

Case Report

HUMAN SUBCUTANEOUS DIROFILARIASIS IN THE PALM, AN UNUSUAL PRESENTATION: CASE REPORT

Pawan Kumar K.M.¹, Ragini Ananth Kashid², Vanitha S.² and *Chowda Reddy N.³

¹Department of Orthopedics, BGS Global institute of Medical Sciences, Bangalore

²Department of Microbiology, BGS Global institute of Medical Sciences, Bangalore

³Department of Pediatrics, Shridevi Institute of Medical Sciences & Research Hospital, Tumkur, Karnataka

*Author for Correspondence

ABSTRACT

Human subcutaneous dirofilariasis caused by *Dirofilaria* species, is a rare zoonotic infection. In recent times, there have been several reports worldwide, of infections caused by *Dirofilaria repens*. Dirofilariasis is now identified as an emerging zoonosis. Few cases of human dirofilariasis are reported from India, with Kerala being the focus of infection. Most of the cases that are reported from India had ocular infections and very few had subcutaneous involvement of face, neck, and upper part of the body. We report a case of human subcutaneous dirofilariasis from Bangalore, in the palm of the hand, with an unusual presentation.

Keywords: *Dirofilariasis, Dirofilaria repens, Human, Palm, Subcutaneous, Zoonosis*

INTRODUCTION

Human subcutaneous dirofilariasis is a rare zoonotic infection caused by the filarial worms of the genus *Dirofilaria* (Reddy, 2013; Reshmina *et al.*, 2013; Yaranal *et al.*, 2015). There are about 40 species of *Dirofilaria*, of which six species are known to cause accidental human infections. These six species are: *Dirofilaria immitis*, *Dirofilaria repens*, *Dirofilaria ursi*, *Dirofilaria striata*, *Dirofilaria spectans* and *Dirofilaria tenuis* (Reddy, 2013). *Dirofilaria* species infect several domestic and wild animals. *Culex*, *Aedes*, *Anopheles*, *Armigeres* and *Mansonia* species of mosquitoes are involved in the transmission of *Dirofilaria* species (Reddy, 2013; Singh *et al.*, 2010).

Though human dirofilariasis is considered a rare zoonotic infection, there are considerable numbers of cases being reported world over, making it an emerging zoonosis (Reddy, 2013; Reema *et al.*, 2013; Pampiglione and Rivasi, 2000). In India, the endemic focus for human dirofilariasis is Kerala. Cases have also been reported from Assam, Orissa and Karnataka (Reddy, 2013; Harish *et al.*, 2011).

Most of the cases which are reported from India presented with ocular manifestations of human dirofilariasis (Harish *et al.*, 2011; Sekhar *et al.*, 2000; Padmaja *et al.*, 2005). *Dirofilaria repens* is the common causative agent of human subcutaneous dirofilariasis in India (Harish *et al.*, 2011). Most cases of human subcutaneous dirofilariasis occur in the face, neck and upper arms (Reddy, 2013). We report a case of human subcutaneous dirofilariasis affecting the palm, with varied clinical presentation, from Bangalore.

CASE REPORT

A 78 year old man hailing from Bangalore presented in the Orthopedic OPD of our teaching hospital, with a swelling on the palmar aspect of the left hand. There was a solitary swelling, located 2 cm above the distal palmar crease, between the third and fourth metacarpal, of the right hand. The swelling was 2x1 cm. in size, insidious in onset and was progressive over 4 weeks.

It was a firm, non-tender and freely mobile swelling. It was associated with pruritus, for which he was on anti-histaminic drugs for a month.

A clinical diagnosis of foreign body granuloma / ganglion was made. On an outpatient basis, under local anesthesia, the swelling was excised. On excision, a yellow colored, cheesy material oozed out, along with which, a single, whitish worm was seen wriggling. The worm was carefully extracted mechanically

Case Report

and recovered intact. The worm was preserved in formalin solution and sent to the laboratory for identification (Figure 1a – 1b).

