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RESEARCH ARTICLE

PERIOCULAR DIROFILARIASIS IN A PATIENT ON ANTICANCER CHEMOTHERAPY

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ABSTRACT

Dirofilaria repens is an endemic parasite in Mediterranean countries that mostly affects animals. Rarely, it can infect humans causing inflammatory subcutaneous swelling. We present a 67 years old male patient with history of chemotherapy for carcinoma bladder who presented with painless swelling about 2x1.5cm approximately 4cm from the lower lid. The mass was freely mobile with no signs of inflammation. The excision biopsy showed three filarial parasites of Dirofilaria Repens species in the mass with mild granulation around them. The serology was negative. The case report showed that ocular filariasis can present as a non inflammatory tumour of the eyelid in non endemic regions (Tamil Nadu, where it is not often reported) also with blood serology being negative due to chronic immunosuppresion due to anticancer medications and radiotherapy.

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INTRODUCTION

Post Dirofilaria is a genus of the family Onchocercidae of the super family Filaroidea, order Spirurida in the subphylum Nematoda. It is most common in the Mediterranean countries like Italy. In India it is most commonly found in Kerala followed by Assam, Gujarat, Maharashtra, Punjab. The first case presented in Kerala in 1976 (Patel, 2014). Human ocular dirofilariasis is sporadically reported in different parts of the world. The species affecting humans are D. Immitis, D. Repens, D. Striata, D. Tenius, D. Ursi, D. Spectans (Nath, 2010). The most common presentation is inflammatory swelling of the ocular adnexa. It rarely can present as a non inflammatory swelling. We report non inflammatory presentation of dirofilariasis in lid mimicking as lipoma on anticancer chemotherapy in Tamil Nadu, state in sourthern India where it is not often reported.

Case History

A 67 year old male belonging to Tamilnadu state in India presented with the complaints of painless swelling in the lower lid of right eye gradually increasing in size. Best corrected visual acuity in right eye was 6/9 and in left eye was 6/6.

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Other ocular findings were within the normal limits, except for previous history of cataract surgery. He was a known case of carcinoma bladder treated with chemotherapy earlier.No H/O of previous pain, tenderness or fever.There was no history suggestive of moving larva migrans. Slit lamp examination showed a swelling of size 2×2cm (Fig 1). On palpation the swelling was non tender, well defined, soft in consistency, freely mobile in all directions. The differential diagnosis considered was a lipoma and excision biopsy was undertaken. It was a well encapsulated mass with yellowish tinge about 2×1.7 cm, and was soft in consistency (Fig 2). The histopathological report showed fibroadipose tissue and skeletal muscle bundles with cut sections of 3 filarial parasites. All 3 parasites showed a smooth thick multilayered cuticle, muscle layer with transverse striations morphological features suggestive of Dirofilariasis Repens (Fig 3). Each parasite was surrounded by a dense mixed inflammatory infiltrates comprising of lymphocytes, neutrophils, plasma cells, eosinophils, and histiocytes. Cross sections showed double uterus and central intestinal tubes. A routine blood examination was sent and was within normal limits except for elevated ESR of 21 mm at 1 hour postoperatively. Blood serology for Dirofilariasis (by ELISA) was found to be negative. Patient was sent to infectious disease department for systemic evaluation of Dirofilariasis and there was no systemic filariasis detected. Post operatively he was advised CT scan of orbit and brain was found to be normal.

Fig. 1. Preoperative picture of right lower lid subcutaneous mass

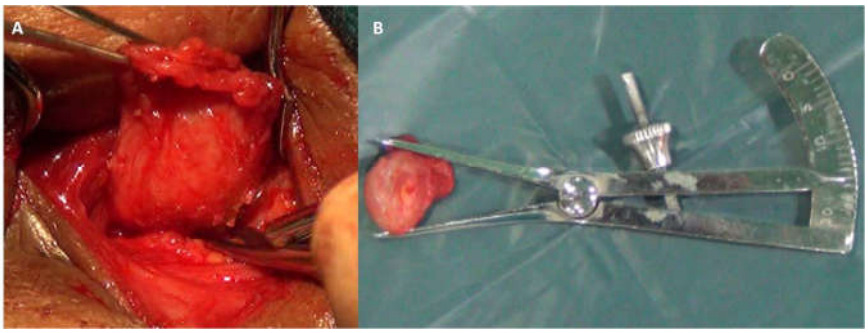

Fig. 2. A. Intraoperative picture showing the plane of excision and B. Mass removed in toto measured size 20mm x17mm

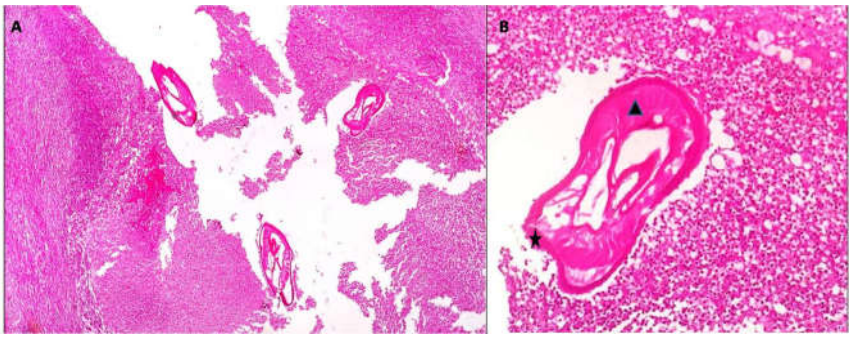

Fig. 3. A. Histopathology section of the mass showing three Dirofilarial parasites surrounded by inflammatory exudates (Hematoxylin & Eosin stain, 40X). B.Cross section showing thick cuticle (star) and muscular layer (triangle) (Hematoxylin & Eosin stain, 100X). Double uterus and centrally placed intestinal tube seen on cross section

