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Clinical case

Black hairy tongue associated with squamous cell carcinoma of the esophagus

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Abstract

Black hairy tongue (BHT) is a benign disorder characterized by dark coloration and hypertrophy with hyperkeratosis in filiform papillae on the surface of the tongue. Several intrinsic and extrinsic factors, including poor dental hygiene, smoking, drinking dark beverages, indiscriminate use of antibiotics, and some malignant diseases, have been proposed as potential causes. This is the case of a patient with advanced squamous cell carcinoma of the mid esophagus in conjunction with BHT, a previously unknown association.

Keywords

Black hairy tongue; Black tongue; Hyperpigmentation of the tongue; Lingual pigmentation.

INTRODUCTION

Black hairy tongue (BHT) is a benign condition in which the center of the tongue changes its color, from black to yellow, and has a hairy appearance due to hypertrophy and elongation of the filiform papillae (1). It has a variable prevalence and its occurrence is associated with multiple factors such as poor oral hygiene, tobacco use, antibiotic use (penicillin, erythromycin, doxycycline [2], linezolid [3], tetracycline [4]), and irritating mouthwashes (5). An association between BHT and systemic diseases such as HIV and malignancies has been described (6). We present the case of an elderly woman with squamous cell carcinoma of the middle esophagus and in whom BHT was an associated, incidental, unaesthetic, and asymptomatic finding.

PRESENTATION OF THE CLINICAL CASE

This is the case of a 78 years old woman born in Ubalá, Cundinamarca and living in Bogotá, with a history of hypothyroidism (treated with levothyroxine) and arterial hypertension (treated with enalapril at the time of consultation). The patient attended the digestive endoscopy service due to having experienced the following signs and symptoms for 1 month: dysphagia with retrosternal choking sensation and a 5 kg weight loss. The patient reported having black tongue for 7 years, a condition that was ignored in several consultations.

On physical examination she was in an acceptable general condition; vital signs: blood pressure (BP): 145/85 mm Hg; heart rate (HR): 76/min; respiratory rate (RR): 18/min;

oxygen saturation (SaO2): 92%; fraction of inspired oxygen (FiO_2): 24%; weight: 40 kg; height: 1.5 m; body mass index (BMI): 17.78 kg/m^2; and body temperature: 36°C.

During endoscopy, the tongue was found to have a dark blackish color on its dorsum, with a hairy appearance, without involvement of the borders or the tip, with a whitish depigmentation area in the central posterior part (**Figures 1** and **2**). In the middle esophagus (25 cm), on the right, left and anterior lateral walls, there is a nodular, infiltrative and friable lesion causing a 12 mm stenosis that can be crossed and reaches up to 30 cm, without distal esophageal involvement (**Figures 3** and **4**).

A non-keratinizing, infiltrating, poorly differentiated and ulcerated large squamous cell carcinoma was described in the pathology report.

At the time this case report was written, the patient is undergoing further testing to determine the treatment and management approach of the BHT syndrome by the odontology service.

DISCUSSION

Black hairy tongue (BHT) is a benign condition consisting of the darkening and hairy appearance of the dorsum of the

Figure 1. Patient in which the dorsum of the tongue is black, without involvement of the lateral borders of the tip.

Figure 2. Patient with black hairy tongue (BHT) with whitish depigmentation in the central posterior part.

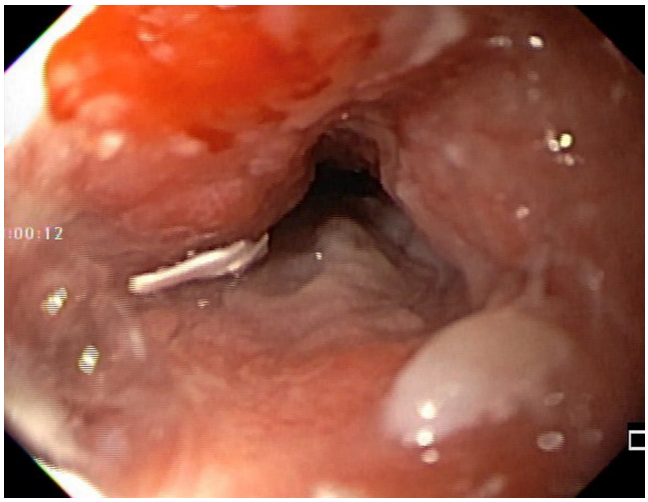

Figure 3. Endoscopy. Proximal border of squamous cell carcinoma in the middle part of the esophagus involving 80% of the wall and lumen.

