

Case Report

# Unmasking Hypoglossal Nerve Schwannomas Mimicking Submandibular Salivary Gland Tumors: Case Report of a Rare Presentation and Surgical Management

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**Abstract:** Background: Schwannomas are solitary neurogenic tumors originating from the myelin-producing cells of the neural sheath. Hypoglossal nerve schwannomas are exceedingly rare, particularly those extracranially originating and mimicking a submandibular salivary gland tumor. Methods: We report the case of a 31-year-old female who presented to our ENT department with a painless swelling in her left submandibular region that has persisted for approximately five months. Discussion: Due to the rarity of these tumors and their unique nature, a comprehensive diagnostic workup is imperative for accurate diagnosis. Surgical excision remains the gold standard treatment. Conclusions: Extracranial hypoglossal nerve schwannomas represent a rare clinical entity requiring a thorough diagnostic process for precise identification. The preferred treatment strategy for managing hypoglossal gland schwannomas involves complete tumor excision while preserving the facial nerve.

**Keywords:** schwannoma; submandibular; surgery; facial nerve; neck; tumor; case report



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## 1. Introduction

Schwannomas are solitary, benign, slow-growing, and well-encapsulated neurogenic tumors arising from the myelin-producing cells of the neural sheath [1]. These tumors can occur anywhere in the body, most located in the head and neck region.

Hypoglossal nerve schwannomas, particularly those originating extracranially and mimicking a submandibular salivary gland tumor, are extremely rare [2,3]. The submandibular gland, one of the major salivary glands, is found beneath the mandible, near the anterior edge of the sternocleidomastoid muscle. The first case of hypoglossal schwannoma was reported by Stout in 1951. Since then, only a few cases have been documented in the literature due to the rarity of this type of tumor. Most hypoglossal schwannomas have been observed in patients aged between the third and sixth decades of life, with a slight female predominance [4].

Extracranial hypoglossal nerve schwannomas typically present as a gradually enlarging, painless mass in the neck, which can easily be mistaken for a submandibular salivary gland tumor [2,3]. Depending on the tumor’s location and size, some patients may experience symptoms like dysphagia, hoarseness, or tongue atrophy. Given their indolent nature, slow growth, and clinical and radiological resemblance to other benign and malignant salivary gland lesions [5], these tumors can remain asymptomatic for extended periods, making diagnosis even more challenging [1]. Understanding hypoglossal schwannomas’ clinical, radiological, and histopathological features is crucial for accurate diagnosis and appropriate treatment.

Imaging techniques such as computed tomography (CT) and magnetic resonance imaging (MRI) play a vital role in the preoperative assessment of hypoglossal schwannomas [6]. MRI, in particular, can provide detailed information about the tumor's nature, size, and anatomical relationships with surrounding nerves and tissues [6]. Due to the rarity of these tumors and their lack of specific radiological features, the preoperative diagnosis often remains elusive [6]. Thus, the definitive diagnosis relies on the histopathological examination of the surgical specimen [5].

The primary treatment for hypoglossal schwannomas is complete surgical excision [5]. In some cases, nerve-sparing surgery, which aims to preserve the nerve's function, may be attempted if the preoperative diagnosis of schwannoma has been established [6]. However, nerve-sparing surgery may not always be possible because of the difficulty in preoperative diagnosis and the infiltrative nature of these tumors [6]. In such cases, nerve reconstruction can be considered to minimize potential postoperative complications, such as facial nerve dysfunction or loss of sensation in the innervated territory [5].

Despite the benign nature of hypoglossal schwannomas, careful postoperative follow-up is essential, as these tumors can recur if not completely excised [5]. Moreover, malignant transformation has been reported in some cases of schwannomas, although this is extremely rare [7]. Notable case studies of submandibular schwannomas include the work of Batsakis et al. [8], which discussed a case in a 67-year-old patient, and Murthy et al. [9], which reported a case in a 53-year-old patient. Both studies underscored the importance of a multidisciplinary approach in diagnosing and treating these rare tumors.

