Case study

Submental swelling due to Dirofilaria Repens in Western India- A rare case report.

Abstract

Dirofilariasis is a zoonotic disease transmitted by mosquitoes. ~~D repens~~ *Dirofilaria repens* causes subcutaneous swelling or infections of bulbar conjunctiva and is rare in India. *Dirofilaria* ~~D~~*immitis* causing pulmonary infections is limited to the New World. We present a case of a ~~40 year old~~40-year-old man presenting with a submental cystic swelling that was excised and confirmed as *Dirofilaria repens* on histology.

Introduction- Dirofilariasis is an ~~infection~~ infestation caused by nematode, *Dirofilaria*. It is a natural parasite of dogs, cats, foxes and wild mammals~~.~~(1). Causative species include, *D.repens, D.immitis, D.tenius, D.ursi*among others~~.~~(2). Although worldwide in distribution, Italy in Europe and Sri Lanka in Asia, have higher incidence rates~~.~~(3). Adult female mosquitoes of species *Culex, Andes, Anopheles* and *Armigeus* are vectors for this parasite, taking up the microfilaria during a blood meal and thereafter transmitting it to the next host, after a bite(4). Clinical manifestations include subcutaneous infection, typically of face and neck, ocular infection, as well as pulmonary disease~~.~~(5). Although rare, subcutaneous and ocular infections have been reported from Southern India~~.~~(6). Treatment of subcutaneous lesions is surgical excision of nodule.

Case Report- A 40 year old male, presented to Dr D Y Patil Hospital, Navi Mumbai with a swelling in the submental region since 1 month. Swelling was sudden in onset, gradually progressive, not associated with pain.It was a 1x1 cm , non tender, nodule in submandibular region. Examination of oral cavity was unremarkable and there was no cervical lymphadenopathy. An ultrasound of the nodule revealed a small, hypoechoic, cystic mass, measuring 1.05 x 0.95 x 0.7cm (vol 0.38cc). It showed mobile, tubular structure within, with well defined echogenic wall suggestive of live parasitic worm. Excision of the nodule was done under general anesthesia. A solitary cystic nodule was excised, with a live, 3 cm worm within. On microscopy, cross sections of parasite displayed hyalanised cuticle layer and inner thick muscle layer and body cavity showing intestinal and genital tubules suggestive of *Dirofilaria*. Thee was a presence of inflammatory granulation tissue with fibroblast infection reaction.

Discussion- *Dirofilariae* are helminths of the family *Onchocerciae*.(7). Several subspecies have been reported, the commonest being D repens and *D. immitis*, which commonly presents as a subcutaneous infection in man. Numerous mammals such as dogs, foxes, coyotes, ~~cats ,wolverines~~cats, wolverines, muskrats ~~etc~~etc. serve as hosts for the *Filaria*~~.~~(8)(9).*Mosquitoes* are vectors for the parasite, taking up the microfilaria during a blood meal. The microfilaria develops in the malphigian tubule of mosquitoes into third stage larva, which then migrates to proboscis through the body cavity and transmitted to hosts man, ~~dog~~dog, and other hosts after a bite~~.~~(10). In humans the worms don't reach maturity, therefore can't express larva in blood stream, hence the rarity of microfilaria in humans. There is total of 27 valid species and 15 of questionable validity~~.~~ (11). Other subspecies are *D. tenuis, D. immitis* (dog heart worm) including rare species such as *D. striata, D. ursi* and the recently discovered *D. hongkongensis*~~.~~(12).*D. immitis* is a disease of the old world, while infections with *D. repens* are seen in the new world~~.~~ (13)~~.~~.*D. repens* is responsible for most of thee cutaneous and subcutaneous infections. In Asia, Srilanka is the worst affected country with as many of 60%

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population of dogs being infected whereas in ~~Europe,the~~Europe, the highest number of cases have been recorded in Italy~~.~~ (14).

D.repens, D. tenuis and D. ursi~~is~~are responsible for subcutaneous infections, while *D. immitis* causes pulmonary infections. The pulmonary lesions are often misdiagnosed at presentation~~.~~(15)(16). Other locations such as bulbar conjunctiva have also been reported. Here they may present as vermiform mobile mass underneath the conjunctiva~~.~~(17).*D. repens* causes infections of the bulbar conjunctiva. *D.~~H~~hongkongensis* presented with a recurrent shoulder mass in an Austrian man after a visit to India~~.~~(18). A rare case of *D. immitis* of spermatic cord has also been reported from California~~.~~ (19). In Brazil, a case of *D. spectans* from a digital artery was reported. (20). Infections can spread to deeper tissues such as lymph nodes, abdominal ~~cavity~~cavity, and dura matter~~.~~ (21)(22).

