

with acute symptoms of a concomitant a respiratory infection and lung abscess might be presented with chronic symptoms of weight loss and anorexia. There are, however, some radiologic findings that can help: rim enhancement of the walls on contrast-enhanced CT scan suggests abscess and satellite nodules might occur in infectious etiologies.⁵

Traditionally, squamous cell carcinoma was the most frequent histology found in cavitated tumors; however, an increasing number of adenocarcinomas have been found to form cavitations.⁶ Despite cavitation of primary lung cancer being relatively common, cavitation of lung metastases is infrequent. Only 4% of lung metastases cavitate,⁷ which makes this case a rare presentation of stage IV lung adenocarcinoma.

The differential diagnosis of a cavitary lung disease is challenging. Radiologic findings may help distinguish benign from malign disease, but a high suspicion of neoplasm is essential for preventing misdiagnosis.

References

1. Hansell DM, Bankier AA, MacMahon H, McLoud TC, Müller NL, Remy J. Fleischner Society: glossary of terms for thoracic imaging. *Radiology*. 2008;246:672-97.

2. Woodring JH. Pitfalls in the radiologic diagnosis of lung cancer. *AJR Am J Roentgenol*. 1990;154:1165-75.
3. Gadkowski LB, Stout JE. Cavitary pulmonary disease. *Clin Microbiol Rev*. 2008;21:305-33.
4. Deng H, Zhang J, Xue X. Thin-wall cyst lung cancer: a study of 45 cases. *Oncol Lett*. 2018;16:755-60.
5. Walker CM, Abbott GF, Greene RE, Shepard JO, Vummidi D, Digumarthy SR. Imaging pulmonary infection: classic signs and patterns. *AJR Am J Roentgenol*. 2014;202:479-92.
6. Chu X, Guo J, Liang C. Lung cancer presenting as thin-walled cysts: an analysis of 15 cases and review of literature. *Asia-Pacific J Clin Oncol*. 2006;12:e105-12.
7. Boitsios G, Bankier AA, Eisenberg RL. Diffuse pulmonary nodules. *AJR Am J Roentgenol*. 2010;194:W354-66.

Luís Carreto,^{a,*} Carlos Alves,^a João Eurico,^b Paulo Cavinho^b

^a *Pneumology Department, Hospital Professor Doutor Fernando Fonseca E.P.E, Portugal*

^b *Cardiothoracic Surgery Department, Hospital Santa Marta, Centro Hospitalar Lisboa Central E.P.E, Portugal*

* Corresponding author.

E-mail address: luis.carreto@hff.min-saude.pt (L. Carreto).

1579-2129/

© 2019 SEPAR. Published by Elsevier España, S.L.U. All rights reserved.

IGG4-Related Pleural Bridging Fibrosis, Retrosternal Fibrosclerosis and Retroperitoneal Fibrosis in an Asbestos-Exposed Patient



Fibrosis pleural en puente, fibrosclerosis retroesternal y fibrosis retroperitoneal relacionadas con IGG4 en un paciente expuesto a asbesto

Dear Editor:

We report the case of a 78-year-old man with a previous history of asbestos exposure for 13 years, who presented with dyspnea. The patient packed asbestos-containing waste materials until the age of 50. Thus, the latency period was 28 years. Chest computed tomography showed an irregular, uncalcified pleural plaque in the anterior right hemithorax. One year later, the plaque had flattened, extending axially to the retrosternal mediastinum and contralateral pleural space (Fig. 1A and B). A small soft-tissue mass surrounding the abdominal infrarenal aorta was also identified (Fig. 1C). Transthoracic percutaneous biopsy revealed storiform fibrosis with polyclonal lymphoplasmocytic and plasma cell infiltration. Additionally, immunoglobulin G4 (IgG4) expression was found in plasma cells, with an IgG4/IgG ratio >50% (Fig. 1D and E). The serial IgG4 concentration was found to be high (5920 mg/L), and a final diagnosis of IgG4-related bridging pleural fibrosis (BPF) with retrosternal fibrosclerosis (RF) was established.

BPF refers to continuous bilateral anterior pleural thickening that “bridges” the hemithoraces and involves the retrosternal area, resulting in RF.^{1,2} BPF and RF have been described in association with retroperitoneal fibrosis, and in the patient described here a subtle retroperitoneal soft-tissue mass surrounding the infrarenal aorta was also observed. These patterns have been described in asbestos-exposed patients and in those with immunoglobulin G4-related sclerosing disease (IgG4SD).^{1,4}

Asbestos-related pleural fibrosis usually appears as bilateral, partially calcified plaques involving the parietal pleura, and is quite different from the pleural thickening observed in our patient.^{1,5}

Uibu et al.⁴ studied pleuro-pulmonary manifestations in a series of 48 asbestos-exposed patients with RF and found atypical pleural plaques extending to the anterior mediastinum in three patients, recognizing such plaques as rare findings and speculating that asbestos may lead to severe fibrotic changes in some susceptible individuals.

