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ORIGINAL ARTICLE

Clinical aspects of tongue subgemmal neurogenous plaque: Surgical treatment of two symptomatic cases

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Abstract:

Subgemmal neurogenous plaque (SNP) is a regional anatomic variation characterized by subepithelial nerve plexus and ganglion cells close to the taste buds at the posterolateral border of the tongue, and is usually asymptomatic. However, SNP may exhibit some diverse and non-specific clinical features that may challenge the diagnosis and, consequently, the appropriate management. We present two cases of papular and symptomatic lesions on the posterior lateral border of the tongue, which were diagnosed as SNP. The objective of this study is to report these two cases of SNP with a review of the clinical characteristics of SNP reported in the literature.

Keywords: Nerve Tissue, Taste Buds, Tongue

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INTRODUCTION

Subgemmal neurogenous plaque (SNP) is characterized by the presence of a subepithelial nerve plexus and ganglion cells near the taste buds and lymphoid tissue at the posterolateral border of the tongue.[1-3] Currently, SNP is not considered a pathogenic condition, but a regional anatomic variation associated with the circumvallate, fungiform and foliaceous lingual papillae, and the lingual tonsil.[1-4]

The clinical features of SNP are not specific, and usually present as an asymptomatic lesion. In this setting, SNP may present from nodules to ulcers, reddish or white colored. Similarly, symptoms may be present, such as a burning sensation and pain.[1,3] The histopathological features of SNP are represented by the nerve plexus along the epithelium, which is composed of neural spindle cells and occasionally scattered ganglion cells through this arrangement. The association with neuroepithelial embryologic remnants (juxtaoral organ of Chievitz) is also described in some cases.[5]

SNP has only recently been described[2] and few studies have been published in the literature. Its diagnosis may be challenging for clinicians and pathologists due to this fact and the variety of its clinical and nonspecific histological characteristics.[1,3,6] Therefore, the goal of this study is to report two cases of SNP with a brief review of the clinical characteristics of SNP cases reported in the literature.

CASE REPORTS

CASE 1: A previously healthy 48-year-old Black female complained of a 2-month history of the presence of blood clots on her tongue. She also referred discomfort, pain, and many traumas in this region. An intraoral examination revealed an erythematous and papular lesion on the right posterior lateral border of the tongue, measuring 15 mm at its longest axis. The lesion was sessile and round-shaped with a smooth surface and soft consistency (Figure 1). During the manipulation of her tongue for clinical examination, the patient suddenly developed a hemorrhagic blister at the contralateral area, which was diagnosed as angina bullosa hemorrhagica, which healed in 1 week. However, the symptoms at the right-sided lesion persisted, and an excisional biopsy was performed with the working diagnosis of SNP, which was confirmed with histological examination. After surgical removal, the patient reported complete resolution of the symptoms. However, on the fourth month of follow-up, the pain relapsed without evidence of any clinical sign, which was diagnosed as a non-specific glossodynia and was managed with acupuncture. The result was successful.

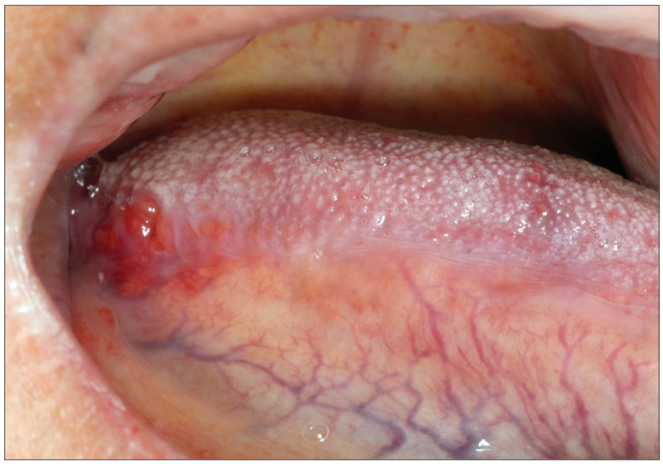

Figure 1. Oral examination of the Case 1: erythematous and papular lesion on the right posterior lateral border of the tongue.

CASE 2: A 56-year-old Caucasian female patient was referred with the working diagnosis of a malignant lesion due to a 10-day history of a bleeding injury at the posterior border of the tongue, which was previously treated with antibiotics. The intraoral examination identified a soft, sessile, papular, round-shaped lesion covered by normal mucosa with a smooth surface at the right posterior border of the tongue measuring 10 mm (Figure 2). An excisional biopsy was performed with the clinical hypothesis of SNP, which was confirmed by histological analysis. After surgical removal, the patient reported complete resolution of the symptoms with no recurrence by the 6-month follow-up.

