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Case Report

Osseous Choristoma in Child with Strong Vomiting Reflex

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Abstract

Osseous lesions within soft tissue such as the tongue are extremely rare. Here, we report an osseous choristoma on the posterior portion of the tongue in a patient with a strong vomiting reflex. The patient was an 11-year-old boy who presented with the chief complaint of swelling on the posterior portion of the tongue. A pedunculated tumor 8-mm in diameter with distinct borders was observed slightly to the right of the midline of the dorsum of the tongue and slightly anterior to the circumvallate papillae. The clinical diagnosis was a right lingual circumvallate papilla fibroma. A further examination conducted under general anesthesia in July 2012 confirmed a pedunculated and solid mass in the area of the circumvallate papillae. As these results suggested a benign tumor, the mass was resected. Histopathological findings on harvested bone and fibrous connective tissue covered with a layer of squamous cells led to a diagnosis of osteoma. At 18 months postoperatively, there were no signs of recurrence.

Key words: Osseous choristoma — Tongue — Circumvallate papillae

Introduction

Lesions between the dorsum and the base of the tongue occupy the border zone between otolaryngology and dentistry, and therefore involve a greater range of medical fields than other sites in the oral cavity. Moreover, osseous lesions in soft tissue such as the tongue are extremely rare, and as they are frequently located in the posterior portion of the tongue, examination can often be made difficult by the vomiting reflex.

Here we report an osseous choristoma that had formed in the posterior portion of the tongue in an 11-year-old boy who experienced pronunciation and swallowing difficulties and which proved difficult to examine due to a strong vomiting reflex. We also provide a brief review of the literature.

Case

Patient: 11-year-old boy.

Chief complaint: Mass on posterior dorsum of the tongue. Difficulty in swallowing and

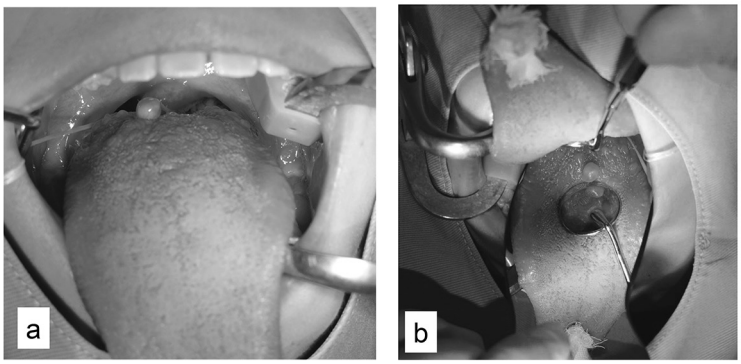

Fig. 1 Oral cavity under general anesthesia

a: Front, b: Mirror image.
Pedunculated mass 8 mm in diameter with distinct borders was observed slightly right of midline and slightly anterior to circumvallate papillae. Tumor was healthy mucosal color and had smooth surface.

pronouncing words.

First visit: May 2012.

Current medical history: The patient became aware of a mass on the posterior portion of the tongue in 2007. He was examined at a Department of Internal Medicine in 2009, after which he was placed under observation. The patient became aware of swallowing and pronunciation difficulties from April 2012 onward and consulted a local dentist, after which he was referred to our department for a more detailed examination and treatment.

Previous medical history: Nothing of note.

Present illness:

Facial appearance; Symmetrical, with good complexion.

Oral cavity; A pedunculated tumor 8-mm in diameter with distinct borders was observed slightly right of the midline and slightly anterior to the circumvallate papillae. The area had a healthy mucosal color and the surface was smooth. A single solid mass was detected upon palpation (Fig. 1).

MRI findings; Both T1 weighted imaging (WI) and T2WI (fat saturation) revealed an oval no-signal area measuring $7 \times 6 \times 8$ mm on the right side of the dorsum of the tongue (Fig. 2). The inside was uniform and it had clear boundaries. No signs of invasion were observed, and it was judged to be bone or calcified tissue.

Clinical diagnosis; Right lingual circumvallate papilla fibroma.

