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Case Report

NEURILEMMOMA OF TONGUE: CASE REPORT AND LITERATURE REVIEW

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ABSTRACT

Neurilemmoma is a benign, slow growing, solitary, encapsulated perineural tumor. Approximately 25-45% of all neurilemmomas occur in the head & neck region. Of these 1-12% occur intraorally, commonest site being tongue. The other sites include palate, floor of the mouth, buccal mucosa, lips and jaws. Its etiology is unknown. Schwann cell is the common precursor of most nerve shealth tumors.. Usually this lesion will not be taken into account clinically and the differential diagnosis includes numerous benign neoplasm originating from connective tissues. Microscopic picture helps in clear diagnosis. Immunohistochemical features are useful in determining the neural differentiation and thus confirming the diagnosis of neurilemmoma. We hereby report a case of neurilemmoma in the tip of the tongue, in a 60yrs old female patient.

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INTRODUCTION

Neurilemmoma also called as Schwannoma, neurinoma, and perineural fibroblastoma is an encapsulated nerve sheath tumor[1]. A benign neoplasm originating from Schwann cells of the nerve shealth was first described by Verocay in 1910. He named it as "Neurinoma" then, in 1935, the term "Neurilemmoma" was coined by Stout[2].

Schwannoma is usually a solitary, nodular soft tissue or intrabony lesion that is slow growing, encapsulated lesion and is often associated with the peripheral nerve attachment [3]. The etiology is unknown, but it is postulated that the lesion arises by proliferation of Schwann cells at one point inside the perineurium. The lesion will cause the displacement and compression of the surrounding normal nerve tissue[4]. Approximately 25–45% of all schwannomas occur in the head and neck. Of these, approximately 1–12% occur intraorally and the most common site is tongue followed by palate, floor of the mouth, buccal mucosa, lips and jaws[5].

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[6]. 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 [7].

Oral schwannomas are found to exhibit two types. The common type is the submucosal nodule, which is encapsulated, well defined, firm in consistency, thus resembling a cyst. The second type is the nonencapsulated, where the tumor is found below the basal layer of the mucous membrane[8].

The clinical differential diagnosis includes any other benign connective tissue lesions such as fibroma, lipoma, neurofibroma, rhabdomyoma, leiomyoma or salivary gland tumors. However, the histological differential diagnosis is made with other neural origin lesions, which could be neurofibroma and neuroma.

Microscopically, the characteristic histological features for schwannomas are complete tumor encapsulation and lesional tissue comprising of alternating regions of Antoni A and Antoni B, respectively. An immunohistochemical examination of the tumor may show positive results with S100 antigen[9].

Radiographical examination such as Computed Tomography (CT) and Magnetic Resonance Imaging (MRI) helps in identifying the extension of the lesion. Conservative surgical excision is the treatment of choice and the prognosis is good[10]. The aim of the article is to report a clinical case of neurilemmoma of tongue whose diagnosis was based upon clinical, histological and immunohistochemical features.

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Case report

A 60yrs old female patient presented to the Department of Oral and Maxillofacial Pathology with a chief complaint of swelling on the tip of the tongue for past 6 months. The swelling was initially smaller in size and has gradually attained the present size with no history of pain.

No significant medical, family or personal history was elucidated. On intraoral examination, a submucosal nodular swelling measuring about 1.5cm X 1cm was present on the tip of the tongue. Colour of the overlying mucosa was normal. On palpation, the swelling was firm in consistency and non- tender with well defined borders. (Figure 1)

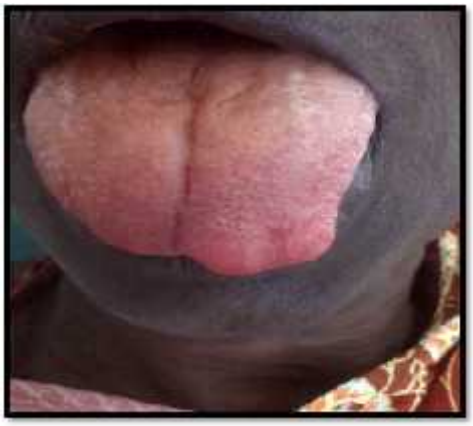

Figure 1 A submucosal nodular swelling measuring about 1.5cm X 1cm was present on the tip of the tongue.

