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Case Report

Basal Cell Adenoma of The Lingual Anterior Salivary Gland (Blandin-Nühn Gland): A Rare Entity Arising in An Unusual Location. Bibliography Review and Case Report

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Abstract

Basal cell adenoma (BCA) of the tubular-trabecular subtype is a rare benign tumor of the major and minor salivary glands. This adenoma rarely occur in the minor salivary glands and one case has been reported in the tongue, but it could not be determined whether it was in the anterior lingual gland (Blandin Nühn gland).
The clinical case of a female patient in her fifth decade of life is presented. The mass on the ventral surface of the tongue that was diagnosed as a basal cell adenoma of the tubular-trabecular subtype was removed by complete oral excision.
We did a bibliographic review of BCA in the anterior lingual gland, reported from 1991 to December 2024, review of the anatomy and histopathology of this gland was carried out, and a bibliographic review was conducted to present the number of cases of this type of salivary tumor in the tongue.

Keywords: *Benign tumor, Minor salivary gland, adenoma basal cells, intraoral salivary gland, Tongue.*

INTRODUCTION

Intra-oral minor salivary gland tumors are rare tumors comprising approximately < 1% of all head and neck tumors and <4% of all salivary gland tumors (SGTs) [1].

The percentage of basal cell adenomas (epithelial neoplasia of the salivary glands) is even lower, ranging from 0.2 to 2% of all salivary gland tumors [2-5]. No cases have been described in the anterior lingual gland.

The anterior lingual glands were initially mentioned by Gaspare Bauhin (1560-1624), although Bahuin mentioned it *(...qui fub mufculo ceratogloffo fitifunt, plurimae glandulae carnosae, fub lingula circa eus frenum occurrunt...)* in 1590 [6], this small structure, without surgical interest and also written in Latin made it go unnoticed by the scientific community, centuries later in 1823 Philippe-Frédéric Blandin (1798-1849) describes it and it remains with his name [7], until, in 1845, Anton Nuhn from the University of Heidelberg noticed that this gland had not been thoroughly investigated and made a monograph where he gives more details about this gland [8].

Anatomy

The anterior lingual glands (also called apical glands) are seromucous glands located deep near the tip of the tongue on either side of the *lingual frenulum (fig. 1)*. They lie on the undersurface of the apex of the tongue and are covered by a bundle of muscle fibers derived from the *styloglossus* and *inferior longitudinalis*. They measure between 12 and 25 mm long and approximately 8 mm wide, and each open by three or four ducts on the undersurface of the apex of the tongue [9].

Between the muscles covering it and the gland near the medial side of the latter, runs the anterior end of the ranine artery, sometimes completely surrounded by the lobes of the gland and always sending several branches to the gland itself. In the same way, the ranine vein, which accompanies the artery, receives most of the venous branches returning from the

gland. The anterior end of the lingual nerve runs along the medial side of the gland and penetrates with several of its branches even between the lobules of the gland [10].

