Case Report

Tongue Osseous Choristoma in an 11-Year-Old Female: A Case Report and Literature Review Focusing on Pediatric Cases

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Osseous choristoma is an uncommon benign lesion characterized by the presence of ectopic mature bone within soft tissue. In most cases, these lesions occur on the dorsum of the tongue in patients in their third and fourth decades of life. This article describes a case of lingual osseous choristoma in a pediatric patient. An eleven-year-old girl with a lingual mass was referred to our hospital from a dental clinic. Total excisional biopsy and histological examination were performed, and osseous choristoma was diagnosed. The postoperative course was uneventful with no signs of recurrence during the 12 months after surgery. Moreover, a literature review focusing on pediatric cases with lingual osseous choristoma was performed to know the etiology, clinicopathological characteristics, and course of treatment of the lesion.

1. Introduction

Choristoma is defined as a tumor-like lesion that is composed of normal tissue in an abnormal location. In 1971, Krolls et al. proposed the term “osseous choristoma” for soft tissue osteoma in the head and neck region [1], and this term has been widely used since. Osseous choristoma is a rare benign lesion characterized by the presence of ectopic mature bone within soft tissue and is more often composed of bone and cartilage [2]. Lingual osseous choristoma is a rather rare entity with less than 100 reported cases in the literature [3]. The pathogenesis of these lesions has remained unexplained [4]. Most cases of intraoral osseous choristoma occur in the tongue (especially its dorsal surface) [2]. Most patients with lingual osseous choristoma are women in their third or fourth decade of life [2]. These lesions are considered self-limiting in their growth. On oral examination, they frequently appear as painless, pedunculated nodules on the tongue that are firm on palpation [5]. Lingual masses can include osseous choristoma or other lesions such as fibroma, papilloma, pyogenic granulomas, squamous cell carcinomas, or hemangiomas [6]. Even though some patients may be asymptomatic, a wide array of symptoms, including gagging, dysphagia, foreign body sensation, throat irritation, discomfort, and pain, have been reported [7]. Physical examination and diagnostic imaging may assist in identifying the mass; however, a definitive diagnosis requires histologic examination. The microscopic features of osseous choristoma include a well-circumscribed mass of viable lamellar bone with haversian canals, a well-developed mass of mature viable cartilage, or a mixture of bone and cartilage surrounded by dense fibrous connective tissue with thin stratified squamous epithelium. Only a few pediatric patients with lingual osseous choristoma have been reported so far [4]. On the other hand, no previous reports of lingual osseous choristoma have highlighted the features of pediatric patients. This report is aimed at presenting another case of lingual osseous choristoma in a pediatric patient and at reviewing the relevant literature focusing on pediatric cases. A thorough literature search was carried out on PubMed and Google Scholar.
using search terms like “osseous choristoma,” “soft tissue osteoma,” and “lingual” or “tongue.”

2. Case Presentation

An 11-year-old Japanese girl told her dentist about a mass in her tongue and was referred to our hospital. She had noticed an asymptomatic nodule at the dorsum of the tongue. However, the fear of being diagnosed with a malignant condition prevented her from consulting a doctor, at least for a while. She had been aware of its existence for 2–3 years before her first visit. She was diagnosed with pneumonia at the age of one year but had no other remarkable medical history. She was not on any long-term medications. Her clinical examination revealed a pedunculated mass covered with normal mucosa in the tongue’s posterior portion (Figure 1). The lesion was approximately 7 mm in diameter. Although the lesion was asymptomatic and clinically diagnosed as a benign soft tissue tumor, the patient and her parents were concerned about the possibility of malignancy. A total excisional biopsy was thus performed under general anesthesia. Our patient’s lesion was composed of mature bone tissue surrounded by fibrous stroma and lined by normal squamous epithelium. This lesion was regarded as ectopic bone tissue localized far away from the maxilla-mandibular bone, and the histological diagnosis of osseous choristoma was made microscopically (Figure 2). Since the pathological specimen’s preparation required the resected sample’s decalcification, the final diagnosis could not be determined until ~30 days postsurgery, when the histological diagnosis was revealed as osseous choristoma. This waiting period was difficult for the patient and her family. Twelve months postoperatively, no symptoms of recurrence have been observed.