Treatment and cure: The mass was extremely difficult to examine visually or by palpation due to a strong vomiting reflex, so magnetic resonance imaging (MRI) was conducted instead and intraoral examination and treatment under general anesthesia planned. Subsequently, a first examination by palpation in July 2012 under general anesthesia confirmed the presence of a solid, pedunculated mass in the circumvallate papillae. The border with the surrounding tissue was distinct, no induration of the base was observed, and no solid mass was palpable in the stem. This suggested a benign tumor, so the mass was resected. This involved injecting local anesthetic into the base of the tumor and resecting the stem base with a scalpel. The tumor was easily resected *en bloc*. Due to a strong vomiting reflex, one-stitch suturing was performed with 4-0 absorbent stitches (Fig. 3-a).

The resected lesion, which measured 6×8 mm, was uneven, of an elastic hardness, and had a smooth surface covered with a partially white mucous membrane (Fig. 3-b).

Histopathological findings: The tumor was

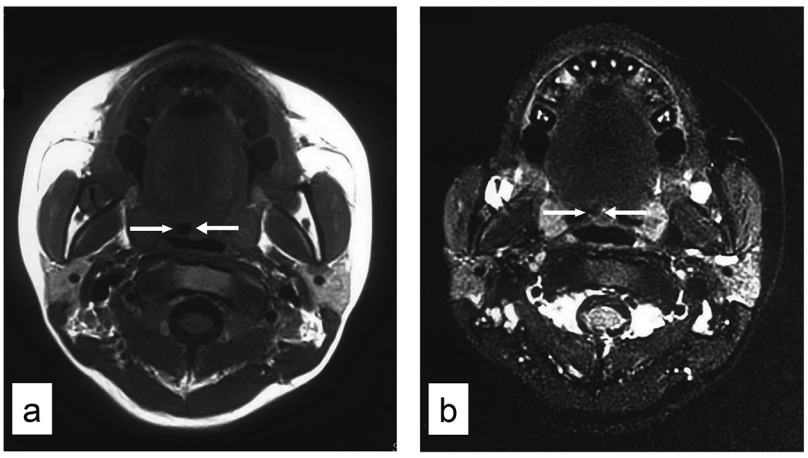

Fig. 2 Preoperative MRI

a: T1WI, b: T2WI (fat saturation).
Both T1WI and T2WI (fat saturation) revealed oval no-signal region measuring 7×6×8 mm on right side of dorsum of tongue.

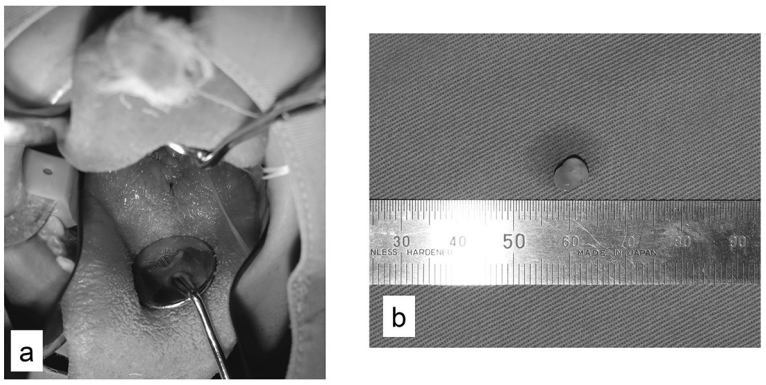

Fig. 3 Post-resection oval cavity

a: Mirror image. Tumor was easily resected *en bloc*. Due to strong vomiting reflex, one-stich suturing with 4-0 absorbent stitches was performed.
b: Resected specimen. Lesion, which measured 6×8 mm, was uneven and had an elastic hardness and smooth surface covered in partially white mucous membrane.

composed of bone tissue and fibrous connective tissue covered in a layer of stratified squamous epithelium (Fig. 4). Bone tissue with a lamellar structure containing bone cells within the lacunae was growing into a mass within the subepithelial connective tissue. Few bone marrow cells were present, however, and imaging indicated compact bone. The stratified squamous epithelium was contiguous with the tongue tissue, and the borderline between the two was unclear. These abnormalities led to a diagnosis of osteoma.

Postoperative course: The postoperative course was favorable and there was no recurrence at 18 months postoperatively. The patient's subjective symptoms of swallowing and pronunciation difficulties have also improved.

