SAGE Open Medical Case Reports
Volume 12: 1–4

DOI: 10.1177/2050313X241260177
journals.sagepub.com/home/sco
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Lingual osteoma presenting as a solitary painless lesion: Report of a rare case with review of the literature

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Abstract

Lingual osteoma, a rare, benign bone tumor that primarily affects the posterior tongue, can be difficult to diagnose. This study aims to report a case of osteoma affecting the tongue in a 17-year-old female. The patient had a foreign body sensation and a progressively growing lesion for 3 years and underwent clinical examination and diagnostic procedures. A well-defined, smooth-surfaced, white mass was discovered in the posterior third of the tongue. The $1.5 \times 1 \times 0.4$ cm mass was completely excised under local anesthesia and histopathologically confirmed as a benign lingual osteoma. The 2-month post-operative outcome was uneventful. The rarity of lingual osteoma, as well as the fact that it is often asymptomatic, makes diagnosis difficult. The diagnosis entails a proper clinical examination, imaging studies, and histopathological analysis. Surgical intervention, primarily aimed at complete excision while preserving tongue function, remains the primary treatment option. Successful excision entails educating healthcare professionals about this rare benign bony tumor to ensure the best possible patient outcomes.

Keywords

Lingual, surgical excision, tongue, osteoma

Date received: 4 April 2024; accepted: 22 May 2024

Take home message

Lingual osteoma, though rare, can present with symptoms such as a foreign body sensation and progressive growth, requiring proper diagnosis and treatment. Clinical examination, imaging studies, and histopathological analysis are crucial for accurate diagnosis. Surgical excision, while preserving tongue function, is the primary treatment, emphasizing the importance of educating healthcare professionals about this condition for optimal patient care.

Introduction

Osteomas are benign bone tumors composed of mature osseous tissue that infrequently occur in the head and neck region, particularly within the oral cavity. There are three types of osteomas based on the site of origin: central, peripheral, and extra-skeletal osteomas.[1,2] An osteoma of the tongue is an extremely rare condition, with fewer than 100 cases reported.[3] These tumors are often pedunculated and covered by normal mucosa. Approximately 40% of patients exhibit no symptoms, whereas nearly a quarter express the sensation of a lump in the throat.[4] The pathogenesis of lingual osteoma remains obscure.[5] Most commonly affected are individuals between 30 and 40 years old (average age at diagnosis: 28.7 years) with a marked female predilection.[6] While a combination of physical examination and imaging modalities is necessary for the initial workup, the definitive

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diagnosis is ultimately dependent on the histological examination of the excised tissue. The dependence on post-operative confirmation emphasizes difficulties in pre-surgical differentiation from other lingual lesions, especially considering the tongue's anatomy and functions. As a result, surgical excision is frequently used as the primary treatment option despite its potential difficulty.[7]

The current study aims to present a case of lingual osteoma recorded in a 17-year-old female patient.

Case presentation

Patient information

A 17-year-old female patient presented with a complaint of a posterior tongue lesion. Remarkably, she experienced no associated symptoms such as pain, bleeding, dysphagia, voice change, dyspnea, or difficulty swallowing. Her only concern was a foreign body sensation and the increasing size of the lesion over the past 3 years. Her past medical and surgical history was unremarkable, including a negative family history of malignancy and an absence of smoking habits.

Clinical finding

Clinical examination revealed a single well-defined, smooth-surfaced, white mass, measuring approximately 1×1 cm, on the posterior third of the tongue, visible upon tongue protrusion. Palpation revealed a hard, non-tender mass without any associated cervical lymphadenopathy.

Diagnostic approach

A neck ultrasound revealed no abnormalities. The serum Thyroid Stimulating Hormone (TSH) level was normal.

