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CASE REPORT

Mixed choristoma on the anterior dorsal tongue: a new case and review of the literature

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Abstract

A choristoma is a benign tumour-like growth consisting of normal tissue in an abnormal site. Seven cases of mixed osseous and cartilaginous choristoma of the tongue have been reported in the literature, only one on the anterior tongue. Here we review the literature regarding osseous and cartilaginous choristomas and report only the second case of mixed osseous choristoma to occur on the anterior dorsal tongue.

Clinical relevance

It is possible that mixed choristoma may occur on any region of the tongue. Therefore, it should be included in the differential diagnosis of any firm swelling on the tongue. Oral surgeon should be aware of the benign behaviour of these lesions to avoid concern to patients and aggressive surgical management.

Introduction

A choristoma is a benign tumour-like growth consisting of normal tissue in an abnormal site[1]. Osseous or cartilaginous choristomas are rare lesions occurring mainly in head and neck region[2,3]. The osseous choristoma was first reported on the tongue by Monserrat in 1913, who used the term 'lingual osteoma' referring to a neoplastic lesion[4], while this lesion behaves benignly and can be treated by surgical excision with extremely rare recurrence[5,6].

In 1971, Krolls *et al*. introduced a more appropriate term 'osseous choristoma' to describe a hamartomatous tumour-like mass[7].

In this article, we describe the clinical and microscopic features of osseous and cartilaginous choristomas with brief review of the literature and report the second case of mixed osseous choristoma to occur on the anterior dorsal tongue.

Case report

A 57-year-old male patient was seen in May 2008 for investigation of an asymptomatic pedunculated swelling on the anterior right dorsal aspect of the tongue. The patient reported that the lesion had been present for 3 years and could not elicit a history of trauma. The lesion measured approximately 0.8 cm in greatest dimension, and blanched slightly under pressure but appeared to arise from the underlying connective tissue. The surface mucosa was slightly keratotic showing evidence of minor chronic trauma, and the lesion was hard on palpation.

Clinically, the lesion appeared consistent with a traumatic fibroma (fibroepithelial polyp) although a peripheral ossifying fibroma was also entertained (Fig. 1). The lesion was excised completely under local anaesthesia and examined by routine histopathology

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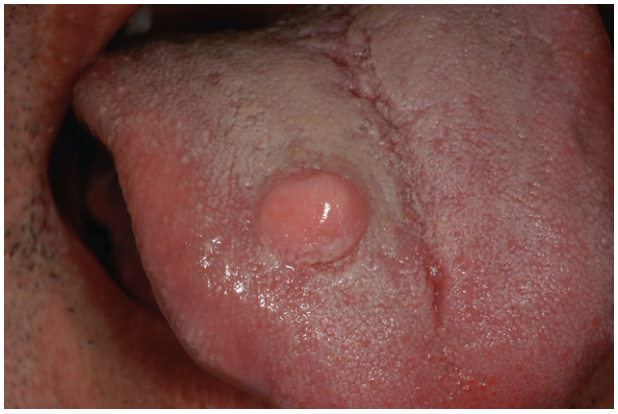

Figure 1 Firm soft tissue lesion located on the anterior right dorsal tongue.

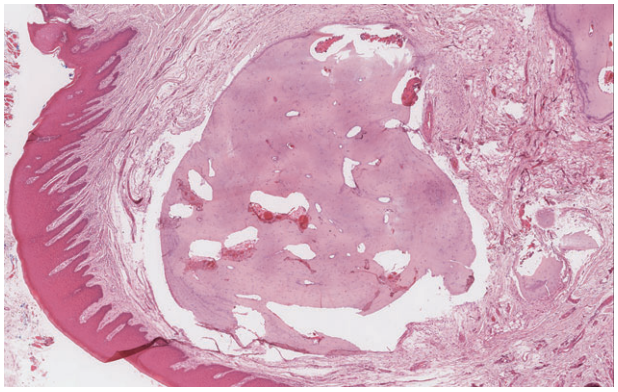

Figure 2 Histological section reveals a polypoid mass of well encapsulated cartilage and dense bone beneath a non-ulcerated non-papillated lingual mucosa (final magnification × 100; stain H&E).

with haematoxylin and eosin. The surgical site healed well without complication.

Microscopic examination revealed a polypoid mass of well encapsulated cartilage and dense bone beneath a non-ulcerated non-papillated lingual mucosa. There was no evidence of active chondroblasts, osteoblasts or osteoclasts (Fig. 2). The lesion was diagnosed histologically as an osseous choristoma with cartilaginous components.

Discussion

Osseous choristomas and cartilaginous choristomas are relatively rare benign lesions with approximately 61 and 28 cases having been reported in the literature, respectively[5,6,8], however, there are seven cases reported to contain a mixture of these tissues[9].

Regarding the osseous lesion, there is an obvious predominance in females with a male to female ratio of 1:5. This gender difference is not found in cartilaginous lesions. Clinically, the majority of osseous choristomas of the tongue present as hard pedunculated smooth-surfaced lesions and occur at the base of the tongue adjacent to foramen caecum and the circumvallate papillae[10,11]. No osseous choristomas have been reported on the ventral tongue. Osseous choristomas have been reported in other sites including the buccal mucosa[12], submental region[13], retromolar pad area[14] and masticatory muscles[15]. Lingual osseous lesions are generally less than 2.5 cm in diameter[16], with the age at time of initial diagnosis ranging from 8 to 73 years. Histologically, the lesion consists of a well-circumscribed, lamellated mass of dense viable bone with a haversian system, as well as osteocytes in lacunae[2]. The bony mass is surrounded by dense fibrous connective tissue covered with thin stratified squamous epithelium[11,17].