3. Discussion

Lingual osseous choristoma is rare among pediatric patients. In this manuscript, we present another pediatric case of lingual osseous choristoma and review the relevant literature. In Japan, most children attend pediatric clinics until the age of ~12 years. Thus, we focused on patients below the age of 13 years with osseous choristoma in our literature search. To our knowledge, in the literature, 62 cases have been described on patients above the age of 13 years (Table 1) [1, 3, 6, 8–45]. On the other hand, 16 cases have been described in children below the age of 13 years (Table 2) [1, 2, 4, 6, 31, 44, 46–55]. On the other hand, only one pediatric case with intraoral nonlingual osseous choristoma was found [34]. We could not detect the crucial differences in clinicopathological features between pediatric cases and the others. We summarized the characteristics of the 17 cases with pediatric lingual osseous choristoma including our case in Table 2. Most pediatric patients with lingual osseous choristoma are females (4 males, 13 females). Although these findings are consistent with previous reports [4, 54], we could not identify the reason for the sexual predisposition. The patients’ ages ranged from 5 years to 11 years (mean 9.3 years, median 10 years using Excel function). It has been demonstrated that most of the lesions develop as symptomless 3–50 mm masses located in the tongue’s posterior third in the area of circumvallate papillae or close to the foramen caecum [4, 54]. The findings reported in our manuscript are in line with previous reports. It has been reported that dysphagia, a gapping sensation, pain, vomiting reflex, and nausea are the most frequent symptoms of this condition [4]. Five patients had a history of these symptoms (29.4%). Moreover, a systematic review reported a correlation...
Table 1: Reports of cases of intraoral osseous choristomas of the tongue on >13-year-old patients.