Therapeutic approach

The surgical procedure began with the excision of the mass under carefully administered local anesthesia, ensuring the patient's comfort throughout. Hemostasis was skillfully achieved without any complications, allowing for a smooth and controlled surgical process. The wound was meticulously sutured using absorbable sutures, promoting optimal healing and minimizing post-operative discomfort. The mass measured $1.5 \times 1 \times 0.4$ cm and was hard to palpate. Histopathological examination (HPE) showed a well-defined, bony mass composed of thick, compact, trabecular mature bone covered by stratified squamous epithelial cells, diagnostic of a benign lingual osteoma (Figure 1).

Follow-up and outcome

The patient was discharged in good health on the first post-operative day. A 6-month follow-up showed no sign of recurrence clinically.

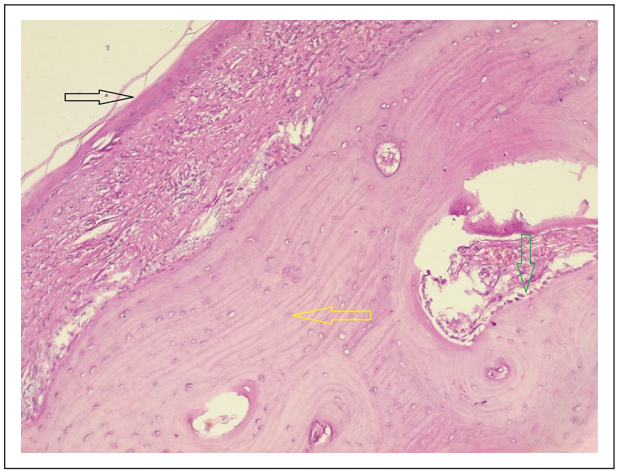

Figure 1. Sections show mucosa covered by benign squamous epithelial cells (dark arrow), with the underlying bone, trabeculae (yellow arrow), and osteoblasts (green arrow).

Discussion

Both malignant and benign lesions of the tongue are rare conditions.[8,9] Lingual osteoma, a form of soft tissue osteoma, is an infrequent tumor-like lesion primarily affecting the posterior part of the tongue.[10] It is characterized by the presence of mature, compact bone with haversian systems.[1] In the head and neck region, these tumors typically originate in the paranasal sinuses, maxilla, and mandible.[11] They were first described by Monserrat in 1913.[6] The reported lesions so far have measured between 3 and 50 mm in their greatest dimensions.[7] The patients range from 5 to 73 years of age at the time of diagnosis,[9] with a female-to-male ratio of 3.25 to 1.00.[1] According to the genuine literature, the pathogenesis of lingual osteoma is still unknown, but three main theories have been outlined.[12] One theory is that post-traumatic reactions, possibly caused by frequent irritation in the posterior third of the tongue (such as swallowing, articulation, or trauma), can cause local inflammation and calcium deposition, resulting in dystrophic calcification. A second theory attributes them to the ossification of undescended lingual thyroid remnants. Lastly, some attribute their occurrence to the ossification of remnants of branchial arches.[11] The current case was a 17-year-old female patient who presented with a posterior tongue lesion.

A significant portion of individuals with this condition remain asymptomatic, contributing to the diagnostic challenges.[7] However, certain patients may exhibit symptoms such as a foreign body sensation in their throat (25.8%), dysphagia (6.9%), gagging (5.1%), nausea (3.4%), and irritation (3.4%).[10] The size, location, and inflammation of the surrounding tissues are correlated to the symptoms.[13] Liu[1] Described a patient with a 0.5-cm, pedunculated, painless mass on the dorsal tongue that was covered by normal mucosa and was not associated with cervical lymphadenopathy. Other cases have presented with a severe vomiting

reflex[14] and respiratory distress caused by airway obstruction.[15] In the current study, the patient presented with a persistent foreign body sensation and observed growth of the lesion over 3 years, while examination revealed a well-defined, hard, painless mass on the posterior third of the tongue, visible upon protrusion.

