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Schwannoma of the tongue: two case reports and review of the literature

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Abstract The aim of this study was to describe clinicopathologic and radiographic features of two cases of schwannoma involving the oral tongue and to review the literature of this unusual clinical entity. Case reports with review of the pathologic, radiologic and clinical data for two patients with schwannoma of the tongue are reported. Review of the literature of case reports of schwannomas (neurilemmomas) of the tongue from 1955 to 2006 with analysis of the patient's age, gender, presenting symptom(s), tumor size, and surgical approach was undertaken. The two patients in our series presented with painless swelling of the tongue. Transoral excision was performed and pathologic examination confirmed the diagnosis of schwannoma in both the cases. A total of 126 cases of schwannoma of the tongue have been reported in the English literature over the past 51 years. Schwannomas of the tongue typically present in the third decade of life (33%), display no gender predilection (52.8% female; 47.2% male) and often present as a painless mass (69.6%). Schwannomas are likely to elicit distressing symptoms when they occur in the posterior one-third of the tongue (63.2 vs. 13.5%) or approach 3 cm in greatest dimension (33.0 vs. 18.2 mm). The vast majority of cases have been treated with transoral excision (94.8%). Recurrence after surgical excision has not been reported. Schwannoma of the tongue is a relatively rare tumor of the head and neck. Transoral resection allows for removal of this tumor in a

manner that precludes recurrence, avoids causing morbidity of tongue function, and remains the standard approach for the treatment of the vast majority of these tumors.

Keywords Schwannoma · Neurilemmoma · Lingual · Tongue

Introduction

Schwannoma (neurilemmoma) is a benign tumor of nerve sheath origin. These tumors can arise from any nerve covered with a Schwann cell sheath, which include the cranial nerves (except for the optic and olfactory), the spinal nerves, and the autonomic nervous system [1]. When the nerve of origin is small, its association with a given tumor may be difficult to demonstrate. On the other hand, if a larger nerve is the site of origin, the nerve fibers are found to be splayed out over the outer aspect of the capsule rather than incorporated within the mass of the tumor [2].

Approximately 25–45% of all schwannomas occur in the head and neck [3]. Of these, approximately 1–12% occur intraorally [4, 5] with the tongue being the most common site [5, 6]. Although several case reports of schwannomas of the tongue exist in the literature, there has been no comprehensive review of the literature since Hatziotis et al. [6]. We present two cases of schwannoma of the tongue and review the available literature of the last 51 years (1955–2006).

Materials and methods

A PubMed search of the terms “tongue schwannoma,” “lingual schwannoma,” “tongue neurilemmoma,” and “lingual neurilemmoma” was performed with the date range of

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Fig. 1 A well-circumscribed mass is located at right posterior tongue as illustrated by the axial T1 (a), sagittal T2 (b), and coronal T1 (c) magnetic resonance images

1955–2006. The search was limited to case reports in English. Schwannomas of the floor of mouth were not included unless the ventral tongue was also involved. All of the case reports had histologically confirmed the identity of the masses as schwannomas.

The following elements were extracted from the case reports for data analysis: age, gender, location of schwannoma (anterior one-third vs. posterior two-thirds of tongue), presenting symptoms, size of tumor, and treatment modality.

Case reports

Case report #1

An 19-year-old female presented to the Head and Neck Surgery Clinic with complaints of a slowly enlarging painless mass in the back of her throat. A submucosal mass was identified at the right posterolateral tongue. Magnetic resonance (MR) imaging was obtained which illustrated a well-circumscribed mass in the right posterolateral tongue that was isointense on T1, hyperintense on T2, and enhanced with gadolinium (Fig. 1a–c). The patient was taken to the operating room for a transoral excision under general anesthesia. The mass was submucosal and once a mucosal flap was raised, the tumor was readily shelled out using blunt dissection. The mass had a smooth surface, was tan-white in color, and measured $18 \times 13 \times 11$ mm. Final pathology was consistent with schwannoma (Fig. 2a–c).

Case report #2

A 77-year-old male was referred to the Head and Neck Surgery Clinic after his dentist noted an 1 cm firm, sessile mass at the right lateral border of his tongue. The patient reported that the mass had been there for approximately

5 years and had not grown significantly over those years. The patient denied any pain associated with the mass, dysphagia, or change in voice. The mass was excised in clinic under local anesthesia. The mass was gray-tan in color, smooth in texture, dome-shaped, and measured $7 \times 5 \times 5$ mm. A final diagnosis of schwannoma was ascribed.

