CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 22 (2016) 8–11

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

A rare case of human pulmonary dirofilariasis with a growing pulmonary nodule after migrating infiltration shadows, mimicking primary lung carcinoma

Akira Haro [a,b,*], Sadafumi Tamiya [c], Akira Nagashima [b]

[a] Department of Thoracic Surgery, Nakatsu Municipal Hospital, Japan
[b] Department of Thoracic Surgery, Kitakyushu Municipal Medical Center, Japan
[c] Department of Diagnostic Pathology, Kitakyushu Municipal Medical Center, Japan

ARTICLE INFO

Article history:
Received 19 January 2016
Received in revised form 15 March 2016
Accepted 15 March 2016
Available online 19 March 2016

Keywords:
Pulmonary dirofilariasis
Primary lung carcinoma
Interstitial pneumonia

ABSTRACT

INTRODUCTION: Pulmonary dirofilariasis is a rare pulmonary parasitic infection by the nematode *Dirofilaria immitis*. It is characterized by an asymptomatic pulmonary nodule usually seen on chest X-ray. The differential diagnosis of pulmonary dirofilariasis includes other pulmonary diseases, primary lung carcinoma and metastatic lung tumor.
CASE PRESENTATION: Pulmonary dirofilariasis was diagnosed in a woman who presented with interstitial pneumonia. Growth of the pulmonary nodule was detected subsequent to hemoptysis. The histological diagnosis was made based on a wedge resection performed under video-associated thoracic surgery (VATS).
CONCLUSION: Pulmonary dirofilariasis often varies in its clinical course. The diagnosis is best made using wedge resection under VATS.

1. Introduction

Human pulmonary dirofilariasis is a sporadic zoonotic infection caused by *Dirofiria immitis* [1]. This parasitic nemotode primarily infects dogs, whereas humans are accidental hosts [2]. Pulmonary dirofilariasis is initially asymptomatic in humans, with the most common clinical finding being a pulmonary granuloma [3]. Here we present a rare case of pulmonary dirofilariasis in a woman with unusual radiological findings, mimicking primary lung carcinoma.

2. Case presentation

A 66-year-old woman who was a non-smoker presented with hemoptysis. She was examined by chest computed tomography (CT), which showed a pyramid-shaped infiltration by pulmonary hemorrhage in S1 of the right upper lobe (Fig. 1). Seven years later, she had a second episode of hemoptysis. Chest CT revealed a 9-mm, well-demarcated, round nodule in S4 of the right middle lobe against a background of interstitial pneumonia (Fig. 2). She was admitted to our hospital for further examination and treatment. Diagnostic sputum cultures showed no findings indicative of a bacterial infection, including TB. Tumor markers, including carcinoembryonic antigen and cytokeratin 19 fragments, were normal. A blood examination showed no evidence of eosinophilia. During 3 months of observation, the mass continued to grow, reaching a size of 11 × 10 × 8 mm. An 18-fluorodeoxyglucose positron emission tomography/computed tomography (18F-FDG PET/CT) scan showed low 18F-FDG uptake by the nodule. The maximum standardized uptake value was 0.94 (Fig. 3). As neither primary lung carcinoma of a particular histological subtype, including mucinous adenocarcinoma, nor a low-grade malignant tumor could be ruled out, a diangnostic wedge resection of the right middle lobe (S4) was performed (Fig. 4). The intraoperative histopathological diagnosis was a benign granuloma without malignant cells. The postoperative histopathological diagnosis was granuloma with pulmonary dirofiliasis (Fig. 5). The patient's postoperative clinical course was good, and she was discharged on postoperative day 6.

3. Discussion

Dirofilaria immitis is a parasitic nematode that infects dogs, with mosquitoes serving as the vectors [1]. Humans are accidental hosts, and human dirofilariasis is a sporadic infection [2]. A filarial infection in a pulmonary blood vessel of a patient was reported by Faust

* Corresponding author at: Department of Thoracic Surgery, Nakatsu Municipal Hospital, 173 Ohjishimoikenaga, Nakatsu, Oita 871-8511, Japan.
E-mail address: aharo@surg2.med.kyushu-u.ac.jp (A. Haro).

