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Case Report

Unusual dental symptoms in Tornwaldt cyst: a case report

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Abstract Tornwaldt (Thornwaldt) or nasopharyngeal cyst is a cyst occurred at the connection site of pharyngeal endoderm and notochord remnant, usually located in the midline postero-superior wall of nasopharynx. Nasopharynx's obstruction, recurrent infection or trauma may exacerbate mucous secretion by the potential space lining. Although rare and is usually symptomless, it is not uncommon for the patients to have some nasal, ear, cervical or oral symptoms. We report a case of Tornwaldt cyst with rare associated dental symptoms. The patient's initial presentations were dry mouth, cheek biting, post nasal drip and few nasal symptoms including frequent snoring, sleep apnoea, mouth breathing, sneezing, sore throat and itchiness. These symptoms were tremendously and immediately relieved once the cyst was surgically removed.

Keywords: Cheek biting; dry mouth; nasopharyngeal cyst; post nasal drip; Thornwaldt cyst; Tornwaldt cyst.

Introduction

Tornwaldt (Thornwaldt) or nasopharyngeal cyst (TC) is an uncommon developmental benign cyst, usually located in the midline postero-superior wall of nasopharynx. It occurred in the potential space at the site where the notochord remnant connected to pharyngeal endoderm (Fig. 1) (Baisakhiya *et al*., 2011; Voth *et al*., 2011). Reported incidence in general population is 3% (Palacios and Valvassori, 2000). TC developed when the epithelial lining of the potential space secrets mucus following obstruction, recurrent infection or trauma (Moody *et al*., 2007; Voth *et al*., 2011). Although majority of the cases are asymptomatic, patients may present with clinically-significant nasal, ear or cervical symptoms. However, patient presented with dental symptoms has never been reported before. The closest reported oral symptoms were halitosis, foul taste in mouth and difficulty in swallowing (Baisakhiya *et al*., 2011; Voth *et al*., 2011).

Case report

A 38-year-old Malay gentleman was referred by a general dental practice to Oral Medicine Clinic for dry mouth and cheek biting associated with bruxism for three months. Patient claimed that his cheek biting habit was getting worst following swelling of the cheek especially on the right side. In addition, he also complained of snoring problem with frequent awaking at night, recurrent post nasal drip for more than one-year, bilateral nasal congestion, frequent sneezing, recurrent throat itchiness and soreness. Upon oral examination, he was presented with bilateral hyperkeratosis of buccal mucosa consistent with chronic cheek biting, slight generalised right buccal mucosa swelling, mild generalised teeth attrition, and clinically dry mouth. Prominent Fordyce's granules on the lips and bilateral

buccal mucosa was also observed. His condition was slightly improved after using the prescribed upper soft splint which help to protect the buccal mucosa from being continuously traumatised from the cheek biting habit. Used of natural saliva replacement containing sodium monofluorophosphate slightly improved his dry mouth condition.

This patient was then referred to Otorhinolaryngologist for further management of sleep apnoea, sinusitis, postnasal drip with excessive phlegm and bilateral nasal congestion. Cystic lesion at midline posterior nasopharynx was found upon rhinoscopy. White discharge from the swelling noted thus throat cell swab for culture and sensitivity was performed, revealed the presence of Staphylococcus aureus. Surgical removal of the cyst was done, and sample was sent for histopathological examination (HPE). Microscopic findings showed tissue lined with respiratory-type epithelium, composed of heterogenous population of mature lymphoid cells in the underlying stroma, with no sign of dysplasia and malignancy. Diagnosis of Tornwaldt cyst was made based on these clinicopathological findings.

Upon three weeks post-surgical review, patient claimed that his nasal symptoms were tremendously improved, with much less mouth breathing, snoring and post nasal drip problem. Moreover, his dry mouth problem was much subsided. There was no more cheek biting as patient felt that his right flabby cheek was decreased. Three months review revealed that the patient is free of oral and nasal symptoms as experienced before.

