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Case Report

Lingual schwannoma of submandibular region - a unique case in a rare location: a case report and literature review

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ABSTRACT

Schwannomas are uncommon benign nerve sheath tumors that rarely arise in the oral cavity. Among those found in the oral cavity, tongue is the predominant site of involvement. Lingual schwannomas originating in the submandibular region are exceptionally rare and pose a challenge in diagnosis owing to their nonspecific presentation and resemblance to salivary gland pathologies. Authors report the case of a 15-year-old girl presenting with a two-year history of a painless, progressively enlarging swelling in the left submandibular region, initially suspected to be a submandibular gland lesion. Radiological evaluation with ultrasonography and contrast-enhanced MRI demonstrated a well-defined, lobulated mass adjacent to the gland. The patient underwent surgical excision via a transcervical approach, that revealed the tumor arising from the lingual nerve near the submandibular ganglion. Histopathological examination confirmed the diagnosis of schwannoma, characterized by biphasic Antoni A and Antoni B areas with nuclear palisading. A literature review revealed 125 reported cases of lingual schwannomas, with no gender predilection and a wide age range. Given their infrequent and varied clinical presentation, it is emphasized that lingual schwannomas be considered in the differential diagnosis of submandibular and tongue-associated swellings. Complete surgical excision remains the treatment of choice and offers an excellent prognosis with minimal recurrence risk.

Keywords: Lingual schwannoma, Neurilemmoma, Submandibular schwannoma, Benign nerve sheath tumor

INTRODUCTION

Neurogenic tumors, including neurofibromas and schwannomas, represent a small fraction of head and neck neoplasms. Among these, schwannomas also referred to as neurilemmomas are even less frequently encountered.[1] These tumors are composed entirely of Schwann cells, are typically solitary, well-encapsulated, and slow-growing.[2] Extracranial schwannomas account for approximately 25–48% of all cases in the head and neck region; however, only about 0.2–1% of these tumors originate within the oral cavity, where the tongue represents the most frequently affected site3.[3] Authors report a case of a 15-year-old, with lingual schwannoma, initially diagnosed as a submandibular gland swelling.

CASE REPORT

A 15-year-old girl presented with a two-year history of a painless, progressively enlarging swelling on the left side of the neck. She reported no pain, altered sensation, or functional disturbances of the tongue. Examination revealed a firm, non-tender, well-circumscribed mass in the left submandibular region. The mass was also detectable on bimanual palpation. Ultrasonography revealed a well-defined, homogeneously hypoechoic nodular lesion in the left submandibular region, measuring 4.0×2.0 cm, inseparable from the submandibular gland, with no significant internal vascularity. Contrast-enhanced MRI (CEMRI) (Figure 1 and 2) showed a well-defined, lobulated soft tissue lesion measuring 3.7×2.6×2.4 cm in the left submandibular

space, anterosuperior to the submandibular gland. The lesion appeared heterogeneously hyperintense on T2-weighted and hypointense on T1-weighted images (Figure 1 and 2). Fine needle aspiration cytology (FNAC) was inconclusive, revealing benign salivary tissue. Surgical excision was performed via a transcervical approach under general anesthesia. A 5 cm skin crease incision was made 2 cm below the angle of the mandible. Subplatysmal flaps were elevated, and the marginal mandibular nerve was preserved using the Hayes Martin maneuver. On dissection, the tumor was found to arise from lingual nerve fibers near the submandibular ganglion and was emerging from medial to the submandibular gland. The tumor was excised in toto and sent for histopathological examination. The postoperative course was uneventful. Histopathological examination revealed a 6×3×2 cm grey-brown soft tissue mass. Microscopy showed an encapsulated tumor with biphasic architecture: hypercellular Antoni A areas and myxoid, hypocellular Antoni B areas, indicated by blue and red arrows in Figure 3 and 4 respectively. Nuclear palisading around fibrillary processes was noted, strongly indicative of schwannoma.

