

# Lingual Osseous Choristoma Causing Odynophagia in a Young Female

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A 37-year-old female was referred to our clinic for evaluation of an enlarged circumvallate papilla. The patient had seen her primary care physician with 3 months of symptoms of odynophagia, globus sensation, a sore throat, and hoarseness, associated with eating greasy foods. She denied unintentional weight loss or otalgia. She was a nonsmoker and used alcohol on rare occasion. There was no family history of head and neck cancer. No lymphadenopathy was appreciated on physical examination. At that time, she had an enlarged circumvallate papilla on oral examination. A clinical diagnosis of gastroesophageal reflux disease was made and she was started on ranitidine and omeprazole. She was then subsequently referred to our Ear Nose and Throat clinic for further evaluation of the enlarged circumvallate papilla. A fiberoptic laryngoscopy confirmed the presence of enlarged circumvallate papilla and short-interval follow-up was planned to determine whether this resolved after the initiation of reflux medications. Two months later, she was compliant with the medications, and most of her symptoms improved, although the odynophagia did not resolve. A repeat transnasal fiberoptic laryngoscopy demonstrated a decrease in size of the enlarged circumvallate papillae and a new midline lesion protruding from the tongue base (Figure 1). A thyroid ultrasound was unremarkable. An excisional biopsy using direct laryngoscopy demonstrated a hard, pedunculated nodule attached to the surface of the tongue base by a thin strip of mucosa. Gross pathologic examination revealed a single 0.7 × 0.4 × 0.3 cm tan-white calcified soft tissue lesion. Histologic examination showed squamous mucosa with underlying metaplastic bone (Figure 2), confirming the diagnosis of lingual osseous choristoma. To date, 26 months postoperatively, the patient has experienced neither symptom nor disease recurrence. The enlarged circumvallate papilla has resolved.

Lingual osseous choristomas are benign neoplasms or tumor-like masses originating from normal osteogenic tissue in an ectopic soft tissue location.<sup>1,2</sup> They are extremely rare, with only 69 cases reported in the literature.<sup>3</sup> Most occur in the second and third decades of life and are 4 times more frequent in females. Patients typically present with an asymptomatic

tongue mass (40%) or globus sensation (26%).<sup>4</sup> They are usually subcentimeter, hard, pedunculated, and calcified lesions, most commonly of the posterior two-thirds of the tongue near the foramen cecum. The mass may be present for years prior to diagnosis,<sup>5</sup> as size appears unrelated to symptom severity.<sup>4,6</sup>

Two main theorems exist to explain the etiology; developmental<sup>7,8</sup> and reactive or posttraumatic.<sup>7</sup> During embryologic development, the first and third branchial arches join to form the tongue, and via endochondral ossification, the incus, malleus, and hyoid bone. As lingual osteomas are commonly located at the posterior midline, the developmental theory seems plausible. The most common histological features of lingual osseous choristomas are mature bone with all the components covered by normal stratified epithelium and no inflammatory cells.<sup>6,9</sup> This also supports the developmental theory. However, this theory does not explain the other potential locations of lingual osseous choristomas. The traumatic theory suggests that these lesions may be a consequence of reactive ossification. It is thought that pluripotent or ectopic mesenchymal cells are likely present at the location, and when stimulated by chronic trauma such as swallowing,<sup>10</sup> form osseous, or cartilaginous components. When traumatic in origin, histology reveals irregular areas of ossification lacking normal bone architecture, as well as inflammatory cells and cartilage. It is most likely that the true etiology is multifactorial, or, that the etiology is different depending on location. It is also possible