Cartilaginous choristomas are frequently found on the dorsal tongue, but four cases have been reported on the ventral tongue[6,8]. Other intraoral sites for cartilaginous choristomas are gingiva[18], soft palate[19], buccal mucosa[20] and palatine tonsil[21]. Time of presentation ranges between 3 to 75 years of age but occurs most commonly during the third and fourth decades of life. The largest lingual cartilaginous choristoma was 4.5 cm in greatest diameter[22]. Microscopically, these lesions consist of a circumscribed nodule composed of hyaline cartilage with well-defined lacunae, showing small chondrocytes[23].

With regards to lesions with mixed osseous and cartilaginous components as noted in Table 1, the age at presentation ranges from 20 to 67 years, with a male to female ratio of 6:1. The size of such lesions varies from 0.7 to 2.5 cm. Four cases (57%) are located on the base of the tongue and two cases (28%) on the ventral tongue, while only one case has been recorded on the anterior third of the tongue[24]. To our knowledge, this is the second reported case of choristoma with mixed osseous and cartilaginous components to be located on the anterior third of the dorsal tongue.

The majority of choristomas of the tongue are asymptomatic, however, there have been some reports of dysphagia, gaging, nausea, irritation and swelling in the throat[1,2]. The present case was asymptomatic.

Differential diagnosis of choristomas should include a thyroid nodule, hyperplastic tonsils, fibroma, granular cell tumour and neural neoplasm. In the current case, the lesion appeared to be most consistent with a fibroma, given its location and tissue consistence. The current case draws attention to the

Table 1 Summary of the main clinical data of mixed lingual choristoma cases

Authors	Sex	Age (years)	Location	Diameter (cm)	Duration	Symptom
Roy *et al*.[28]	F	20	Near foramen caecum	1.0	10 years	Bothersome
Gabriele & Kaufman[29]	F	22	Near foramen caecum	1.0–1.5	Found during routine examination	Asymptomatic
Wesley & Zielinski[30]	F	57	Ventral surface of the tongue	1.0	Entire life	Asymptomatic
Landini *et al*.[24]	F	35	Left lateral border of the tongue	0.8	23 years	Asymptomatic
Watson *et al*.[31]	F	67	Junction between anterior one third and posterior one third	0.7–1.0	6 months	Sensation of a lump in the throat
Piattelli *et al*.[32]	F	64	Right ventral tongue	0.8	2 years	Not mentioned
Lee[33]	M	26	Posterolateral border of the tongue	2.5	8 years	Asymptomatic
AbdulMajeed & Farah	M	57	Anterior right dorsal aspect of the tongue	0.8	3 years	Asymptomatic

need of including choristoma as a differential diagnosis for such lesions.

Treatment of choice for osseous and cartilaginous choristomas is surgical excision, with recurrence being extremely rare[5,6,12]. No follow-up complication of lingual choristomas has been described in the literature.

The aetiology of choristomas remains obscure[3] but several theories have been suggested to explain this. Generally, these theories can be designated either as developmental or reactive[2]. The developmental theory proposes that the complex embryology of the tongue leads to a developmental malformation which includes ossification of the branchial arch remnants[4] or calcifying of the thyroid gland remnants[25]. This theory could explain why osseous choristomas are so widely distributed in the area of foramen caecum and the circumvallate papillae. The developmental theory for cartilaginous choristomas postulates that the lesion originates from heterotopic cartilage remnants from any of the first four branchial arches. It is believed that chondroblastic cells are misplaced during development and sequestered in the tongue.

The reactive theory proposes that the lesion is reactive in nature, and this results in central ossification similar to myositis ossification[26,27]. In the case of cartilaginous choristomas, the reactive theory postulates that trauma can stimulate metaplastic change[23].

In the current case, branchial remnants or thyroid tissue were not found histologically. In addition, the history given by the patient and the position of the lesion on the anterior third of the dorsal tongue makes trauma the most possible cause. We suggest that this lesion is preceded by a traumatic event such as a tongue bite that led to chronic inflammation, stimulating metaplastic change to fibrous, cartilaginous and finally to osseous tissue, and this may explain why the lesion contained a mixture of cartilaginous elements within a solid osseous mass.

Previous literature has considered osseous and cartilaginous choristomas as separate entities, however we propose that these two types of choristoma have a common aetiopathogenesis. Lesions start with a traumatic episode such as mastication or a tongue bite that leads to local inflammation, development of hyperplastic tissue, and fibroma formation. As the fibroma bulges above the surface of the tongue, it is subjected to further trauma. Frequent traumatic episodes may lead to metaplastic ossification of a degenerating fibroma that begins with cartilaginous formation. Continuous trauma and metaplastic ossification leads to formation of bone within the lesion. It is feasible then, through this suggested pathway that osseous and cartilaginous choristomas have the same aetiopathogenesis but osseous choristomas represent a late presentation of the lesion. This may explain why most choristomas reported in the anterior part of tongue are cartilaginous, as they are more likely to be identified by the patient or clinician early, with ensuing diagnosis and surgical removal. Conversely, when the lesions develop at the base of the tongue, they have more time for osseous formation given that they are asymptomatic and not identified at an early stage.

In conclusion, osseous and cartilaginous choristomas should be included in the differential diagnosis of any firm swelling on the tongue. It is possible that this lesion may occur on any region of the tongue. Both lesions may have the same aetiopathogenesis but osseous choristomas represent a late stage in the development of this entity. Awareness of its benign behaviour is important to avoid aggressive surgical management.

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