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 causes subcutaneous swelling or infections of bulbar conjunctiva and is rare in India. D immitis causing pulmonary infections is limited to the New World. We present a case of a 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 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, muskrats 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 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% population of dogs being infected whereas in Europe,the highest number of cases have been recorded in Italy. (14)

D.repens, D tenuis and D ursi is 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 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 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, dirofilaria 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) 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 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.

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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