The major congenital variants include an abnormal number or diameter of pulmonary veins, abnormal drainage, and abnormal connections with the pulmonary arterial tree. Acquired abnormalities include hypertension, thrombosis, calcifications, collateral circulation, and stenosis or obstruction. The latter 2 may be caused by cancer, fibrosing mediastinitis, tuberculosis, or complications after radiofrequency ablation.[3]

Complications arising from the ablative procedure are caused by thermal injury to the vessel wall.[5] Pulmonary vein stenosis occurs in 0.5% of patients, and usually develops about 3 months after ablation.[2] Thermal lesions produce scarring and contraction of the vessel wall, causing architectural remodeling and hyperplasia of the intima, producing venous stenosis. Patients may have non-specific respiratory symptoms (dyspnea, cough, chest pain, or hemoptysis) and the severity of symptoms is associated with the number of veins affected, and the degree, length, and duration of stenosis.[5,6]

Mild stenoses may be difficult to detect. CT angiogram clearly shows pulmonary venous occlusion, but this complication is rarer, since anticoagulation starts immediately after the procedure.[2]

Pulmonary parenchymal abnormalities are indirect signs of significant stenosis or venous occlusion, which can include multifocal opacities or peripheral consolidations that might indicate alveolar infarction or hemorrhage, or interstitial septal thickening.[5] Venous occlusion is frequently accompanied by perivenous infiltrate and locoregional lymphadenopathies caused by thermal damage.[2]

Stenoses are managed according to the grade of severity compared to the pre-ablation findings. If stenosis is 50%–70%, follow-up in 3–6 months is recommended; if stenosis is 75%, a repeat CT in 3 months is recommended, and if it is 90%, urgent treatment is required within 3–6 weeks, since the lesion could progress. The treatment of choice in this case is angioplasty followed, if necessary, by stent placement.[2]

In conclusion, pulmonary vein stenosis and occlusion are increasingly rare complications of ablation for AF, but it is important that they are investigated, since a good prognosis depends on early diagnosis and prompt treatment. Imaging techniques such as CT angiography play a fundamental role in the management of these lesions, thanks to their good anatomical resolution, rapid results, and availability. In-depth understanding of the anatomy of the pulmonary veins and radiological findings of the complications of ablative surgery are therefore essential.

References

1. Ghaye B, Szapiro D, Dacher JN, Rodriguez LM, Timmermans C, Devillers D, et al. Percutaneous ablation for atrial fibrillation: the role of cross-sectional imaging. Radiographics. 2003;23:S19–33.
2. Madrid Pérez JM, García Barquín PM, Villanueva Marcos AJ, García Bolao JI, Bastarrika Alemañ G. Complicaciones asociadas a la ablación mediante radiofrecuencia de venas pulmonares. Radiología. 2016;58:444–53.
3. Varona Porres D, Persiva Morenza O, Pallisa E, Roque A, Andreu J, Martínez M. Learning from the pulmonary veins. Radiographics. 2013;33:999–1022.
4. Ravenel JG, McAdams HP. Pulmonary venous infarction after radiofrequency ablation for atrial fibrillation. AJR Am J Roentgenol. 2002;178:664–6.
5. Kwon M, Lee HY, Cho JH, Um SW. Lung infarction due to pulmonary vein stenosis after ablation therapy for atrial fibrillation misdiagnosed as organizing pneumonia: Sequential changes on CT in two cases. Korean J Radiol. 2015;16:942–6.
6. Yataco J, Stoller JK. Pulmonary venous thrombosis and infarction complicating pulmonary venous stenosis following radiofrequency ablation. Respir Care. 2004;49:1525–7.

Laura Fernández-Navarro,[a,*] Elena Moya-Sánchez,[a] Diego Segura-Rodríguez,[b] Eduardo Ruiz-Carazo[a]

[a] *Departamento de Radiodiagnóstico, Hospital Universitario Virgen de las Nieves, Granada, Spain*
[b] *Departamento de Cardiología, Hospital Universitario Virgen de las Nieves, Granada, Spain*

* Corresponding author.
E-mail address: laurafn2617@gmail.com
(L. Fernández-Navarro).