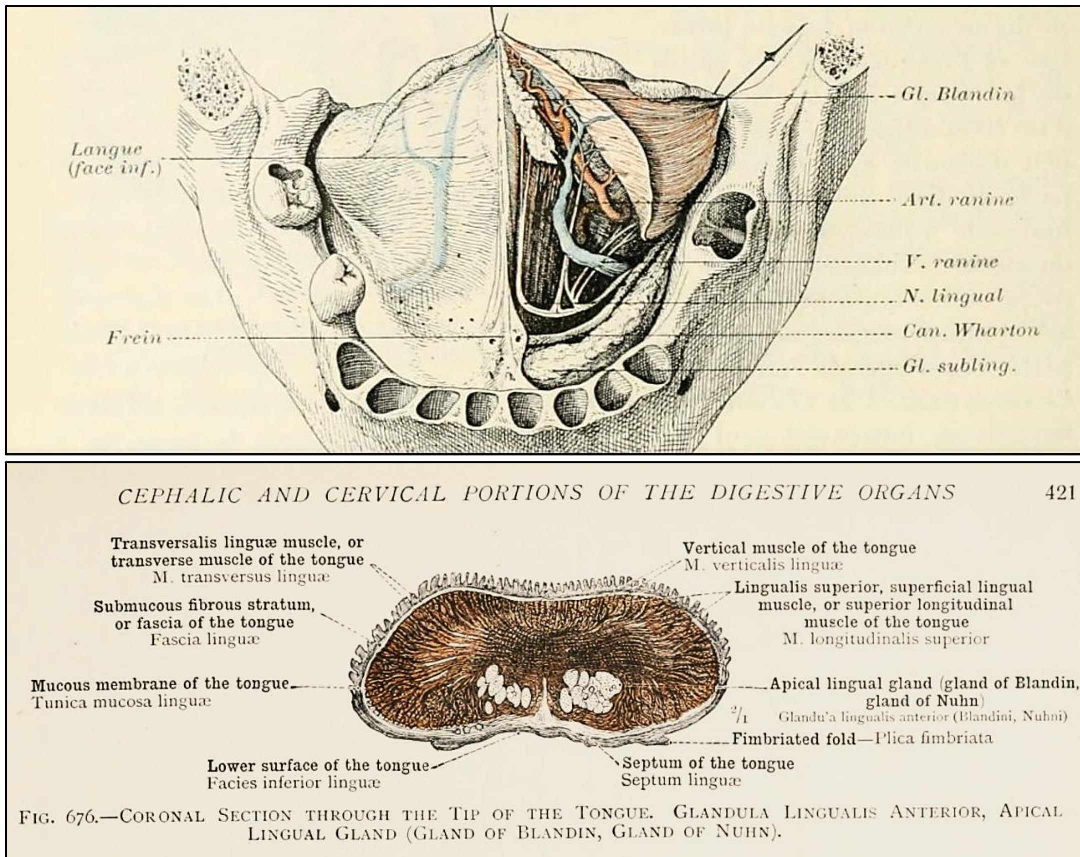

Fig. 1.: Coronal section through the tip of the tongue. Glandulae lingualis anterior. Told, 1904 [11].

Histology

The anterior lingual salivary glands are composed predominantly of mucous tubules (which come in two distinct sizes: large and small), seromucous demilunes, and rare seromucous acini. Regardless of tubule size, mucous cells are typically in appearance and, like mucous cells in other human salivary glands, contain filamentous bodies [12].

Histopathology

The term "monomorphic adenoma" of the salivary glands was first proposed by Kleinsasser and Klein in 1967 in order to distinguish it from pleomorphic adenoma. Histogenetically, they can be divided into four groups:

1) tumors of terminal duct origin (*basal cell adenoma* and *canalicular adenoma*),
2) tumors of terminal or striated duct origin (*sebaceous adenoma* and *sebaceous lymphadenoma*),
3) tumors of striated duct origin (*oncocytoma* and *papillary cystadenoma lymphomatosum*), and
4) tumors of excretory duct origin (*sialadenoma papilliferum* or *inverted ductal papilloma*) [13].

The basal cell adenoma (BCA) can be divided on the basis of their morphologic appearances into four subtypes [14]:

- Solid
- Tubular
- Trabecular
- Membranous

The incidence of intraoral minor salivary gland tumors varies among populations, as demonstrated in studies conducted in Asia [15-20], Europe [1,21,22], North America [23-26], South America [25-33], Africa [34-37], and other multi-institutional [38,39].

Regional differences in the occurrence of salivary gland tumors may reflect genetic and cultural variations among people from different geographic regions.

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Although already was known from 1928 to 1971, eight BCA arising from minor salivary glands had been reported. Christ & Crocker series (1972) brings the total to 11. The majority 8 of these 11 cases have occurred in the lip [40].

We present a patient with basal cell adenoma of the trabecular-tubular subtype located in the anterior lingual gland (Blandin-Nuhn gland), and we review the literature.

MATERIAL AND METHODS

Retrospective and cross-sectional study. All intraoral cases with a histopathological diagnosis of basal cell adenoma, recorded between 1991 and December 2024, were included.

The PubMed, SciELO and Google Scholar search in English, Portuguese and Spanish with the words: "salivary gland tumors", "intraoral gland tumors", "basal cell adenoma", "minor salivary glands", "lingual anterior gland adenoma", "Blandin-Nuhn glands adenoma", resulted in 14877 articles from 1991 to 2025, of which 74 articles reported at least one case of minor salivary gland basal cells adenoma.

