Asian Pacific Journal of Cancer Care
Vol 4 No 4 (2019), 145-149
Original Research

Lingual Osseous Choristoma: A Case Report and Review of the Literature

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Introductıon: Osseous choristoma is a benign lesion that is characterized by proliferation of mature osseous tissue in an abnormal anatomical location. The lesion, which was first described by Monserrat in 1913 as “lingual osteoma” has been called “osseous choristoma” since 1971. Here, we present a rare case of osseous choristoma located on the tongue and a review of the literature.
Case Report: A 26 year-old male patient was referred to the Department of ENT with the complaint of swelling of the tongue. Physical examination showed a nodular, sessile lesion on the base of the tongue and the lesion was removed. Histopathological examination of the material revealed a well-defined, submucosal lesion consisting of mature bone tissue and was reported as “osseous choristoma”.
Conclusıon: Osseous choristoma is a benign lesion that occurs mostly in women in their second or third decades of life, although it can be seen in a wide age range. Osseous choristoma may occur in different locations of the oral cavity and maxillofacial region such as the tongue, buccal mucosa, alveolar mucosa, submandibular region, submental region, masseter muscle and palate. This rare entity should be kept in mind because it may be confused with other benign lesions of the tongue like hemangioma, lymphangioma, hamartoma and malignant tumors.

Introduction

Choristoma is defined as the proliferation of normal tissue in an abnormal anatomical location. Although choristomas are usually asymptomatic, it is important they recognize them as they may clinically mimic neoplasms when they give symptoms. Choristomas located in the oral cavity may consist of various tissues such as bone, cartilage, glial tissue, gastric mucosa and sebaceous glands [1].

Osseous choristoma is a benign lesion that develops with the proliferation of mature bone tissue in an abnormal region in which bone tissue is not normally found. This entity was first defined in 1913 by Monserrat as “lingual osteoma”. The term “osseous choristoma” was used by Krolls et al. in 1971 for the first time [2][3].

Case Report

A 26-year-old male patient was referred to the XXX Department of Otolaryngology in May 2018 with the complaint of swelling on his tongue.

Physical examination revealed a nodular lesion at the tongue base and endoscopy was planned. The endoscopic study showed a sessile, nodular lesion that was located on the midline of the posterior region of the tongue. It was considered to be hypertrophy of the circumvallate papilla. Medical treatment and follow-up were recommended. After one month, no regression was seen and the

patient underwent excisional biopsy.

The surgical specimen was 0,6x0,5x0,3 cm sized, gray-white, firm nodular lesion. The entire material was sampled after decalcification process.

Histopathological examination revealed a submucosal, well-demarcated nodular lesion consisting of osseous tissue beneath the non-keratinizing squamous epithelium (Figure. 1).

Figure 1. Submucosal Nodular Lesion Consisting Mature Bone (H&Ex40).

The bone tissue showed lamellation with well-developed Haversian system. Osteoblastic, osteoclastic activity, mitosis or cytological atypia was not seen (Figure. 2).

Figure 2. A. Non-keratinizing Squamous Epithelium on the Surface of Osseous Choristoma (H&Ex200). B. Lamellar mature bone showing no atypia or mitosis (H&Ex400).

The histological findings were consistent with the diagnosis of lingual osseous choristoma. After six months of follow-up, there is no evidence of recurrence.

Discussion

Discussion and Review of the Literature

We searched the literature in PUBMED, MEDLINE, Google Scholar and XXXXX Library database using the keywords "lingual", "osseous", "choristoma", "tongue", "lesion" and their combinations. We also examined the content and bibliography of all the available publications manually. Only the cases with the histopathological diagnosis of “lingual osseous choristoma” were included. The diagnosis of “osteoma” were excluded from the review. Our review revealed 77 lingual osseous choristoma cases reported between 1971 – 2017 (Table 1)[1-53].

