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Case report

Subgemmal neurogenous plaque of posterolateral region in tongue: A case report and review of literature

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ABSTRACT

Introduction: The subgemmal neurogenous plaque is a well-defined subepithelial neural plexus associated with taste buds, located within foliate, fungiform, and circumvallate papillae of the human tongue, that is poorly known by dentists and medical professionals. The hyperplasia of subgemmal neurogenous plaque may occur and it manifests as a papule, nodule, plaque, or ulcer in the lateral posterior border of the tongue inducing the overdiagnosis of reactive, cystic, or neoplastic lesions. The aims of this study are to report a case of a patient with subgemmal neurogenous plaque, and to provide a comprehensive review of other subgemmal neurogenous plaque in the posterolateral tongue with challenging diagnosis.

Presentation of case: A 28-year-old female with a little yellowish nodule, asymptomatic, located in posterior left lateral border of the tongue. Accurate diagnosis was based in excisional biopsy and histopathological examination showing a superficial neurofibroma-like pattern, and a neuroma-like in the deep zone. The final diagnosis was subgemmal neurogenous plaque.

Discussion: The subgemmal neurogenous plaque has an erythematous nodule or papule in the border of the tongue that are frequently misdiagnosis as foliate papillitis and lymphoepithelial cyst. Despite the subgemmal neurogenous plaque be considered a normal structure, the biopsy is recommended for differential diagnosis with other benign or malignant lesions in tongue.

Conclusion: Contributing with the dentists and medical professionals in their clinical practice, we reviewed the literature and added a case of subgemmal neurogenous plaque in the posterolateral region of the tongue, highlighting the importance of its recognition to avoid misdiagnosis.

1. Introduction

The subgemmal neurogenous plaque (SNP) is a normal morphologic structure found in the foliate, fungiform, and circumvallate papillae of human tongue, characterized by subepithelial neural plexuses associated with the taste buds [1]. Each papilla usually contains one plaque that is identified in tongue biopsies [2].

Clinically, the hyperplasia of the SNP may occurs manifestation as an asymptomatic small nodule or papule, white plaque or ulcer in border of the tongue, occasionally, associated with local burning [2–7]. These non-specific clinical features may be confused with cyst lymphoepithelial, inflammatory fibrous hyperplasia, or squamous cell carcinoma that affects the posterolateral region of tongue [2,8]. The development of hyperplastic SNP has been associated to trophic influence from gustatory nerve fibers or reactive lesion in foliate papillae of tongue frequently submitted to trauma [2].

The histopathological features of the SNP show a biphasic pattern with a superficial layer presenting neurofibroma-like proliferation, and a deep area composed of tortuous neuronal bundles with ganglion cells as neuroma-like [1,2,9]. Inexperienced oral pathologists can microscopically confuse, the SNP with other pathologies, such as neurofibroma, ganglioneuroma, or traumatic neuroma [3,10].

In order to contribute to the knowledge of dentists and medical professionals in identifying the clinical and microscopic aspects of the SNP in the posterolateral region of tongue, we reviewed the English-language literature adding the present case report. This case report has been reported in line with SCARE 2020 criteria [11].

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Fig. 1. Asymptomatic yellow nodule in left posterolateral border of tongue. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

2. Case presentation

A 28-year-old Caucasian female, student was referred to the dentist for evaluation of a lesion in tongue within one month of duration. Intraoral examination revealed a little 0.4 cm yellowish nodule, asymptomatic and with regular contour, located in posterior left lateral border of the tongue (Fig. 1A). The history medical irrelevant. There was no history trauma associated to lesion or no abnormal alterations in extraoral examination. The clinical diagnosis was a lymphoepithelial cyst or hyperplastic lingual tonsil. An excisional biopsy with conventional scalpel and local anesthesia was performed by the dental surgeon and submitted to histopathological analysis. Microscopic features of the lesion revealed a foliate papilla covered by parakeratinized stratified squamous epithelium containing taste buds (Fig. 2A and B) and underlying spindle cells organized in cords surrounded by collagen in a pattern neurofibroma-like proliferation (Fig. 2C). More deeply, several irregular neural bundles circumscribed by the perineurium layer distributed in a pattern neuroma-like were also observed (Fig. 2D). In addition, focuses of mononuclear inflammatory infiltrate in subepithelial region were found. The final diagnosis was subgemmal neurogenous plaque and the patient was oriented about her clinical condition. After one month of follow-up, the patient presented with normal healing and no complaints.