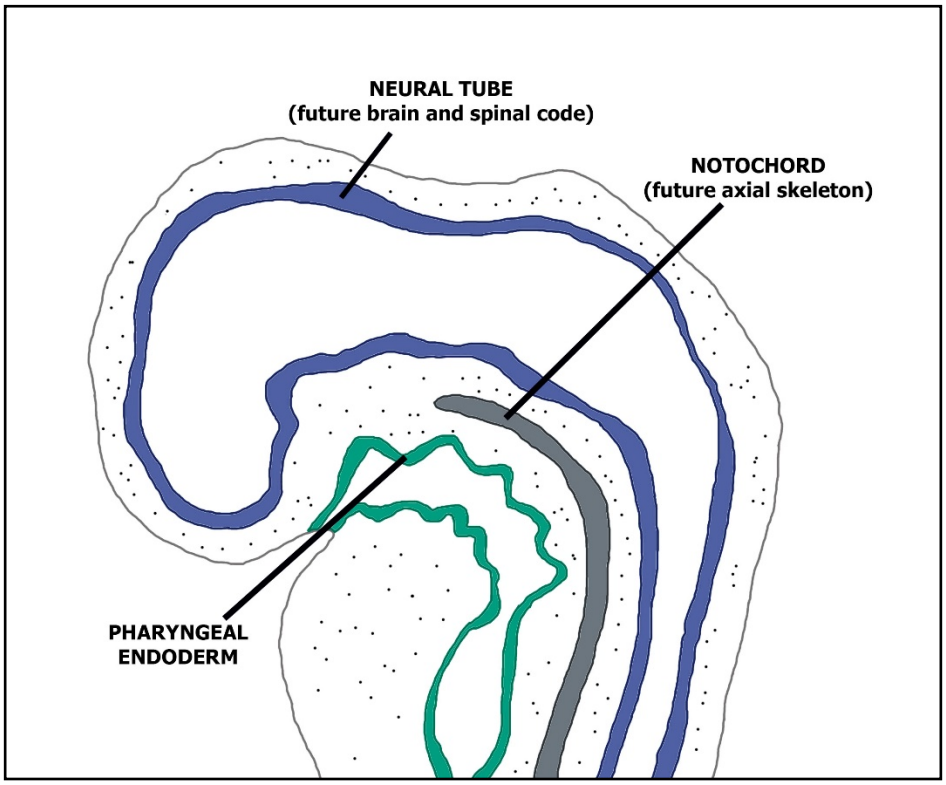

Fig. 1 Schematic diagram of the pharyngeal endoderm, notochord and neural tube position in a developing embryo. If the notochord remnant connected with the pharyngeal endoderm, it will form a potential space for a TC to happen.

Table 1 Summary of TC reported symptoms vs their presence in this case

Symptom categories	Reported symptoms	Symptoms present in this case
Nasal symptoms	Nasal obstruction/congestion	*Yes*
	Recurrent cold	*No*
	Frequent sneezing	*Yes*
	Postnasal drip (clear, or purulent in infected TC)	*Yes* (recurrent, purulent)
	Epistaxis	*No*
		Snoring
		Mouth breathing secondary to nasal congestion
Ear symptoms	Fullness sensation in the ear	*No*
	Tinnitus	*No*
	Eustachian tube dysfunction	*No*
	Otitis media	*No*
Cervical symptoms	Sore throat (pharyngitis)	*Yes*
	Neck pain (cervical myalgia)	*No*
	Neck/cervical stiffness	*No*
		Frequent itchy throat
Neurological symptoms	Giddiness (vertigo)	*No*
	Slurred or slow speech (dysarthria)	*No*
	Difficulty in speaking (dysphonia)	*No*
	Hoarseness (odynophagia)	*No*
	Any kind of all headache (cephalgia) particularly occipital headache	*No*
Oral symptoms	Halitosis (associated with leakage of cyst content)	*No*
	Foul taste in mouth	*No*
	Difficulty in swallowing (dysphagia)	*No*
		Dry mouth (with clinically evidenced xerostomia)
		Cheek biting (with clinically bilateral buccal mucosa hyperkeratosis)
		Teeth grinding habit

Discussion

TC is an uncommon, benign, developmental cyst (Baisakhiya *et al*., 2011; Voth *et al*., 2011). The overall incidence has not been clearly established (Voth *et al*., 2011) but the peak incidence is reported in the second or third decade of life (De Loof and Bachert, 2016). Reported incidence was 1.4 to 3.3% in autopsy specimens (Moody *et al*., 2007; Voth *et al*., 2011), and as low as 0.06% to as high as 6% in radiographic review findings (Moody *et al*., 2007; De Loof and Bachert, 2016).