http://dx.doi.org/10.1016/j.ijscr.2016.03.023

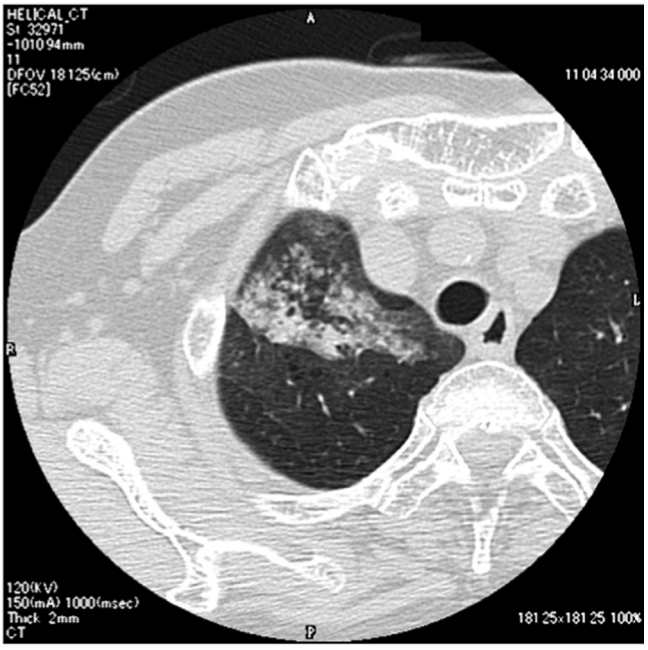

Fig. 1. Chest computed tomography (CT) images show a pulmonary hemorrhage spreading radially in the right upper lobe.

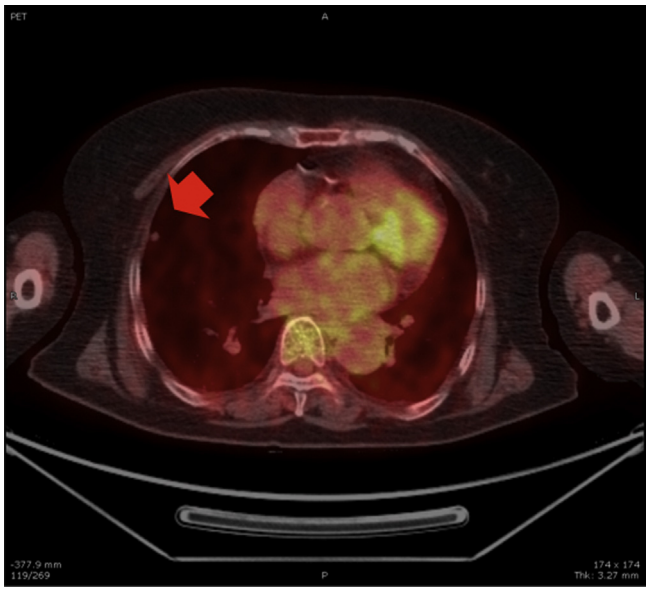

Fig. 3. Positron emission tomography/CT images show the low signal accumulation of signal in a pulmonary nodule (arrow).

in 1952 [4]; Dashiell described the first case of human pulmonary dirofilariasis in 1962 [1].

The most common clinical finding in human pulmonary dirofilariasis is a pulmonary granuloma, which develops when the immature worms reach a branch of the pulmonary artery and then settle in the lung. A small nodule often forms, caused by an inflammatory reaction that remains asymptomatic [3]. The pulmonary nodule is usually subpleural, often speculated or cavitated, has a predilection for the right lower lobe of the lung [5], and a size <3 cm [6]. The differential diagnosis includes pulmonary dirofilariasis but also other pulmonary diseases, such as primary lung carcinoma, metastatic lung tumors, tuberculosis, and hamartomas [1,4,7].

Most patients with pulmonary dirofilariasis are asymptomatic, but ~38% will have symptoms [8] of cough, chest pain, fever, eosinophilia, and/or hemoptysis [9]. In one report, eosinophilia was described in 11% of dirofilariasis patients [8]. The main symptom in our patients was hemoptysis. Chest CT showed a pulmonary hemorrhage in S1 of the right upper lobe on the same side. These findings suggested that the worm had migrated from the right upper lobe to the right middle lobe, where it triggered granuloma formation. Previous reports described new coin lesions together with migrating infiltration shadows in patients with *D. immitis* [4] and *Paragonimus Miyazakii* [10] infections. The former report did not find an association between the nematode and a migrating infiltration shadow, whereas the latter concluded that if the worms fail to migrate, secondary effects will not be seen.

Primary lung carcinoma often occurs against a background of interstitial pneumonia [11]. For this reason, a tumor could not be ruled out in our patient, who presented with a slowly growing pulmonary nodule and interstitial pneumonia. A rare case of pulmonary dirofilariasis with coexisting primary lung carcinoma has been also reported [12]. Wedge resection allowed an operative histopathological diagnosis. Following successful surgical resection, postoperative chemotherapy was unnecessary [13].

