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Congenital Epulis Of The Tongue In An Adult Female: A Rare Case Report

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Abstract
Congenital epulis is a rare benign soft tissue tumor predominantly seen in neonates, usually arising from the alveolar ridges. Its occurrence in adults is exceedingly rare, and involvement of the tongue is exceptional. We report a rare case of congenital epulis of the tongue in a 39-year-old female patient who reported to the Department of Oral Medicine and Radiology, Government Dental College, Alappuzha. The lesion presented as a painless, well-circumscribed nodular mass on the dorsal surface of the tongue. Histopathological examination, including review of archived biopsy sections obtained three years earlier, confirmed the diagnosis. This case highlights the importance of considering congenital epulis in the differential diagnosis of tongue lesions in adults.
Keywords: *Congenital epulis, granular cell tumor, tongue, adult female, rare oral lesion*

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I. Introduction

Congenital epulis (CE), also known as congenital granular cell tumor, is an uncommon benign lesion first described by Neumann in 1871.[1] It typically presents at birth or during the neonatal period and shows a marked female predilection.[2] The lesion most commonly arises from the anterior alveolar ridge of the maxilla, followed by the mandible.[2] Extra-alveolar occurrence is extremely rare, and presentation in adulthood is exceptional.[7] Due to its rarity and clinical resemblance to other soft tissue tumors, congenital epulis may be misdiagnosed in adult patients.[8] The present report describes a rare case of congenital epulis occurring on the tongue in a 39-year-old female patient.

II. Case Report

A 39-year-old female patient reported to the Department of Oral Medicine and Radiology, Government Dental College, Alappuzha, with a chief complaint of a painless growth on the tongue noticed incidentally. The patient could not recall the exact duration of the lesion but reported a slow and gradual increase in size. There was no history of pain, bleeding, ulceration, dysphagia, speech difficulty, or functional impairment. The patient's medical, dental, and family histories were non-contributory. No history of trauma, parafunctional habits, or similar lesions during childhood was reported. Extraoral examination did not reveal any facial asymmetry, swelling, or cervical lymphadenopathy.

On intraoral examination, a solitary, well-defined nodular mass was noted on the dorsal surface of the tongue, located close to the midline in the anterior two-thirds region. The lesion measured approximately 2.5 × 2 cm and appeared oval in shape with well-demarcated margins. The surface was smooth and intact, with no evidence of ulceration, crusting, secondary infection, or surface keratinization. The lesion was pinkish in color, similar to the adjacent normal mucosa, with no areas of erythema, blanching, pigmentation, or surface vascularity. The surrounding lingual mucosa appeared healthy and non-inflamed, with preservation of normal papillary architecture. There was no evidence of surface fissuring, trauma from opposing teeth, indentation marks, active bleeding, or discharge. On palpation, the mass was firm in consistency, non-tender, and non-fluctuant. The lesion was non-compressible and non-pulsatile, with no evidence of thrill or bruit. The margins were well delineated from the surrounding tissues, and the surface texture was smooth. The lesion was freely movable over the underlying structures, suggesting a superficial soft tissue origin without deep tissue fixation. No blanching on pressure or bleeding on manipulation was observed. The temperature of the overlying mucosa was normal, and no induration was noted in the surrounding tissues.

Based on the clinical findings, a provisional diagnosis of a benign soft tissue tumor of the tongue was considered. Review of the patient's records revealed that an incisional biopsy had been performed three years earlier, and archived histopathological sections were available for evaluation.

Differential Diagnosis

The differential diagnosis included adult-type granular cell tumor, fibroma, neurofibroma, schwannoma, lipoma, and lingual thyroid.

Histopathological Findings

Histopathological examination of the archived hematoxylin and eosin–stained sections obtained three years earlier revealed a well-circumscribed, non-encapsulated lesion composed of sheets and nests of large polygonal cells separated by scant connective tissue stroma. The tumor cells exhibited abundant eosinophilic granular cytoplasm with centrally placed round to oval nuclei and inconspicuous nucleoli. No mitotic figures, cellular pleomorphism, or necrosis were observed. The overlying stratified squamous epithelium was thin and atrophic, without evidence of pseudoepitheliomatous hyperplasia. These features are characteristic of congenital epulis and help distinguish it from adult-type granular cell tumor.

III. Discussion

Congenital epulis is a rare benign soft tissue tumor that is classically encountered in the neonatal period, with more than 90% of cases reported at birth or within the first few days of life and a strong female predilection.[5] The lesion most frequently arises from the anterior alveolar ridge of the maxilla, followed by the mandible.[2] Extra-alveolar locations are distinctly uncommon, and adult presentation of congenital epulis is exceptional.[7] Involvement of the tongue represents one of the rarest documented sites of occurrence. The present case is therefore unusual both because of the patient's age and because of the extra-alveolar lingual location, making it a diagnostically challenging entity in routine oral clinical practice.

The etiopathogenesis of congenital epulis remains uncertain. Several hypotheses have been proposed regarding its cellular origin, including derivation from undifferentiated mesenchymal cells, fibroblasts, histiocytes, myogenic cells, and pericytes.[2] Although hormonal influence has been proposed to explain the marked female predilection, immunohistochemical studies demonstrating absence of estrogen and progesterone receptor expression in tumor cells weaken this hypothesis.[3] Furthermore, the occurrence of congenital epulis in adult patients, as observed in the present case, further challenges the concept of a hormonally driven lesion confined to the neonatal period.[7] The long-standing presence and indolent behavior of the lesion in this patient suggest that congenital epulis may represent a developmental hamartomatous proliferation with the capacity for long-term persistence rather than a purely congenital, transient entity.