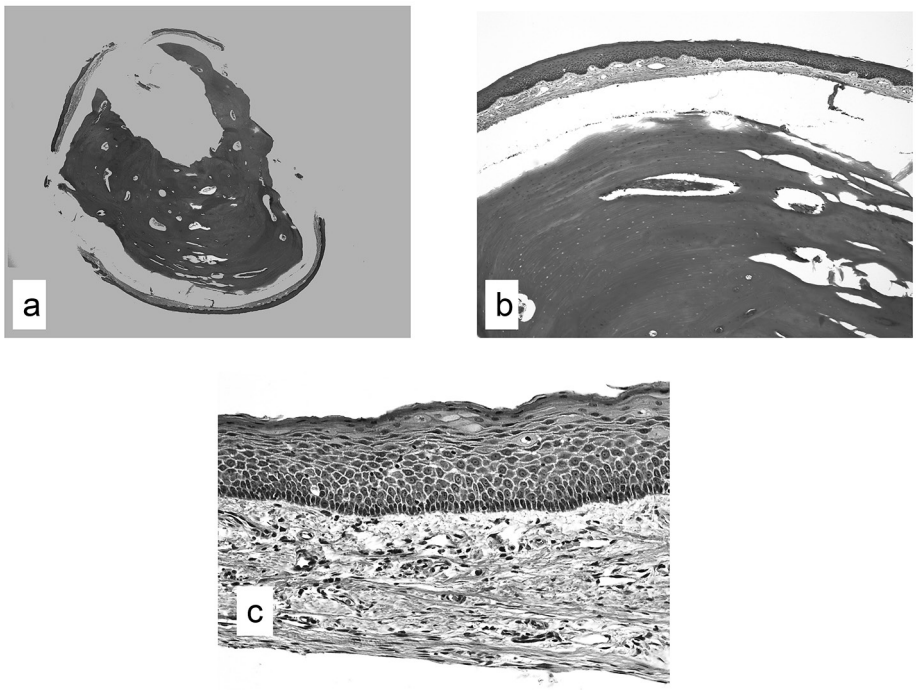

Fig. 4 H-E staining

a: Low-power (H-E staining). Tumor consisted of stratified squamous epithelium and bone tissue.
b: Middle expansion (H-E staining). Tumor was composed of bone tissue and fibrous connective tissue covered in layer of squamous cells. Bone tissue with lamellar structure containing bone cells within lacunae was growing into mass within subepithelial connective tissue; bone marrow cells exhibited poor compact bone image.
c: High-power (H-E staining). No thyroid tissue was found in tegumentary epithelium.

Discussion

In 1913, Monserrat[21] was the first to report a bone tumor forming in the tongue. Bone tumors forming within soft tissue separate from the original tissue and requiring differentiation from osteomas and hamartomas were first referred to as osseous choristomas by Krolls *et al.*[15] in 1971. In the present case, the tumor was histopathologically no different from an osteoma and had formed within soft tissue separate from the original tissue; thus, the clinical diagnosis was an osseous choristoma. Various theories have been raised as to the cause of this disease. The branchial arch persistence theory suggests that they arise from residual undifferentiated mesenchymal cells in the foramen cecum in the fetus later ossifying as they undergo differentiation. The lingual thyroid ossification theory, on the other hand, suggests that they arise from thyroid gland tissue developing in the foramen cecum in the fetus. Here, we believed that the theory of residual tissue in the pharyngeal arches was the most likely candidate, as the tumor was superficial, the site of occurrence was near the union of the first, second, and third branchial arches, no thyroid tissue was seen in the resected specimens, and the patient was young.