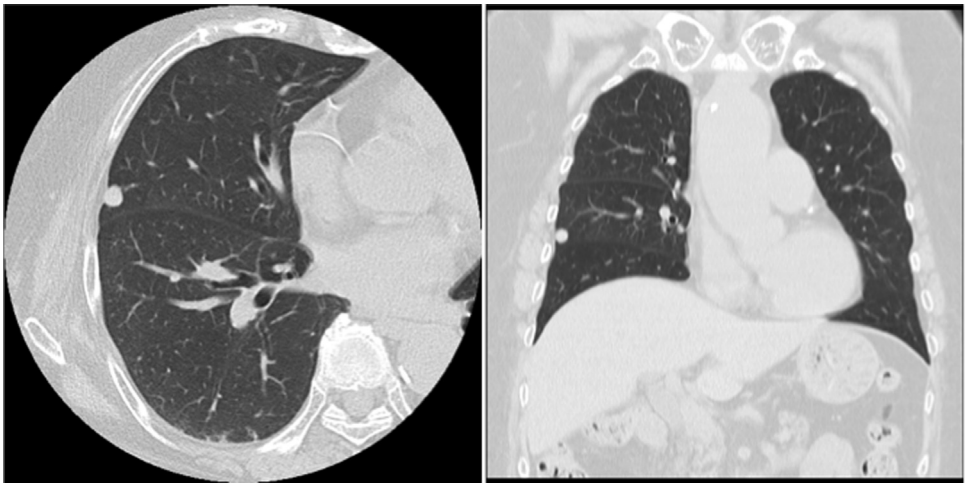

Fig. 2. Transverse and coronal chest CT images show a growing pulmonary nodule in the right middle lobe.

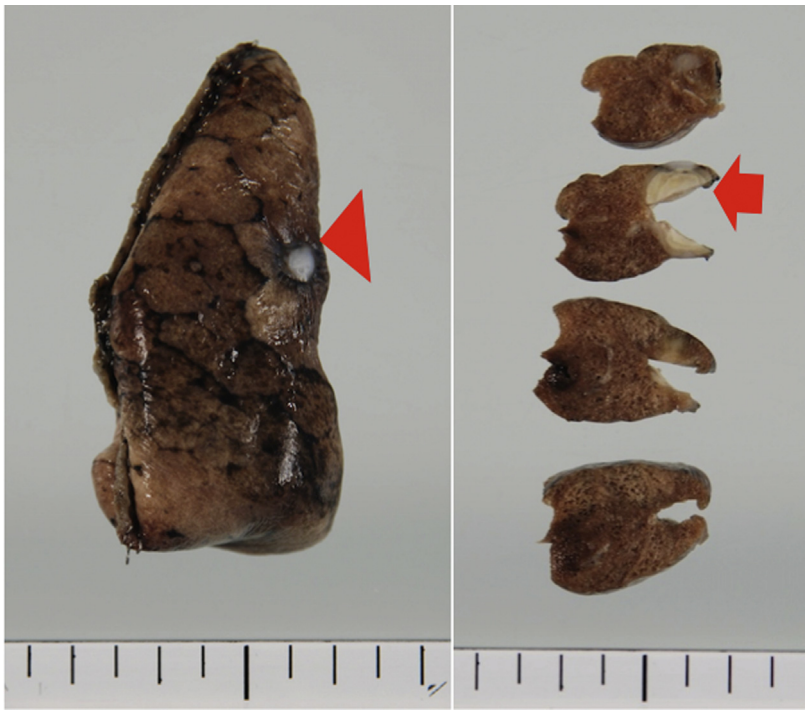

Fig. 4. Macroscopic findings of the surgically resected pulmonary nodule. The nodule exhibits white pleural change (arrowhead); its cut surface is also white (arrow).

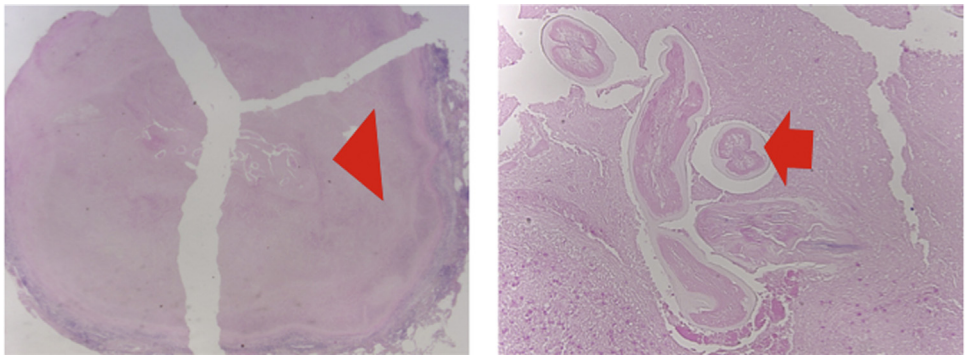

Fig. 5. Histopathological findings of the pulmonary nodule. *Dirofilaria immitis* is present in the center of the necrotic granuloma (left; arrowhead). The worm has the characteristic internal longitudinal cuticular ridges (right; arrow).

