

Case Report

A case report on facial subcutaneous dirofilariasis with intraoral extension

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ABSTRACT

Dirofilariasis is parasitic nematodes of domestic and wild animals that can infect humans accidentally via vectors. This disease is endemic in South Eastern United States, Australia, Europe and Central and Southern Asia. *Dirofilaria immitis* and *Drepens* are the common mosquito borne filarial nematodes that cause infection. The most frequent presentation of human dirofilariasis is a single submucosal nodule without signs of inflammation. Oral dirofilariasis is extremely rare and only a few cases have been documented. We report an interesting case of subcutaneous dirofilariasis in a patient who presented with a facial swelling. Laboratory investigations and radiographs were non-contributory to diagnosis. This paper stresses the importance of considering dirofilariasis as differential diagnosis for subcutaneous swelling of the face with intraoral extension, especially in areas where it is endemic.

Keywords: Dirofilariasis, Facial, Intraoral, Nematode, Subcutaneous

INTRODUCTION

Dirofilariasis is a rare parasitic infection caused by helminthic nematode belonging to the genus dirofilariasis. It is a habitual zoonotic parasite of canine. Human beings are rarely affected by accidental infection and usually present as pulmonary, peritoneal, ocular or subcutaneous lesions. Among the 40 species recognized, fewer than six are known to cause human infections.^{1,2} The species responsible for subcutaneous dirofilariasis is *Drepens*. Dirofilariasis has not been widely recognized in India, but a few cases have been reported from Kerala.^{3,4}

Dirofilariasis is transmitted to man through the bite of potential mosquito vectors. Exposed part of the body including the head, neck and lower extremities form the common involved subcutaneous site. Dirofilariasis presenting as a facial swelling is extremely rare and only a few cases have been documented. We report an interesting case of subcutaneous dirofilariasis of the face.

Diagnosis was based on the identification of dirofilariasis worm in the excised tissue sections.

CASE REPORT

A 58 years F presented with a non-progressive, painless swelling of 5 months duration over right cheek of sudden onset with a history of single episode of enlargement of right side of the face, associated with pruritis. Local examination revealed an oval, non-tender, firm swelling of 1×0.5 cm with ill-defined margin in the right cheek with no facial asymmetry and overlying normal skin. Intraorally palpable in the buccal mucosa (Figure 1a). CBC and ESR were within normal limits. USG showed 9.8×6.0 mm inflammatory parasitic cystic lesion over right cheek lying within masseter muscle with foci of calcification.

On the basis of above findings most probable diagnosis was thought to be calcified lymph node or tuberculous lesion. Other differentials considered were sialolithiasis,

pleboliths and traumatic myositis ossificans. For confirmation excision biopsy was done.

Intra-operative findings

Location of swelling and proposed skin was marked. Adrenaline was infiltrated. Submandibular linear skin incision was made. Meticulous dissection done subcutaneously. Nodular swelling was gently separated from buccal nerve and other adhesions, excised and sent for HPE. Suction drain was placed and skin closure done. Review of the patient after 2 weeks showed complete resolution of postoperative edema with healing of sutured wound Figure 1 (b-d).

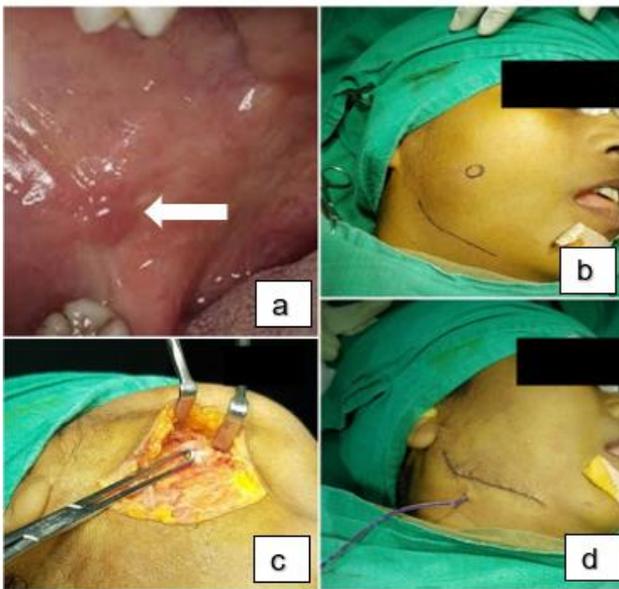


Figure 1 (a-d): Clinical and intra-operative findings.

