

Case report

Human pulmonary dirofilariasis: uncommon cause of pulmonary coin-lesion

Ottavio Rena^{a,*}, Monica Leutner^b, Caterina Casadio^c

^aThoracic Surgery Department, University of Torino, Ospedale San Giovanni Battista, Via Genova 3, 10126 Torino, Italy

^bPathology Department, University of Eastern Piedmont "A. Avogadro", Ospedale Maggiore della Carità, Novara, Italy

^cUnit of Thoracic Surgery, University of Eastern Piedmont "A. Avogadro", Ospedale Maggiore della Carità, Novara, Italy

Received 10 January 2002; received in revised form 2 April 2002; accepted 4 April 2002

Abstract

Pulmonary dirofilariasis is a rare entity caused by *Dirofilaria*, a dog worm that is transmitted to humans by mosquitoes. The filarial nematode enters the subcutaneous tissue, travels to the right ventricle, dies and then embolizes the pulmonary vessels causing a small pulmonary infarction, which subsequently appears as a solitary nodule. Although these nodules are usually identified incidentally by chest radiography in asymptomatic patients, the lesion is generally presumed to be neoplastic. Diagnosis is made by surgical excision. Awareness of this benign entity is important in the differential diagnosis of pulmonary coin-lesions. © 2002 Elsevier Science B.V. All rights reserved.

Keywords: Dirofilarial granuloma; Dirofilariasis; Pulmonary infarct; Pulmonary nodules

1. Introduction

Pulmonary dirofilariasis is a rare, zoonotic disease, caused by *Dirofilaria immitis* or *Dirofilaria repens* that is usually transmitted from dogs or other carnivores to humans by mosquitoes, with a tropical and subtropical distribution in Southern Europe, Asia, Australia and North and South America [1]. Only 12 cases of human pulmonary dirofilariasis have so far been reported in Europe out of a world-wide total of about 180 cases [2,3]. We report about two new cases due to *D. repens* which is a parasite of the dog which is present only in the Old World.

2. Case reports

2.1. Patient 1

In June 1997, a 56-year-old woman was evaluated for arthroscopy and found to have a new pulmonary nodule in the right lower lobe by chest roentgenogram. The patient was asymptomatic and past pathologic history was uneventful. Serologic and pulmonary function tests were normal. Computed tomography (CT) of the chest demonstrated a non-calcified, well-defined 1.5 cm mass in the posterior

segment of the inferior pulmonary lobe without pleural reaction or hilar or mediastinal lymph nodal enlargements. Bronchoscopy was negative, as was cytological examination of the bronchoalveolar lavage. Trans-thoracic fine-needle aspiration showed necrotic lung parenchyma, but necrotic primitive or secondary lung neoplasm could not be excluded. The patient underwent right lateral muscle sparing mini-thoracotomy and wedge resection of the lesion in the right inferior lobe. Frozen sections revealed the absence of neoplastic cells and negativity of the resection margins. The operation was terminated, the postoperative stay was uneventful and the patient was discharged home on day 4. Pathologic analysis confirmed a pulmonary infarction due to *D. repens*. Four years after surgery, the patient is well.

2.2. Patient 2

In August 2001, a 62-year-old asymptomatic male ex-smoker was found to have a 1.0 cm right upper lobe pulmonary nodule on routine chest radiography. CT confirmed a non-calcified right upper lobe nodule: the mediastinum resulted negative as did the pleural space. The patient's past history revealed an episode of high fever of unknown cause 20 years before. All serologic evaluations were negative. Bronchoscopy and cytology on bronchoalveolar lavage were both negative. Trans-thoracic fine-needle aspiration revealed 'non-diagnostic' specimens. The patient under-

* Corresponding author. Tel.: +39-11-6336775; fax: +39-11-6960170.
E-mail address: ottavio.rena@tiscalinet.it (O. Rena).



Fig. 1. Cross-section of the parenchymal granuloma with the pulmonary artery centered by fragments of the parasite (hematoxylin and eosin stain with Weighert reaction for the elastic fibers, $\times 20$).

went right lateral muscle sparing mini-thoracotomy and wedge resection of the lesion in the right upper lobe: frozen section excluded primary or secondary lung neoplasms; the operation was terminated and the patient discharged home 3 days after surgery. Pathologic analysis confirmed pulmonary infarction caused by *D. repens* (Fig. 1).

3. Comment

D. immitis and *D. repens*, filarial nematodes, are an unusual cause of solitary pulmonary nodules. The first case of human pulmonary dirofilariasis due to *D. immitis* was described by Dashiell in the USA in 1961 [4]. The majority of patients described world-wide reside in the south-eastern United States. Other countries that have reported cases are Japan, Australia, Italy, Costa Rica and The Netherlands. In Italy in general, and in the area of Piedmont in particular, the prevalence of dirofilariasis is one of the highest reported world-wide and the zoonosis is due to *D. repens* [5].