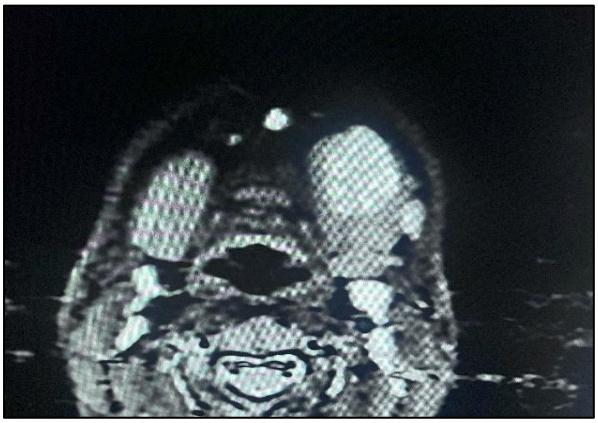

Figure 1: T2 weighted axial MR image of well-defined hyperintense lesion in left submandibular region.

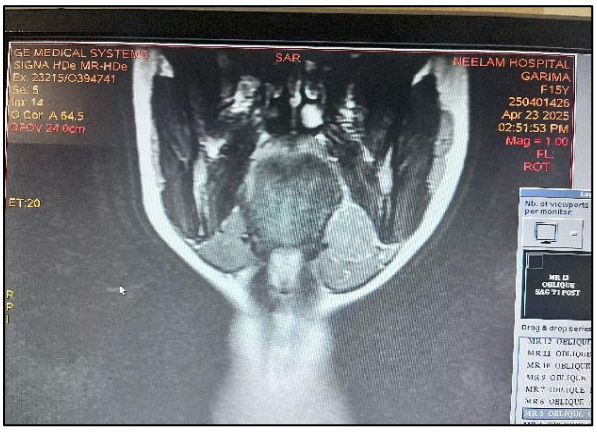

Figure 2: T1 weighted coronal MR image of well-defined hypointense lesion in left submandibular region.

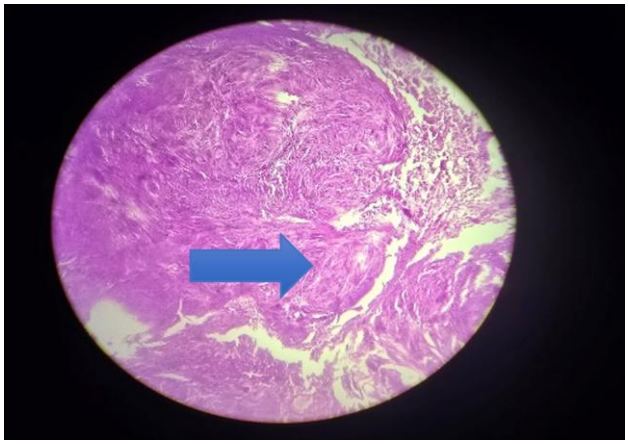

Figure 3: Antoni A cells.

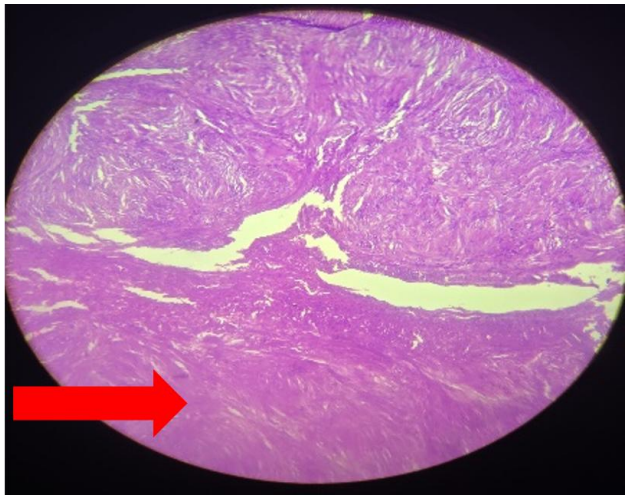

Figure 4: Antoni B cells.

