LINGUA VILLOSA NIGRA WITH CHRONIC KIDNEY DISEASE- A CASE REPORT

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PRESENTATION OF THE CASE

A 55-year-old married female was referred from the department of Nephrology as a case of chronic kidney disease due to hypertensive nephropathy with blackish lesions on the dorsum of tongue. The discolouration was of two months duration. Patient was a known case of hypertension and diabetes for the past 12 years, on regular treatment with oral antihypertensives- telmisartan 40 mg once daily, and oral hypoglycaemic agents- glimepiride 1 mg twice daily, metformin 500 mg once daily. Patient had undergone 10 sittings of haemodialysis for the renal failure in the past before the aetiology of BHT. There was no history of tobacco abuse or any use of mouthwash. None of her siblings or her parents had similar lesions in the tongue. General examination revealed anasarca and pallor. Oral mucosal examination revealed poor oral and dental hygiene, xerostomia and blackish hairy projections on the dorsum of tongue. [Fig:-1]. There was no regional lymphadenopathy. Scraping of the tongue and KOH examination was found to be negative for fungal infection. Bacterial and fungal culture of the tongue lesion did not reveal any growth. Biopsy was not done because patient did not give consent for biopsy. Routine investigations revealed proteinuria (albumin 3+), severe anaemia (Hb: 6 g/dL), and abnormal renal function tests (Serum urea: 87 mg/dL, Serum creatinine 8 mg/dL). Serology was negative for HIV, Hepatitis B and C. Hence, this case was clinically diagnosed as lingua villosa nigra. Patient was advised to maintain proper oral hygiene with regular brushing of tongue with soft brushes, and advised to maintain oral hydration. The case was followed up periodically for 1 month. The lesion disappeared significantly.

Aetiopathogenesis

The aetiopathogenesis of BHT is not clearly understood and is multifactorial. Risk factors for developing BHT include mostly males with the use of tobacco, alcohol and intravenous drugs, poor oral hygiene, xerostomia, general debilitation, fissured tongue and macroglossia, recent radiation therapy to the head and neck regions, advanced age, and neurological disorders like trigeminal neuralgia[1,2] affecting tongue movement and mastication. Prolonged use of oxidising mouthwashes containing sodium perborate, sodium peroxide, and hydrogen peroxide has also been associated with the development of BHT. Dietary consumption of herbal tea and sugars may lead to lowering pH on the dorsum of the tongue promoting chromogenic bacterial overgrowth producing BHT.[3] Drugs like topical or systemic antibiotics including penicillin, erythromycin, doxycycline, neomycin, and linezolid have been associated with this disorder. Antipsychotics (fluoxetine, olanzapine causing dry mouth) as well as psychotropic agents can also predispose to the development of BHT. Additionally, other medications, including fluoxetine, olanzapine, lansoprazole, and bismuth, can precipitate BHT.[4,5] Recently, it has been reported in association with the use of Tyrosine kinase inhibitors like Erlotinib.[6] The exact mechanism for BHT is unknown, but the theory propagated delineates to defective desquamation of dorsal surface of the tongue. This defective desquamation prevents normal debridement leading to accumulation of keratinised layers or delayed shedding of the cornified layer that results in excessive growth and thickening of the filiform papillae[7] and then secondarily collection of debris, bacteria, fungi or other foreign materials, which contribute to its discolouration. This collection can include residue from tobacco, tea, coffee and other foods, as well as porphyrin-producing chromogenic organisms in the oral flora that lends its characteristic hue. Another theory put forth by Prinz in 1925 regarding main source of pigmentation was that of a local reaction between decomposed food products and iron (secondary to blood within the mouth).[8] Interference of the melanin at local level could be another factor to be taken in to consideration. In our patient, the predisposing factors for the development of BHT would have been xerostomia due to chronic kidney disease, and poor oral hygiene due to general debility.