Results

A PubMed search from 1955 to 2006 identified 124 cases of schwannoma of the tongue (Table 1). Of the 126 cases (including the two patients reported in this paper), 106 identified the gender of the patient which showed an almost equal gender predilection with 50 males (47.2%) and 56 females (52.8%). Tongue schwannomas were also noted at all age ranges, but had a significantly higher incidence between the second and fourth decades of life (Fig. 3).

The location of the schwannoma was categorized as either anterior two-thirds or posterior one-third of the tongue. This classification was possible in 56 of the 126 cases with 37 cases occurring anteriorly and 19 cases identified posteriorly.

Descriptions of the patient's clinical symptoms were only found in 57 case reports. The majority of patients presented with an asymptomatic mass (39/57; 69.6%). Of the patients that did present with a symptomatic mass, the most common complaints were throat pain or discomfort, dysphagia, and voice change (Table 2). When the location of the schwannoma and the presence of symptoms were examined, a correlation was identified. Of the 19 patients with posterior one-third tumors, 12 patients reported symptoms (63.2%). On the other hand, in the anterior two-thirds group, only five patients reported symptoms (13.5%).

The exact size of the schwannoma was documented in 53 patients. The average size of the excised tumor in all

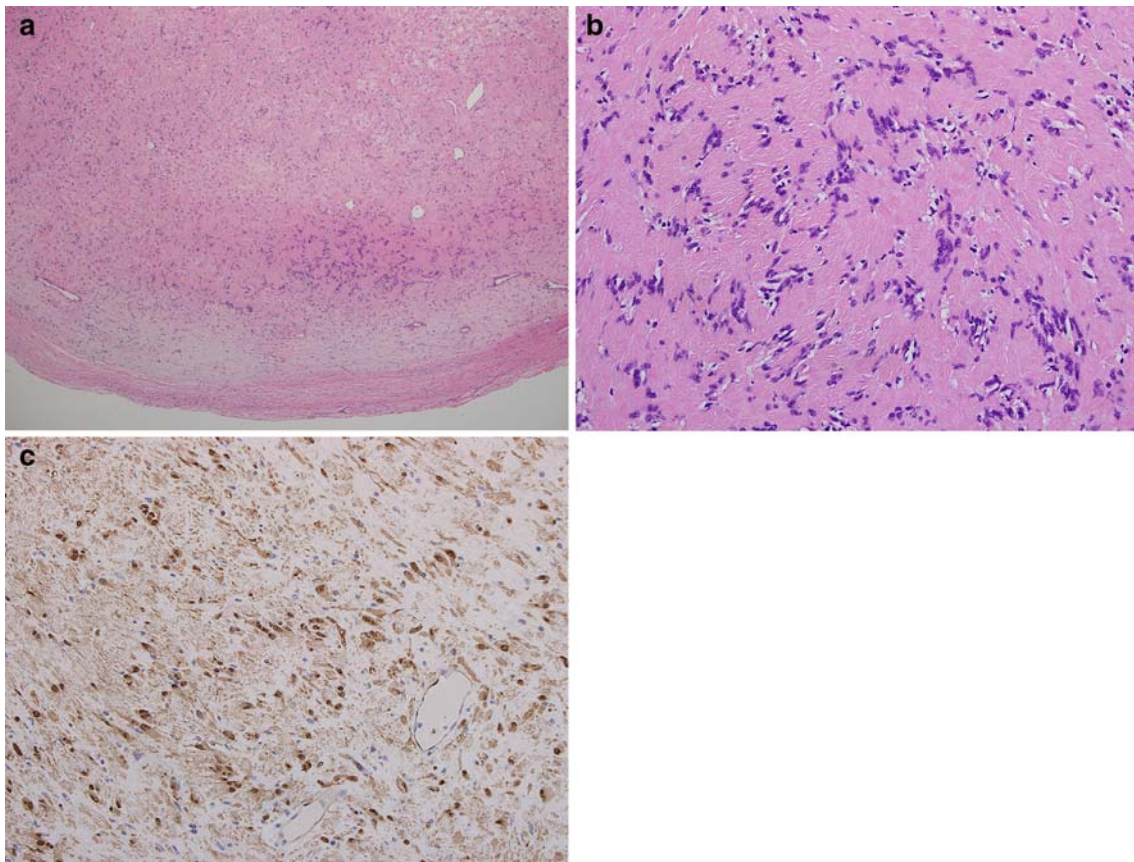


Fig. 2 Schwannoma of the tongue. **a** At low power (H&E, $\times 4$), both Antoni A and Antoni B morphologies can be seen. **b** At high power (H&E, $\times 20$), palisading nuclei and Verocay bodies are noted. **c** S-100 is diffusely positive (S-100, $\times 20$)

