The Radiological Diagnosis of Lingual Osseous Tumor-like Lesion

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ABSTRACT

A lingual osseous tumor-like lesion is a rare curiosity of unknown etiology. No malignant transformation has been reported. A 40-year-old healthy woman suffered chronic rhinosinusitis. An otorhinolaryngological physical examination revealed that the bilateral inferior turbinates were hypertrophic, while others were normal. However, sinus computed tomography incidentally showed the presence of an osseous lesion about 1.0 × 0.9 × 0.6 cm at the tongue base. At this point, it became problematic as to whether surgical excision should be recommended for the pathological diagnosis of an asymptomatic lingual lesion. The computed tomography demonstrated that the mass had a homogeneous bony density over the tongue base and therefore a lingual osseous tumor-like lesion was diagnosed that could be differentiated with other osseous neoplasms. As the radiological demonstration was enough to diagnose a lingual osseous tumor-like lesion, then surgical excision and pathological diagnosis was found to be unnecessary for such a benign and slow-growing lesion.

CASE REPORT

A 40-year-old healthy woman presented with posterior nasal dripping and stuffy nose that she had suffered from for several years. On visiting our clinic, she underwent an otorhinolaryngological physical examination that showed the bilateral inferior turbinates were hypertrophic, and but that the others were normal. However, a Water’s radiograph demonstrated bilateral maxillary sinuses that were opacified. A follow-up sinus computed tomography without contrast demonstrated (1) bilateral maxillary sinuses and ethmoid sinuses that were full of low density material, (2) a nasal septum that was ridged to the left side, and (3) the presence of a small high density mass, about 1.0 × 0.9 × 0.6 cm, at the tongue base (Fig. 1, filled arrows). Based on the above, chronic rhinosinusitis and nasal septal deviation were diagnosed, and a lingual neoplasm was suspected. At this point, it became problematic as to whether surgical excision should be recommended for the pathological diagnosis of an asymptomatic lingual lesion. Over the following half year, treatment with a corticosteroid nasal spray and other forms of symptomatic control was able to relieve the nasal disease. The tongue-base lesion was still asymptomatic. It did not increase in size and its covering mucosa kept normal, so regarded as a still mass.
DISCUSSION

The diagnosis of osseous lesion remains relatively straightforward and usually relies on a combination of clinical history and radiography [1], complemented by computed tomography, scintigraphy or magnetic resonance imaging [2, 3]. Computed tomography remains more useful than scintigraphy or magnetic resonance imaging because of its low cost, the immediate availability and the ease of diagnosis [2, 3]. The patient’s lingual mass was embedded in the lingual tonsil and covered with normal mucosa, so escaping from the otorhinolaryngologist’s physical examination. The computed tomography demonstrated a mass having homogeneous bony density over the tongue base and therefore a lingual osseous tumor-like lesion was diagnosed. This could be differentiated from other osseous neoplasms, such as (1) osteosarcoma (or “osteogenic sarcoma”), which shows local bony destruction or superficial ulceration as part

Figure 1

Figure 1. a. the coronal view of a sinus computed tomography. b. the axial view of the computed tomography. c. the saggital view of a sinus computed tomography. d. the volume rendering of mandible, hyoid bone and the lingual osseous tumor-like lesion.
Lingual osseous choristoma

of the gross appearance [4, 5], and (2) osteolipoma, which shows a mixture of soft tissue density and bony density on imaging [6-8]. The final possibility, osteoblastoma, has a heterogeneous bony dense on imaging and has never been reported to affect the tongue [9].

In 1913, Monserrat first reported a case of an osseous lesion in the tongue base and termed it “lingual osteoma”, which implies a neoplasm [10]; however, this lesion is benign, slow growing and composed of well-circumscribed dense bone beneath the epithelial surface. This type of biological behavior is unlike a neoplasm and therefore “osseous choristoma” has been applied as a better term for this lesion as it implies that there is a normal tissue growing in the wrong organ [11]. In fact, its pathogenesis remains obscure, so Vered et al [12] suggested a more descriptive term “osseous tumor-like lesion of the tongue”. Nonetheless, only the use of the terms “lingual osteoma” and “lingual osseous choristoma” were able to identify associated articles from the literature.

