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Infected lingual osseous choristoma. Report of a case and review of the literature

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Abstract

Osseous choristoma is a rare, benign lesion of the oral cavity occurring usually in the tongue. It appears as a tumorous mass of normal bony structure with mature cells in an ectopic position. The case of a 72 years Caucasian male is presented and analyzed along with 52 similar cases reported in the English literature between 1967 and 2007. Lingual choristoma shows a female predilection, whereas the commonest anatomic location is the posterior third of the tongue, occurring at or close to the foramen caecum and the circumvallate papillae. Histologically the lesions show signs of a well-circumscribed mass of vital bone located under the surface oral epithelium. Some lesions represent developmental malformations, whereas others may be reactive lesions after trauma or chronic irritation. Treatment of lingual osseous choristoma consists of simple excision.

Key words: *Lingual choristoma, osteoma of the tongue, ectopic tissue, oral developmental anomalies.*

Introduction

Choristoma is defined as a tumor-like mass of normal cells or tissue that develops in an ectopic location (1). Several different tissue types may occur in the mouth as choristomas. These include bone, cartilage, gastric mucosa, glial tissue, and tumor-like masses of sebaceous glands (2). However the most frequently observed choristomas of the oral cavity are those that consist of bone (3, 4). These lesions have also been called soft tissue osteomas, but osseous choristoma is a more accurate term as the lesions are not true neoplasms (5). Choristomas occur most frequently in the tongue (6) and less commonly in other sites such as buccal mucosa (7, 8) and alveolar mucosa (3, 9). The case of an infected lingual osseous choristoma is reported and the literature from this rare entity is reviewed.

Case Report

A 72-year old white male was referred to our department with a chief complaint of pain and swelling of the posterior tongue in combination with dysphagia. The patient suffered from a Type A diabetes which was not under control. He had been aware of the lesion for several years and connected that with local trauma from the dentures that he wore. Intra-oral clinical examination revealed a pedunculated well circumscribed swelling measuring 2 cm in the posterior one-third of the dorsal area of the tongue (Fig. 1). The lesion was situated just anterior to the foramen caecum near the midline. It was covered with a thin normal looking mucosa, and in the middle of the lesion there was a fistula from where a purulent discharge could be observed. Palpation showed a painful, firm mass, fixed to the underlying tissue. The patient was put on antibiotics and symptoms resolved within four days. Excision biopsy

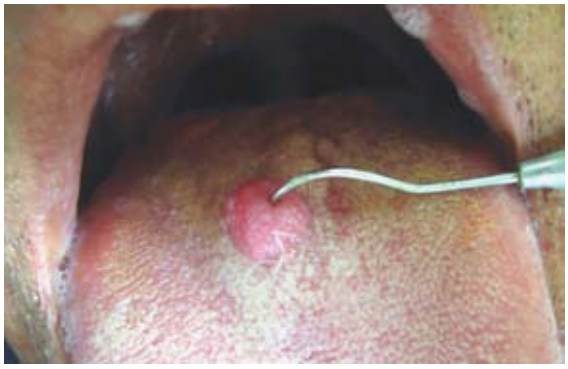

Fig. 1. A tumorous mass in the posterior dorsum of the tongue covered from healthy looking mucosa is clearly evident.

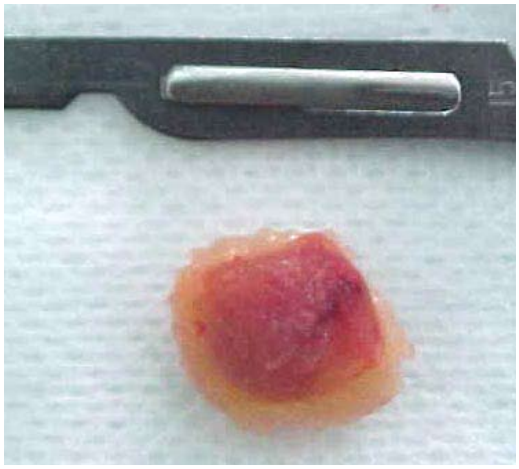

Fig. 2. The excised specimen. The tumor mass is covered by oral mucosa without any invasion