From the clinical findings, the provisional diagnosis was established as benign soft tissue mesenchmal neoplasm.

The routine blood investigations were done and excisional biopsy was performed under LA.

Histopathological findings revealed an encapsulated lesional tissue. The lesional tissue was composed of spindle shaped cells arranged in short bundles of interlacing fascicles that characterize Antoni A areas. Few spindle cells exhibited mild pleomorphism & nuclear hyperchromatism. Whorling of spindle cells were also seen in few areas. In few foci the spindle cells were loosely arranged in a fibrinous background that characterize Antoni B areas. Mixed inflammatory cell infiltrate comprising of lymphocytes, plasma cells and mast cells admixed with blood capillaries were seen. (Figure 2)

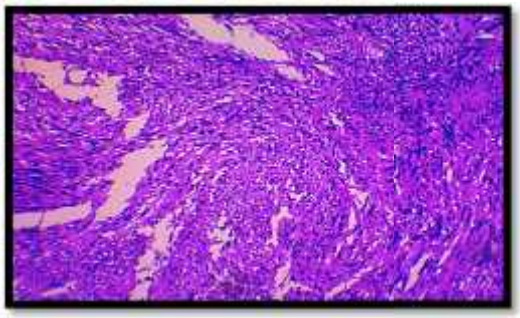

Figure 2 Spindle shaped cells arranged in short bundles of interlacing fascicles that characterize Antoni A areas. In few foci the spindle cells were loosely arranged in a fibrinous background that characterizes Antoni B areas.

Considering the non organized pattern of schwann cells, immunohistochemical analysis was done using S-100 antibody. S100 showed intense positivity in the tumoral cells.(Figure 3)

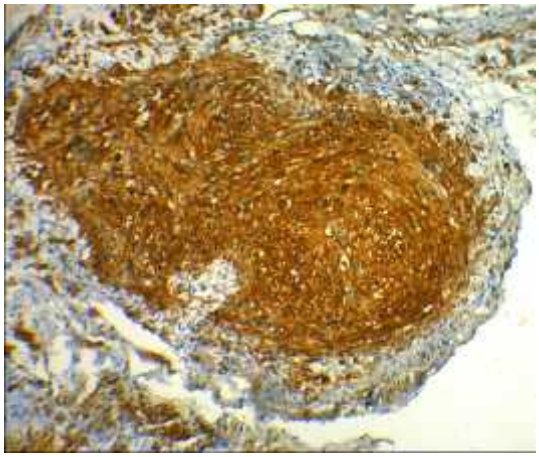

Figure 3 Immunohistochemical analysis using S-100 antibody showing intense positivity in the tumoral cells.

Based on the correlation with clinical, histopathological and immunohistochemical findings, the final diagnosis of Neurilemmoma was given.

DISCUSSION

Schwannoma is a benign ectodermal peripheral nerve sheath tumor arising from Schwann cells. 25 to 45% of Schwannomas occur in the head and neck and only 1% demonstrate an intraoral origin. Upon which greatest frequency, in the mobile portion of the tongue followed by floor of the mouth, palate, gingiva, vestibule, lips, salivary glands and mental nerve region[11].The origin of the Schwann cell has always been a debate till date. Few suggest that, they arise directly from the neural tube whereas few were convinced that they originate from the neural crest, cells that lie lateral to the neural tube and beneath the ectoderm of the developing embryo[12]. But now it is believed to originate from the proliferation of Schwann cells in the perineurium causing displacement and compression of the adjacent nerve[13]. It could also arise from nerves covering schwann cell sheath which includes cranial nerves except from cranial nerves I and II because they lack Schwann cells, spinal nerves, autonomic nervous system[14].