All cases of minor salivary glands that were not basal cell adenomas and in which it could not be determined whether the BCA were minor salivary glands were excluded, in addition to those of the oropharynx, base of tongue and paranasal sinuses.

Additionally, those prior to the new WHO classification of 1991 and Update from the 5th Edition of the World Health Organization Classification of Head and Neck Tumors: Salivary Glands (2022) [41] were excluded. There were several studies that were discarded because the data collected were prior to 1991.

RESULTS

Have been reported from the 1991 to 2024 herein 216 cases of minor salivary gland BCA (216 cases from the literature review and 1 new case reported herein) and only 2 cases in the tongue (1%), and our case the only one confirmed in the anterior lingual glands (Blandin-Nühn glands).

Basal cell adenomas in minor salivary glands were distributed as follows: in the upper lip 76 cases (35%), in the hard palate 47 cases (21.6%) in the buccal mucosa 36 cases (16.6%), in the lip without specifying the anatomical site 3 cases (1.4%), in the retromolar triangle 2 cases (0.9%), in the alveolar region 2 cases (0.9%), on the floor of the mouth 2 cases (0.9%), in the lower lip 1 case (0.45%), on the soft palate 2 cases (0.9%), on the tongue 2 cases (0.9 %), of these last 1 case on the tongue without determining the anatomical location and our case on the lingual gland anterior (0.45%); and 45 cases (20.7%) without determining the anatomical site.

Table 1. Distribution of case of Basal Cell Adenoma in minor salivary glands.

DISTRIBUTION	CASES	% off Basal Cell Adenoma
UPPER LIP	76	35
HARD PALATE	47	21.6
BUCCAL MUCOSA	36	16.6
LIP/ ND	3	1.4
RETROMOLAR TRIGONE	2	0.9
ALVEOLAR REGION	2	0.9
FLOOR MOUTH	2	0.9
SOFT PALATE	1	0.45
LOWER LIP	1	0.45
TONGUE /ND	1	0.45
ANT. LINGUAL GLAND*	1	0.45
ND	45	20.7
TOTAL	217	100

Regarding sex, it was found that 51 (23.5%) cases were female, 17 male (7.8%) and 149 (68.6%) undetermined. Among the decade of life of the 67 cases where sex was determined, 4 cases (3.5%) were between 20-29 years; 4 cases (6%) between 30-39 years; 11 cases (16.4%) were between 40-49 years; 10 cases (14.9%) between 50-59; 11 cases (16.4%) between 60-69; 7 cases (10.5%) between 70-79 years and 1 case (1.5%) between 80-89.

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CASE REPORT

We reported a 51-year-old woman was referred to the Department of Oral and Maxillofacial Surgery at the San Vicente de Paul University Hospital in April 2024 by a private dentist. The patient had a 5-month history of mild inflammation on the ventral surface of the tip of the tongue (fig. 2).

On palpation, the swelling was non-tender, firm, with a smooth surface and well-defined margins and painful to palpation. It was non-compressible, non-fluctuant and non-pulsatile. There was no restriction of tongue mobility. No cervical lymphadenopathy was evident.

Based on the clinical appearance, a provisional diagnosis of extravasation mucocele was given, with a differential diagnosis of pleomorphic adenoma, traumatic neuroma, schwannoma and myofibroma.

An MRI is performed, observing a signal hyperintense (bright or white) in the anterior part of the body of the tongue (fig. 3). Given the size of the tumor, it was decided to perform an excisional biopsy.

