hun Mo**
Department of Otorhinolaryngology-Head and Neck Surgery,
Dankook University College of Medicine, 201 Manghyang-ro,
Dongnam-gu, Cheonan 330-180, Korea
Tel: +82-41-550-3933, Fax: +82-41-556-1090
E-mail: jihunmo@gmail.com

Copyright © 2013 by Korean Society of Otorhinolaryngology-Head and Neck Surgery.

This is an open-access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (<http://creativecommons.org/licenses/by-nc/3.0>) which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

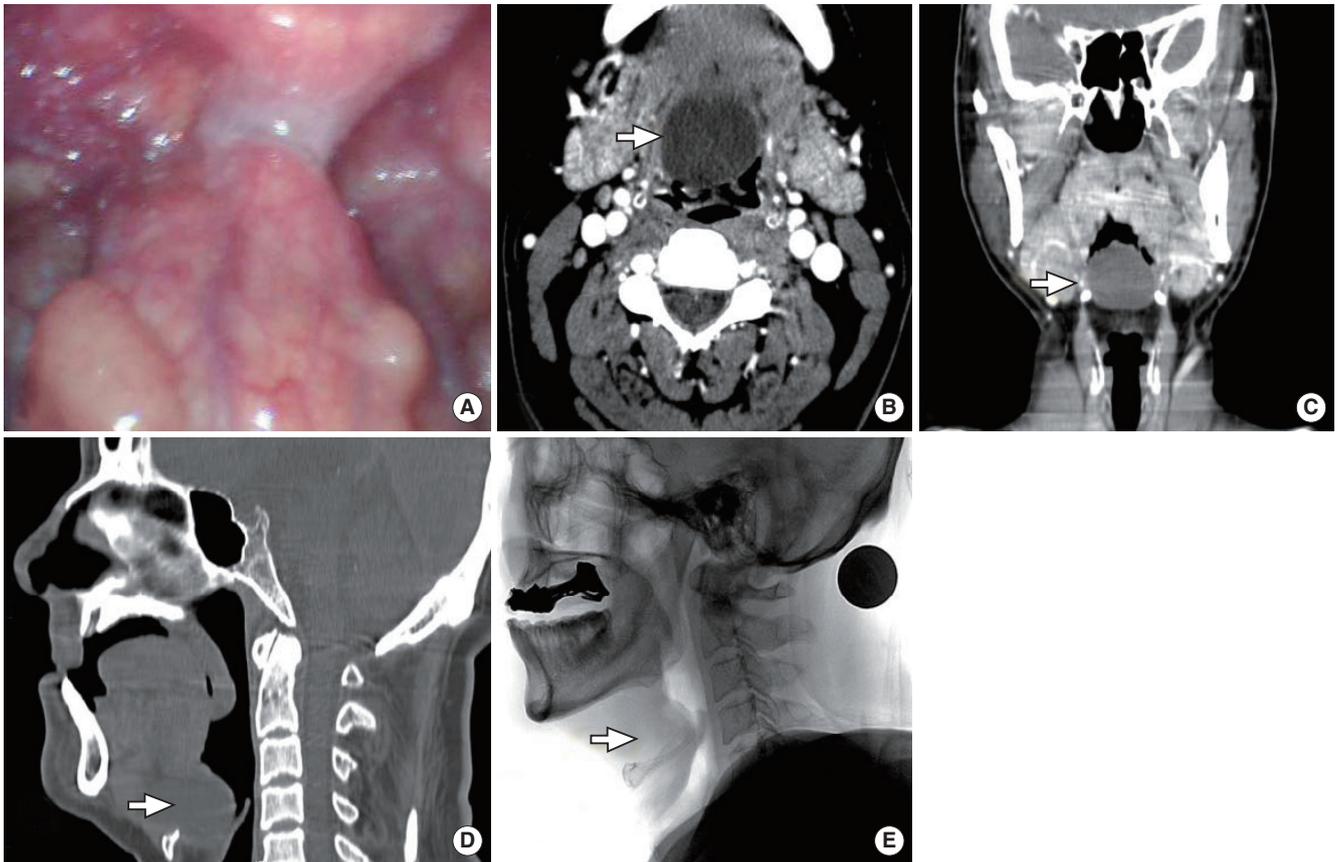


Fig. 1. Preoperative findings of lingual thyroglossal duct cyst. (A) Stroboscopic findings of huge thyroglossal duct cyst (TGDC) in the tongue base, (B) axial, (C) coronal, and (D) sagittal computed tomography scan showed midline cystic mass in the tongue base. (E) Video fluoroscopic image showed a round shadow pushing the epiglottis posteriorly.