| Year | Author | Pathogenesis | Age (y) | Sex | Location on tongue | Size | Symptom | Duration | Course of discovered events | Diagnostic imaging | Preoperative diagnosis | Local recurrence | Follow-up | Reference number |
|------|--------|--------------|---------|-----|-------------------|------|---------|----------|----------------------------|-------------------|----------------------|-----------------|-----------|-----------------|
| 1950 | Breckenridge and Lukens | Developmental malformation theory | 23 | F | Right anterior 2/3 | 1 cmØ | None | Un. | Un. | None | Fibroma | Un. | Un. | 8 |
| 1956 | Peimer et al. | Developmental malformation theory | 27 | F | Left anterior 2/3 | 0.7 × 0.6 × 0.3 cm | Gagging, foreign body sensation | 5 months | Un. | None | Fibroma | Un. | Un. | 9 |
| 1967 | Cataldo et al. | Developmental malformation theory | 39 | F | Posterior tongue | 1 cmØ | None | 4 months | Physical examination | None | Un. | Un. | Un. | 10 |
| 1968 | Jahnke and Daly | Not trauma, developmental malformation theory | 22 | F | Posterior to CP | 1.3 × 0.8 × 0.7 cm | Lump | 13 years | Asymptomatic | None | Un. | Un. | Un. | 11 |
| 1968 | Begel et al. | Developmental malformation theory | 22 | F | Area of CP | 1 × 0.5 cm | Dysphagia | 2 years | Slowly getting bigger | None | Fibroma | Un. | Un. | 12 |
| 1968 | Kaye | | 26 | F | Base of the tongue | 1 × 1 cm | Lump | Childhood | Slowly getting bigger | Un. | Un. | Un. | Un. | 13 |
| 1971 | Krolls et al. | | 22 | F | Anterior to CP | 0.75 cmØ | None | 2 years | Un. | None | Papilloma | Un. | Un. | 1 |
| 1971 | Krolls et al. | | 23 | M | Area of CP | 0.5 × 0.5 × 0.5 cm | Un. | Un. | Un. | None | Hyperplast papilla | Un. | Un. | 1 |
| 1971 | Krolls et al. | | 23 | M | Area of FC | Un. | Un. | Un. | None | Papilloma | Un. | Un. | 1 |
| 1971 | Krolls et al. | | 25 | F | Posterior tongue | 0.5 cmØ | Un. | 4 months | Un. | None | Fibroma | Un. | Un. | 1 |
| 1971 | Krolls et al. | | 39 | M | Area of CP | 0.6 × 0.6 cm | None | Un. | Un. | None | Hyperplast papilla | Un. | Un. | 1 |
| 1971 | Krolls et al. | | 73 | M | Posterior tongue | Un. | Gagging | Several years | Un. | None | Papilloma | Un. | Un. | 1 |
| 1971 | Goldberg et al. | | 65 | M | Lateral border | 1 cmØ | None | Un. | Un. | Un. | Un. | Un. | Un. | 14 |
| 1975 | McClendon | | 15 | F | Area of FC | 1.4 × 0.6 × 0.5 cm | Lump | None | Physical examination | Thyroid scan: failed to show ectopic thyroid tissue, a thyroid scintigram: normal | Lingual thyroid | Un. | Un. | 15 |
| 1975 | McClendon | | 20 | M | Posterior tongue | 0.7 cmØ | None | None | Physical examination | None | Un. | Un. | Un. | 15 |
| 1976 | Engel and Cherrick | Developmental malformation theory | 31 | M | Mid third right border | 2 cmØ | Lump | 3 years | Oral cavity examination | Dental X-P | Un. | No | 2 years | 16 |
| 1979 | Sugita et al. | | 29 | F | Area of FC | 0.8 × 0.8 × 0.5 cm | None | 12 months | Slowly getting bigger | Dental X-P | Un. | No | 6 months | 17 |
| Year | Author          | Pathogenesis                                      | Age (y) | Sex | Location on tongue | Size (cm) | Symptom          | Duration | Course of discovered events | Diagnostic imaging | Preoperative diagnosis | Local recurrence | Follow-up | Reference number |
|------|-----------------|---------------------------------------------------|---------|-----|--------------------|-----------|------------------|----------|-----------------------------|-------------------|----------------------|-------------------|-----------|------------------|
| 1981 | Ohno et al.     | Un.                                               | 44      | M   | Posterior to FC    | 0.4 × 0.8 | Un.              | 6 years  | Slowly getting bigger       | Dental X-P         | Benign tumor         | Un.               | Un.       | 18               |
| 1981 | Sato et al.     | Un.                                               | 14      | F   | Anterior to CP     | 0.4 cmØ  | Lump             | 4 years  | Slowly getting bigger       | None              | None                 | Un.               | Un.       | 19               |
| 1982 | Esguep et al.   | Developmental malformation theory                 | 63      | F   | Right border       | 0.5 cmØ  | Lump             | Un.      | Un.                         | None              | Un.                  | Un.               | Un.       | 20               |
| 1983 | Wasserstein et al. | Developmental malformation theory | 50      | F   | Mid third           | 1.5 × 0.75 | Lump             | 3 months | Oral cavity examination    | None              | Un.                  | Un.               | Un.       | 21               |
| 1984 | Main            | The anterior tongue: trauma or inflammation       | 54      | F   | Posterior to FC    | 1.5 cmØ  | Lump             | Childwood | Un.                         | None              | Un.                  | No 1 month       | 22        |
| 1984 | Sheridan        | Un.                                               | 20      | F   | Anterior to CP     | 1 cmØ    | Lump             | From birth | Un.                         | None              | Un.                  | Un.               | Un.       | 23               |
| 1984 | Shimono et al.  | Not trauma, developmental malformation theory     | 37      | F   | Area of FC         | 1.5 × 1.5 | Lump             | 8 years   | Un.                         | None              | Lingual thyroid      | Un.               | Un.       | 24               |
