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內文：

Introduction

The tonsils form part of a circular band of adenoid tissue known as **Waldeyer's ring**, which guard the opening of the digestive and respiratory tracts.

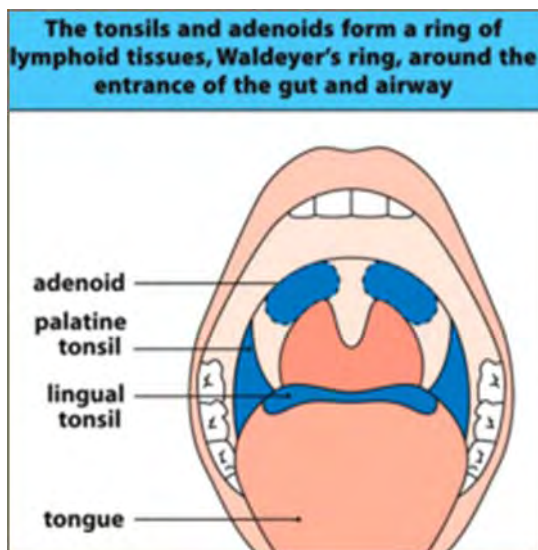
This circular band is comprised of four major tonsil groups: **the palatine, lingual, pharyngeal, and tubal tonsils.**

An **ectopic tonsil** is tonsillar tissue that develops in areas outside of these regions.

The existence of ectopic oral tonsils was described by **Knapp in 1970.**

It was shown that such structures, resembling pharyngeal and other tonsils, can be found within the oral cavity.

Ectopic tonsils have been reported in different anatomic locations of the oral cavity, for example,**on the floor of the mouth, ventral surface of the tongue, and soft palate, and in other parts of the aerodigestive tracts, for example, larynx, hypopharynx, nasal septum, or in the orbit.**



Here we report a rare case of bilateral symmetric ectopic oral tonsillar tissue observed on the ventral surface of the tongue and two other solitary cases arising from floor of the mouth along with a review of the literature.

Case presentations:

Case 1



A **53-year-old Japanese male**, referred by his general dental practitioner, presented with **small, bilaterally symmetric masses on the ventral surface of the tongue, noticed during a routine dental examination 2 months ago.**

The areas affected were **painless** and **remained unchanged in size** over the previous 2 months.

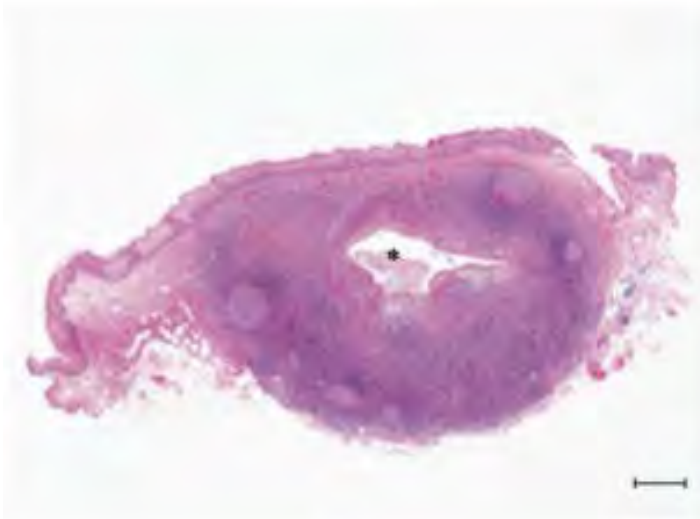
Intraoral examination revealed hard masses of **8 mm diameter (right) and 6 mm diameter (left)** on the ventral surface of the tongue.

The surface covering of these masses was **slightly red** and was **hard on palpation.**

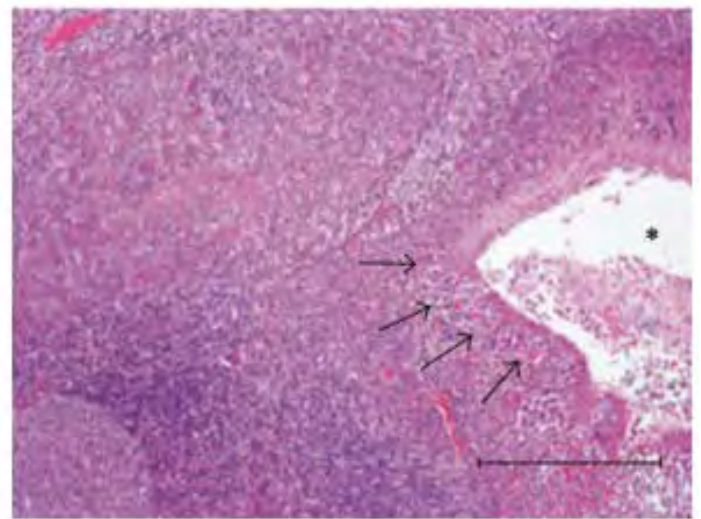
Clinically, **a small pit** was evident at the tip of both masses; a provisional diagnosis of **bilateral benign tumors of salivary origin** was made.