documented patients was 24.04 mm. When the size of the tumor was examined between the symptomatic and asymptomatic groups, a clear distinction was noted. The average schwannoma size in the asymptomatic groups was 18.2 versus 33.0 mm in the symptomatic group.

Almost all schwannomas were excised via a transoral approach (55/58; 94.8%). The remaining patients had their tumors excised via a transhyoid, suprahyoid, or submandibular approach. One patient had a transoral laser excision of the schwannoma.

Discussion

Schwannoma is a slow-growing benign tumor of the nerve sheath. It originates from the Schwann cell of the peripheral, autonomic, and cranial nerve. It is usually a single, circumscribed, firm, painless lesion of variable size [7]. The etiology of schwannomas is unknown. Approximately 25–45% of all schwannomas occur in the head and neck [3] with the parapharyngeal space being the most common site [8]. Less commonly, schwannomas present in the oral cavity, and of this subset, the tongue is the most frequently involved [5, 6].

In this literature review of the past 51 years, we have identified 126 cases of tongue schwannomas and elucidated some of their characteristics. Schwannomas of the tongue demonstrate an equal gender predilection. The majority of them present as a painless mass. However, as they approach 3 cm in greatest dimension, they are more likely to produce symptoms such as throat discomfort, dysphagia, and voice changes. Moreover, if the schwannomas arises in the posterior two-thirds of the tongue, they are more likely to produce debilitating symptoms.

Schwannomas usually present as a solitary lesion. When multiple, however, they can be associated with neurofibromatosis. The differentiation between schwannoma and neurofibroma is essential because an apparently “solitary” neurofibroma may be a manifestation of neurofibromatosis [9]. Approximately 15% of patients with neurofibromatosis will have malignant transformation in one or more lesion, which is in marked contrast to the typical behavior of a schwannoma [10].

Histologically, schwannomas display several features. Virtually, all of these tumors are encapsulated. Beneath this capsule, two main patterns are seen intermingled but sharply defined from each other. The first pattern is referred

