



Case Report

Case Report and Literature Review on Tongue Schwannoma

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Abstract: Schwannoma is a neoplasm originating from cells surrounding and insulating axons in peripheral nerves. It usually presents benign behaviour with slow growth. A significant portion of cases occur in the head and neck region but rarely in the oral cavity, where the tongue is the most frequently affected organ. This article describes the case of a man presenting an asymptomatic mass on the dorsal aspect of the tongue that sought attention at the Integrated Therapies in Otorhinolaryngology Department of the Policlinico Campus Bio-Medico Foundation in Rome. After clinical and radiological examinations, the patient underwent surgical treatment under local anaesthesia. A literature search was conducted on PubMed and Google Scholar. Only complete case reports published in English from 1923 to 2023 were selected. A total of 183 cases were considered after the selection of relevant articles and the elimination of duplicates. The resulting data confirm that the most common presentation of this pathology consists of a painless mass in the oral tongue; usually, this lesion is removed surgically via a transoral approach, but different variations were described depending on the dimensions and position of the lesion.

Keywords: schwannoma; neurinoma; neurilemmoma; tongue; local anaesthesia



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1. Introduction

Schwannoma, formerly known as a neurilemmoma, is a benign tumour developing from Schwann cells. This pathology was identified by Verocay, initially named “Neurinom”, in 1910 [1]. Its name derives from the mass proliferation of Schwann cells and the basement membrane structure separating cells and the interstitium. No discernible variation in schwannoma occurrence depending on gender has been observed, and its aetiology is linked to the loss of function of the NF2 gene on chromosome 22, encoding for the protein Merlin. It is linked to genetic pathologies like Neurofibromatosis type 2 (NF2) and Carnay Complex [2]. Schwannomas can appear at any age; however, they are most frequently seen in people between the ages of 10 and 40, with an overall incidence suggested around 1.2:100,000 [2]. A portion between 25 and 45 per cent of all schwannomas occur in the head and neck region [3]. The tongue is the most common location for schwannomas in the oral cavity, which accounts for around 1% of all schwannomas [4]. Most schwannomas are asymptomatic but sensory and motor disturbances are possible and most of the symptomatology is ascribable to its mass effect [2].

Schwannoma usually emerges as a hard–elastic mass with a smooth nodular outline, sometimes with a visible nerve of origin. A tan or yellow colour is visible on the cut surface, sometimes with areas of haemorrhage and cystic change. Microscopically, a capsule separating the mass from the surrounding tissues is observable; the Schwann cells display spindle morphology and can be organized into fascicles forming an “Antoni A” area or can be arranged in a loose microcystic area, defined as an “Antoni B” area. Other histological elements, defined as “Verocay bodies”, display patterns of parallel

nuclear arrays [5]. Schwannomas can be categorized into distinct subgroups based on histological findings, such as “Ancient”, “Plexiform”, “Cellular” [6], “Melanotic” [7], and “Psammomatous” [8] variants.

2. Detailed Case Description

A 61-year-old man visited the outpatient clinic of the Integrated Therapies in Otorhinolaryngology Department of the Policlinico Campus Bio-Medico Foundation in Rome, presenting an asymptomatic mass on the dorsal aspect of the tongue for approximately 3 months. The clinical history of the patient reported tonsillectomy, inguinal hernia reduction, abdominal lipoma removal, cutaneous melanoma, and kidney stones. No clinical features, such as skin pigmentation abnormalities and Lisch nodules, or familial predisposition related to neurofibromatosis were reported. A small ulcer measuring approximately 3 mm was identified on the dorsum of the tongue within a nodular area of approximately 1 cm along the median raphe. A contrast-enhanced magnetic resonance imaging (MRI) was performed, identifying a $13 \times 7 \times 11$ mm mass located medially in the posterior third of the oral tongue (Figures 1–3). The patient agreed to undergo surgical treatment and provided informed consent for the use of his data.