Neurilemmoma is usually a solitary, nodular, aymptomatic lesion. If multiple lesions are seen, it is called as “Schwannomatosis” which may either involve cranial, spinal or peripheral nerves and is associated either with or without neurofibromatosis or Von Recklinghausen’s disease[15].

In our case, the neurilemmoma presented as a solitary, well circumscribed swelling without any specific characteristics to differentiate it from other soft tissue lesions. Clinical evaluation of provisional diagnosis is been quite difficult for identification of a particular lesion. The provisional diagnosis was established as benign soft tissue mesenchymal neoplasm.

Histopathological examination helps in providing a definitive diagnosis for the case. There are many histopathological variants of schwannoma. The two refined histological patterns present are, Antoni-A with hyper cellularity and Antoni-B with hypo cellularity. The ancient schwannoma comprises of various degenerative changes like cyst formation, haemorrhage and hyalinization along with nuclear atypia. Pseudo glandular schwannoma shows cystic spaces lined by schwann cells which assumes a round or epitheliod structures resembling glands or

dilated lymphatics. Plexiform schwannoma exhibits conventional or mixed pattern of appearance with increased cellularity and mitosis. Epitheliod schwannoma also shows schwann cells which are round with sharp cytoplasmic borders and nuclear inclusions resembling epitheliod cells[16].

Microscopically, the histological differential diagnosis of neural origin lesions are schwannoma and the neurofibroma. Both contain elongated cells with irregular nuclei interspersed between the bundles of collagen fibers. They differ histologically and histogenetically. The schwannoma is derived from the Schwann cells and the neurofibroma arises from the fibroblasts of the perineurium. Neurofibroma is not encapsulated, consisting of a mixture of Schwann cells, perineurial cells, and endoneurial fibroblasts while schwannoma is encapsulated[17].

The histological features of the present case are almost similar to those cases which are reported earlier, comprising of spindle cells arranged in short bundles of interlacing fascicles indicating Antoni A areas and loosely arranged spindle cells in a fibrinous background indicating Antoni B areas. Antoni A pattern was predominantly present in the entire specimen

Immunohistochemistry helps in confirmatory diagnosis of the lesion. For all neural tumors S100 protein shows positivity but immunohistochemical examination could assist in lesion differentiation. Chrysomaly *et al*., observed an intense positive reaction to S100 in schwannoma and palisaded encapsulated neuroma. Intensive reaction to CD57 was observed in traumatic neuroma, while capsular epithelial membrane antigen (EMA), and CD34 positivity were also observed in few cells of schwannoma[18]. In our study, S100 shows intense positivity in the tumoral cells.

Passador-Santos *et al*.,in their study observed that S100 protein, EMA, laminin, fibronectin, and collagens I and II are all expressed in benign neural neoplasms. They found that schwannoma cells were intensively positive for S100 regardless of the growth pattern. EMA was seen only in the capsule staining perineurial cells. Laminin was expressed in the basement membrane around the tumor cells. Fibronectin and collagens I and III were stained in the capsule. In neurofibroma, the cells expressed S100 whereas EMA was expressed only in a few scattered cells. Laminin and fibronectin showed a diffuse positivity[19]. Because it is a well encapsulated tumor, treatment of choice could be conservative surgical enucleation. Periodic follow up is necessary. Recurrence is usually uncommon.

CONCLUSION

Schwannoma is not often encountered in the clinical practice, but still it should not be omitted on observing a tumor involving the oral cavity. Schwannoma can also reach out to complications such as involving a nerve or rarely leading to a malignant transformation when left untreated. Thus definitive diagnosis can be made with the aid of histopathological and immunohistochemical findings to initiate prompt treatment.

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