

Case Report

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Walnut consumption as a cause of black tongue: A case report

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Abstract

Extrinsic stain and black hairy tongue need to be considered in the differential diagnosis of discoloration of the tongue. Both are benign conditions, characterized by altered coloration of the dorsum of the tongue. However, they differ in the macro- and microscopic appearance, with black hairy tongue presenting with hypertrophy and elongation of the filiform papillae. In the absence of such manifestation, extrinsic stain is a more likely diagnosis. Several factors have been associated with extrinsic stain including poor oral hygiene, dark beverage intake, smoking habit, yeast infections, topical or systemic medications. There are few reports of food substances contributing to extrinsic stain. In this report, a persistent black discoloration of the tongue completely resolved within 2 weeks after discontinuation of daily walnut consumption (15-20 per day). Clinicians should familiarize with this unusual side-effect of walnuts, especially when all the other etiological factors have been excluded.

Keywords: Tongue hairy; Juglans; Pigmentation.

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Background

Oral staining can be secondary to intrinsic or extrinsic environmental factors [1]. Extrinsic staining is recognized mostly on dental surfaces and is commonly associated with the use of chlorhexidine mouthwash [2-4], certain toothpastes [5], and frequent intakes of tea and/or coffee [6,7]. When the discoloration appears on a mucosal surface, the clinician should be careful to take a thorough history and build a differential diagnosis based on the findings.

Similar to extrinsic strain, Black Hairy Tongue (BHT) is a benign and self-limiting condition, clinically characterized by a brown-black pigmentation, although yellow greenish discoloration has been reported [8]. What distinguishes the two conditions is the presence of elongated and hypertrophic keratinized

filiform papillae in BHT, that result in a hairy appearance of the dorsum of the tongue [9,10]. The pathophysiology of BHT is unclear, although a lack of adequate desquamation of keratin along with the presence of porphyrin-producing chromogenic bacteria or yeast are the most accredited hypotheses [9]. Many factors are associated with the development of this condition, including medications (antibiotics, antipsychotics, proton pump inhibitors, immunomodulators, antidepressants, antineoplastics), infections (yeast, hepatitis), medical conditions (malignancies, trigeminal neuralgia, graft-versus-host disease, amyotrophic lateral sclerosis), habits (smoking, excessive alcohol intake, high consumption of beverages such as coffee or black tea), and environmental factors (poor oral hygiene, xerostomia, oxidizing mouthwashes) [9].

Regardless of the differential diagnosis, many patients with discoloration of the tongue seek treatment for aesthetic concern [11]. Commonly, once the offending agent has been discontinued and the stain removed with manual or electric toothbrushes [6], the condition resolves. However, this implies a correct identification of the initiating factor, among those known to be associated with this condition.

The aim of this report is to present the case of a patient suffering from a mucous membrane pemphigoid well-controlled with topical medication, presented at a recall visit with an unexplainable appearance of black discoloration of the tongue that had persisted for 12 months. The condition did not respond to the reduction in coffee intake nor to daily brushing with a solution of toothpaste mixed with pumice, but completely resolved within 2 weeks after discontinuation of daily walnut consumption (15-20 per day). To the best of our knowledge, this is the first report of extrinsic stain secondary to walnut intake that appears in the scientific literature.

Case report

A 75-year-old female presented at the Orofacial Pain Clinic (University of Kentucky, College of Dentistry, Lexington, USA), where she was evaluated by an oral medicine specialist (C.S.M.) for a chief complaint of painful and cyclic intra-oral ulcers on the right posterior mandibular alveolar ridge and buccal mucosa, started few months before.

Medical history and medication regimen of the patient at the time of the first visit are presented in Table 1. A cranial nerve screening did not reveal any abnormalities. Palpation of temporomandibular joint and musculoskeletal orofacial muscles was negative. An intraoral examination demonstrated the presence of painful irregular shaped ulcers with yellowish slough, surrounded by an erythematous halo, located at the right posterior mandibular alveolar ridge and buccal mucosa.