DIFFERENTIAL DIAGNOSES

Differential diagnoses of BHT includes oral hairy leukoplakia, pseudo-hairy tongue, pigmented fungiform papillae of the tongue and acanthosis nigricans.[3,9] "Pseudo-black hairy tongue" appears as a darkly stained tongue in absence of elongated filiform papillae seen in BHT. Oral hairy leukoplakia can be seen in the immunocompromised patients and has a white plaque appearance on the dorsal and ventral surfaces of the tongue. Pigmented (Due to melanin laden macrophages) fungiform papillae are rare, characterised by isolated hypertrophied lesions primarily on the lateral aspect

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and apex of the tongue that has a predilection to dark skinned patients.

Acanthosis nigricans in the oral cavity manifests as multiple dark and demarcated papillary lesions on the dorsum and lateral region of the tongue with frequent labial involvement and may be associated with underlying malignancy. Detailed history and physical examination is important for diagnosis, with particular emphasis on identifying known aetiological factors.

CLINICAL DIAGNOSIS

BHT is usually asymptomatic, but occasionally it can produce nausea, halitosis, dysgeusia. The diagnosis of BHT is by visual intraoral examination. It shows a predilection for the dorsal aspect of tongue, anterior to the circumvallate papillae and sulcus terminalis.[3] Microscopic examination may be used as an adjunct to diagnosis; demonstrating elongated filiform papillae on the dorsal tongue more than 3 mm in length. Cultures may be considered to rule out superimposed bacterial or fungal infections associated with BHT.[3,9] Tongue biopsy is supportive but not usually required if the lesion appears characteristic for BHT. Our patient was asymptomatic, and on oral examination, discolouration was found to involve the dorsum of tongue anterior to the circumvallate papillae. There was xerostomia too. The lesion was not scrapable. KOH scraping was negative for fungal elements. Bacterial and fungal cultures of the swab taken from the lesions did not reveal any growth. Hence, it was clinically confirmed as BHT.

DISCUSSION OF MANAGEMENT

Black hairy tongue (BHT) was originally described by Amatus Lusitanus in 1557 as hairs on the tongue that would regrow upon being removed. BHT has been described with various names- hyperkeratosis of the tongue, lingua villosa nigra, nigrites linguae, keratomycosis linguae, and melanotrichia lingua. It is an asymptomatic benign medical condition characterised by the appearance of abnormally hypertrophied and elongated filiform papillae on the dorsal surface of the tongue. The name is a misnomer because of its classical presentation as a superficial black and hairy carpet-like lingual growth. Hairy tongue may also appear in varied colours like brown, yellow, green, blue, or even unpigmented. Prevalence of BHT in various studies has been reported in a wide range varying from 0.6% to 11.3%.[3] The youngest ages where BHT has been reported was in two week and two-month-old babies.[10,11]

Mechanical debridement and removal of the suspected precipitating factor leads to resolution of the condition within few weeks. Patient education on proper oral hygiene and lifestyle modifications is important. Therapeutic options of modest benefit include increasing hydration and salivation, discontinuing smoking, brushing the tongue with a soft toothbrush enhanced by previous application of 40 percent urea, applying topical retinoids or salicylic acid, or undergoing surgical excision.[12] BHT is a self-limiting disease with a good prognosis, but rarely may cause aesthetic anxiety, hence timely recognition of the condition, and early intervention with reassurance to the patient is mandatory.[3]

FINAL DIAGNOSIS

A wide range of oral mucosal lesions were described in individuals with end stage renal disease (ESRD) receiving dialysis and in renal transplant patients. These include white patch, erythematous patch, ulceration, lichen planus, oral hairy leukoplakia, uremic stomatitis, macules/nodules, fibroepithelial polyps, geographic tongue, black hairy tongue, papilloma, pyogenic granuloma.[13]

In this patient, probably general debility and xerostomia due to chronic kidney disease have predisposed for the development of lingua villosa nigra. Clinicians should be aware of the prevalence, the predisposing factors and drug classes that may play a role in the development of BHT and its treatment.

Figure. 1 Black Hairy Tongue on the Dorsum of the Tongue

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