Case Report

Osseous Choristoma of the Tongue: A Review of Etiopathogenesis

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Osseous choristoma is a normal bone tissue in an ectopic position. In the oral region lingual localization occurs more frequently and the mass is generally localized on the dorsum of the tongue. Definitive diagnosis is obtained only after histopathologic examination. The etiology remains already debatable. The treatment of choice is surgical excision. In this paper we present a case of tongue osseous choristoma and a review of the literature.

1. Introduction

The term choristoma is used to describe the growth of normal tissue in an abnormal position. Choristoma of the mouth may be composed of several different tissue types. These include bone, cartilage, gastric mucosa, glial tissue, and tumor-like masses of sebaceous glands. Osseous choristoma is a well circumscribed benign growth of normal, mature osseous tissue in ectopic sites.

Osseous choristoma of the tongue is an extremely rare condition, of which only 66 cases have been reported in the literature till now. The osseous histotype is the most frequently described among choristomas. In the oral region lingual localization occurs more frequently and the mass is generally localized on the dorsum of the tongue. The etiology remains already debatable. The treatment of choice is surgical excision.

2. Case Report

A 10-year-old girl was referred to our ENT unit for a whitish 1 cm sessile swelling of the paramedian dorsum of the tongue, near foramen caecum (Figure 1). Upon palpation this not ulcerated lesion was firm. The patient did not complain of any symptom for many years. In the last two months she complained of lump. The mother of the girl reported that she noticed the little swelling since first months of life. She underwent neck ultrasound that showed a normal thyroid gland in shape and position. Although she was paucisymptomatic we proposed and performed a surgical excision of the lesion in order to obtain a histological diagnosis. Histological examination described an osseous choristoma of the tongue. The specimen was totally included for histological examination. Histologically, it appeared as a polypoid nodule of cortical bone tissue (mm 10 in maximum diameter) beneath the mucous membrane of the tongue covered by orthokeratinised squamous epithelium. The bone tissue was lamellar type with well developed haversian systems. The bone tissue was with sharply demarcated edges and in the surrounding tissue there was no inflammation or scar tissue (Figure 2).

After 1-year follow-up there is no evidence of recurrence.

3. Discussion and Review of the Literature

The term osseous choristoma was introduced by Krolls et al. in 1971 [1]. By definition it is a growth of normal tissue in an abnormal position. Choristoma of the mouth may be composed of several different tissue types. These include bone, cartilage, gastric mucosa, glial tissue, and tumor-like masses of sebaceous glands [2, 3].