Table 1 Patient and tumor characteristics of tongue schwannomas

Author	Year	Age/ gender	Location of tumor	Size (greatest dimension, mm)	Presenting symptoms	Surgical approach
Cameron [24]	1959	25M	Anterior	15	Painless mass	Transoral
Mercantini et al. [25]	1959	22M	Anterior	10	Intermittent pain	Transoral
Kragh et al. [2]	1960	Report of 5 cases; patient and tumor characteristics not described				
Chadwick [26]	1964	20F	Posterior	22	Globus	Transoral
Craig [27]	1964	8F	Posterior	30	Painless mass	Transoral
Chhatbar [28]	1965	29M	Posterior	50	Throat discomfort	Transoral
Pantazopoulos [29]	1965	25M	Anterior	<10	Painless mass	Transoral
		45F	Posterior	45	Dysphagia, change in voice	Transoral
Firfer et al. [30]	1966	28F	Anterior	30	Anterior	Transoral
Hatziotis et al. [6]	1967	25M	Posterior	“Hazelnut size”	Painless mass	Transoral
		60F	Anterior	“Pea size”	Painless mass	Transoral
Report of 46 more cases (18 males, 28 females); patient and tumor characteristics not described						
Oles et al. [31]	1967	52M	Anterior	10	Painless mass	Transoral
Paliwal et al. [32]	1967	32M	Anterior	25	Painless mass	Transoral
Uj [33]	1967	13F	n/a	n/a	Painless mass	Transoral
Crawford et al. [34]	1968	24M	Anterior	10	Painless mass	Transoral
		23M	Anterior	5	Painless mass	Transoral
Bititci [35]	1969	40M	Anterior	25	Slight discomfort	Transoral
Das Gupta [5]	1969	21F	Posterior	50	Pain	Transoral
Report of 7 more cases; patient and tumor characteristics not described						
Cherrick et al. [11]	1971	Report of 8 cases; patient and tumor characteristics not described				
Eversole et al. [12]	1971	58F	Anterior	25	n/a	Transoral
Sinha et al. [36]	1971	23M	Posterior	15	Odynophagia	Transoral
Mosadomi [37]	1975	19M	Anterior	30	Pain	Transoral
Swangsilpa et al. [38]	1976	24M	Anterior	30	Painless mass	Transoral
Sharan et al. [39]	1978	30F	Anterior	15	Change in voice	Transoral
Barbosa [40]	1984	12M	n/a	5	Painless mass	Transoral
Piatelli et al. ^a [18]	1984	18F	Anterior	n/a	Painless mass	Transoral
Akimoto et al. [41]	1987	14M	Anterior	10	Painless mass	Transoral
Malden [42]	1988	51F	Anterior	n/a	Painless mass	Transoral
Siar et al. [43]	1988	17F	Posterior	30	Painless mass	Transoral
		13F	n/a	44	n/a	Transoral
Wilson et al. [44]	1988	78F	Posterior	n/a	n/a	Transoral
Flickinger et al. [19]	1989	28F	Anterior	30	Painless mass	Transoral
Talmi et al. [45]	1991	75F	Posterior	10	Painless mass	Transoral
Lopez et al. [4]	1993	33M	n/a	12	Painless mass	Transoral
		24M	Anterior	6	Painless mass	Transoral
Williams et al. [46]	1993	28M	Anterior	5	Painless mass	Transoral
		58M	Anterior	10	Ulceration	Transoral
Haring [47]	1994	49F	Anterior	20	Painless mass	Transoral
Nakayama et al. [13]	1996	40F	Anterior	55	Painless mass	Transoral
Spandow et al. [48]	1999	37M	Posterior	79	Throat discomfort	Transoral
de Bree et al. [7]	2000	24F	Posterior	50	Dysphagia	Submandibular
Pfeifle et al. [49]	2001	30F	Anterior	10	Painless mass	Transoral
		18M	Anterior	20	Painless mass	Transoral
Mevio et al. [50]	2002	35F	Anterior	50	Painless mass	Transoral

Table 1 continued

Author	Year	Age/ gender	Location of tumor	Size (greatest dimension, mm)	Presenting symptoms	Surgical approach
Bassichis et al. [51]	2004	9M	Posterior	23	Snoring, difficulty breathing	Transoral
Cinar et al. [52]	2004	7M	Anterior	10	Painless mass	Transoral
Hwang et al. [53]	2005	23M	Anterior	28	Painless mass	Transoral
Vafiadis et al. [54]	2005	18M	Anterior	31	Painless mass	Transoral
Hsu et al. [21]	2006	39F	Anterior	10	Painless mass	Transoral
		39F	Posterior	40	Dysphagia	Transhyoid
		9M	Anterior	12	Painless mass	Transoral
		32F	Posterior	18	Globus	Transoral
		25M	Anterior	9	Painless mass	Transoral
		15F	Anterior	12	Painless mass	Transoral
		38M	Anterior	30	Painless mass	Transoral
		12F	Anterior	16	Painless mass	Transoral
		45M	Anterior	5	Painless mass	Transoral
Mehrzad et al. [22]	2006	20M	Posterior	50	Globus	Transoral
		49M	Posterior	22	Throat discomfort	Transoral CO ₂ laser
Ying et al. [20]	2006	26F	Posterior	40	Dysphagia, snoring, change in voice	Suprahyoid
Cohen and Wang	Current study	18F	Posterior	18	Painless mass	Transoral
		77M	Anterior	7	Painless mass	Transoral