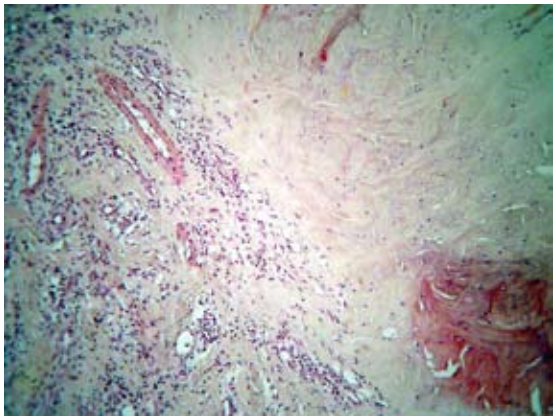

Fig. 3. Microphotograph of the histological slide from the excisional biopsy. Under the oral epithelium an osteoid mass with sparse osteocytes and a few reversal lines is evident. The underlying lamina propria was infiltrated by inflammatory cells mainly lymphocytes, plasmacells and macrophages. (H and E x 25)

was performed under local anesthesia (Fig. 2). The histopathological examination of the specimen showed that the lesion consisted of a white-tan mass, which measured 1,5X1X1 cm. Microscopic examination showed under the oral epithelium an osteoid mass with sparse osteocytes and a few reversal lines. The underlying lamina propria was infiltrated by inflammatory cells mainly lymphocytes, plasmacells and macrophages (Fig. 3).

Discussion

A literature review from 1967 to 2007 yielded 53 cases of lingual choristomas (2-6, 9-36). Inclusion criteria in this study were cases with adequate clinical information and histological diagnosis of tumor-like masses in the tongue which exhibited bone formation without neoplastic features. The present study was added (Table 1).

The ages of the patients at the time of initial diagnosis ranged from 5 to 73 years with a mean age of 29,8. Twenty nine cases (54.7%) were patients in their third and forth decades of life. In four patients the lesion was present at birth or during early childhood (9, 14, 23, 24). The male-female ratio was 1:2.8 in favor of females. The anatomic location of the tumor was in the posterior third of the tongue in 41 cases (77%), at the lateral border in 9 cases (17%), and in middle third of the tongue in 3 cases (6%). The lesions in the posterior third were at or in close proximity to the foramen caecum and the circumvallate papillae. The sizes of the lesions varied from 3 mm to 5 cm at their largest diameter. Clinically, they presented as a hard mass, either pedunculated or sessile. In four cases, the lesions were lobulated (4, 5, 9, 32). In most cases, the covering mucosa exhibited a normal clinical appearance, ulceration was present in one case (19) and another lesion was described as a mass with verrucal surface (3). The duration of the tumors ranged from 3 days (32) to 50 years (23). In 9 cases the lesions showed an increase in size (12, 14, 19, 20, 31-34). In 18 cases the lesion was detected as incidental finding on routine clinical oral examination and 22 cases complained of a painless lump in their tongue.

Histologically, the lesions were composed of a well circumscribed, lamellated mass of dense, vital bone with a well-developed haversian canal system, surrounded by dense, fibrous connective tissue and covered with stratified squamous epithelium in the exophytic part of swelling (2, 29, 32, 33). Cellular activity of osteoblasts at the periphery of osseous mass is inconspicuous and was reported in only four cases, (19, 22, 33, 34) three of which had a history of increasing in size lesions (19, 33). The presence of bone marrow spaces filled with hematopoietic or fatty tissue was uncommon and was described in only three cases (21, 22, 26).

All lesions were treated by surgical excision with uneventful healing. Recurrence or malignant transformation has not been reported.

The pathogenesis of lingual choristoma remains obscure

Table 1. Age, sex, duration, location, size, form and main symptom of patients with lingual osseous. choristoma.