ward. The tip of epiglottis was in contact with the posterior pharyngeal wall irrespective of respiratory cycle (Fig. 1E).

Sistrunk operation was performed via external approach instead of oral approach, because the mass was somewhat large. Round cystic mass was removed uneventfully. Soon after the operation, the signs and symptoms of OSA such as snoring and daytime sleepiness ceased and his voice was normalized. His ESS score reached 4.

Postoperative flexible laryngoscopy showed that the mass was disappeared completely (Fig. 2A). Postoperative PSG (3 months after operation) showed that all the parameters were nearly normalized or improved. The RDI decreased to 4.0 events per hour and the supine RDI decreased to 13.4 events per hour. The longest apnea duration was 26.5 sec. The lowest SaO₂ was 92%, and the average SaO₂ saturation was 97.8%.

Postoperative SVF showed that the displaced epiglottis returned to normal position and the pharyngeal airway was not collapsed, however, during inspiration the hypopharyngeal airway was partially obstructed at the level of the epiglottis, which could explain the remnant RDI of 4.0 (Fig. 2B).

The histopathology showed that the cyst was lined by stratified squamous epithelium, and that the deeper tissue showed fi-

brosis, skeletal muscle fragments and laryngeal gland cysts, which was consistent with a TGDC (Fig. 2C). There has been no evidence of recurrence for 1 year.

DISCUSSION

We demonstrated a case of TGDC at the tongue base, resulting in OSA syndrome with SVF findings of dynamic changes in the upper airway.

OSA is a highly prevalent disease that is typified by functional narrowing of the pharynx. The common causes of sleep apnea are bulky or retropositioned soft tissues at the palate, the base of the tongue or the retropharynx. Patients with OSA frequently have multiple anatomic abnormalities and possibly neuromuscular dysfunction causing airway collapse. The less common causes of obstructive sleep apnea include mass lesions of the pharynx, larynx and base of the tongue [3-11]. These include retropharyngeal lipoma, aryepiglottic fold cyst, parapharyngeal tumor such as carotid body tumor, lingual thyroid, supraglottic cyst and epiglottic cyst etc. Theoretically, any mass-like lesions around the upper airway have a potential to cause airway ob-