Since a report by Muta and Ogata[22] in 1938, to date 61 cases of osseous tumor have been reported in Japan[14]. Only 46 cases in the literature, however, have specified details such as subjective symptoms and site of occurrence[1,2,4–14,16–20,22–40,42–44]. Worldwide, 34 cases

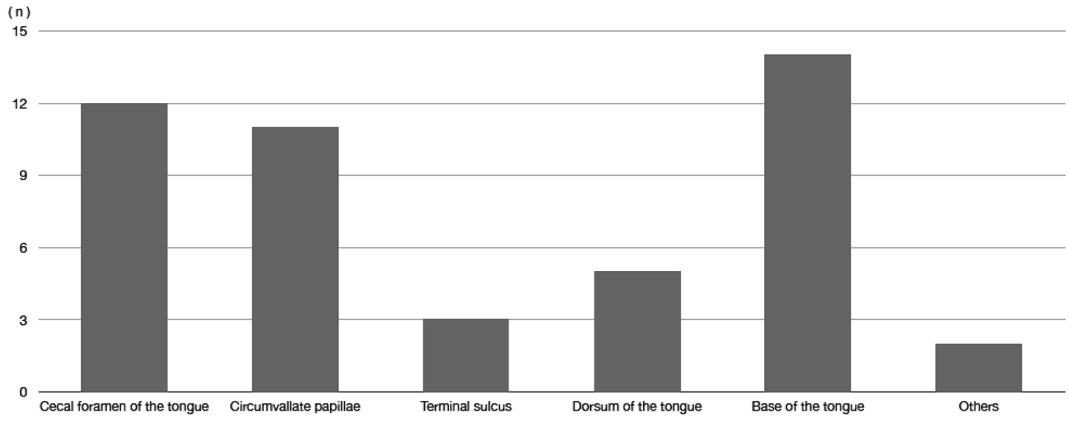

Fig. 5 Sites of occurrence

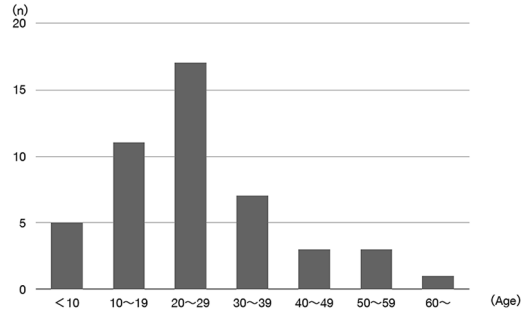

Fig. 6 Age

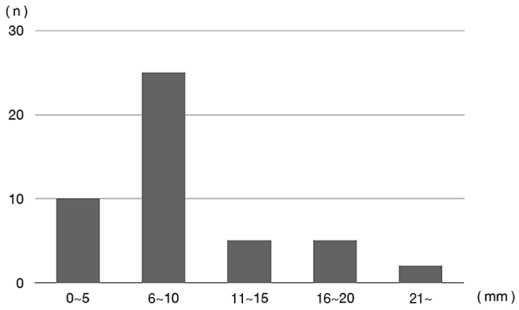

Fig. 7 Size

were confirmed between 1996 and 2006[3]. In this report, we discuss only the 46 cases of intraoral choristoma with identified subjective symptoms and site of occurrence, and compare the presence or absence of subjective symptoms and the relationship between site of occurrence and subjective symptoms with those in the present case.

The departments responsible for treatment in these cases included the Department of Oral and Maxillofacial Surgery and Department of Otolaryngology. Many of these patients visited the Department of Oral and Maxillofacial Surgery (64%) before attending the Department of Otolaryngology (32%) or Dentistry (4%) for treatment. The most common sites of occurrence were, in order of frequency, the midline of the base of the tongue, the cecal foramen of the tongue, the circumvallate papillae, the dorsum of the tongue, and the terminal sulcus of the tongue (Fig. 5). The male-to-female ratio was 1:2–3, with female cases more common[4,14,38]. However, the incidence in males appears to be gradually increasing. Among the total number of patients, 30% were male and 70% female. Young patients under the age of 30 years were common (Fig. 6), with tumors often less than 10 mm in size[38,39] (Fig. 7). However, many adult cases have been observed worldwide, and young cases are rare[3]. The tumor in the present case measured 6×8 mm, the most common size in the literature.