Author	Age(y)/Sex	Duration	Location	Size	Form	Symptom
Cataldo et al [11]	39/F	4 months	Posterior tongue	1.0 cm diameter	Pedunculated	None
Begel et al [12]	22/F	2 years	Area of CP	1.0 x 0.5 cm	Sessile	Dysphagia
Jahnke & Daly [13]	22/F	13 years	Posterior to CP	1.3x0.8x0.7 cm	Pedunculated	Lump
Kaye [14]	26/F	Since childhood	Base of tongue	1.0x1.0 cm	Pedunculated	Lump
Goldberg et al [15]	65/M	Unknown	Lateral border	1.0 cm diameter	Hard mass	None
Krolls et al [5]	22/F	2 years	Anterior to CP	0.75 cm diameter	Papillomatous	None
	23/M	Unknown	Area of FC	Unknown	Pedunculated	Unknown
	73/M	Years	Posterior tongue	Unknown	Pedunculated	Gagging
	9/F	2.5 months	Area of FC	Unknown	Pedunculated	Gagging
	25/F	4 months	Posterior tongue	0.5 cm diameter	Sessile	Unknown
	11/F	1 year	Posterior tongue	2.0 cm diameter	Pedunculated, lobulated	Unknown
	23/M	Unknown	Area of CP	0.5x0.5x0.5 cm	Sessile	None
	39/M	Unknown	Area of CP	0.6x0.6 cm	Pedunculated	None
Singh & Doyle [16]	14/F	Unknown	Left border	Unknown	Unknown	Unknown
	22/F	Unknown	Area of CP	0.5 cm diameter	Pedunculated	None
McClendon [17]	15/F	Unknown	Area of FC	1.4x0.6x0.5 cm	Pedunculated	None
	20/M	Unknown	Right border	0.7 cm diameter	Sessile	None
	46/F	Unknown	Area of FC	0.6 cm diameter	Pedunculated	None
Patel & Dane [18]	42/M	Unknown	Lateral border	1.0 cm diameter	Papillomatous	None
Engel & Cherrick [19]	31/M	3 years	Mid third, right border	2.0 cm diameter	Ulcerated mucosa	Lump
Busuttil [20]	8/F	9 months	Left border	Pea-sized	Unknown	Lump
Esguep et al [21]	63/F	2 months	Right border	0.5 cm diameter	Sessile	Lump
Wasserstein et al [22]	50/F	3 months	Mid third	1.5x0.75 cm	Movable	Lump
Shimono et al [4]	37/F	8 years	Area of FC	1.5x1.5x0.7 cm	Pedunculated, lobulated	Lump
Main [23]	54/F	Since childhood	Posterior to FC	1.5 cm diameter	Pedunculated	Lump
Sheridan [9]	20/F	From birth	Anterior to CP	1.0 cm diameter	Pedunculated, Lobulated	Lump
Cabbabe [24]	5/F	2 years	Base of tongue	0.6x0.5x0.3 cm	Pedunculated	Lump
Nash et al [25]	31/M	Unknown	Right border	2.5 cm diameter	Sessile	None
Weitzner [26]	52/F	Unknown	Mid third	Small nodule	Sessile	None
	25/F	Unknown	Posterior tongue	0.8x0.4x0.4 cm	Unknown	Lump
	27/F	Unknown	Posterior tongue	0.8x0.7x0.3 cm	Unknown	Lump
Tohill et al [3]	31/F	Unknown	Anterior to CP	1.0x0.8x0.7 cm	Unknown	None
Markaki et al [27]	25/F	5 months	Posterior to CP	0.8x0.4x0.3 cm	Pedunculated	Lump
van der Wal & van der Waal [6]	31/F	Unknown	Area of FC	1.0 cm diameter	Unknown	Lump
Cannon & Niparko [28]	51/F	20 years	Posterior tongue	Unknown	Unknown	Lump
Bernard et al [29]	27/F	12 years	Area of FC	2.0 cm	Pedunculated	Lump
Maqbool et al [30]	8/F	6 months	Right vallecula	5.0x4.0 cm	Pedunculated	Dysphagia airway distress

Lutcavage & Fulbright [31]	11/F	1 year	Posterior to FC	1.0 cm diameter	Unknown	Lump
Ishikawa et al [32]	53/F	3 days	Area of FC	0.8 cm diameter	Pedunculated	Foreign body sensation
	5/F	1 month	Anterior to CP	3 mm	Pedunculated, lobulated	Lump
Lee et al [33]	35/M	3 years	Lateral border	Unknown	Sessile	Lump
Vered et al [34]	44/M	Months	Left border	0.7x0.7x0.6 cm	Sessile	Gagging, nausia, dysphagia
	27/M	Months	Posterior to CP	1.0x0.5 cm	Pedunculated	Pain, gagging
Supiyaphun et al [35]	28/F	4 years	Area of FC	1.0x0.8x0.6 cm	Pedunculated	Throat irritation
	25/F	1 year	Area of FC	0.7x0.5x0.4 cm	Pedunculated	Lump
	9/F	Unknown	Area of FC	0.7x0.6x0.5 cm	Pedunculated	None
	35/F	Unknown	Area of FC	0.7x0.5x0.5 cm	Pedunculated	None
	27/F	Unknown	Area of FC	1.2x0.9x0.6 cm	Pedunculated	None
	21/F	5 years	Area of FC	1.5x1.3x0.8 cm	Pedunculated	Lump
	22/M	Unknown	Area of FC	0.9x0.8x0.6 cm	Pedunculated	None
	19/F	11 years	Area of FC	1.1x0.7x0.7 cm	Pedunculated	None
Horn et al [36]	11/F	1 year	Posterior tongue	Unknown	Unknown	Lump
Present case	72/M	Years	Anterior to CP	1.5x1.0 cm	Pedunculated	Pain, dysphagia

FC: foramen caecum
CP: circumvallate papillae

and a variety of theories have been proposed to explain its etiology. These theories can be divided into two main categories: the developmental malformation theory and the reactive or posttraumatic theory (34).