Although such lingual lesions are rare, there are eight case reports available in the Taiwanese literature since 1993 and these consist of twelve patients (five males and seven females) with an average age of 29 years (range: 15~47) (Table. 1) [13-20]. The intraoral osseous tumor-like lesion occurred predominantly in females with a female to male ratio of 1:4: 1 and in the posterior dorsal tongue near or at the circumvallate papillae or the foramen cecum. Other locations for the tumor occurrence included the middle third of the dorsal tongue, ventral tongue, buccal mucosa, retromolar pad and lingual aspect of the alveolar process of the anterior mandible. Microscopically, the lesion presents as a circumscribed mass of dense lamellar bone [13-20].

Most sufferers were asymptomatic but a few may present with globus pharyngeus, gagging, nausea, eating disturbances and even upper airway obstruction [21]. Pathogenic theories including (1) an embryologically developmental abnormality [12], (2) thyroid remnant transformation [22] and (3) traumatically reactive metaplasia [23] have been suggested in the literature. However, the first one is the most widely accepted because it is able to explain why the lesion usually occurs near the foramen cecum. It has been suggested that some pluripotential cells from primitive mesenchymal tissue are accidentally embedded near foramen cecum as the first, second and third branchial arches fuse to form the embryological tongue; these uninvited cells then differentiate into osteoblasts, and grow to form an osseous tumor-like lesion after birth.

In any case, this osseous tumor-like lesion is quite benign, slow growing and of unknown etiology. No malignant transformation has been reported. Surgical excision is the only method available to cure it and make a full pathological diagnosis, such as was the case with the twelve patients presented in the eight Taiwanese case reports [13-20]; Non-surgical management with a radiological diagnosis has been suggested for those patients whose lesions are less suitable to surgery, whose symptoms are readily controlled by conservative treatment [24], and whose lesions are asymptomatic, such as is the case with our patient. Therefore, a radiological demonstration is enough to diagnose a lingual osseous tumor-like lesion, and surgical excision with pathological diagnosis should be unnecessary for an asymptomatic incidental lingual osseous lesion such as the one described here.

<table>
<thead>
<tr>
<th>No</th>
<th>Age/Gender</th>
<th>Characteristic, size and location</th>
<th>Author (publishing year)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>30/F</td>
<td>Sessile, 0.7 × 0.5 × 0.3 cm, on right tongue base, posterior to circumvallate papillae</td>
<td>Wang et al (1993) [13]</td>
</tr>
<tr>
<td>2</td>
<td>27/F</td>
<td>Pedunculated, 0.8 × 0.8 × 0.5 cm, at foramen cecum</td>
<td>Lee (1994) [14]</td>
</tr>
<tr>
<td>3</td>
<td>39/F</td>
<td>Pedunculated, 1 × 0.6 × 0.6 cm, at foramen cecum</td>
<td>Lee (1994) [14]</td>
</tr>
<tr>
<td>4</td>
<td>15/F</td>
<td>Pedunculated, 1.5 cm in diameter, on dorsal tongue, anterior to circumvallate papillae</td>
<td>Weng et al (1996) [15]</td>
</tr>
<tr>
<td>5</td>
<td>21/F</td>
<td>Pedunculated, 1.2 × 0.8 × 0.5 cm, on midline dorsal tongue, anterior to circumvallate papillae</td>
<td>Lin et al (1998) [16]</td>
</tr>
<tr>
<td>6</td>
<td>45/M</td>
<td>Sessile, 2.0 × 2.0 × 1.5 cm, on right buccal mucosa</td>
<td>Lin et al (1998) [16]</td>
</tr>
<tr>
<td>7</td>
<td>22/M</td>
<td>Sessile, 1 × 1.0 cm, on midline tongue base</td>
<td>Huang et al (1999) [17]</td>
</tr>
<tr>
<td>8</td>
<td>25/M</td>
<td>Pedunculated, 1 × 0.5 cm, on right tongue base</td>
<td>Huang et al (1999) [17]</td>
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<tr>
<td>9</td>
<td>22/F</td>
<td>Pedunculated, 0.5 cm in diameter, on midline dorsal tongue near foramen cecum</td>
<td>Chu et al (2000) [18]</td>
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<td>10</td>
<td>24/M</td>
<td>Pedunculated, 1.5 × 1.5 × 1 cm, at foramen cecum</td>
<td>Su et al (2007) [19]</td>
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<td>11</td>
<td>47/F</td>
<td>Protruding, 1 × 0.5 × 0.4 cm, at the right tongue base near foramen cecum</td>
<td>Liu et al (2010) [20]</td>
</tr>
<tr>
<td>12</td>
<td>31/M</td>
<td>Exophytic, 0.7 × 0.5 × 0.5 cm, at right tongue base, posterior to the circumvallate papillae</td>
<td>Liu et al (2010) [20]</td>
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REFERENCES