Schwannoma of Floor of the Mouth: A Rare Case at Unusual Location

Shweta Channavir Saddu¹, D Sujatha², Nidhin J Valappila³

¹Post-graduate Student, Department of Oral Medicine & Radiology, The Oxford Dental College, Hospital & Research Centre, Bengaluru, Karnataka, India, ²Professor, Department of Oral Medicine & Radiology, The Oxford Dental College, Hospital & Research Centre, Bengaluru, Karnataka, India, ³Senior Lecturer, Department of Oral Medicine & Radiology, Royal Dental College, Chalissery, Palakkad, Kerala, India

Corresponding Author: Shweta Channavir Saddu, Department of Oral Medicine and Radiology, The Oxford Dental College, Bommanahalli, Hosur Road, Bengaluru - 560 068, Karnataka, India. Phone: +91-9747672871. E-mail: shwetasaddu@gmail.com

Abstract
Schwannomas are benign, encapsulated, slow-growing and usually solitary tumor originating from Schwann cells of the peripheral nerve sheath. Approximately, 25-45% of cases are seen in the head and neck region being uncommon in the oral cavity. We report a rare case of schwannoma in a 24-year-old male who had a 4-year history of swelling on the right side floor of the mouth, with details of computerized tomography imaging and clinicopathologic characteristics of the tumor. Complete excision of the tumor with primary closure was carried out.

Keywords: Computerized tomography scan, Floor of mouth, Immunohistochemistry, Schwannoma

INTRODUCTION
The term schwannoma has numerous synonyms such as neurilemmoma, neurinoma, neurolemoma, peripheral glioma, perineural fibroblastoma and peripheral nerve sheath tumor, but among these neurinoma, neurilemmoma and schwannoma are presently used.¹⁻²

Schwannoma is solitary, slow growing, benign encapsulated neural tumor arising from the nerve sheath Schwann cells of the peripheral, cranial or autonomic nerves, and it has a predilection for sensory nerves.²⁻³

Approximately 25-45% of schwannomas occur in the head and neck area the intracranial region being the most common site. Only 1% of the schwannomas occur intra-orally the tongue being the most common site, but schwannoma in the oral floor is extremely rare. Many authors reported that schwannoma occurs regardless of age and sex, grows gradually and painlessly. Schwannoma does not recur, and the malignant transformation is rare.⁴ Here, we present a rare case of schwannoma of the floor of the mouth.

CASE REPORT
A 24-year-old male was referred to our outpatient department with complaints of painless, progressive swelling on the floor of the mouth for 4 years. He underwent incisional biopsy and Computerized tomography (CT) scan 1 week before visiting our institution. There was no history of trauma, local infection or systemic illness. The patient did not report any discomfort while talking and swallowing. On examination, there was well-defined large swelling in the right floor of the mouth measuring 4 cm × 3 cm in size, with a smooth surface. Overlying adjacent mucosa revealed no abnormality. The swelling was free from alveolar mucosa and involved the ventral surface of the tongue not crossing the midline; posteriorly it extended up to right lower second molar (Figure 1). An enlarged salivary duct opening was seen on posterio-superior aspect of swelling that showed saliva pooling on palpation (Figure 2). The swelling was firm in consistency, non-tender, and mobile. Tongue mobility was normal.

CT scan revealed ill-defined mildly non-homogeneously enhancing soft tissue density space occupying lesion
measuring about 3.8 cm × 3.5 cm × 3.6 cm seen in the right side floor of the mouth, in the sublingual region. Minimal smooth scalloping of adjacent body of the mandible was also noted. Based on history, clinical appearance and CT findings benign tumor of sublingual salivary gland tumor was made (Figure 3).

Excisional biopsy was performed under general anesthesia (Figure 4). Macroscopic appearance of specimen revealed a solitary, pinkish, well-encapsulated mass measuring 3.5 cm × 3 cm which was sent for histopathological examination (Figure 5).

On microscopic examination, the specimen was noted to be composed of alternating antoni A and B areas. The antoni A areas were composed of spindle cells with indistinct cytoplasmic borders and nuclear palisading associated with Verocay bodies. Antoni B areas were hypocellular, with a spindle to oval shaped cells arranged haphazardly in loosely textured matrix. The tumor showed degenerative changes characterized by extensive hyalinization, hemorrhage and nuclear atypia though there was an absence of mitoses. The margins were clear of tumor though an area of tumor extension into the normal salivary gland component was seen (Figure 6). Immunohistochemistry showed cells positive for protein S-100, a marker for neural cell origin (Figure 7). Finally, the tumor was diagnosed as a schwannoma. Postsurgical recovery of the patient was uneventful.