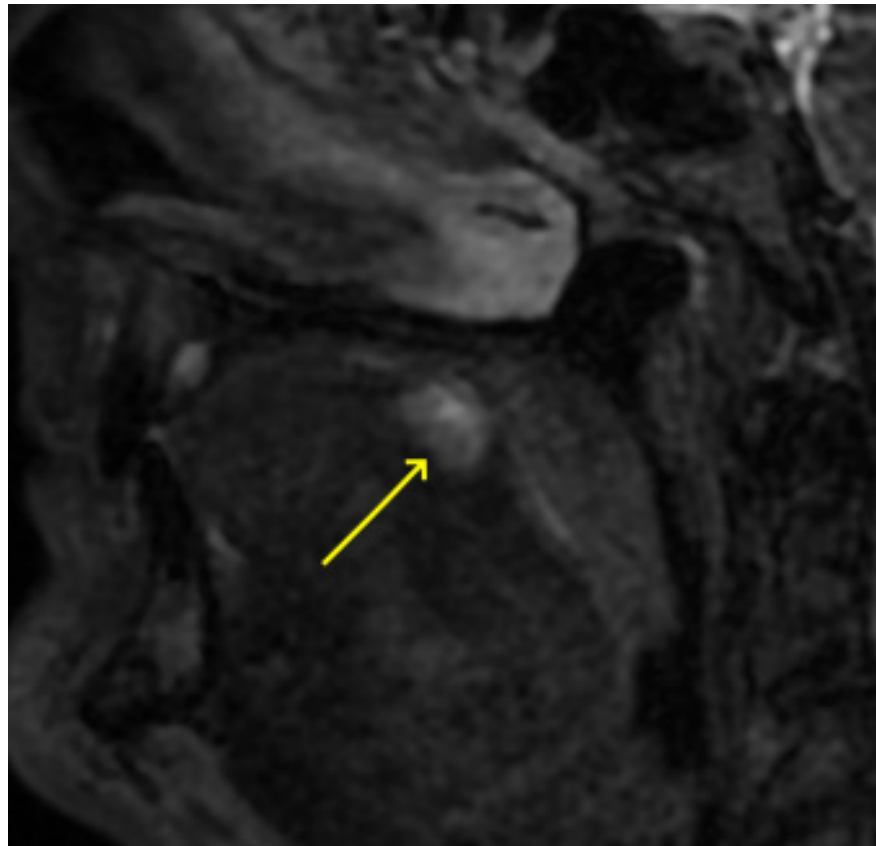


Figure 1. T2-weighted sequence, sagittal view, showed an oval-shaped lesion (yellow arrow) presenting a heterogeneous hyperintense signal between the central and the posterior thirds of the tongue.

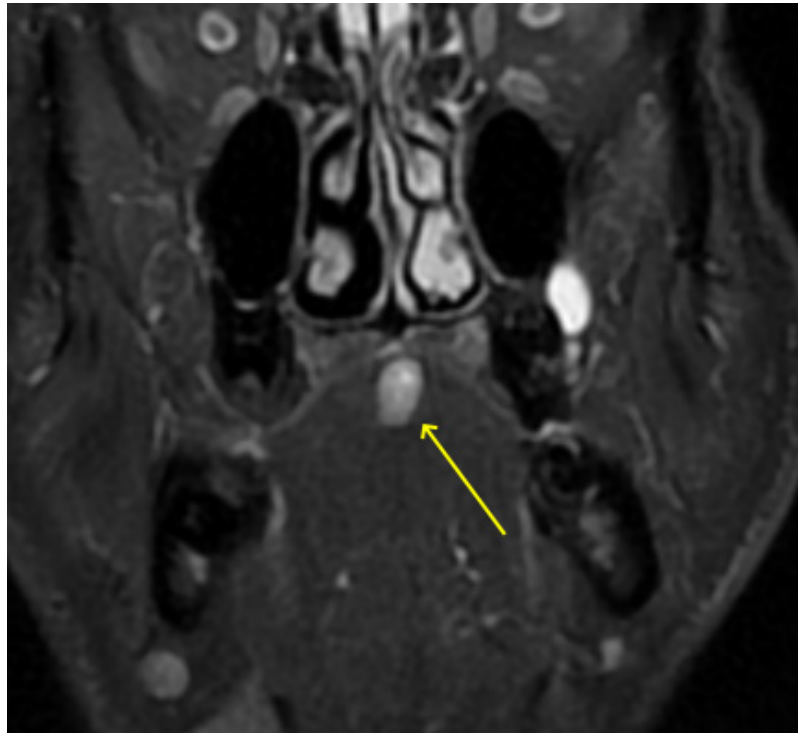


Figure 2. Short Tau Inversion Recovery (STIR) sequence, coronal view, showing an oval-shaped lesion (yellow arrow) in the median portion of the tongue presenting a hyperintense signal.