On macroscopic examination the worm was whitish in color, 12 cm long and 0.5mm in thickness (Figure 2). It had a short, rounded tail. The microscopic view of the worm revealed that the worm had a thick cuticle. Longitudinal cuticular ridges and transverse striations were seen. The paired uteri of the worm were seen. Based on these findings the worm was identified as female worm of *Dirofilaria repens* (Harish et al., 2011; Cook and Zumalal, 2010; Khurana et al., 2010).

In the past, the patient had no history of travel to Sri Lanka or Kerala, both of which are endemic areas for dirofilariasis. He did not have any animal, as a pet at home. The patient's general physical examination revealed that there was no other swelling. Routine haematological and biochemical tests were normal. The patient's blood was sent for microfilaria examination; however, no microfilaria was detected.



Figure 1a: Whitish worm seen on incision in the swelling present on the palm



Figure 1b: Whitish worm seen on incision in the swelling present on the palm



Figure 2: The extracted *Dirofilaria* worm



Figure 3: The incision closed after worm extraction

Case Report

RESULTS AND DISCUSSION

Discussion

Human dirofilariasis is now recognized as an emerging zoonosis in India (Reddy, 2013; Khurana *et al.*, 2010). The filarial worms of *Dirofilaria* species are natural parasites of dogs, cats, foxes and wild mammals. These filarial worms are transmitted to man by the bite of mosquitoes belonging to *Culex*, *Aedes*, *Anopheles*, *Armigeres* and *Mansonia* species. Humans are the dead end hosts and not the natural hosts for *Dirofilaria* species. Hence, it is thought that in most cases, the infective larvae that are injected through mosquito bites perish before attaining maturity (Yaranal *et al.*, 2015). Most cases of dirofilariasis are asymptomatic, while the symptomatic one's manifest with lung parenchymal disease or subcutaneous nodules (Reshmina *et al.*, 2013). In the Asian subcontinent and in India, *Dirofilaria repens* is the main causative agent of human subcutaneous dirofilariasis (Reshmina *et al.*, 201; Singh *et al.*, 2010; Reema *et al.*, 2013; Padmaja *et al.*, 2005). The risk factors, identified in the acquisition of human dirofilariasis are: mosquito density, warm tropical climate with extended mosquito breeding season, outdoor activities and the abundance of microfilaraemic dogs (Sanjeev *et al.*, 2011; Khurana *et al.*, 2010). There are studies which suggest that humans are at an enhanced risk of acquiring *Dirofilaria* infection from dogs (Khurana *et al.*, 2010). As per two recent surveys, the prevalence of microfilaraemic dogs in Kerala and Karnataka are 7% and 21% respectively (Sanjeev *et al.*, 2011; Sabu *et al.*, 2005; Ananda *et al.*, 2006). Zoonotic filariasis occurs in those individuals who handle cats or dogs, either as pets or as a part of their profession (Sathyan *et al.*, 2006). This patient had no contact with animals either at his residence or at workplace.

In India, several studies have documented ocular dirofilariasis, while the numbers of case reports of subcutaneous dirofilariasis are few (Singh *et al.*, 2010; Harish *et al.*, 2011; Sekhar *et al.*, 2000). Cases of human subcutaneous dirofilariasis have been reported from head, neck, extremities, thoracic wall, lip, breast, axilla, peri-orbital, abdominal wall and male genitalia (Reddy, 2013; Singh *et al.*, 2010). We are reporting a case of human subcutaneous dirofilariasis, in the palm of the left hand, from Bangalore, which is very rare.

