

Thyroglossal duct cysts and obstructive sleep apnoea: three case reports and review of the literature

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Abstract

Background: Thyroglossal duct cysts and obstructive sleep apnoea are commonly occurring medical conditions which appear to present independently in patients. However, we noted three cases where the thyroglossal duct cysts influenced the development and/or therapy of obstructive sleep apnoea. In this article, these three case studies are presented, as is a study of the association between thyroglossal duct cysts and obstructive sleep apnoea, preceded by a literature review.

Case reports: The patient in the first case study underwent hyoidthyroidpexia for obstructive sleep apnoea, which revealed an unexpected thyroglossal duct cyst. The second patient had previously undergone Sistrunk surgery for the removal of a thyroglossal duct cyst and subsequently presented with obstructive sleep apnoea. Finally, the third patient, who had previously undergone Sistrunk surgery, presented with obstructive sleep apnoea and underwent alternative surgery as hyoidthyroidpexia was no longer possible.

Conclusion: To our knowledge, the association between thyroglossal duct cysts and obstructive sleep apnoea has not been addressed previously. The results indicate that the relationship is much stronger than previously thought, and further research is required to investigate the extent of the association and possible causal relations.

Key words: Thyroglossal Duct Cyst; Obstructive Sleep Apnoea; Child

Introduction

Thyroglossal duct cysts and obstructive sleep apnoea (OSA) are commonly occurring medical disorders, which appear to present independently in patients. However, we noted three cases where the thyroglossal duct cysts influenced the development and/or therapy of OSA. We suspected a causal connection between the two and decided that a more in-depth study could yield important insights. In this article, we set the context for further investigation before presenting the three case studies. A study of the association between thyroglossal duct cysts and OSA was also performed, and a literature review was conducted.

In the context of the following case studies, it is important to understand the pathogenesis and the aetiology of the two disorders in order to comprehend the possible causal relationships between them. Here, we present relevant background information regarding thyroglossal duct cysts and OSA, including the possible treatments.

Thyroglossal duct cysts originate from persistent epithelial remnants of the thyroglossal duct present during the descent of the thyroid gland from the foramen caecum to its final position in the neck. In the seventh week of gestation, the thyroglossal tract obliterates. Epithelial remnants in the tract from the foramen caecum to the thyroid can lead to cyst formation.¹ In 60.0–66.7 per cent of cases, the cyst is localised beneath the hyoid.² The prevalence of thyroglossal duct cysts is 7 per cent.¹ A thyroglossal duct cyst usually presents as an asymptomatic

swelling in childhood (in those under 15 years of age), and one-third present after the second decade of life.² The typical clinical presentation is a painless lump in the midline of the neck; this lump moves upwards and downwards when swallowing due to its close proximity to the hyoid.^{1,2}

Cysts are removed for cosmetic reasons, because of functional complaints or when malignancy is suspected.^{1,2} Differential diagnoses include dermoid cyst, sebaceous cyst, lateral neck cyst, thyroid nodules, lymph nodes, ectopic thyroid gland (1–2 per cent) or malignancy (1 per cent).¹

The Sistrunk procedure has been the ‘gold standard’ for surgical removal of a thyroglossal duct cyst since 1920. Given that the cyst originates from an embryonic remnant of the duct, there is a good chance that after simple cystectomy a new cyst will form from remaining tissue.³ Sistrunk therefore developed a technique in which the whole tract of the thyroglossal duct – from the centre of the hyoid up to the foramen caecum (with a margin of 3–4 mm) – is removed, reducing the recurrence rate to 2–6 per cent.^{2,3}

Obstructive sleep apnoea is a sleep-related breathing disorder caused by repeated partial or repeated complete collapse of the upper airway. Loud snoring is one of the most common complaints. Obstructive sleep apnoea is defined by the presence of excessive daytime sleepiness, or two or more of the following complaints: gasping during sleep, recurrent awakenings, unrefreshing sleep, daytime fatigue or impaired concentration, and the presence of five or more

respiratory events per hour of sleep. Surgery for OSA is indicated in patients who do not tolerate continuous positive airway pressure (CPAP) or oral appliances, or in cases where a surgically correctable upper airway anatomy exists.⁴