Diagnosis and management

Table 2 summarizes the work-up that led to a diagnosis of Hyposalivation, Herpes Virus (HSV-1) infection and Mucous Membrane Pemphigoid (MMP). The hyposalivation was addressed by providing a dry mouth protocol, with all the recommendations aiming at increasing the daily fluid intake and the use of fluoride-rich toothpaste for enamel protection. The HSV-1 infection was treated with an antiviral therapy (as illustrated in Table 2). The clinical and histopathological diagnosis of MMP was managed with a daily therapeutic protocol (Table 2), along with a referral to an ophthalmologist to rule out ocular complications. After 1 month of daily medication regimen for MMP, the patient reported a decrease in size and symptomatology associated with her intraoral ulcers.

Follow-up visits

At 3-month appointment, the patient reported a complete resolution of her intraoral lesions, except for rare and occasional symptoms lasting a day or two. Management for MMP included blood tests repeated every 3 months, follow-ups with the oral medicine specialist and ophthalmologist every 6 months, and long-term systemic therapeutic protocol consisting of doxycycline 100 mg BID, clotrimazole 100 mg BID, niacinamide 400 mg TID and clobetasol 0.05% TID. One year later, the systemic

medication regimen was suspended, due to the development of cutaneous photosensitivity reactions on her face and arms. Since then, only topical application of clobetasol 0.5% was continued, with the recommendation of introducing budesonide rinse 3 mg in case of symptomatic flare-ups.

Over the following 7 years, the patient was mostly asymptomatic with successful management with topical clobetasol 0.5% and budesonide rinse 3 mg.

At her routine 8-year follow-up, the patient complained of dry mouth. At that time, she indicated enjoying 9 cups of caffeinated coffee per day. In light of the presence of a heart rate above 100 beats per minute and generalized coffee stains noted on the teeth, a recommendation to reduce the daily consumption of caffeinated drinks was provided, along with a referral to her primary care physician for evaluation of the rapid heart-beat. Dry mouth protocol as introduced at her first visit was reinforced.

Appearance of a new chief complaint

Six months later, the patient returned to the Orofacial Pain Clinic with a complaint of a “black tongue”, accompanied by xerostomia and tingling sensation on the dorsum of the tongue (Figure 1A). Her updated medical history revealed supraventricular tachycardia, which has been managed by her primary care physician with daily metoprolol (Table 1). Dietary assessment was performed and revealed a daily intake of 6 cups of caffeinated coffee, 2-3 glasses of water, 15-20 walnuts, eggs, fruit and vegetables, and high consumption of high-processed sugary food (cookies). An extraoral examination excluded evidence of swelling, asymmetry or lymphadenopathy. Results of chairside assessments were consistent with hyposalivation (Table 2) and ruled out oral fungal infection. The diagnosis of hyposalivation was confirmed and the dry mouth protocol was reiterated. As for the main chief complaint of the patient, BHT was considered, but the appearance lacked hypertrophy and elongation of filiform papillae; hence, a diagnosis of extrinsic stain seemed more likely. Education regarding the benign nature of the condition was provided, along with the identification of possible offending agents, which in her case pointed to the excessive coffee intake. A recommendation to reduce daily consumption of coffee and sugar consumption was reinforced, and a solution of toothpaste mixed with pumice for daily brushing of the tongue was provided, to decrease tongue coating [12].

At 6-month follow-up from the onset of the chief complaint, no change in tongue pigmentation was observed despite being compliant with the daily regimen of tongue brushing, although the patient admitted only slightly decreasing her coffee intake to a total of 4 cups per day. She was encouraged to decrease her coffee consumption and consider eliminating other food items from her diet.

At 1-year follow-up, the patient returned to the Orofacial Pain Clinic presenting with a complete resolution of her tongue discoloration (Figure 1B) after discontinuing the consumption of walnuts two weeks before. When she reintroduced the walnuts in her daily diet, the extrinsic stain returned. After a second interruption of walnut consumption, her tongue returned to a normal pink color and she was satisfied with the appearance.