In this report, we aim to present a rare case of a submandibular schwannoma masquerading as a submandibular gland tumor, highlight the diagnostic and management challenges associated with this type of tumor, and discuss the crucial clinical, radiological, and histopathological features necessary for its precise identification.

## 2. Case Report

### 2.1. History and Examination

A 31-year-old female consulted our ENT department with a complaint of painless swelling in the left submandibular region that was present for about five months. A single swelling, measuring approximately  $4 \times 3 \times 2.2$  cm, was identified upon physical examination. The swelling was smooth to the touch, with a soft and elastic consistency. It had well-defined margins, was slightly mobile, and tender upon palpation. Notably, there was no palpable laterocervical lymphadenopathy, and the cranial nerve function appeared normal. An examination was conducted to assess the integrity and functioning of the facial and hypoglossal nerves. No abnormalities, such as depression of the nasolabial fold or palpebral fissure, were detected. When asked to smile, the patient exhibited no facial expression asymmetries. Despite initial treatment with anti-inflammatory medications and antibiotics, the patient did not experience any marked improvement. As shown in Table 1, laboratory tests did not reveal any inflammatory findings.

**Table 1.** Baseline hematological values. Abbreviations: ESR, Erythrocyte sedimentation rate; CRP, C-reactive protein.

Blood Parameters	Values
Erythrocytes	$4.85 \times 10^6/\mu\text{L}$
Hemoglobin	14.3 g/dL
Platelets	222,000/ $\mu\text{L}$
Leukocytes	7200/ $\mu\text{L}$
Neutrophils #	$3.27 \times 10^3/\mu\text{L}$
Lymphocytes #	$2.90 \times 10^3/\mu\text{L}$
CRP	0.92 mg/dL
ESR	13 mm/h

Upon the attending physician's advice, the patient underwent a CT scan. The scan, assessed by an ENT specialist, showed an enlarged left submandibular gland compared to the contralateral side, with homogeneous density (Figure 1). Consequently, we performed ultrasonography of the neck. This revealed a hypoechogenic area measuring  $19 \times 19 \times 15$  mm in the left submandibular region, with evidence of peri- and intralesional vascularization per the Doppler signal.



**Figure 1.** Axial view of the CT scan shows enlargement and homogeneous density of the left submandibular space gland compared to the contralateral.

Fine needle aspiration cytology (FNAC) of the submandibular mass revealed cohesive clusters of benign spindle cells with bland nuclei and bipolar cytoplasmic processes, features consistent with a benign salivary gland tumor. These findings suggested the possibility of a benign pleomorphic adenoma.

The clinical and radiological findings indicated a potential benign submandibular salivary gland tumor, such as a pleomorphic adenoma typically characterized by hypodensity on CT and bright hyperintensity on T2-weighted images [10,11].

## 2.2. Surgery

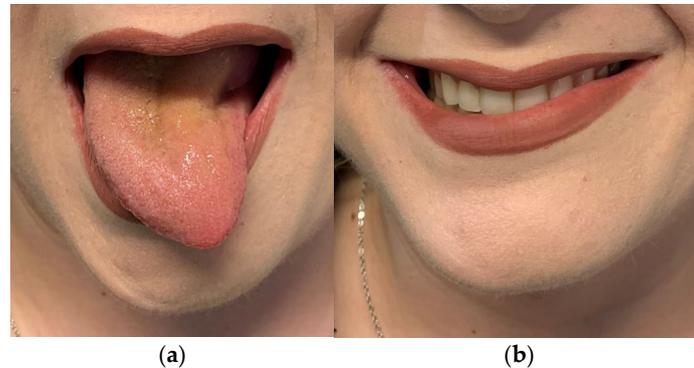
The patient underwent surgical resection of the mass via a transcervical approach. After mobilizing the neoplasm, we isolated it from the digastric muscle and proceeded with the ligation of the facial artery and vein. The dissection continued two centimeters from the mandible, with the mass being detached from the mylohyoid muscle. Although a clear cleavage plane was absent, it was possible to ligate Wharton's duct while preserving the lingual nerve. A suction drainage tube was applied, followed by an intradermal suture. The exposure of the submandibular gland was achieved using a "pull-through" approach, enabling the identification of the facial, lingual, and hypoglossal nerve. Notably, the hypoglossal nerve appeared encased by the neoplasm, and a minor nerve injury was noted during its isolation. However, considering the substantial integrity of the nerve, it was not deemed necessary to implement reconstruction techniques.