The prevalence of OSA in children is 1–4 per cent.⁵ Complaints attributable to OSA are usually reported by worried parents of children over the age of five years. These include: excessive sleepiness, habitual changes, concentration impairment, changes of character and morning headaches. Obstructive sleep apnoea is more often seen among children with neurological disorders and craniofacial dysmorphism.⁶

In the following sections, we present three patients with both OSA and thyroglossal duct cysts. A summary of the three patients is as follows: the first patient underwent hyoidthyroidpexia for OSA, which revealed an unexpected thyroglossal duct cyst; the second patient had previously undergone Sistrunk surgery for the removal of a thyroglossal duct cyst, and subsequently presented with OSA; the third patient, who had previously undergone Sistrunk surgery, presented with OSA and underwent alternative surgery as hyoidthyroidpexia was no longer possible.

Case one

Medical history

A 49-year-old man presented with complaints of hypersomnolence, snoring and gasping for air at night. His body mass index was 33.5 kg/m², his neck circumference was 43 cm and he had a Mallampati score of 2. His apnoea–hypopnoea index was 51.

After multiple failed attempts to lose weight and proven intolerance to CPAP, the patient was re-evaluated and underwent drug-induced sleep endoscopy.⁷ Following sleep endoscopy, we decided to perform multilevel surgery, which consisted of Z-palatoplasty (a variation of uvulopalatopharyngoplasty used in cases where a tonsillectomy has been

performed previously), radiofrequency ablation of the tongue base and hyoidthyroidpexia.

The hyoidthyroidpexia procedure consists of the dissection of the infrahyoid muscles (sternohyoid muscle, omohyoid muscle and thyrohyoid muscle) and a suprahyoid disconnection of the hyoid bone. The hyoid bone is mobilised in the antero-caudal direction and fixated to the thyroid cartilage using permanent sutures. The purpose of this procedure is to create more space at the retrolingual level (Figure 1).⁸

There were no indications necessitating pre-operative imaging for any of the interventions mentioned above. Z-palatoplasty and radiofrequency ablation of the tongue base were performed without any complications. During hyoidthyroidpexia, after the dissection of the infrahyoid muscles, a yellow-white liquid appeared, resembling the content of a (thyroglossal duct) cyst. The cyst, measuring approximately 1.5 cm in length, was not detected pre-operatively, and was not compressing the tongue posteriorly. A Sistrunk procedure was considered during the surgery, but this would have interfered with the planned hyoidthyroidpexia. Therefore, we decided to perform a simple cystectomy and proceed with the hyoidthyroidpexia. The cyst was sent for histological study and the procedure was completed without further complications. Subsequent pathology reports confirmed the diagnosis of a thyroglossal duct cyst.

Discussion

The decision to opt for a simple cystectomy instead of a Sistrunk procedure was made based on the following considerations. Firstly, hyoidthyroidpexia had already been discussed with, and was agreed to, by the patient. To abort this surgery and perform a Sistrunk procedure would require further consultation with the patient, prior to carrying out the procedure, in order to obtain informed consent. Secondly, it was taken into account that a Sistrunk procedure might be required in the future, as the origin of the cyst was unknown.

