Sleep Medicine 54 (2019) 113–115

Contents lists available at ScienceDirect

Sleep Medicine

journal homepage: www.elsevier.com/locate/sleep

Images in Sleep Medicine

Large posterior lingual thyroglossal duct cyst pneumatically splinted with auto-continuous positive airway pressure at low pressures

Romy Hoque*, Donald L. Bliwise

Emory Sleep Center, Emory University School of Medicine, Atlanta, GA, USA

ARTICLE INFO

Article history:
Received 20 August 2018
Received in revised form
26 September 2018
Accepted 3 October 2018
Available online 19 October 2018

Keywords:
Thyroglossal duct cyst
Auto-CPAP
Pneumatic splint

1. Introduction to the case

The patient is a 58-year-old Iranian American man with long-standing snoring that progressively worsened 4 years ago. Home sleep apnea testing (HSAT) showed an apnea hypopnea index (AHI) of 51.1 events per hour, and patient was started on auto-CPAP with pressure range of 5–20 cm delivered through a nasal mask. Recently while attempting to undergo elective cosmetic facial surgery, his anesthesiologist found his “tongue too large,” and the surgery was cancelled. Subsequent otolaryngology consultation, including computerized tomography of the neck soft tissue revealed a large lingual thyroglossal duct cyst (TDC), which was resected along with central hyoid bone using a combined trans-oral robotic surgery (TORS)/Sistrunk procedure. Surgical specimen histopathology revealed a sub-acutely inflamed cyst lined by respiratory and squamous epithelium abutting resected hyoid bone, histologically consistent with a TDC. Patient denied pre-surgical neck mass, throat pain or soreness, voice hoarseness, or cutaneous fistula; he did however endorse gradually worsening dysphagia with globus sensation.

2. Image analysis

Computerized tomography of the soft tissue of the neck with intravenous contrast revealed a 5.8 cm anterior-posterior diameter, by 5.1 transverse diameter, by 4.4 cm cranial-caudal diameter, homogenous, hypo-dense round/oval base of the tongue lesion, slightly to the right of midline, displacing the epiglottic vallecula posteriorly, and intimately associated with the hyoid bone, consistent with a large TDC (Fig. 1). No internal nodularity, or suspicious enhancement was noted.

Auto-continuous positive airway pressure (auto-CPAP) download for 50 days prior to TDC surgical resection showed an average AHI of 3.6 events per hour, with auto-CPAP mean pressure 7.4 cm H_2O, and average device pressure $\leq$90% of usage time 9.3 cm H_2O (Fig. 2). HSAT after TDC surgical resection revealed an AHI of 10.5 events per hour.

3. Discussion

TDCs originate from persistent epithelial remnants of the thyroglossal duct (TD) that are present during the descent of the thyroid gland from the foramen cecum at the base of the tongue to its final position in the anterior neck. The TD usually atrophies and disappears between the 5th and 10th gestational week; with the potential for TDC development occurring when the TD does not undergo complete involution. Though the most common age of presentation is the first decade of life, there is similar incidence of TDC in children and adults.

The TDC can be situated anywhere along the developmental tract from the tongue base to the suprasternal area. Potential vertical locations of TDCs are: lingual, suprahyoid (including submental), thyrohyoid (between the hyoid bone and thyroid cartilage), or suprasternal. Our patient's TDC was large and extended from the tongue base caudually towards the hyoid bone. Lingual is the least common location for a TDC, given that involution of the TD starts from the cranial end.

Midline neck mass, which moves upward and downward with swallowing given proximity to the hyoid; and throat infection are the two most common presenting symptoms of a TDC. In adults TDCs may present insidiously with additional complaints, such as throat pain or soreness, voice hoarseness, dysphagia, globus sensation, and/or cutaneous fistula formation. Our patient had no

* Corresponding author. Emory Sleep Center, 12 Executive Park Drive, NE, Fourth Floor, Atlanta, 30329, GA, USA.
E-mail address: romy.hoque@gmail.com (R. Hoque).