Hosts, other than the dog, include cat, fox, muskrat, wolf, otter, coyote, sea lion and jackal [1]. The adult female nematode lodges in the right ventricle of the dog, attaining a length of up to 30 cm and a width of 3–4 mm; the male develops to about half that size. The female sheds up to several thousand microfilaria daily, and an infected dog may circulate several hundred per millimeter of blood. The microfilaria are transported by a mosquito vector (*Culex*, *Aedes* or *Anopheles* [6]) to another animal's integument through which they migrate into subcutaneous tissues and muscle sheaths where they reside, molt and mature for 80–120 days. They further migrate through capillaries to the heart, where the worm reaches maturity in about 6 months to initiate the cycle again. A similar course has been postulated to explain human infections. However, the human environment is apparently unsuitable for propagation of this cycle:

the adult worm dies and embolizes to the lung causing a pulmonary infarction [7,8].

Most human infections have been in adults. The patients are generally asymptomatic, and a single pulmonary nodule from 1 to 3 cm is identified in the periphery of the lung on routine chest radiography [3,5,9]. Occasionally, multiple nodules miming lung metastatic disease can be observed. The thoracic CT scan helps to better characterize the nodules and some patients have multiple and/or bilateral lesions that could suggest histoplasmosis, Wegener's granulomatosis, septic emboli, or metastatic carcinoma, and in these cases, a biopsy specimen is necessary to establish the final diagnosis. Clinical symptoms may include cough, chest pain, hemoptysis, fever, chills and malaise [3,5,9].

Macroscopically, the lesion appears as a well-circumscribed, grayish-yellow rounded nodule, containing a dead parasite within a thrombotic pulmonary artery, surrounded by a normal lung [3]. Microscopic analysis reveals that the center of the involved parenchyma has undergone coagulation, necrosis, enveloped by a granulomatous zone composed of epithelial cells, plasma cells, lymphocytes and occasional giant cells [3,9]. Peripherally, the lesion is demarcated by fibrous tissue, and the lung parenchyma contains scattered collections of lymphocytes, macrophages and eosinophils [3,9]. Pulmonary vessels, in the area of the nematode or even in tissue located some distance away, may have varying degrees of endoarteritis [3,9].

Dirofilariasis should be included in the differential diagnosis in patients with asymptomatic, small, solitary pulmonary nodules. Serologic tests, trans-thoracic fine-needle biopsy, bronchial washing, biopsy and sputum cytological analysis lack specificity for accurate diagnosis [10]. Diagnosis by fine-needle aspiration biopsy is rare, although parasite fragments have been identified in two reported cases [7], obviating the need for excisional biopsy. Risks of dissemination when needle biopsy is used are null, because the parasite embolizes to the lung after its death. Simple surgical excision is recommended unless dirofilarial fragments are found by needle aspiration [9].

Human pulmonary dirofilariasis is a benign condition, transmitted by mosquitoes to humans as an accidental host, that results in small, solitary peripheral pulmonary nodules. Awareness of this entity is important in the differential diagnosis of pulmonary coin-lesions in the areas of the world in which the prevalence of the dirofilariasis is reported to be significant.

References

- [1] Neafie RC, Connor DH, Meyers WM. Dirofilariasis. In: Binford CH, Connor DH, editors. Pathology of tropical and extraordinary diseases, Armed Forces Institute of Pathology: Washington, DC, 1976. pp. 391–396.
- [2] Pampiglione S, Del Maschio O, Pagan V, Rivasi F. Pulmonary dirofilariasis in man: a new Italian case. Review of the European literature. Parasite 1994;1:379–385.

- [3] Pampiglione S, Rivasi F, Paolino S. Human pulmonary dirofilariasis. *Histopathology* 1996;29:69–72.
- [4] Dashiell GF. A case of dirofilariasis involving the lung. *Am J Trop Med Hyg* 1961;10:37–38.
- [5] Pampiglione S, Rivasi F, Angeli G, Boldorini R, Incensati RM, Pastormerlo M, Pavesi M, Ramponi A. Dirofilariasis due to *Dirofilaria repens* in Italy, an emergent zoonosis: report of 60 new cases. *Histopathology* 2001;38:344–354.
- [6] Ciferri F. Human pulmonary dirofilariasis in the West. *West J Med* 1981;134(2):158–162.
- [7] Panayiotis JA, Katras A, Christie B. Pulmonary dirofilariasis, the largest single-hospital experience. *Chest* 1992;102:851–858.
- [8] Roy BT, Chirurugi VA, Theis JH. Pulmonary dirofilariasis in California. *West J Med* 1993;158:74–76.
- [9] Milanez de Campo JR, Valente Barbas CS, Brito Filomeno LT, Fernandez A, Minamoto H, Valente Barbas Filho J, Biscegli Jatene F. Human pulmonary dirofilariasis. Analysis of 24 cases from Sao Paulo, Brazil. *Chest* 1997;112:729–733.
- [10] Glickman LT, Griere RB, Schawtz PM. Serologic diagnosis of pulmonary dirofilariasis. *Am J Med* 1986;80:161–164.