3. Review of literature of subgemmal neurogenous plaque in margin of tongue

A review of the English-language literature was performed including the term "subgemmal neurogenous plaque," and "subepithelial nerve plexus + human tongue" from 1999 to 2021, in the PubMed, Scopus and Google Scholar databases. In first step, a primary selection was performed including studies such as: (1) manuscripts in English language; (2) all duplicates' articles are excluded; (3) complete published articles: Original research, retrospective studies, letter to the editor and cases reports. A total of 16 manuscripts with 117 cases of SNP were retrieved and fully read. Posteriorly, the followed eligibility criteria were applied: (1) diagnosis of SNP in the posterolateral region of tongue (foliate papilla), confirmed by histopathologic analysis; (2) complete patient's clinical information (sex, age, and location of the lesion); (3) photomicrography of the subgemmal neurogenous plaque. Fifty cases of SNP in 8 published manuscripts were selected using the eligibility criteria (Fig. 3).

The SNP patients' informations are summarized in Table 1. The cases of SNP associated with lymphoepithelial cyst, which it was not possible to confirm the specific patient informations (sex, age, location), were excluded.

4. Discussion

The subepithelial nerve plexus was, initially, characterized as a normal structure associated with taste buds in papillae of tongue [1] appears to be more common than previously considered [7]. Besides the tongue, the SNP was also described in the glottis and tonsillar fossa [2,7].

This neural structure can become hyperplastic and it manifests as a

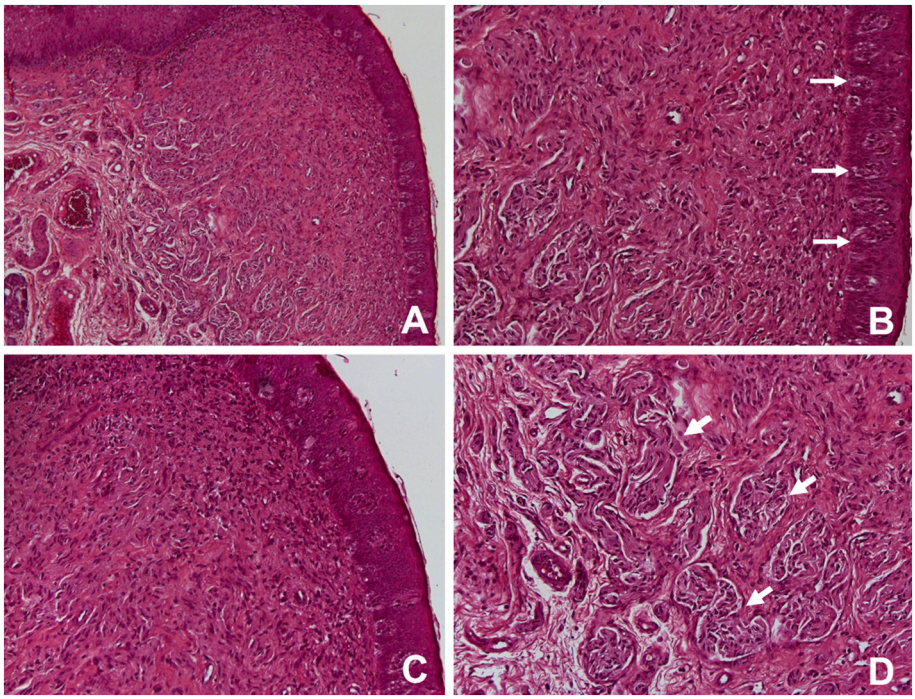

Fig. 2. Photomicrography of subgemmal neurogenous plaque. (A) Panoramic microscopic features of subgemmal neurogeous plaque located in foliate papilla. (B) Detail of the foliate papilla covered by parakeratinized stratified squamous epithelium containing taste buds (white arrows). (C) Superficial subgemmal neurogenous plaque containing spindle cells organized in cords surrounded by collagen in a pattern neurofibroma-like proliferation. (D) Deep subgemmal neurogenous plaque with irregular neural bundles circumscribed by the perineurium layer distributed in a pattern neuroma-like (white arrows).