An excision biopsy of the mass on the right side was subsequently performed under local anesthesia.

Histopathological findings showed a **germinal center, lymphoid tissue, and lymphoepithelial symbiosis in the crypt.**



(a)



Case 2

A **63-year-old Japanese female** presented at our hospital with a small swelling **on the left side of the floor of the mouth.**

She first noticed this lump **10 days** previously. The affected area was **painless** and its **size remained unchanged.**



Intraoral examination revealed a **well-circumscribed mass (5 mm diameter) on the left side of the floor of the mouth.**

TABLE 2: Clinicopathological characteristics of three cases of ectopic tonsils.

	Case number		
	1	2	3
Gender	Male	Female	Female
Age	53	63	38
Localization	Ventral surface of the tongue	Floor of the mouth	Floor of the mouth
Number of lesions	Bilateral	Solitary	Solitary
Lesion size (mm)	8/6	5	6
Color of oral mucosa	Slightly red	Slightly red	Normal
Palpation	Hard	Hard	Soft
Clinical diagnosis	Benign salivary tumor	Benign salivary tumor	Mucocele
Histopathological findings			
Crypt architecture	+	+	+
Encapsulation	+	+	+
Lymphoepithelial symbioses	+	+	+
Lymphoid follicle	+	+	+
Crypt obstruction	-	-	-
Cyst formation	-	-	-

The mass was **slightly red** and **hard on palpation** and was clinically diagnosed as a **benign salivary tumor of the floor of the mouth**.

It was resected under local anesthesia and at excision was found to be **encapsulated** and appeared **fairly close to the sublingual salivary gland**.

Histopathology revealed characteristic features of a **tonsil with a germinal center, a mass of lymphoid tissue, and a crypt with lymphoepithelial symbiosis**. These findings were suggestive of **ectopic tonsillar tissue**.

Case 3

A 38-year-old Japanese female visited our clinic complaining of a **small painless lump on the right side of the floor of the mouth**.



She first noticed this lesion **2 days ago**.

Intraoral examination revealed a **well-circumscribed mass (6 mm diameter) covered by intact normal-appearing mucosa**.

The mass was **soft on palpation** and was clinically diagnosed as a **mucocele of the floor of the mouth**.

It was resected under local anesthesia and at excision it was completely detached from the sublingual salivary gland and Wharton's duct **by its own capsule**.

Pathological characteristics were similar to the earlier described cases.

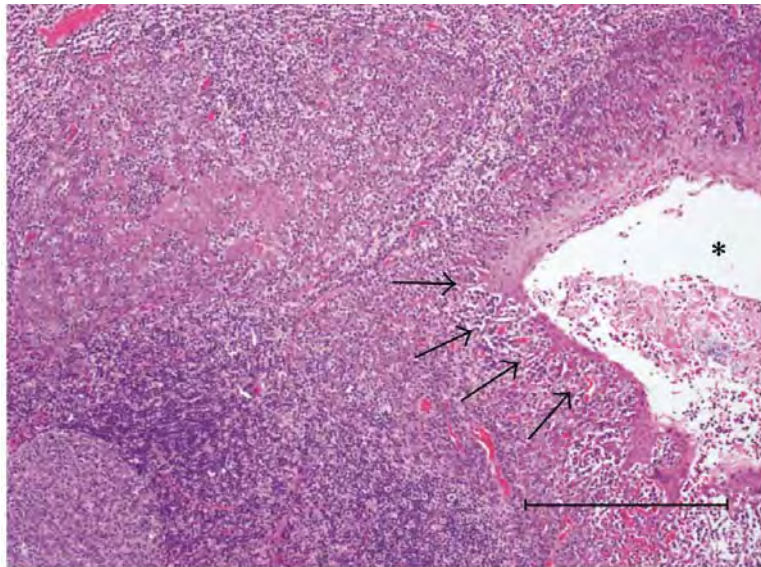
Discussion

Ectopic tonsils are comprised of a **single or branched crypts** containing **lymphoid follicles lined with stratified squamous epithelium**.

In Table 1, we present single cases and case series of ectopic tonsils.

A literature search was conducted in **August 2014** using the electronic databases PubMed and Scopus and hand-searching using the search term of **ectopic tonsil**.