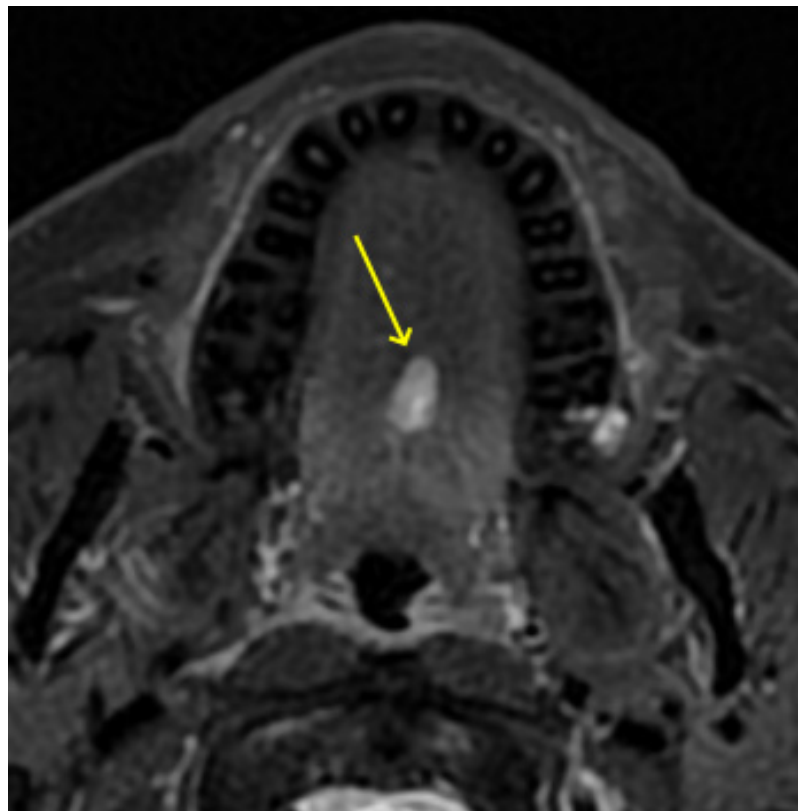


Figure 3. STIR sequence, axial view, showing an oval-shaped lesion (yellow arrow) presenting a hyperintense signal in the median portion of the tongue, between the central and the posterior thirds of the tongue.

The procedure was performed under local anaesthesia through the perilesional infiltration of lidocaine and adrenaline. The mass was easily separable from the surrounding tissues for the majority of its surface and it appeared whitish, with a firm and elastic consistency. It measured $2 \times 18 \times 15$ mm (Figures 4–7). No complications were reported during the 6 months of follow-up (Figure 8).

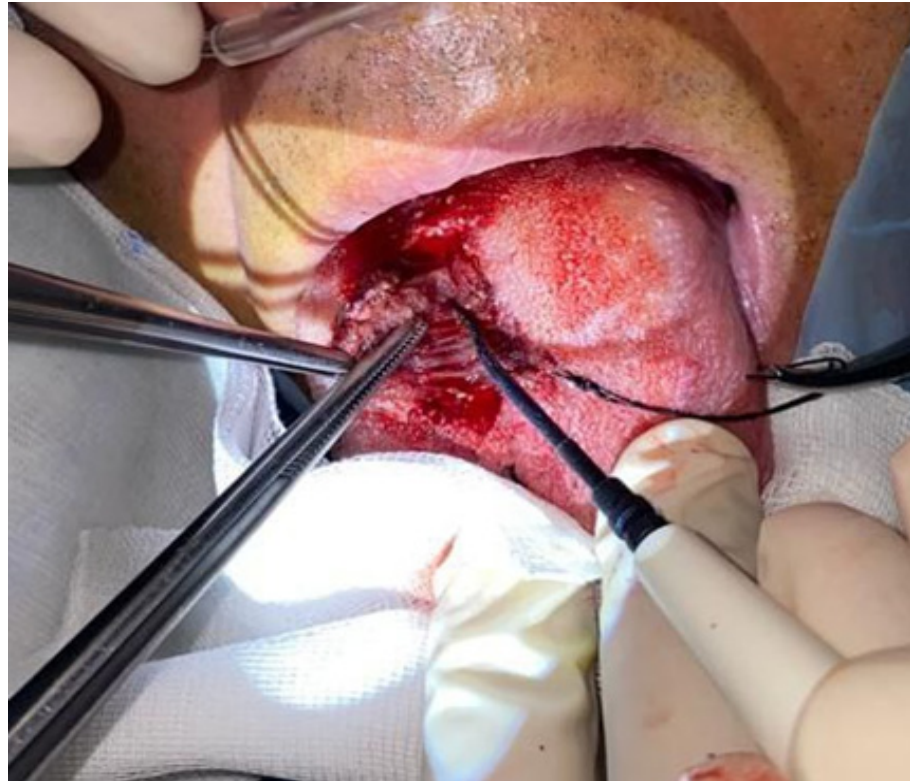


Figure 4. Lesion excision under local anaesthesia.



Figure 5. Macroscopic aspect of the lesion after excision.



Figure 6. Tongue after the excision.



Figure 7. Macroscopic aspect of the lesion during the procedure.