1579-2129/

Angiolymphoid Hyperplasia With Eosinophilia of the Lung

Hiperplasia angiolinfoide con eosinofilia del pulmón

Dear Editor:

Angiolymphoid hyperplasia with eosinophilia (ALHE), also named epithelioid hemangioma, is a benign vascular tumor with unknown pathogenesis, characterized by the presence of well formed, but often immature vessels, and by the proliferation of epithelioid endothelial cells with prominent lymphocytic infiltration.[1] The majority of the ALHE lesions affects the subcutaneous tissue of the head and neck.[2] Reported cases of the pulmonary involvement are extremely rare.[3,4]

We report the case study of a 27-year-old Caucasian woman, non-smoker. The patient performed an abdominal computed tomography (TC) for a history of abdominal pain that showed bilateral nodules in the pulmonary bases, reason why the patient has been referred to our hospital for study. The patient reported a history of cough and asthenia with 1-year evolution, which have devalued over time. Physical examination, including skin observation, was unremarkable and the laboratory investigation was normal. The chest CT showed the presence of multiple bilateral pulmonary nodules (ranging between 10 and 14 mm), some of which in ground glass (Fig. 1A and B). The fiberoptic bronchoscopy was normal and the bronchoalveolar lavage showed a normal cell count. Histopathological examination of TC-guided transthoracic core needle biopsies of one of the left lung nodule have revealed proliferation of numerous small-caliber vessels with hyperplastic endothelial lining, lymphoid follicles and prominent eosinophilic background (Fig. 1C). Immunohistochemical study showed tumor cell positivity for CD34 (endothelial marker) (Fig. 1D). Overall, these findings indicated the diagnosis of angiolymphoid hyperplasia with eosinophilia. Given the rarity of the pulmonary involvement by ALHE, together with the presence of multiple pulmonary nodules (more common in epithelioid hemangioendothelioma), the possibility of surgical biopsy was discussed with Thoracic Surgery. Nevertheless, surgical option was discarded because of the low density of the nodules as well due to location constrains. Instead, we repeated TC-guided transthoracic core needle biopsies in one right lung nodule. The histopathological and immunohistochemical

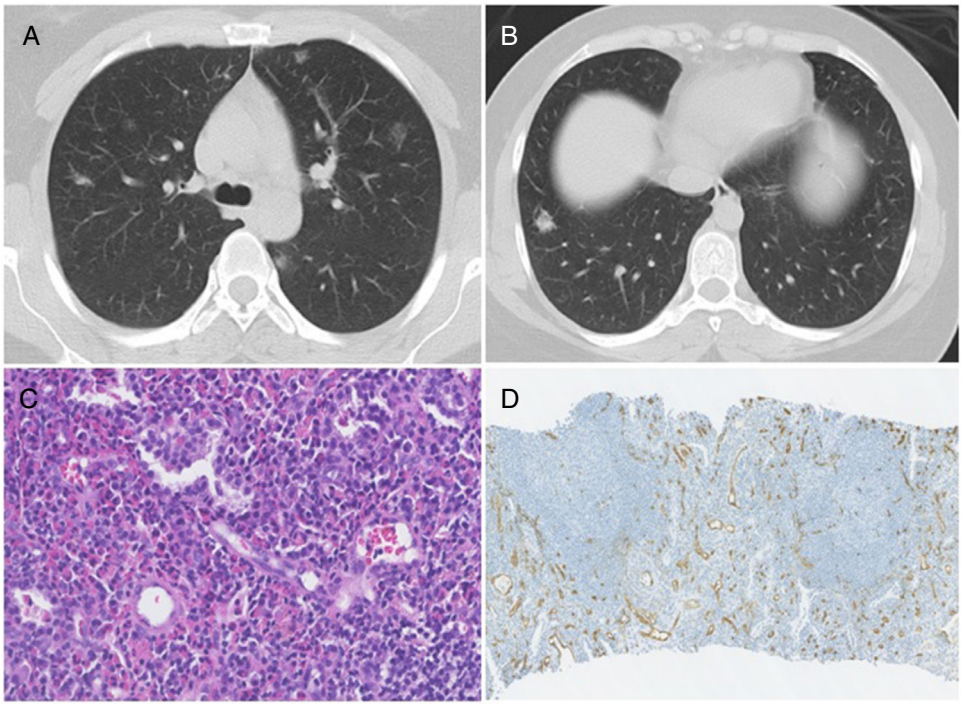

Fig. 1. (A) Chest CT showing bilateral pulmonary nodules, predominantly in ground-glass. (B) Chest CT showing a nodule at the base of the right lung, which was biopsied. (C) Photomicrograph illustrating lymphocytes and prominent eosinophilic background; Stain: hematoxylin and eosin (HE); magnification: 40í. (D) Immunohistochemistry photomicrograph showing numerous small-caliber vessels positive for CD34; magnification: 10í.

studies confirmed the diagnosis of ALHE. Given its benign nature, we have decided to maintain only clinical and imaging follow-up, and the monitoring resulted in no changes during 2 years.