| 1984 | Shimono et al.  | Not trauma, developmental malformation theory     | 47      | F   | Posterior tongue   | 1 cmØ    | Lump             | 12 months | Slowly getting bigger       | Dental X-P         | Benign tumor         | Un.               | Un.       | 24               |
| 1985 | Weitzner        | Developmental malformation theory                 | 25      | F   | Posterior tongue   | 0.8 × 0.4 | Lump             | None      | Physical examination        | None              | Cyst                 | No 6 weeks       | 25        |
| 1985 | Weitzner        | Developmental malformation theory                 | 27      | F   | Posterior tongue   | 0.8 × 0.7 | Lump             | None      | Physical examination        | None              | None                 | No 2 weeks       | 25        |
| 1985 | Weitzner        | Developmental malformation theory                 | 52      | F   | Mid third           | 1 × 0.6  | None             | None      | Physical examination        | None              | Benign tumor         | No 2 months      | 25        |
| 1987 | Markaki et al.  | Un.                                               | 25      | F   | Posterior to FC    | 0.8 × 0.4 | Lump             | 5 months  | Asymptomatic                | The thyroid gland was normal to palpation | Un.                  | Un.               | 26               |
| 1987 | Tohill et al.   | Un.                                               | 26      | F   | Right anterior 2/3  | 0.9 × 0.9 | None             | None      | Oral cavity examination     | None              | Fibroma              | Un.               | Un.       | 27               |
| 1987 | Tohill et al.   | Un.                                               | 31      | F   | Anterior to CP     | 1 × 0.8  | None             | Examination | None                        | None              | Lingual thyroid, fibroma, salivary gland neoplasm | Un. | Un. | 27 |
| 1987 | Tohill et al.   | Un.                                               | 68      | M   | Left posterior 1/3  | 0.7 × 0.5 | None             | 2 years   | Oral cavity examination     | None              | Papilloma             | Un.               | Un.       | 27               |
| 1987 | Van Der Wal and van der Waal | Developmental malformation theory | 61      | F   | Anterior to CP     | 2 cmØ    | None             | 15 years  | Slowly getting bigger       | None              | Un.                  | No 2.5 years     | 28        |
| Year       | Author                        | Pathogenesis                              | Age (y) | Sex | Location on tongue | Size       | Symptom         | Duration | Course of discovered events | Diagnostic imaging          | Preoperative diagnosis | Local recurrence | Follow-up | Reference number |
|------------|-------------------------------|-------------------------------------------|---------|-----|--------------------|------------|-----------------|----------|-----------------------------|---------------------------|----------------------|-------------------|-----------|-----------------|
| 1988       | Cannon and Niparko           | Un.                                       | 51      | F   | Posterior tongue   | Un.        | Lump            | 20 years | Un.                         | Un.                      | Un.                  | Un.               | Un.       | 29              |
| 1989       | Bernard et al.               | Un.                                       | 21      | F   | Area of FC         | 2 cmØ      | Lump            | 12 years | Physical examination        | CT: densely ossified mass | Un.                  | No                | Un.       | 30              |
| 1993       | Ishikawa et al.              | Developmental malformation theory         | 53      | F   | Area of FC         | 0.8 cmØ    | Foreign body sensation | 3 days   | Foreign body sensation     | None                     | Benign tumor         | No                | Un.       | 31              |
| 1996       | Wei Cheong et al.            | Developmental malformation theory         | 23      | F   | Posterior to FC    | 1.5 cmØ    | Un.             | 13 years | Slowly getting bigger       | The thyroid gland was normal to palpation | Lingual thyroid, fibroma | No                | 12 months | 32              |
| 1996       | Manganaro                    |                                            | 27      | M   | Posterior tongue   | 1.0 × 0.5 cm | Gagging          | Un.       | Slowly getting bigger       | None                     | Un.                  | Un.               | Un.       | 33              |
| 1996       | Manganaro                    |                                            | 44      | M   | Posterior tongue   | 0.7 × 0.6 cm | Gagging          | Several months | Slowly getting bigger       | None                     | Un.                  | Un.               | Un.       | 33              |
| 1996       | Lin et al.                   | Not trauma, the posterior tongue: developmental malformation, other site: trauma | 21      | F   | Posterior tongue   | 1.2 cmØ    | Lump            | 5 years | Asymptomatic                | None                     | Fibroma              | No                | 4 years   | 34              |
| 1998       | Supiyaphun et al.            | Un.                                       | 19      | F   | Area of FC         | 1.1 × 0.7 cm | None            | 11 years | Un.                         | None                     | Un.                  | No                | Un.       | 6               |
