



ORIGINAL RESEARCH PAPER

ENT

SUBCUTANEOUS HUMAN DIROFILARIASIS : RELEVANCE OF THIS EMERGING ZONOSIS IN OTORHINOLARYNGOLOGY

KEY WORDS: Human dirofilariasis, subcutaneous nodules

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ABSTRACT

Objective : The aim of this study is to highlight the need to be aware of an emerging zoonosis that could be the cause for common presentations in otorhinolaryngology. There is a need to think out of the box for diagnosing some common presentations where the causative agent is the nematode *Dirofilaria*. Human dirofilariasis is an emerging zoonosis caused by the filarial nematode of genus *Dirofilaria*. Humans are accidental dead end hosts wherein the infective larvae injected through mosquito bites perish before attaining maturity.

Study Design : This is a retrospective study where we present five such cases that presented to the department of ENT in a tertiary care centre over a period of one year.

Results : The study reveals that a high index of suspicion in patients presenting with subcutaneous nodules especially in an endemic area helps in early diagnosis which is mainly assisted by radiological evaluation. The diagnosis enables prompt treatment which is complete surgical excision and confirmation by histopathology.

Conclusion : The common clinical manifestations of human dirofilariasis is either as subcutaneous nodules in various regions in the upper part of the body or as lung parenchymatous lesions. There is an increase in the number of cases of subcutaneous dirofilariasis with varied presentations in otorhinolaryngology. The study well emphasises the avoidance of unnecessary investigations once appropriately diagnosed with role for further studies to understand the aetiopathogenesis and outline definitive guidelines with management strategies.

INTRODUCTION :

Dirofilariasis is an emerging zoonotic infection caused by the filarial nematode of genus *Dirofilaria* which inadvertently infects humans. There are about 40 recognized species of *Dirofilaria* and atleast six of them i.e *Dirofilaria immitis*, *Dirofilaria repens*, *Dirofilaria striata*, *Dirofilaria tenuis*, *Dirofilaria ursi* and *Dirofilaria spectans* are known to cause accidental infections in humans.[1] The subcutaneous lesion is caused mostly by *Dirofilaria repens* and the lung lesion by *Dirofilaria immitis* respectively. The canines are the principal reservoir host and it naturally infects several domestic and wild animals. The transmission to man is by the bite of mosquitoes especially of the anopheles, culex and aedes species.

The review of literature revealed increasing report of cases worldwide with majority of cases reported from southern and eastern Europe, Sri Lanka, Italy, France, Greece and Spain. In India, Kerala is an endemic area; with cases also reported from states of Karnataka, Assam and Orissa. A study of 21 cases of subcutaneous dirofilariasis over 8 years has been reported from Kolenchery, Ernakulam District, Kerala which highlights the increasing number of these cases with predominance of the lesions over the face and anterior chest wall. [2] As the clinical presentation of subcutaneous dirofilariasis may mimic various benign and malignant lesions it is important that the clinician should be familiar of this entity and include it in the differential diagnosis of subcutaneous nodules. Here we present five cases of subcutaneous dirofilariasis that presented to the Department of Otorhinolaryngology in a one year period stressing the varied likely clinical presentations in otorhinolaryngology with ways to help in early diagnosis and appropriate treatment

Materials and Methods : The study was a retrospective review of all cases of subcutaneous dirofilariasis that were diagnosed from August 2015 to August 2016 in the department of otorhinolaryngology at Govt. Medical College Kozhikode; a tertiary care hospital in Northern Kerala. As per the departmental protocol all patients were assessed by detailed history followed by complete otorhinolaryngological examination. The patients were further investigated radiologically with an ultrasonography and/or contrast enhanced CT Scan as deemed necessary. All patients were treated by a complete surgical excision of the lesion with confirmation of diagnosis by the histopathological report.

