Mount Sinai Subgengmal Neurogenous Plaque: Report of Two Cases and a Review of the Literature
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Introduction

Originally described as a “hyperplastic subepithelial (subgengmal) nerve plexus” by McDaniel (1999), the subgengmal neurogenous plaque (SNP) is a subepithelial proliferation of neural fibers likely associated with the overlying taste buds. Prior to the description of the SNP, the normal neural structures of the taste buds were well-described. It consists of a subgengmal nerve plexus with intergengmal (between taste buds) and intragengmal (within taste buds) branches of nerves. The proposed etiology of the SNP is that the nervous tissue structures become hyperplastic following chronic irritation and subsequent inflammation; this is disputed, with other authors suggesting that it may merely be a normal structure of the taste buds.

The diagnosis of subgengmal neurogenous plaque was recently rendered in two cases at our institution. We present these cases as well as a review of the literature on the subject.

Methods

We reviewed recent cases of diagnosed SNP at our institution (The Mount Sinai Hospital, New York, New York). For these cases, immunohistochemistry was performed. S100 (Ventana, 10 µg/mL) and neuron-specific enolase (NSE, Cell Marque Sigma, 1 µg/mL) were utilized. We also searched PubMed for all relevant articles using the phrases “subgengmal neurogenous plaque” and “subgengmal nerve plexus)” that discuss the SNP in the oral cavity of humans.

Results

Case 1: A 64-year-old female presented to an oral surgeon for a biopsy of a painful ulcer on the lateral tongue that had not resolved over the course of the previous three months. Histopathological examination showed multiple tongue papillae, each exhibiting multiple taste buds. Some papilla had a proliferation of neural-appearing tissue that stained positively with S100. The taste buds and ganglion cells (located deep to the taste buds) showed strongly positive staining for NSE. Other areas of the lesion showed a pseudoneural inflammatory component composed of eosinophils, plasma cells, lymphocytes, and occasional neutrophils; this inflammatory component was diagnosed as a traumatic ulcerative granuloma with stromal eosinophilia, and primary cutaneous CD30+ lymphoproliferative disorder was ruled out with the demonstration of CD30 negativity. Selected images of this case are presented in Figure 2.

Discussion

The taste bud is a structure that transmits sensory information about chemicals in the oral environment to the brain; in order to do so, it must have neural structures present. It is reasonable to expect that biopsies in areas where taste buds are predominant will have at least a minor degree of neural tissue present. One article found what they called “pseudoganglioneuromas” in the plaque, which they described as traumatic neuromas occurring in areas with pre-existing pharyngeal neural structures. The identification of the ganglion cells near a neurofibroma-like area may give the impression of these being neurofibromas, though the descriptions for each are functionally similar to ganglion cells, such as ballooning degeneration and multinucleation. The identification of the ganglion cells near a neurofibroma-like area may give the impression of these being neurofibromas, though the descriptions for each are functionally similar to ganglion cells, such as ballooning degeneration and multinucleation.

The reported case involves a 64-year-old female with a painful ulcer on the lateral tongue that had not resolved over the course of the previous three months. The biopsy revealed a pseudoneural inflammatory component composed of eosinophils, plasma cells, lymphocytes, and occasional neutrophils. The authors hypothesized that these cells are not actively proliferating and that the lesion is a reactive proliferation of postganglionic gustatory nerves with axonal branching and endoneurial fibroblasts, which eventually disappear, resulting in a loose mixture of Schwann cells, axons, and basement membrane material. Alternatively, other authors suggest that this entity may merely be a normal neural structure of the taste buds.

Most publications on the subject agree that the SNP should have the following histopathological features:

- Circumscription and non-encapsulation
- Composed of eosinophilic cellular and fibrous structures, containing Schwann cells (which stain with S-100 and laminin) and axons (which stain with PGP9.5 and NFi)
- Somewhat rectangular in shape, roughly 0.5-3.5 mm in diameter
- Often, a focal lymphoplasmacytic inflammatory infiltrate
- Can be located in foliate papillae (usually one plaque per papilla, but can have two separated by a groove in the papilla, forming a “mirror image” of itself, perhaps because it is forming an annular structure)

Triantafylou and Couter, in their review of 16 cases on the subject, developed a useful description of the “progressive and subtle” zonal pattern in these lesions:

- Superficial: primary component of the plaque, nerve fascicles are inconspicuous and set in a loose background of fibroblastic collagen.
- Intermediate: composed of nerve fascicles, but with attenuated perineurial sheaths and increased endoneurial components that are both growing less organized as they progress to the superficial regions.
- Deep: often endoneural, or composed of small nerve fascicles, often with myelinated axons and occasionally ganglion cells.

Guers et al. considered this to be a biphasic lesion, categorizing the histological patterns as “superficial” and “deep,” though the descriptions for each are functionally similar to Triantafylou and Couter’s descriptions, with the latter authors preferring to have a transitional area in their description.

It is important that these lesions are not confused with other hyperplastic neural entities such as a neuroma, neurofibroma, or ganglioneuroma. This is particularly true in the case of neurofibroma, wherein multiple misdiagnosed SNPs could potentially lead to an erroneous diagnosis of neurofibromatosis. The superior portion of the subgengmal plaque is often less well-defined, and the macroscopic features of collagenous fibers and neural cells may appear very similar to a neurofibroma. However, the subgengmal neurogenous plaque tends to be more well-defined, has the aforementioned zonal distribution of features, and lacks the feature of continuous growth that the neurofibroma more often demonstrates. The identification of the ganglion cells near a neurofibroma-like area may give the impression of these being neurofibromas, though these are very uncommon in the head and neck. Usually, in those aged 10-30, are usually encapsulated, and often display alterations in the ganglion cells, such as ballooning degeneration and multinucleation.

Because this is likely either a normal structure or a hyperplasia of a normal structure, conservative excision with histologic examination to rule out other entities in the list of differential diagnostic possibilities should be adequate therapy. According to the study that identified two cases possibly associated with burning mouth syndrome, while initial efforts to remove irritants and rule out systemic causes of burning tongue were ineffective, removal of the subgengmal plaque resulted in a complete resolution of symptoms in both cases.

References


Table 1. Patient characteristics from these cases and the literature review.