

Palisaded Encapsulated Neuroma of the Tongue Clinically Mimicking a Pyogenic Granuloma: A Case Report and Review of Literature

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Abstract

Palisaded encapsulated (solitary circumscribed) neuromas (PENs) are relatively common intraoral neurogenic tumors, which occur most frequently on the hard palate. Herein, we describe the clinicopathological characteristics of a palisaded encapsulated neuroma of the tongue. This tumor was an exophytic sessile mass measuring 0.3×0.4 cm with rubbery consistency on the anterior one-third of the dorsum of the tongue. The tumor was excised under the impression of a pyogenic granuloma (PG). No recurrence was reported at 12 months postoperatively. Histopathological examination showed a well-circumscribed mass that composed of interlacing fascicles of spindle cells. The cells were S-100 positive. The nuclei, showing parallel orientation within the fascicles, were wavy and pointed and showed no sign of mitotic activity. Giemsa staining revealed no mast cells within the stroma.

Key Words: Neuroma; Nerve Sheath Neoplasms; Pyogenic Granuloma

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INTRODUCTION

In 1972, Reed and colleagues described a distinctive neural tumor as PEN. Palisaded encapsulated neuromas clinically manifest as solitary, firm, non-pigmented, dome-shaped nodules on the face of adult patients [1]. Palisaded encapsulated neuroma is known as a benign tumor of the facial skin, and is rarely found in the oral mucosa [2]. Microscopically, the tumors are characterized by moderately

cellular, fascicular proliferation of spindle cells that show some areas of parallel nuclei [3]. A bundle of nerves interposed between the Schwann cells typically aggregates in palisades and is identified by S-100 protein immunohistochemical stain [4]. An alternate designation i.e. solitary circumscribed neuroma (SCN) was proposed by Fletcher in 1989 [5]. Irrespective of the nomenclature, PEN/SCNs are considered as reactive hyperplastic

processes. In 2010, Koutlas and Scheithauer considered PEN/SCNs as relatively common true neuromas of the skin or mucosa [6].

As a peripheral nerve sheath tumor, PEN accounts for only 0.04 to 0.05% of oral biopsy specimens. Other peripheral nerve sheath tumors are neurofibroma, schwannoma (neurilemmoma), mucosal neuroma associated with multiple endocrine neoplasia III, nerve sheath myxoma and granular cell tumor [7].

In the mouth, PEN is mostly found on the hard palate and maxillary labial mucosa as a small, superficial and usually painless nodule. The lesion is frequently diagnosed between the 5th and 7th decades of life, with equal sex predilection. The cause is uncertain; although trauma is presumed to play an etiological role [8]. A preferred treatment for PEN is conservative local surgical excision [8]; although gross total resection has been recently claimed to be the treatment of choice [6].

CASE REPORT

A 48-year-old man was referred to the Oral Medicine Department of Babol Dental School, complaining of a tongue mass persisting for one year. The lesion was painless, but sensitive to hot food and drink. His medical and family history was unremarkable. Physical examination revealed an exophytic sessile mass measuring 0.3× 0.4 cm with rubbery consistency on the anterior one-third of the dorsal surface of the tongue (Fig. 1). Clinically, the overlying mucosa was depapillated and had increased vascularity. Under the impression of a PG, excision of the mass was done and no recurrence was reported at 12 months postoperatively. Histopathological sections showed an encapsulated mass within the connective tissue, composed of interlacing fascicles of spindle cells that were consistent with Schwann cells. The cells showed a positive immunohistochemical reaction to S-100 protein (Fig. 2).

The nuclei, showing a parallel orientation within the fascicles, were characteristically

wavy and pointed, with no significant pleomorphism or mitotic activity. There was scant fibrous stroma among these nests. The overlying epithelium was atrophic and no rete ridges were seen (Figs. 3 and 4). Giemsa special stain revealed no mast cells within the stroma, ruling out the differential diagnosis of neurofibroma.

DISCUSSION

Mucosal neuroma can be distinguished histologically from other neurogenic tumors such as neurofibroma, neurilemmoma and PEN. Briefly, mucosal neuroma is not encapsulated and does not have palisading nuclei; whereas neurofibroma and PEN are encapsulated [9,10].

Mucosal neuromas are usually associated with the multiple endocrine neoplasia syndrome III (MEN III), a rare syndrome with potentially fatal consequences such as medullary carcinoma of the thyroid. Other clinical signs of the patients with MEN III can be considered in the diagnosis of oral lesions such as a mucosal neuroma.

However, when oral lesions are present in absence of other diagnostic signs, histopathological evaluation can be helpful. The microscopic examination of the mucosal neuroma shows nerve bundles in various sizes surrounded by normal connective tissue, which are not usually seen in PEN [11].

A traumatic neuroma is not a true tumor, yet it develops as a proliferation of neural tissue that is caused by injury to a peripheral nerve. Traumatic neuromas are usually associated with pain, ranging from pain on palpation to a constant severe pain [12]. Substantial histomorphological differences exist between PEN/SCN and traumatic neuroma. These include the presence of perineural cells surrounding individual microfascicles, the greater abundance of interstitial collagen, mucoid matrix and myelin components, and the more orderly parallel arrangement of axons in traumatic neuroma [6].



