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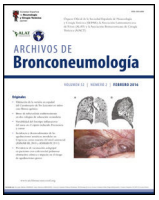

Scientific Letter

Multimodality Imaging With Confocal Laser Endomicroscopy, Red Dichromatic Imaging, and Virtual Bronchoscopic Navigation Guided Recanalization of Complete Airway Obstruction Caused by Tracheobronchial Amyloidosis

To the Director,

We report the case of a 58-year-old woman diagnosed with tracheobronchial amyloidosis (TBA) 4 years ago, based on tissue biopsy with positive staining for Congo red. She had been treated with multiple bronchoscopic interventions about every 6–9 months due to recurrent diffuse stenosis of the left main bronchus (LMB), but had persistent symptoms and frequent exacerbations. A chest X-ray showed atelectasis of the left lung (Fig. 1A), which was complemented with a chest computed tomography (CT) that demonstrated irregular thickening of the wall of the LMB with extensive calcifications and severe stenosis of the LMB (Fig. 1B). Bronchoscopy performed at that time revealed roughness, micronodular and friable appearance of tracheal mucosa, and complete obstruction of the LMB (Fig. 2A). Bronchoscopic probe-based confocal laser endomicroscopy (pCLE) (*Cellvizio® Lung*, *Mauna Kea Technologies*, *France*) examination showed a patchy "cotton wool"-like appearance (Fig. 2B), consistent with TBA, which was further verified by pathological finding with Congo red staining that produced apple-green birefringence under polarized light (Fig. 2C).

We empirically conjectured the site of LMB ostium by evaluating the indentation and puckering of LMB mucosa (Fig. 2D), however which was proven inaccurate after the unsuccessful recanalization and confirmation via virtual bronchoscopic navigation (VBN, DirectPath V2.0, Cybernet Systems; Olympus, Japan) (Fig. 2E). Thus, the patient was re-scheduled for bronchoscopic recanalization of the LMB using electrocautery and balloon dilatation on the basis of the proposed LMB ostium by VBN. During the procedure, active bleeding was found in the LMB. Under the white-light imaging (WLI), it is difficult to maintain clear bronchoscopic visibility and accurately located the bleeding points (Fig. 2F), which was improved by using EVIS X1 system-based red dichromatic imaging (RDI) (*CV-1500; Olympus, Japan*), which provided clear bronchoscopic visibility and could facilitate endoscopic hemostasis (Fig. 2G). Significant improvement in the patient's condition was observed after the procedure. Repeat chest X-ray revealed recruitment of the left lung (Fig. 1C).

TBA, as a rare disorder with a proportion of 0.5% of all symptomatic tracheobronchial lesions, is characterized by amyloid deposits limited specifically to the submucosa of the respiratory tract, which could be definitively diagnosed by pathological finding with Congo red staining that produced apple-green birefringence under polarized light.[1] To note, pCLE with a combination of high-resolution and real-time microscopic analysis of tissue architecture, as a novel "optical biopsy" technique, shows a patchy "cotton wool"-like appearance when employing over areas of TBA, which appears to be a promising tool for patients with TBA.[2]

There are no standardized guidelines regarding the treatment of TBA. Bronchoscopic interventions have been demonstrated to be effective in restoring airway patency, and thus serve as the most common approach in TBA associated airway stenosis.[3] However, for patients who present with complete airway obstruction with no visible ostium, performing the bronchoscopic recanalization remains challenging. As reported in our case, postulating empirically the invisible ostium by evaluating the indentation and puckering of LMB mucosa may be deceiving, even performed by an experienced bronchoscopist. VBN, as a noval technique of improving the diagnostic yield for peripheral pulmonary lesions,[4] can

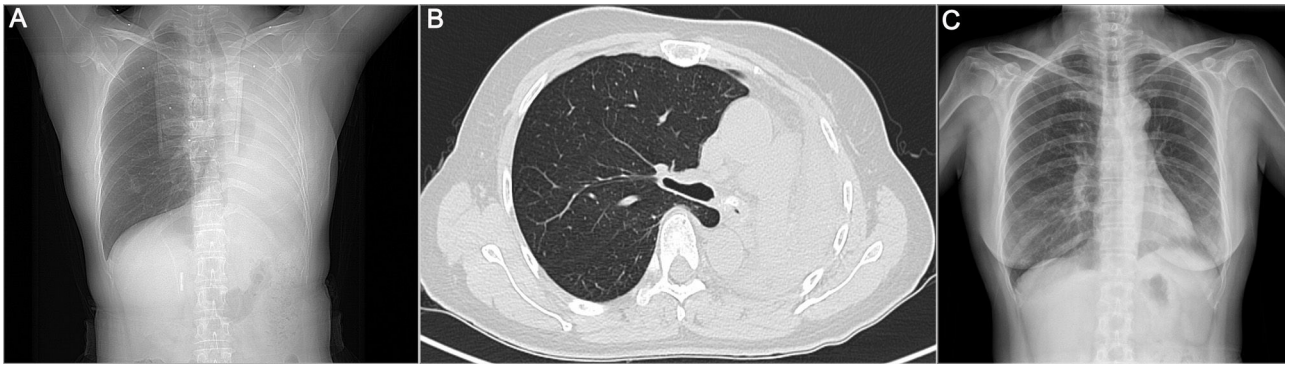

Fig. 1. (A) Chest X-ray shows atelectasis of the left lung. (B) CT scan of chest performed on admission reveals irregular thickening of the wall of LMB with extensive calcifications and severe stenosis of the LMB. (C) Chest X-ray 1 day post-operative shows recruitment of the left lung.