Author/Year	No. of cases	Age/Sex			Symptom	Size (mm)	Localization
Present Case	1	26/M			Lump	6	Posterior
Yoshimura/2017 10	1	7/M			None	6	Posterior
Heinz/2017 2	1	21/F			Lump	5	Posterior
Rezende/2017 13		9/F			None	3	Posterior
Turan/2016 9	1	41/F			Lump	10	Posterior
Tran/2016 14	1	30/F			Lump, Gagging	5	Posterior
Adhikari/2016 12	2	15/F			Lump	5	Posterior
		21/F			Pain	5	Posterior
Davidson/2016 15	1	11/M			Lump	-	Posterior
Ginat/2016 16	1	33/F			None	-	Posterior
Valle /2015 6	1	21/F			None	8	Posterior
Saniasiaya /2015 17	1	25/F			None	10	Posterior
Tachasuttirut /201518	1	27/M			None	9	Posterior

Kaplan/2015 19	1	44/F			-	-	Posterior
Stanford /2015 20	1	11/M			None	11	Posterior
Gorini/2014 7	1	10/F			Lump	10	Posterior
Yamamoto/20 14 8	1	11/M			Lump	8	Posterior
Lin /2013 21	1	15/M			Lump	5	Posterior
Chen/2012 22	1	57/F			None	10	Posterior
Toda/2012 23	1	20/F			-	-	Posterior
Kobori/2011 24	1	37/F			None	8	Posterior
Hironaka/201 0 25	1	25/F			None	-	Posterior
Naik/2009 26	1	25/F			Lump	12	Posterior
Andressakis/2 008 1	1	72/M			Pain, Lump, Disphagia	15	Posterior
Hibi/2007 27	1	32/F			None	5	Posterior
Benamer/2007 28	1	14/F			Lump, Gagging	10	Posterior
Velez/2003 29	1	-			-	-	Lateral
Horn/2001 30	1	11/F			None	-	Posterior
Piattelli/2000 31	1	64/F			None	8	Floor
Supiyaphun/2 000 32	3	-			-	-	Posterior
Kim/1999 33	1	17/F			-	-	Posterior
Lin/1998 4	1	21/F			Lump	12	Posterior
Supiyaphun/1 998 34	8	28/F			Irritation	10	Posterior
		25/F			Lump	7	Posterior
		9/F			None	7	Posterior
		35/F			None	7	Posterior
		27/F			None	12	Posterior
		21/F			Lump	15	Posterior
		22/M			None	9	Posterior
		19/F			None	11	Posterior
Vered/1998 35	2	44/M			Gagging, Nausea, Disphagia	7	Lateral
		27/M			Pain, Gagging	10	Posterior
Horie/1998 36	1	25/F			Disphagia	2	Posterior
Pineau/1997 37	1	-			None	-	-
Nakanishi/199 6 38	1	7/F			-	5	Posterior
Manganaro/19 96 39	1	-			-	-	-
Ngeow/1996 40	1	23/F			Lump	15	Posterior
Takahashi/199 5 41	1	9/F			Lump	5	Posterior
Wang/1993 42	1	30/F			-	-	Posterior
Nozoe/1993 43	1	11/F			-	8	Posterior
Ishikawa/1993 44	2	53/F			Lump	8	Posterior

		18/F			Lump	9	Middle
Machino/1990 45	1	14/F			-	-	Posterior
Shintani/1990 46	1	23/F			-	8	Posterior
Mizukami/1988 47	1	28/F			-	5	Middle
Tohill/1987 11	1	31/F			None	7	Posterior
Ioroi/1986 48	1	14/F			-	-	Posterior
Cabbabe/1986 5	1	5/F			None	-	Posterior
Weitzner /1986 49	3	-			None	-	-
Shimono/1984 50	2	47/F			Lump	16	Posterior
		37/F			Lump	15	Posterior
Azuma/1984 51	1	27/M			None	10	Posterior
Sheridan /1984 52	1	20/F			Lump	10	Middle
McClendon /1975 53	2	15/F			None	14	Posterior
		20/M			None	10	Posterior
Krolls/1971 3	8	22/F			None	7	Posterior
		23/M			-	-	Posterior
		73/M			Gagging	-	Posterior
		9/F			Gagging	-	Posterior
		25/F			None	5	Posterior
		11/F			None	20	Posterior
		23/M			None	5	Middle
		39/M			None	6	Middle

Table 1. Review of the literature.

