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

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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: Dirofilariais, 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 worldwide, 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.
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 ZumlaI, 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 Dirofilarial worm

Figure 3: The incision closed after worm extraction
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 (Sathyen 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 vagina 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; Sathyen 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; Sathyen et al., 2006).
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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.

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