Received: November 9, 2023
Accepted: November 26, 2023

Acad Med J 2023;3(3):104-107
UDC: 616.313-006.03
DOI: https://www.doi.org/10.53582/AMJ2333104p
Case report

OSSEOUS CHORISTOMA OF THE TONGUE: A CASE REPORT

Panchevski Goran, Iliev Aleksandar, Kirkov Antonio, Dvojakovska Suzana, Stamatoski Aleksandar, Koneski Filip

University Clinic for Maxillofacial Surgery, Skopje, Faculty of Dentistry,
Ss. Cyril and Methodius University, Skopje, Republic of North Macedonia
e-mail:

Abstract

Osseous choristoma of the tongue is a rare condition characterized by the appearance of exophytic mature bone tissue in an ectopic site, mostly on the dorsal surface of the tongue. In this study we present a case with lingual osseous choristoma in an 11-year-old girl, treated surgically under local anesthesia. After the treatment, the patient was symptom free, and follow-up showed no recurrence.

Keywords: osseous choristoma, ectopic, osteoma, tongue

Introduction

Choristomas are defined as tumor-like conditions or growth of normal, matured tissues, located in regions in which they are not normally found. Oral choristomas are rare and most often derived from bone and cartilage. The first report on osseous formation of dorsal surface of the tongue was by Monserrat in 1913[1,2], and the term intraoral and lingual osseous choristoma was introduced in 1971 by Krolls *et al*. reporting nine cases[1,3]. Since then, about 100 cases of lingual osseous choristoma have been documented and published. Arimoto S. *et al.* reported that lingual osseous choristoma in pediatric patients below the age of twelve is extremely rare, consisting of seventeen published cases so far[4]. It presents as a well circumscribed lump consisting of lamellar bone covered with squamous epithelium. Even though few theories are widely accepted for pathogenesis, the etiology of lingual osseous choristoma is still unknown. While the exact etiopathogenesis remains debatable, we are presenting a case of this rare entity.

Case description

An 11-year-old girl was referred to the Clinic for Maxillofacial Surgery complaining on a lump, located in the posterior 1/3 of the tongue in front of *papillae circumvallate* on the left side, with no symptoms, except occasional swallowing discomfort felt lately (Figure 1).

Her parents reported that she had noticed the asymptomatic node more than three years ago. Physical examination revealed adherent sessile mass of the dorsal aspect of the tongue covered with smooth mucosa, measuring 7-8 mm. It was firm on palpation, painless, suggesting an initial diagnosis of a benign formation. Surgical excision, actually excisional biopsy, was performed under local anesthesia and a solid circumscribed tumor was removed.

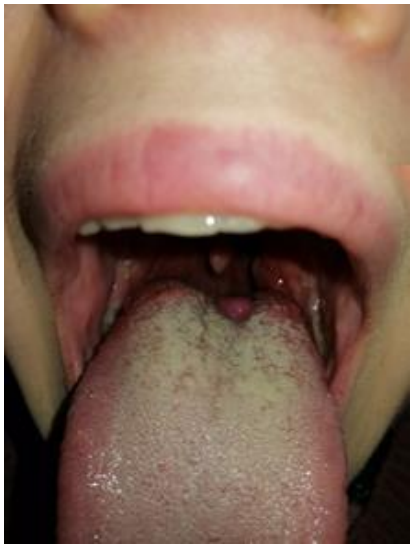

Fig. 1. Clinical appearance of tumor
of the posterior dorsum of the tongue

The specimen was histologically analyzed. The report described a well circumscribed nodule measuring 7 x 5 x 3 mm, composed of cortical lamellar bone, Haversian canals, osteocytes, peripheral fibrous tissue lined with normal squamous epithelium (Figure 2).

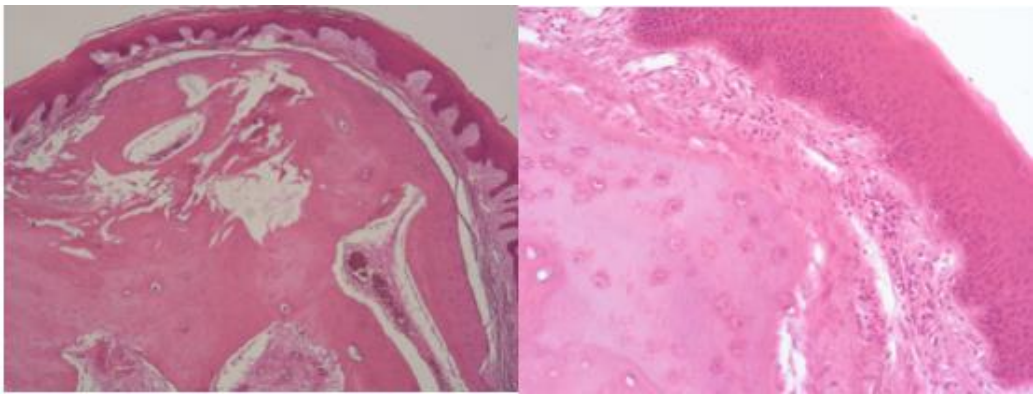

Fig. 2. Histological examination: mature lamellar bone with Haversian system, osteocytes, stratified squamous epithelium (H&E, magnification 50X, 200X)

Diagnosis revealed osseous choristoma. Postoperatively, after 6 months and 3 years follow-up, the patient was free of symptoms and showed no recurrence.

