

Lingual osteoma: case report and literature review

Liu J Y, Tan K K H

ABSTRACT

Lingual osteomas are extremely rare. Fewer than 100 cases have been reported in the medical literature. This article reports a case of lingual osteoma, together with a literature review of its clinical features and pathogenesis so as to enhance our understanding of this unusual osseous lesion.

Keywords: glossal, lingual, osseous choristoma, osteoma, tongue

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INTRODUCTION

Osteomas are benign bony neoplasms consisting of normal mature osseous tissue. They are uncommonly found in the head and neck region, and rarely occur in intraoral tissues. Here, we report a case of lingual osteoma. The original description of this lesion is attributed to Monserrat in 1913; since then, fewer than 100 cases have been reported.⁽¹⁾ As this is a rare lesion, its pathogenesis remains a topic of controversy.

CASE REPORT

A 17-year-old boy presented with a painless mass measuring 0.5 cm in diameter on his dorsal tongue for eight years. It had remained the same size throughout the years. There was no history of trauma, and the patient had never experienced bleeding from the mass or dysphagia. Examination revealed a pedunculated mass covered with normal mucosa on the dorsal tongue, posterior to the line of circumvallate papillae and to the right of the foramen caecum. There was no cervical lymphadenopathy. The thyroid gland was normal. The lesion was excised under general anaesthesia via a per-oral approach. It measured 0.7 cm × 0.5 cm × 0.3 cm and was hard to palpate (Fig. 1). Histology confirmed a diagnosis of osteoma with dense sclerotic lamellar bone underneath the tongue epithelium (Fig. 2).

DISCUSSION

Lingual osteomas are extremely rare. To date, less than 100 cases have been reported since its first description by Monserrat in 1913.⁽¹⁾ Most occur in women, with a

female to male ratio of 3.25 to 1.00.⁽²⁾ The age of diagnosis is 5–73 years, but lingual osteomas are most commonly seen in the third and fourth decades of life. The most common location of the lesion is in the posterior third of the dorsal tongue. In most cases, the patients are asymptomatic but may occasionally experience a foreign body sensation in their throat (25.8%), dysphagia (6.9%), gagging (5.1%), nausea (3.4%) and irritation (3.4%).⁽³⁾ Reported lesions are 0.5–2.0 cm in diameter, with most being pedunculated. All cases have been unilateral, except for a case of two osteomas, one on either side of the foramen caecum, reported by Jung.⁽⁴⁾ Histologically, these lesions are well circumscribed and are composed of mature compact bone with Haversian systems. Osteoblastic cellular activity has been reported in only four cases, three of which had a history of the lesions increasing in size.⁽⁵⁾ Treatment in all cases was surgical excision, with no reported cases of recurrence.

The origin of lingual osteomas is uncertain, although three pathogenetic mechanisms have been proposed: post-traumatic reaction,⁽³⁾ developmental malformation^(1,3,5) and ossification of an embryologic thyroid remnant.^(6,7) The first hypothesis of post-traumatic reaction suggests that the posterior third of the tongue is a reactive centre of ossification, since it is susceptible to trauma and irritation from swallowing.⁽³⁾ Bony metaplasia then occurs due to previous injury, haematoma or chronic inflammation; this is similar to the pathologic process seen in post-traumatic myositis ossificans.⁽³⁾ However, this theory fails to explain why lingual osteomas are all composed of well-developed mature bone, rather than an inflammatory irregular bony pattern, which is to be expected in a traumatic lesion.

The second hypothesis of developmental malformation was proposed by Monserrat. Lingual osteomas are thought to be ossified branchial arch remnants due to their close proximity to the foramen caecum. Foramen caecum represents the junction between the first and third branchial arch derivatives, and is also the site where the second branchial arch disappears. Osteomas arise from either a congenital remnant of the second branchial arch or from the first or third arch remnants.⁽⁷⁾ During embryological development, multipotential mesenchymal cells may be entrapped in this

Department of
Otolaryngology,
Khoo Teck Puat
Hospital,
100 Yishun Central,
Singapore 768826

Liu JY, MMed,
MRCS
Registrar

Department of
Otolaryngology,
KK Women's and
Children's Hospital,
100 Bukit Timah
Road,
Singapore 229899

Tan KKH, MBBS,
MD, FRCS
Senior Consultant
and Head

Correspondence to:
Dr Liu Jiaying
Tel: (65) 8121 1670
Fax: (65) 6295 6339
Email: liu.jiaying@
singhealth.com.sg