Postoperatively, a moderate degree of ipsilateral tongue deviation and deviation of the right buccal rhyme angle were observed, attributable to the involvement of the marginal mandibular branch of the facial nerve.

## 2.3. Clinical Course and Follow-Up

The postoperative course spanned three days, during which the suction drainage was maintained for 48 h. The patient experienced no postoperative complications and resumed

eating on the second day. Intravenous antibiotic therapy was administered throughout the hospital stay. At a three-month follow-up, there were no signs of local recurrence. However, the same degree of ipsilateral tongue deviation and buccal rhyme deviation persisted, as shown in Figure 2. Consequently, the patient was enrolled in a speech therapy rehabilitation protocol.

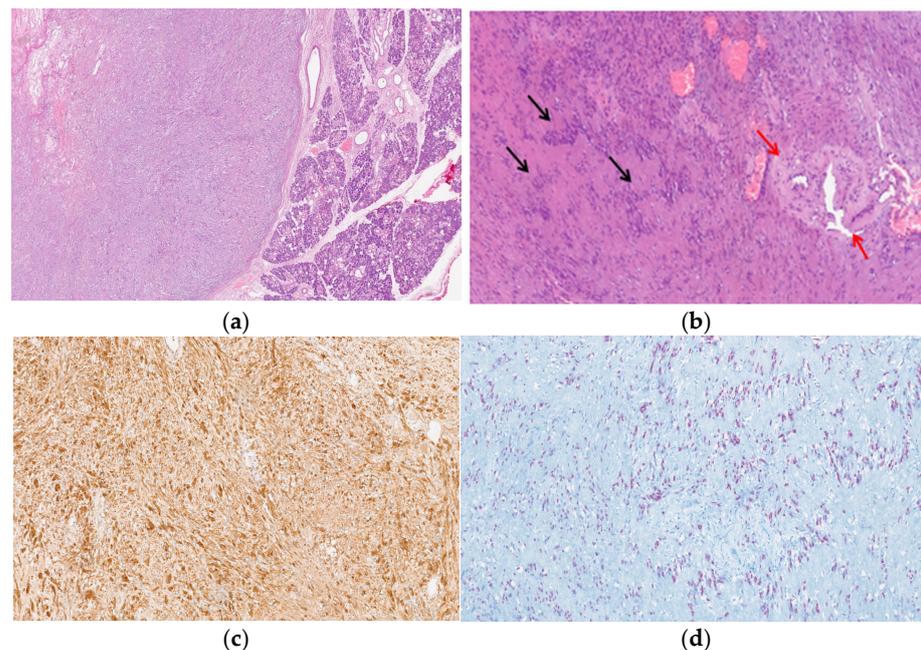


**Figure 2.** (a) Postoperative paresis of the left hypoglossal nerve. (b) Postoperative paresis of the marginal branch of the left facial nerve.

#### 2.4. Histopathological Examination

The surgical sample was fixed in 10% neutral-buffered formalin, embedded in paraffin, sectioned to a thickness of 5  $\mu\text{m}$ , and stained with hematoxylin and eosin (H&E). Histological examination revealed large-sized cysts, accounting for about 80% of the entire lesion, which caused a multilocular cystic appearance of the tumor. A nodular multi-cystic lesion was seen closely adjacent to the submandibular gland at low magnification.