| 1998       | Supiyaphun et al.            | Un.                                       | 21      | F   | Area of FC         | 1.5 × 1.3 cm | Lump            | 5 years   | Un.                         | None                     | Un.                  | No                | Un.       | 6               |
| 1998       | Supiyaphun et al.            | Un.                                       | 22      | M   | Area of FC         | 0.9 × 0.8 cm | None            | Un.       | Examination                 | None                     | Un.                  | No                | Un.       | 6               |
| 1998       | Supiyaphun et al.            | Un.                                       | 25      | F   | Area of FC         | 0.7 × 0.5 cm | Lump            | 12 months | Un.                         | None                     | Un.                  | No                | Un.       | 6               |
| 1998       | Supiyaphun et al.            | Un.                                       | 27      | F   | Area of FC         | 1.2 × 0.9 cm | None            | Un.       | Examination                 | None                     | Un.                  | No                | Un.       | 6               |
| 1998       | Supiyaphun et al.            | Un.                                       | 28      | F   | Area of FC         | 1 × 0.8 cm | Throat irritation | 4 years   | Un.                         | None                     | Un.                  | No                | Un.       | 6               |
| 1998       | Supiyaphun et al.            | Un.                                       | 35      | F   | Area of FC         | 0.7 × 0.5 cm | None            | Un.       | Examination                 | None                     | Un.                  | No                | Un.       | 6               |
| 1998       | Vered et al.                 | Un.                                       | 27      | M   | Posterior to CP    | 1 × 0.5 cm | Gagging          | None     | Slowly getting bigger       | None                     | Un.                  | Un.               | Un.       | 35              |
| 1998       | Vered et al.                 | Un.                                       | 44      | M   | Posterior to CP    | 0.7 × 0.6 cm | Gagging          | Several months | Slowly getting bigger       | None                     | Un.                  | Un.               | Un.       | 35              |
| 2007       | Benamer and Elmangoush       | Un.                                       | 14      | F   | Midline posterior 1/3 | 1 cmØ      | Gagging          | More than ten years | Painless but slowly getting bigger | None                     | Un.                  | Un.               | Un.       | 36              |
| 2007       | Demirseren and Aydin         | Not trauma                                | 28      | M   | Anterior 2/3       | 2 × 1.5 cm | Gagging          | 4 years   | Slowly getting bigger       | None                     | Pyogenic granuloma   | No                | 48 months | 37              |
| Year | Author | Pathogenesis | Age (y) | Sex | Location on tongue | Size | Symptom | Duration | Course of discovered events | Diagnostic imaging | Preoperative diagnosis | Local recurrence | Follow-up | Reference number |
|------|--------|--------------|---------|-----|-------------------|------|---------|----------|---------------------------|-----------------|---------------------|----------------|----------|-----------------|
| 2008 | Andressakis et al. | Local trauma from dentures | 72 | M | Area of CP | 1.5 × 1.0 cm | Pain, dysphagia | Several years | Asymptomatic | None | Un. | Un. | Un. | 38 |
| 2009 | Naik et al. | Not trauma | 25 | F | Posterior tongue | 1.2 × 1.1 × 0.5 cm | Lump | 5 years | Slowly getting bigger | Un. | Un. | Un. | Un. | 39 |
| 2011 | Liu et al. | Not trauma | 17 | M | Area of FC | 0.5 × 0.5 cm | Lump | Several years | Asymptomatic | None | Un. | No | Un. | 40 |
| 2016 | Adhikari et al. | Not trauma | 15 | F | Area of FC | 0.5 cmØ | Throat irritation | 12 months | Painless but gradually swelling | None | Fibroma | No | 5 months | 41 |
| 2016 | Adhikari et al. | Not trauma | 21 | F | Area of FC | 0.5 mØ | Pain | Un. | Oral cavity examination | None | Un. | No | 48 months | 41 |
| 2016 | Turan et al. | Un. | 41 | F | Posterior tongue | 1 × 0.5 cm | Throat irritation | 6 months | Examination | Ultrasonographic evaluation: normal thyroid gland | Lingual thyroid, mucocele, lingual thyroglossal duct cyst | No | 4 months | 42 |
| 2017 | Heinz et al. | Not trauma | 21 | F | Base of the tongue | 0.5 cmØ | Lump | Un. | Asymptomatic | Fiberoptic examination | Un. | No | 3 months | 43 |
| 2020 | Sun et al. | Un. | 23 | M | Base of the tongue | 0.8 × 0.7 × 0.5 cm | Lump | Un. | Un. | None | Benign tumor | Un. | Un. | 3 |
| 2020 | Sun et al. | Un. | 27 | M | Base of the tongue | 0.8 × 0.5 × 0.5 cm | Lump | 3 months | Un. | None | Benign tumor | Un. | Un. | 3 |
| 2020 | Leigh et al. | Developmental malformation theory | 37 | F | Base of the tongue | 0.7 × 0.4 × 0.3 cm | Gagging | 3 months | Physical examination | Ultrasonographic evaluation: normal thyroid gland | Un. | No | 26 months | 44 |
| 2020 | Hemmi et al. | Not trauma | 89 | M | Base of the tongue | 1 cmØ | Cough | Un. | Prolonged cough | Cervical spine CT: well-defined, rounded, high-density mass | Un. | No | 15 months | 45 |