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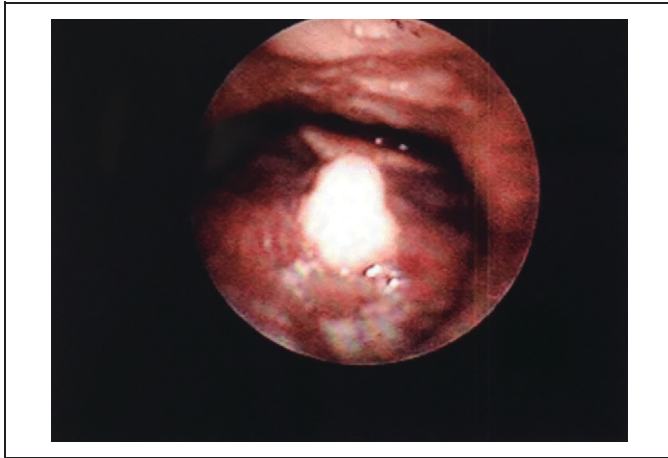
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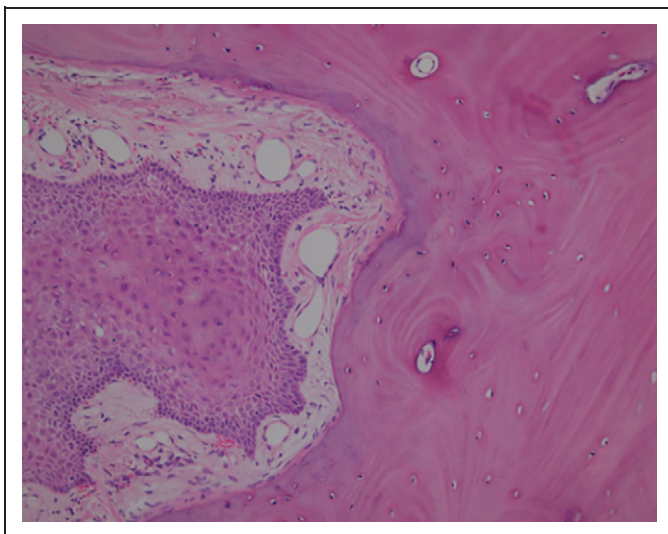
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**Figure 1.** Photograph of the tongue base lesion seen on laryngoscopy.



**Figure 2.** Histologic section of the base of tongue lesion showing squamous mucosa with underlying metaplastic bone (hematoxylin and eosin stain,  $\times 20$  magnification).

that repeated, prolonged trauma causes progressive maturation and ossification and that there is a spectrum of ossified tongue lesions with osseous choristomas being the most mature.<sup>11</sup> It is generally thought that lesions of the posterior midline tongue are developmental in origin, whereas those on the anterior tongue or buccal mucosa are posttraumatic.<sup>12</sup> In our patient, pathologic examination revealed normal mature bone without chronic inflammation, lending support to the developmental theory for the formation of her posterior midline lesion.

Given that lingual osseous choristomas are most commonly located in the posterior midline at the foramen cecum, it is crucial to exclude a lingual thyroid. Other differentials for a posterior tongue mass include a thyroglossal duct cyst and benign (hemangioma, hamartoma, teratoma) and malignant tumors (sarcomas, salivary gland tumors, squamous cell carcinoma). Rarely, an ingested fish or animal bone which has

become impacted and covered by granuloma formation<sup>13</sup> can mimic a lingual choristoma.

Diagnosis is primarily clinical and laryngoscopy provides better visualization. Imaging studies may be useful prior to operative intervention; however, these are not routine. On magnetic resonance imaging, osseous choristomas are low signal intensity lesions on T1- and T2-weighted images owing to their ossified and calcified nature.<sup>14</sup> Surgical excision is the most common treatment strategy,<sup>5</sup> although potassium titanyl phosphate laser therapy has also been attempted.<sup>15</sup> There are no reported cases of malignant transformation or postoperative recurrence,<sup>5</sup> thus surgical resection appears to be curative. Our patient was successfully managed with surgical excision and there is no evidence of postoperative recurrence. Pathology revealed benign bony tissue. With regard to the enlarged circumvallate papillae, no clear etiology was found, and it resolved after initiation of reflux medications and removal of the osseous choristoma. It is possible that it was secondary to irritation from reflux or the osseous choristoma; however, it might also have been a coincidental finding.

#### Authors' Note

Informed consent for publication of their clinical details and/or clinical images was obtained from the patient. The authors declare their responsibility for the content of this publication. All authors contributed to the conception and content of the article and approved the final version as well as for submission.

#### Authors' Note

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