

Coexistence of localized tracheobronchial amyloidosis and chronic hepatitis B infection

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A 44-year-old man presented to our department with a 3-year history of dyspnea and cough. He was an ex-smoker with a cigarette smoking history of 35 pack-years. On initial examination, his vital signs were normal. Chest auscultation revealed normal breath sounds. Physical examination of other organ systems was not remarkable.

Thoracic computed tomography showed diffuse nodular thickening and calcification of the submucosal tracheal and the right upper lobe bronchial walls (FIGURE 1A and 1B). Bronchoscopy revealed completely irregular surface of tracheal and right upper lobe bronchial mucosa (FIGURE 1C). Bronchial alveolar lavage samples were negative for bacteria, acid-fast bacilli, and fungi. The tracheal biopsy specimens showed amorphous deposits, which were identified as amyloid by apple-green birefringence in sections stained with Congo red, under polarized light (FIGURE 1D). According to immunohistochemical amyloid protein identification, the amyloidosis type was serum amyloid A. Further etiological workup of serum amyloid A amyloidosis revealed negative test results for rheumatological markers. Proteinuria was not

detected in 24-hour urine collection, and cardiac evaluation was normal. Pulmonary function tests revealed the following findings: forced vital capacity (FVC) of 97% of predicted value, forced expiratory volume in 1 second (FEV₁) of 73% of predicted value, and a FEV₁/FVC ratio of 0.62. Liver function tests were abnormal: total bilirubin, 2.1 mg/dl (reference range, 2.8–5.8 mg/dl); direct bilirubin, 0.53 mg/dl (0–0.5 mg/dl), serum aspartate aminotransferase, 67 IU/l (11–25 IU/l); and serum alanine transaminase, 104 IU/l (7–28 IU/l). Viral studies showed chronic hepatitis B infection and a hepatitis B virus DNA titer of 31 130 922 IU/ml. Liver biopsy confirmed chronic hepatitis B infection, and amyloid-stained liver and rectal biopsies were negative. Tenofovir treatment (245 mg/d) was started.

Amyloidosis can be systemic or confined to a single organ.¹ Tissue biopsy is required for the diagnosis of amyloidosis. Congo red stain used with polarized light is the gold standard test.² Amyloidosis can appear in the lungs in 3 different clinicopathological forms: nodular pulmonary amyloidosis; diffuse, alveolar-septal amyloidosis;

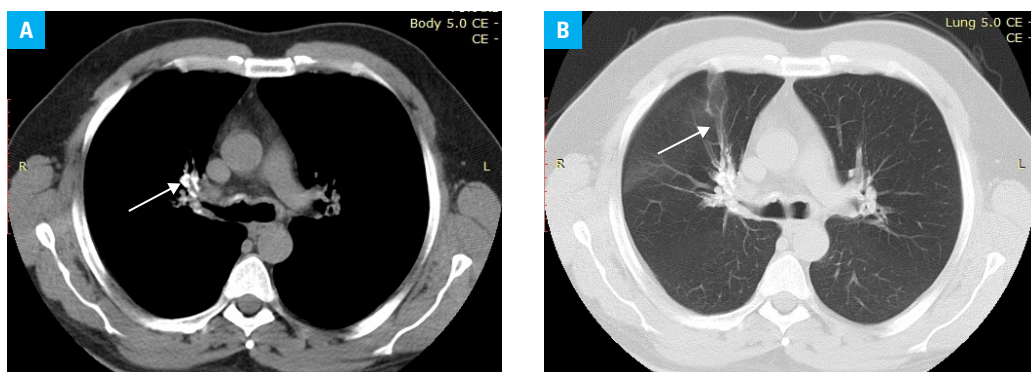


FIGURE 1 A, B – thoracic computed tomography showing diffuse nodular thickening with calcification of submucosal tracheal and right upper lobe bronchial walls and segmental atelectasis due to bronchial stenoses (arrows)

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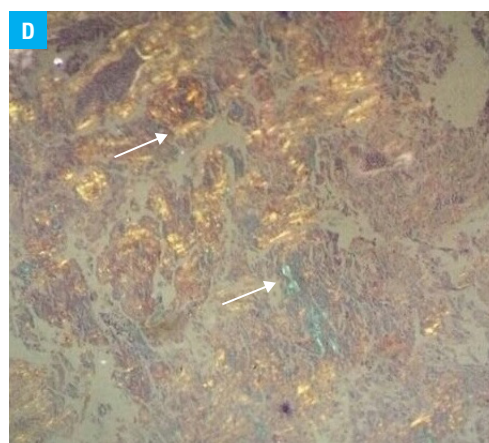
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FIGURE 1

C – bronchoscopy showing diffuse whitish deposits (arrows) with completely irregular surface of tracheal and right upper lobe bronchial mucosa;
D – a histopathological section stained with Congo red and viewed under polarized light, showing apple-green birefringence (arrows)



and tracheobronchial amyloidosis (TBA).³ Tracheobronchial amyloidosis is a rare organ-limited form of localized amyloidosis, which most often presents as multifocal submucosal plaques. The pulmonary parenchyma is characteristically not involved.⁴ A computed tomography scan may reveal tracheal and bronchial wall thickening with possible calcification, which is usually located in the posterior tracheal wall. Atelectasis due to bronchial stenoses may be found.⁵ Tracheobronchial endoscopy usually shows irregular, diffuse whitish deposits or submucosal plaques.

The management of TBA is largely dependent on symptoms, and there is no proven drug therapy. Endoluminal narrowing is usually treated with endoscopic therapies such as debridement or external beam radiation.

To our knowledge, we report a first case of localized TBA with chronic hepatitis B infection. Viral infections have been infrequently reported with amyloidosis. Literature review has revealed a case of hepatitis C virus-associated glomerulonephritis and amyloid light-chain amyloidosis, a case of Still disease complicated with amyloid A amyloidosis and hepatitis B, and a case of common variable immunodeficiency with hepatitis C and systemic amyloidosis.

Tracheobronchial amyloidosis should be considered in cases with unexplained chronic cough and persistent dyspnea, and bronchoscopic biopsy should be employed for definitive diagnosis.

ARTICLE INFORMATION

CONFLICT OF INTEREST None declared.

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