CP: circumvallate papillae; FC: foramen caecum; M: male; F: female; CT: computed tomography; Un.: unknown.
| Year | Author          | Age (y) | Sex | Location on tongue | Size            | Symptom       | Duration   | Pathogenesis                                  | Course of discovered events | Diagnostic imaging | Preoperative diagnosis | Local recurrence | Follow-up | Reference number |
|------|----------------|---------|-----|-------------------|-----------------|---------------|------------|-----------------------------------------------|-----------------------------|--------------------|---------------------|------------------|----------|------------------|
| 1964 | Church         | 11      | F   | Area of FC        | 0.5 cmØ        | Dysphagia     | Un.        | Slowly getting bigger                         | None                        | Un.                | Un.                 | Un.              | Un.      | 46               |
| 1971 | Krolls et al.  | 9       | F   | Area of FC        | Un.             | Gagging       | 2.5 years  | None                                          | None                        | Fibroma            | Un.                 | Un.              | 1        |                  |
| 1971 | Krolls et al.  | 11      | F   | Posterior tongue | 2 cmØ          | Un.           | 12 months  | None                                          | None                        | None               | Papilloma           | Un.              | 1        |                  |
| 1977 | Busuttill      | 8       | F   | Area of CP        | Pea-sized       | Lump          | 9 months   | Slowly getting bigger                         | None                        | None               | Un.                 | Un.              | Un.      | 47               |
| 1986 | Cabbabe et al. | 5       | F   | Base of the tongue| 0.6 × 0.5 × 0.3 cm | Lump         | 2 years    | Not trauma, developmental malformation theory | Asymptomatic                | None               | Fibroma            | Un.              | Un.      | 48               |
| 1992 | Maqbool et al. | 8       | F   | Right vallecula   | 5 × 4 cm        | Dysphagia, distress | Un.     | Developmental malformation theory             | Asymptomatic                | None               | Fibroma            | No               | Un.      | 49               |
| 1993 | Ishikawa et al.| 5       | F   | Anterior to CP    | 0.3 → 0.8 cmØ  | Lump          | 1-month follow-up: 16 months                 | Asymptomatic                | None               | Fibroma            | No               | Un.      | 31               |
| 1993 | Lutcavage and Fulbright | 11 | F   | Area of FC        | 1 cmØ          | Lump          | 12 months  | Enclavement of mesenchymal cells               | Slowly getting bigger        | A thyroid scintigram: normal | Un.              | Un.      | 50               |
| 1998 | Supiyaphun et al. | 9  | F   | Area of FC        | 0.7 × 0.6 × 0.5 cm | None       | Un.        | Examination                                   | None                        | Un.                | No                  | Un.              | 6        |                  |
| 2001 | Horn et al.    | 11      | F   | Posterior tongue  | Un.             | Lump          | Un.        | Thyroid scan: failed to show ectopic thyroid tissue | Un.                        | No                 | 12 months           |                  |          |                  |
| 2014 | Gorini et al.  | 10      | F   | Area of FC        | 1 cmØ          | Lump          | Since the first months of life               | Asymptomatic                | Un.                | No                 | 12 months          | 4        |                  |
| 2014 | Yamamoto et al.| 11      | M   | Posterior to CP   | 0.8 × 0.6 cm    | Dysphagia     | 5 years    | Developmental malformation theory             | Slowly getting bigger        | MRI: T1 and T2WI (fat saturation): oval no-signal area | Papilla fibroma | No      | 52               |
| 2015 |                 | 11      | M   |                  | 1.1 × 0.9 × 0.8 cm | Un.        | Un.        | Examination                                   | Un.                        | Un.                | Un.                 |                  |          | 53               |
| Year  | Author            | Age (y) | Sex | Location on tongue | Size          | Symptom                  | Duration | Pathogenesis                                                                 | Course of discovered events                                                                 | Diagnostic imaging                                                                 | Preoperative diagnosis | Local recurrence | Follow-up | Reference number |
|-------|-------------------|---------|-----|-------------------|---------------|--------------------------|----------|-----------------------------------------------------------------------------|-------------------------------------------------------------------------------------|--------------------------------------------------------------------------------------|-----------------------|-----------------|------------|--------------|
| 2016  | Davidson et al.   | 11      | M   | Base of the tongue| Un.           | Foreign body sensation | Un.      | Un.                                                                         | Slowly getting bigger                                                              | Thyroid uptake scan: normal<br>CT: densely calcified lesion<br>Thyroid scan: failed to show ectopic thyroid tissue<br>CT: a calcified ovoid mass<br>T1- and T2-weighted images: low-signal intensity<br>Dermoscopy<br>oral cavity examination | No                    | No                | Un.     | Un.      | 2                                                                      |
| 2018  | Yoshimura et al.  | 7       | F   | Posterior tongue  | 0.5 cmØ → 0.6 cmØ | None                     | Un.      | Ectopic bone formation by secretion of BMPs                                | Slowly getting bigger                                                              | Benign tumor<br>Dermoscopy<br>oral cavity examination | No                    | 36 months | 54          |
| 2018  | Macêdo et al.     | 9       | M   | Posterior 1/3     | 2.3 × 2.0 × 0.8 cm | Symptomatic             | 3 months | Developmental malformation theory<br>oral cavity examination<br>CT: pseudotumor | Oral cavity examination<br>oral cavity examination | None<br>Papilloma | No         | 2.5 years  | 55          |
| 2021  | Present case      | 11      | F   | Posterior tongue  | 0.7 cmØ       | Lump                     | 2–3 years| Developmental malformation theory                                             | Oral cavity examination                                                                 | None                    | Papilloma     | No         | 12 months   |

