

A new Tunisian case of subcutaneous dirofilariasis

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Abstract

Introduction: Dirofilariasis is a cosmopolitan zoonosis of dogs and cats, accidentally affecting humans. Human dirofilariasis is a clinical entity caused by infection with nematode species of the genus *Dirofilaria*. *D. Repens* is the most implicated species. It is responsible for benign skin manifestations but often poses problems of differential diagnosis. Although *Dirofilaria* infections are rare, we should be aware of their possibility. We reported a new Tunisian case of dirofilariasis presenting as a subcutaneous periorbital and frontal nodule caused by *D repens* in an old Tunisian man.

Observation: A 71-year old male without underlying medical conditions consulted us for a periorbital subcutaneous nodule and a painful swelling on the forehead which had been evolving for 20 days without fever. He denied redness or pain. On physical examination, there was a periorbital skin nodule and a frontal swelling of 3 and 4 cm in diameter, which was firm, fixed and painless. Blood counts were normal with no eosinophilia. The Sedimentation rate and the C reactive protein were normal. A facial CT scan had showed a right subcutaneous lesion of 25 mm, peripherally enhanced, hypodense and liquified in the center, with a thickening of the right periorbital soft tissues, without any bone lesion and without extension to the intraorbital space. The ophthalmological examination was normal. Surgical excision was performed. Direct examination showed a whitish filamentous structure which parasitological examination concluded to be fragments of adult nematode which was identified as *Dirofilaria repens*. Microfilaria was not detected in peripheral smear. The treatment of choice consisted of complete surgical resection of the nodule. There is no relapse three months after discharge.

Conclusion: Subcutaneous dirofilariasis is rare in our country, but we should be aware of their possibility. The prevalence is probably underestimated due to a non-specific symptomatology. The detection and treatment of the canine reservoir must be done in order to reduce the number of human cases.

Keywords: Dirofilariasis; *Dirofilaria repens*; subcutaneous nodule.

Introduction

Dirofilariasis is an emerging helminthic zoonosis seldom reported. Human involvement is a clinical entity caused by infection with nematode species of the genus *Dirofilaria*. This cosmopolitan zoonosis is usually affecting dogs and cats. Humans are accidental hosts and rarely affected. We reported a new Tunisian case of dirofilariasis presenting as a subcutaneous periorbital and frontal nodule.

Case report

A 71-year-old male without underlying medical conditions living in the North of the country and had not traveled outside of Tunisia, consulted us at the beginning of September 2021 for painful swelling in the temporo frontal region associated with a right periorbital nodule that had been evolving for 20 days without fever. He denied redness or pain. The patient had revealed the notion of mosquito bites one month before. On physical examination, there was a periorbital skin nodule and

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a frontal swelling of 3 and 4 cm in diameter, which was firm, fixed and painless. Blood counts were normal with no hypereosinophilia. The Sedimentation rate and the C reactive protein were normal. A facial CT scan had showed a right subcutaneous lesion of 25 mm, peripherally enhanced, hypodense and liquified in the center, with a thickening of the right periorbital soft tissues, without any bone lesion and without extension to the intraorbital space. The ophthalmological examination was normal. Microscopic study of the nodule biopsy revealed eosinophils and fragments of adult nematode. The worm was identified as *Dirofilaria repens* based on morphological features (**Figure 1**). Microfilaria was not detected in peripheral smear. The treatment of choice consisted of complete surgical resection of the nodule and removal of the worm. There is no relapse three months after discharge.



Figure 1: *Dirofilaria repens* with central digestive tract (A), cuticle with a well-developed muscle layer (B).