Table 1. Demographic distribution of subcutaneous Dirofilariasis in India

Nagdir *et al* (2001)	Subconjunctival dirofilariasis in India.	Karnataka
Ittyerah TP *et al* (2004)	A case of subcutaneous dirofilariasis of the eyelid in the South Indian state of Kerala	Kerala
G Mahesh *et al* (2005)	A Case of Periocular Dirofilariasis Masquerading as a Lid Tumour	Kerala
Nath R *et al*(2010)	Ocular dirofilariasis	Assam
Khurana *et al*(2010)	Human subcutaneous dirofilariasis in India: a report of three cases with brief review of literature	Karnataka & Maharashtra
Pauly *et al*(2013)	Periocular dirofilariasis mimicking lacrimal sac mucocoele	Kerala
kyreim *et al* (2013)	Subcutaneous dirofilariasis	Meghalaya
Kombade *et al* (2015)	Subcutaneous human Dirofilariasis in Vidarbha.	Maharashtra
Premi *et al* (2011)	Subcutaneous Human Dirofilariasis Due to *Dirofilaria Repens*: Report of Two Cases	Kerala

DISCUSSION

The life cycle of *Dirofilaria* species comprises a definitive vertebrate host and a vector. Both *D.immitis* and *D. repens* demonstrate poor vertebrate host specificity given that they can infect numerous mammalian species. Among mammalian hosts, they are best adapted to domesticated and wild dogs, which function as reservoirs. Humans and cats are less suitable hosts, in which parasite development is dramatically modified compared with the patterns in dogs.

The vectors are females of various mosquito species of the *Culicidae* family. The dirofilaria are accidentally transmitted to humans by bite of mosquitoes carrying infective larvae. Dirofilaria cannot mature fully in human tissue and dies before producing microfilaria. Most cases with ophthalmic infection present with pain in the eye, redness, sometimes blurred vision, and swelling of eyelids, which coincides with the worm entering the subconjunctiva. Ocular presentation of Ocular Dirofilariasis can be in the form of subconjunctival cysts, eyelid swellings, anterior chamber, nodules, painful inflammatory swelling, abscess in subcutaneous tissue. In

published reports of ocular dirofilariasis, most of the cases were located under the conjunctiva (>60% of all cases) followed by orbital/eyelid dirofilariasis (approximately 25%). Most reports of ocular dirofilariasis originate from Europe (Italy, France, Greece, Croatia, Serbia, Denmark, and Russia), Africa (Tunisia), Mediterranean countries and South Asia. To the best of our knowledge, in India commonly affected states are Kerala, Assam, Gujarat, Maharashtra, and Punjab (Table 1) (Nadgir, 2001; Mahesh *et al*., 2005; Font, 1980; Khurana, 2010; Pauly, 2013; Khyriem, 2013 and Kombade, 2015). There is one report of Dirofilariasis in Tamil Nadu which was clinically visible under the teno-conjunctiva (Sathyan, 2006). Pauly *et al* has reported a case of Dirofilariasis mimicking lacrimal mucocele and Mahesh *et al*. (2005) as lid tumour (Mahesh, 2005 and Pauly, 2013). The case is presented as there no reports of periocular dirofilariasis in this part of India (Tamil Nadu) and presenting with non inflammatory signs in a patient on chronic chemotherapy and radiotherapy. Most of the eyelid subcutaous dirofilariasis swelling were associated with inflammatory signs of the eye (Patel, 2014; Nath, 2010 Nadgir, 2002; Font, 1980; Khyriem, 2013 and Kombade, 2015).

However, it can rarely present as a painless swelling also (Mahesh *et al*., 2005 and Pauly, 2013). Since the patient in our case was treated with chemotherapy and radiotherapy earlier, the immune system has not reacted by producing inflammatory response to the parasite. Chemotherapeutic agents are used widely in clinical medicine and oncology for the treatment of conditions where cytotoxicity activity on the neoplastic cell is the objective. The chemotherapy is given in the form of Ionizing radiation or systemic drugs. In both ways, they have the tendency to affect humoral and cellular response of the body. One of the unfortunate sequelae of chemotherapy induced immunosuppression is an increased susceptibility of the host to opportunistic pathogens. Apart from Malignancy, chemotherapy is indicated for immunologically mediated disease, lymphoproliferative diseases, and prevention of graft rejection. The differential diagnosis considered in this case was non inflammatory mass like lipoma. Our case report is also about the unusual presentation of an non-inflammatory swelling with blood serology being negative for dirofilariasis rarely due to immunosuppresion as a part of comorbidity (in this case Carcinoma Bladder). Hence clinicians should be aware regarding the possibility of this zoonotic infection in cases of localized nodules in any part of the body.

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