ENT specialist was requested, in view of suspected upper airway stenosis. Fiberoptic laryngoscopy showed paralysis of the vocal cords (VC) in adduction, and whistling masses consistent with tophi on the VC and arytenoids (Fig. 1C), requiring urgent tracheostomy. Fiberoptic bronchoscopy showed whitish, excretory lesions in the LMB, similar to those observed in the larynx (Fig. 1D). Bronchial biopsies revealed fibrinous bloody material with isolated atypical epithelial cellularity and fragments of bronchial wall with no significant morphological changes. Laryngeal biopsies showed mucosa with squamous epithelium, with no changes and no evidence of malignancy. Of note in the clinical laboratory tests was uric acid 12 mg/dl. After oxygen therapy, bronchodilators, and antibiotics, the patient was discharged with a permanent tracheostomy. During his hospital stay, a respiratory polygraphy with open tracheostomy was performed, showing episodes of severe hypoventilation, and an AH1 of 8.5/h. Treatment began with invasive mechanical ventilation via the tracheostomy in the hospital, followed by night time ventilation at home (VIVO 50 VA/C: Vt 1000 ml, Fr 12 bpm, EPAP 6 cmH2O, Ti 1.5 s, inspiratory trigger 3, and descending ramp), with good compliance. One year later, the patient was admitted for hemoptysis, with soft tissue density visualized on CT around the LMB, origin of the intermediate bronchus and the LUL bronchus. In addition to the LMB lesions, fiberoptic bronchoscopy showed almost complete stenosis of the lingular bronchus and partial stenosis of the culmen with necrotic mucosa. Biopsies revealed well-differentiated squamous cell carcinoma.

Head and neck tissue involvement in CTG is exceptional, and VC paralysis due to acute gouty arthritis is even rarer. Moreover, bronchial involvement has only been described in 1 patient, and there are no more than 20 published cases of laryngeal involvement, none of which appear in the Spanish literature. Although very few studies have reported laryngeal involvement in gout, which is more commonly observed in association with the cricothyroid joint. Garrod, in 1863, described a few “specks” of sodium urate in the arytenoid cartilage in the autopsy of a man with CTG. Years later, Virchow reported the presence of monosodium urate in the VC of a patient with extensive tophaceous deposits. VC paralysis is a potential cause ARF, and exceptionally occurs with acute arthritis. As in our case, VC paralysis is caused by atrophy and denervation of some of the laryngeal muscles of the cricoarytenoid joint, rather than the direct deposit of tophi, causing fibrosis and inflammation on the perineural tissue, that ultimately leads to atrophy of the innervated muscle fibers and hypotrophy of the healthy tissue. Our patient also presented endobronchial tophi, although he ultimately developed squamous cell carcinoma. Of course, no cause–effect relationship between gout and the carcinoma caused by smoking can be established. The diagnosis of cricoarytenoid gout can be established by signs and symptoms observed in patients with extensive CTG, such as dysphonia, dysphagia, dyspnea, etc. Gouty laryngeal involvement should be considered in a case of history of hyperuricemia and acute or chronic gouty arthritis, accompanied by snoring, odynophagia, dysphagia, stridor, dyspnea, and/or dysphonia. Sometimes the upper airway is acutely compromised, requiring urgent tracheostomy, and bronchial lesions can also develop.

References

Mar Arlandis, Virginia Molina, Sandra Vañes, Eusebi Chiner
Servicio de Neumología, Hospital Universitari Sant Joan d’Alacant, Alicante, Spain
* Corresponding author.
E-mail address: echinervives@gmail.com (E. Chiner).

1579-2129/ © 2017 SEPAR. Published by Elsevier España, S.L.U. All rights reserved.

Pleural Lymphoma Associated With Chronic Empyema

Linfoma pleural asociado a empiema crónico

To the Editor,

We report the case of a 78-year-old man, former smoker, with a history of pulmonary tuberculosis treated with left therapeutic pneumothorax and chronic recurrent left empyema, who presented with worsening of his general status in recent months, with dyspnea, weight loss, asthenia, generalized skin dryness, symptoms of right heart failure, and chronic hypercapnic respiratory failure.

Of note on clinical laboratory tests were anemia and mild liver enzyme changes. Chest X-ray revealed a significant increase in previous left pleural effusion with mediastinal shift, so thoracentesis was performed, which yielded cloudy pleural fluid, consistent with exudate, predominantly polynuclear, with low glucose, raised proteins, LDH and ADA, and normal CEA. Cytology was negative for malignant cells and culture was negative for bacteria and mycobacteria.