Osseous choristoma of the tongue is an extremely rare condition. In the literature 66 cases have been described.
| Author                  | Age (y)/sex | Location            | Size                 | Symptom                        |
|-------------------------|-------------|---------------------|----------------------|--------------------------------|
| Cataldo et al. [4]      | 39/F        | Posterior tongue    | 1 cm Ø               | None                           |
| Begel et al. [5]        | 22/F        | Area of CP          | 1 × 0.5 cm           | Dysphagia                      |
| Jahnke and Daly [6]     | 22/F        | Posterior to CP     | 1.3 × 0.8 × 0.7 cm   | Lump                           |
| Kaye [7]                | 26/F        | Base of the tongue  | 1 × 1 cm             | Lump                           |
| Goldberg et al. [8]     | 65/M        | Lateral border      | 1 cm Ø               | None                           |
| Krolls et al. [1]       | 23/M        | Area of FC          | Un.                  | Un.                            |
|                          | 73/M        | Posterior tongue    | Un.                  | Gagging                        |
|                          | 9/F         | Area of FC          | Un.                  | Gagging                        |
|                          | 25/F        | Posterior tongue    | 0.5 cm Ø             | Un.                            |
|                          | 11/F        | Posterior tongue    | 2 cm Ø               | Un.                            |
|                          | 23/M        | Area of CP          | 0.5 × 0.5 × 0.5 cm   | None                           |
|                          | 39/M        | Area of CP          | 0.6 × 0.6 cm         | None                           |
| Singh and Doyle [9]     | 14/F        | Left border         | Un.                  | Un.                            |
|                          | 22/F        | Area of CP          | 0.5 cm Ø             | None                           |
| McClendon [10]          | 15/F        | Area of FC          | 1.4 × 0.6 × 0.5 cm   | None                           |
|                          | 20/M        | Right border        | 0.7 cm Ø             | None                           |
|                          | 46/F        | Area of FC          | 0.6 cm Ø             | None                           |
| Patel and Dane [11]     | 42/M        | Lateral border      | 1 cm Ø               | None                           |
| Engel and Cherrick [12] | 31/M        | Mid third right border | 2 cm Ø             | Lump                           |
| Busuttil [13]           | 8/F         | Left border         | Pea-sized            | Lump                           |
|                          |            | Dorsum of the root of the tongue | Un. | Un. |
| Sugita et al. [15]      | Un.         | Un.                 | Un.                  | Un.                            |
| Sato et al. [16]        | Un.         | Un.                 | Un.                  | Un.                            |
| Esguep et al. [17]      | 63/F        | Right border        | 0.5 cm Ø             | Lump                           |
| Wasserstein et al. [18] | 50/F        | Mid third           | 1.5 × 0.75 cm        | Lump                           |
| Shimono et al. [19]     | 37/F        | Area of FC          | 1.5 × 1.5 × 0.7 cm   | Lump                           |
| Main [20]               | 54/F        | Posterior to FC     | 1.5 cm Ø             | Lump                           |
| Sheridan [21]           | 20/F        | Anterior to CP      | 1 cm Ø               | Lump                           |
| Cabbabe et al. [22]     | 5/F         | Base of tongue      | 0.6 × 0.5 × 0.3 cm   | Lump                           |
| Nash et al. [23]        | 31/M        | Right border        | 2.5 cm Ø             | None                           |
| Weitzner [24]           | 52/F        | Mid third           | Small nodule         | None                           |
|                          | 25/F        | Posterior tongue    | 0.8 × 0.4 × 0.4 cm   | Lump                           |
|                          | 27/F        | Posterior tongue    | 0.8 × 0.7 × 0.3 cm   | Lump                           |
| Tohill et al. [25]      | 31/F        | Anterior to CP      | 1 × 0.8 × 0.7 cm     | None                           |
| Markaki et al. [26]     | 25/F        | Posterior to CP     | 0.8 × 0.4 × 0.3 cm   | Lump                           |
| Van Der Wal and van der Waal [27] | 31/F | Area of FC | 1 cm Ø | Lump |
| Cannon and Niparko [28] | 51/F        | Posterior tongue    | Un.                  | Lump                           |
| Bernard et al. [29]     | 21/F        | Area of FC          | 2 cm Ø               | Lump                           |
| Maqbool et al. [30]     | 8/F         | Right vallecula     | 5 × 4 cm             | Dysphagia, distress            |
| Lutcavage and Fulbright [31] | 11/F | Posterior to FC | 1 cm Ø | Lump |
| Ishikawa et al. [32]    | 53/F        | Area of FC          | 0.8 cm Ø             | Foreign body sensation         |
| Lee et al. [33]         | 35/M        | Lateral border      | Un.                  | Lump                           |
| Ngeow et al. [34]       | Un.         | Un.                 | Un.                  | Un.                            |
| Manganaro [35]          | Un.         | Un.                 | Un.                  | Un.                            |
| Vered et al. [36]       | 44/M        | Left border         | 0.7 × 0.7 × 0.6 cm   | Gagging, nausea, and dysphagia |
|                          | 27/M        | Posterior to CP     | 1 × 0.5 cm           | Pain, gagging                   |
| Supiyaphun et al. [3]   | 28/F        | Area of FC          | 1 × 0.8 × 0.6 cm     | Throat irritation               |
Table 1: Continued.

