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Palisaded encapsulated neuroma of the oral cavity: The case report of a rare entity

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Abstract
Peripheral Nerve Sheath Tumors are neoplasms or reactive lesions involving of the peripheral nerves. Such lesions are an uncommon finding in the oral cavity. The lesion can present in different forms such as neurofibroma, neurilemmoma, traumatic neuroma and palisaded encapsulated neuroma. These lesions are benign and show less chances of recurrence. Here we report a case of Palisaded Encapsulated Neuroma on the lower jaw.

Keywords: Peripheral Nerve Sheath Tumor, Palisaded Encapsulated Neuroma, Reactive Lesion

Introduction
Peripheral Nerve Sheath Tumors (PNST) are uncommon lesions of the oral cavity [1] It comprises of neoplastic or reactive lesions that arise due to the proliferation of axons or Schwann cells or their lining structures. PNSTs are classified based on it's cellular composition and organization. Neurofibromas, Schwannoma, traumatic neuroma, Palisaded Encapsulated Neuroma, Granular cell tumor, Nerve Sheath Myxomas, perineurioma are some of the PNSTs reported apart from malignant nerve sheath tumors [2].
Palisaded Encapsulated Neuroma (PEN) is a benign PNST which appears as a solitary, small peripheral nerve lesion that occurs on the facial skin [3]. Here we report a case of oral PEN which presented as a sessile soft tissue swelling at the mental foramen region.

Case Report
A 34 year old female patient reported with the chief complaint of swelling in the lower left back teeth region for past 5 months. A history of trauma was ruled out. The swelling was initially small and gradually increased in size to the present form and it was asymptomatic. Clinical examination revealed a well-defined, dome shaped, soft and non-tender swelling which measured 2x3cm in it's greatest dimension on the buccal alveolar mucosa in relation to 33, 34. (Fig.1). No evidence of paraesthesia in the region of interest. Radiographic evaluation was non-contributory. Based on history and clinical examination a provisional diagnosis of Benign soft tissue neoplasm was arrived at. Considering the size of the lesion, an excisional biopsy was performed under local anaesthesia.

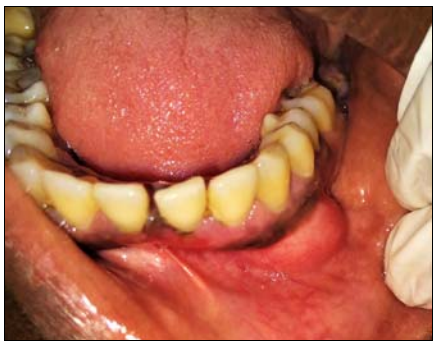

Fig 1: Swelling in 33, 34 region

Histopathological examination revealed partially encapsulated dense fibrous connective tissue stroma interspersed by cells exhibiting wavy elongated nuclei with presence of numerous nerve bundles and mast cells. A histopathological diagnosis of palisaded encapsulated neuroma was given. (Fig 2). There was good healing of the region without any scar.

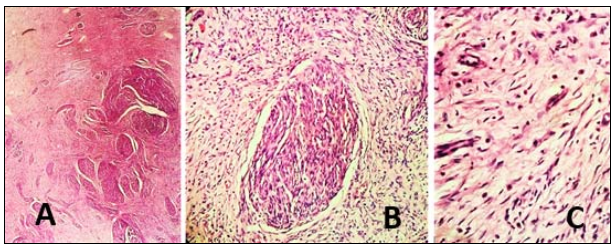

Fig 2: Photomicrograph of the lesional tissue; A-4X Shows multiple nerve bundles; B-10X Single nerve bundle showing palisaded arrangement of nuclei; C-40X Shows wavy elongated nuclei

Discussion

PEN constitute to 4.5% of all oral neoplasms. The patients usually are middle aged with no sex predilection [4] The first case of PEN was described by Dr. Richard J. Reed in 1972 occuring ossn the surface of the skin.[5] Later, in 1989, Fletcher used the term 'Solitary circumscribed neuroma' to describe the lesion. His report included a case of PEN occurring on antero-lateral border of the tongue. It is considered as one of the first reported cases of intra-oral PEN. Although, it is predominantly a cutaneous lesion, it's occurrence in the oral cavity had also been reported [6].

The etiology of PEN had been a matter of debate. It was proposed that the tumor is an attenuated manifestation of multiple endocrine neoplasia type 2B (MEN2B). MEN2B is characterized by development of neoplasms in the eyes, mouth and endocrine glands. However, marked difference existed in the histopathological features of PEN to that of MEN2B. History of trauma had been proposed as an etiological factor. However, most cases of PEN does not reveal a history of trauma [7].

Clinically intraoral-PEN usually present as solitary, sessile, dome shaped growth which has a smooth surface. The mucosa over the growth would be similar in color to the adjacent region. It is a slow growing tumor that varies in size between 2 to 6 mm in size [1]. It is usually detected during the 5th to 7th decade of life and has equal predilection to occur in both genders. The most common intra-oral site for PEN is hard palate [8].

Histopathologically, PEN demonstrates a solitary well-differentiated partially encapsulated nodule, which is made up of spindle shaped cells that is consistent with Schwann cells. It is grouped in interlacing fascicles. Tumor cells have an eosinophilic cytoplasm with wavy nuclei. There are different histopathological presentation of the tumor which include nodular pattern, epithelioid, plexiform and multinodular types. Nodular pattern is the most common type of presentation. Immunohistochemistry shows strong positivity for S-100 protein and collagen IV and negative for GFAP [9].

Based on the clinical presentation and histopathologic features, the closest differential diagnosis that was considered for the present case was traumatic neuroma. Traumatic neuroma is a hyperplastic response of the nerve tissue to trauma in the region. The lesion is characterized by firm nodular growth with severe tenderness in the region [9].

Schwannoma which is also called Neurilemmoma is another benign peri-neural tumor that can a similar presentation in the oral cavity. It presents as a slow growing, asymptomatic mass and it is commonly found among females. The commonest site of occurrence is on tongue. It is characterized microscopically by the presence of Verocay bodies and nuclear palisading. These features were absent in the present case [10].

Kuyama *et al*. (2012) reported a case of intraoral PEN presenting on the upper lip mimicking a fibroma [11]. Manchanda *et al*. (2015) reported PEN presenting as growth originating from the gingiva [1]. Mortazavi *et al*. (2015) reported a case of PEN occurring on the tongue that clinically mimicked pyogenic granuloma [8]. In the case by Moghadam *et al*. (2017) reported case of PEN presenting as pedunculated growth on the buccal mucosa [12]. Cunha *et al*. (2018) reported case of intraoral PEN mimicking mucocele of lower labial mucosa [13]. In all the cases surgical excision was performed and recurrence was not detected during the follow up period. This is the first case to the best of our knowledge with the presentation of the lesion on the alveolar mucosa.

Complete surgical excision is the treatment of choice for PEN and there is very less chance of recurrence of the lesion. In the present case, based on the provisional diagnosis of benign soft tissue neoplasm, surgical excision of the lesion was done under local anesthesia. The healing was uneventful.

Our Patient had been under review since 1 year and does not reveal any signs of recurrence.

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