

Case Report

Appearances are deceptive: a case report on subcutaneous dirofilarial granuloma mimicking a thyroglossal cyst

Adheela Nusrin K. K.*, Amal John Jacob, Ramachandran Kizhakkethil, Nivee Narayanan

Department of ENT and Head and Neck Surgery, KMCT Medical College, Mukkam, Kozhikode, Kerala, India

Received: 25 April 2023

Revised: 01 June 2023

Accepted: 02 June 2023

*Correspondence:

Dr. Adheela Nusrin K. K.,

E-mail: adheelanusrin@gmail.com

Copyright: © the author(s), publisher and licensee Medip Academy. This is an open-access article distributed under the terms of the Creative Commons Attribution Non-Commercial License, which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

ABSTRACT

Dirofilarial granuloma is a nematode infestation of subcutaneous tissue transmitted by mosquito bite. This is a case of 24 year old male who presented with complaints swelling in mid region of anterior neck and on examination patient had a firm midline neck swelling 2x2cms size which had minimal movement with deglutition respiration and no movement on protrusion of tongue. Initial ultrasonogram of lesion was suggestive of a subcutaneous swelling probably thyroglossal cyst. As the clinical findings were discrepant with ultrasonogram findings we repeated imaging and repeat ultrasonogram suggested possibility of worm granuloma. Patient underwent excision and worm could be isolated from the specimen. The specimen along with worm was sent for HPE and diagnosis of dirofilarial granuloma was confirmed.

Keywords: Neck swelling, Thyroglossal cyst, Dirofilariasis

INTRODUCTION

A thyroglossal duct cyst is the commonest congenital cervical anomaly. It is an embryologic remnant formed in a persistent thyroglossal duct somewhere along the course of descent of thyroid gland which is closely associated with hyoid bone.^{1,2} Dirofilariasis is a zoonotic infection which is caused by nematode *Dirofilaria*, *D. immitis*, *D. repens*, *D. tenuis* and *D. ursi*. It is a vector borne disease mainly transmitted by *Culex*, *Aedes* or the *Anopheles* mosquito species. Human dirofilariasis is rare though its incidence is increasing owing to global environmental change. Most common presentation is nodular lesions in the lung, subcutaneous tissues/eyes.³

CASE REPORT

A 27 year old male presented with c/o swelling in mid region of anterior aspect of neck for past 4 months with

no h/o pain, dyspnea, dysphagia. On examination it was a 2x2 cm firm non tender anterior midline neck swelling which had minimal movement with deglutition and no movement on protrusion of tongue. He had done an ultrasonogram from elsewhere which was suggestive of a subcutaneous swelling probably being a thyroglossal cyst with respect to its site. Initial plan was to go for a Sistrunk procedure. But there was a high degree of suspicion as the clinical findings were not in conjunction with that of a thyroglossal cyst. Although the site of the swelling was in favour of a thyroglossal cyst, there was only a minimal movement with deglutition and no movement on protrusion of tongue.

A repeat ultrasonogram was done which revealed a well-defined thick-walled cystic lesion towards right of midline in subcutaneous plane anterior to strap muscles with hyperechoic tubular rod like structure within.



Figure 1: Anterior midline neck swelling.



Figure 2: USG neck showing well defined thick-walled cystic lesion in subcutaneous plane.

He underwent excision under local anaesthesia and careful dissection of mass revealed a 6 cm long worm within.



Figure 3: Excised specimen along with the dissected-out worm.

The excised specimen along with worm was sent for histopathological examination which revealed fibro-cartilaginous tissue fragments composed of a cyst lined by lymphoplasmacytic infiltrate and cross section of dead worm surrounded by granulomas composed of foreign body giant cells, foamy macrophages and dense mixed inflammatory infiltrates.