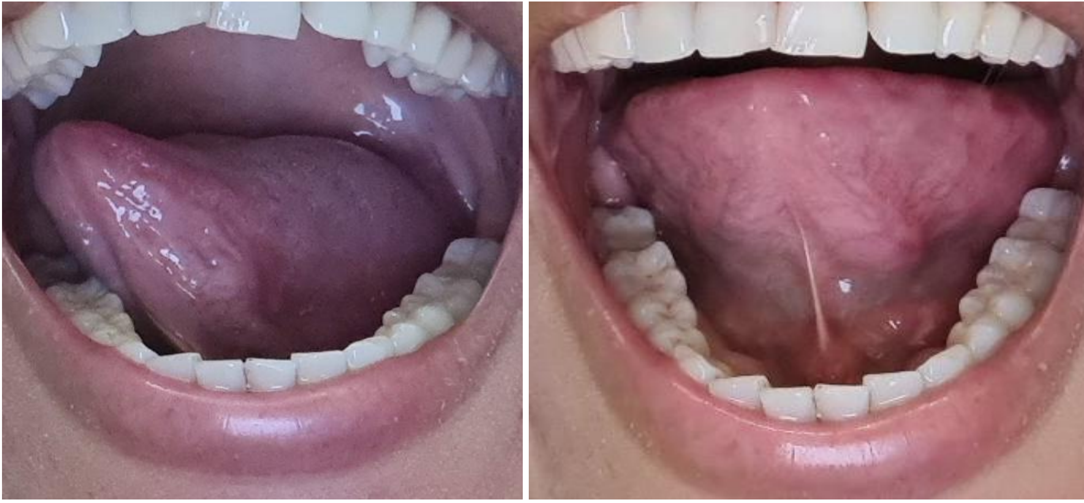

Fig. 2: Clinical image where the small elevation in the mucosa of the ventral side of the tongue, lateral to the frenulum, can be observed.

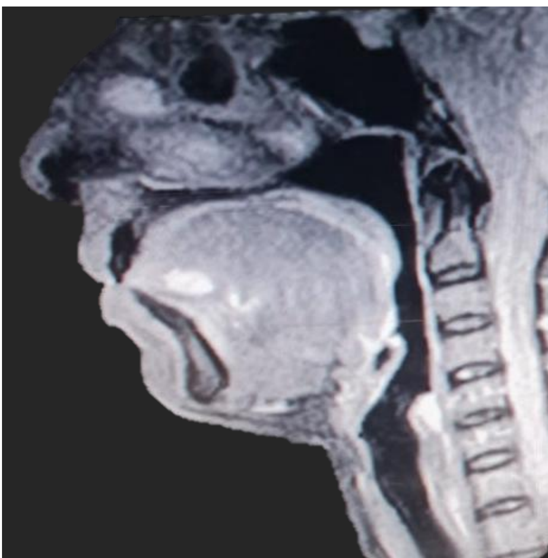

Fig. 3: Characteristics of the contrast-enhanced magnetic resonance imaging hypointense nodular lesion in T1 sequence, and intense in STIR and T2 sequence, approximately 11 mm to the side of the midline in the anterior and basal part of the tongue, where homogeneous images with well-defined and encapsulated margins are shown.

Surgical technique:

After asepsis and antisepsis of the operating field and the placement of surgical fields, under local infiltration anesthesia with 2% lidocaine with epinephrine 1:100,000 IU, the tongue was stabilized with a traction suture.

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A linear paramedian incision was made on the ventral surface of the tongue and blunt dissection of the soft tissues was performed until reaching the tumor. On gross examination, the resected specimen included a well-circumscribed lesion with a whitish appearance in all its extension (fig. 4).

Using blunt dissection, the lesion was removed, the bleeding was controlled with electric cautery and sutured with 4-0 vicryl. The patient healed uneventfully and no recurrence was detected on the regular 1-year follow-up.