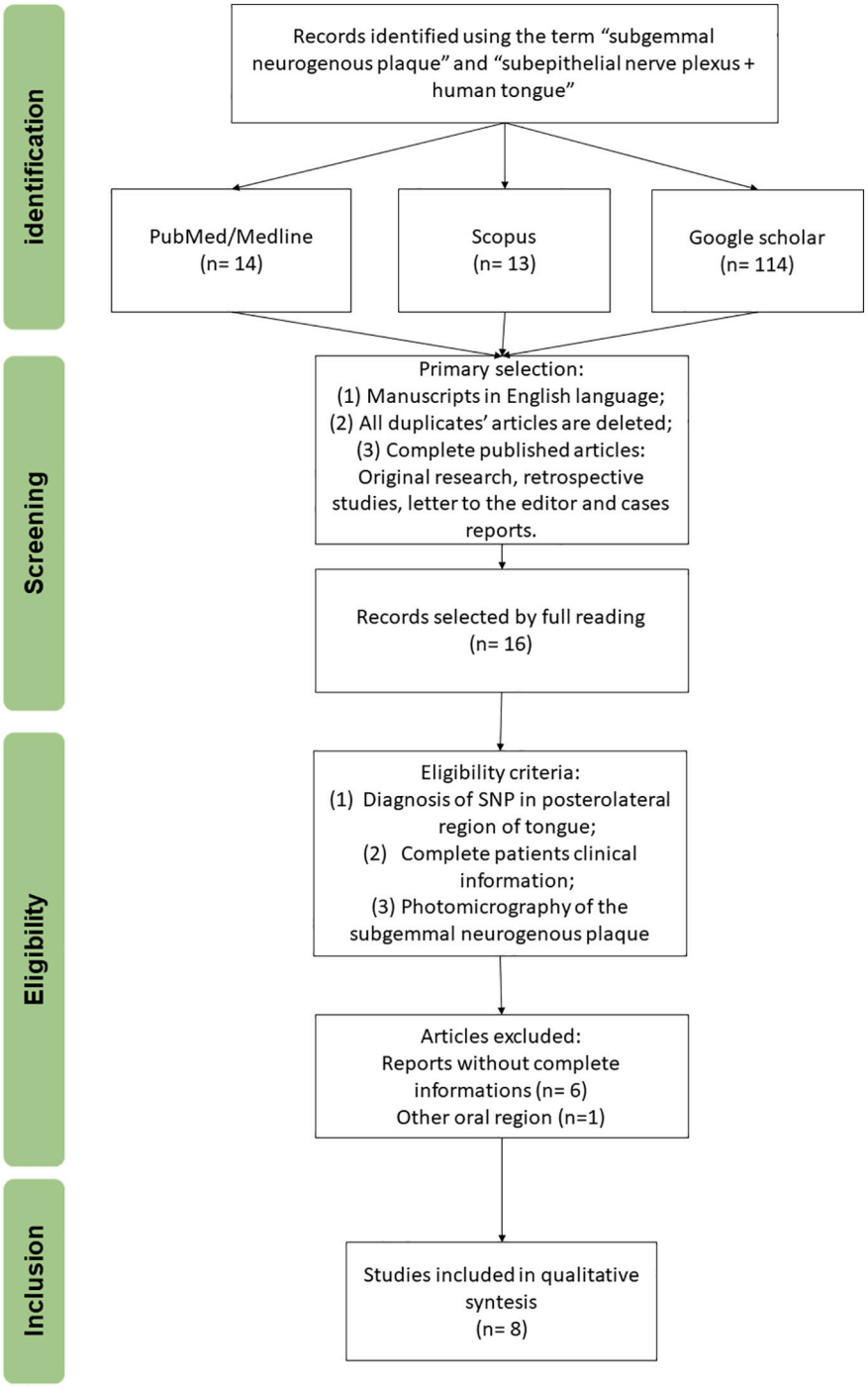

Fig. 3. Flowchart of the steps established for literature review.

Table 1
Clinical features of 51 patients with subgemmal neurogenous plaque located in posterolateral region of tongue.