Subcutaneous dirofilariasis can mimic benign and malignant lesions, hence it's worthwhile to consider it as a differential diagnosis for individuals presenting with subcutaneous nodules in endemic areas~~.~~(23). Patient may present with a creeping eruption due to migration of the worm under the skin. In infected individuals, *D~~d~~irofilaria* may migrate for prolonged periods of time (even months) without producing significant symptoms~~.~~ (24). It later, stops migrating and forms a subcutaneous nodule. Location of this nodule could be anywhere over the body,but typically face, perioral and periorbital areas are affected~~.~~ (25)(26). Sites such as scrotum and uncommonly breast have also been reported, probably due to lower temperatures at these sites~~.~~ (27)(28). Diagnosis depends on clinical findings, with visible live worm sometimes seen in ocular infections. Patients with subcutaneous nodules from endemic areas should be treated with a high index of suspicion. Ultrasound can help confirm diagnosis, at times demonstrating mobile worm~~.~~ (29).Polymerase Chain Reaction~~PCR~~ (PCR~~Polymerase Chain Reaction~~) can be used to confirm diagnosis~~.~~ (30). Eosinophilia is an inconsistent finding.Treatment is excision of subcutaneous nodule and microscopic analysis for definitive diagnosis(31). Although some authors suggest tetracycline or Diethycarbamazine~~DEC~~ (DEC~~Diethycarbamazine~~)or Albendazole as an adjunct to surgical excision, there are others who question its benefit as microfilaremia is rare in humans~~.~~(32). Some authors suggest using antihelminthics for recurrent disease.

Conclusion-

Dirofilariasis is an uncommon zoonotic disease that is often misdiagnosed on presentation. Ultrasound may aid in the diagnosis. It's infrequent incidence in Western India makes it a disease of interest to the General Surgeon.

References-

1. Orihel, Thomas C., and Mark L. Eberhard. "Zoonotic filariasis." Clinical microbiology reviews 11.2 (1998): 366-381

2. Capelli, Gioia, et al. "Recent advances on Dirofilaria repens in dogs and humans in Europe." Parasites & Vectors 11.1 (2018): 1-21.

3. Pampiglione, Silvio, and F. Rivasi. "Human dirofilariasis due to Dirofilaria (Nochtiella) repens: a review of world literature." Parassitologia 37.2-3 (1995): 149-193.

4. Fuehrer HP, Auer H, Leschnik M, Silbermayr K, Duscher G, Joachim A. Dirofilaria in humans, dogs, and vectors in Austria (1978–2014). From imported pathogens to the endemicity of Dirofilaria repens. PLoS Negl Trop Dis. 2016;10:e0004547

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5. Jelinek T, Schulte-Hillen J, Löscher T. Human dirofilariasis. Int J Dermatol. 1996 Dec;35(12):872-5. doi: 10.1111/j.1365-4362.1996.tb05054.x. PMID: 8970844

6. Padmaja, P., et al. "Subcutaneous dirofilariasis in southern India: A case report." Annals of tropical medicine & Parasitology 99.4 (2005): 437-440.

7. Grandi, Giulio, Tatjana Zˇivicˇnjak, and Relja Beck. "Pathogenesis of Dirofilaria spp. infections." Dirofilaria immitis (2007): 60-61.

8. Orihel TC: Dirofilaria lutrae sp. n. (Nematoda:Filarioidea) from otters in the southeast United States. J Parasitol.

9. Noronha D, Vicente JJ, Pinto RM: A survey of new host records for nematodes from mammals deposited in the Helminthological Collection of the Oswaldo Cruz Institute (CHIOC). Rev Bras Zool. 2002, 19: 945-949. 10.1590/S0101-81752002000300032.

10.Genchi, Claudio, et al. "Climate and Dirofilaria infection in Europe." Veterinary parasitology 163.4 (2009): 286-292.

11. Canestri Trotti G, Pampiglione S, Rivasi F: The species of the genus Dirofilaria, Railliet & Henry, 1911. Parassitologia. 1997, 39: 369-374.

12. Xing, Fanfan, et al. "Dirofilaria hongkongensis infection presenting as recurrent shoulder mass." Parasitology international 77 (2020): 102117.