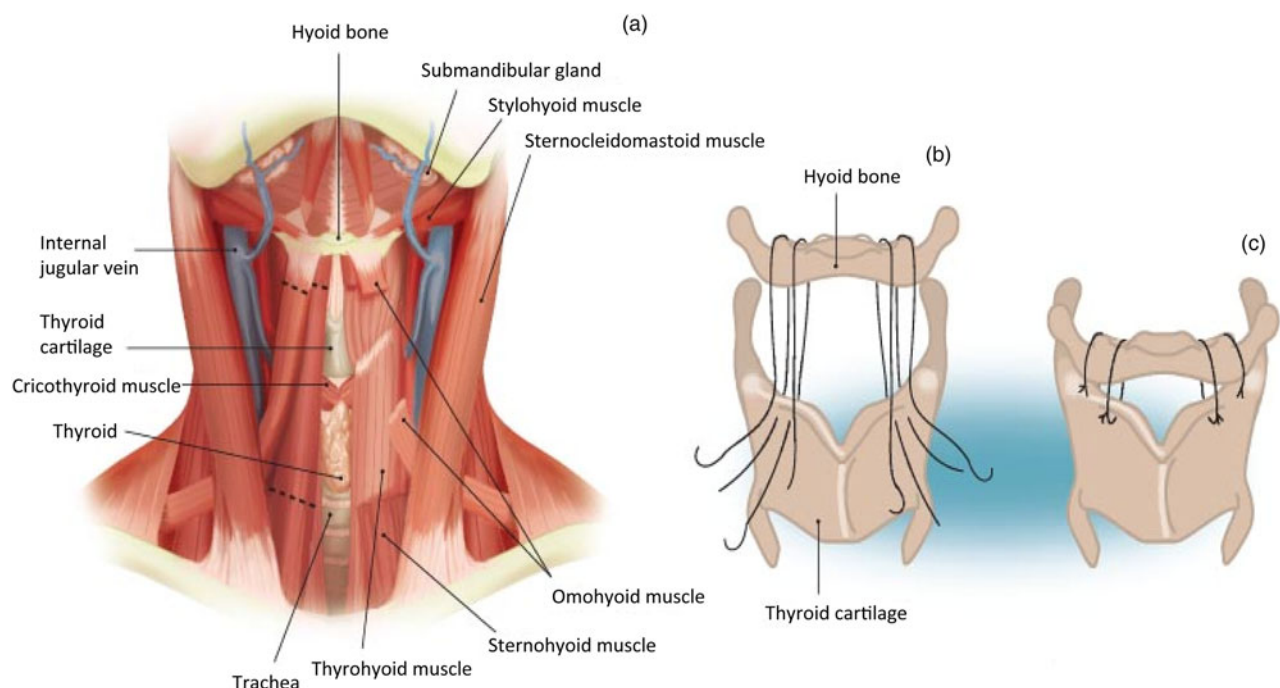


FIG. 1

Illustration of the hyoidthyroidpexia procedure, showing cleaving of muscles (dashed lines in a) and suturing of the hyoid–thyroid gap (b and c). Reproduced with permission.¹¹

In retrospect, it is likely that the unexpected situation that occurred during surgery could have been avoided if pre-operative imaging had been performed. With this course of action, alternatives could have been discussed with the patient beforehand. This prompts the question of whether pre-operative imaging should be performed prior to hyoidthyroidpexia.

Case two

Medical history

A nine-year-old boy was referred to the clinic because of complaints regarding snoring and apnoea. His medical history consisted of adenoidectomy and adenotonsillectomy at the ages of two and three years respectively. At the age of seven years, he underwent three operations for the removal of

a thyroglossal duct cyst, the first being a Sistrunk procedure, with the other two being revisions. After the initial surgery, he suffered recurrent infections in the surgical area. The complaints of snoring and apnoea presented after the first surgery and increased in severity after the second and third revisions. His quality of sleep deteriorated, consisting of several awakenings per night.

The Epworth sleepiness scale score for the patient was 8. A transoral inspection of the tongue revealed bilateral hypertrophic tonsils; further ENT examinations excluded other abnormalities. A polysomnography revealed an apnoea–hypopnoea index of 4.8 (with a normal value at this age being 0.8).⁹ Drug-induced sleep endoscopy showed a complete collapse of the upper airway at the tongue base level (Figure 2) caused by hypertrophy of the lingual tonsils. This was confirmed by a computed tomography scan. The scan also showed cervical lymphadenopathy (Figure 3).

Subsequently, our patient underwent two sessions of transoral radiofrequency ablation of the tongue base within a six-week period, resulting in a significant improvement in his sleep quality. An ENT examination showed a reduction of the lingual tonsil hypertrophy. Three months after the operation, his Epworth sleepiness scale score had reduced to 0 and the polysomnography revealed an apnoea–hypopnoea index of 1.2.

Discussion

The development of OSA caused by reactive hypertrophy of the lingual tonsil after a Sistrunk procedure has not been described in the literature before. Our patient did not have any predisposing factors (i.e. obesity, macroglossia, adenotonsillar hypertrophy or mandibular retrognathia) for OSA.

