Glomus tumor in the floor of the mouth: a case report and review of the literature

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Abstract

Background: Glomus tumors are rare benign neoplasms that usually occur in the upper and lower extremities. Oral cavity involvement is exceptionally rare, with only a few cases reported to date.

Case presentation: A 24-year-old woman with complaints of swelling in the left floor of her mouth for 6 months was referred to our institution. Her swallowing function was slightly affected; however, she did not have pain or tongue paralysis. Enhanced computed tomography revealed a 2.8 × 1.8 × 2.1 cm-sized well-defined, solid, heterogeneous nodule above the mylohyoid muscle. The mandible appeared to be uninvolved. The patient underwent surgery via an intraoral approach; histopathological examination revealed a glomus tumor. The patient has had no evidence of recurrence over 4 years of follow-up.

Conclusions: Glomus tumors should be considered when patients present with painless nodules in the floor of the mouth.

Keywords: Glomus tumor, Floor of mouth, Oral surgery

Background

The glomus body is a special arteriovenous anastomosis and functions in thermal regulation. Glomus tumors are rare, benign, mesenchymal tumors that originate from modified smooth muscle cells of the normal glomus body [1]. Glomus tumors account for only 1.6% of all soft tissue tumors and typically present as blue-red nodules (sized < 1 cm) that occur in the deep dermis or subcutis region [2]. These tumors are relatively common in the upper and lower extremities, particularly in the subungual site, but rarely occur in mucinous regions or the viscera [3]. Oral cavity involvement is exceptionally rare, with very few cases having been reported to date. Here, we present a case of an unusual glomus tumor that originated from the left floor of the mouth.

Case presentation

A 24-year-old woman with a 6-month history of swelling in the left floor of her mouth was referred to our institution. Although she experienced slight difficulty in swallowing, she did not experience pain or tongue paralysis. Her medical and family histories were unremarkable. Intraoral examination revealed a well-defined 3.5 × 3 × 2 cm-sized solid, spherical submucosal nodule adjacent to the sublingual gland; the nodule was covered with light bluish smooth mucus (Fig. 1a). The patient experienced slight pain when pressure was applied to the tumor. Mobility and sensory functions of the tongue were normal, and no lymphadenopathy in the submandibular region was detected on palpation. All relevant laboratory test results were normal. Enhanced computed tomography revealed a 2.8 × 1.8 × 2.1 cm-sized well-defined, solid, heterogeneous nodule that did not appear to involve the mandible (Fig. 1b). In addition, a three-dimensionally reconstructed image showed a nodular lesion occupying the left floor of the mouth with abundant blood flow (Fig. 1c). No enlarged lymph nodes were found in the submental or submandibular regions.

The initial clinical impression was a benign salivary gland tumor, dermoid cyst, or benign connective tissue neoplasm. The patient was scheduled for surgery via an
intraoral approach. First, an elliptical incision was made around the periphery of the sublingual gland through only the oral mucosa, and a full-thickness tissue flap was prepared along the lingual aspect of the sublingual gland. After the sublingual gland was freed from its surrounding tissue with blunt dissection, a well-circumscribed tumor without capsular extension was found beneath the body of the sublingual gland and located above the submandibular gland duct and lingual nerve. The submandibular gland duct and lingual nerve were carefully freed from the tumor surface, and the complete tumor was excised along with the sublingual gland (Fig. 2a). The tissue sample was fixed with 10% formalin and submitted for histopathological diagnosis.

Microscopically, the tumor cells were round, oval, polyhedral, or fusiform and were arranged in organoid and sheet-like patterns with vascular lumens. Most of their nuclei were small and round within an amphiphilic or slightly eosinophilic cytoplasm. Nuclear atypia was rare (Fig. 2b).

Immunohistochemistry revealed that the tumor cells yield positive results for vimentin and alpha-smooth muscle actin, but negative results for desmin, anticytokeratin (AE)1 or AE3, cluster of differentiation (CD)31 and CD34, and S-100, and exhibited a Ki-67 index of 5%. These findings were consistent with those for a glomus tumor.

After surgery, the patient had an uneventful recovery with primary healing and had no evidence of recurrence over 4 years of follow-up.