^a Case report of malignant schwannoma of the tongue

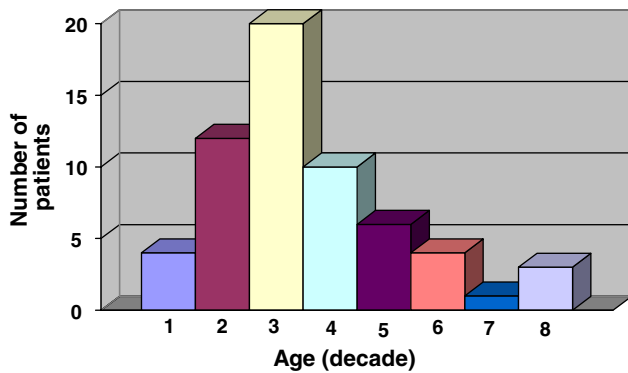


Fig. 3 Age at presentation of patients with tongue schwannomas

to as *Antoni type A* which consists of closely packed Schwann cells that form bundles or are arranged in rows with palisading, elongated nuclei. Free bands of amorphous substance between rows of nuclei constitute the *Verocay bodies*. The second pattern is known as *Antoni type B* and is composed of very loosely arranged Schwann cells set in a meshwork of reticulum fibers and microcysts [11]. In addition to these characteristic patterns, diagnosis is aided by immunohistochemical markers, S-100 and Leu 7, which support the Schwann cell nature of these tumors [4].

Two cases of schwannoma of the tongue were referred to as “ancient” schwannoma [12, 13]. The term “ancient”

Table 2 Presenting symptoms of patients with tongue schwannomas

Symptom	Number of patients (n = 56)
Painless mass	39
Throat pain/discomfort	7
Dysphagia	4
Change in voice	3
Globus sensation	2
Difficulty breathing	2
Snoring	2
Ulceration with pain	1

schwannoma was introduced by Ackerman and Taylor [14]. These authors, in addition to others, noted extensive degeneration within the schwannoma and attributed this change to the aging process of the schwannoma. Aside from nuclear atypia, additional changes associated with the degenerative process include formation of cysts, stromal edema, xanthomatous change, and fibrosis. The theorized mechanism is that increasing tumor size results in vascular insufficiency. This hypothesis is supported by the finding that the degenerative changes correlate with tumor size [15]. It is now generally accepted that all schwannomas can demonstrate some degenerative changes and the “ancient” variety probably represents the far end of the continuum of histologic appearance [16].

Several cases of malignant transformation of head and neck schwannomas have been reported [2, 17], including one occurrence in the tongue [18]. To support a diagnosis of malignant transformation in benign schwannoma, the following features should be confirmed: (a) the tumor demonstrates, to some extent, benign schwannoma; (b) the tumor contains unequivocal malignant foci as manifested by the presence of increased cellularity, numerous mitoses, anaplastic cells, and invasiveness; (c) transitional features between malignant and benign areas can be seen; and (d) the patient has no evidence of von Recklinghausen's disease [13].

The imaging modality of choice for schwannomas of the tongue is MR. MR is superior to computed tomography (CT) in several aspects. The MR image is not degraded by dental amalgam or the beam-hardening artifacts that plague CT scanning of the oral cavity. In addition, MR allows an accurate measurement of tumor size and precise localization in relation to other structures. On MR, tongue schwannomas appear isointense to muscle on T1-weighted images and homogeneously hyperintense on T2-weighted images. Moreover, these tumors usually appear smooth, well demarcated, and do not invade the surrounding musculature [19].

All reported cases of schwannomas of the tongue have been treated by surgical excision. The most common approach was the transoral route. This is an obvious choice for approaching these tumors since most are easily accessible via this route. Several other approaches have also been reported to have success including submandibular [7], suprahyoid pharyngotomy [20], and transhyoid [21] approaches. All of these approaches were used for base of tongue schwannomas that were deemed difficult to approach by the transoral route. More recently, the use of CO₂ laser for excision of a base of tongue schwannoma has also been reported [22]. The goal of surgical therapy is to complete resection. If this is accomplished, recurrence is rare [21].

In evaluating a patient with a slow-growing tongue mass that has been present for a long period of time, benign soft tissue neoplasms and reactive lesions need to be considered. In addition to schwannomas, the differential diagnosis should include neurofibromas, granular cell tumors, irritation fibromas, leiomyomas, rhabdomyomas, hemangiomas, lymphangiomas, lipomas, pyogenic granulomas, and benign salivary gland tumors [23]. Malignant etiologies should always be considered, but are unlikely to present with the slow clinical course that is typical of a schwannoma.

Conclusions

Schwannoma of the tongue is a relatively rare tumor of the head and neck. Transoral resection allows for removal of

this tumor in a manner that precludes recurrence, avoids causing morbidity of tongue function, and remains the standard approach for treatment of the vast majority of these tumors. The chance of malignant transformation of these tumors is exceedingly unlikely.

Conflict of interest statement None.

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