ALHE is an unusual lesion that mainly occurs between the third and fifth decades of life and appears to have a predilection for women.[1] It mostly affect the subcutaneous tissue, however, cases of the involvement of other organs, like lung, are extremely rare.[3,4] In 2005, Moran and Suster described the first two cases of ALHE located in the lung of a 60-years-old man with a history of cough and dyspnea and a woman with 27-years-old with clinical history of asthma.[3] The two cases only presented a single pulmonary lesion, with 20 and 30 mm, respectively, very different from the TC findings of our patient, Moreover, Dulohery et al. have published a study about the lung involvement in hypereosinophilic syndromes that included the case of a 32-years-old woman with a history of cough and rash presented, similar to our case, numerous bilateral nodules, and the diagnosis of ALHE was achieved after a surgical biopsy.[4] In this case-study, the patient was treated initially with prednisone followed by interferon-α2b, with symptomatic and radiologic improvement, however details like dose or time of treatment were undisclosed. Contrary to this case, our patient did not have peripheral eosinophilia. To the best of our knowledge, this are the only three published cases describing the pulmonary involvement by ALHE. Overall, all the published cases presented very different clinical, laboratory and radiologic findings, highlighting that much remain unknown regarding this entity.

Pathological features is a key tool for the diagnosis of ALHE. Histopathology features include prominent proliferation of small, capillary-sized vessels with immature appearance, lined by plump, epithelioid endothelial cells. An inflammatory background rich in eosinophils and lymphocytes is present in the overwhelming majority of cases and, frequently, as in our case, a prominent lymphoid reaction with follicle formation is present. Immunohistochemical studies for CD31 and VIIIrAg stained the epithelioid endothelial cells. Immunoreactivity for CD34 is also present, though often to a lesser degree.[1] ALHE must be distinguish from epithelioid hemangioendothelioma. The latter, is a rare vascular tumor of intermediate behavior, but potentially malignant, also with predilection for middle-aged women and similarly to ALHE, it is radiologically characterized by the presence of multiple, bilateral, perivascular nodules with well-defined or blurred margins.[5] Histologically, epithelioid hemangioendothelioma is characterized by the presence of vacuolated endothelial cells growing singly or in linear streaks or cords, separated by a myxohyaline stroma, and lack of lymphoid or eosinophilic inflammatory infiltration.[2]

Due to the scarcity of published cases, no ideal treatment is defined. We decided to keep only clinical and imaging follow-up.

Herein, we have described one rare case study of pulmonary involvement by ALHE, a diagnosis that must be added to the large list of differential diagnoses of multiple lung nodules, also extending the spectrum of vascular and lymphoid lesions that can occur within the lung.

References

1. Fletcher CDM, Unni KK, Mertens F, editors. World health organization classification of tumours. Pathology and genetics of tumours of soft tissue and bone. IARC Press: Lyon; 2002.
2. Guo R, Gavino A. Angiolymphoid hyperplasia with eosinophilia. Arch Pathol Lab Med. 2015;139:683–6.
3. Moran CA, Suster S. Angiolymphoid hyperplasia with eosinophilia (epithelioid hemangioma) of the lung: a clinicopathologic and immunohistochemical study of two cases. Am J Clin Pathol. 2005;123:762–5.
4. Dulohery MM, Patel RR, Schneider F, Ryu JH. Lung involvement in hypereosinophilic syndromes. Respir Med. 2011;105:114–21.
5. Sardaro A, Bardoscia L, Petruzzelli MF, Portaluri M. Epithelioid hemangioendothelioma: an overview and update on a rare vascular tumor. Oncol Rev. 2014;8:259.