**Discussion and conclusions**

Glomus tumors are rare mesenchymal tumors that occur due to glomus body hyperplasia or hamartomatous
the tumors were located on the lip (from 10 to 85 years (median, 52 years). In most cases, Table 1. The neoplasms developed in 23 women and 14
alties of all 37 cases (including our case) are shown in
upper 10), followed by the buccal mucosa (n
= 39). The patient characteristics of all 37 cases (including our case) are shown in
Table 1. The neoplasms developed in 23 women and 14
men (male to female, 1:6:1), and patient age ranged from 10 to 85 years (median, 52 years). In most cases, the tumors were located on the lip (n = 14, lower 4, upper 10), followed by the buccal mucosa (n = 7), tongue (n = 5), hard palate (n = 6), gingiva (n = 2), maxilla (n = 1), the floor of the mouth (n = 1), and multiple locations (n = 1). Unfortunately, many of the cases documented had an unknown clinical presentation or medical history. Due to inadequate figures or illustrations in these articles, the histologic types of the tumors were not included in our table. Of the cases with available information, the size of the glomus tumor ranged from 0.3 to 4.5 cm. Some lesions were painful, and most were asymptomatic. All lesions were completely excised. Follow-up information was only available in 11 cases ranging from 2 months to 7 years; local recurrence was noted in two cases. Owing to the low incidence rate of glomus tumors in the head and neck region, accurate information on the peak incidence period and sex ratio remain unclear.

Atypical performance may be the main reason why patients with head and neck glomus tumors postpone visiting the maxillofacial surgery clinic. Although cold sensitivity, spontaneous intermittent pain, and pinpoint tenderness are hallmarks of extraoral glomus tumors, few patients with oral glomus tumors who are referred to the maxillofacial clinic have these symptoms [32]. The lack of such sensations may be attributable to the varying distribution of nerve fibers in different anatomical regions; this notion remains to be explored.

The accurate preoperative diagnosis of intraoral glomus tumors remains challenging. Inaccurate diagnoses are largely attributed to this tumor’s rarity and the lack of distinguishing clinicomorphologic characteristics. Furthermore, such lesions have nonspecific and heterogeneous appearances on radiologic images. A glomus tumor may initially be diagnosed as a salivary tumor, sebaceous cyst, neurofibromatosis, dermoid cyst, teratoid tumor, vascular malformation, or another type of mesenchymal neoplasm [42]. Although vascular malformations and cystic soft tissue lesions can usually be ruled out using color duplex ultrasonography, the differential diagnosis of solid tumors remains challenging. Recently, 18fluorodopa (F-DOPA) positron emission tomography was used for detecting glomus tumors [43]; however, the validity and specificity of this technique for tumors in the head and neck region requires verification. As formal diagnostic guidelines are absent, histologic examination and immunohistochemical analysis remain the gold standards.

Histologically, the appearance of glomus tumors depends on their cellular compositions and differentiation levels. A typical solid glomus tumor is composed of small vascular channels surrounded by clusters of well-defined round cells with lightly eosinophilic cytoplasm, and a large central round or oval nucleus with no atypia. The immunohistochemical profile of glomus tumor cells includes positivity for vimentin, smooth muscle actin, and muscle-specific actin; moreover, positivity for desmin, CD34, and BRAF mutations has been identified in some cases [44]. Conversely, these tumors yield negative results for S-100, myoglobin, neurofilaments, and factor VIII-related antigen [27].

Glomus tumors should first be differentiated from tumors originating from the sublingual gland, where acinar and ductal structures can be observed histologically in such neoplasia [45]; these structures were not observed in our cases. Meanwhile, the sublingual gland is a common site for epithelial tumors; we found that epithelial markers were negative in this case, indicating that the tumor was not a neoplasm of epithelial original.