4. Conclusion

Pulmonary dirofilariasis is a rare disease but it should be included in the differential diagnosis of diseases characterized by an asymptomatic pulmonary nodule. Surgical biopsy by wedge resection under minimally invasive video-associated thoracic surgery is useful for the diagnosis.

Conflicts of interest

The authors have no conflicts of interest to declare.

Sources of funding

None.

Ethical approval

Not requested.

Consent

Written informed consent was obtained from the patient to publish this case report and the accompanying images.

Author's contribution

AH acquired the data and wrote the article. ST and AN coordinated and critically revised the study. Both authors read and approved the final manuscript.

Guarantor

Akira Haro.

Acknowledgment

The authors thank Dr. Fumiko Kojima (Department of Health Sciences, School of Medicine, Kyushu University) for her help in the histopathological diagnosis.

References

[1] K. Hiroshima, A. Iyoda, T. Toyozaki, T. Fujisawa, F. Aosai, M. Kobayashi, et al., Human pulmonary dirofilariasis: report of six cases, Tohoku J. Exp. Med. 189 (1999) 307.
[2] M. Benzaquen, D. Brajon, M. Delord, N. Yin, F. Bittar, I. Toga, et al., Cutaneous and pulmonary dirofilariasis due to *Dirofilaria repens*, Br. J. Dermatol. 173 (2015) 788.
[3] F. Simon, M. Siles-Lucas, R. Morchon, J. Gonzalez-Miguel, I. Mellado, E. Carreton, et al., Human and animal dirofilariasis: the emergence of a zoonotic mosaic, Clin. Microbiol. Rev. 25 (2012) 507.
[4] T. Kuraki, H. Kobayashi, S. Shikata, Y. Uwabe, N. Nagata, M. Watanabe, et al., [Pulmonary dirofilariasis with cavity formation and spontaneous regression], Nihon Kyobu Shikkan Gakkai Zasshi 34 (1996) 685.
[5] D.B. Flieder, C.A. Moran, Pulmonary dirofilariasis: a clinicopathologic study of 41 lesions in 39 patients, Hum. Pathol. 30 (1999) 251.
[6] T. Miyoshi, H. Tsubouchi, A. Iwasaki, T. Shiraishi, K. Nabeshima, T. Shirakusa, Human pulmonary dirofilariasis: a case report and review of the recent Japanese literature, Respirology 11 (2006) 343.
[7] T. So, R. Mitsueda, T. Miyata, A. Sekimura, T. Yoshimatsu, N. Nose, et al., Pulmonary dirofilariasis in a 59-year-old man, J. Surg. Case Rep. 2014 (2014).
[8] T.S. Bailey, A. Sohrabi, S.S. Roberts, Pulmonary coin lesions caused by *Dirofilaria immitis*, J. Surg. Oncol. 44 (1990) 268.
[9] A. Biswas, P. Reilly, M.H. Perez At Yassin, Human pulmonary dirofilariasis presenting as a solitary pulmonary nodule: a case report and a brief review of literature, Respir. Med. Case Rep. 10 (2013) 40.
[10] S. Katagiri, M. Kondo, T. Ohnuki, J. Tamaoki, A. Nagai, [A case of Paragonimus miyazakii with migrating infiltrates: use of ELISA in diagnosis and treatment], Nihon Kokyuki Gakkai Zasshi—J. Jpn. Respir. Soc. 40 (2002) 225.
[11] R. Hubbard, A. Venn, S. Lewis, J. Britton, Lung cancer and cryptogenic fibrosing alveolitis: a population-based cohort study, Am. J. Respir. Crit. Care Med. 161 (2000) 5.
[12] E.A. Mulanovich, V.E. Mulanovich, K.V. Rolston, A case of *Dirofilaria* pulmonary infection coexisting with lung cancer, J. Infect. 56 (2008) 241.
[13] A. Echeverri, R.F. Long, W. Check, C.M. Burnett, Pulmonary dirofilariasis, Ann. Thorac. Surg. 67 (1999).