Author/year	Sex	Age	Clinical features	Clinical diagnosis
McDaniel 1999	7M 5 F	33 to 61 years	NA	Foliate papillitis, Lymphoid hyperplasia, Lymphoepithelial cyst, Incidental findings
Triantafyllou & Coulter 2004	5 M 8 F	31 to 74 years	Exophytic, with plaque, erythematous, papillomatous, nodule, and normal tissue	Foliate papillitis, Lingual tonsil, Dysplasia, Squamous cell carcinoma, Lichen planus
Gueiros et al. 2008	2 F	34 and 61 years	Erythematous and red swelling	Lingual papillitis, Contact allergy
Gueiros et al. 2009	3 M 4 F	34 to 58 years	NA	Candidosis, Hyperplasia lingual tonsil, Squamous cell carcinoma
Gonzaga et al. 2017	2 M 1 F	41 to 77 years	Nodule, papula, and erythematous	Fibrous hyperplasia, Leukoplakia, Foliate papillitis
Agrawal et al. 2018	1 M	28 year	Nodule	NA
Heguedusch et al. 2019	2 F	48 and 56 years	Papula	Subgemmal neurogenous plaque
Alnajar et al. 2019	2M 8 F	30 to 66 years	Fluctuating lesion	NA
Present case 2022	1 F	28 year	Nodule	Lymphoepithelial cyst or hyperplastic lingual tonsil

M: masculine, F: female, NA: not available.

nodule, papule, white plaque, or ulcer in posterolateral region of tongue [2,3,5–7,12].The present case report illustrates the usual clinical and histopathological characteristics of SNP in posterior border of tongue (Figs. 1A and 2A), confirming the findings of our literature review using the PubMed, Scopus, and Google Scholar databases (Table 1).

Clinically, the SNP occurs, mainly, as an erythematous nodule or papule in border of the tongue of the adult female patient with age varying from 31 to 77 years (Table 1), confirming the findings of other studies [6,10,13]. Although the SNP in tongue can be asymptomatic, it has been suggested that the burning sensation and pain in region of lesion reported by patients [2–7,13] probably is due to chronic trauma that cause stimulation of the nerve fibers directly or via inflammatory mediators [2,3].

According to our review of literature, the SNP in lateral border of the tongue is frequently confused with other oral benign and malignant lesions including foliate papillitis, lymphoepithelial cyst, lingual tonsil, and squamous cell carcinoma (Table 1). Despite the SNP be a normal structure usually without signs, symptoms or significant clinical behavior, the biopsy is necessary for differential diagnosis with other benign or malignant lesions in posterolateral region of the tongue [2,3,7,10].

Recent studies identified the SNP in posterolateral region of the tongue as an incidental finding adjacent or in association with the lymphoepithelial cyst [9,13,14]. The retrospective study of Cunha et al. 2021, based on the clinical and histopathological features of 132 oral lymphoepithelial cysts, found the SNP in 11.3% of the 80 cysts located in the tongue [13]. In our case reported, the histopathological analysis confirmed the SNP as a reactive lesion in foliate papilla of tongue, not associated with lymphoepithelial cyst and with the presence of mononuclear inflammatory infiltrate. Therefore, the present case suggests that the etiopathogenesis of isolated and hyperplastic SNP seems to be related to trophic influence from gustatory nerve fibers or to trauma in posterior border of the tongue [2,6].

Microscopically, the characteristics of SNP of tongue with spindle cells proliferation surrounded by collagen and deep region with irregular neural bundles suggest the other benign neural tumors as neurofibroma or neuroma traumatic and it can confuse the inexperienced oral pathologists [10,15]. Additionally, Van Bernal et al. 2006, reported a case of SNP in a lingual circumvallate papilla of tongue associated with pseudoepitheliomatous hyperplasia at autopsy and, reinforced that the knowledge of SNP morphological spectrum is essential to avoid the misdiagnosis as squamous cell carcinoma [8].

This review of literature, including the present case report, reinforces that diagnosis of SNP remain challenger, particularly, when the lesion manifests as an asymptomatic nodule or plaque in posterior border of the tongue. In fact, further cases of SNP should be published in order to contribute with the clinical and histopathological features of this anatomic structure associated with the taste buds of tongue.

Despite of few cases published of SNP in border of tongue, the dentist and medical professionals should be able to recognize its clinical characteristics and the histopathological analysis can be necessary to avoid misdiagnosis providing adequate management of patient.

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Ethical approval

Written informed consent was obtained from the patients for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Consent

Written informed consent was obtained from the patients for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Author contributions

G.L.S and D.T.O. wrote the manuscript with inputs from all authors; D.T.O. analyzed the microscopic imagens and contributed to the diagnosis; C.LC. provided care for the patient. All authors discussed, revised, and contributed final approval of the manuscript.

Registration of research studies

None.

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Declaration of competing interest

No conflict of interest.

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