Fig. 1. Clinical view of a PEN on the dorsum of the tongue.

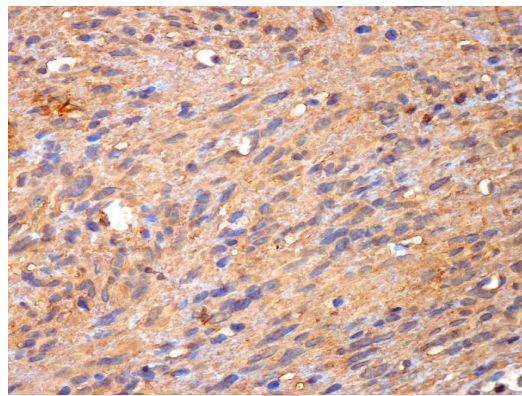


Fig. 2. Positive immunoreactivity of tumoral cells with S100 antibody ($\times 400$).



Fig. 3. Photomicrograph showing an encapsulated proliferation of neoplastic cells (hematoxylin and eosin staining; original magnification $\times 40$).

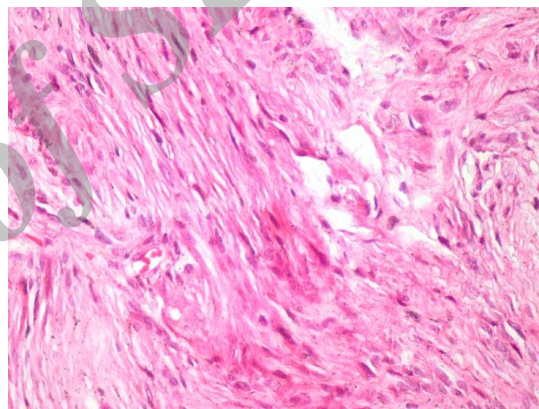


Fig. 4. The tumor composed of spindle cells in distinct fascicular pattern. No evidence of malignancy is observed (hematoxylin and eosin staining; original magnification $\times 400$).

Several microscopic criteria have been introduced to differentiate PEN from schwannoma. Antoni A, organized spindle cells in palisaded whorls, and Antoni B, haphazardly distributed neoplastic cells, are two common patterns which are often found during histopathological examination of schwannomas. Other microscopic criteria include Verocay bodies and the more definite palisading in the nuclei than that in PEN [13].

Contrary to the latter, it is extremely difficult to microscopically differentiate neurofibroma from PEN, especially when an incisional biopsy has been performed. The absence of a marked fibrous capsule and the irregular arrangement of the neoplastic cells are the main differential clues seen in neurofibroma as compared with PEN. The significant presence of mast cells, usually observed among tumoral cells of the neurofibroma, is also detectable

using histochemical or immunohistochemical staining methods [8]. As Regezi and colleagues stated, PEN/SCN may be misdiagnosed clinically once identified somewhere in the mouth other than the palate [11]. This may be an obvious clinical impression since intraoral PEN/SCNs are mostly found on the hard palate [3,8]. Conversely, the tongue involvement comprises less than 8% of PEN/SCN cases [6]. As for this case, the PEN resembled a PG on the dorsal surface of the tongue (a common site for neurilemmoma and neurofibroma, but not for PEN) [14]. An erythematous lesion in a less commonly affected site rarely happens to be a PEN. Besides, the tongue is a potential site for PG. Regezi et al. have reported that PG is most commonly seen on the attached gingivae, tongue, lower lip and buccal mucosa [11]. The age of patient may be an important clinical parameter when the list of differential diagnoses of a lesion is formulated [15]. Our patient was 48 years old, which was close to the recently reported average age for PG patients (52 years). Pyogenic granuloma usually occurs in patients older than 39 years, with equal gender distribution [16].

Soft tissue enlargements of the oral cavity often present a diagnostic challenge because a diverse group of pathological processes can produce such lesions. Pyogenic granuloma is among the most common entities responsible for causing soft tissue enlargements [17]. Pyogenic granuloma can manifest as a painless smooth or lobulated mass with a surface that bleeds quite easily. Because of their high level of vascularity, young PGs are red, whereas older lesions are more collagenized and appear pink or normal colored [18]. Clinically, oral PG occurs as an exophytic lesion manifesting as small, erythematous papule on a pedunculated or sometimes sessile base [19]. Pyogenic granuloma arises in response to various stimuli such as chronic low-grade irritation, traumatic injury and hormonal factors [20]. However, the effect of female hormones on oral PG was questioned by Bhaskar and Jacoway since they

found lesions both in males and females with no sex predilection [21].

CONCLUSION

Palisaded encapsulated neuromas may be misdiagnosed clinically once they appear somewhere in the oral cavity other than the palate. A PEN arising on the dorsum of the tongue may mimic a PG with similar clinical morphology. Even the patient's age may be misleading. On the other hand, the dorsal surface of the tongue is a common site for neurilemmomas and neurofibromas. Peripheral nerve sheath tumors must therefore be included in the list of differential diagnoses for a PG-like lesion on the tongue.

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