RESULT :

The retrospective case study yielded 5 patients with subcutaneous dirofilariasis who presented to the department of otorhinolaryngology during the one year period from Aug 2015 to Aug 2016. There were 3 female (60%) and 2 male(40%) patients. The patients were of the age group 15 – 45 years (mean 31.6) with one patient 13 years and another 60 years of age. Of the 5 patients 3 of them presented with subcutaneous swellings adjacent to the nose and orbit with the remaining two presenting as swelling in the neck and cheek respectively. (Table No. 1 summarises the demographic data of patients)

Table No. 1 : Demographic data of patients with subcutaneous dirofilariasis

Sr. No.	Age of the Patient (years)	Sex	Site of the lesion	Duration (months)
1	66	Male	Lateral to the root of the nose (Rt)	3
2	33	Female	Neck (Lt)	2
3	25	Female	Root of nose (Rt)	6
4	13	Female	Cheek (Rt)	2
5	27	Male	Medial to medial canthus of eye(Rt)	4

The need to distinguish this condition from other likely diagnosis; frontoethmoid mucocoele, complicated sinusitis, malignancy becomes important as in our 60 year old patient with a swelling lateral to the root of the nose on the right side. (Fig. 1)



FIG. 1 Clinical presentation of human subcutaneous dirofilariasis in ENT mimicking other likely conditions

The clinical presentation in patients showed that gradually increasing, painless swellings in the head and neck region for a duration of 2 to 6 months was the common symptom with which all the patients presented. There was no history of trauma, fever, pain, dental or orbital complaints. The patients underwent complete hemogram which revealed a high ESR in 3 patients with elevated absolute eosinophil count in one patient. USG was done in 3 patients wherein the report revealed hypoechoic area in the concerned region (i.e the strap muscle on the left side in patient with neck swelling and in the subcutaneous plane in proximity to medial canthus in patient with swelling adjacent to the medial canthus) with hyperechoic tubular structures within suggestive of parasitic granuloma. (Fig 2)

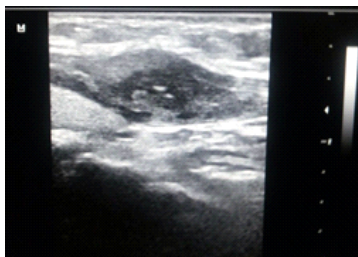


FIG. 2 Ultrasonography : the radiological investigation that helps in early diagnosis. The hypoechoic area with hyperechoic tubular structures is suggestive of parasitic granuloma as seen above in USG Neck of our patient.

The CT Scan was done in two patients where the need to rule out any intraorbital or intracranial extension was felt and the CT Scan done in the 60 year old male patient showed partially enhancing hyperdense lesion with non enhancing area of fluid density in superomedial part of the right eye with no intraorbital lesion. (Fig



FIG. 3 CECT Scan of the 60 year old patient with subcutaneous dirofilariasis showing enhancing hyperdense lesion in the superomedial part of the orbit(Rt) with no intraorbital extension. (3)

All patients underwent complete surgical excision of the lesion with the worm isolated in two of them (Fig 4) and the histopathological report confirmed subcutaneous dirofilariasis in all.



FIG. 4 The worm isolated from an excised lesion in a patient with subcutaneous dirofilariasis

DISCUSSION : Human dirofilariasis can present as subcutaneous nodules in the head and neck region and this makes it necessary to consider the entity as a differential diagnosis in patients who present with this complaint. The comprehensive evaluation of clinical features with blood and radiological investigations helps in early diagnosis. This would enable prompt treatment which is complete surgical excision thereby avoiding unwarranted tests and morbidity.

Subcutaneous human dirofilariasis is caused by zoonotic filarial nematode of the genus *Dirofilaria*. *D. repens* is the main causative agent of subcutaneous dirofilariasis in Asia. [3] Most of the cases reported from India are due to infection with *D. Repens*, some of the *D. immitis* and *D. tenuis* infections have also been reported.[4] The epidemiology of human dirofilariasis is directly correlated to the prevalence of canine dirofilariasis infection.[1] Dogs, monkeys and cats are the primary host and mosquitoes are the intermediate host and vector. Some species of lice, fleas and ticks may also act as potential vectors.[5]