In most cases, the clinical presentation of human subcutaneous dirofilariasis present as a single, solitary swelling and the diagnosis has often been missed. In this case, based on the clinical presentation, a diagnosis of foreign body granuloma/ ganglion was made, and the actual diagnosis was missed. Based on the morphological features of the extracted worm, it was identified as female worm of *Dirofilaria repens*. An adult male worm is 5- 7 cm long, 370- 450µm wide, with 2- 6 pre anal papillae on the right side and 4-5 on the left. The left spicule is 460- 590 µm and the right ones are 180- 210 µm. The female worm is 10- 17 cm long and 460 – 650 µm wide (Sanjeev *et al.*, 2011). The female reproductive system has the vulva at the anterior end, the proximal portion of the vagina is bulbous, and it loops and ends in the uterine bifurcation (Khurana *et al.*, 2010). The ovaries and oviducts are highly coiled. The identity of the worm was confirmed as female worm of *Dirofilaria repens*, at the National Centre for Disease Control, Bangalore. Few cases of subcutaneous dirofilariasis caused by *Dirofilaria immitis* have been reported. However, *Dirofilaria immitis* does not have longitudinal and transverse striations (Singh *et al.*, 2010; Padmaja *et al.*, 2005; Sathyan *et al.*, 2006).

The clinical implication of subcutaneous human dirofilariasis is that, it is often misdiagnosed as malignant tumours, which further warrants for invasive diagnostic procedures or surgery (Sabu *et al.*, 2005; Ilyasov *et al.*, 2013). This could lead to detrimental physical and psychological effects on the patient (Ilyasov *et al.*, 2013). Excision of this swelling was both diagnostic and therapeutic (Sanjeev *et al.*, 2011).

Several studies have observed that eosinophil counts and measurement of total Ig E levels are of limited value for screening human subcutaneous dirofilariasis (Ananda *et al.*, 2006). In our case also the patient's eosinophil counts were normal and IgE level was not elevated. To confirm the diagnosis, DNA analysis is done using panfilarial PCR that targets the mitochondrial 12S rRNA gene (Sanjeev *et al.*, 2011; Maltezos *et al.*, 2002; Poppert *et al.*, 2009). The cost involved for this kind of identification of the parasite is a deterrent, for developing countries. In human subcutaneous dirofilariasis, microfilaraemia is extremely rare and hence there is no need for chemotherapy (Singh *et al.*, 2010; Padmaja *et al.*, 2005; Sathyan *et al.*,

Case Report

2006). The patient did not have microfilaraemia, hence chemotherapy was not started. Human subcutaneous dirofilariasis is associated with negligible to mild inflammatory symptoms and hence many cases go undiagnosed (Reddy, 2013).

Most cases of human dirofilariasis remain underreported, as they are undiagnosed or unpublished (Sathyan *et al.*, 2006; Srinivasamurthy *et al.*, 2012). This case report re-emphasises the need to consider human subcutaneous dirofilariasis as a differential diagnosis for solitary subcutaneous swelling. It also reiterates the need to increase the awareness about human dirofilariasis. This will help the clinicians to initiate appropriate treatment, to develop cost-effective, diagnostic tools for species identification, and to deliver effective chemoprophylaxis in animals, especially in endemic areas, to conduct systematic epidemiological surveys and to step up preventive measures (Harish *et al.*, 2011; Gupta *et al.*, 2013).

Conclusion

Human dirofilariasis is an emerging zoonotic disease caused by infection with any of the several species of worms belonging to the genus *Dirofilaria*. Clinicians, pathologists, and microbiologists should have an awareness of this and also the possibility of presentation of these infections both in uncommon and unusual presentations.

ACKNOWLEDGEMENT

We kindly acknowledge Dr. M.E. Mohan, our Principal and Professor of Medicine for his valuable suggestion and guidance.