Discussion

With less than 100 reported cases, lingual osseous choristoma is considered a rare pathology. Analyzing the age of patients, it ranges from 5 to 89 years, mostly affecting young patients in the second and third decade of life[4,5,6]. This lesion has female predilection with a ratio of almost 2.7:1, and in 2/3 of cases it is located in the posterior tongue region of the dorsum anterior to *papillae circumvallate* and *foramen cecum*. Literature reports cases with osseous choristomas located in the lateral edge, medium third of the tongue, anterior third and base of the tongue, according to frequency in that order[3,4]. Dimensions can vary from 3 mm to 50 mm in the largest diameter. Lingual choristomas are pedunculated or sessile, covered with almost normal epithelium, whitish, firm, mostly asymptomatic. In symptomatic cases, authors reported foreign body sensation and discomfort (68%), dysphagia (28,9%), tongue

swelling (15%) and in some cases gagging and nausea. Symptoms are related to localization and size of choristomas in the oral cavity[1,3,7].

Considering preoperative diagnosis, CT can be used to clear dilemmas in differential diagnosis despite infrequent use of imaging (14%)[7]. Endoscopic examination can contribute to determining the appearance, localization, and dimensions of lumps located on the tongue base and around *foramen cecum*, while the benefit of ultrasound is limited.

Differential diagnosis of lingual osseous choristoma includes benign tumors, hamartomas, pyogenic granuloma, lingual thyroid, malignant tumor and should be based on appearance, consistency, and some of them are more frequent on certain locations[8,9,,10]. Definitive diagnosis is confirmed with histopathological examination, where typical findings include mature bone, Haversian canals, osteocytes, stratified squamous epithelium[7].

There are several most frequently used theories to explain pathogenesis. One is the theory of developmental malformation related to fusion of the first and third branchial arches from which anterior two and posterior third of the tongue originate. The second theory says that traumatic irritation and inflammation as a reactive center of ossification are reasonable to be used for explaining choristomas on the lateral tongue. The third theory suggests that the mass is due to calcification of remnant undescended thyroid tissue, trying to explain the more frequent appearance around *foramen cecum*. Appearance on other locations and presence of mature bone, not calcification, is not in favor of the last two. In our case, the developmental theory is more plausible, because of the missing chronic irritation or trauma. However, pathogenesis is still unclear[3,8].

The treatment of choice is a surgical excision under local or general anesthesia depending on clinical judgement, considering tumor localization and patient age. There are two reported recurrences for osseous choristoma, one of a buccal soft tissue presented by Long (1991) and another one of the masseter muscle described by Dalkiz (2001)[8,7,11]. Until now, there is no reported recurrence or malignant transformation for lingual osseous choristoma.

Conclusion

Lingual osseous choristoma can be treated successfully with surgical excision by oral and maxillofacial or ENT specialists who are familiar with the complex regional anatomy and diverse symptoms of oropharyngeal pathology. When evaluating tongue lesion or growth, surgeons should consider this rare entity in the differential diagnosis.

Conflict of interest statement: None declared.

References

1. Benamer MH, Elmangoush AM. Lingual Osseous Choristoma Case report and review of literature. *Libyan J Med* 2007; 2(1): 46-48. doi: 10.4176/061211.
2. Liu JY, Tan KKH. Lingual osteoma: case report and literature review. *Singapore Med J* 2011; 52(10): e198-e200. PMID: 22009406.
3. Gorini E, Mullace M, Migliorini L, Mevio E. Osseous choristoma of the tongue: a review of etiopathogenesis. *Case Rep Otolaryngol* 2014; 2014: 373104. doi: 10.1155/2014/373104.
4. Arimoto S, Shigeoka M, Akashi M. Tongue Osseous Choristoma in an 11-Year-Old Female: A Case Report and Literature Review Focusing on Pediatric Cases. Case Rep Dent 2021; 2021: 8021362. doi: 10.1155/2021/8021362.
5. Hemmi T, Suzuki J, Sato S, Tabata M, Watanabe K, Sugawara M, *et al*. A Case of an Incidentally Removed Lingual Osseous Choristoma. *Case Rep Otolaryngol* 2020; 2020: 3498915. doi: 10.1155/2020/3498915.

6. Stanford JK 2nd, Spencer JC, Reed JM. Case presentation and images of lingual osseous choristoma in a pediatric patient. *Am J Otolaryngol* 2015; 36(6): 753-754. doi: 10.1016/j.amjoto.2015.07.008.
7. Shareef ZJ, Shareef SJ, Kerndt CC, Aughenbaugh A, Di Ponio A. Lingual Osseous Choristoma: A Comprehensive Systematic Review of Lesion Presentation, Histology, and Morphology. *Spartan Med Res J* 2020; 5(2): 17543. doi: 10.51894/001c.17543.
8. Adhikari BR, Sato J, Morikawa T, Obara-Itoh J, Utsunomiya M, Harada F, *et al*. Osseous choristoma of the tongue: two case reports. *J Med Case Rep* 2016; 10: 59. doi: 10.1186/s13256-016-0840-8.
9. Heinz MJ, Peters SM, Caruana SM, Yoon AJ. Lingual Osseous Choristoma of the Tongue Base: Unusual Presentation of a Rare Entity. *Case Rep Otolaryngol* 2017; 2017: 3234086. doi: 10.1155/2017/3234086.
10. Turan Ş, Pınarbaşlı MÖ, Açıkalın M, Kaya E, Özüdoğru E. Lingual Osseous Choristoma. *Turk Arch Otorhinolaryngol* 2016; 54(2): 86-88. doi: 10.5152/tao.2016.1448.
11. Gkouveris I, Kapranos N, Mitrou GG. Lingual Osseous Choristoma: Case report and literature review of this rare entity. *J Clin Exp Dent* 2023; 15(1): e79-e81. doi: 10.4317/jced.59225.