Figure 4: Longitudinal section of the worm on hematoxylin and eosin staining showing thick cuticle and internal tubular structures

DISCUSSION

Francesco Birago made the first known reference to filarial infestation in dogs in the 17th century by describing the presence of adult *Dirofilaria immitis* worms inside the hearts of his hunting dogs.⁴ Human dirofilariasis was first reported in 1962 from Sri Lanka in a 15-year-old boy.⁵ Dogs, monkeys, and cats are the primary host and mosquitoes such as *Mansonia uniformis*, *Mansonia annulifera*, and *Aedes aegypti* are considered potential vectors. The worms found in humans were unable to mature and hence cannot express larvae in the bloodstream. Anti-filarial/ anthelmintic drugs are ineffective in majority of the documented cases as there are no microfilariae in the blood stream.⁵

The most common presentations of dirofilariasis are subcutaneous and submucosal lesions. Diagnosis is usually based on a high clinical suspicion in patients from endemic areas. Enzyme linked immunosorbent assay (ELISA) for antibody response to *D. repens* somatic antigen is a useful adjuvant test in endemic areas with a strong clinical suspicion prior to surgery, although it has been found to be negative in many cases. High-resolution ultrasonography is the imaging modality of choice, as live motile worms can be visualized in real time. As a result, full nodule excision combined with worm extraction is usually successful. If secondary lesions are suspected in deep body sites such as chest or abdomen, a course of ivermectin and diethylcarbamazine (DEC) is advisable to prevent further invasive surgery. Treatment with tetracyclines has been reported to damage *D. immitis*, even causing death of adult worms.⁷

Although human infection with *D. repens* has been increasing in South Asia and other parts of the world, many of them remain undiagnosed or unpublished. This can create a disease epidemic situation with the presence of potential vectors for transmission. Hence, there is need for increased awareness about this infection, diagnostic tests and vector controlling measures, in order to reduce the disease prevalence through suppressing vector densities, proper diagnosis and patient care

CONCLUSION

Even though subcutaneous dirofilariasis is rare, it can be included in the differential diagnosis, when patient presents with a firm submucosal nodule without overlying inflammatory signs which does not completely respond to routine therapy, especially in patients from endemic areas.

Funding: No funding sources

Conflict of interest: None declared

Ethical approval: Not required

REFERENCES

1. Garcia E, Osterbauer B, Parham D, Koempel J. The incidence of microscopic thyroglossal duct tissue superior to the hyoid bone. *Laryngoscope*. 2019;129(5):1215-7.
2. Ma J, Ming C, Lou F, Wang ML, Lin K, Zeng WJ, et al. Misdiagnosis analysis and treatment of pyriform sinus fistula in children. *Zhonghua Er Bi Yan Hou Tou Jing Wai Ke Za Zhi*. 2018;53(5):381-4.
3. Pampiglione S, Rivasi F. Human dirofilariasis due to *Dirofilaria* (Nochtiella) repens: an update of world literature from 1995 to 2000. *Parassitologia*. 2000;42(3-4):231-54.
4. Birago F. Trattato cinegetico, ovvero della caccia. Sfondrato V, Milan, Italy. 1626;77.
5. Iddawela D, Ehambaram K, Wickramasinghe S. Human ocular dirofilariasis due to *Dirofilaria repens* in Sri Lanka. *Asian Pac J Trop Med*. 2015;8(12):1022-6.
6. Acharya D, Chatra PS, Padmaraj SR. Subcutaneous dirofilariasis. *Singapore Med J*. 2012;53(9):184.
7. Giannelli A, Ramos RA, Traversa D, Brianti E, Annoscia G, Bastelli F et al. Treatment of *Dirofilaria repens* microfilariaemia with a combination of doxycycline hyclate and ivermectin. *Vet Parasitol*. 2013;197(3-4):702-4.

Cite this article as: Adheela NKK, Amal JJ, Ramachandran K, Narayanan N. Appearances are deceptive-a case report on subcutaneous dirofilarial granuloma mimicking a thyroglossal cyst. *Int J Otorhinolaryngol Head Neck Surg* 2023;9:596-8.