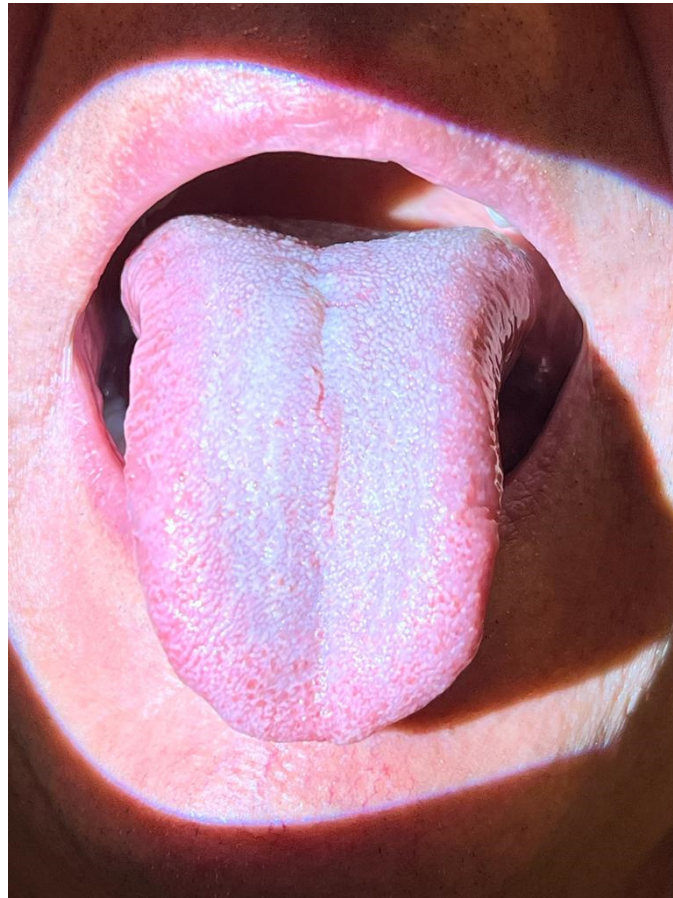


Figure 8. Tongue at 6 months follow-up.

A histological analysis documented the presence of a proliferation of spindle cells with nuclei arranged in palisades. An immunohistological analysis reported S100 and Sox10 positivity. The overall anatomopathological findings are consistent with a diagnosis of Schwannoma (Figures 9–11).

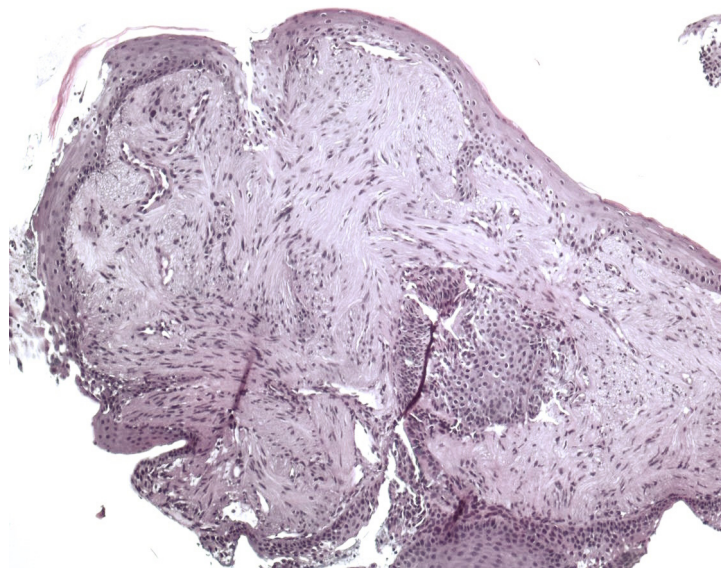


Figure 9. Haematoxylin eosin (100×).

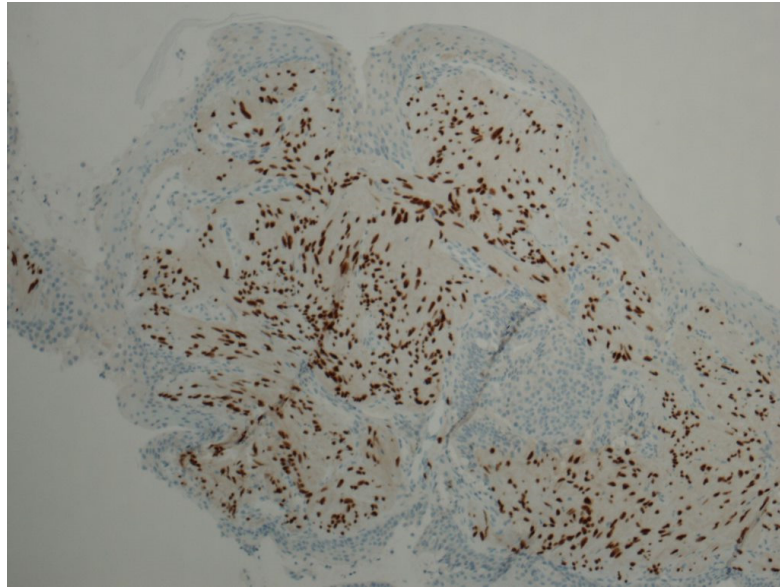


Figure 10. Sox-100 positivity (100×).