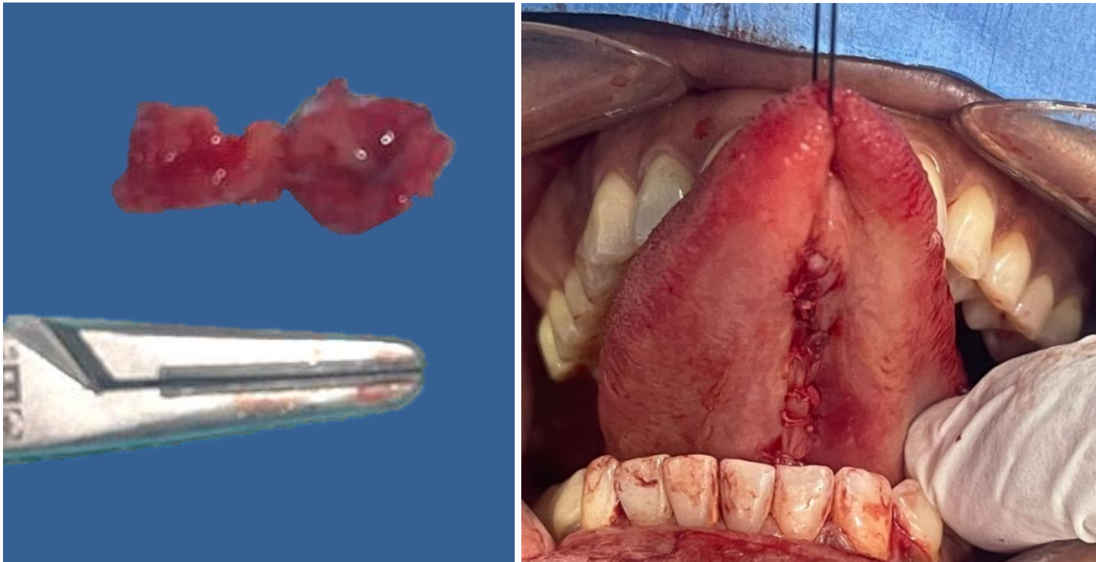

Fig. 4: Left: Excised specimen: Elastic and hard tumor with size 15 × 8 × 4 mm. Right: Paramedian incision suture

The specimen was analyzed at the Pathology Service of the San Vicente de Paul Regional Hospital in the Dominican Republic, which reported: numerous monotonous cubic cell trabeculae and another component of small and medium-sized tubular structures with epithelium that varies from cubic to flat that do not show atypia. The stroma is sparse and predominantly loose (fig. 5). The tumor has a fibrous capsule with no evidence of malignancy. The diagnosis was concluded as BCA subtype trabecular-tubular.

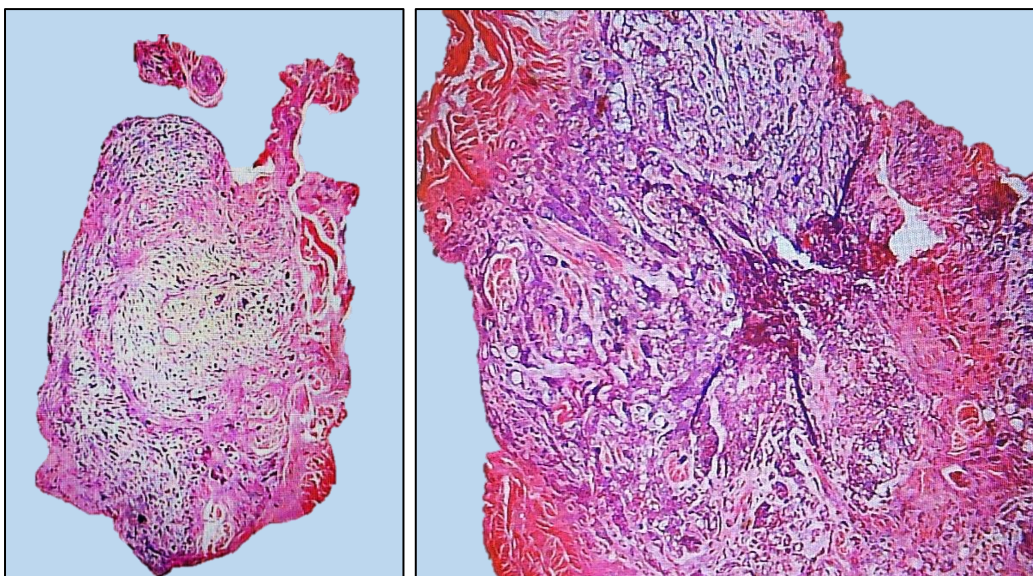

Fig. 5: Mixed tubular-trabecular pattern of basal cell adenoma type showing numerous trabeculae of monotonous cubic cells and another component of small and medium-sized tubular structures with epithelium that varies from cubic to flat without atypia. The stroma is scant and predominantly loose, with a fibrous capsule. Hematoxylin-eosin, x10-x80.

Immunohistochemistry is rarely needed to establish a diagnosis of BCA or BCAC in the separation of either from basaloid salivary gland neoplasms if the tumor is completely excised. In the case of limited material, immunohistochemical studies may help point to the correct diagnosis [41].

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The immunohistochemistry shows the immunoreactivity to S-100 and cytokeratin varied. All basaloid cell adenomas were positive for cytokeratin, while S-l 00 positivity was found mostly in the stroma along with the varied reaction and intensity in the epithelial nests.

