Osseous Choristoma of the Tongue — A Case Report

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Abstract

Osseous choristoma is a rare benign lesion in head and neck region. Here, we report a case of osseous choristoma on dorsum of the tongue in a 21 year-old female who presented with an asymptomatic pedunculated nodule on the posterior third of the tongue. She underwent total excision of the lesion under general anesthesia and there were no complications upon 2 weeks follow-up and no recurrence noted 1 year post-operatively. In reviewing previous articles, osseous choristomas are proliferations of the histopathologically normal tissue in an ectopic location. They may mimic other benign lingual soft tissue swellings and the only determinant of these lesions is their histopathology where it consists of developed harversian canal system beneath the stratified epithelium. They are benign and rarely recur after excision.

Key words: Osseous choristoma, Harversian canal system.

Introduction

Osseous choristoma is a tumor–like growth of microscopically normal tissue in an abnormal location. It is a rare benign lesion of the head and neck. The frequently observed choristomas of the oral cavity can be of bone, cartilage or both with a size around 0.5 to 2 cm in diameter and are also commonly found along the dorsal area of the tongue. The occurrence of this lesion is rare, and Kroll and his colleagues reported 24 cases in 1971, which firstly introduced the term of osseous choristoma. Here, we report a case of this unusual lesion on the dorsal tongue of a 21-year-old female patient, by reviewing her clinical process and histopathology of the lesion.

Case Report

A 21-year-old female, in good health upon physical examination and routine laboratory work up and denied any medical and family history, was seen in Oral and Maxillofacial Surgery, Department of Stomatology, Taipei Veterans General Hospital for consultation in October 2013. She presented an asymptomatic, hard pedunculated nodule on the posterior third of the tongue (Fig. 1). The lesion has been noticed for 5
years but the patient did not pay much attention to it. Until 1 month prior to consultation, there was a sudden increase of the lesion. Intraoral findings showed a verrucous nodular lesion with pedicle at right side posterior third of tongue around $1 \times 1$ cm in size, hard upon palpation, no obvious ulceration and currently in full orthodontic appliance.

Initial clinical impression was a benign tumor over posterior third of tongue. The differential diagnosis consist of focal fibrous hyperplasia, pyogenic granuloma, neurilemmoma, granular cell tumor, neurofibroma, fibroma, giant cell fibroma, fibrous histiocytoma and lingual thyroid.

Due to difficulty when tried to access the lesion, total excision was conducted under general anesthesia. The whole specimen sent for histopathologic examination composed of pink soft tissue and a small piece of bone measuring $0.8 \times 0.7 \times 0.6$ cm and $0.7 \times 0.6 \times 0.4$ cm. Microscopic findings revealed benign squamous mucosa and an osseous choristoma in the submucosal area (Fig. 2), S-100 stain is non-reactive and no evidence of malignancy.

The patient recalled after 2 weeks and 1 month post-operation that showed with fair wound healing and was given instructed to return if noticed any condition. There were no recurrences of the lesion for 1 year after surgery.

**Discussion**

As choristomas, there are several different tissues that may occur in the oral cavity which includes bone, cartilage, gastric mucosa, glial tissue and tumor-like masses of sebaceous glands\(^1,2\). The most frequent can either be osseous or cartilaginous or both depending on the histological features. The choristoma does not appear as true neoplasms that they are sometimes called soft tissue osteomas or soft tissue chondromas\(^1\). The clinical feature of the said lesion is usually firm, smooth-surfaced can either be sessile or pedunculated with a size around 0.5 to 2.0 cm. It can occur anywhere in the head and neck area. In the oral cavity, it is commonly seen along the posterior tongue along the area of foramen caecum, but there are rare cases that it can be seen in other locations such as anterior and ventrolateral surface of the tongue, lingual alveolus of the mandible, masseter and buccal mucosa\(^1,4\). According to Gorinin and his colleagues, most of the osseous choristomas at tongue presented with asymptomatic swelling, and most frequently complained was lump, which accounted for 46% of 66 reviewed cases\(^5\). And there are some patients presented with other symptoms such as dysphagia, gagging, pain, foreign body sensation, vomiting, nausea and sudden increase in growth\(^5,6\). The presented case is asymptomatic with a chief complaint of sudden increase of size. It occurs in any age, but commonly seen in young adults and children\(^1\). The majority of being diagnosed are women around the age between 20 and 40 years\(^2\). Youngest victim being reported is at the age of 5 that is found at the buccal mucosa\(^3\).