A study of the aetiopathogenesis shows that the adult nematode lives in subcutaneous tissues of their natural hosts and attain full size after which it deposits microfilaria in blood. Vector arthropods ingest the first stage of larva (L1) while feeding on an infected host and L1 microfilaria develop to infective third stage (L3) and migrate to the proboscis. Transmission occurs when the mosquito vector carrying the infective third stage larva(L3) penetrates a new host, which can be either a human or a natural host. Since the humans are dead end host, the nematode in humans does not reach sexual maturity and remains non fertile. Consequently microfilaria are not released into the peripheral blood in humans and due to the presence of an adult nematode in the subcutaneous tissue, chronic inflammatory infiltration can occur in the surrounding tissues forming a parasitic granuloma. As natural transmittance of dirofilariasis is through microfilariae, which anyway does not occur in humans, dirofilariasis cannot be transmitted from person to person.[1]

The prevalence of this disease is increasing and it continues to emerge as a significant health problem in different areas of the world. It is commonly encountered in the 2nd – 4th decade of life with a female predilection as noted in our study too. Human dirofilariasis is categorised into two groups : pulmonary and extra-pulmonary dirofilariasis. Extra-pulmonary dirofilariasis is classified further into four groups: cardiovascular, subcutaneous, visceral and ophthalmic dirofilariasis.[6] The clinical presentation in subcutaneous human dirofilariasis is usually as a single non tender nodule. The patients are mostly asymptomatic with no particular sensation attributable to insect bite. Very rarely satellite lymphadenopathies with hyperpyrexia can occur or subcutaneous migration of parasite in tissues of head can cause trigeminal neuralgia.

The hematological investigations revealed elevated ESR with peripheral eosinophilia mainly owing to chronic inflammatory process. Of the radiological investigation; ultrasonography may contribute significantly to the diagnosis as it does reveal the worm like structure which displays movement within the cyst raising strong suspicion of dirofilariasis. In our study too the suspicion on ultrasonography especially in case of the lesion in the neck; helped an early diagnosis with avoidance of unnecessary investigations. However CT scan is essential to find the extent of the lesion and decide on the approach of surgery based on the site and extent of lesion. Investigations like Giemsa stain and fine needle aspiration cytology followed by excisional biopsy are useful in confirming the diagnosis. Once the excision of the lesion is performed parasitological evaluation can be performed on the worm specimen.[7,8] The serological investigations like measuring the reduction in the level of anti-*D. repens* serous antibodies by immunoenzymatic means for 3 to 6 months from the time of surgery can be used in the follow-up care of patients. It is important that pathologists too must be familiar with the histopathological features of the parasite. The precise identification of species may be achieved with DNA analysis based on polymerase chain reaction, but the large number of species

probes limits the usefulness of this method.

The appropriate treatment is complete surgical excision of the lesion with removal of the adult worm. The diagnosis of dirofilariasis can be made with certainty only after excision biopsy and the species identification is based on the microscopic features of the parasite. Diagnosis of dirofilariasis by histopathology has disadvantages such as when the morphology of the nematode is changed due to an inflammatory response, when the nematode is immature or when the specimen is affected by surgical manipulations. There is no role of chemotherapy as microfilaraemia is extremely rare. In a small number of cases ivermectin and /or diethylcarbamazine has been tried with good results. Antibiotic treatment of the filarial nematodes results in sterility and inhibits larval development and adult worm viability.[9] Treatment with ivermectin (6 mcg/kg weekly) combined with doxycycline (10 mg/kg/day orally) resulted in a significantly faster decrease of circulating microfilariae and higher adulticidal activity compared with either ivermectin or doxycycline alone.[10] Anthelmintics are not usually recommended after surgery. However oral treatment with diethylcarbamazine(DEC) 2mg/kg/day over a period of 4 weeks and /or oral ivermectin (150 mcg per kg) may be given post surgery to prevent recurrences.[11]

CONCLUSION : Human subcutaneous dirofilariasis is a fast emerging zoonosis in India with a definite increase in incidence especially in endemic areas. The increased awareness of this entity is necessary to raise a high index of suspicion in patients presenting with subcutaneous nodules in the upper part of the body. An otorhinolaryngologist needs to be aware of the likely varied presentations so that relevant investigations can be done and appropriate treatment planned thereby reducing morbidity. The treatment of choice is complete surgical excision and removal of the worm. The confirmation of diagnosis is essentially by histopathology. A prospective study in the future with species identification could help elucidate further facts with regard to this zoonosis.

Conflicts of Interest : None

Compliance with Ethical Standards : This is a retrospective study. "For this type of study formal consent is not required."

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