Upon higher magnification, the cysts were found to be devoid of any epithelial layer (pseudocysts), and the surrounding tissue comprised a proliferation of bland-looking, spindle-shaped cells with wavy nuclei, primarily arranged in bundles. Focal palisading of nuclei and a mild-to-moderate degree of nuclear atypia were observed. Mitoses were absent, and no atypical mitoses or necrosis were found. Lymphocytes and foamy macrophages were interspersed among the neoplastic cells. The stroma also featured hemorrhages and hemosiderin deposits (Figure 3).



**Figure 3.** (a) Low magnification showing a spindle cell mesenchymal neoplasm arising within salivary gland parenchyma (on the right) (H&E; original magnification 50 $\times$ ). (b) Tumor is composed of elongated

fascicles set in a fibrous stroma and exhibits areas of nuclear palisading around the fibrillary process (“Verocay bodies”) (black arrows); medium-sized vessels with hyalinized walls (red arrows) are seen (H&E; original magnification 200×). (c,d) Neoplastic cells are diffusely and strongly stained with S-100 (c) and SOX10. (d) (immunoperoxidase; original magnification 200×). Abbreviations: SOX10, SRY-related HMG-box 10.

### 3. Discussion

This clinical case highlights a rare schwannoma originating from the hypoglossal nerve, disguised as a submandibular gland tumor. Clinical examination revealed the standard characteristics of a progressively enlarging, painless neck mass. Originating from the myelin sheath of peripheral nerves, schwannomas are benign tumors representing 35% of head and neck district tumors [12]. However, schwannomas affecting the salivary gland are exceedingly rare [13].

Schwannomas exhibit several histological subtypes: ancient, microcystic–reticular, epithelioid, cellular, psammomatous, and melanotic [14]. Histopathologically, submandibular schwannomas typically demonstrate a biphasic growth pattern comprising densely aggregated spindle cells (Antoni A) and areas of looser, disorganized cells (Antoni B) [4]. Verocay bodies—layered structures of spindle cells separated by thin reticular fibers—are considered a distinctive schwannoma feature [4]. Additionally, immunostaining for the S-100 protein, highly expressed in Schwann cells, can be useful in confirming a schwannoma diagnosis [5].

These tumors generally grow slowly and affect both sexes but show a higher prevalence in women. While they are usually asymptomatic, they can occasionally result in symptoms such as paresthesia, a foreign body sensation, dysphagia for solids, and sometimes pain [12]. The diagnosis of salivary gland schwannoma is often made postoperatively.

Due to the rarity of extracranial hypoglossal nerve schwannomas and their resemblance to submandibular salivary gland tumors, a comprehensive diagnostic workup is necessary.

The diagnostic work-up includes imaging studies such as ultrasound, CT, and MRI. Moreover, it is crucial to thoroughly understand the hypoglossal nerve’s anatomy, particularly its extracranial portion and its relationship with nearby structures [15,16].

Despite MRI being the diagnostic test of choice for both mass detection and extension assessment, patients often undergo CT scans [17]. In one of the earliest reported submandibular gland schwannoma cases, Susan et al. emphasized the importance of preoperative imaging for accurate diagnosis and surgical planning [18]. In our patient, while MRI aided in evaluating the neck mass, a definitive diagnosis was only achieved after surgical resection and histopathological examination.

Histopathological examination after surgical excision often confirms a schwannoma diagnosis. Baranović M. et al. [19] presented a case report of a 53-year-old female with a painless swelling in the left submandibular region, stressing the importance of considering schwannomas in the differential diagnosis of submandibular masses and the need for complete surgical excision. Other authors, such as Gaffar et al. [20], suggested that preoperative FNAC could facilitate diagnosis.

Our study confirms the importance of preoperative cytology using FNAC, which provided clues to a benign spindle cell process but did not lead to a specific diagnosis. This underscores the need for a multimodal diagnostic approach when schwannoma is suspected but reinforces that tissue diagnosis is required for definitive diagnosis.