Due to its rarity, it is usually misdiagnosed as soft tumors. Differential diagnosis included focal fibrous hyperplasia, pyogenic granuloma, neurilemmoma, granular cell tumor, neurofibroma, fibroma, lipoma, giant cell fibroma, fibrous histiocytoma and lingual thyroid as they are ruled in due to similarities to clinical presentations.

There are several hypotheses to explain the occurrence of choristomas of the tongue such as ossification of brachial arch remnants, congenital abnormality or developmental malformation,
Fig. 1. Pedunculated mass on the posterior third of the tongue (black arrow), measuring $0.8 \times 0.7 \times 0.6$ cm.

Fig. 2. Benign squamous mucosa (yellow arrow) and compact mature bone with normal Haversian canals in the submucosa (blue arrow).
ossified lymphatic tissue, calcifying remnants of the thyroid gland, degenerating fibroma undergoing ossification, neoplasm or teratoma with cartilage preponderance, metaplasia, derivation from pluripotential cells and cartilaginous embryonic rests\textsuperscript{6, 7}. Origin of the lesion is also debatable that it can be developmental or traumatic, but currently, the exact origin of choristoma is unknown\textsuperscript{2}. It can be developmental by embryonic rests because of its close proximity to the foramen cecum or traumatic by differentiation of pluripotential mesenchymal cells into chondrocytes or cartilaginous metaplasia of the connective tissue\textsuperscript{7}. Metaplastic theory is supported by patients with the history of trauma and chronic inflammation\textsuperscript{7, 8}.

Histologically, it is a well-circumscribed mass of viable lamellar bone with well-developed Harversian canal system, as well as developed mass of viable cartilage, or a mixture of both bone and cartilage surrounded by dense fibrous connective tissue. Occasionally there is a presence of central fatty or hematopoietic marrow\textsuperscript{1, 2}. Tumor-like growth that has developed from groups of primordial cells located at the site remote from the original tissue or organ. Final diagnosis of this lesion presented by patient reported determines of its histopathological findings, which is osseous choristoma.

The best treatment of osseous and cartilaginous choristoma is total surgical excision. Recurrence is very rare. As of to date, there are only 2 reported cases showed recurrences of choristoma. A 22 year old male showed recurrence 1 year after surgical excision in the masseter area and the other one recurred 12 years after the surgery, which the lesion located in the buccal soft tissue connected to the mandibular ramus\textsuperscript{4, 6}.

**Conclusion**

Osseous choristoma may mimic other benign lingual soft tissue swellings, but determinant of this lesion is its histopathological presentation. And, the best treatment is total excision. The recurrence of the lesion is rare.

**References**

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舌上骨性迷芽細胞瘤—病例報告

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摘 要

骨性迷芽細胞瘤為罕見之頭頸部良性腫瘤。本篇報告討論一位21歲女性患者因舌上一無痛突起物而求診，詳細理學檢查後發現為一含柄狀之突起物於舌根處。經全身麻醉手術切除病灶後，病理報告證實為骨性迷芽細胞瘤。術後一年追蹤並無復發病灶。骨性迷芽細胞瘤於組織切片鏡檢是正常組織異位增生所致，常與舌側軟組織類似，唯一差異是病理組織切片下可見在複層上皮下，有已發育的哈氏管。骨性迷芽細胞瘤在手術切除後極少復發。提出此病例以供研究及治療之參考。

關鍵詞：骨性迷芽細胞瘤，哈氏管。

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