Lingual osseous choristomas can occur at any age but the majority of the patients are in their second or third decades of life [4]. Among the cases reported in the literature, the youngest patient was 5 and the oldest was 73 (mean age: 25,2+14,8) [3][5]. There is a strong female predilection (F/M: 50/17). Our case is a 26-year-old male patient.

Osseous choristomas of the oral cavity and maxillofacial region are most frequently localized in the tongue. 88,2% of the cases, the lesion was located in the posterior one-third of the tongue, 7,4% in the middle one-third of the tongue, 2,9% in tongue lateral borders and 1,5% on the floor of the tongue. In addition, buccal mucosa, buccal vestibule, alveolar mucosa, submandibular region, submental region, retromolar region, masseter muscle and palate localization can be seen [6].

The etiopathogenesis of lingual osseous choristoma is not fully understood. There are two theories widely recognized to explain the development of osseous choristomas. These are the “embryological” and “post-traumatic” (reactive) theory. According to the embryological theory, the lesion develops from the pluripotent cells in the 1st and 3rd branchial arches. The post-traumatic theory is based on the fact that posterior one-third of the tongue is the most common site of traumatic irritation of the oral cavity. The theory suggests that local inflammation and calcium deposition in the trauma site give rise to the lesion [7]. In our case, the lesion consisted of mature bone tissue with fully developed Haversian system and not just calcification. Also, there was no evidence of local inflammation in the surrounding tissue. We believe that post-traumatic theory alone is insufficient to explain the etiopathogenesis of lingual osseous choristoma.

Osseous choristoma of the tongue is mostly asymptomatic (46%). In symptomatic cases, lump (31,7%) was the most common complaint. Additionally, gagging (9,5%), dysphagia (4,8%), pain (4,8%), nausea (1,6%) and throat irritation (1,6%) were the other symptoms reported [8].

Grossly, osseous choristomas are usually under one cm in size, pedunculated or sessile, well-demarcated, firm, smooth surfaced, gray-white, pink colored nodular lesions [9]. 67,2% of the osseous choristomas reported in the literature are pedunculated and 32,8% are sessile. The size of the lesion ranged from 2 to 20 mm and the mean size was 8,7+3,6 mm. In our case, the patient was complaining of a lump and physical examination revealed a six mm sessile lesion.

Histopathologically, lingual osseous choristoma is submucosally located and consist of well-developed lamellar bone and Haversian system. Osteoblastic and osteoclastic activity, cytological atypia or mitosis are not observed in the bone tissue [10].

Differential diagnosis of lingual osseous choristomas should be made according to where the lesion is located on the tongue. For a lesion that is situated on posterior one-third of the tongue, lingual thyroid, thyroglossal ductus cyst, mucocele, pyogenic granuloma, hemangioma, lymphangioma, hamartoma, salivary gland tumors, and sarcomas should be included in the differential diagnosis. Anterior and lateral localization should be differentiated from fibroma, granular cell tumor, and neural origin tumors. Salivary gland tumors, mucus retention cysts, neural tumors, and lipomas are differential diagnosis with osseous choristomas located under the tongue [11].

The treatment of osseous choristoma is the total excision of the mass. Recurrence after excision is not expected. In the literature, recurrence of two cases of osseous choristoma has been reported [12].

In conclusion, lingual osseous choristoma is a rare benign lesion. This lesion, which can be confused with benign and malignant tumors in the oral cavity and maxillofacial region, should be considered when a clinicopathological diagnosis is made. In this review, available cases of lingual osseous choristomas were gathered and our case is presented with the literature.

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