Histological findings

Histological examination showed the presence of ovoid to spindle-shaped cells diffusely distributed throughout the lamina propria. Taste buds were embedded in the overlying epithelium (Figure 3). The cells were strongly positive for S100 (Figure 4).

DISCUSSION

In 1999, McDaniel[2] described SNP based on 12 examples from a large sample of human tongue biopsies. Since then, nine studies covering 75 cases on this issue have been published[1,3-10] according to a review at the PubMed database. Regarding the general characteristics, along with the presently reported cases, 28 SNP cases (37%) were diagnosed in male patients, while 49 (63%)

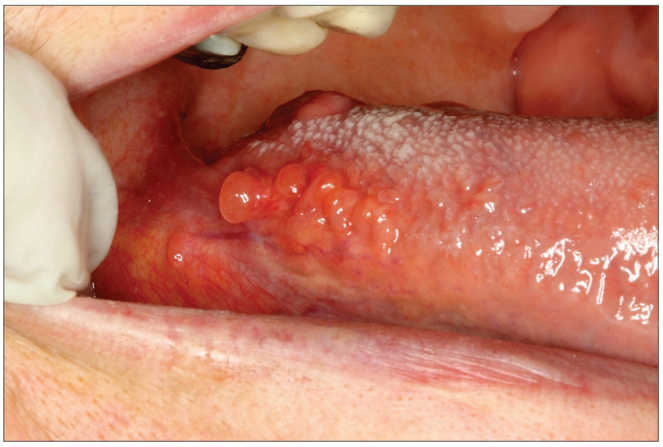

Figure 2. Oral examination of the Case 2: 10 mm papular lesion on the right posterior lateral border of the tongue recovered by normal oral mucosa in B, which was bilateral.

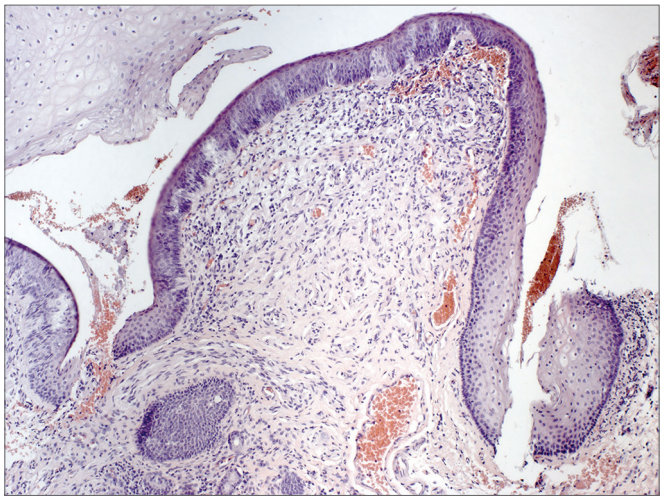

Figure 3. Photomicrograph of the both biopsies showed similar microscopic features represented by ovoid to spindle-shaped cells diffusely distributed throughout the lamina propria (H&E, 100X).

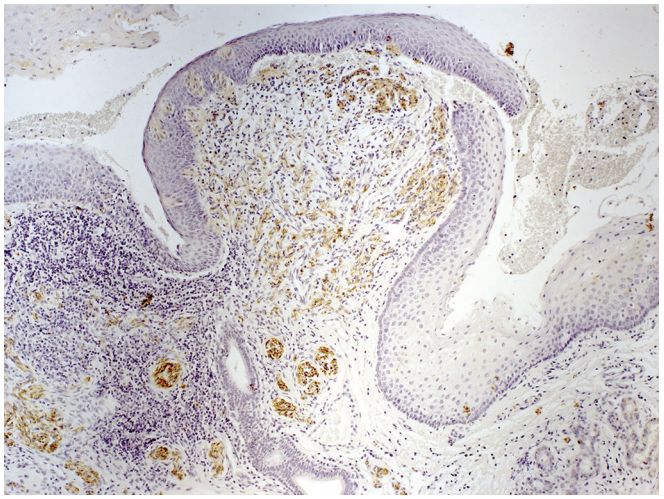

Figure 4. Photomicrograph of the immunohistochemical features of both cases showed the positivity for S100 in the cells of interest (100X).

were observed in female patients. The age ranged from 28 to 78 years, with a mean age of 56 years.