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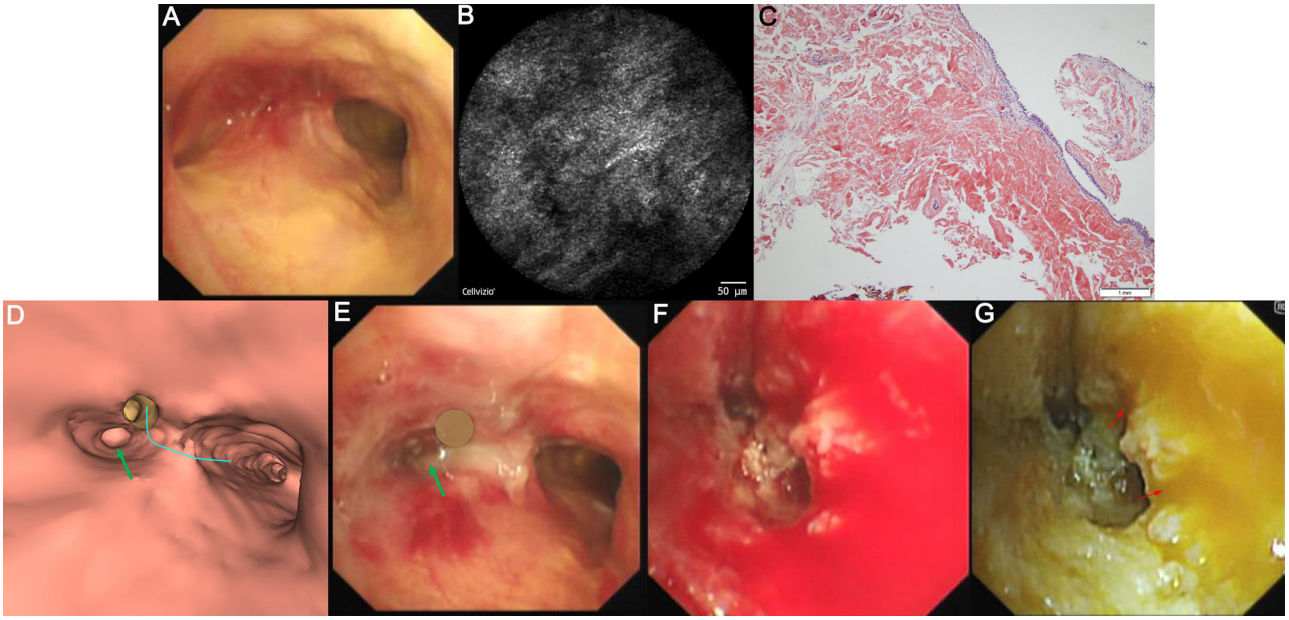

Fig. 2. (A) Bronchoscopic visualization shows roughness, micronodular and friable appearance of tracheal mucosa. (B) Bronchoscopic pCLE imaging demonstrates a patchy "cotton wool"-like appearance. (C) Viewing this tissue with polarized light after Congo red staining demonstrates apple-green birefringence. Correlation of bronchoscopic (D) and virtual bronchoscopic views (E) reveals the speculative site of LMB ostium based on experience (green arrow) and the actual site by VBN (yellow dot). (F) Fresh blood is observed in the LMB during bronchoscopic recanalization. (G) The bleeding point is observed as deep yellow (red arrow) compared to surrounding blood, and that allows us to recognize it precisely after switching to RDI mode.

also plan and localize the site of recanalization ostium with high confidence level, which has the potential to be added to the armamentarium of bronchoscopists who deal with this century-old problem.

Intraoperative bleeding is unavoidable during the bronchoscopic recanalization due to the friable nature of the amyloid affected mucosa. However, under the conventional WLI mode, it is usually difficult to maintain clear visibility and rapidly identify the bleeding points because of the absence of contrast between the blood and bronchial mucosa. Hopefully, RDI, installed in the latest endoscopy system, EVIS X1, has been to resolve the clinical dilemma. Where all the previous studies of RDI are confined to gastrointestinal diseases,[5] we examined for the first time the utility and effectiveness of RDI in the case of a patient with TBA. In the present case, the addition of RDI enabling distinct visualization of the bleeding points, improves the endoscopic visibility, determines the ongoing bleeding or cessation, and reduces the mental stress, thereby reducing the risk of bleeding during procedure.

In conclusion, multimodality imaging should be considered as a safe and novel approach in this patient population.

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Conflict of Interests

The authors state that they have no conflict of interests.

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Sen Tian [a,b,1], Yifei Zhang [a,1], Shunmin Zhang [c], Chong Bai [a,*], Haidong Huang [a,*]

[a] *Department of Respiratory and Critical Care Medicine, The First Affiliated Hospital of Naval Medical University, Shanghai, China*
[b] *Department of Respiratory and Critical Care Medicine, No. 906 Hospital of the Chinese People's Liberation Army Joint Logistic Support Force, Ningbo, China*
[c] *Department of Pathology, The First Affiliated Hospital of Naval Medical University, Shanghai, China*

*Corresponding authors.
E-mail addresses: Chongbai@smmu.edu.cn (C. Bai), hhdongbs@126.com (H. Huang).
[1] These authors have contributed equally to this work and share corresponding authorship.