A biopsy is necessary for a definitive diagnosis.[16] Distinguishing lingual osteoma from an ectopic thyroid gland near the foramen cecum is crucial.[17] Patients with multiple osteomas need to be assessed for Gardener's syndrome.[18] A combination of clinical examination and imaging studies is useful for diagnosis. A clinical examination may reveal a palpable mass in the posterior tongue. Imaging studies, including X-rays and computed tomography (CT) scans, provide detailed information about the mass. A CT scan is superior to magnetic resonance imaging (MRI) for detecting the hallmark calcification and cortical bone since osteoma is primarily composed of mature lamellar bony tissue,[3] and MRI mainly helps to differentiate it from soft tissue mimics. This multifaceted approach ensures correct diagnosis and appropriate management.[4] In cone beam computed tomography (CBCT) imaging, lingual osteomas typically manifest as well-defined, round, or irregular-shaped radiopaque masses within the lingual soft tissues. The utilization of CBCT provides three-dimensional visualization of the lesion, offering valuable information for precise diagnosis and guiding the surgical excision approach.[19]

A comprehensive assessment of the patient's overall health and medical history is paramount for determining the most suitable treatment course. Factors like age, existing medical conditions, and individual treatment preferences should all be considered when developing a personalized care plan. Collaboration with multidisciplinary teams, including oral and maxillofacial surgeons, otolaryngologists, and speech therapists, can be invaluable in addressing the unique challenges posed by lingual illness.[20] Differential diagnosis of lingual neoformation plays an important role in the management of tongue surgery, as many different neoformations may arise, such as schwannoma, osteoma, lipoma, and ossifying lipoma.[21–25] Surgical excision is the gold standard of treatment for osteoma of the tongue. This approach prioritizes complete tumor removal while preserving maximal tongue function and integrity. The size of the osteoma dictates the chosen technique, with per-oral or endoscopic access preferred for smaller lesions and hot instruments like diathermy chosen for larger ones.[11] HPE should reveal mature lamellar bone with osteocytes, cement lines, and haversian systems covered by stratified squamous epithelium.[10] In the present case, the mass was successfully excised under local anesthesia with no complications, and the subsequent HPE confirmed a benign lingual osteoma composed of mucosa benign squamous epithelial cells with underlined bone trabeculate.

Post-operative follow-up involves periodic clinical examinations and, in some cases, radiological studies to monitor for potential recurrence.[26] In this case, the patient recovered well post-surgery, and a 6-month follow-up showed no signs of recurrence clinically.

The main limitation of this study was the lack of pre-, intra-, post-operative picture and radiological imaging of the tongue lesion. This could have aided in formulating a more precise surgical plan. Knowing the exact location and depth of the lesion would allow for targeted access and minimize the risk of damage to surrounding structures, especially vital nerves or blood vessels in the tongue. Despite the lack of imaging, the lesion was completely excised, and the post-operative period was uneventful.

Conclusion

This case report contributes to the existing literature on lingual osteoma by highlighting the significance of early detection, accurate diagnosis, and timely intervention in managing this rare benign bony tumor.

Acknowledgements

Not applicable.

Author contributions

A.M.S. was a major contributor to the conception of the study, as well as to the literature search for related studies. F.H.K. and A.A.Q. were involved in the literature review, study design, and writing the manuscript. S.H.H., Y.A.S., H.M.D., A.S.M., and B.O.H. were involved in the literature review, the design of the study, the critical revision of the manuscript, and the processing of the figures. R.M.A. and A.M.A. were the pathologists who performed the histopathological diagnosis. A.J.Q. was the radiologist who assessed the case. S.H.H. and A.S.M. confirm the authenticity of all the raw data. All authors have read and approved the final manuscript.

Data availability statement

The datasets used and/or analyzed during the current study are available from the corresponding author upon reasonable request.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding

The author(s) received no financial support for the research, authorship, and/or publication of this article.

Ethics approval

Our institution does not require ethical approval for reporting individual cases or case series.

Informed consent

Written informed consent was obtained from a legally authorized representative(s) for anonymized patient information to be published in this article.

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