Imaging findings for mediastinal fibrosis were also atypical in this case; the characteristic features of this entity are an often-calcified invasive mass in the middle mediastinal compartment, encasing vascular and bronchial structures. In the case described here, we observed a different pattern of isolated, uncalcified retrosternal fibrosis.¹

IgG4SD is a multisystem clinical-pathological entity described in the last decade, characterized by fibro-inflammatory lesions rich in IgG4-positive plasma cells.⁶ Thoracic involvement may occur in several patterns, comprising pulmonary lesions (nodules, ground-glass opacities, alveolar-interstitial disease and bronchovascular disease), pleural thickening and mediastinal lesions (mediastinal fibrosis and lymphadenopathy).^{6,7} Two sets of diagnostic criteria have been proposed: the Japanese criteria, which involve clinical, serological IgG4 levels and histopathological features of the disease, and the Boston criteria, which are histopathological.^{8,9} Recently, the Japanese team updated and proposed an organ-specific approach.¹⁰

Some authors have speculated about a possible association between asbestos exposure and IgG4SD, arguing that the latter may arise due to hypersensitivity of the regulatory immune system to atopic conditions induced by asbestos.^{3,11} IgG4SD shares some characteristics with allergic disorders, such as elevated T helper 2 (Th2) cell, regulatory cytokine and serum immunoglobulin E (IgE) levels; additionally, mast cells in IgG4SD show strong positivity for IgE.¹² Thus, IgG4SD has been considered to be a potentially allergic condition, and asbestos could be the trigger inducing persistent Th2 responses and mast cell activation by sustained IgE production.¹² The literature contains some case reports and case series describing IgG4SD in asbestos-exposed patients.^{3,11,13} In conclusion, we here describe a clinicopathologically proven case of IgG4SD-related

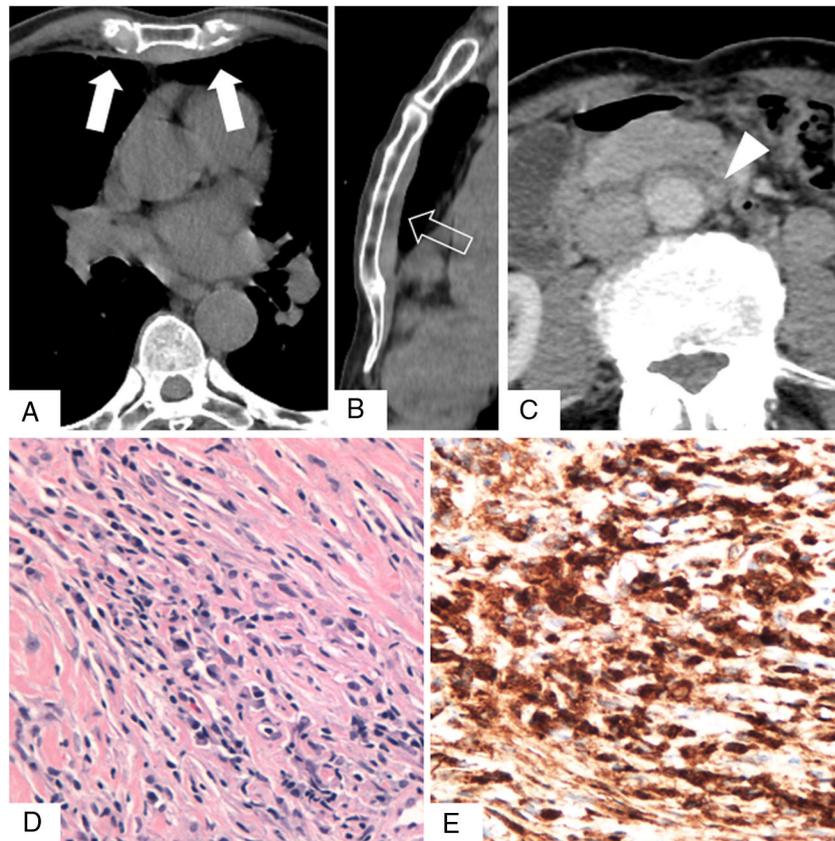


Fig. 1. Unenhanced chest computed tomography scans obtained in the axial plane (A) and with sagittal reformatting (B), showing a flat, uncalcified anterior pleural plaque “bridging” the hemithoraces (arrows in A, open arrow in B). (C) Axial contrast-enhanced abdominal scan showing the periaortic soft tissue mass (arrowhead). (D) Photomicrograph of the pleural plaque showing storiform fibrosis and lymphoplasmacytic infiltrate (hematoxylin-eosin stain). (E) Immunohistochemical staining showing immunoglobulin G4 positivity in plasma cells.

