References

1. Franquet T, Rodríguez S, Hernández JM, Martino R, Giménez A, Hidalgo A, et al. Air-leak syndromes in hematopoietic stem cell transplant recipients with chronic GVHD: high-resolution CT findings. J Thorac Imaging. 2007;22:335-40.
2. Ratanatharathorn V, Ayash L, Lazarus HM, Fu J, Uberti JP. Chronic graft-versus-host disease: clinical manifestation and therapy. Bone Marrow Transplant. 2001;28: 121-9.
3. Macia I, Moya J, Ramos R, Morera R, Escobar I, Saumench J, et al. Spontaneous pneumomediastinum: 41 cases. Eur J Cardiothorac Surg. 2007;31:1110-4.
4. Padley SP, Adler BD, Hansell DM, Muller NL. Bronchiolitis obliterans: high resolution CT findings and correlation with pulmonary function tests. Clin Radiol. 1993;47:236-40.
5. Suzuki T, Saijo Y, Ebina M, Yaekashiwa M, Minegishi M, Tsuchiya S, et al. Bilateral pneumothoraces with multiple bullae in a patient with asymptomatic bronchiolitis obliterans 10 years after bone marrow transplantation. Bone Marrow Transplant. 1999;23:829-31.
6. Wintermark M, Schnyder P. The Macklin effect: a frequent etiology for pneumomediastinum in severe blunt chest trauma. Chest. 2001;120:543-7.

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Kikuchi-Fujimoto Disease in an Atypical Site

Enfermedad de Kikuchi-Fujimoto: una localización atípica

To the Editor:

Kikuchi-Fujimoto disease or histiocytic necrotizing lymphadenitis is a rare entity that is generally associated with cervical lymphadenopathy and fever. We report a case of this disease in an atypical location, the anterior mediastinum, associated with multiple pulmonary nodules.

A 27-year-old woman was evaluated in the internal medicine department for a 3-month history of fever, mainly in the evening, night sweats, and nonpruriginous maculopapular lesions. Physical examination was normal, with no evidence of cervical or supraclavicular adenopathy. Laboratory tests showed no abnormalities. Agglutination tests for *Typhi* and *Brucella* were negative, as were antibody titers against Epstein-Barr virus, rheumatic and immunological tests, and the Mantoux test. A computed tomography scan of the chest and abdomen revealed small peripherally distributed subpleural nodules (the largest measuring 9 mm in diameter) in the right lung, discrete paratracheal and bilateral hilar lymph nodes, and an enlarged subcarinal lymph node (Figure). Fiberoptic bronchoscopy was normal. Microbiology and cytology of transbronchial and bronchial biopsy samples and bronchoalveolar lavage fluid did not provide conclusive findings.

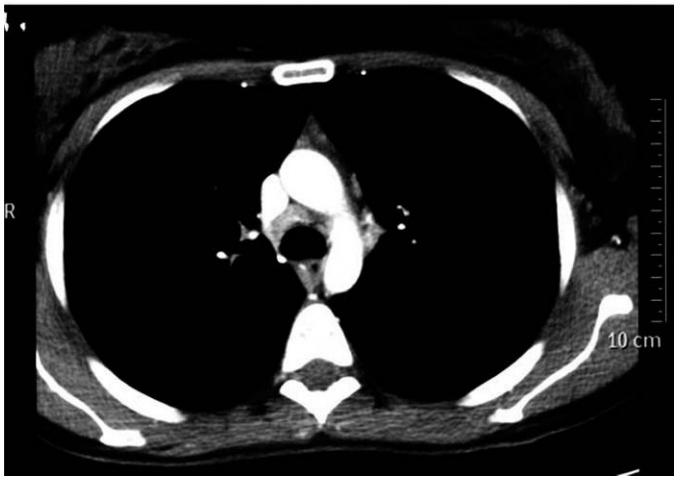

Figure. Computed tomography.

Mediastinoscopy revealed a level 4R lymph node conglomerate, which was biopsied for microbiological and pathological examination. The microbiological analysis was negative, and the pathology report showed reactive lymphadenitis with nonneutrophilic necrosis suggestive of Kikuchi's disease. One month after surgery, the patient had no fever and was asymptomatic.

Kikuchi-Fujimoto disease, nonneutrophilic necrotizing lymphadenitis or histiocytic necrotizing lymphadenitis is a self-limiting disorder that runs a benign course. It mainly affects young Asian women generally less than 30 years of age. Of unknown origin, the disease has been associated with viruses such as parvovirus B19, human immunodeficiency virus, Epstein-Barr virus, cytomegalovirus, and herpes simplex virus type 6, and with microorganisms such as *Toxoplasma*, *Yersinia*, and *Brucella* species. Some authors have described an exaggerated immune response to different agents as the cause of disease.[1]

In 70% to 90% of cases, Kikuchi-Fujimoto disease manifests as cervical lymphadenopathy, which is generally unilateral. The parotid gland is a less common location. Fever is another frequently observed symptom, occurring in 30% to 50% of cases. Other symptoms that may accompany this disease include fatigue, night sweats (as in our patient), weight loss, gastrointestinal disorders, and various skin alterations such as rash.[2,3]

The diagnosis of this disease is complex as there are no conclusive laboratory or imaging test findings, though they may serve to exclude other disorders. Diagnosis can only be made by lymph node biopsy, since fine needle aspiration does not provide sufficient information.[4]

From a pathologic point of view, the disease has 3 phases: proliferation (with an increase in mononuclear cells), necrosis (with necrosis and a predominance of histiocytes), and xanthoma. This means that diseases such as systemic lupus erythematosus, lymphomas, Hodgkin's disease, and tuberculosis should be considered in the differential diagnosis of Kikuchi-Fujimoto disease.

Kikuchi-Fujimoto disease usually runs a benign, self-limiting course, though recurrence has been reported in some cases. Patients should be closely monitored, however, as there appears to be a clear association between lupus and Kikuchi-Fujimoto disease.[1,6]

References

1. López C, Oliver M, Olavarria R, Sarabia MA, Chopite M. Kikuchi-Fujimoto necrotizing lymphadenitis associated with cutaneous lupus erythematosus: a case report. Am J Clin Dermatol. 2000;22:328-33.
2. Scagni P, Peisino MG, Bianchi M, Morello M, Sardi N, Linari A, et al. Kikuchi-Fujimoto disease is a rare cause of lymphadenopathy and fever of unknown origin in children: report of two cases and review of the literature. Am J Pediatr Hematol Oncol. 2005;27:337-40.
3. Chiang YC, Chen RM, Chao PZ, Yang TH, Lee FP. Intraparotid Kikuchi-Fujimoto disease masquerading as a parotid gland tumor. Am J Otolaryngol. 2005;26:408-10.

4. Lin HC, Su CY, Huang CC, Hwang CF, Chieu CY. Kikuchi's disease: a review and analysis of 61 cases. Otolaryngol Head Neck Surg. 2003;128:650-3.
5. Kuo TT. Kikuchi's disease (histiocytic necrotizing lymphadenitis). A clinicopathologic study of 79 cases with an analysis of histologic subtypes, immunohistology, and DNA ploidy. Am J Surg Pathol. 1995;19:798-809.
6. Mugnanini EN, Watson T, Guccion J, Benator D. Kikuchi disease presenting as a flu-like illness with rash and lymphadenopathy. Am J Med Sci. 2003;325:34-7.

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