

A Unique Tongue Presentation Revealing a Bronchobiliary Fistula

Une présentation unique de la langue révélant une fistule bronchobiliaire.

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A 45-year-old chronic smoker (20 pack-years) with a history of two previous resections for a left colonic tumor and tumorectomy of liver metastases presented three months after the last operation with respiratory distress, fever and biliopytosis. At the clinical examination, the tongue takes on a dark yellow colour (Figure A). The magnetic resonance imaging (MRI) revealed the presence of a bronchobiliary fistula (BBF) (Figure B). BBF represents an abnormal communication between the bile ducts and the bronchial tree (1). The etiologies are diverse: congenital, malignant, abscessed, traumatic and iatrogenic causes (1). Diagnosis is typically based on clinical presentation, and is confirmed through imaging modalities such as computed tomography (CT) or MRI. MRI, with its superior soft tissue contrast, is the preferred imaging modality for BBF (2). It allows direct visualization of the fistula and any associated biliary or hepatic pathology. CT with contrast may serve as an alternative when MRI is unavailable, providing insights into associated pulmonary complications and offering faster acquisition times in urgent clinical situations (2).

Initially, the patient underwent scan-guided percutaneous drainage and broad-spectrum antibiotic therapy, resulting in partial clinical improvement with resolution of fever but persistent biliopytosis. The progression of the underlying tumor complicated the patient's prognosis, and made further surgical intervention difficult. It was therefore decided to pursue an endoscopic treatment with endoscopic retrograde cholangiopancreatography (ERCP). ERCP revealed a BBF between the bile ducts and the bronchial tree, and an endoscopic biliary

sphincterotomy was performed to relieve pressure on the biliary tree, facilitate bile drainage into the duodenum, and promote fistula closure. Two months later, the patient demonstrated significant improvement with no recurrence of biliopytosis. Endoscopic techniques such as ERCP and percutaneous transhepatic cholangiography are crucial for the diagnosis and initial management of BBF (1). ERCP facilitates management of the bile flow by stenting and sphincterotomy, with efficacy influenced by the complexity of the fistula (3). Surgical intervention remains pivotal for complex scenarios or treatment failure. Treatment selection considers fistula characteristics, underlying causes, patient health status, and clinical expertise (3). Non-surgical approaches are preferred in contexts of poor oncological prognosis and advancing tumor conditions.

The appearance of the patient's tongue described in our observation has not been previously illustrated in the literature. The clinical presentation may suggest other differential diagnoses, such as pneumonia (fever with yellow sputum). However, the authors believe this distinct tongue presentation provided a unique clinical sign and may aid in suspecting a BBF during the clinical examination of patients with a compatible history. The exact mechanisms behind the dark yellow discoloration of the tongue in BBF are not fully understood. This discoloration may be due to the migration of bile pigments such as bilirubin across the BBF, with some bile reaching the mouth during biliopytosis and causing visible pigmentation on the tongue. While direct contact with bile can stain tissues yellow, this usually fades quickly with

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normal oral hygiene practices. One hypothesis suggests a mechanism that causes a localised high concentration of bilirubin in the mouth, implying continuous or repeated exposure. In our case, the frequency and abundance of biliptysis, along with poor oral hygiene or a tendency to retain bile in the oral cavity, may contribute to this presentation. Another mechanism, though less likely, could be related to the specific anatomy of the fistula in our patient. Further investigation and additional case observations are needed to fully understand this phenomenon, to potentially validate this sign as a specific symptom of BBF and to elucidate the mechanisms behind this coloration.

Consent of the patient: Written informed consent was obtained from the patient for the publication of this case report and the use of clinical images.

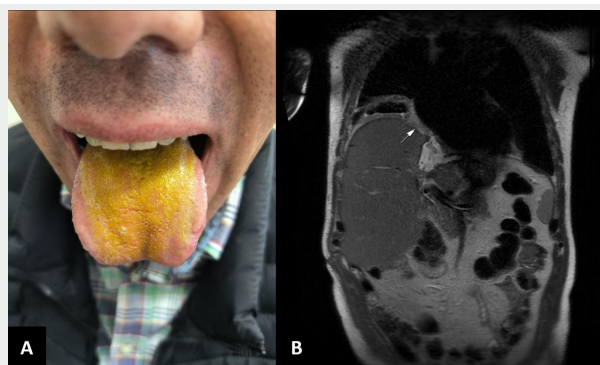


Figure 1. A) At the clinical examination, the tongue takes on a dark yellow colour. B) Magnetic resonance imaging revealed a bronchobiliary fistula (arrow)

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