The patient presented paresthesia of the lateral border of the tip of the tongue, which improved with Nucleo C.M.P. FORTE™ (Cytidine 5-monophosphate (CMP)- 5 mg, Uridine 5-triphosphate (UTP)- 3 mg). Despite this treatment, a year later a slight paresthesia persists on the lateral edge of the tip of the tongue and a slight deformity on the lateral edge of the tongue.

DISCUSSION

Although the distribution of salivary gland tumors varies from series to series, its prevalence is estimated to be between 3 and 6% of all head and neck tumors [42,43]. The majority (68%) of the salivary gland tumor presented in major and 32% in the minor glands. The parotid gland was the most common location (70%) for benign and minor glands (47%) for malignant tumors [39].

The minor salivary glands are important components of an oral cavity, present in most parts of the mouth, and their secretions directly bathe the tissues [44]. Development of these tumors in the buccal mucosa, palate, or lower lip is unusual. Although these tumors are generally described as freely movable, asymptomatic swellings or masses, there have been a number of reports of pain associated with major and minor gland tumors.

Since the first report detailing the clinicopathologic features of monomorphic adenomas appeared in the English-language literature in 1970 [45].

In the 1972 World Health Organization (WHO) histological classification of tumors, salivary gland adenomas were divided into two categories: monomorphic and pleomorphic adenomas. According to this classification, basal cell adenoma (BCA) was categorized as a subtype of monomorphic adenoma [13].

The World Health Organization classification of salivary gland tumors has been updated three times, with the most recent update published in 2017 [46] and update in 2022 [47].

Compared to its previous version, the nomenclature and histological subtypes have changed significantly. Therefore, accurate histopathological diagnosis is of fundamental importance for the evaluation of these patients, to establish the appropriate treatment.

In a study of 6982 cases in minor salivary gland tumors in a Chinese population, BCA was extremely rare, only 5 cases (0.07%) BCAs were reported in minor glands [2], but less so in another population according to an international multicenter study with 5379 cases analyzed the BCA 95 cases (1.77%) were reported in minor glands [38].

Wang *et al*. (2012) found that the proportions of minor salivary glands tumors in 1176 gland tumors, the benign are 397 (53.9%), and the basal cells adenoma only 4 cases (0.4%), 1 on the tongue, but it was not determined in which part of the tongue it was located [48].

Since basal cells adenoma was classified as an independent salivary gland tumor in 1991, only 217 cases of BCA of the minor salivary glands, including the present case, have been reported.

In our review the upper lip is the most common site followed by hard palate. Tongue is an unusual site. A total of 2 cases, including the cases in this article have been reported in the tongue, and possible this one unique case reported of a basal cell adenoma subtype tubular/trabecular in the anterior lingual gland (Blandin-Nuhn glands), in the other case, its anatomical location could not be determined.

It is possible that by existing considerable disagreement in previous years over further subclassification of monomorphic adenomas.

BCA accounts for 1.8–5% of all salivary gland tumors. It is predominantly a tumour of major salivary glands with approximately 75–80% of cases found in the parotid glands and at least 5% in the submandibular glands [49].

Most studies have shown that minor salivary gland tumors are more common in females than males [23,50, 51,52,53] with a male-to-female ratio ranging from 1:1.02 [54] to 1:2.0 [26].

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CONCLUSIONS

The anterior lingual glands were mucous-rich mixed glands; and mucous and serous acinar cells and ductal cells were observed [55].

Since BCA was first classified as an independent tumor in 1991, only 216 reported cases of BCA have been reported in the minor salivary gland, most commonly occurs on the upper lip followed by the palate and the buccal mucosa with no cases reported on the anterior lingual gland. BCA of the anterior lingual gland are a rare neoplasm as demonstrated by our case.

The varied presentation of tumors of the minor salivary glands makes diagnosis a challenge. However, as seen in the present case, despite its rarity, another salivary gland tumor can also be considered in the differential diagnosis of swellings of the tongue.

The adenomas, specifically the BCA should be considered in the differential diagnosis of lingual masses in both young and adult patients. Complete local surgical excision is the treatment of choice to treat this pathology.

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