Although SNP occurrence was slightly more common in females, it does not appear to be gender related.[4] This entity is usually observed between the fifth and seventh decades of life, but five cases were reported under the age of 40 years,[1,2,7,9,10] and none was reported in childhood. Ethnical aspects have been described in our series, and by McDaniel[2] and Gonzaga et al.,[8] with a mild predominance of Caucasian patients (58%).

SNP occurs exclusively on the tongue, mainly at the posterolateral border, and has been established as a microanatomical variation.[3] The clinical presentation of SNP in published studies is diverse, varying from discoloration of the mucosa to erythematous or white plaque, and is associated with a papule/nodule or an ulcerated area. Some studies[2,3,7,9] consisted of retrospective evaluations of archived biopsies of the tongue lesions, and SNP was diagnosed in association with other lesions, such as fibroepithelial polyp,[2,7] neurofibroma,[2] focal fibrous hyperplasia,[2,3] squamous papilloma,[2] hemangioma,[2] oral squamous cell carcinoma,[3,7,9] oral lichen planus,[3,7] smoker's keratosis with dysplasia,[7] candidosis,[9] leukoplakia, and erythroplakia.[3]

The published case reports[1,5,8] and all the retrospective studies[2,3,7,9] described SNP as exophytic or nodular-shaped lesions occasionally associated with local erythema, which were surgically removed due to patients' symptomatology, and were clinically diagnosed as foliate papillitis or hyperplastic lingual tonsil. Two reports[4,6] refer to SNP diagnosed in autopsied cases without evidence of significant clinical changes, since the histological analyses were performed from samples of tongue papillae regions, circumvallate and foliate papillae, respectively, which can exhibit variations in shape and size. In the study by Val-Bernal et al.,[4] where they investigated an eventual oral primary tumor for a pulmonary malignant lesion (which caused the patient's death), SNP was an incidental finding. The Fonseca et al.[6] study aimed to demonstrate the importance of autopsy-based studies for oral pathologists, describing SNP as a normal anatomical structure.

In addition, Palazzolo et al.[5] reported two SNP cases that were diagnosed in biopsies of the tongue's base, also without any evidence of an apparent lesion. The biopsies were performed in pursuit of the primary site of a malignancy detected on cervical metastatic lymph nodes, which were diagnosed with squamous cell carcinoma. McDaniel[2] and Brito et al.[10] reported the most atypical cases regarding the location of SNP; the

former described two cases of SNP at the anterior portion of the tongue, and the latter at the dorsum. Since SNP is associated with taste buds, this regional variation is infrequent according to this review.

The clinical hypothesis for tongue lesions involves benign neoplasia from connective tissue origin, such as neuronal, vascular, and muscular lesions; or from reactive lesions, such as giant cell fibroma and focal fibrous hyperplasia. They are usually represented by asymptomatic submucosal masses that can change the color and surface of the overlying mucosa.[10] According to this review, the symptomatic cases tended to be clinically diagnosed as hyperplastic lingual tonsil or foliate papillitis, which is a form of transient lingual papillitis. The latter is usually described in fungiform papillae and is characterized by recurrent symptomatic and multiple white or erythematous papules[11]—the etiology and clinical relevance of which still remain controversial.[12]

Generally, there is a recommendation for surgical removal of SNP in symptomatic cases.[1,3,9] In our review, only one study[1] reported the follow-up (from 3 to 6 months) of SNP patients after the surgical procedure. In this study, no recurrences were observed. However, one of our cases had a relapse of symptoms after 4 months of follow-up, which required additional surgical intervention.

As SNP can be incidentally found as a microanatomical variation in biopsies undertaken with other diagnostic focus—and therefore cannot be described in the histopathological report—the actual incidence of this entity may be imprecise.

Similarly, there is a lack of knowledge on the effect of the surgical removal of SNP on the patient's quality of life. In this setting, ethical restrictions seem to hamper the design of a prospective study to demonstrate the frequency of SNP and the surgical therapy results.

Regarding the histopathological diagnosis, the pathologist should be aware of the similarity of SNP with other neuronal lesions, such as neuromas, neurofibromas, ganglioneuromas, ganglioneurofibromas, and traumatic neuromas, shown in previous studies.[3,9] For the clinician, this hypothesis should be included in biopsies of nodular lesions on the tongue, and symptomatic cases may benefit from surgical excision.

Finally, the identification of SNP is not common, and the dissemination of knowledge of clinical and histopathological characteristics contributes to avoiding misunderstandings or even underdiagnoses.

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