For our patient, the best explanation for the hypertrophic lingual tonsils seems to be the recurrent infections.¹⁰ An alternative explanation for the OSA occurring after Sistrunk surgery could be the absence of the hyoid corpus. This alone could increase collapsibility of the tongue, in the absence of hypertrophy. However, based on drug-induced sleep endoscopy findings and imaging, this seems unlikely.

- The combination of obstructive sleep apnoea (OSA) and thyroglossal duct cysts is more prevalent than previously thought
- Pre-operative ultrasound should be considered prior to hyoidthyroidpexia to rule out a thyroglossal duct cyst
- A previous Sistrunk procedure is a contraindication for hyoidthyroidpexia as a surgical treatment for OSA

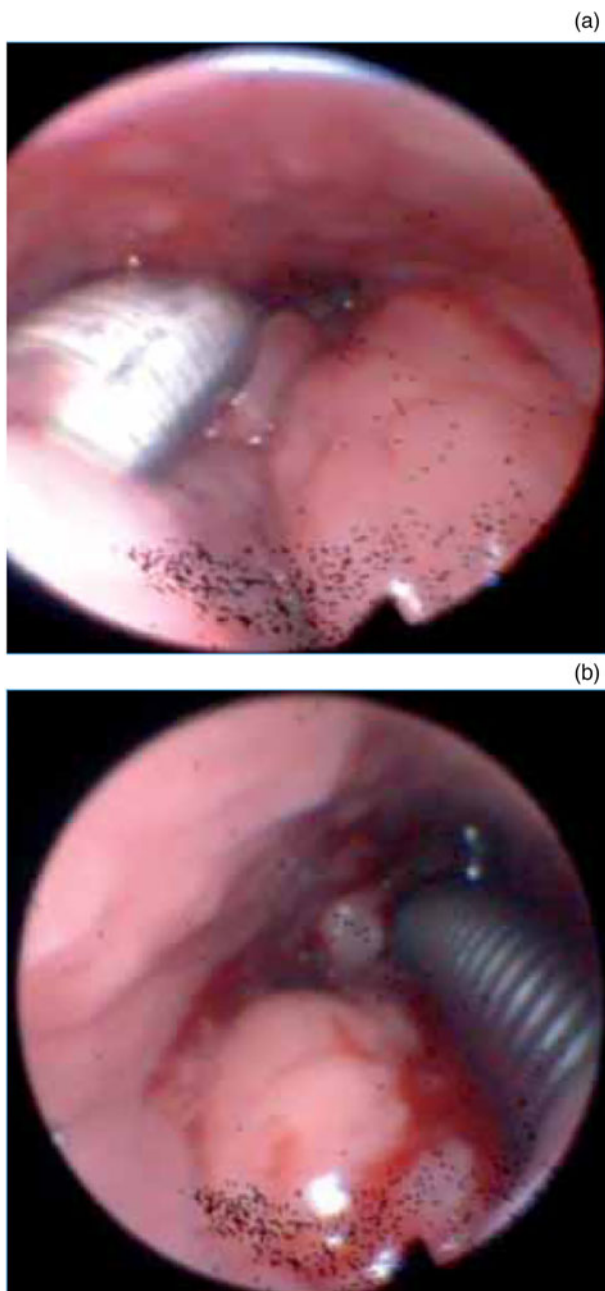


FIG. 2

Endoscopic views of the left (a) and right (b) hypertrophic lingual tonsils.

Case three

Medical history and discussion

In 2006, a 51-year-old man presented with moderate OSA (apnoea–hypopnoea index of 27), with failure of CPAP therapy. His complaints had been present for several years. Drug-induced sleep endoscopy revealed a complete tongue base collapse. Twenty years earlier, the patient had undergone a Sistrunk procedure. Because of the long period between the surgery and the development of OSA, we