TC occurs in the midline of the nasopharynx posterior wall, just above the superior pharyngeal constrictor muscle when a potential space existed following persistent communication remnant at the site of where nasopharynx endoderm connected with the notochord remnant during the 6th-to-10th week of developmental (Baisakhiya *et al*., 2011; Voth *et al*., 2011). Following trauma, post-surgery, scarring or inflammation, the epithelial lining of the potential space pouch may become partially or totally obstructed (Moody *et al*., 2007; Voth *et al*., 2011), or underwent damage and changes which eventually leads to mucus secretion, thus resulting of the cyst formation (Baisakhiya *et al*., 2011; Voth *et al*., 2011).

Most TC is relatively small and asymptomatic which usually diagnosed as an incidental finding upon rhinoscopy or imaging (conventional or advanced imaging including computerized tomography (CT) or magnetic resonance imaging (MRI)) and need no treatment (Baisakhiya *et al*., 2011; Voth *et al*., 2011;

De Loof and Bachert, 2016). But large-sized, symptomatic or infected TC may present with nasal, ear, cervical, neurological and oral symptoms (Table 1) (Palacios and Valvassori, 2000; Moody *et al*., 2007; Baisakhiya *et al*., 2011; Voth *et al*., 2011; Chang *et al*., 2015; De Loof and Bachert, 2016). Symptomatic TC may require surgical intervention regardless of the size.

History taking must include questions regarding present of any nasal, ear, cervical, oral or other relevant symptoms. Nasal rhinoscopy usually revealed smooth, pink colour submucosal mass occupying midline posterior nasopharynx region. Ear otoscopy usually revealed signs of Eustachian tube obstruction (Baisakhiya *et al*., 2011). Conventional or advanced radiographic investigations will show present of mass in the nasopharynx area (De Loof and Bachert, 2016). Advanced radiographic is preferable than conventional, whilst MRI is preferable than CT scan (Baisakhiya *et al*., 2011). TC will appear as a well-defined radiographic enhancement mass in MRI, but as a low density well capsulated mass in CT. Histological examination of biopsy sample revealed the TC is lined with ciliated pseudostratified columnar epithelium and has none or very little presence of lymphoid tissue (Baisakhiya *et al*., 2011; Chang *et al*., 2015; De Loof and Bachert, 2016). But significant inflammatory cells infiltration is presented in an infected TC (Chang *et al*., 2015). Examination and biopsy was done for our patient by the otorhinolaryngologist, except performing advance radiographs, were consistent with the findings of a Tornwaldt cyst. Differential diagnosis of TC or midline nasopharyngeal cyst included branchial cleft cyst, Rathke's pouch cyst, adenoid retention cyst, oncocytic cyst, seromucinous cyst, sphenoid sinus mucocele, meningoceles, meningoencephalocele and meningomyelocele (Baisakhiya *et al*., 2011; Voth *et al*., 2011; De Loof and Bachert, 2016).

We reported the unusual oral symptoms of dry mouth and flabby or hyperplastic buccal mucosa with subsequent hyperkeratosis of buccal mucosa, following constant biting habit of the patient. These two symptoms were never reported before. The dry mouth condition was probably secondary to mouth breathing habit resulting from nasal congestion. The hyperplastic mucosa represents a reaction to a kind of irritation or low-grade injury like chewing (Zarei *et al*., 2007). In addition, the patient's dry mouth condition may promote the buccal mucosa to be sucked in towards the tongue region thus interfere with the biting, as seen in many xerostomia patients. Once the cause removed as in this case the TC, nasal congestion improved leads to subsided dry mouth and the flabby mucosa problems.

No treatment is needed in a small and asymptomatic TC. However, in symptomatic TC regardless of the size, surgical intervention is required which options included marsupialization, excision, aspiration or electrocoagulation (Baisakhiya *et al*., 2011; Voth *et al*., 2011). Most patients diagnosed with TC have a very good, promising prognosis once treated. Majority of patients reported improved or totally free and with complete resolution of their nasal obstruction, postnasal drip, ear blockage, headache, halitosis and dysphagia symptoms post-surgery (Moody *et al*., 2007; Baisakhiya *et al*., 2011). However, infected TC may progress to a deep neck infection (retropharyngeal abscess) which could compromise the upper airway and caused life-threatening situation (Chang *et al*., 2015). Surgical excision of the cyst showed a good outcome for this patient in our experience.

Conclusion

We reported this case to create awareness among dental colleagues about the existence of Tornwaldt cyst since this lesion is usually diagnosed and managed by Otorhinolaryngologist. Nevertheless, the mouth can still be the first clinical manifestation site of other medical conditions. Thus, one should always consider medical referral of the patient after ruling out oral causes, which subsequently enable us to diagnose and manage the patient appropriately.

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