REFERENCES

- Ananda KJ, D'Souza PE and Jagannath MS (2006).** Methods of identification of microfilaria of *Dirofilaria repens* and *Dipetalonema reconditum*. *J Vet Parasitol* **20** 45- 7.
- Cook CG and Zumla A (2010).** *Filariases*. In: *Manson's Tropical Diseases*, 22nd edition (Philadelphia: Saunders Elsevier) 1477- 1505.
- Gupta CN, Shetty BS, Prathap AG and Bhandary AS (2013).** Case report; Ophthalmic parasitic zoonosis caused by *Dirofilaria repens*. *Journal of Clinical and Biomedical Sciences* **3**(3) 140-42.
- Harish S Permi, Veena S, Kishan Prasad HL, Sunil Kumar Y, Rajashekhar Mohan, Jayaprakash Shetty K (2011).** Subcutaneous human dirofilariasis due to *Dirofilaria repens*: Report of two cases. *Journal of Global Infectious Diseases* **3**(2) 199-201.
- Ilyasov B, Kartashev V, Batrikov N, Morchon R, Gonzalez Miguel J and Simon F (2013). Delayed diagnosis of Dirofilariasis and complex ocular surgery, Russia. *Emerging Infectious Diseases* **19**(2) 326-8.
- Khurana S, Singh G, Bhatti HS and Malla N (2010).** Human subcutaneous dirofilariasis in India: a report of three cases with review of literature. *Indian Journal of Medical Microbiology* **28** 394- 396.
- Maltezos ES, Sivridis EL, Giatromanolaki AN and Simopoulos CE (2002).** Human subcutaneous dirofilariasis. A report of three cases manifesting as breast axillary nodules. *Scottish Medical Journal* **47** 86- 8.
- Padmaja P, Kanagalakshmi, Samuel R, Kuruvilla PJ and Mathai E (2005).** Subcutaneous dirofilariasis in southern India: A case report. *Annals of Tropical Medicine and Parasitology* **99** 437- 40.
- Pampiglione S and Rivasi F (2000).** Human dirofilariasis due to *Dirofilaria (Nochitella) repens*. An update of world literature from 1995 to 2000. *Parasitologia* **42**(231) 54.
- Poppert S, Hodapp M, Krueger A, Hegasy G, Niesen WD, Kem WV et al., (2009).** *Dirofilaria repens* infection and concomitant meningo encephalitis. *Emerging Infectious Diseases* **15** 1844- 6.
- Reddy MV (2013).** Human dirofilariasis: An emerging zoonosis. *Tropical Parasitology* **3**(1) 2- 3.
- Reema Nath, Sorbeswar Bhuyan, Himadri Dutta and Lahari Saikia (2013).** Human subcutaneous dirofilariasis in Assam. *Tropical Parasitology* **3**(1) 75-78.
- Reshmina D'Souza, Ramakrishna PaiJekribettu, Sunil H Sudarshana and Satyamurthy P Aithala (2013).** Subcutaneous nodule: A case of *Dirofilaria*. *International Journal of Applied and Basic Medical Research* **3**(1) 64-65.

Case Report

Sabu L, Devada K and Subramanian H (2005). Dirofilariasis in dogs and humans in Kerala. *Indian Journal of Medical Research* **3** 121-691.

Sanjeev H, Rajini M and Prasad SR (2011). Human Dirofilariasis: An uncommon case of subcutaneous infection with *Dirofilaria repens* with a brief review of literature. *Nitte University Journal of Health Sciences* **1** 1-3.

Sathyan P, Manikandan P, Bhaskar M, Padma S, Singh G and Appalaraju B (2006). Subtenons infection by *Dirofilaria repens*. *Indian Journal of Medical Microbiology* **24** 61- 2.

Sekhar HS, Srinivasa H, Batru BR, Mathai E, Shariff S and Macaden RS (2000). Human ocular Dirofilariasis in Kerala, Southern India. *Indian Journal of Pathology and Microbiology* **43**(77) 9.

Singh R, Shwetha JV, Samantaray JC and Bando G (2010). Dirofilariasis: A rare case report. *Indian Journal of Medical Microbiology* **28**(75) 7.

Srinivasamurthy M, Rao MS, Thejaswini MU and Yoganand (2012). Human subcutaneous dirofilariasis. *Annals of Tropical Medicine and Public Health* **5** 349- 51.

Yaranal PJ, Priyadarshini MM and Purushotham B (2015). Human subcutaneous dirofilariasis of forearm an unusual presentation. *Indian Journal of Dermatology* **60**(103).