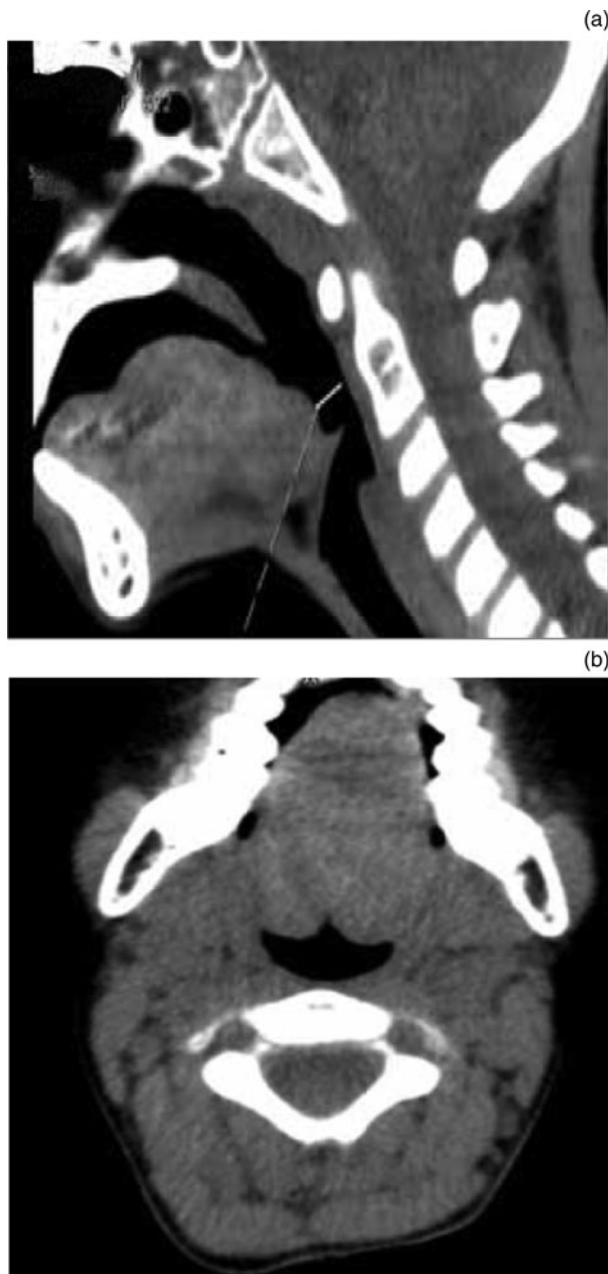


FIG. 3

Sagittal (a) and axial (b) computed tomography images showing hypertrophic lingual tonsils and lymphadenopathy (indicated by the dashed line in a).

assumed that the Sistrunk procedure did not cause the OSA and would not limit the therapeutic options.

Hyoidthyroidpexia with or without radiofrequency ablation of the tongue base is the procedure most often used at our department for the treatment of OSA with complete tongue base collapse.⁸ However, the hyoidthyroidpexia procedure requires an intact corpus of the hyoid. As the patient had already undergone a Sistrunk procedure, hyoidthyroidpexia was not possible.

Apneoplasty, an alternative experimental surgery for treating OSA, was conducted. During this procedure, a magnet was placed in the tongue, with a second attracting or repelling magnet implanted or attached elsewhere. Unfortunately, this procedure did not result in any lasting improvements. Other interventions are presently being considered.

Conclusion

Obstructive sleep apnoea and thyroglossal duct cysts are both common disorders that are not normally associated with each other. However, following a literature search, and considering these three case reports, we can conclude that the combination of OSA and thyroglossal duct cyst is more prevalent than was previously thought. The thyroglossal duct cyst itself does not cause the obstruction of the upper airway leading to OSA. However, thyroglossal duct cyst treatment, including the Sistrunk procedure, can lead to OSA as a result of (possible) reactive hypertrophy of the lingual tonsils associated with recurrent infections in the surgical area. A Sistrunk procedure, which entails removal of the corpus of the hyoid bone and possible subsequent loss of support of the tongue base muscle, could lead to OSA because of a tongue base collapse. However, further research is required to support this theory.

Thyroglossal duct cysts and OSA 'share' the same upper airway anatomy. Therefore, a pre-operative ultrasound should be considered before hyoidthyroidpexia is performed in order to rule out a thyroglossal duct cyst. Because of the close anatomical proximity, a Sistrunk procedure restricts the therapeutic armamentarium in the case of OSA caused by possible tongue base collapse.

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