Extracranial hypoglossal nerve schwannomas, while rare and benign, can potentially result in various long-term complications. These complications largely depend on the tumor size, location, and the treatment approach that is taken.

One of the significant complications relates to the neurological effects. Given that the hypoglossal nerve controls the movements of the tongue, a schwannoma that impinges on this nerve can affect the tongue’s functionality [3–6]. This can lead to changes such

as tongue weakness or atrophy, altered speech (dysarthria), and even difficulty swallowing (dysphagia). These symptoms might persist after the surgical removal of the tumor, especially if the nerve is damaged during the procedure [8–10].

Beyond the physical complications, there is also a potential psychosocial impact to consider. Alterations in speech and swallowing can significantly affect a patient's quality of life. This can lead to various psychosocial challenges, including reduced self-esteem, depression, and anxiety.

The preferred treatment is surgical, aiming for complete excision while preserving the integrity of the hypoglossal nerve. However, due to their very nature, neural damages after surgical resection of salivary gland schwannoma are common, seen in more than 50% of cases [13]. A clinical study by Keon Woo Park et al. [21] analyzed 61 cases diagnosed with schwannomas at the final biopsy after surgery. Three schwannomas (4.9%) developed in the submandibular space. They found that the most common surgical approach was transcervical, with complete tumor excision being the primary treatment. The authors noted that most cases had minimal postoperative complications, with only transient facial nerve palsy reported in some instances.

Surgical complications are another area of concern. While the surgical removal of these tumors is generally safe, there is always a potential risk of complications. These could include infection, bleeding, and potential damage to surrounding nerves or structures. The complexity of the surgery can often depend on tumor size and location and its proximity to other critical structures.

The possibility of recurrence also exists [21]. Recurrence is infrequent, and malignant transformation is rare. In the present case, the resection margins were free from pathology, further emphasizing that complete excision is curative and the preferred treatment when feasible without significant morbidity.

Although schwannomas are usually slow-growing and benign, there is a chance of recurrence after surgery [22]. This is especially true if the tumor could not be entirely removed due to its location or size.

In such cases, clinicians may opt for alternative treatment options such as Watchful Waiting, Radiation Therapy, or Stereotactic Radiosurgery.

Given the benign nature and slow growth of schwannomas, it might be an option to monitor the tumor's size and growth over time without immediate intervention, especially if the tumor is small and asymptomatic [15]. This approach would involve regular follow-up appointments and imaging studies.

Although radiation therapy is more commonly used for malignant tumors, it can sometimes be used for benign tumors like schwannomas when surgery is not an option. Radiation therapy might be considered if the tumor is in a location that makes surgery particularly risky or if the patient has other health conditions that make them a poor candidate for surgery. However, this is not a commonly used treatment for these tumors due to their benign nature and the potential side effects of radiation [19].

On the contrary, in Stereotactic Radiosurgery, delivering precise radiation doses to the tumor helps to minimize exposure to the surrounding healthy tissues [10–12]. This approach is usually considered for brain or spinal cord schwannomas, but in particular scenarios, it may be used for submandibular schwannomas.

In our patient, total tumor excision was possible together with preserving the nerve via a transcervical approach, allowing optimal exposure and no postoperative adverse effects. Complete excision is curative and is the preferred treatment when feasible without significant morbidity. Nerve-sparing should be attempted given the indolent tumor growth.

#### 4. Conclusions

This case highlights the diagnostic pitfall posed by extracranial schwannomas due to their rarity and resemblance to more common salivary gland tumors. Submandibular schwannomas should remain on the differential diagnosis for submandibular masses.

A stepwise diagnostic approach utilizing imaging, cytology, and excisional biopsy is useful in achieving an accurate diagnosis. Nerve-sparing surgical excision is the optimal therapeutic strategy when possible. When feasible, total surgical excision is the treatment of choice and should be aimed for while attempting to preserve neural function, given the characteristically indolent course of these tumors.

Further research is needed to establish more standardized surgical techniques and long-term outcomes.

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