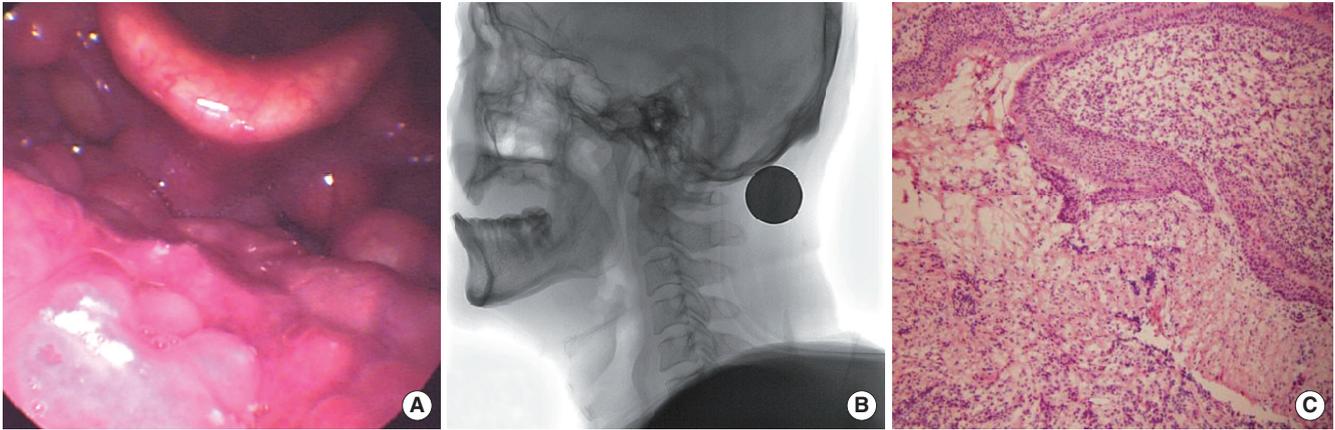


Fig. 2. Postoperative findings. (A) Stroboscopy showed that the cystic mass was removed and (B) video fluoroscopic image showed that the round shadow of cystic mass was disappeared. (C) Histopathology showing the cyst lined by stratified squamous epithelium.

struction, resulting in OSA.

TGDCs are the most common congenital midline neck masses that arise from a tubal remnant of thyroid descent during development [12], however, lingual TGDC, which has an isolated tongue lesion without neck mass, has been rarely reported. The differential diagnosis of this lesion could be a TGDC or a vallecular cyst. When a cystic lesion was buried in the tongue base and close to the hyoid bone or the foramen cecum, a lingual TGDC is more likely than the vallecular cyst. Vallecular cysts are more often attached to the epiglottis or vallecular sulcus with broad base [13]. Pathologic studies cannot differentiate the tongue base cystic lesions, because the epithelial lining of lingual TGDCs and vallecular cysts are quite similar and pathologists tend to rely on the clinical impressions. In our case, the large cystic lesion lied in the midline in close proximity to hyoid bone so that it favors a lingual TGDC more than a vallecular cyst.

There have been some reports on TGDC causing apnea in infants [14-16], however, a TGDC case causing apnea in adult have not been reported, to our knowledge. Most of lingual TGDC cases were pediatric patients and there has been few case reports of adult lingual TGDC with dyspnea [17] or with asphyxia [18].

The position and size of the cysts determine the clinical presentation and cystic lesions could be asymptomatic in whole life. These cysts may be found incidentally in adults or they may present as swelling as a result of associated infection. Once the lesion is identified, complete cyst excision is the treatment of choice. The treatment of lingual TGDC includes external surgical resection, endoscopic laser-assisted resection or marsupialization.

The use of flexible laryngoscopes allows a thorough examination of the larynx and pharynx, and this can be easily performed in the outpatient setting. Our case points out the necessity of a full upper airway examination, including endoscopic and radiological evaluation for patients who present with symptoms of OSA.

SVF is a valuable tool for the evaluation of the upper airway obstruction and was first introduced in 1981 with the name of

somnofluorography [19]. It has been recently introduced to evaluate the changes and obstruction sites of the upper airway in patients with OSA in our hospital [2,20,21]. In this patient, SVF provided dynamic information on the position of the mass in the upper airway, which was displacing the epiglottis posteriorly to the posterior pharyngeal wall, resulting in obstruction of the upper airway. After surgical removal of the mass, SVF showed that the displaced epiglottis returned to natural position and showed the patent airway.

In conclusion, for patients with OSA symptoms, a thorough examination of the upper airway including flexible laryngoscopes is needed and mass lesion such as TGDC should be included in the differential diagnosis. SVF could give us additional information on dynamic changes of the upper airway.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

ACKNOWLEDGMENT

The present research was conducted by the research fund of Dankook University in 2010.

REFERENCES

1. Friedman M, Ibrahim H, Joseph NJ. Staging of obstructive sleep apnea/hypopnea syndrome: a guide to appropriate treatment. *Laryngoscope*. 2004 Mar;114(3):454-9.
2. Lee CH, Mo JH, Seo BS, Kim DY, Yoon IY, Kim JW. Mouth opening during sleep may be a critical predictor of surgical outcome after uvulopalatopharyngoplasty for obstructive sleep apnea. *J Clin Sleep Med*. 2010 Apr;6(2):157-62.