The search was restricted to



Author	Year	Anatomic location	Number of cases	Clinical presentation		Microscopic findings
				Clinical features	Lesion size (mm)	
Kashima et al. [6]	2012	Floor of the mouth	1	Well-circumscribed, smooth, round, painless, swelling covered by intact normal-appearing mucosa	4	(i) Abundant reactive lymphoid aggregates with well-formed germinal centers (ii) A nondilated central crypt lined with stratified squamous epithelium and containing desquamated epithelial cells (iii) Keratin debris in a central lacuna-like space
Present cases (Kimura et al.)	2014	Ventral surface of the tongue	1	Well-circumscribed, slightly red, hard on palpation, and bilateral presentation A small pit was evident at the tip (Case 1)	8/6	Shown in Table 2
		Floor of the mouth	2	Well-circumscribed, slightly red, and hard on palpation (Case 2)	5	
		Soft palate	1	Well-circumscribed and soft on palpation mass covered by normal mucosa (Case 3)	6	
Wolter and Roosenberg [10]	1977	Orbit	1	A smooth surface, an oval shape, and a rubber-like consistency	24 × 15 × 10	(i) Many primary lymphoid nodules with germinal centers
Paslin [5]	1980	Floor of the mouth	1	Oval, pink, lucent, rounded, and firm papule on the sublingual fold just to the right of the frenulum.	3 × 3	(i) Circumscribed masses of lymphoid cells forming germinal centers surrounding the central crypt of stratified squamous epithelium
Pelletiere et al. [7]	1980	Larynx	1	Firm and freely movable and covered by normal appearing, smooth, and intact mucosa	15	(i) Moderately well delineated germinal center
Furukawa et al. [9]	1983	Nasal septum	1	Firm and greyish-white mass	28 × 22 × 14	(i) The surface epithelium of the tumour was fibrous tissue covered with squamous cells which invaginated into the lymphoid tissue producing crypts surrounded by lymphoid follicles
Mogi [3]	1991	Floor of the mouth	1	Small, dark red, and soft tumor with no tender	6 × 3 × 3	(i) A germinal center surrounded by fibrous tissue invaded by squamous epithelium
Patel et al. [4]	2004	Floor of the mouth	1	Three small, red, and circular lesions in the mucosa of the floor of the mouth	3	(i) Aggregation of lymphoid tissue within the lamina propria (ii) Well-defined lymphoid follicles
		Ventral surface of the tongue	1	White, soft, and nontender mucosal nodule of the frenum of the ventral surface of the tongue	4	(i) A focus of lymphoid tissue including follicles with well-formed germinal centers (ii) A cystic lesion lined with stratified squamous epithelium filled with keratinous debris
Baba et al. [8]	2010	Hypopharynx	1	Smooth mucosal swelling in the right pyriform recess	No mention	(i) Germinal center, lymphoid tissue, and crypt involving lymphoepithelial symbiosis

published articles containing **clinicopathological features**.

As a result, the most frequently affected area is the **floor of the mouth (59% of cases)**, followed by the **soft palate (24.6%)** and **ventral surface of the tongue (9.8%)**.

In the clinical findings, the size of the lesions ranged from **3 to 28mm with rounded shape**, and the surface covering of the lesions was occasionally and **slightly red**.

It may be misdiagnosed as tumors that arise from the sublingual gland.

According to Patel et al. inflamed ectopic tonsils may **swell** and **become tender**, thus requiring resection.

Usually, however, ectopic oral tonsils remain **asymptomatic and can be left untreated**, but surgical exploration is indicated to establish a tissue diagnosis.

In Case 1, excisional biopsy of one mass led to a histopathological diagnosis of ectopic tonsillar tissue. Thus, the need for surgical resection of the contralateral lesion was avoided.

The **pathogenesis** of ectopic tonsils in this region remains unclear.

Lymphoid tissue is also found in fetal salivary glands, and occasionally remnants of lymphoid tissue are found in adult salivary glands.

The masses in Cases 2 and 3 appeared close to the **sublingual gland** but were completely separated from the salivary tissues, whereas the masses in Case 1 were placed distant from the salivary tissues, and thus the origins of these masses remained obscure.

It is reported that ectopic tonsillar tissue in the nasal septum may result from **persistent infection**.

Further cadaveric study is required to clarify the presence of ectopic tonsillar tissue on these anatomical sites, particularly with regard to its **developmental mechanisms**, and to assess its **prevalence** and to **study the clinical significance of the immune system and its response**.

Ectopic tonsils appear to occur more frequently than are generally recognized, probably because they are usually asymptomatic and are thus easily overlooked.

題號	題目
1	Where is the most frequently affected area of ectopic tonsils? (A) floor of the mouth (B) soft palate (C) ventral surface of the tongue (D) nasal septum

答 案 (A)	出處：Oral and maxillofacial pathology third edition P185~186
題 號	題 目
2	Which is not the four major tonsil groups in the circular band?
	(A) palatine tonsils (B) lingual tonsils (C) Pharyngeal tonsils (D) laryngeal tonsils
答 案 (D)	出處：Oral and maxillofacial pathology third edition P185~186