DISCUSSION

Schwannomas, first identified by Virchow in 1908 and later described by Verocay, are uncommon benign nerve sheath tumors arising from Schwann cells, which insulate peripheral, autonomic, or cranial nerves-excluding the olfactory and optic nerves.[4,5] Schwannomas grow along the length of the nerve, thus assuming a fusiform appearance. They don't compromise the functional and morphological integrity of the nerve, allowing an easy surgical separation from nerve of origin.

Since, neurofibromas also have a similar perineurium origin, the diagnosis between the two is established histopathologically, as schwannomas are composed solely of Schwann cells while neurofibromas contain a mix of cell types.[6,7] They can occur either sporadically or as a part of genetically inherited disorders like neurofibromatosis type 1, type 2 and schwannomatosis.[8] Etiology remains unclear, though some factors like external trauma, chronic irritation, exposure to radiation and spontaneous growth can be implicated. Their occurrence in frequently injured areas like the tongue supports trauma as a possible factor.[9,10]

Table 1: Clinical presentation.

Symptom	Cases
Painless lump/ swelling/ nodule/ mass	92
Ulcer	2
Foreign body sensation in throat	8
Dysphagia	8
Odynophagia	5
Change in voice	2
Snoring	2
Deviation of tongue	2
Paraesthesia	2
Papillomatous/ exophytic growth	2
Dysgeusia	1

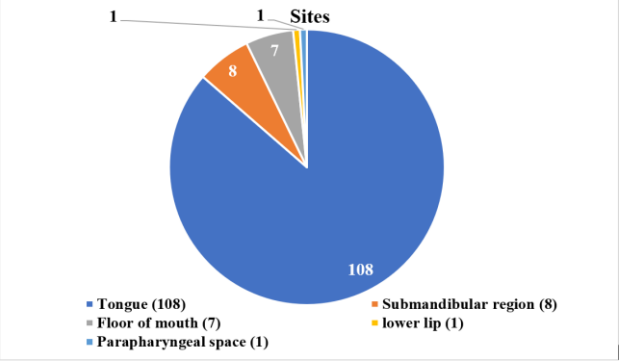

Figure 5: Site of origin of schwannoma.

A review of 125 lingual schwannoma cases from 1959 to 2024 revealed no gender predilection and a mean age of 30.77 years, with cases ranging from 7 to 82 years. Most cases involved the tongue (108 cases), particularly the anterior two-thirds (77 cases), while only eight involved the submandibular region. The other sites of involvement are represented in Figure 5. In over half of tongue schwannoma cases, the nerve of origin could not be identified intraoperatively; when specified, the lingual nerve was most commonly involved. Other nerves of origin were hypoglossal in 2 cases and parasympathetic fibers of submandibular ganglion in 1 case. The different presentations are summarized in Table 1. Radiologically, a schwannoma can be identified as a well circumscribed tumor with homogenous/ heterogenous enhancement on CT scan12. The most useful investigation is MRI, showing isointense signal on T1 and high signal intensity on T2 weighted images.[13]

The differential diagnosis is broad, including carcinomas, sarcomas, granular cell tumors, neurofibromas, salivary gland tumors, and others, making histopathological examination essential for definitive diagnosis14. Microscopically, there are 2 identified patterns- Antoni A and Antoni B. More densely packed spindle cells are found in Antoni pattern, whereas Antoni B has a more myxoid stroma. A positive staining for S-100, a neural crest marker, is important for diagnosis.[15] Complete surgical excision is the treatment of choice, as schwannomas are radio-resistant and recurrence is rare if removal is complete.[16-18] The surgical approach depends on tumour size and location.[19] In this case, a submandibular swelling was found intraoperatively to arise from the lingual nerve and was confirmed histologically as a schwannoma, underscoring the importance of considering nerve sheath tumours in the differential diagnosis of neck swellings.

CONCLUSION

Lingual schwannomas of the submandibular region are rare, benign, and typically slow-growing tumors that can present across a wide age range without gender predilection. Their diagnosis is challenging due to nonspecific imaging features and a broad differential, making histopathological confirmation essential. Surgical excision remains the treatment of choice, with excellent prognosis and low recurrence. Awareness of this entity and careful evaluation are crucial for accurate diagnosis and management.

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