https://doi.org/10.1016/j.sleep.2018.10.005

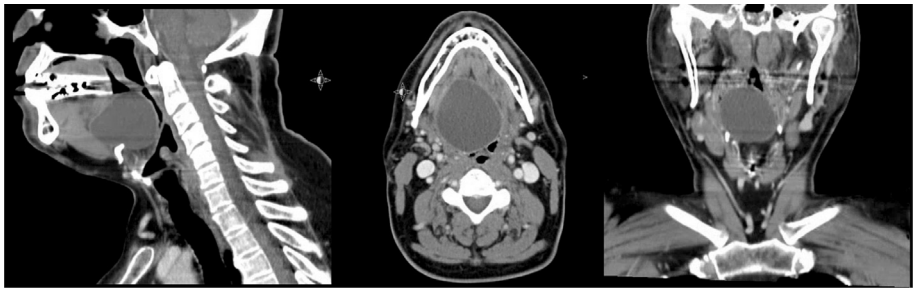

Fig. 1. Computerized tomography of the soft tissue of the neck revealed a 5.8 cm anterior-posterior diameter, by 5.1 transverse diameter, by 4.4 cm cranial-caudal diameter, homogenous, hypo-dense round/oval base of the tongue lesion, slightly to the right of midline, displacing the epiglottic vallecula posteriorly, consistent with a large lingual thyroglossal duct cyst.

palpable neck mass on pre-operative physical examination, given the posterior location of the large lingual TDC.

In adults TDCs are generally a benign condition that does not cause serious health problems, with an insidious presentation compared to children and infants. Nevertheless, on rare occasions, these cysts, particularly lingual TDCs, by virtue of their posterior position and size, may produce significant airway obstruction in both children and adults; and in tragic cases may result in sudden infant death, or fatal asphyxia in adults by valve effect on the epiglottis, leading to trachea closure by a pressed epiglottis [1,2].

To our knowledge this is the first case of TDC pneumatically splinted by auto-CPAP. The 50-day auto-CPAP average AHI, prior to

Fig. 2. Download from patients Philips Respironics auto-continuous positive airway pressure (auto-CPAP) device. Auto-CPAP was delivered through a nasal mask, with pressure range from 5 to 20 cm H_2O. Over the 50-day range, prior to thyroglossal duct cyst surgical resection, patient used the device 96% of days, with percentage of days with usage $\geq$4 h at 86%. Auto-CPAP mean pressure was 7.4 cm H_2O, and average device pressure $\leq$90% of usage time 9.3 cm H_2O. At applied pressures the obstructive airway index was 1.1 events per hour of sleep, and the average apnea hypopnea index was 3.6 events/hour. Response to proprietary device square wave pressure pulse of 2 cm H_2O revealed an obstructed airway index of only 1.1 events/hour, further indication of adequate airway patency with auto-CPAP.

TDC surgical resection, of 3.6 events per hour, being less than <10 events per hour is highly predictive of manually scored polysomography AHI <10 events per hour [3]. The relatively low amount of pressure required to maintain airway patency, may be in part due to the compressible fluid filled nature of the TDC. In a single rare reported case fixed pressure nasal CPAP enlarged a prior TDC, resulting in painless anterior neck swelling [4]. Our patient had no such issues, as his sleep quality improved with auto-CPAP, and he denied palpable painless neck mass either before of after initiation of auto-CPAP.

Conflict of interest

The authors have no conflicts of interest regarding this manuscript. Patient has signed informed consent.

The ICMJE Uniform Disclosure Form for Potential Conflicts of Interest associated with this article can be viewed by clicking on the following link: https://doi.org/10.1016/j.sleep.2018.10.005.

References

[1] Kanawaku Y, Funayama M, Sakai J, et al. Sudden infant death: lingual thyroglossal duct cyst versus environmental factors. Forensic Sci Int 2006;156(2–3):158–60.
[2] Sauvageau A, Belley-Cote EP, Racette S. Fatal asphyxia by a thyroglossal duct cyst in an adult. J Clin Forensic Med 2006;13(6–8):349–52.
[3] Li QY, Berry RB, Goetting MG, et al. Detection of upper airway status and respiratory events by a current generation positive airway pressure device. Sleep 2015;38(4):597–605.
[4] Poza-Aldea JJ, Morello A, Santamaria J. CPAP cyst. N Engl J Med 1993;328(18):1357.