Secondly, the differential diagnosis for glomus tumors includes vascular tumors such as hemangioma,
| Case | Authors                      | Year | Gender | Age(years) | Site                                | Size(cm)         | Clinical presentation          | Treatment   | Follow-up time | Outcome   |
|------|------------------------------|------|--------|------------|-------------------------------------|------------------|-------------------------------|-------------|----------------|-----------|
| 1    | Langer [6]                   | 1949 | M      | 52         | Hard palate                         | NA               | NA                            | NA          | NA             | NA        |
| 2    | King [7]                     | 1954 | M      | 32         | Gingiva                             | NA               | No symptoms                    | NA          | NA             | NA        |
| 3    | Kirschner and Strassburg [8] | 1962 | M      | 56         | Gingiva/Alveolar mucosa             | NA               | NA                            | NA          | NA             | NA        |
| 4    | Frenkel [9]                  | 1965 | M      | 13         | Buccal mucosa                       | NA               | NA                            | NA          | NA             | NA        |
| 5    | Harris and Griffin [10]      | 1965 | F      | 35         | Maxilla                             | 0.5 × 0.25 cm    | Pain                          | Surgery     | 2 years        | NED       |
| 6    | Sidhu [11]                   | 1967 | F      | 10         | Hard palate                         | NA               | Painless neoplasm              | NA          | NA             | NA        |
| 7    | Charles [12]                 | 1976 | F      | 17         | Hard palate                         | NA               | NA                            | NA          | NA             | NA        |
| 8    | Lele [13]                    | 1977 | M      | 35         | Hard palate                         | 1.5 × 1 cm       | Painless neoplasm              | Surgery     | 6 months       | NED       |
| 9    | Sato et al. [14]             | 1979 | F      | 29         | Tongue                              | NA               | Painless neoplasm              | NA          | NA             | NA        |
| 10   | Tajima et al. [15]           | 1981 | M      | 63         | Tongue                              | NA               | Painless neoplasm              | NA          | NA             | NA        |
| 11   | Saku et al. [16]             | 1985 | M      | 45         | Buccal mucosa                       | 4.5 × 3.3.5 cm   | No symptoms                    | Surgery     | NA             | NA        |
| 12   | Ficarra et al. [3]           | 1986 | F      | 51         | Upper lip                           | NA               | No symptoms                    | NA          | NA             | NA        |
| 13   | Moody et al. [17]            | 1986 | F      | 65         | Upper lip                           | 1 × 0.5 × 0.5 cm | No symptoms                    | Surgery     | NA             | NA        |
| 14   | Stajicic and Bojic [18]      | 1987 | F      | 55         | Tongue                              | NA               | Painless neoplasm              | Surgery     | NA             | NA        |
| 15   | Geraghty et al. [19]         | 1992 | M      | 71         | Hard palate                         | 1.5 cm           | No symptoms                    | Surgery     | NA             | NA        |
| 16   | Kusama et al. [20]           | 1995 | M      | 57         | Upper lip                           | NA               | Painless swelling              | Surgery     | 4 years        | NED       |
| 17   | Savaci et al. [22]           | 1996 | M      | 55         | Buccal mucosa                       | 1 cm             | Pain                          | Surgery     | NA             | NA        |
| 18   | Sakashita et al. [23]        | 1997 | M      | 54         | Upper lip                           | 1.2 × 1 cm       | Painless swelling              | Surgery     | NA             | NA        |
| 19   | Yu et al. [24]               | 2000 | F      | 54         | Left mandibular area, lip, anterior | NA               | Painless neoplasm              | Surgery     | NA             | NA        |
| 20   | Kessaris et al. [25]         | 2001 | F      | 46         | Hard palate                         | 1.8 cm           | Painless swelling              | Surgery     | 3 years        | NED       |
| 21   | Rallis et al. [27]           | 2004 | F      | 85         | Upper lip                           | 1.3 × 1 × 1 cm   | Painful swelling               | Surgery     | 1.5 years      | NED       |
| 22   | Quesada et al. [26]          | 2004 | M      | 61         | Tongue                              | 3 cm             | Painless swelling              | Surgery     | 7 years        | Recurrence |
| 23   | Lanza et al. [28]            | 2005 | M      | 65         | Lower lip                           | NA               | Painful mass                   | Surgery     | NA             | NA        |
| 24   | Ide et al. [30]              | 2008 | M      | 57         | Upper lip                           | 0.8 cm           | NA                            | Surgery     | NA             | NA        |
| 25   | Ide et al. [30]              | 2008 | M      | 54         | Upper lip                           | 1.2 cm           | NA                            | Surgery     | NA             | NA        |
| 26   | Wang et al. [31]             | 2008 | M      | 51         | Buccal mucosa                       | NA               | NA                            | Surgery     | NA             | NA        |
| 27   | Wang et al. [31]             | 2008 | F      | 58         | Buccal mucosa                       | NA               | NA                            | Surgery     | NA             | NA        |
| 28   | Boros et al. [32]            | 2010 | M      | 34         | Lower lip                           | NA               | NA                            | Surgery     | NA             | NA        |
| 29   | Yoruk et al. [34]            | 2010 | F      | 30         | Buccal mucosa                       | 2 × 1.1 × 0.5 cm | Painless swelling              | Surgery     | 1 years        | NED       |
| 30   | Dérand et al. [33]           | 2010 | F      | 11         | Lower lip                           | 0.3 cm           | No symptoms                    | Surgery     | 7 years        | NED       |
| 31   | Veros et al. [35]            | 2012 | F      | 24         | Buccal mucosa                       | 1 × 1 cm         | Painful mass                   | Surgery     | 2 months       | Recurrence |
| 32   | Chou et al. [36]             | 2015 | M      | 39         | Upper lip                           | NA               | NA                            | Surgery     | NA             | NA        |
| 33   | Monaghan [37]                | 2017 | M      | 73         | Upper lip                           | 1 cm             | No symptoms                    | Surgery     | NA             | NA        |
| 34   | Vasconcelos et al. [39]      | 2018 | M      | 67         | Upper lip                           | 1 cm             | Painful swelling               | Surgery     | 3.3 years      | NED       |
| 35   | Smith et al. [38]            | 2018 | M      | 26         | Lower lip                           | 1.5 × 0.5 × 0.5 cm | Painful mass                   | Surgery     | NA             | NA        |
| 36   | Smith et al. [38]            | 2018 | F      | 58         | Tongue                              | 2 × 1 cm         | No symptoms                    | Surgery     | NA             | NA        |
| 37   | Our case                     | M    | 24     | The floor of the mouth              | 2.8 × 1.8 × 2.1 cm | Painful swelling               | Surgery     | 4 years        | NED       |