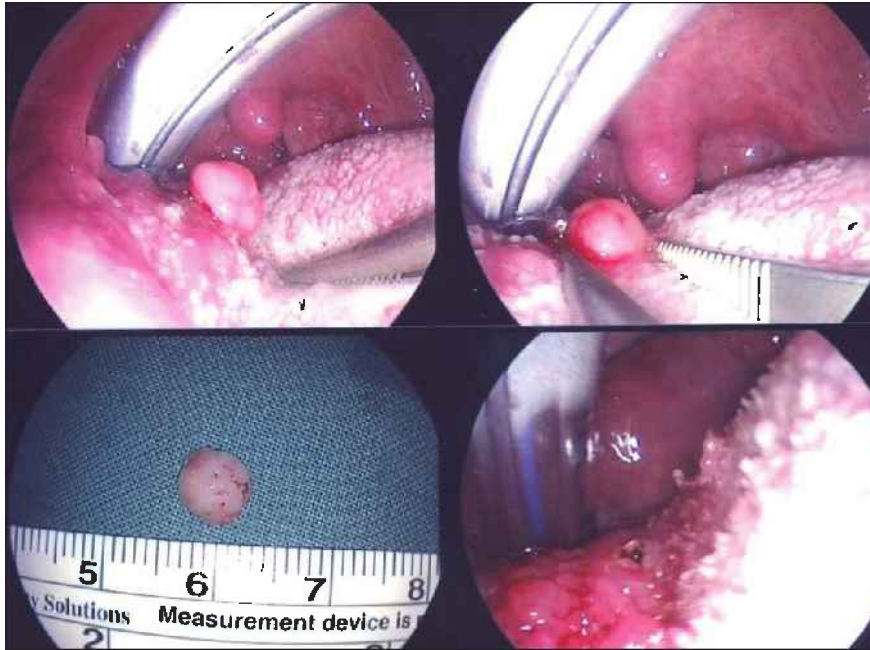


Fig. 1 Intraoperative photographs show the osteoma located at the posterior third of the tongue pre- and post excision.

region, and by some stimulus in the milieu, give rise to osteomas over time.⁽⁵⁾ This hypothesis explains the anatomic location but does not account for its predilection in females.

The final theory involves thyroid remnant calcification.^(6,7) It is an attractive theory, as it accounts for the distinct anatomic relationship between these osteomas and the foramen caecum, as well as the higher predilection seen in females. Foramen caecum is also the site where the thyroid anlage arises and descends into the neck. Remnants of undescended intraglossal thyroid tissue can lead to the development of this osseous lesion.⁽⁸⁾ Moreover, lingual thyroid remnants and embryologically displaced intralaryngeal thyroid tissue are all found predominantly in females.⁽⁹⁾ Metaplastic ossification in thyroid tissue is also known to occur in colloid goitres and thyroid cysts. Hence, lingual osteomas may be ossified derivatives of thyroid remnants.

Apart from aetiology, controversy pertaining to the correct terminology for this group of lesions also exists. Kroll et al⁽¹⁰⁾ believed that the term lingual choristoma was more appropriate than lingual osteoma. They argued that osteomas are benign, progressively enlarging neoplasms originating from osteogenic tissue, and thus closely associated with the skeleton. Lingual osteomas are neither closely related to the skeleton nor proven to be of osteogenic origin. Kroll recommended the term choristoma, as it applies to a cohesive, tumour-like mass consisting of normal cells in an abnormal location.^(10,11) In spite of the debate over its pathological features, the terms

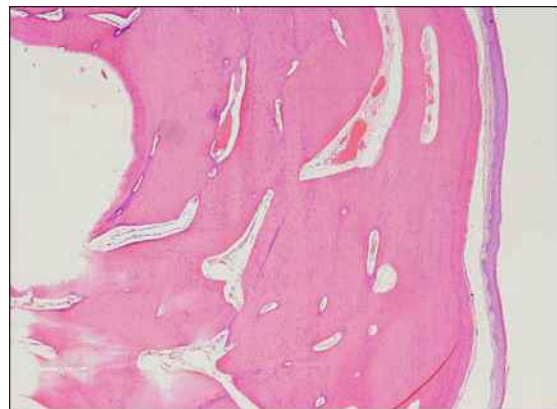


Fig. 2 Photomicrograph of the specimen shows the nodule of lamellar bone beneath the attenuated tongue mucosa (Haematoxylin & eosin, $\times 40$)

‘lingual osteoma’ and ‘lingual choristoma’ continue to be used interchangeably in the medical literature.

In conclusion, lingual osteomas are rare benign lesions commonly found in the posterior third of the dorsal tongue, close to the foramen caecum. They usually occur in females and have varied presentations. It is suggested that they represent either a developmental malformation or a post-traumatic reactive lesion. Definitive management is by surgical excision, with no cases of recurrence having been reported to date.

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