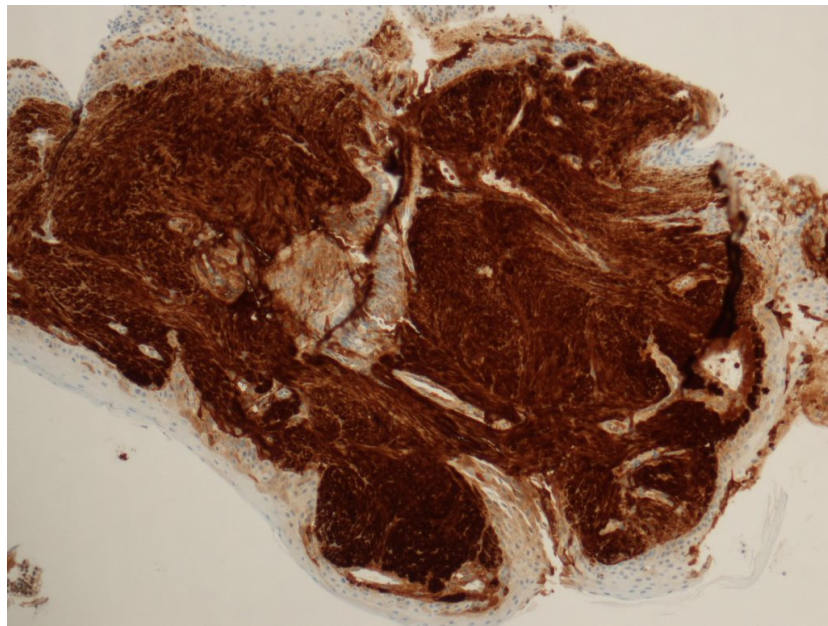


Figure 11. S100 positivity (100×).

3. Discussion

3.1. Literature Review

A literature review was performed using the keywords “lingual schwannoma”, “tongue schwannoma”, “lingual neurilemmoma”, and “tongue neurilemmoma” on the PubMed and Google Scholar databases. A secondary search was conducted in the bibliographies of the identified articles. Only complete case reports published in English from 1923 to 2023 were selected. A total of 908 results were considered, and after excluding works that did not meet the inclusion criteria, duplicates, and articles unrelated to the purpose of the review, 136 articles were obtained. An additional 19 articles were identified through the citations of the initially obtained works. Six articles were excluded due to insufficient information regarding the described cases. A total of 183 cases were recorded; for each case, the sex, age, dimensions, site, presenting symptoms, type of surgical approach, and follow-up period were considered Chart 1 (Supplementary Table S1).