3. Hockstein NG, Anderson TA, Moonis G, Gustafson KS, Mirza N. Retropharyngeal lipoma causing obstructive sleep apnea: case report including five-year follow-up. *Laryngoscope*. 2002 Sep;112(9):1603-5.
4. Picciotti PM, Agostino S, Galla S, Della Marca G, Scarano E. Aryepiglottic fold cyst causing obstructive sleep apnea syndrome. *Sleep Med*. 2006 Jun;7(4):389.
5. Smadi T, Raza MA, Woodson BT, Franco RA. Obstructive sleep apnea caused by carotid body tumor: case report. *J Clin Sleep Med*. 2007 Aug;3(5):517-8.
6. Barnes TW, Olsen KD, Morgenthaler TI. Obstructive lingual thyroid causing sleep apnea: a case report and review of the literature. *Sleep Med*. 2004 Nov;5(6):605-7.
7. Veitch D, Rogers M, Blanshard J. Parapharyngeal mass presenting with sleep apnoea. *J Laryngol Otol*. 1989 Oct;103(10):961-3.
8. Walshe P, Smith D, Coakeley D, Dunne B, Timon C. Sleep apnoea of unusual origin. *J Laryngol Otol*. 2002 Feb;116(2):138-9.
9. Chen MF, Fang TJ, Lee LA, Li HY, Wang CJ, Chen IH. Huge supraglottic cyst causing obstructive sleep apnea in an adult. *Otolaryngol Head Neck Surg*. 2006 Dec;135(6):986-8.
10. Henderson LT, Denny JC 3rd, Teichgraber J. Airway-obstructing epiglottic cyst. *Ann Otol Rhinol Laryngol*. 1985 Sep-Oct;94(5 Pt 1):473-6.
11. Dahm MC, Panning B, Lenarz T. Acute apnea caused by an epiglottic cyst. *Int J Pediatr Otorhinolaryngol*. 1998 Jan;42(3):271-6.
12. Koeller KK, Alamo L, Adair CF, Smirniotopoulos JG. Congenital cystic masses of the neck: radiologic-pathologic correlation. *Radiographics*. 1999 Jan-Feb;19(1):121-46.
13. Suh MW, Sung MW. Apnea spells in an infant with vallecular cyst. *Ann Otol Rhinol Laryngol*. 2004 Sep;113(9):765.
14. Diaz MC, Stormorken A, Christopher NC. A thyroglossal duct cyst causing apnea and cyanosis in a neonate. *Pediatr Emerg Care*. 2005 Jan;21(1):35-7.
15. Eom M, Kim YS. Asphyxiating death due to basal lingual cyst (thyroglossal duct cyst) in two-month-old infant is potentially aggravated after central catheterization with forced positional changes. *Am J Forensic Med Pathol*. 2008 Sep;29(3):251-4.
16. Hanzlick RL. Lingual thyroglossal duct cyst causing death in a four-week-old infant. *J Forensic Sci*. 1984 Jan;29(1):345-8.
17. Wu PY, Friedman M, Huang SC, Lin HC. Radiology quiz case 1: Adult lingual thyroglossal duct cyst (TGDC). *Arch Otolaryngol Head Neck Surg*. 2009 Jul;135(7):716, 718.
18. Sauvageau A, Belley-Cote EP, Racette S. Fatal asphyxia by a thyroglossal duct cyst in an adult. *J Clin Forensic Med*. 2006 Aug-Nov;13(6-8):349-52.
19. Fodor J 3rd, Malott JC, Colley DP, Walsh JK, Kramer M. Somnofluoroscopy. *Radiol Technol*. 1981 Sep-Oct;53(2):105-9.
20. Lee CH, Mo JH, Kim BJ, Kong IG, Yoon IY, Chung S, et al. Evaluation of soft palate changes using sleep videofluoroscopy in patients with obstructive sleep apnea. *Arch Otolaryngol Head Neck Surg*. 2009 Feb;135(2):168-72.
21. Lee CH, Kim JW, Lee HJ, Yun PY, Kim DY, Seo BS, et al. An investigation of upper airway changes associated with mandibular advancement device using sleep videofluoroscopy in patients with obstructive sleep apnea. *Arch Otolaryngol Head Neck Surg*. 2009 Sep;135(9):910-4.