F female, M male, NA not available, NED no evidence of death
hemangioendothelioma, epithelioid hemangioma, kaposiform angiodermatitis, reactive angioendotheliomatosis, and angiosarcoma [25]. These tumors can be easily excluded based on histomorphologic features and the expression of endothelial cell markers [46].

Glomus cells, myopericytes, vascular smooth muscle cells, and myofibroblasts are derivatives of pericytes [47]; therefore, glomus tumors should also be distinguished from the most common of the perivascular tumors, including myofibromas, glomangiopericytoma, and myopericytoma. These tumors share many histologic, immunophenotypic, and cytogenetic features, and it is difficult to distinguish them from one another solely by immunohistochemical examination [30]. However, myopericytoma can be differentiated from the glomus tumor based on concentric perivascular growth of spindle neoplastic cells [16]. Myofibromas have a biphasic zonation pattern with light staining fascicles to whorls of myofibroblastic, and dark-staining zones of polygonal cells associated with hemangiopericytoma-like vessels [48]. Glomus tumors are composed of cuboidal cells with distinct cell borders, and a round, centrally located nucleus, and they lack spindle cell morphology. The absence of both a multinodular and biphasic pattern would help to exclude myofibroma. Angioleiomyomas have a predominant vascular smooth muscle cell component. On histopathological examination, proliferation of vascular channels was noted, along with thick walls of circumferentially arranged spindle cells [32]. The histomorphological characteristics and positive expression of both actin and desmin can be used to positively identify a glomus tumor.

The majority of glomus tumors are entirely benign; hence, en bloc resection is an effective treatment. While incomplete resection may result in recurrence, local recurrence is very uncommon. Malignant glomus tumors are very rare and require multimodal integrated treatments [26].

We report the rare case of a glomus tumor in the floor of a patient’s mouth that showed no marked symptoms, which complicated its early diagnosis. Glomus tumors should be included in the initial differential diagnosis in patients presenting with painless nodules in the floor of the mouth. En bloc resection is an effective treatment, and patients should receive long-term counseling regarding the risk of recurrence.

**Abbreviations**

AE1: Anti-cytokeratin 1; AE3: Anti-cytokeratin 3; CD: Cluster of differentiation; F-DOPA: 18Fluorodopa

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**Availability of data and materials**

All data generated or analyzed during this study are included in this published article.

**Authors’ contributions**

All authors provided intellectual contribution to this manuscript. HZ wrote the manuscript. LS and MJ analyzed and interpreted the patient data. LW confirmed the histopathological examination results. YS reviewed the clinical notes and edited the document. All authors read and approved the final manuscript.

**Ethics approval and consent to participate**

This study was approved by the Research Ethics Committee of the Hospital of Stomatology of Wuhan University.

**Consent for publication**

Written informed consent was obtained from the patient for the publication of this case report and any accompanying images. A copy of the patient’s written consent is available for review by the Editor-in-Chief of this journal.

**Competing interests**

The authors declare that they have no competing interests.

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