In view of the patient’s poor progress after evacuation of pleural fluid and findings suggestive of right heart failure, transthoracic echocardiography was performed, which revealed a hypoechoic mass in the inferolateral wall of the left ventricle, with dilatation of the left atrium, slight pericardial effusion, preserved LVEF, and dilated inferior vena cava without inspiratory collapse. Chest–abdomen computed tomography (CT) was performed, showing a soft tissue mass in the medial portion of the posterior chest wall, measuring 6 × 3 cm, with pericardial infiltration, mass effect, and associated small pericardial effusion. A significant increase in the pleural collection compared to previous studies was visualized, occupying practically the entire hemithorax, causing compressive atelectasis of the lung with contralateral mediastinal shift and cardiac compression (Fig. 1).

A CT-guided biopsy of the mass was performed, which according to the pathology report was consistent with non-Hodgkin’s diffuse large B-cell lymphoma, with a proliferation of 70%, no positivity for Epstein Barr virus (EBV) or c-myc, and no bone marrow

infiltration. HIV, HBV, HCV and CMV serologies were negative. The patient’s clinical situation worsened rapidly in a few weeks, so he was referred for palliative care and follow-up, and died 2 months after diagnosis.

Primary pleural lymphoma is an uncommon entity, accounting for approximately 7% of all lymphomas. It usually affects patients with HIV or chronic pyothorax (CP), and occurs only exceptionally in immunocompetent patients. Although the incidence of CP is similar in both sexes, men are more susceptible to developing non-Hodgkin’s lymphomas (NHL) than women (ratio of 5:2:1).

Long-term inflammatory stimulation has been identified as an important etiological factor in the development of malignant lymphomas, and studies have reported longer periods between the onset of CP and the development of NHL (>20 years) in these patients compared to patients with autoimmune diseases or renal transplants (9.5 and 4 years, respectively). In fact, Aozasa et al. used the results of their study to differentiate and establish a characteristic clinico-pathological entity called pyothorax-associated lymphoma (PAL). This entity is defined as a B-cell NHL that develops in the pleural cavity of patients with CP of more than 20 years’ standing, in which an exclusive molecular profile has been determined, consisting of overexpression of interferon alpha-inducible protein 27 that plays a role in chronic inflammation. EBV causes latent infection in PAL, with type III expression of EBV-related proteins in the tumor cells, a phenomenon not observed in our patient.

Although the most common symptom is chest pain, dyspnea may also develop in the presence of significant pleural effusion, as occurred in our case. Radiological signs include diffuse nodular pleural thickening, accompanied by pleural mass. A soft tissue mass in the pleura adjacent to the edge of a coexisting empyema cavity is suggestive of pyothorax-associated lymphoma. Isolated pleural effusion may occasionally appear before the pleural mass develops. Knowledge of the typical radiological findings and location assists in diagnosing this rare disease. In line with the literature, in the months before diagnosis, our patient required repeated evacuating thoracentesis for recurrent empyema, and the pleural mass was only visualized subsequently, located (unusually) in the mediastinal portion of the left posterior chest wall, extending to the pericardium.

For diagnosis, pleural biopsy should be obtained under ultrasound or CT-guidance or by video-assisted thoracoscopy. In histological terms, all cases of PAL are NHL, the most common being diffuse large B-cell type NHL, as found in our patient. Aggressive surgical treatment with pleuropneumonectomy is highly effective in early-stage disease, but is therefore only possible in a very small number of patients. Systemic chemotherapy based on CHOP combinations is required, but efficacy is variable. Radiation therapy is effective for local and primary control and for rescue therapy after chemotherapy. Prognosis is poor, with a 5-year survival rate of 20%–30%. Although this is an uncommon problem, a diagnosis of pleural lymphoma should be taken into account in the long-term follow-up of patients with chronic pleural infection, in order to avoid therapeutic delay.

References


Ana Cerezo-Hernández,* María Victoria García-Gallardo Sanz, Carmen Ainhoa Arroyo Domingo, Félix del Campo Matías

Servicio de Neumología, Hospital Universitario Río Hortega, Valladolid, Spain

*Corresponding author.

E-mail address: ana.cerezo.barqueros@gmail.com

(A. Cerezo-Hernández).

1579-2129/ © 2018 SEPAR. Published by Elsevier España, S.L.U. All rights reserved.