CP: circumvallate papillae; FC: foramen caecum; M: male; F: female; CT: computed tomography; MRI: magnetic resonance imaging. Un.: unknown.
between these symptoms and lesion size [4]; however, another review concluded that there was no correlation between them [6]. Hemmi et al. reported an adult case of lingual osseous choristoma with prolonged cough. They concluded that the cough was due to gastroesophageal reflux disease. Regarding the correlation in pediatric cases, we could not conclude from only 5 cases (29.4%). To resolve this discordance, it is necessary to recruit more cases [45]. The follow-up period ranged from one year to three years. No evidence of spontaneous loss or malignant transformation has been reported. No case of pediatric lingual osseous choristoma showed any sign of recurrence [7], while only two recurrent nonpediatric cases of the buccal mucosa lesion were reported [56, 57]. Long et al. reported that the recurrent lesion could have arisen as a result of the surgical trauma caused by the removal of the original lesion; however, this theory could not explain the occurrence of the original lesion because the patient denied any history of trauma [56]. Besides, according to Dalkiz et al., lesions do not recur once excised and the recurrent lesion might have been caused by a new fibrotic region that underwent ossification an uncalcified lesion that subsequently ossified [57]. Although the mechanisms of recurrence remain uncertain, cases of extralingual lesions should have a longer follow-up period. Our patient’s clinical findings were consistent with previous reports. Taking into account the fact that our patient was referred from a private dental clinic, not only head and neck clinicians but also dentists should be familiar with the clinical features of this disease.