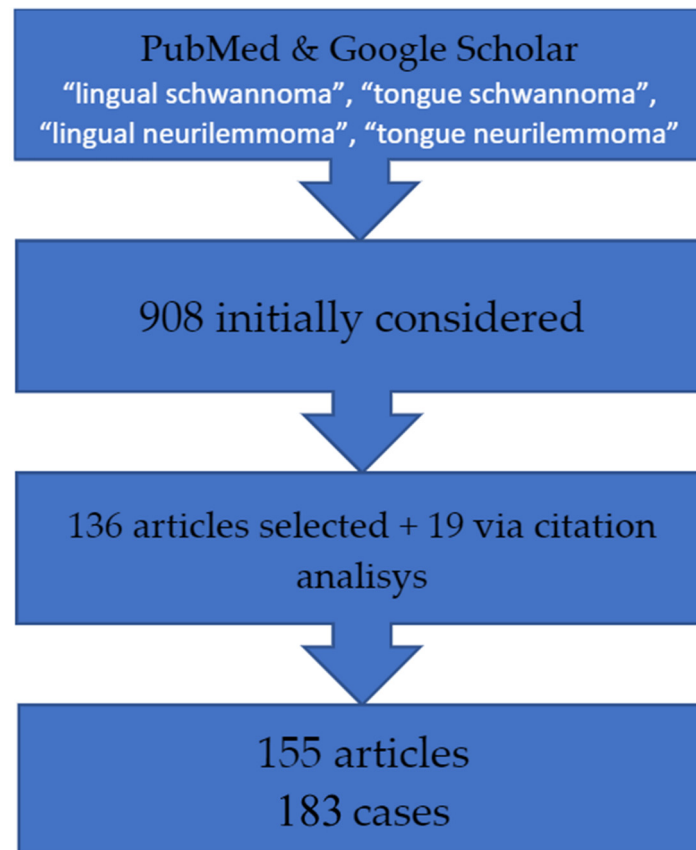
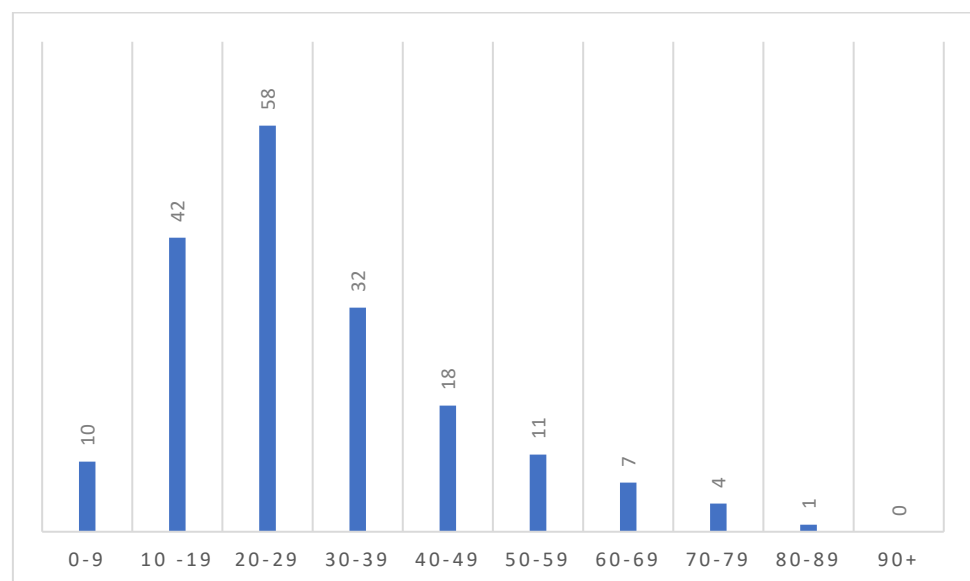


Chart 1. Flowchart outlining the paper selection.

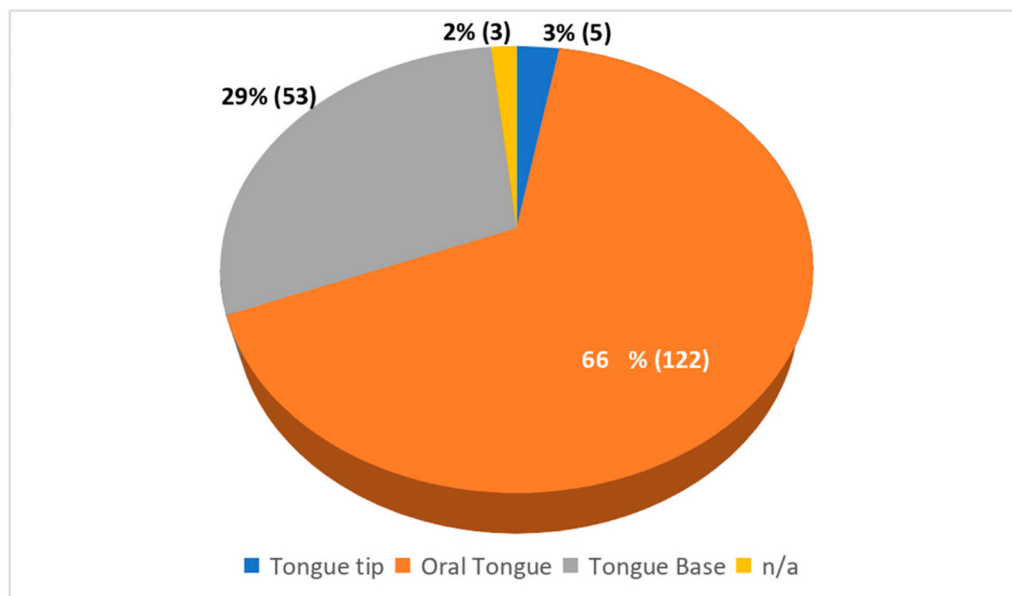
3.2. Pathology Presentation

The male-to-female ratio resulted as 1.1:1, with a mean age of 29.5 ± 15.8 years, ranging from 4 [9] to 81 years [10]. In 35 cases (19.4%) (Graph 1), patients were 16 years or younger. The average size of described schwannomas is 3.2 ± 2 cm, with a maximum of 16 cm along the major axis [11]. No episodes of recurrence were observed among the considered cases.