BPF, RF and retroperitoneal fibrosis in an occupationally asbestos-exposed patient, highlighting, as previously suggested, a possible association between these conditions.

References

- Cottin V, Brillet P-Y, Combarrous F, Duperron F, Nunes H, Cordier J-F. Syndrome of pleural and retrosternal “bridging” fibrosis and retroperitoneal fibrosis in patients with asbestos exposure. *Thorax*. 2008;63:177–9.
- Debray M-P, Marchand-Adam S, Soufan R, Ostinelli J, Brillet P-Y, Claeys-Schouman E, et al. Retrosternal and pleural “bridging” fibrosis mimicking malignancy in a patient exposed to asbestos. *Eur Respir Rev*. 2010;19:158–60.
- Kamiya K, Yoshizu A, Nkazato T, Hayashi H, Hida N, Okamoto H. High serum immunoglobulin G4-related retrosternal fibrosclerosis. *J Thorac Imaging*. 2012;27:W190–2.
- Uibu T, Jarvenpaa R, Hakomaki J, Auvinen A, Honkanen E, Metsärinne K, et al. Asbestos-related pleural and lung fibrosis in patients with retroperitoneal fibrosis. *Orphanet J Rare Dis*. 2008;3:29.
- Ryu JH, Yi ES. Immunoglobulin G4-related disease and the lung. *Clin Chest Med*. 2016;37:569–78.
- Alegria AMA, Baleato-González S, García-Figueiras R, Bermúdez-Naveira A, Abdulkader-Nallib I, Díaz-Peromingo JA, et al. IgG4-related disease from head to toe. *Radiographics*. 2015;35:2007–25.
- Zen Y, Inoue D, Kitao A, Onodera M, Abo H, Miyayama S, et al. IgG4-related lung and pleural disease: a clinicopathologic study of 21 cases. *Am J Surg Pathol*. 2009;33:1886–93.
- Umehara H, Okazaki K, Masaki Y, Kawano M, Yamamoto M, Saeki T, et al. Comprehensive diagnostic criteria for IgG4-related disease (IgG4-RD), 2011. *Mod Rheumatol*. 2012;22:21–30.
- Deshpande V, Zen Y, Chan JKC, Yi EE, Sato Y, Yoshino T, et al. Consensus statement on the pathology of IgG4-related disease. *Mod Pathol*. 2012;25:1181–92.
- Umehara H, Okazaki K, Nakamura T, Sathoh-Nakamura T, Nakajima A, Kawano M, et al. Current approach to the diagnosis of IgG4-related disease • combination of comprehensive diagnostic and organ-specific criteria. *Mod Rheumatol*. 2017;27:381–91.
- Toyoshima M, Chida K, Kono M, Kaida Y, Nakamura Y, Suda T, et al. IgG4-related lung disease in a worker occupationally exposed to asbestos. *Intern Med*. 2010;49:1175–8.
- Onishi Y, Nakahara Y, Hirano K, Sasaki S, Kawamura T, Mochiduki Y. IgG4-related disease in asbestos-related pleural disease. *Respirol Case Rep*. 2016;4:22–4.
- Gajewska ME, Rychwicka-Kielek BA, Sorensen K, Kubik M, Hilberg O, Bendstrup E. Immunoglobulin G4-related pleuritis • a case report. *Respir Med Case Rep*. 2016;19:18–20.

Pedro Paulo Teixeira e Silva Torres,^a
 Lorena Barbosa de Moraes Fonseca,^b Kim-Ir-Sen Santos Teixeira,^c
 Marcelo Fouad Rabahi,^{b,c} Edson Marchiori^{d,*}

^a *Multimagem Diagnósticos, Goiânia, Brazil*

^b *Hospital Geral de Goiânia, Goiânia, Brazil*

^c *Universidade Federal de Goiás, Goiânia, Brazil*

^d *Universidade Federal do Rio de Janeiro, Rio de Janeiro, Brazil*

* Corresponding author.

E-mail address: edmarchiori@gmail.com (E. Marchiori).

1579-2129/

© 2019 SEPAR. Published by Elsevier España, S.L.U. All rights reserved.