Table 1: Medical history, review of system, medication and allergies of the patient at first visit and at the onset of the new chief complaint.

Evaluation	Time	Medical history	Review of systems	Medication	Allergy
First visit	June 2012	GERD Stomach ulcer Osteoarthritis (knee)	Pulmonary: hay fever Neurological: dizzy spell	glucosamine QD, chondroitin QD, aspirin 81 mg QD, calcium 500 mg BID, multivitamins (Vit B ₁₂ , Vit C, Vit D ₃) QD	Protonix, Biaxin
Onset of new chief complaint	September 2020	GERD Stomach ulcer Osteoarthritis (knee) Supraventricular tachycardia	Pulmonary: snoring, hay fever Neurological: tingling of the tongue	Metoprolol 25 mg QD, multivitamins (Vit B ₁₂ , Vit C, Vit D ₃) QD, clobetasol 0.5% prn, 2% viscous lidocaine 100 mL PRN	Protonix, Biaxin

BID: Two Times Per Day; GERD: Gastro-Esophageal Reflux Disease; QD: Once Per Day; PRN: As Needed.

Table 2: Work-up and laboratory tests performed with correspondent diagnosis and treatment management at first visit and at the onset of the new chief complaint.

Evaluation	Time	Laboratory test	Results	Diagnosis	Treatment
First visit	June 2012	Salivary flow measurement	Negative salivary production with milking parotid and submandibular glands	Hyposalivation	Dry mouth protocol: Biotene® fluoride toothpaste, 6 glasses of water daily, drops of lemon juice with water, sugarless gum and candies, ice chips. Reduce caffeine, tobacco, alcohol, acid food and beverages
		Intraoral fungal swab cultured in Sabouraud dextrose agar	No candida infection detected		
		PCR of 7 consecutive daily salivary sample	HSV-2 DNA not detected; HSV-1 DNA detected	HSV-1 infection	valacyclovir 1g BID for 14 days, and then 0.5 g BID for 14 days
	July 2012	Blood tests with CBC with differential and CIFS 8052 for cutaneous immunofluorescence antibodies serum	WNL; cutaneous autoantibodies not detected		
		Biopsy for H&E and immunofluorescence	Immunological epithelial lesions	Mucous membrane pemphigoid	doxycycline 100 mg BID; clotrimazole 100 mg BID; niacinamide 400 mg TID; clobetasol 0.05% ointment TID; Orogel (Benzocaine 20%) PRN
August 2012	Blood tests with serum level, liver enzymes, urea nitrogen, GFR, creatinine, CBC	WNL			
Onset of new chief complaint	September 2020	Salivary flow measurement	UWS: 0.10 mL/min	Hyposalivation	Reinforcement of dry mouth protocol
		Intraoral fungal swab cultured in Sabouraud dextrose agar	No candida infection detected		

BID: Two Times Per Day; CBC: Complete Blood Count; GFR: Glomerular Filtration Rate; H&E: Hematoxylin And Eosin Stain; PCR: Polymerase Chain Reaction; HSV: Herpes Simplex Virus; PRN: As Needed; TIB: Three Times Per Day; UWS: Unstimulated Whole Salivary Flow; WNL: Within Normal Limit.



Figure 1: A). Black pigmentation of the dorsum of the tongue at its initial appearance; B). Resolution of the extrinsic discoloration within 2 weeks of interruption of walnut consumption.

Discussion

This report describes the successful management of a sudden and long-lasting tongue discoloration in a patient with a positive history of mucous membrane pemphigoid. To the best of our knowledge, this is the first report in the literature that describes the contribution of walnut consumption in the pathogenesis of extrinsic stain of the dorsum of the tongue.