Due to the patient having a strong vomiting reflex performing a visual inspection was difficult. The results of a visual inspection

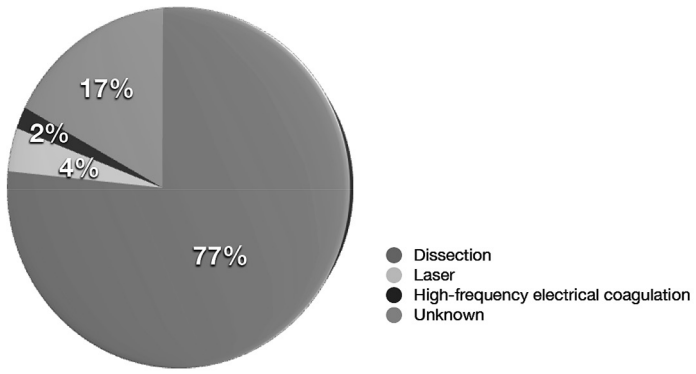

Fig. 8 Resection method

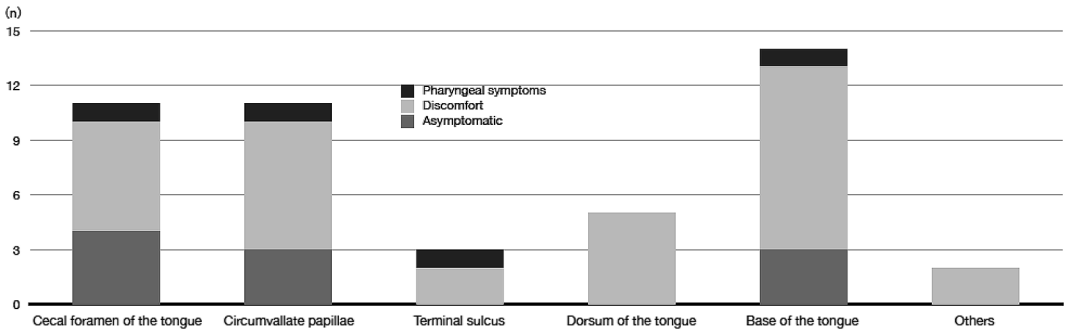

Fig. 9 Sites of occurrence and subjective symptoms

suggested a fibroma. However, we could not rule out the possibility of heterotopic hard tissue disease. Indeed, the MRI findings strongly indicated hard tissue disease and, despite difficulties with palpation, we were able to sufficiently predict the contents. Reports of postoperative MRI examination are only few. However, we believe that MRI offers a useful method of examination in such diseases, and with only minimal physical impact.

Surgical resection is often chosen as the treatment method for this disease. Resection can be performed with a scalpel, laser, or electronic scalpel, depending on the facility (Fig. 8). Recurrence is uncommon and no recurrence has been observed in the present case at 18 months postoperatively.

Many patients complained of subjective symptoms such as discomfort or were asymptomatic, whereas only few exhibited pharyngeal symptoms. Some patients complained of pain and some were aware of the tumor increasing in size. Apart from the present case, the only other example we could find of a patient presenting with swallowing difficulties was that reported by Muta and Ogata[22]. Thus, patients complaining of swallowing and pronunciation difficulties such as in the present case are rare, suggesting that the strong vomiting reflex in the present patient exacerbated their subjective symptoms. This suggests functional impairment in some cases.

In terms of the relationship between site of occurrence and subjective symptoms, patients who complained of clear pharyngeal symptoms tended to have developed a tumor pos-

terior to the circumvallate papillae (Fig. 9). In other countries, vomiting was reported as a subjective symptom[41]. In one patient presenting with swallowing difficulties, the site was in the vicinity of the circumvallate papillae, while in another patient complaining of pharyngalgia it was in the foramen cecum. In one case where the increase in the size of the tumor was noticed, the tumor was located in the area of the circumvallate papillae. The site of occurrence in the present case was near the circumvallate papillae, which is presumed to be a site where pharyngeal symptoms are easily felt. Clinical resection was chosen as it was feared that if left in place the tumor would grow back from the circumvallate papillae and cause further problems with function.

Few reports have described patients presenting with the complaint of poor function. The present patient became aware of swallowing and pronunciation difficulties. Postoperatively, however, an improvement was experienced in subjective symptoms. Is believed that such patients are generally able to subjectively notice oncogenic change through subjective symptoms and an increased vomiting reflex.

Conclusions

We have reported an osseous choristoma located near the right circumvallate papillae in an 11-year-old boy who became aware of swallowing and pronunciation difficulties and which presented difficulties in diagnosing due to a strong vomiting reflex. We have also included a brief review of the literature.

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