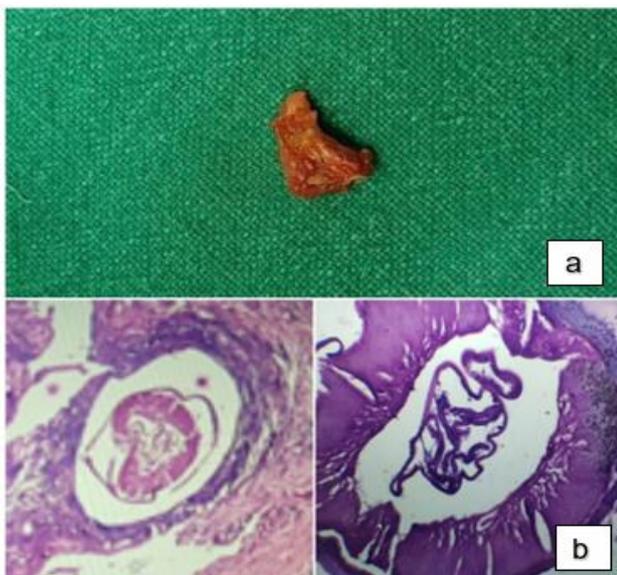


Figure 2: (a) Excised specimen and (b) histology of dirofilariasis.

Histopathological examination

Histopathological examination suggested histiocyte rimmed necrotic nodule containing fragments of parasites, morphologically consistent with features of dirofilariasis. Inflammatory cell infiltration seen. The parasite is composed of a thick cuticle layer with longitudinal ridges, myoid tissue fibres, single gut tube and genital tubule; suggestive of subcutaneous *Drepens* (Figure 2).

DISCUSSION

Dirofilaria repens is a common subcutaneous parasite of dogs. They live in subcutaneous tissues of their hosts and produce microfilariae which are accidentally transmitted to man through the bite of *Aedes*, *Anopheles* or *Culex* mosquitoes. In man, infection by *Dirofilaria repens* usually present as a subcutaneous nodule localized around eyes, eyelids and conjunctiva. Intraoral involvement is extremely rare. The limited number of cases documented, showed a predilection for buccal mucosa followed by lips.^{3,5,6} Our patient presented with an ill-defined facial swelling with intra oral manifestation, associated with pruritis. Due to rarity of the lesion, dirofilariasis was not considered a diagnostic possibility, clinically. The specks of calcification within the hypoechoic lesion seen in USG, turned out to be the subcutaneous inflammatory nodule harbouring the dirofilarial worm on microscopic examination; which is the gold standard for diagnosis. A thick laminated cuticle with external longitudinal ridges and the presence of a well-developed circumferential musculature interrupted by two lateral cords are characteristic features of *Dirofilaria*.⁷ The differentiation among the species is done based on the size, thickness of the cuticle and its structure, ridges and type of muscle cells. *D. repens* are identified by longitudinal ridges separated by a distance wider than the ridge itself, 95-105 ridges on the circumference of the body and 2-5 chord nuclei in each cross section of worm.⁸ However, species identification is not always possible because of strong inflammatory response and in such situations parasites are correlated with its geographic distribution. In this case, even if the granulomatous tissue reaction was noticed, the morphology of the worm was well-preserved and identification was possible. Human dirofilariasis is usually regarded as infection by a single worm. Hence, excisional biopsy is curative and no further treatment is required.

CONCLUSION

Dirofilariasis is a rare emerging zoonosis and may present as facial subcutaneous swelling. Making a diagnosis for such swelling may be difficult because of multiple differentials and asymptomatic characteristics of the nematode. For definitive diagnosis, it is also equally important for the pathologists to be familiar with the diagnostic method for this disease.

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