Discussion

Outside the areas where exotic human filariasis are rife, more than 27 species of filaria usually parasitic on domestic or wild animals can be accidentally transmitted to humans [1]. *Dirofilaria* are nemathelminths belonging to the Onchocercidae family. Two subgenus have been identified: *Dirofilaria*, including *D. immitis*; and *Nochtiella*, five species of which may be responsible for human infections [2]. *D. repens*, the most common species in Europe, *D. tenuis*, *D. ursi*, *D. subdermata* and *D. striata* in America. Only two species can infest man: essentially the subgenus *Nochtiella* represented by *D. repens* and also the subgenus *Dirofilaria* represented by *D. immitis*. The latter is a cosmopolitan wireframe. It is the agent of human pulmonary dirofilariasis and exceptionally other visceral localizations. As for *D. repens*, widespread in Asia, Europe and Africa and particularly around the Mediterranean basin. It is the cause of subcutaneous or conjunctival damage, more rarely genital, peritoneal, and even pulmonary [1, 3]. Our observation describes a benign nematodosis, subcutaneous heartworm disease caused by *D. repens*. The definitive host is a carnivorous mammal (especially dogs or cats). The intermediate and vector host is a Diptera of the Culicidae family, belonging mainly to the genus *Aedes*. However, the genera *Anopheles* and *Culex* may be involved. This vector transmits the microfilariae in the blood of the animal during an infesting meal. These develop in the muscles of the insect to give infesting larvae

which migrate to the tubes to be transmitted by a new bite to the animal but also accidentally to humans. Heartworm is generally immature in humans which is considered a parasitic dead-ends. Human infections with *D. repens* are increasingly reported in Europe and in countries around the Mediterranean area, with a major focus in Italy [4]. In Tunisia, there have been 16 cases since 1990, including eight with subcutaneous localization [5, 6]. Other cases have been reported with different location (scrotal, ocular, axillary, breast, paraumbilical) [7, 8]. The patients came from different regions of our country. Around the Mediterranean basin, canine reservoirs and vectors are abundant. But, human cases of heartworm disease are rare. It is therefore possible that this pathology is underestimated due to a non-specific and benign symptomatology [9]. Clinically, subcutaneous heartworm disease is manifested in most cases by a little or no painful nodule, sometimes associated with local inflammatory signs. Incubation does not exceed 6 months (1 month for our patient) followed by the appearance of a subcutaneous nodule of 1 to 4 cm, little or not painful, sometimes preceded by a sensation of painful edema [10]. This nodule is most often unique. The bite usually goes unnoticed; moreover, it is only rarely reported. The diagnosis is very often of accidental discovery.

Pathological examination reveals sections of *D. repens* generally in the form of adult female worms, generally immature, as was described in our patient. Morphological analysis of the parasitic sections made it possible to conclude on the diagnosis of dirofilariasis caused by *D. repens* on the diameter of the nematode (sections from 300 to 600 μm in the female and from 250 to 450 μm in the male), the presence of ten papillae cephalic on the anterior extremity, the well-developed muscular structure of the polymyax type, the internal organs that do not fill the cavity and the presence of a multilamellar cuticle, surmounted by parallel longitudinal ridges and regularly spaced 3 at 4 μm , or about 25 per quadrant [11]. The diagnosis is rarely made by direct examination of an adult worm. It is essentially histological, brought to pathological examination of the resected nodule by revealing a section of the parasite surrounded by a polymorphic inflammatory infiltrate including lymphocytes, plasma cells, histiocytes and eosinophils, which constitute a granuloma [2, 12, 13]. Serological diagnosis of the *D. repens* and *D. immitis* subgenera may be possible but remains insensitive and unspecific [12]. Biological diagnosis makes it possible to detect the parasite in humans before resection and could help in species identification. DNA analysis by PCR is considered the gold standard, even when small or altered larval fragments are used [14].

These new methods can be of interest for the diagnosis of visceral forms inaccessible to surgery. These methods are not common practice and they are unavailable in Tunisia. The diagnosis of subcutaneous heartworm is based on the morphological study of the filaria. Treatment is based on surgical removal of the nodule [12, 15]. In our observation, the diagnosis was histological by the demonstration of a section of a nematode following surgical exeresis. The spontaneous evolution was favorable.

Conclusion

Although *Dirofilaria* infections are rare, we should be aware of their possibility. Definitive cure can be achieved with surgery. Antihelminthic drugs may not be required in case of *D. repens*. Finally, We suggest that epidemiological investigations of vectors and canine dirofilariasis should be implemented. The detection and treatment of the canine reservoir must be done in order to reduce the number of human cases.

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