13. McCall, John W., et al. "Heartworm disease in animals and humans." Advances in parasitology 66 (2008): 193-285.

14. Dissanaike, A. S., et al. "Human dirofilariasis caused by Dirofilaria (Nochtiella) repens in Sri Lanka." Parassitologia 39.4 (1997): 375-382

15.Simón F, Siles-Lucas M, Morchón R, González-Miguel J, Mellado I, Carretón E, Montoya-Alonso JA: Human and animal dirofilariasis: the emergence of a zoonotic mosaic. Clin Microbiol Rev. 2012, 25: 507-544.

16.Asimacopoulos, Panayiotis J., Anthony Katras, and Byron Christie. "Pulmonary dirofilariasis: the largest single-hospital experience." Chest 102.3 (1992): 851-855.

17. Redon-Soriano, María, et al. "Subconjunctival human dirofilariasis by Dirofilaria repens in the Mediterranean Basin." American Journal of Ophthalmology Case Reports (2022): 101570.

18.Riebenbauer, Katharina, et al. "Human dirofilariosis in Austria: the past, the present, the future." Parasites & vectors 14.1 (2021): 1-10.

19. Theis, J. H., et al. "Case report: Unusual location of Dirofilaria immitis in a 28-year-old man necessitates orchiectomy." The American journal of tropical medicine and hygiene 64.5 (2001): 317-322.

20. Freitas, J. F. T., and R. Mayall. "Fenómeno de Raynaud na mão esquerda, provocado por Dirofilaria spectans." Rev Bras Med 10 (1953): 463-467

21. Révész E, Markovics G, Darabos Z, Tóth I, Fok E. Dirofilaria in the abdominal cavity. Magy Seb. 2008;61:281–4.

22. Perret-Court A, Coulibaly B, Ranque S, Bouvier C, Lena G, Coze C, et al. Intradural dirofilariasis mimicking a Langerhans cell histiocytosis tumor. Pediatr Blood Cancer. 2009;53:485–7

23. Joseph, Elizabeth, et al. "Subcutaneous human dirofilariasis." Journal of Parasitic Diseases 35.2 (2011): 140-143.

24. de Vries PJ, Visser LG, Vetter HCM, Muller HP, Polderman AM. Migrating subcutaneous swellings due to dirofilariasis after a visit to the South of France. Ned Tijdschr Geneeskd. 2003;147:566–9.

25. Kaouech E, Becheur M, Cheikh M, Belhadj S, Kallel K, Chaker E. Dirofilariose sous-cutanée en Tunisie: une observation à localisation labiale. Santé. 2010;20:47–8

26. Desai RS, Pai N, Nehete AP, Singh JS. Oral dirofilariasis. Indian J Med Microbiol. 2015;33:593–4.

27. Leccia N, Patouraux S, Carpentier X, Boissy C, Del Giudice P, Parks S, et al. Pseudo-tumor of the scrotum, a rare clinical presentation of dirofilariasis: a report of two autochtonous cases due to Dirofilaria repens. Pathog Glob Health. 2012;106:370–2.

28. Vucaj Cirilovic V, Dobrosavljev M, Niciforovic D, Donat D, Bogdanovic-Stojanovic D, Jukovic M. Dirofilariasis of the breast: sonographic appearance. J Clin Ultrasound. 2014;42:433–5

29. Ilyasov, Boris, et al. "Thirty cases of human subcutaneous dirofilariasis reported in Rostov-on-Don (Southwestern Russian Federation)." Enfermedades Infecciosas y Microbiologia Clinica 33.4 (2015): 233-237.

30. Haim, Andreas, et al. "A case of human Dirofilaria repens infection, causing an asymptomatic subcutaneous nodule." Parasitology Research 119.5 (2020): 1703-1705

31. Houston, Peter M. MD, MPH; Parks, Amanda L. MD† Subcutaneous Dirofilariasis in the United States, Infectious Diseases in Clinical Practice: January 2022 - Volume 30 - Issue 1 - e1074

doi: 10.1097/IPC.0000000000001074

32. Lupşe, M., et al. "Recurrent subcutaneous human dirofilariasis due to Dirofilaria repens after surgical removal of the worm and anthelmintic treatment." Parasites & Vectors 7.1 (2014): 1-1.

33. Pampiglione, Silvio, and F. Rivasi. "Human dirofilariasis due to Dirofilaria (Nochtiella) repens: a review of world literature." Parassitologia 37.2-3 (1995): 149-193.

Fig 1: Ultrasound image showing mobile worm within the cyst.

Fig 2: Preoperative image of submental cyst

Fig 3: Intraoperative image

Fig 4: Post operative image of cyst and live worm respectively