Both extrinsic stain and BHT were considered in the differential diagnosis. The diagnosis of BHT is based on a thorough history, clinical inspection of the tongue, and positive finding of elongated filiform papillae on dermoscopy [10] or on widefield optical fluorescence [13] with no biopsy necessary [14]. Most

of the time it is a self-limiting and asymptomatic condition, although in some patients it can be associated with burning of the tongue, dysgeusia, halitosis and nausea [15]. As evidence of hypertrophic filiform papillae was not observed in this instance, extrinsic stain appeared as the most appropriate diagnosis. A further laboratory investigation was not deemed necessary as discontinuation of the offending factor along with a gentle brushing of the discolored area is advocated as first-line treatment for both conditions, regardless of the precise diagnosis [16].

The work-up that led to the resolution of the chief complaint included the exclusion of all the known factors implicated in the etiology. First of all, fungal infection was ruled out by culture, as few reports have described the chromogen organism *Candida albicans* as a possible predisposing factor [17,18]. Moreover, despite her good oral hygiene, the patient was scheduled on a 3-month oral hygiene recall, as chromogen bacteria have been implicated in the literature, although most frequently on dental surface [19-21]. Smoking habit was not reported by the patient [22,23]. Lastly, the positive history of autoimmune disease of the patient was also considered [24], although a contribution of MMP to the onset of the new chief complaint was unlikely, as the condition was well-controlled for more than 8 years with topical administration, and none of her medications were linked to reports of extrinsic stain [25]. Studies have shown a possible

darkening of the mucosal area due to the deposition of melanin within the connective tissue due to chronic inflammatory conditions, such as MMP [26]. However, the lesions attributed to MMP in the case of our patient were consistently located at the right retromolar buccal mucosa and alveolar ridge, and a shift of location to the dorsum of the tongue was improbable. After excluding all the above contributors, our first approach was to address her complaint of xerostomia and hyposalivation, by reinforcing daily fluid intake, use of coconut oil to oral mucosa before meals and sugar-free gums and candies. At the same time, a reduction in the amount of caffeinated coffee was advocated, as studies have consistently identified dark beverages such as coffee, black tea and red wine, as possible etiologic factors [6,7,9,17,27-30]. As no benefit was seen after reduction of caffeine intake, the patient was encouraged to discontinue her daily use of alcohol-free mouthwash, although the literature has pointed at chlorhexidine containing in these products as the most implicated in extrinsic stain of teeth and tongue [31-33]. However, the patient's daily rinse with alcohol-free mouthwash dated back to 12 years before and discontinuation of her mouthwash did not affect her discolored tongue.

Excessive walnut consumption proved to be the direct causal factor, as disappearance and reappearance of the stain occurred when she stopped and reinitiated the dietary habit. To our knowledge, no study has ever reported extrinsic stain secondary to walnuts. Few reports in the literature indicate food as responsible of tongue discoloration, including sunflower seeds [34], and the consumption of pureed or soft diet in the appearance of BHT [10]. However, the higher predisposition of such food in staining the tongue seems more implicated in the lack of tongue movement with consequent impairment of desquamation of the keratinized filiform papillae [10], rather than the soft food itself. One plausible explanation of walnuts as a precipitating factor for extrinsic stain is the presence of tannins, which are also present in coffee and tea [28]. Tannins only accounts for 0.01-0.88% of walnuts composition, although the proportion can vary according to the specific seed [35]. Discoloration secondary to tannins is suggested to derive from metallic salts precipitating in solution, there by producing a black color [36]. This is in contrast to another study which found that iron needs to be present with tannic acid in order to produce discoloration [37]. A combination between tannins with chromogens from beverage has also been reported, although in most of the cases it affects hard tissue, such as enamel and dentin [36]. As observed in this atypical case presentation, clinicians should familiarize with this unusual side-effect of walnuts, or the combined effects of eating walnuts with coffee as a potential cause of mucosal stain, especially when all the other etiological factors have been excluded.

Conclusions

This case report identified walnut consumption as a factor influencing the appearance of extrinsic stain of the tongue. Clinicians should be aware that walnut consumption may be implicated in the pathogenesis of this benign condition.

Declarations

Declarations of interest: none.

Practical implications: Clinicians should be aware that walnut consumption may be implicated in the pathogenesis of extrinsic stain on the tongue.

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