Monserrat was the first to suggest the developmental malformation theory, who attributed the lesion's origin to the ossification of branchial arch remnants, basing his theory on the anatomic location of the lesion in the foramen caecum area (10). During embryologic development of the tongue, the union between its anterior two thirds and its posterior third takes place in the region of the foramen caecum and the sulcus terminalis. The anterior two thirds of the tongue originate from the first branchial arch, and the posterior third originates from the third branchial arch. Also this region is the site where the second branchial arch normally disappears. It is interesting to note that certain normal osseous structures derive from each of the branchial arches that contribute to the formation of the tongue: i.e., the incus and malleus from the first; the stapes, styloid process, and the lesser horn of the hyoid bone from the second; and the remainder of the hyoid bone from the third arch. Therefore, the possibility of enclavement of mesenchymal pluripotential cells originate from these embryonic branchial arches, and subsequent development of an osseous lesion in the tongue seems an attractive theory for its origin. This theory is strongly supported by Begel et al (12), and Engel and Cherick (19).

Cataldo et al (11) and Jahnke and Daly (13) have proposed a developmental theory, which is associated with remnants of thyroid tissue. The foramen caecum is the site where the anlage of the thyroid gland develops in embryologic life and from this site the glandular tissue descents to the neck to take its normal place. They suggested that, remnants of underdescended intraglossal thyroid tissue, either as primordial endodermal or as differentiated thyroid parenchymal cells, can produce unusual osseous proliferating lesions later in life, mainly during puberty and adolescence. Furthermore, the embryologically displaced intralaryngeal thyroid tissue, lingual thyroid and osseous lingual choristoma, all three conditions predominantly occur in women between the ages of 20 and 30 and therefore, are intenerated. Also, metaplastic ossification in thyroid tissue is not an uncommon event in colloid goiters and in thyroid cysts (13).

Other theories include epignathous formation, and that of degenerating fibroma undergoing ossification (37). The latter theory suggests that osseous lesions of the tongue represent a reactive or posttraumatic center of ossification (38). These types of lesions have been reported in other muscles of the body under the term "myositis ossificans" (4, 12, 25). It is indicated that either pluripotential cells or ectopic mesenchymal cells were probably present at these sites and, when stimulated by trauma, produced bone or cartilage (29, 33). It is generally believed that such lesions

of the buccal mucosa and the anterior aspect of the tongue are posttraumatic centers of ossification whereas these of the posterior tongue are developmental abnormalities (3, 7). Although in such cases one finds chronic inflammatory reaction, residual cartilage and irregularity of the bone pattern, these changes are never seen in lingual osseous choristomas.

Controversy also exists as to the adequate terminology for this group of lesions. Choristoma is a more accepted term and it is used first time from Krolls et al (5). According to these investigators choristoma refers to a tumorlike growth that has developed from groups of primordial cells located at a site remote from the original tissue or organ (2). The term osteoma is defined as a benign, progressively enlarging, neoplasm of bone originating from osteogenic tissue and closely associated with a part of the skeletal structure. However most investigators claim that the biologic behavior of the lesion does not fulfill the criteria of a true neoplasm, and there isn't a close relation to the skeleton (34). Also it defers from a hamartoma which is a focal overgrowth of normal cells that occurs at a normal site of involvement (5, 39).

The differential diagnosis of the lingual choristoma depends on the location of the lesion (3). When the lesion is located near the foramen caecum, the most important condition that should be entertained is the presence of single or multiple foci of ectopic thyroid gland in the tongue and a thyroid function test and scanning may be required before treatment (6, 17, 31). Lingual thyroids are usually globular and reddish nodules of varying sizes which occur in the median line between the foramen caecum and the epiglottis, in an anatomical location more posterior than the most lingual osseous lesions. An inadvertent removal of the lingual thyroid may cause permanent functional hypothyroidism if it happens to be the only thyroid tissue present in that patient (22). Hyperplastic lingual tonsil and salivary gland neoplasms should also be included in the differential diagnosis. When the lesion is located on the anterior and lateral aspect of the tongue, fibroma, granular cell tumor, neural tumor, and foreign body glanuloma, should be considered. Lesions on the ventral surface of the tongue may resemble salivary gland neoplasms, mucous retention phenomena, lipomas, and neural tumors. When the lesion is pedunculated and has a verrucal surface, it may clinically resemble a papilloma (3).

Lingual osseous choristoma shows a benign clinical behavior, and no recurrence or malignant transformation has been reported. Although two cases, one with location in the buccal mucosa (7), and one with location within masseter muscle (40) have been reported as recurrent, simple surgical excision of the tumor and follow-up would be satisfactory in the majority of cases.

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