The pathogenesis of osseous choristoma is not yet known, a “developmental malformation hypothesis” and a “chronic trauma-associated reactive hypothesis” were proposed [4, 16, 41, 54, 58]. The involvement of systemic diseases has not been reported. With respect to the former, the lesion arises at the line of fusion of the first and third brachial arches between the anterior two-thirds and posterior one-third of the tongue [41]. Additionally, some researchers indicated that the lingual thyroid remnant ossification is associated with developmental malformation theory due to it occurring at the posterior tongue near the foramen cecum [12, 30]. However, no thyroid tissue was observed in the current case. With respect to the latter, on the other hand, the osseous lesion on the tongue appeared due to a reactive or posttraumatic center of ossification [41]. There were no previous cases that support the reactive hypothesis. In the current case, there was no evidence of irritational factors. Moreover, our patient’s microscopic findings showed no reactive epithelial change, including acanthosis and cell atypia, and little inflammatory cell infiltration and fibrosis were observed. From these clinicopathological features, the “developmental malformation hypothesis” seems likely to apply in this case.

Finally, it cannot be overlooked that the 30-day waiting period until the histological diagnosis of the patient’s lesion was stressful for the patient and her family. Although the reason for the long waiting time was the need to decalciﬁy the bone tissue, the psychological care we provided was insufficient. It was speculated that the information provided by imaging modalities can help reduce patients’ anxiety. Diagnostic imaging was conducted in five pediatric cases. Given that no attending surgeons considered the possibility of an osseous choristoma, we did not conduct any imaging examination in this case. An earlier study reported computed tomography (CT) images are useful for the radiological diagnosis of lingual osseous choristoma [45]. Also, Yoshimura et al. reported the usefulness of not only radiographic examination for the surgical specimen. Additionally, they proposed developing a miniaturized, ﬂexible dermatoscope that enabled the detailed examination of the whole oral cavity [54]. However, it is difficult for children to follow instructions when undergoing a CT scan, which often leads to motion artifacts [45]. It is also important to consider the effect of radiation exposure in pediatric patients. Furthermore, a case that occurred in the choroid was diagnosed using ultrasonography [59]; however, there are no reports of ultrasonography being used to diagnose lingual osseous choristoma as it might be diﬃcult to use ultrasonography on the base of the tongue. Therefore, radiographic examination of the surgical specimen might be the most useful tool in the diagnosis of lingual osseous choristoma in pediatric patients.

In conclusion, we presented a pediatric case of lingual osseous choristoma and conducted a review of the literature to identify the characteristics of pediatric cases of the lesion.

Data Availability
The data used to support the ﬁndings of this study are available from the corresponding author upon request.

Conflicts of Interest
The authors declare that there is no conﬂict of interest regarding the publication of this article.

Authors’ Contributions
S.A. was responsible for the manuscript drafting, literature search, and collection of clinical records. M.S. was responsible for the histological evaluation and the revision of the manuscript. M.A. supervised the work. All authors reviewed and accepted the ﬁnal version of the manuscript.

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