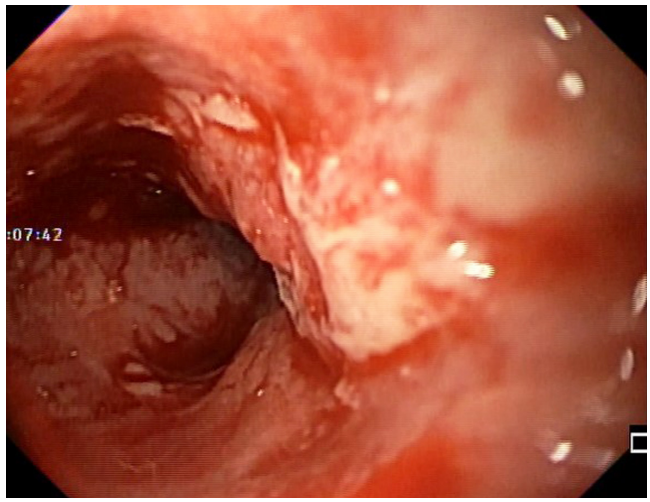

Figure 4. Endoscopy. Distal border of the carcinoma affecting the middle part of the esophagus.

tongue. It was first described by Amatus Lusitanus in 1597 (7, 8). Its prevalence is variable, although a prevalence as high as 11.3% has been described in some oral health studies, being more common in elderly men, heavy smokers, and black tea and coffee consumers (9, 10), HIV positive people, edentulous individuals, people with prostate cancer or B-cell lymphoma (6), and in patients undergoing antibiotic treatment (2-4). Clinically, a black plaque or membrane is observed on the dorsum of the tongue, without lateral borders or tip involvement (**Figure 1**) and, as in this case, without involvement of the central and posterior part of the tongue (**Figure 2**). Black is the most common color change, but can vary to brown, green or yellow (11).

BHT is usually asymptomatic, although symptoms such as nausea, loss of taste, halitosis, burning or tingling of the tongue have been described (12); however, the main annoyance for patients is its unpleasant esthetic appearance (13).

Hairy appearance is due to inadequate keratin desquamation (hyperkeratosis) over the filiform papillae, which elongate up to 18 mm × 2 mm. On the other hand, color change is caused by yeasts or anaerobic (such as *Porphyromonas gingivalis*) (14), chromogenic, amino acid-fermenting, and porphyrin-producing bacteria, and modifications resulting from environmental factors (tobacco, alcohol, oxidizing mouthwashes, antibiotics, antipsychotics, proton pump blockers, chemotherapeutic agents, radiotherapy, anti-HIV drugs, and xerostomia predisposing drugs) (7), which can be associated with alterations of the oral microbiome present in HIV infection, graft-versus-host disease, amyotrophic lateral sclerosis, trigeminal neuralgia or malignancy (11).

Differential diagnoses include "pseudo-BHT" conditions such as acanthosis nigricans, congenital lingual melanotic macules, congenital melanocytic nevi, premalignant leukoplakia, squamous cell carcinoma and hypertrophic infection by herpes simplex virus (15).

Clinical diagnosis is made by asking the patient in detail about the presence of the abovementioned predisposing factors. Gentle scraping of the tongue with a tongue depressor or a toothbrush may suggest BHT if the pigment dissipates; however, this procedure is not always easy, as it may require multiple attempts (16). A biopsy is usually not necessary, except when the lesion becomes atypical, refractory to treatment or symptomatic, thus suggesting a potential malignancy or systemic disease (17); however, when biopsy is performed, hyperkeratosis and accumulation of parakeratosis at the tip of the filiform papillae are reported (18). Dermoscopy is the diagnostic tool of choice; it allows the identification of shape and color changes of the filiform papillae; it is also useful in the evaluation of therapeutic success (18, 19).

The first line of treatment is based on suspending the medications associated with the condition, having excellent oral hygiene, quitting predisposing habits, and the generous brushing or scraping of the tongue to promote the desquamation of the papillae (17). Topical use of 3% hydrogen peroxide, suspension of oxidizing mouthwashes; fitting of dentures in edentulous patients (which promotes mechanical abrasion of the desquamated papillae when chewing solid food) have shown to have a good efficacy (20). In BHT resistant cases, papillae can be removed by burning-off or electrodesiccation with carbon dioxide laser (21). Second-line treatments, which are anecdotal and have no supporting evidence, include oral retinoids, antifungal agents, antibiotics, topical urea solution or triamcinolone, salicylic acid and gentian violet.

No cases of BHT and esophageal cancer association were found in the literature review conducted by us, so this is an association that should be taken into account.

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