| Author                | Age (y)/sex | Location          | Size                  | Symptom              |
|-----------------------|-------------|-------------------|-----------------------|----------------------|
| 50                    | 25/F        | Area of FC        | 0.7 × 0.5 × 0.4 cm    | Lump                 |
| 51                    | 9/F         | Area of FC        | 0.7 × 0.6 × 0.5 cm    | None                 |
| 52                    | 35/F        | Area of FC        | 0.7 × 0.5 × 0.5 cm    | None                 |
| 53                    | 27/F        | Area of FC        | 1.2 × 0.9 × 0.6 cm    | None                 |
| 54                    | 21/F        | Area of FC        | 1.5 × 1.3 × 0.8 cm    | Lump                 |
| 55                    | 22/M        | Area of FC        | 0.9 × 0.8 × 0.6 cm    | None                 |
| 56                    | 19/F        | Area of FC        | 1.1 × 0.7 × 0.7 cm    | None                 |
| 57                    | Lin et al. [37] | Un. Posteriortongue | Un.                  | Un.                  |
| 58                    | Horn et al. [38] | 11/F Posteriortongue | Un.                  | Lump                 |
| 59                    | Benamer and Elmangoush [39] | 14/F Mid third | 1 cm Ø               | Lump                 |
| 60                    | Carvalho et al. [40] | 22/F Posteriortongue | 1 cm Ø               | None                 |
| 61                    | Andressakis et al. [2] | 72/M Anterior to CP | 1.5 × 1 cm           | Pain dysphagia       |
| 62                    | Naik et al. [41] | 25/F Posteriortongue | 1.2 × 1.1 × 0.5 cm   | Lump                 |
| 63                    | Chen et al. [42] | 57/F Posteriortongue | 1 cm Ø               | Lump, dysphagia, and odynophagia |
| 64                    | Spencer and Reed [43] | 11/M Posteriortongue | 1.1 × 0.9 × 0.6 cm   | None                 |
| 65                    | Lin et al. [44] | 15/M Area of FC | 0.5 × 0.5 cm         | Lump                 |
| 66                    | Qin et al. [45] | Un. Anterior to FC | Un.                  | Un.                  |
| 67                    | Present case | 10/F Anterior to FC | 1 cm Ø               | Lump                 |

(CP: circumvallate papillae, FC: foramen caecum, and Un.: unknown).

Figure 1: Paramedian dorsum of the tongue choristoma.

Figure 2: Histological examination shows mature lamella lined by mucous membrane of the tongue.

(Table 1) [1–45]. In our review the patient age ranged from five to seventy-three years (mean age: 28.7 years), with the majority of the patients being in the second or third decades of life. Choristomas of the tongue occur more frequently in women (M:F 16:44).

The most frequent affected region is the posterior third of the tongue dorsum near to the foramen caecum and circumvallate papillae. Pathogenesis of choristoma is still unknown and remains already debatable. Several theories tried to explain the pathogenesis of this disease. Some authors suggested that remnants of the undescended thyroid tissue might produce an osseous lesion but in some rare case choristoma is localized not in midline but on the border of the tongue [46]. In these cases some authors [36, 47] suggested a traumatic pathogenesis. They consider that the posterior third of the tongue is site of traumatic irritation by different lingual movement during swallowing and articulation and that frequent trauma leads to local inflammation with deposit of calcium. This theory cannot explain the formation of osseous choristoma, because this lesion contains fully developed bone with haversian system and not just calcifications.

In our opinion embryologic theory sounds true. Embryologically the tongue is a very complex structure. First and third branchial arches give rise, respectively, to the anterior two-thirds and posterior third of the tongue. It was suggested that pluripotential cells from these arches give origin to the osseous choristoma [5, 12].

Choristoma appears as a sessile or pedunculated mass usually covered by normal mucosa. The sizes of the lesions vary from 3 mm to 50 mm at their largest diameter.

Most of osseous choristoma of the tongue presents as a frequently asymptomatic swelling. The most frequent symptom is lump (46% of cases). Rarely patient complains of dysphagia (5 cases), gagging (4 cases), pain (4 cases), and nausea (1 case). Symptoms are correlated to lesion size, tumour
localization, and surrounding tissues flogosis. The differential clinical diagnosis can be also based on the tumor location. When the lesion is located on the dorsal tongue near the foramen caecum we should consider in differential diagnosis benign tumours (hemangioma, lymphangioma, teratoma, hamartoma, and leiomyoma), thyroglossal duct cyst, lingual thyroid, mucocoele, pyogenic granuloma, and malignant tumours (rhabdomyosarcoma, other sarcomas, and epidermoid carcinoma) [25]. Traumatic neuroma, neurofibroma, schwannoma, fibroma, and cartilaginous choristoma usually are located on the tongue margin. Pyogenic granuloma, mucocoele, and cartilaginous choristoma frequently involve the anterior portion of the tongue. Nevertheless definitive diagnosis is obtained only after histopathologic examination. The treatment of choice is surgical excision. Recurrence or malignant evolution has not been described.

**Conflict of Interests**

There are not present facts which may be considered as potential conflict of interests and significant financial contributions to this work. The authors wish to confirm that there is no known conflict of interests associated with this paper and there has been no significant financial support for this work that could have influenced its outcome.

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