Clinically, congenital epulis typically presents as a smooth-surfaced, sessile or pedunculated, painless mass.[2] In neonates, large lesions may interfere with feeding or respiration; however, in adults, the lesion is usually asymptomatic and discovered incidentally. In the present case, the lesion was slow growing, non-ulcerated, and asymptomatic, consistent with the benign biological behaviour reported in the literature.[9] The absence of functional impairment and the lack of regional lymphadenopathy further supported a benign diagnosis. The long-standing nature of the lesion, supported by the availability of archived histopathological sections obtained three years earlier, underscores the indolent course of congenital epulis and highlights that these lesions may remain clinically stable for prolonged periods without malignant transformation.[12]

The most important diagnostic consideration in this case is differentiation from adult-type granular cell tumor (GCT), which commonly involves the tongue and shares a similar granular cytoplasmic morphology.[11] Adult-type GCT typically exhibits pseudoepitheliomatous hyperplasia of the overlying epithelium and shows immunoreactivity for S-100 protein, reflecting its Schwann cell origin.[2] In contrast, congenital epulis lacks pseudoepitheliomatous hyperplasia, is S-100 negative, and demonstrates a distinct immunohistochemical profile, supporting a non-neural origin.[11] These histopathological and immunohistochemical differences are crucial, as misinterpretation of epithelial hyperplasia in adult-type GCT may lead to an erroneous diagnosis of squamous cell carcinoma. The absence of pseudoepitheliomatous hyperplasia in the present case, along with the classic granular cytoplasm and bland nuclear features, favoured a diagnosis of congenital epulis over adult-type granular cell tumor.

Other lesions considered in the differential diagnosis of a nodular mass on the dorsal tongue include fibroma, schwannoma, neurofibroma, lipoma, leiomyoma, vascular malformations, and ectopic lingual thyroid.[4] However, these entities demonstrate distinct clinical or histological features that aid in differentiation. The firm consistency, absence of blanching or pulsation, lack of vascular surface changes, and characteristic granular histomorphology effectively excluded vascular lesions and adipocytic or neural tumors in the present case. Lingual thyroid, although rare, typically presents as a midline posterior tongue mass associated with dysphagia or dysphonia and shows thyroid tissue histologically, features not observed in this patient.[4]

From a biological behavior standpoint, congenital epulis is regarded as a non-aggressive lesion with an excellent prognosis. Notably, spontaneous regression has been reported in neonates, and there are no documented cases of malignant transformation or true recurrence, even following incomplete excision.[9] In adults, surgical excision remains the treatment of choice, primarily for definitive diagnosis, elimination of potential functional interference, and patient reassurance. The stability of the lesion over several years in the present case further reinforces the indolent nature of congenital epulis and supports conservative surgical management when indicated.

This case emphasizes the importance of maintaining a broad differential diagnosis when evaluating long-standing, asymptomatic nodular lesions of the adult tongue. Although congenital epulis is traditionally regarded as a neonatal lesion of the alveolar ridges, rare extra-alveolar and adult presentations do occur and may be easily misdiagnosed as more common entities such as adult-type granular cell tumor. Awareness of this rare presentation, combined with careful histopathological evaluation, is essential for accurate diagnosis and appropriate management. Documentation of such atypical cases contributes to a better understanding of the biological spectrum of congenital epulis and expands the existing literature on rare oral soft tissue tumors.

IV. Conclusion

Congenital epulis of the tongue in an adult female is an extremely rare clinical entity. This case underscores the importance of including congenital epulis in the differential diagnosis of tongue masses in adults. Histopathological evaluation remains essential for accurate diagnosis and appropriate management. Documentation of such rare presentations contributes valuable information to the existing literature.

Declaration of Patient Consent

The authors certify that appropriate patient consent was obtained for clinical examination and publication of clinical photographs and histopathological images, ensuring patient anonymity.

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Nil.

Conflicts of Interest

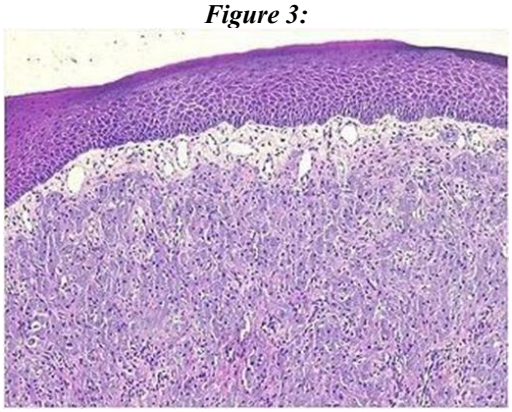
There are no conflicts of interest.

Figure Legends

Figure 1 ***Figure 2***

Clinical photographs showing a well-defined, smooth-surfaced nodular and pedunculated mass arising from the dorsal surface of the tongue in a 39-year-old female patient.

Figure 3:



Histopathological photomicrograph showing thin atrophic stratified squamous epithelium overlying sheets of large polygonal cells with abundant eosinophilic granular cytoplasm and centrally placed nuclei, consistent with congenital epulis (Hematoxylin and Eosin stain, ×10).

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