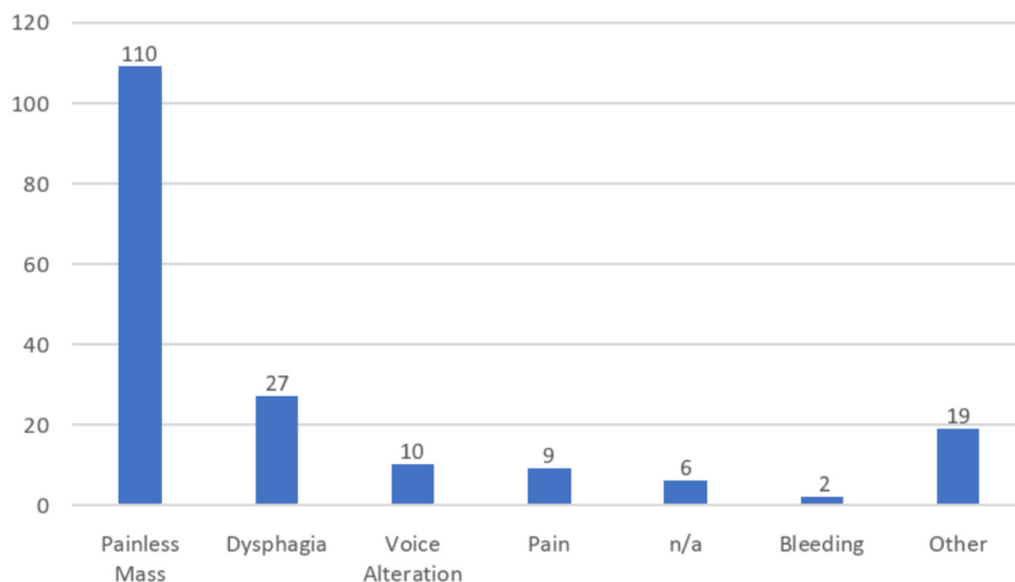


Graph 1. Age of reported patients.

The most affected region of this pathology is the lingual body (66.6%—122 cases), followed by the lingual base (29%—53 cases), while a minority manifested at the tip (3%—5 cases); in 3 cases, the site was not specified (Graph 2). In the majority of reported cases (65%—120 cases), the only reported symptom was a painless mass effect. The second most reported symptom was dysphagia (14.5%—27 cases), often associated with onset at the base of the tongue (Graph 3).



Graph 2. Sites of onset (n/a: not reported/absent).

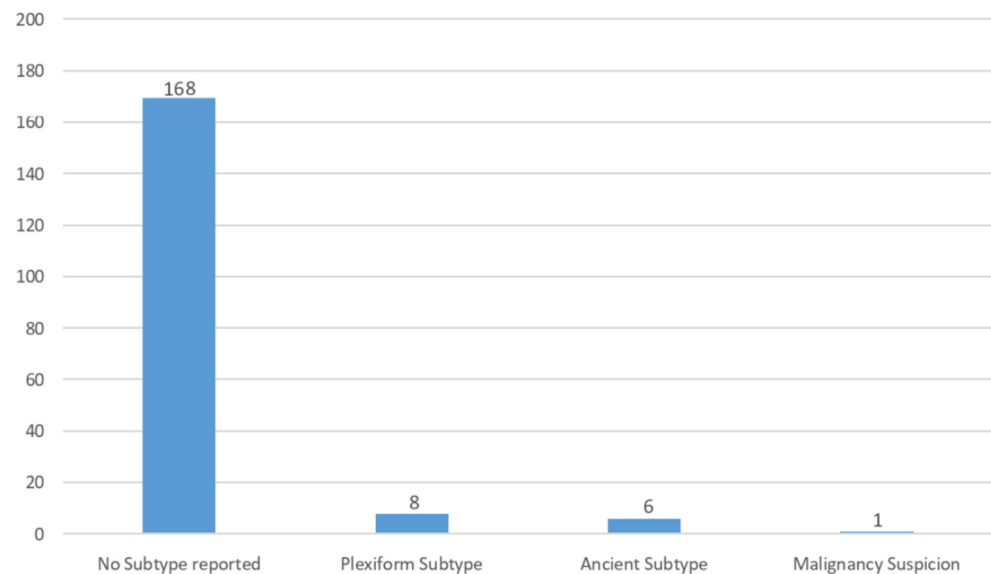


Graph 3. Presenting symptoms (n/a: not reported/absent).