DISCUSSION

Schwannoma was first described in 1910 by Verocay, and he named it neurinoma. According to Parikh & Desai, the term neurilemma was suggested by Stout in 1935. Schwannoma is a rare, benign, neurogenic neoplasm composed of Schwann cells. Embryologically, Schwann cells arise during the 4th week of development from neuroectoderm.

Most reports suggest that the majority of tumors are present between the ages of 20 and 40 years and are equally distributed between the two sexes. In the head and neck region, the tongue is the most common site, followed by the palate, floor of mouth, buccal mucosa, lips and jaws. Other common sites include the flexor surface of upper and lower extremities and less often the mediastinum and peritoneum. Occasionally the tumor can arise centrally within bone and may produce the bone expansion.
Schwannomas are benign, slow-growing; usually solitary encapsulated tumors. The etiology is unknown. They can arise from nerves 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. More commonly it develops from the sensory nerves and rarely from the motor nerves. If the nerve of origin is small, its association with a given tumor may be difficult to demonstrate. Whereas, if it originates from a larger nerve, it appears to be splayed out over the outer aspect of the capsule rather than incorporated within the tumor. Rarely, the tumor can cause displacement and compression of the surrounding normal nerve tissue associated with pain and paresthesia.

Schwannomas have two clinical forms, the most frequent being the encapsulated one in which the tumor is surrounded by dense fibrous connective tissue; the other is pediculate, resembling a fibroma.

The lesion normally appear as well-circumscribed circular-type mass with a smooth margin, as observed in the presenting case. Numerous diseases come in the differential diagnosis of swelling of floor of mouth such as fibroma, lipoma, mucocele, epithelial hyperplasia, benign salivary gland tumors, hemangiomma, granular cell tumor, neurofibroma, neuroma, nerve sheath myxoma, leiomyoma, rhabdomyoma. Although schwannoma in the oral floor is rarely observed, it should be taken into consideration while making a differential diagnosis.

Diagnostic investigations include an ultrasound scan, CT, magnetic resonance imaging (MRI) and fine needle aspiration cytology. MRI is the best choice in detecting the extent of the tumor and correlates well with operative findings. MRI was not advised in the present case as patient already underwent CT scan.

Identification of the originating nerve may be difficult as in the present case. In more than 50% of intraoral lesions, it is not possible to differentiate between tumors arising from the lingual, hypoglossal and glossopharyngeal nerves. Also, there are reported cases of schwannoma...
arising from sublingual gland,\textsuperscript{11} mylohyoid nerve,\textsuperscript{12} and hypoglossal nerve.\textsuperscript{13}

Ideally two histological patterns are defined, antoni A and antoni B. Antoni type A consists of Schwann cells arranged in compact, twisted bundles, associated with delicate reticulin fibers and spindle-shaped nuclei aligned in parallel rows forming a typical palisading pattern. Between the rows there are fine cytoplasmatic fibrils with acellular, eosinophilic masses called Verocay bodies. Antoni Type B tissue is formed by irregularly arranged masses of elongated cells and fibers similar in appearance to neurofibroma, with areas of cystic degeneration and edema. Immunostaining analysis is critical in the diagnosis of these neoplasms. Immunohistochemical tests can reveal a high affinity of the Schwann cells to S-100. The histopathologic examination and immunostaining provided a definitive diagnosis in the present case.\textsuperscript{6-8}

Surgical excision is the treatment of choice and relapse is uncommon in the well-encapsulated variety. The encapsulated form is enucleated easily, whereas the non-encapsulated requires normal tissue margins to avoid relapse. If the nerve of origin is visualized, an attempt should be made to separate carefully to preserve function, although this is sometimes not possible. The prognosis of schwannoma is quite favorable. Malignant transformation of benign schwannoma has been controversial, with a few isolated cases documented. Malignant transformation was not likely in our patient because examination of the excised mass revealed benign microscopic features and complete removal was confirmed.\textsuperscript{1,6,8}

CONCLUSION

The schwannoma represents a lump not often encountered in clinical practice. The sub mucosal form of this lesion is, usually, indistinguishable from other benign neoplasm that also, usually, seen in the same region. Therefore, schwannoma should be included in the differential diagnosis of well-circumscribed mucosal masses. The final diagnosis should be done after appropriate investigations, histopathological examination and in some cases after immunohistochemical analysis.

REFERENCES


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