Liliana Ribeiro,[a,*] Márcia Souto,[b] Ana Loureiro[a]

[a] *Department of Pulmonology, Centro Hospitalar de Trás-os-Montes e Alto Douro, Vila Real, Portugal*
[b] *Department of Internal Medicine, Centro Hospitalar de Trás-os-Montes e Alto Douro, Chaves, Portugal*

* Corresponding author.
E-mail address: liliana.sc.ribeiro@hotmail.com (L. Ribeiro).

1579-2129/

Assessment of Midazolam Sedation in Ultrasound-Guided Percutaneous Biopsy of Peripheral Lung Lesions☆

Valoración de la sedación con midazolam en las punciones pulmonares periféricas dirigidas por ecografía torácica

To the Editor,

Thoracic ultrasound (TU) is a complementary tool in respiratory medicine that has many applications in patients with peripheral pulmonary parenchymal and pleural disease. One of these is TU-guided biopsy of peripheral lung lesions (PLL) for the diagnosis of suspected malignancy.[1–5]

Over the years, the respiratory community has become aware of the importance of conscious sedation in patients undergoing interventional techniques in respiratory medicine. The most commonly used benzodiazepine in sedation is midazolam, given its sedative, anxiolytic, amnesic, and muscle relaxant properties. No studies have yet been published investigating the degree of sedation in TU-guided PLL biopsy and patient satisfaction with or without midazolam.

We report our experience in 2 patient groups undergoing TU-guided PLL biopsy: group A without midazolam; and group B with midazolam.

This was an ambispective, observational case–control study. Data from the control group (A) who did not receive sedation were collected retrospectively, and those from group (B), who received sedation, were collected prospectively.

Control data, including the satisfaction survey, vital signs, complications, and diagnosis, were reviewed retrospectively. For the cases, the satisfaction survey and all other variables were collected prospectively. The satisfaction survey was based on previous publications which reported patient satisfaction with respiratory endoscopic techniques.[6–10]

The sample size required for group B was calculated from the score obtained on the satisfaction survey of the historical control group.

In total, 39 patients with no contraindications for PLL biopsy or for sedation were included. They were assessed previously by a nurse and a pulmonologist with experience in interventional respiratory medicine. Patients were considered for inclusion if they had PLL in contact with the chest wall, previously visualized on chest computed tomography, suspected to be lung cancer in any disease stage, with an area of contact between the lesion and the chest wall of at least 2 cm.

Patients had to meet all inclusion criteria and none of the exclusion criteria (less than 18 years of age, coagulation, liver or kidney disorders, unstable ischemic heart disease, COPD with FEV1 <30%, ASA (American Society of Anesthesiologists) status > III, or hemodynamic instability). At least 2 and at most 3 passes were performed for both fine needle aspirations and biopsies. The patient survey comprised 12 questions, of which 10 required Likert-type responses (A lot, Quite a lot, A bit, A little, Very little). The other 2 questions were multiple-choice. The interventional pulmonologist completed a 3-question survey, 2 of which had multiple numbered choices (0 = None, 1 = A little, and 2 = A lot), and a third question with alternative responses.

Group B received midazolam at a dilution of 1 mg/ml, and doses of 1 mg were administered during the process at 3-min intervals if required by the patient, to a maximum dose of 5 mg. Both groups received local anesthesia with lidocaine 2% applied to the subepidermis and the parietal pleura. No patients in either group used oral anxiolytics before the procedure.

Nineteen patients were included in group A, and 20 in group B.

Demographic variables showed no statistically significant differences between the baseline values of both groups, with the exception of age. Nor were differences observed in vital signs or number of biopsies. Procedure duration was shorter, but not statistically, in the sedation group (Table 1).

Scores for each of the Likert-type responses were examined to evaluate the patients' perception of the procedure. Scores for each question showed a higher feeling of discomfort during the biopsy in group A. Patients who received midazolam were less nervous at the prospect of repeating the procedure. The perception of pain, memory of the procedure, and the perception of the length of the procedure were greater in the group that did not receive midazolam. Patients' perception of care received and confidence and trust in the staff were similar in both groups.

In the multiple-choice questions, patients in group A reported that the worst moment was receiving the anesthesia, and in group B, the worst moment was going into the procedure room. Group A would probably repeat the biopsy, and group B would definitely repeat it.

☆ Please cite this article as: Wangüemert Pérez AL, González Expósito H, Pascual Fernández L. Valoración de la sedación con midazolam en las punciones pulmonares periféricas dirigidas por ecografía torácica. Arch Bronconeumol. 2018;54:342–343.