3.3. Histological Variants

Schwannomas of big dimensions or that underwent prolonged physical stress may present areas expressing a degenerative change with micro-vascular thrombosis and degenerative nuclear atypia; when such anomalies are dominant among the histological specimen, the schwannoma is defined as “Ancient” [5]. Another notable histological variant of schwannoma reported in the literature is the “Plexiform” variant, which is more common in dermal schwannomas and presents an intraneural multinodular growth pattern,

typical in young individuals. Histologically, it shows a prevalence of Antoni A areas and immunoreactivity to the glial fibrillary acidic protein (GFAP) [6]. The scarcity of reported subtypes could be attributed to the heterogeneity in the field of scientific articles found. The relative incidence among the reported subtypes is attributable to the lower attention given to the ancient variant, which could be under-reported (Graph 4). The only case reported of malignant transformation lacks immunohistochemical evidence to support the suspicion provided by histological observation. Six cases of “Ancient schwannoma” were identified in this literature review [12–17]. Eight cases of “Plexiform schwannoma” were reported [18–20], with one case associated with NF2 [21]. One case of malignant behaviour in a schwannoma was documented [22].



Graph 4. Histological variants reported.

3.4. Surgery and Anaesthesia Techniques

Regarding imaging, preoperative biopsy, surgical technique, and anaesthesia, only the cases published in the last 24 years were considered to reduce the bias brought by the evolution of techniques over time. This decision reduced the number of considered publications to 118, describing 139 cases.

The transoral surgical approach was adopted in the majority of the cases, while the transcervical approach was adopted in cases with considerable proportions of tongue base pathologies. The use of transoral robotic surgery (TORS) [23], laser [10,24], and endoscopy [25] has been reported. In total, 11 cases were approached with transcervical methods: 3 cases with a submandibular approach [26–28], 3 with paramedian mandibulotomy [29–31], 2 with lateral pharyngectomy [32,33], one suprahyoid approach [34] and one transhyoid approach [35] were mentioned, in one case mandibulotomy was performed [36]. In different cases, a temporary tracheotomy was adopted to preserve the airways [29,33,36,37]; in one case, guided nasotracheal intubation using a flexible bronchoscope within the ventilation tube is described [38]. Local anaesthesia was reported in seven cases, mostly in patients with small-sized intraoral or tongue tip pathologies [39–44]. No intraoperative or postoperative complications were reported.

In 75 cases, no follow-up period was mentioned. The reported follow-up periods varied from 1.5 months to 11 years and 5 months; the median period resulted in 12 months.

The preoperative imaging exams considered were lingual ultrasound, computer tomography (CT), and MRI; in some cases, more than one exam was used on the same patient. In 50 cases, none of these exams was reported. The preferred preoperative exam was MRI, used in 48 cases, followed by CT, used in 31. Tongue ultrasound use was reported in four cases.

In 29 cases, a preoperative biopsy was performed, with the diagnosis of schwannoma. When the lesion has small dimensions and appears easily separable from the tongue, many clinicians prefer to perform an excisional biopsy without preoperative imaging.

3.5. Strengths and Limitations

Overall, this study contributes to the existing literature by providing a comprehensive overview of tongue schwannoma, its clinical manifestations, and management approaches. The findings underscore the importance of proper preoperative assessment and tailored surgical interventions to achieve favourable outcomes for patients with tongue schwannoma.

The selection of publications only in the English language and using only two search engines reduced the number of cases considered. The time frame taken into account exposes different methodologies in the diagnostic and therapeutic choices; this variability was partially managed considering only the last 24 years for imaging, surgical technique, and anaesthesia. The risk of human error in the data collection is also another possible cause of limitations for this article.

4. Conclusions

The findings of this review confirm that tongue schwannoma predominantly affects individuals between their third and fifth decades of life, with a similar incidence among both sexes. The intraoral portion of the tongue is the most frequently affected site. Schwannomas often presented as asymptomatic masses, although problems with deglutition were commonly reported when the tongue base was involved. The most commonly used preoperative exam is reported to be MRI. In some cases, it has been replaced by CT, probably due to availability. The transoral approach is the preferred method due to its less invasive nature. The recent introduction of TORS and laser excision has been reported but does not appear to be widely employed. A combined approach has been used for particularly expansive lesions.

Further research could employ the methodology of this article to schwannomas in other anatomical regions. The integration of the gathered data with those reported in this article would contribute to the understanding of the epidemiology and therapeutic strategies of schwannoma.

Supplementary Materials: The following supporting information can be downloaded at <https://www.mdpi.com/article/10.3390/ohbm5020011/s1>: Table S1: Cases considered from 1923 to 2023 on the PubMed and Google Scholar databases. Refs [45–160] are cited in the Supplementary Materials.

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